The Conceptualisation, Development and Validation of a Generic Health-Related Family Quality of Life Measure

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Catherine Golics

School of Pharmacy and Pharmaceutical Sciences & School of Medicine

Cardiff University

Cardiff

UK

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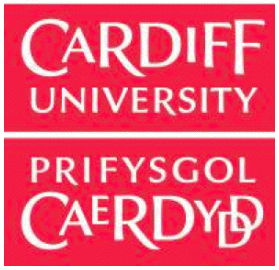

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ABSTRACT

Chronic conditions have an impact on the quality of life (QoL) of families as well as patients themselves, and the two are often linked; the greater the effect on the patient, the more the QoL of the family members is reduced. Research into family QoL exists in several medical specialties, but studies have usually been focused on carers or families of patients with one specific disease. Currently, there is no generic instrument that can be used to measure the impact of illnesses on the partner or family members of patients. This study describes the development of the Family Reported Outcome Measure (FROM-16)©. The aims of this study were to investigate the impact of disease on family members of patients over a wide range of specialties, identify key impact areas and develop a generic family quality of life measure.

Semi-structured interviews were carried out with 133 family members of patients from 26 medical specialties. Family members were invited to discuss all the areas of their lives that had been affected by having an unwell relative. Thematic analysis was carried out using NVivo9© software. A preliminary 31-item measure was developed from the content of the interviews with family members. Content validity was assessed using qualitative and quantitative data from expert panels involving clinicians and family members. A separate cohort of 240 family members was recruited for both Rasch analysis and factor analysis to reduce items. A further 120 family members completed the final version of the FROM-16 for full psychometric testing including construct validity and reliability.

Most family members interviewed were female (61%), the partner or spouse of the patient (56%) or the parent (22%). The mean age was 56.1 years (range= 21-85) and the mean duration of the patient's disease was 8.9 years (range= one month to 60 years). 10 key themes of family quality of life were identified from interviews. The median number of themes reported by family members was 6 (range= 1-10). The key themes included: emotional impact (mentioned by 92% of subjects), daily activities (91%), family relationships (69%), sleep and health (67%), holidays (62%), support and medical care (61%), work and study (52%), financial impact (51%), social life (37%), and time planning (14%). Relationships between the themes were identified.

A 31-item generic family quality of life instrument, the Family Reported Outcome Measure (FROM)©, with a 5-point Likert response scale was developed. The content validity panel's ratings of each item on a 4-point scale for the four attributes showed either "strongly agreed" or "agreed" (88%), with an ICC value of 0.98 (CI=0.97-0.99) suggesting a high agreement between the panel members' responses.

Collapsing response categories, removing misfitting items and combining items with residual correlations produced a good fit to the Rasch model (n=240, Total χ^2 = 56.6, df = 48, p =

0.18). Factor analysis produced a 16-item measure with two factors. The FROM showed high internal consistency (n=120, Cronbach's α= 0.91), high reproducibility (n=51, ICC=0.93) and a mean completion time of two minutes. Construct validity was proven through the correlation between the FROM and the WHOQOL-BREF total scores (n=119, r=-0.55, p<0.001), and the correlation between the FROM and the patient's overall health score (n=120, r=-0.51, p<0.001).

This large scale multi-specialty study has demonstrated the great, yet similar impact that illness can have on the quality of life of family members of patients. Family quality of life is a previously neglected area of healthcare which needs to be addressed in order to provide better support for the patient and for the family unit. The FROM is both reliable and valid for use in family members of patients. It has a potential for wide use, including clinical (all medical specialties), industrial and social sciences.

LIST OF ABBREVIATIONS

ADHD	Attention deficit hyperactivity disorder
AMED	Allied and complementary medicine
ANOVA	One way analysis of variance
CAT	Computer adaptive testing
CFA	Confirmatory factor analysis
CVI	Content validity index
DFI	Differential item functioning
DIF	Differential item functioning
EFA	Exploratory factor analysis
EMA	European Medicines Agency
FDA	Food and drug administration
FDLQI	Family dermatology life quality index
FROM	Family Reported Outcome Measure
GH	Global health
GHS	Global health score
GP	General practitioner
HIV	Human immunodeficiency virus
HMIC	Health management information consortium
HRQoL	Health-related quality of life
ICC	Intraclass correlation
I-CVI	Item content validity index
KMO	Kaiser-Meyer-Olkin
NICE	National Institute of Clinical Excellence
PASW	Predictive Analytics SoftWare Statistics
PRO	Patient reported outcomes
PSI	Person separation index
QALY	Quality adjusted life year
QoL	Quality of life
S-CVI	Scale content validity index
UHW	University Hospital of Wales
WHO	World Health Organisation

GLOSSARY OF TERMS

PRO measure	Any report of the status of a patient's health condition that comes directly from the patient, without interpretation of the patient's response by a clinician or anyone else.
Construct validity	Evidence that relationships among items, domains, and concepts conform to *a priori* hypotheses concerning logical relationships that should exist with other measures or characteristics of patients and patient groups
Content validity	Evidence from qualitative research demonstrating that the instrument measures the concept of interest including evidence that the items and domains of an instrument are appropriate and comprehensive relative to its intended measurement concept, population, and use.
Face validity	Whether, on the face of it, the instrument appears to be assessing the desired qualities.
Factor analysis	The process of summarising or reducing a large set of variables using a smaller set of factors or components.
Rasch analysis	A statistical technique used for investigating what should be expected in responses to items if measurement is to be achieved
Reliability	The ability of a PRO instrument to yield consistent, reproducible estimates.
Practicality	Describing the feasibility of using an instrument in its intended population and clinical setting.
Sensitivity to change	Evidence that a PRO instrument can identify differences in scores over time in individuals or groups who have changed with respect to the measurement concept.
Likert scale	A bi-polar response scale with descriptors ranging from none or little of an attribute at one end, to a lot or maximal at the other end.
Regression analysis	Exploring the predictive ability of a set of independent variables on one continuous dependent measure.
Purposive sampling	Sampling with a purpose in mind, or previous knowledge of the population.
Saturation point	When interviewing patients, the point when no new relevant or important information emerges and collecting additional data will not add to the understanding of how patients perceive the concept of interest and the items in a questionnaire.
Thematic analysis	A method for identifying, analysing, and reporting patterns (themes) within data. It minimally organises and describes a data set in rich detail.
Recall period	The period of time patients are asked to consider in responding to a PRO item or question.

Adopted from: (Bhatti 2011; Braun and Clarke 2006; FDA 2009; Pallant 2005; Pallant and Tennant 2007; Streiner and Norman 2008)

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CHAPTER 1

General Introduction

BACKGROUND

In 1948, the World Health Organisation (WHO) defined health as "a state of complete physical, mental and social well-being and not merely the absence of disease" (The WHOQOL Group 1948). Since then, interest in the concepts of well-being and quality of life have grown, both in clinical and research environments. The definition of Quality of Life (QoL) varies between authors, with the most commonly used definition provided by the WHO: "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (The WHOQOL Group 1997). The definition of QoL often depends on the area of one's life which is being assessed, with the term "Health-related quality of life" (HRQoL) being applied to the impact of illnesses or treatments on the lives of patients or those around them. The term QoL when applied in health studies often only includes those areas of patients' lives which are relevant to their illness. Assessing the effect of health on a patient's life using the term HRQoL helps to ensure that all areas of a patient's life are included in the assessment, and not just the obvious physical effects of the illness. Quality of Life is a multidimensional concept, which can be difficult to define or measure, as it is often made up of a number of different components of a patient's life, which can often vary in importance from patient to patient. Rather than just a measure of health, QoL takes into account social, physical and psychological factors, and is more than just a measure of well-being. For the purpose of this thesis the terms "Quality of Life" and "Health-related quality of life" will be used interchangeably.

Table 1.1 shows the variation in definitions of QoL between authors. At the most basic level, the Oxford English Dictionary (2004) defines QoL as "The standard of living, or degree of happiness, comfort, etc., enjoyed by an individual or group in any period or place; an instance of this." Although, like other definitions, this description relates the term "happiness" to QoL, it does not pick up on the particular areas of an individual's life which are impacted. On the other hand, the definition given by Haas (Haas 1999), places the concept of QoL in context of the person's own environment: "QOL is a multidimensional evaluation of an individual's current life circumstances in the context of the culture and value systems in which they live and the values they hold. QOL is primarily a subjective sense of well-being encompassing physical, psychological, social, and spiritual dimensions. In some circumstances, objective indicators may supplement or, in the case of [people] unable to subjectively perceive, serve as a proxy assessment of QOL." The differences in detail of each definition are clear, but the one thing they all have in common is that they all convey the idea of QoL being a subjective concept, using phrases such as "an individual's perception" and "that individual's present experiences". QoL relates directly to an individual's hopes and expectations; improving an individual's QoL is an attempt to reduce the gap between the

present and that individual's expectations, which can vary greatly from person to person (Bhatti 2011). It is therefore important that quality of life is measured by patients themselves, as measurement by other individuals such as healthcare professionals can be inaccurate (Slevin et al. 1988). The subjective nature of QoL can manifest itself in several ways. The external factors which contribute to an individual's environment or circumstances vary hugely from person to person, for example the makeup of the patient's family, socioeconomic status, job or career, religion and relationships. These factors all have the potential to influence the patient in both a negative and positive way, and each factor will be seen as more or less important by individual patients. Often, the individual's perspective on their QoL is influenced by past experiences relating to these factors. Another way in which QoL is subjective is defined by the individual's personality, character and attitude to life. All three of these aspects have the potential to contribute to how a patient views their QoL, and is unique to each individual. This provides a further reason as to why QoL is best assessed by the individual concerned, as it may be difficult for a proxy measure to recreate the unique balance between environmental factors, personal factors and past experiences and future hopes and expectations. However, there are some circumstances where it may be necessary for another individual (e.g. a member of the healthcare team or family member) to act as a proxy to the patient when measuring QoL, such as when the patient is a very young child, or seriously unwell.

The measurement of QoL falls under the umbrella term "patient reported outcomes" (PROs), which includes information provided by the patient, usually in relation to symptoms, disease, treatment, preferences, satisfaction or experiences (Doward et al. 2010). Quality of life assessment is often used alongside other forms of PROs, for example symptom assessment, or the measurement of health behaviour, to create a true picture of the disease. Alongside PROs, other forms of assessment where QoL can be measured include observer reported outcomes, where the quality of life measurement is made by someone other than the patient (usually a family member or clinician), as described above. The concepts used when measuring QoL can be derived from past literature (Farquhar 1995), or directly from patients (Basra and Finlay 2007), for example through interviews, questionnaires or focus groups. Both methods have their advantages; literature reviews can provide a broad overview of the important issues for patients with a particular disease, whereas interviews with patients can provide "real-life" information including quotes and examples from patients. However, the latter approach reflects the way patients describe themselves in relation to their functional behaviour, both physical and psychosocial. This helps ensure reliability and validity of the resultant QoL instrument.

Table 1.1: Definitions of Quality of Life

Author	Definition
(Calman 1984)	Measures the difference, or the gap, at a particular period of time between the hopes and expectations of the individual and that individual's present experiences.
(Cella and Cherin 1988)	Refers to patients' appraisal of and satisfaction with their current level of functioning compared to what they perceive to be possible or ideal.
(Clinch and Schipper 1993)	The perception of the impact of the disease and is both subjective and culturally bound.
(Cohen et al. 1996)	"Subjective well-being"
(Felce and Perry 1995)	Quality of life is defined as an overall general wellbeing that comprises objective descriptors and subjective evaluations of physical, material, social, and emotional wellbeing together with the extent of personal development and purposeful activity, all weighted by a personal set of values.
(Ferrans 1990)	A person's sense of well-being that stems from satisfaction or dissatisfaction with the areas of life that are important to the individual.
(Haas 1999)	QOL is a multidimensional evaluation of an individual's current life circumstances in the context of the culture and value systems in which they live and the values they hold. QOL is primarily a subjective sense of well-being encompassing physical, psychological, social, and spiritual dimensions. In some circumstances, objective indicators may supplement or, in the case of [people] unable to subjectively perceive, serve as a proxy assessment of QOL.
(Oxford English Dictionary 2004)	The standard of living, or degree of happiness, comfort, etc., enjoyed by an individual or group in any period or place; an instance of this.
(WHOQOL 1993)	An individual's perception of his/her position in life in the context of the culture and value systems in which he/she lives, and in relation to his/her goals, expectations, standards and concerns. It is a broad-ranging concept, incorporating in a complex way the person's physical health, psychological state, level of independence, social relationships, and their relationship to salient features of their environment.

Due to the complex, multidimensional nature of QoL, it is a concept which can be difficult to quantify. Quality of life can be assessed in a number of different ways depending on the type of study and intended outcomes. Using QoL instruments which have been validated and standardised can allow the comparison of results across disease states and groups of patients, whilst taking into account the different factors which influence the patient's QoL, for example social, physical and psychological. In order to account for individual variation in the influence of each factor on different patient's lives, QoL instruments often allow the user to rate the level of the impact of each of the factors on their QoL (e.g. 'not at all' ranging to 'a lot').

Types of Quality of Life Instruments

There are two main types of QoL instruments; generic and disease-specific measures. Generic measures can be used over a large group of patients, and give a measure of QoL which is not related to a specific disease, but can include the effects of a wide range of diseases or treatment. This is particularly useful when measuring the impact on QoL of a community or group of patients, rather than individuals (Huebner et al. 2004), as the scores can be used to directly compare individual patients, or identify population-wide trends. Generic measures tend to include broader items, designed to be applicable to a wide population. In contrast, disease-specific measures are designed to be used to assess QoL in a specified disease, and can detect changes in individuals' QoL following clinical interventions. These measures, which can be used to determine the extent to which a patient has been affected by suffering from a disease, can help clinicians to decide on appropriate treatment, but cannot be used to compare across conditions or treatment programs (Revicki and Ehreth 1997). Disease-specific measures contain items which are particularly relevant to the disease in question, for example items measuring specific symptoms or the impact of the clinical features of the disease. On some occasions, generic measures are used in patient groups suffering from specific diseases, but give a much less detailed picture of the patient's QoL, especially when measuring change in disease state (Revicki and Ehreth 1997). The information gathered by using these measures can be too generalised, especially when using the resulting QoL score to influence patient care or choose a specific drug treatment. Temple et al (2009) argue that a disease-specific instrument can provide better insight into the unique issues faced by patients. Some QoL studies may use both a generic and disease specific measure, in order to capture the different patient viewpoints or to compare the results of using each type of measure (Klassen et al. 2000).

The term "quality of life" can also be used in relation to health utilities, often measured using preference based instruments, which allow the patient to express their preference for different health states in relation to cost or number of years of life. Preference based instruments can are often used in cost-analysis studies, or decision-making, for example by pharmaceutical industry or healthcare providers (Tolley 2001). The most widely used preference based instrument is the EQ-5D (1990), which can be used to derive utility values for specific diseases or disease states. Quality adjusted life years (QALYs) can then be calculated to combine quality with quantity of life, and allow for comparison between the benefits of different treatments in terms of length of time living in a certain health state.

Quality of life measurement can be undertaken in a variety of different settings and for different purposes. Quality of life measures have traditionally been used in research and clinical settings to provide information about the needs of the patient. This includes

assessing the results of healthcare interventions, screening for psychosocial problems, monitoring patient progress, determining choice of treatment and prioritising funding (Fitzpatrick et al. 1992). The use of PRO data in clinical trials is becoming more common and quality of life assessments relating to new products can help support marketing campaigns, measure cost-effectiveness of new drugs, and provide information on the effectiveness of medical interventions (Marquis et al. 2006). Measuring improvement in quality of life using both psychometric and preference-based measures, as well as producing safety and efficacy data may distinguish a product from its competitors (Revicki and Ehreth 1997). Quality of life measurement is especially important when evaluating treatments for chronic diseases, where new drugs are used to reduce disease severity or limit disease progression (Revicki and Ehreth 1997), both of which can be proven by using PRO data alongside clinical data. Over the last four years, the Food and Drug Administration (FDA) have recognised the importance of the inclusion of PROs in clinical trials, and produced guidance in 2009 to attempt to standardise their use, and outline recommendations for instrument development (FDA 2009). The European Medicines Agency (EMA) have also produced guidance on the use of HRQoL measures in trials, but their guidance is more general and does not contain specific information regarding instrument development (EMA 2005).

The FDA guidance contains information and recommendations as to the minimum standards required for development and validation of PRO measures. This helps to ensure that PRO measures are developed to the highest standards and are suitable for their purpose, especially when being used as part of the evidence for licensing of a new drug. These standards include reliability, sensitivity to change and validity. Reliability measures how able the instrument is to yield consistent, reproducible results (FDA 2009) with regards to stability of scores over time when no change is expected in the concept being measured. This could include test-retest reliability, which measures the stability of the instrument over a short period of time, or internal consistency, which measures the extent to which items measuring the same concept correlate. For interviewer-administered PRO measures, reliability testing could also include inter-interviewer reliability, or the inter-rater variability to test consistency (EMA 2005). Validity ensures that the instrument is measuring what it is intended to measure. Content validity tests whether the measure is composed of the elements collected during the initial data collection phase (those which are important to the patient, or concept being measured), and whether these elements are proportionally and clearly represented in the measure. Construct validity tests the outcome of the measure against a pre-formed hypothesis, based on existing literature (EMA 2005). The measure should also be able to identify differences in scores over time in individuals or groups who have changed with respect to the concept being measured (sensitivity to change). This is a particularly important attribute when the measure is used to demonstrate the impact of a new treatment or

intervention on the patient's life. Designing a measure with these attributes in mind, and implementing them into the design process helps to ensure that the resulting measure is suitable and accurate for its intended use.

Although the majority of QoL measures are designed for use with patients, there has been increasing interest in the impact of illness on those surrounding the patient, and how the patient's illness impacts their lives (Poston et al. 2003). The following information provides a focused literature review investigating the impact of illness on the lives of family members of patients. This is the first step in understanding a topic where there is a lack of evidence in most medical specialties.

REVIEW OF THE LITERATURE

The aim of this focused literature review was to identify literature examining the impact of illness on family members of patients across a range of medical specialties. The main purpose of the review was to identify any relevant information relating to the generic impact of illness on families, for example including several specialties in one study, or drawing comparisons. Family quality of life studies in individual specialties or diseases were also identified, along with the key impact areas and common themes throughout studies. The literature review also aimed to obtain information about measuring the impact of illness on the family, and to identify any existing QoL measures.

The first exploratory review of the literature revealed that many of the articles reviewed were written several decades ago, as there is not a large volume of modern literature specifically examining impact on the family. Therefore it was decided that carrying out a systematic review would have lead to a substantial reduction in the number of articles to be reviewed, therefore influencing the intended comprehensive nature of the review. Therefore, a focused literature review was carried out, with search terms chosen that were both broad, and more specific to elicit the required information.

Search strategy and selection criteria

The review was based around the PRISMA Statement; a 27-item checklist designed to ensure the transparent and complete reporting of reviews and meta-analysis (Moher et al. 2009). Although the PRISMA Statement is designed to ensure transparent systematic reviews, many of the principles were also relevant to this literature review, for example "Describe the rationale for the review in the context of what is already known" and "Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated" (Moher et al. 2009). Following this guidance helped ensure that the review was comprehensive and replicable.

The main search term used was “family quality of life” and this was also substituted with “impact/effect on family”, and “secondary impact”, and these were combined with “disease” (Table 1.2). The term “partner” was also used. “Impact on family” was combined with several common medical specialties.

Table 1.2: Results of key search terms

Key search term(s) used	Number of references retrieved
Family quality of life	193
Impact on family	2493
Impact of disease on family	12
Effect on family	1349
Effect of disease on family	3
Family + disease	388
Family scale	491
Family measurement	113
Impact on family + surgery	0
Impact on family + medicine	12
Impact on family + dermatology	0
Impact on family + psychiatry	0
Impact on family + respiratory	0
Impact on family + cardiology	0
Impact on family + renal	0
Impact on family + gynaecology	0
Impact on family + paediatrics	0
Impact on family + urology	0
Impact on family + gastroenterology	0
Impact on family + disability	0
Greater patient	2946
Secondary impact	165
Impact on partner	113
Chronic disease + family	38

The OVIDSP Medline was selected as the primary search platform. This included the following resources: Cardiff University Books and Journals, PsycArticles, AMED (Allied and Complementary Medicine), British Nursing Index 1985-present, Embase 1947-present, HMIC (Health Management Information Consortium), ICONDA 1976 to June 2011, Medline In Process, Medline 1947-present, and PsycINFO 1806 to July Week 1 2011. A Google Scholar search was also carried out using the same search terms. Searches were limited to sources published in English. A search of the Compendium of Quality of Life Instruments was also carried out (Salek 1998, 2007). This compendium lists over 150 questionnaires and profiles, and the search was carried out to identify any existing family quality of life measures. Two measures of possible relevance to the impact of disease on family members of patients were identified. The search was carried out in July 2011, and updated in January 2013 to include more recent relevant publications.

Each abstract identified was read to determine the type of study and its relevance. Where appropriate, the full paper was read in detail. To be included in this review, a source had to be an original paper, summary article or review, in English, concerned with the impact of any illness or disability on the family of patients. A total of 1517 abstracts were screened, and 158 were identified for review. No sources were identified when combining "impact on family" with several major medical specialties and only 13 sources were identified using the term "impact of disease on family". In addition, using the term "greater patient" resulted in 2946 articles but on review, these were not relevant to the "Greater Patient concept" pertaining to the secondary impact on the family and partner (Basra and Finlay 2007). The majority of sources reviewed were concerned with family members of patients of one medical specialty or one specific disease, and were often limited to the effect on one particular family member, for example partners. No information was found regarding the more general impact of disease on families of patients over more than one specialty. However, many of the studies revealed similar ways that family members of patients were impacted by disease.

Definition of family

The term "family" is difficult to define. The mid 20th century concept of family, with heterosexual parents and offspring living under the same roof is seldom used, and many authors now consciously use a wider and more open definition of family. The dynamics between family members are constantly evolving, and there is evidence of many diverse family types in the modern western European society. Scott (2006) describes a decline in "family life", with falling marriage rates but increasing divorce and cohabitation, which she associates with the transformation of the role of women in society and increased secularisation. Poston et al (2003) define family as "people who think of themselves as part of the family, whether by blood or marriage or not, and who support and care for each other on a regular basis", and this definition is thought to acknowledge the diverse social arrangements that may constitute a family (Jokinen 2006). In other studies, the terms "family", "informal caregiver", "carer", and "caregiver" are used interchangeably (Pochard et al. 2001; Swanberg 2006). Many authors use their own definitions of family according to their own values and beliefs. The authors must also decide how to take into account the views of individual family members and the family as a whole (Jokinen 2006). Beutler et al. (1989) outline the concept of "family realm", describing unique relationships which tend to be more permanent than friendships, the strong emotional attachments between family members, and the exclusive ethics and dynamics within the family structure. For this review, we have taken a broad view of the term family and accepted each author's interpretation as valid. Where studies refer to caregivers (or carers), it was ensured that this related to family caregivers. A wealth of literature was found relating to carers/caregivers, but this was excluded from the review as this study focuses on the impact of illness on family members, not always those

who identify themselves as carers, but may still be impacted. Rather than repeat previous work relating to carers, this review aims to uncover some of the hidden impacts on family members of patients, whether they care for the patient, or not. As it is not yet known whether the ways that family members are impacted are the same as the ways that carers are impacted, the two were kept separate, unless specified as “family caregivers”. This literature review does not aim to cover the extensive information available regarding patient carers, but focuses on other family members who are often overlooked in family-related studies, particularly when they are not parents of paediatric patients.

The history of family research

Historically, medicine has been largely physician-centred, with physicians withholding diagnostic information from patients, refraining from discussing treatment options with patients, and making clinical decisions without first consulting the patient, and often without patient consent. Since the 1960s, medicine has undergone a rapid “patient centered shift”, where patients perspectives are now taken into account, and the teaching of psychosocial aspects of medicine are now a requirement in medical schools (Laine and Davidoff 1996). During the late 1970s, high profile law cases, such as that of Cruzan (Lo and Steinbrook 1991) encouraged physicians to share clinical decisions with both the patient and the family members, signifying the beginning of a new approach to medicine which included taking into account the secondary aspects surrounding the patients’ lives such as family and social care. Changes in the 1960s and 1970s in education and health, as well as an increased awareness of patients with special needs, helped to emphasise the great importance of the role that the family plays in modern healthcare. Today, family-centred care is supported by several major studies (Kuhlthau et al. 2011; Radwin et al. 2011), and is a topic which is at the forefront of many clinicians’ minds. Family-centred care is an approach to healthcare which emerged as an important concept in the second half of the twentieth century and includes considering the role of families in promoting the health and well-being of their children (2003). The American Academy of Pediatrics (2003) outline the main principles surrounding family-centred care: recognising the vital role that families play in ensuring the health of all family members, and acknowledging that emotional, social and developmental support are integral parts of healthcare. It is thought that following these principals leads to better health outcomes, wiser allocation of resources and greater patient and family satisfaction.

RESULTS

Several major disease-specific studies have been carried out on the impact of disease on families of patients and these were first reviewed in order to understand what is already known about the area. A longitudinal study of the effects of chronic childhood illness on

families of paediatric patients resulted in the development of the Impact on Family Scale (Stein and Riessman 1980). This is a 24-item scale with four quality of life domains which has been revised to a 15- item scale (Williams et al. 2006). On the basis of interviews with 187 family members of children with a disability, Poston et al.(Poston et al. 2003) proposed ten domains representing family quality of life. These formed the basis of the Beach Center Family Quality of Life Scale (Hoffman et al. 2006).

Several key review articles were identified concerning the impact of illness on the quality of life of the partner (Rees et al. 2001), the impact of cancer on the family (Lewis 1986), and the impact of chronic disease in the elderly on the patient's family (Kriegsman et al. 1994). Several studies included control groups (Table 1.3).

Key impact areas

Most chronic diseases have similar effects on patients' partners and other family members such as psychological and emotional functioning, disruption of leisure activities, effect on interpersonal relationships, and financial resources. However there may be some aspects which attain dominance in one particular disease as compared to other diseases. Several common themes were identified from the studies reviewed, providing an understanding of the extent of the impact of disease on family members of patients. Further examples of less common themes mentioned are summarised in Table 1.4.

Table 1.3: Summary of studies that included a control group

Author	Family member group	Control	Summary
(Goldbeck 2006)	Parents of children diagnosed with cancer, diabetes or epilepsy.	Parents of healthy children.	Parents of children diagnosed with cancer, diabetes or epilepsy reported significantly lower quality of life compared with healthy controls. However they were also more satisfied with their family situation compared with healthy controls.
(Lawson et al. 1998)	Families of children with atopic dermatitis.	Families of healthy children.	Families of children with atopic dermatitis have a lower family function level than families of healthy controls.
(Hagedoorn et al. 2000)	Male and female partners of cancer patients.	Healthy couples.	Female cancer patients and female partners of patients perceived more psychological distress and a lower quality of life than women in healthy couples. Psychological distress and quality of life did not differ between male partners of patients and their healthy controls.
(Coyne et al. 2010)	Family members of overactive bladder (OAB) patients.	Family members of healthy individuals.	The OAB-FIM discriminated between OAB and control family members with OAB family members demonstrating significant impact on quality of life.

Table 1.4: Further examples of less common themes identified from the literature

Affecting sleep (Basra and Finlay 2007; Coyne et al. 2010; Davis et al. 2009; Eghlileb et al. 2007; Elliott and Luker 1997; Ferrario et al. 2004; Kornblith et al. 1994; Lawson et al. 1998; Weitzenkamp et al. 1997)
Concerns about medical treatment (Basra and Finlay 2007; Cappelleri et al. 2008; Eghlileb et al. 2007; Ferrario et al. 2004)
Altered food choices (Basra and Finlay 2007; Elliott and Luker 1997; Komulainen 2010)
Using religion, spiritual and cultural beliefs to cope (Basra and Finlay 2007; Brown et al. 2003; Cohen et al. 2006; Koldjeski et al. 2007; McMillan and Mahon 1994; Palma et al. 2012)
Feeling obliged to give care (Boeije et al. 2003)
Concerns about receiving information about the disease and understanding (Bowen et al. 2011; Ferrario et al. 2004; Koldjeski et al. 2007; Weitzenkamp et al. 1997)
Needing support from others (Brown et al. 2003; Majasaari et al. 2005; Mellon 2002)
Limited freedom (Davis et al. 2009)
Worrying about death of the patient (Ferrario et al. 2004; Osse et al. 2006; Weitzenkamp et al. 1997)

Psychological impact

Family members suffer greatly from the emotional effects of living with, and caring for, a relative with a disease, with the impact of some diseases being felt by every member of the family (Bowen et al. 2011). Emotional impact resulting from having an unwell relative was the most common topic discussed in the literature. The psychological distress felt by family members often results from their feelings of helplessness and lack of control (Basra and Finlay 2007; Sallfors and Hallberg 2003). Many different emotions are mentioned by family members: guilt, anger, worry, upset, frustration, embarrassment, despair, loss and relief. Each emotion affects family members in different ways and to different extents, often depending on the disease severity of the patient (Balkrishnan et al. 2003; Berge et al. 2006) and the period of time that has passed since the diagnosis (Koldjeski et al. 2007). The coping strategies of family members have also been reported (Kempainen, 2007; Wade, 2001; Bush, 1997; Barnett, 2012). In one study, Gauthier et al. (2007) found that the emotional burden of caregivers of patients with amyotrophic lateral sclerosis significantly increased over a period of nine months of the illness, whereas the patient's psychological wellbeing remained relatively stable. In one paediatric study, it was found that children with a family member suffering from multiple sclerosis were three times more likely than the general community to develop a psychological illness (De Judicibus and McCabe 2004).

Hagedoorn et al. (2000) found that female partners of cancer patients had higher psychological distress than male partners. However, no significant difference was seen between genders when measuring overall quality of life of relatives. Several other studies have found gender differences in responses to caregiving (Boeije and Van Doorne-Huiskes 2003; Bristol et al. 1988; Northouse et al. 2000; Pitceathly and Maguire 2003), although Walsh et al. (1999) found no difference in the well-being of the partners of rheumatoid

arthritis patients, based on the gender of the patient. As well as looking at differences in gender, authors have also used the relationship to the patient as a variable; it is not just the parents and partners who are affected emotionally by a relative's disease (O'Brien et al. 2009). For example, Fisman et al. (2000) reported that siblings of children with pervasive developmental disorder suffered from "significant adjustment problems" compared to a control group. In addition, Hardy et al. (Hardy et al. 2008) found no significant difference in the post-traumatic stress symptoms and psychological functioning of parents of adult cancer sufferers and parents of paediatric patients, and that parents of adult patients may remain "psychologically vulnerable" for many years after treatment ends. This is also reflected by Sabo et al. (2013) who found that spouses of patients undergoing haematopoietic stem cell transplantation were still at risk of depression one year after the transplant.

Several studies have focused on the effect on the health of the family member. Family members of female HIV patients with poor health reported feeling a greater burden and family members who reported less family cohesion reported higher depressive mood and greater burden (Demi et al. 1997). Looking after an unwell patient can also cause increased feelings of anxiety for the family member regarding their own health (Osse et al. 2006), especially in cancer, where family members worry about the possibilities of cancer being genetic, or the possibility of recurrence (Mellon 2002). Kornblith et al. (1994) found that spouses of patients with prostate cancer experienced a greater psychological distress than the patients themselves, and this finding is echoed in several similar studies (Rees et al. 2001; Weitzenkamp et al. 1997). Conversely, Huygen et al. (1992) found no difference between the morbidity of spouses of patients diagnosed with a serious disease and a group of matched controls.

Pochard et al. (2001) questioned the ability of family members to make decisions about the care of relatives in intensive care, as they demonstrated that more than two thirds of family members visiting patients suffered from anxiety or depression. This study included "all individuals who visited the patient in the ICU, regardless of their relationship to the patient", so the effects on the quality of life specifically of the family are unclear. Werner at al. (2009) investigated the effect of an out-of-home residential placement on the QoL of families of patients with intellectual disability, and found that many of the common negative emotions felt by families of patients were reduced or eliminated after the placement, including anger, frustration, and resentment.

Financial impact

One of the greatest burdens on family members of patients is the financial cost to the family (Clarke et al. 2009; Martinez-Martin et al. 2012). This can include treatment costs, transport to appointments, the cost of hiring a carer, and adapting their home environment to

accommodate their relative's needs. In a Canadian study, Brown et al (2003) asked families how much they spent on average in a month on care, support, or equipment for their family member with an intellectual disability. On average, these families spent C$624 per month, and many described not having any money left at the end of the month. In a similar USA study, the financial impact on families caring for patients with dementia varied from US$3630 to US$17700 depending on the severity of the patient's dementia (Langa et al. 2001).

The financial strains felt by family members of patients often lead to stress and worry, adding to the already great emotional impact they suffer from as a result of caring for their relative. Family members of dermatology patients increase their working hours in order to support their family financially, and many turn to state benefits to cover the extra costs to them and their family which can often lead to the family having to compromise on the needs of other family members, or alter their lifestyle significantly by opting for cheaper living choices (Basra and Finlay 2007). Davis et al. (2009) interviewed parents of children with cerebral palsy to determine how their quality of life has been affected by their child's illness. The authors found that providing even the basic necessities when caring for a child with cerebral palsy put financial pressure on the parents, and that accessing funding was also challenging, which again increased stress and emotional effects. The difficulties involved in accessing funding are greater in low income families, who often receive minimal support and face greater problems with social functioning and relationships (Lapidus and Kerr 2001). Walton-Moss et al. (2005) interviewed family members with a variety of relationships to patients with mental illness and categorised the families as "hanging on", "being stable", or "doing well" as a result of the interviews. The families who were classed as "hanging on" in the study all shared common problems relating to limited financial resources and limited social support.

Impact on family relationships

Family members of patients experience a negative effect on their family relationships, whether between the relative and the patient, or between other members of the family as a result of the patient's illness. Poor satisfaction with family relationships does not bode well for chronic disease management regardless of the disease and often family members find relationships difficult as they do not know how to emotionally support each other (Lewis 1990). Bowen et al. (2011) interviewed 25 family members of patients with multiple sclerosis. Generally, the families reported negative effects on their relationships with each other resulting in arguments, tension, and a lack of understanding of each other's feelings. In particular, relatives struggle to deal with patients who have altered significantly in terms of beliefs, outlook, and behaviour, as a consequence of their disease. There was little time for relationships between other members of the family, and family members often felt the strain of having to spend a lot of time with the patient, with many reporting this as a reason for their

relationship being strained (DesRosier et al. 1992). Many patient-related quality of life studies mention the negative effects on family relationships and if the unwell member is affected, it is likely that the other family members will feel the negative changes in the family relationships too. For example, Golics et al. (2009) found that 38% of adolescents with dermatological conditions felt that their family relationships had been affected as a result of their condition.

Partners of patients often experience a negative effect on their sex lives as a result of the patient's disease. One relative of a patient in a psoriasis study stated that "my wife feels embarrassed when she undresses with me in the bedroom" (Eghlileb et al. 2007). This can lead to friction between couples, and in some cases can lead to the breakdown of relationships, or partners seeking sexual encounters outside the relationship (Basra and Finlay 2007; Elliott and Luker 1997). In a study of parents of children with chronic health conditions, the decrease in the mothers' marital satisfaction reported in the study was influenced by their perceptions of the impact of their child's condition (Berge et al. 2006). Partners of stroke patients described having to "find a new marital path" after the stroke; describing how they had to rebuild and alter their relationship to account for the impact of the stroke and their new carer role in the relationship (Kitzmuller et al. 2012).

However, in some families relationships can grow stronger (Kim et al. 2007), as the family members work together to help each other and become more closely knit. Brown et al. (2003) used their measure, The Family Quality of Life Survey, to assess the impact of having a child with an intellectual disability on parents. They found that the majority (21 out of 34 interviewed) of families were taking the initiative to maintain good family relations, and engaging in family activities to encourage this. An increase in family closeness was also found by Mellon (2002). One husband of a cancer survivor said *"I look at life differently after that. I feel much closer to her"*. In one study looking at family members of patients with lung cancer, 40% of family members reported a closer relationship with the patient as a result of providing care to them (Mosher et al. 2013).

Education and work

Living with, or caring for, a relative with a disease can have a large impact on the education and careers of family members. This could include disruption of school work in siblings or children of the patients, or the employment of adults being affected by their relative's disease, and the burden of care which is placed upon them. Some families studied by Brown et al. (2003) felt that some of their family members would not be able to attend work or school in the near future. One family member is quoted: *"The unpredictable natures of our children's health and lives does not often fit with a typical, progressive work profile*". In eight of the 34 families studied, one or both parents had given up an education or career to care for their child with a disability. Basra and Finlay (2007) found that 40% of family members of

dermatology patients felt that their employment was affected by their relative's skin condition. This was due to a variety of reasons including needing to look after the patient, taking time off work for hospital appointments, and emotional effects preventing individuals from going to work. Looking after a patient with cancer can also have a huge impact on a family member's work on a day-to-day basis, as discussed in an interview-based study by Swanberg (2006) Twenty seven of the 30 caregivers interviewed were family members of patients, and the study found that they were reporting late for work, missing work, spending time at work talking on the telephone to their relative and, as in the previous studies, some left work due to their carer responsibilities.

Leisure time

An important part of family QoL is family members being able to participate in the hobbies they enjoy (Poston et al. 2003). The majority of families in a large, international Canadian study reported barriers that prevented them from taking advantage of leisure opportunities (Brown et al. 2003). The barriers reported linked into other domains of Family Quality of Life, including lack of time due to the responsibilities of care, limited finance, and lack of support available. However, encouragingly, it has been shown that when family members do take the initiative to plan leisure activities, they usually work out positively, despite the restrictions due to the relative's illness, and families show high satisfaction with this achievement (Brown et al. 2003).

Family members also find difficulty in taking family holidays for a variety of reasons, often depending on the disease state of their relative. Problems with finding suitable accommodation with wheelchair access can make holiday planning "awkward" (Davis et al. 2009). Relatives of patients with skin diseases in one study described limitations of holiday planning, for example not wanting to swim together at the beach or their relative having to wear certain types of clothes (Eghlileb et al. 2007). Similarly, parents of children with food allergies described the restrictions on their holiday plans because of their child's allergy (Komulainen 2010). These are burdens which could also affect family members of patients with other diseases.

An increase in housework load has also been reported as a result of living with an unwell relative, particularly in those with skin disease where more frequent washing of bed linen and more cleaning is required (Basra and Finlay 2007). This impacts on the day-to-day activities of the relative, potentially leading to emotional problems including stress and worry, and takes up time which could be used to socialise or spend time with other members of the family.

Social impact

A large number of the existing studies into the burden of disease on family members note the drastic effect that caring for a patient has on their social lives (Basra and Finlay 2007; Davis et al. 2009; Eghlileb et al. 2007; Poston et al. 2003). Notably, mothers caring for disabled children felt that their lives were so different from their friends and felt that they could only contribute to depressing conversations, and therefore lost friends as a result (Davis et al. 2009). Other family members described friends "drifting away", as they did not understand the family situation (Bowen et al. 2011).

In one study, a large number of individuals with a relative suffering from a skin disease complained of social disruption (Eghlileb et al. 2007). Many of the problems they face could be applicable to a number of diseases, for example feeling embarrassed by their relative's condition in public places, but some are more psoriasis specific, for example feeling embarrassed about their relative's skin flakes. Conditions which result in visible signs of disease (for example basal cell carcinoma on the face or chronic obstructive pulmonary disease requiring oxygen therapy) may have a greater effect on the social lives of patients and their relatives, for fear of strangers' reactions to their visible condition. This is explored in relation to dermatological conditions by Basra and Finlay (2007). Hunfeld et al. (2001) found that mothers of adolescent patients suffering from severe chronic pain reported more restrictions in their social life than mothers of children with less severe chronic pain and the authors suggest that this could be directly related to the illness.

As well as concern about their own social lives, family members also worry about their unwell relative being socially accepted, especially in the case of a parent caring for a child, where many report that their child's acceptance is a "high priority" for them (Poston et al. 2003). Parents also report feeling upset about their child being bullied at school due to their illness (Basra and Finlay 2007).

Instruments to measure family quality of life

Several studies have lead to the development of instruments designed to measure the impact of disease on families of patients. However, these instruments are mostly disease or specialty specific, and can therefore only be used to assess the quality of life of the family of a particular group of patients.

The Family Dermatology Life Quality Index is a ten-item questionnaire designed to measure the quality of life of family members of dermatology patients (Basra et al. 2007). The questionnaire, which has undergone full psychometric evaluation (Basra et al. 2008), contains items such as "Over the last month how much emotional distress have you experienced due to your relative/partner's skin disease (e.g. worry, depression,

embarrassment, frustration)?". The Impact of Pediatric Epilepsy Scale (Breau et al. 2008), is designed to measure the impact of childhood epilepsy on the patient and their family. Using this measure, Breau et al. (2008) found that the severity of seizures correlated directly with the quality of life of the patient and their family. This correlation with disease severity is reflected in a study by Ben Gashir et al. (2002) who found that disease severity negatively influences the quality of life of families of children with atopic dermatitis using the Family Dermatitis Impact Questionnaire (Lawson et al. 1998). Further examples of disease-specific measures include the Psoriasis Family Index (Eghlileb et al. 2009), the Overactive Bladder Family Impact Measure (Coyne et al. 2010), and the Quality of Life in Life Threatening Illness - Family Carer Version (Cohen et al. 2006), which is designed to be used with carers of palliative oncology patients with the aim to develop and deliver the most effective services to these carers. In fact, many of the existing instruments to measure family quality of life have been designed for use in families of patients with cancer (McMillan and Mahon 1994; Weitzner and McMillan 1999) and mental health illness (Baronet 1999).

The Impact-on-Family Scale, as mentioned above, measures the impact of childhood chronic illness on the family (Stein and Riessman 1980). Similarly, the extensively tested Beach Centre Family Quality of Life scale (Hoffman et al. 2006), was designed for and evaluated in families of children with disabilities. The questions in this measure fall under five main categories: family interaction, parenting, emotional well-being, physical/emotional well-being, and disability-related support. The Family Quality of Life Survey is another example of a measure designed for use in family members of patients with or without an intellectual disability (Issacs et al. 2007). This survey takes on average an hour to complete and is designed to assess the aspects of family quality of life which are important to a family and whether these aspects are being adhered to.

The Caregiver Quality of Life Index (McMillan 1996), is a four-item visual analogue scale measure designed to assess the quality of life of primary caregivers of hospice patients receiving homecare, but not specifically family members. The Family Strain Questionnaire (Ferrario et al. 2004) was also identified, but again the population group was defined as "principal caregivers" and not family members. Furthermore, the measure assesses the burdens or problems and needs of caregivers of patients and not the overall quality of life. These generic measures were examined for evidence of psychometric testing, and most were found to demonstrate good evidence of validity and reliability testing. The properties of these generic measures are summarised in Table 1.4, including the areas covered by each measure. No measure was found which can be used to assess and compare the impact of any disease on family members of patients.

Table 1.5: The characteristics of four family quality of life measures

Name of measure (key reference)	Population	No. items	Coverage	Time to complete	Origin	Frame of reference	Scale	Other
Impact-on-Family Scale (Stein and Riessman 1980).	Parents of children with chronic illness.	24	Four dimensions (factors): Financial, Social, Personal strain and Mastery.	10 min	Family member interviews.	"at the present time".	Likert	A revised 15-item version was created in 2003 which should be used in replacement of the earlier instrument.(Stein and Jessop 2003).
Beach Center Family Quality of Life Scale (Summers et al. 2005).	Family members of children with a disability.	25	Five domains: Family interaction, Parenting, Emotional well-being, Physical/material well-being and Disability-related supports.	15 min	Family member interviews and focus groups.	Current e.g. "how satisfied am I that...?"	Likert	The scale was later tested in families of non-disabled children and proved to have psychometric validity (Zuna et al. 2009).
Family Quality of Life Survey (Issacs et al. 2007).	Main caregiver: intellectual or developmental disabilities.	49	9 areas of family life: health, financial, family relationships, support from others and services, influence of values, careers, leisure&recreation, community integration.	60 min	Previous research, expert opinion.	Questions relate to the present and the future.	5-point scales and a variety of response categories .	The measure was updated in 2006 and a version to include families without disability has also been produced.
Family Strain Questionnaire (Ferrario et al. 2004).	Caregivers of patients with any disease.	44	Five factors: emotional burden, problems in social involvement, need for knowledge about the disease, satisfaction with family relationships, and thoughts about death.	20 min	Developed from a stress-appraisal-coping model (Lazarus and Folkman 1984).	Present. Phrases such as "during this period" and "at this moment" are used.	Dichotomous	In 2010, the FSQ-Short Form was developed, which contains 30 items and can be completed in 5 minutes.(Vidotto et al. 2010).

CONCLUSION

There is a wide range of information in the literature about the impact of disease on family members of patients. However, most studies are focused on a specific group of patients, or a specific family member of a patient. It is unknown whether the results of disease-specific studies are applicable to a more general population, or whether family members are affected in similar ways across every medical specialty. For example, the family quality of life domains suggested by Poston et al. (2003) result from a study with family members of disabled children. Kazak (2002) discusses the lack of "reliable and valid family outcome measures" and the negative effects of family outcome studies of one disease or clinical area, including the lack of communication between medical specialties and obscuring commonalities across different disease areas. Although many studies conclude that a more family-centred approach to care (Bowen et al. 2011; Brown et al. 2007; Fisher 2006; Koldjeski et al. 2007), and further education of professionals is needed (Jokinen 2006), no generic measure exists to assess the impact of a variety of diseases on family members of patients.

The impact of disease on families of patients is often unrecognised. Comparing and contrasting information from families of patients with a variety of diseases could uncover new domains of quality of life unique to family members, which, with appropriate support in place, could result in a higher standard of patient and family care. Taking into account the quality of life of families as well as patients can offer the clinician a unique insight into issues such as family relationships and the effect of treatment decisions on the patients' close social group of partner and family, the "Greater Patient" (Basra and Finlay 2007).

To understand the needs of family members of patients and be able to offer appropriate support, we first need to understand the ways in which their lives are affected. This review has highlighted the need for a multi-specialty study investigating the issues faced by families of patients, how these differ between diseases, and exploration of the common themes and ideas.

AIMS OF THE STUDY

- To investigate the impact of a wide range of diseases on family members.
- To develop and validate a quality of life measure for use by individual members of a patient's family to assess the impact on the family member's life of having a person in the family with a disease.

OBJECTIVES

- To identify and understand the research about family quality of life which already exists.
- To identify key dimensions of quality of life for families of patients and design a model or diagram to represent family quality of life.
- To compare and contrast the impact of disease on family members of patients across different clinical specialties.
- To investigate whether the impact of illness on family members varies by age, disease duration or gender.
- To investigate the psychometric properties of the new measure.
- To outline possible uses for the new measure.
- To record and evaluate feedback from family members of patients regarding the interviews, the study and the new measure.
- To further understand the Greater Patient Concept (Basra and Finlay 2007) and the role of the family in the life of a patient with chronic disease.
- To make recommendations as to the specific areas and possible ways in which family members of patients can be supported.

CHAPTER 2

Study Rationale and Methodological Framework

STUDY RATIONALE

The impact of illness on family members of patients has already been explored in many areas of medicine, with the majority of work focused around physical and mental disability, dermatology and oncology (Basra and Finlay 2007; Cohen et al. 2006; Poston et al. 2003). Illness has been shown to have a major impact on families of patients across all of these areas. In fact, the quality of life of family members correlates with that of the patient (Basra and Finlay 2007), and in some cases the quality of life of the family member can be worse than that of the patient themselves (das Chagas Medeiros et al. 2000; Rees et al. 2001; Weitzenkamp et al. 1997). Much of the previous research into family quality of life focuses on those members of the family who are primary caregivers, or use the terms "family" and "caregiver" interchangeably. There is still a lack of understanding of the impact of illness on those members of the family who are not necessarily the primary carer, but are still part of the family and may still be affected.

Disease-specific and specialty-specific questionnaires have been designed to measure the impact of illness on family members of patients. As these measures have been designed to be used with family members of specific patient groups, they cannot be used in a general population, or to compare and contrast the impact of different illnesses.

Generic health-related quality of life instruments exist for patients, such as the SF-36 and the WHOQOL (Brazier et al. 1992; Skevington et al. 2004). These measures are designed to be used to assess the impact of illness on patients, regardless of their illness, and are currently routinely used in clinical practice and policy making, highlighting the importance of considering the effect on the patient's quality of life when making treatment decisions. From previous disease-specific studies, it can be clearly seen that the impact of illness on family members is also widespread and of high importance, but there is currently no way of measuring it in a generic way and being able to compare and contrast between illnesses and specialties. Furthermore, as the subject of family quality of life is a relatively new concept, it is an area which has not been explored in many disease areas, or been highlighted to clinicians.

Exploring the ways in which disease impacts family members over a wide range of conditions will allow comparisons to be drawn between different disease areas. Areas of family members' lives which are affected will be identified, and will give a more clear idea of areas where more support is required. The impact of illness on those members of the family who are not necessarily the primary caregiver will also be explored, where in previous studies they have often been overlooked. The extent of the impact will also be of interest, and the results of a qualitative exploration will form a solid basis for the production of a generic family quality of life measure.

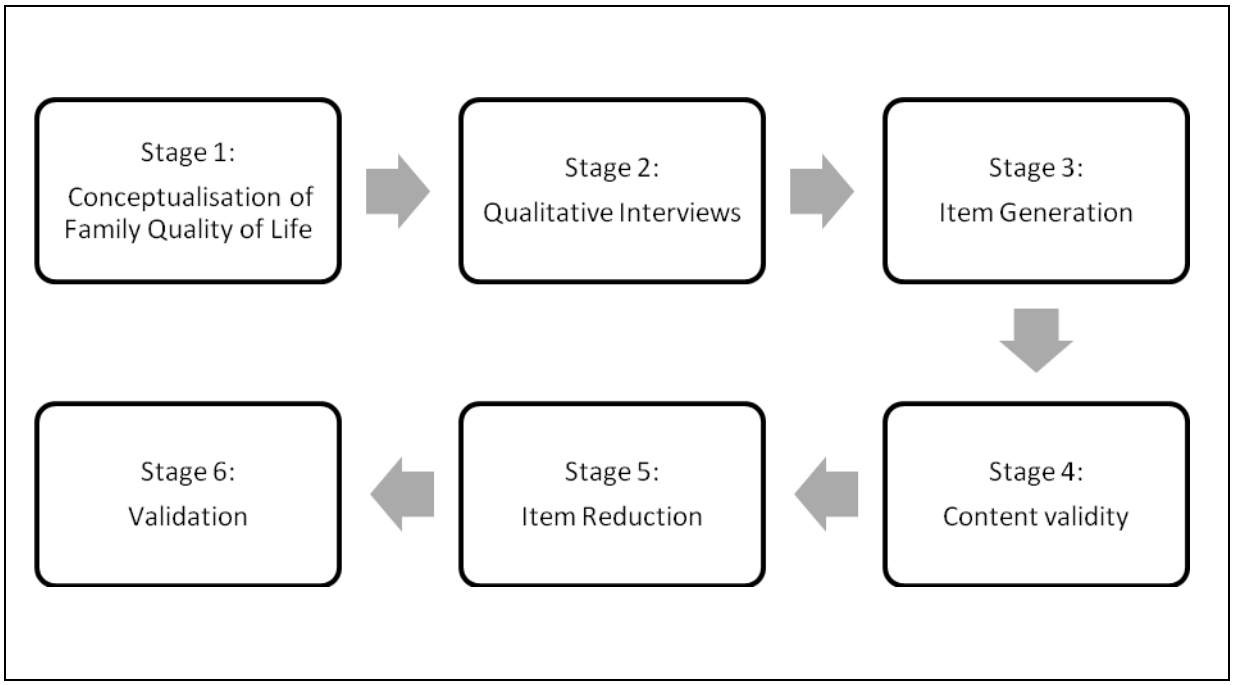
A family quality of life measure will allow the generic impact of illness on family members of patients to be quantified for the first time. In the same way that generic patient QoL measures are used in clinical practice and health research, the impact of illness on families will be able to be used as an additional endpoint, or outcome measure. The impact of treatment decisions on family members of patients will be able to be measured, and the new measure will increase the awareness of the family impact of disease in a clinical setting.

This thesis describes the qualitative investigation into the impact of illness on family members of patients, and the development of a generic family quality of life measure.

STUDY DESIGN

This is a mixed methods study, based around six stages. Figure 2.1 shows the main stages of the study and the processes involved:

Figure 2.1: The six main stages of the study



STUDY POPULATION

The participants in this study were family members or partners of patients attending outpatient clinics and inpatients at hospitals in the Cardiff and Vale University Health Board (University Hospital of Llandough, University Hospital of Wales, Gabalfa Clinic), and Velindre NHS Trust over a range of medical specialties. In order not to restrict the study to the selection biases inherent in studies entirely based in secondary care, family members of patients in primary care were also recruited.

Selecting specialties

As one aim of this study was to identify the impacts of illness on families from a generic point of view, a wide variety of specialties were selected to be included in the study. The aim of this selection process was to widen the recruitment of family members to maximise the number, type, severity and length of diseases considered. The study team obtained from the Health Board website a list of medical and surgical specialties based at the Cardiff and Vale University Health Board and selected 25 of these specialties (Table 2.1). These included medical and surgical specialties, dental surgery (including trauma), mental health (including a community outreach team) and several paediatric specialties. It was felt that excluding family members of paediatric patients would leave the study incomplete, especially given the huge emphasis on the impact of illness on families of children in previous literature (Opperman and Alant 2003; Stein and Riessman 1980; Wennick et al. 2009). General Practice was added as a 26th specialty, to cover the views of relatives of patients being treated in a primary care setting (Table 2.1).

Table 2.1: The specialties included in the study

Cardiology	Infectious Diseases
Care of the Elderly	Mental Health
Chronic Pain	Neurology
Colorectal Surgery	Oncology
Dental Surgery	Ophthalmology
Dermatology & Paediatric Dermatology	Orthopaedics & Paediatric Orthopaedics
Ear, Nose & Throat	Paediatric Endocrinology
Endocrinology & Diabetes	Post-stroke
Gastroenterology	Renal & Renal transplant
General Practice	Respiratory
Genetics	Rheumatology
Gynaecology	Urology
Haematology	Wound Healing

Contact was made with one consultant clinician from each of the 26 specialties identified and the subsequent recruitment process was discussed with each consultant. Each consultant was asked to identify at least five patients who best represented the conditions they saw under their specialty area. At each stage of the study, the consultant first approached the patient and their family member(s) to gain permission for the investigator to then approach them and introduce them to the study.

Inclusion and exclusion criteria

Inclusion criteria for family members

- Age 16 years and above

- Able to understand and read English
- An immediate family member, or partner living with, or caring for, a patient diagnosed with one or more medical conditions under one of the selected specialties (Table 2.1)
- Able to give written informed consent

Inclusion criteria for patients

- Able to understand and read English (if over 16 years old)
- Diagnosed with one or more medical conditions under the selected specialties (Table 2.1)
- Able to give written informed consent (if over 16 years old)
- Attending clinic with a family member, or currently a hospital inpatient.

Exclusion criteria for family members

- Age under 16 years
- Unable to understand and read English
- Not considered by the patient as a family member
- Unable to give written informed consent
- Having a severe handicap or disability which prevents them participating in an interview or completing a questionnaire.

Exclusion criteria for patients

- Unable to understand and read English (unless under 16 years old)
- Unable to give written informed consent (unless under 16 years old)

The definition of family

As explored in Chapter 1, many authors interpret the definition of family in many different ways, and the definition of family is constantly evolving with society. Before this study began, it was important to have a clear definition of our interpretation of the term "family". We agreed with the definition of family given by Poston et al.: *"people who think of themselves as part of the family, whether by blood or marriage or not, and who support and care for each other on a regular basis"* (Poston et al. 2003). This meant that we included unmarried partners of

patients, children and parents-in-law, and cousins. As long as the family member considered themselves as “family”, and offered support to the patient on a regular basis, they could be recruited in the study. This wide definition reflects a modern society, where families are not always made up of two heterosexual parents and their offspring. It also allows different relationships within the family to be explored, and does not restrict those more distant family members who play an active role in the patient’s life. This definition of family also reflects the complexity of family life in Britain today. Furthermore, unlike many of the existing studies which refer to family carers or caregivers, the family members recruited in this study are not necessarily those who provide care to the patient, but their lives may still be impacted by the patient’s illness.

ETHICAL CONSIDERATIONS

During the design of the study, particular attention was paid to the ethical considerations surrounding all stages of the study. These issues of ethical concern are addressed below.

Ethical approval was sought from the South East Wales Research Ethics Committee, and was granted on 21st May 2010 (Appendix A). Local approval was also granted from Cardiff and Vale University Health Board Research and Development Department (Appendix B) and Velindre NHS Trust Research and Development Department (Appendix C). The study was also registered with the Data Protection Officer for Cardiff and Vale University Health Board. An expert in the area of quality of life research was asked to review the project and gave a favourable opinion (Appendix D).

Written informed consent was taken from both the patient and the family member prior to data collection. The consent taken from the patient allowed the family member to talk about, or give information about, the impact of the patient’s medical condition on their life. Separate information leaflets and consent forms were designed for family members and patients, and also for those patients under 16 years of age (Appendix F-I). The study was fully explained to both the patient and the family member and they were given a chance to ask any questions they may have had before taking consent.

Confidentiality was of paramount importance during this study, as many sensitive issues were discussed during the interviews. Patients and family members were given a code number to identify them during the study. Only the study team had access to the codes and data arising from the study, and these have been kept securely within the university building. Interviews and questionnaire completion were carried out in private rooms and participants were assured about the privacy and confidentiality of any information they provided. This was also emphasised in the information leaflets, along with examples of how the information taken from interviews would be quoted anonymously in publications.

The interviewer undertook formal interview training and has previous interviewing experience. Care was taken to use language comprehensible to all study participants. The interviewer was aware that sensitive questions were being asked, and if the family member was too uncomfortable or emotional during the interview or completion of the questionnaire (as determined by asking the participant), the interview would be terminated and information discarded if the family member or patient wished. In that unlikely circumstance, the family member would be offered a further opportunity to be given advice concerning the issues raised.

For the convenience of the patient and the family member, the venue and time of the interview or questionnaire completion was decided by them. The family member had the option to request the interview or questionnaire completion whilst the patient was being seen by the clinician in the clinic.

The ethics of recruitment were also considered during the study design. Patients and family members were approached in clinics or inpatient settings. They were then introduced to the study by the clinician, given the information leaflet to read and given the opportunity to ask questions. They were then given 48 hours (or longer if they wished) to decide whether to take part in the study. If they did decide to take part, the investigator was then available to conduct the interview or administration of the questionnaire in the clinic or on the ward.

PROCEDURE

The study was conducted between August 2010 and August 2012, in six main stages (Figure 2.1).

Stage 1: Conceptualisation of Family Quality of Life

Prior to the design of the study, the concept of family quality of life was examined in depth. This involved the study of available literature on the subject, in particular identifying the gaps in the current knowledge of the area, in order to ensure that this study was not repeating previous work and that the work focused on previously unexplored areas. The related literature was collated into a literature review, the results of which are given in Chapter 1. For each original report, the design and methods were examined, so that the most appropriate and effective methods were written into the protocol for this study. The limitations of each reviewed study were also noted and considered during the design of this study. The available literature on family quality of life also provided information about the themes which had arisen from these studies, which might therefore be of importance in this study. Themes and ideas which arose from previous related studies were used to form the basis for an interview structure, and provided ideas of possible questions to ask family members of patients.

Exploring the concept of family quality of life allowed an advance understanding of the possible themes which may have arisen from the interviews with family members, and how these themes compare to previous studies. A full understanding of the background of the study area was therefore of great importance during the study design.

Stage 2: Qualitative Interviews

It was important to select the most appropriate method for collecting the qualitative data regarding how family members are affected by illness. Several different methods were considered for the qualitative data collection:

Focus groups

This is a type of group interview which can be a quick and efficient way to collect qualitative data. The advantages of focus groups include the benefits of the interactions between members, commenting on each other's ideas and points of view (Kitzinger 1994), with the discussion reminding participants of additional issues. Other advantages include not discriminating against those who cannot read and write and encouraging those who are reluctant to be interviewed on their own (Kitzinger 1995). However, focus groups were considered inappropriate for this study as it was felt that the information being discussed could potentially be very personal to the participants and many may not want to talk about it in front of others, especially if they are not used to talking to anyone about how the patient's illness has affected them. This view was informed by previous recent experience of the research team in gathering qualitative information concerning Major Life Changing Decisions (Bhatti et al. 2011), where there was great difficulty in recruitment for focus groups. It is also important that the views of family members from a variety of specialties are collected, and although it is the role of the moderator to ensure contribution from all members during a focus group meeting, the discussion could be dominated by those family members who have experienced the greatest impact. It is considered extremely important that the participants feel comfortable enough to open up and discuss sensitive issues and it was felt that this may not be achieved during a focus group.

Questionnaires

Questionnaires with spaces for family members to write their answers were also considered as a method of data collection. This method needs relatively few resources and requires little effort to recruit a large number of participants. However, there is the high possibility of a low response rate if the questionnaires are posted (Zelnio 1980), and there is often a lack of sensitivity as the respondent cannot elaborate on answers as easily as in an interview or focus group. As family quality of life over a number of specialties is a new concept which has not been explored for the purpose of development of a measurement method before,

questionnaires were not chosen as the method for data collection, as a rigid, structured set of questions would be required which would be difficult to predict at this stage of the research. Using questionnaires would potentially limit the richness of the data and not allow the investigator to invite the subject to expand upon answers given.

Individual interviews

Interviews with family members were the third potential method of data collection considered. Interviews have the advantage of being able to follow up the information the interviewee gives with further probing, or clarify anything ambiguous (Kvale and Brinkmann 2009). Although individual interviews can be time consuming and potentially induce bias into the data, they allow for in-depth data collection. Individual interviews were chosen as the method of data collection for this study as the method allows flexibility and the topics covered with family members have the potential to be personal or sensitive, therefore a one on one method of data collection is preferable. Steps were taken to reduce interviewer bias and the use of individual interviews worked well within the time constraints of the project.

Qualitative interviews were carried out with family members of patients suffering from a variety of diseases over 26 specialties. Family members were asked about the ways their lives had been affected by having an unwell relative. Interviews were semi-structured, and participants were encouraged to give examples where possible. Effort was made to recruit a spread of family members (e.g. siblings, parents, spouses), in collaboration with the clinician from each specialty. The interviews were audio recorded and transcribed verbatim. Throughout the interviews, and as new themes emerged, potential uses of the new measure were considered and ideas of how family members of patients can be supported were explored.

Stage 3: Item Generation

The interview transcripts were analysed using the qualitative data analysis software NVivo©. This coding software allows the highlighting and sorting of the transcripts into common themes, or codes. Hierarchy of themes can also be established in the NVivo software, and relationships between the different themes can be identified. Examples from transcripts containing similar themes are grouped and easily retrieved in order to calculate the prevalence of each theme and identify specific examples to illustrate each theme. As well as using the NVivo© software, the interview transcripts were also re-coded by hand using a numbering system to ensure reliability between the paper and electronic methods. The process of coding the data followed that outlined by Braun and Clarke (2006), and included familiarising oneself with the data, generating initial codes, searching for themes, reviewing themes, and then defining and naming themes. A flexible approach to coding was used, and

both the coding process and themes were discussed in detail with the study team at regular stages. During coding, the transcripts are often examined for saturation, the point at which no new themes emerge from subsequent interviews, which can be used to determine adequate sample size in qualitative research (Guest et al. 2006). As it is important that this study covers family members of patients with a variety of illnesses, all 26 specialties must be included at interview stage. Therefore, sample size was not calculated as the interviews were carried out, but was calculated once the desired number of interviews from each specialty had been competed (>5), to ensure that saturation point had been reached. If saturation point had not been reached at this stage, further interviews would be carried out. This is explained further in Chapter 3.

The themes generated were then used to form the items of the preliminary version of a generic family quality of life questionnaire. The questions were worded in a simple and comprehensive way, and each question was designed to measure a different concept. The questionnaire was designed so that the answer to each question would be scored to allow quantification of the overall effect their relative's illness has on their quality of life. The questionnaire was developed so that it could be applicable to, and understood by, all members of a patient's family over 16 years of age. The design of the questionnaire was such that the investigator, and after the questionnaire is launched the clinician, would be able to determine the area(s) of the participant's life which are most greatly affected.

Stage 4: Content validity

Content validity ensures that the instrument measures the intended concept and that it is understood and relevant to the intended population (FDA 2009). It is often the first, and arguably the most important step in the validation of an instrument, as it underpins the theory of tests such as reliability and construct validity (Rubio et al. 2003). Members of the intended instrument population are asked to provide feedback regarding the measure, both in terms of individual items, and the content covered in the measure as a whole. This includes the instructions, layout and response options. This feedback is then used to justify that the measure covers the intended concept, and identifies areas where changes could be made. Additionally, clinicians are asked to give their feedback on the measure, and the relevance of items to the family members of the patients they treat, who are often involved in their care.

The preliminary version of the instrument was evaluated by both an expert panel and by a family member panel who were recruited to test the content validity of the questionnaire items, which is considered important during instrument development (Streiner and Norman 2008). The expert panel consisted of academic experts, consultant clinicians and nurses from a variety of specialties, and they rated each item in the scale for its language clarity, completeness, scaling and relevance. They also reviewed the planned layout of the measure

and the instructions given. Agreement amongst the panel members was measured using interclass correlation and the panels suggested several changes to the measure which were then implemented to form the developmental version of the instrument.

Stage 5: Item Reduction

As in Stage 2, patients and family members were recruited over the 26 specialties with guidance from a consultant clinician. The aim of this stage of the study was to begin to validate the newly developed family quality of life instrument using a combination of item response theory and factor analysis. Item response theory was used to identify mis-fitting items, items which showed differential item functioning by age and gender, and items which were clustering and potentially measuring the same concept. Items with low item-total correlation (<0.2) were dropped (Streiner and Norman 2008). Factor analysis was then applied to allocate domains to the instrument, and also to confirm any mis-fitting items. The traditional method of using factor analysis alone has been criticised for assuming that each item contributes equally to the final score, assuming a standard error of measurement across the scale and posing problems with equating tests (Streiner and Norman 2008). Rasch analysis is now the preferred technique for item reduction, and it has been said that "only Rasch analysis constructs the kind of objective linear variables that social scientists need to quantify their constructs, map their fields of study, test their hypotheses, and measure the values of their social programs" (Wright 1996). Comparing both factor analysis and the cutting edge technique of item response theory gives increased confidence that the items, and the questionnaire as a whole, are functioning to a high standard. Comments and feedback from family members was taken into account when removing the items and amending the measure.

Item response theory is a modern psychometric method usually carried out using the Rasch measurement model (Rasch 1960). Although the model has been widely used in education for the last 40 years, it has only recently been applied in the health sciences (Tennant and Conaghan 2007). The Rasch model shows "what should be expected in responses to items if measurement is to be achieved" (Pallant and Tennant 2007), and both polytomous (Andrich 1978) and dichotomous (Rasch 1960) versions of the Rasch model are available. The Rasch model is based around the probabilistic Guttman pattern (Andrich 1985), meaning that if an item of higher difficulty is affirmed by the subject, the probability of an easier item also being affirmed is high (Tennant and Conaghan 2007). This also allows the scores for individual items to be summed to give a total score for a measure, as the Rasch model is based around a unidimensional model. An important part of the process of Rasch analysis is confirming whether a unidimensional scoring system is appropriate for the data set, and this can then be confirmed using classical test theory and factor analysis. Tennant and Conaghan (2007)

explains how the Rasch model transforms individual items scores to interval data, providing a linear relationship, using the example of a pain score: *"The model assumes that the probability of a given respondent affirming an item is a logistic function of the relative distance between the item location and the respondent location on a linear scale. In other words, the probability that a person will affirm an item is a logistic function of the difference between the person's level of, for example, pain and the level of pain expressed by the item, and only a function of that difference".* Rasch analysis can be used when designing a new scale and selecting items to include which contribute to a total score (Court et al. 2007), or when evaluating the properties of an existing measure (Liang et al. 2009). It can also be used in computer adaptive testing when creating an item bank (Lai et al. 2003). The process of Rasch analysis in this study was carried out using the software RUMM2030, which is designed to be able to perform the multiple tests required for exploring the data-model relationship, and produce the formal statistics required to test this relationship (Hagquist et al. 2009). This latest version of the RUMM software has several additional features compared to the earlier RUMM2020: the software can be used to assess dimensionality of the measure, data sets can be created with complete values only, the cumulative person distribution can be referenced to the normal counterpart in terms of the mean and standard deviation, and the clarity of presentation of results and graphs has been improved.

Stage 6: Further Validation

The aim of this stage was to assess the psychometric properties of the newly developed instrument and establish its validity, reliability and sensitivity to change. This would allow the new measure to be used effectively in future studies, and ensure that it is fit for its intended purpose. A new cohort of family members were recruited for this purpose. The following instruments were used during the further validation phase:

The WHOQOL-BREF

The WHOQOL-BREF is a generic quality of life instrument, commonly used across many different countries to assess the quality of life of adults with and without illness (Appendix S). The WHOQOL-BREF was developed from a longer and more detailed instrument, the WHOQOL-100, which has also been validated and used internationally (The WHOQOL Group 1998b). The WHOQOL-BREF was developed for use where time is restricted, where the investigator wants to reduce the burden on the subject, and where less detail is needed in the responses (Skevington et al. 2004). The items selected from the WHOQOL-100 for inclusion in the WHOQOL-BREF were identified using the data from multinational studies of the WHOQOL-100, and were selected by their relationship with the overall WHOQOL model, the discriminant validity and their proportion of variance within their domain (The WHOQOL Group 1998a). The WHOQOL-BREF is a self-administered questionnaire which consists of

26 items and 4 domains: physical, psychological, social and environmental (Skevington et al. 2004). The instrument is scored on a 5-point Likert scale: Very poor, Poor, Neither poor nor good, Good and Very good and individual item scores are added to give a total score (two items are reverse-scored). Higher scores mean a higher quality of life. Scores from the WHOQOL-BREF can be transformed on a scale of 0-100 in order to be compared to the scores from the WHOQOL-100 (Skevington et al. 2004). The WHOQOL-BREF has been extensively tested in a multinational and wide population for validity and reliability (Skevington et al. 2004; 1998a). The instrument has also been developed and validated in many different countries (Izutsu et al. 2005; Leung et al. 2005; Min et al. 2002; Yao et al. 2002). In this study, the WHOQOL-BREF was completed by family members, and was used to assess the construct validity of the convergent type. It was selected for use as it has a short completion time, has been widely validated in a range of populations, is straight forward to score and has been extensively tested in terms of reliability and validity; this increases the confidence in the measure to produce reliable results and put a minimal burden on the family members in the study. Before the measure was selected and this stage of the study was completed, the investigator thoroughly reviewed the content of the WHOQOL-BREF.

The global health (GH) score

The global health (GH) score was used in several stages of the validation. The GH score asked family members about their subjective assessment of the patient's health at the moment on a 0-10 visual analogue scale. 0 indicated worst possible health, and 10 indicated perfect health (Figure 2.1).

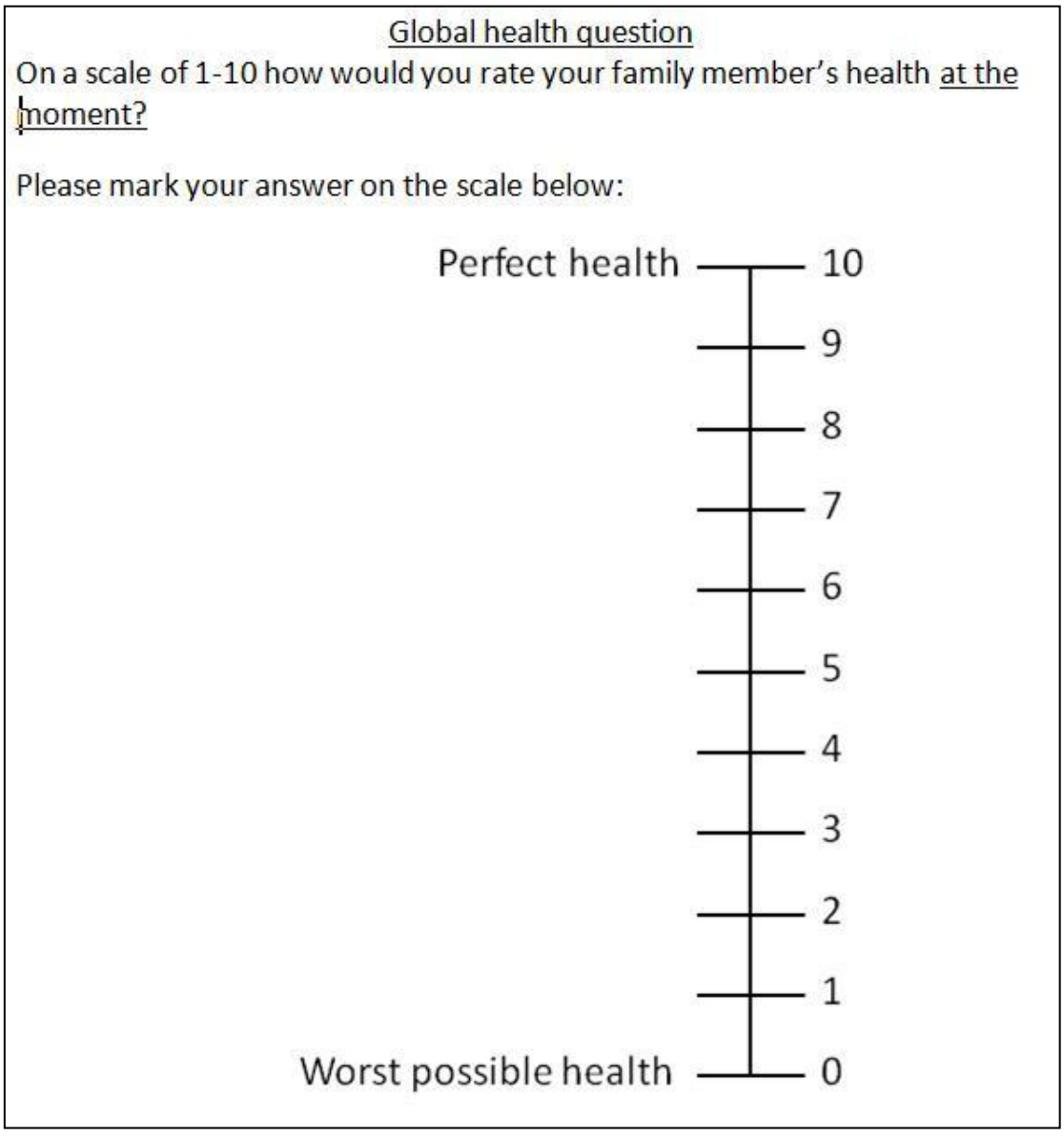
The GH score was correlated to the total score of the family quality of life instrument during the construct validity stage and was used to assess whether the patient's health status had changed during both test-retest reliability and sensitivity to change studies.

Follow up recruitment procedure

Subjects were followed up during both the test-retest reliability (after 7-14 days) and the sensitivity to change test (after 2 months). With the aim of maximising response rate, family members were given the option of being followed up via post or email, and provided postal or email addresses accordingly. Those who were followed up via post were sent a covering letter explaining the importance of returning the questionnaires on time (Appendix U), and a prepaid envelope to return them in. Each of the questionnaires was numbered by the investigators before they were sent out, so that they could be matched up with the correct subject when returned. Those subjects who chose to be followed up via email were sent a covering email with the same wording as the covering letter, with a link to an online version

of the instrument, together with an introductory page and an electronic version of the global health (GH) score.

Figure 2.1: The global health (GH) score completed by family members of patients



Once the subject had submitted their responses they were accessed online, via a password protected system, for analysis. Subjects were asked to enter their age, initials and relationship to the patient as unique identifiers, so that they could be matched up with the correct subject. During both forms of recruitment, no personal details relating to the family member or patient were released. Subjects were provided with contact details for the investigators in case they had any questions or trouble completing the questionnaires. Any subjects who had not completed their questionnaires after a week were followed up in order to increase the response rate.

Reliability

Reliability has been described by Nunnally (1967) as “the extent to which [measurements] are repeatable and that any random influence which tends to make measurements different from occasion to occasion is a source of measurement error”. Reliability was tested using internal consistency and the test-retest method.

Internal consistency

This examines the variance between a subject’s true score, and the total score for a measure (Charter 2003), and the most widely used measure of internal consistency (Streiner 2003) is Cronbach’s alpha (α) coefficient (Cronbach 1951) which measures the degree of consistency of the scale. The α coefficient has a value of between 0-1 and authors differ in their opinions of the acceptable value of α. Most suggest that α should have a minimum value of 0.7 (Heppner et al. 1992; Kaplan and Saccuzzo 1997), which was used as the determinate level for internal consistency during this study. Others suggest that the α value should be above 0.8 (Clark and Watson 1995; Nunnally 1978). Some authors also suggest that if the instrument is to be used clinically, the α value should be above 0.9 (Ponterotto and Ruckdeschel 2007), although this contradicts Streiner and Norman (2008) who argue that if α is above 0.9 then there could be a high level of item redundancy and the scale may be too narrow in scope. Ponterotto and Ruckdeschel (2007) argue that an internal consistency greater than 0.7 should be met in instruments which have more than 11 items.

Test-retest reliability

The purpose of this type of reliability is to examine the reproducibility of results, and therefore the stability of the scale when repeated used under the same conditions. In the same way that it is important for an instrument to be able to detect a change in disease state, it is just as crucial for the instrument to demonstrate stability (Guyatt et al. 1987). The choice of time interval for follow up in a test-retest analysis is an important consideration. If the interval is too long, the disease state may have changed, or other external factors may influence the QoL. Conversely, if the interval selected is too short, the subject may remember their original responses and answer the questions from memory (Streiner and Norman 2008), often overestimating the reliability (Hunt et al. 1981). The test-retest reliability can vary depending on the nature of the disease (Meltzer and Hochstim 1970), and a retest interval of between 2 and 14 days is usually considered acceptable (Streiner and Norman 2008). In this study, family members were sent another copy of the instrument via email or post (depending upon their choice) 7-14 days after the initial recruitment, and asked to complete it again. The interval of 7-14 days falls within the acceptable time period outlined by Streiner and Norman (2008). It was felt that leaving a period of a week would help to prevent subjects answering the questions from memory, but by capping the time period at 14 days it would also help to

reduce the chance of change in the patient's disease state. During this round of recruitment, family members were also asked to complete a global health (GH) score, rating the patient's disease from 0-10 depending on how bad they felt the patient's condition was. Subsequently, 7-14 days later, the global health score was sent to the family member, along with the newly developed family quality of life instrument. The questionnaire results were analysed for family members who had indicated no change, or very little change (no more than one point on the 0-10 scale) on the global health score. Correlation between the results from the initial recruitment and the results from the 7-14 day follow up were analysed using the Intraclass Correlation Coefficient (ICC), which is generally recommended over the Kappa coefficient when testing polytomous data for a number of reasons, including the fact that it can handle missing data (Streiner and Norman 2008).

Validity

Face validity and practicality

Face validity and practicality of the instrument was assessed for a second time after item reduction using feedback questions which were given to the family members during this stage of recruitment. Face validity is important, as if the item appears irrelevant on the surface, the subject may omit it (Streiner and Norman 2008), and an instrument with face validity is known to increase motivation amongst subjects and reduce dissatisfaction (Nevo 1985). Family members were asked to complete four questions about the face validity and practicality using "yes" or "no" tick boxes. They were also asked to report any comments or reasons why they may have disagreed with the statements in the questions. As a further measure of practicality, family members were timed when completing the questionnaire. It is important that the instrument does not put a large time burden on the family member, and it was felt that the mean time taken to complete the instrument should be less than five minutes, so that during future use, it minimises burden on both the family member and the investigators or clinicians. The time taken to complete the instrument was correlated with the family member's age using Pearson's product moment correlation coefficient. Pearson's product correlation was chosen as both values are made up of continuous data. The readability score and item lengths of the new instrument were also assessed. The standard Flesch test for readability was used (Flesch 1948), with the aim to score between 60 and 70, giving it a "standard" readability. The desired item length was "as short as possible, although not so short that comprehensibility is lost"(Streiner and Norman 2008). Holden et al (1985) also found that shorter items have higher validity coefficients.

Construct validity

This was assessed to show that the newly developed instrument is measuring what it is intended to measure. Construct validity examines the relationships between behaviours and attitudes (Streiner and Norman 2008) and is often measured by comparing the scores of the new measure with a similar instrument with established validity. However, new measures are often defining new concepts, and by definition, no similar measure exists. Therefore, construct validity is often assessed by developing a hypothesis which relates the attribute we are measuring to other known attributes (Streiner and Norman 2008). This hypothesis can be accepted as a measure of construct when there is a strong, positive fit between the prediction and the subsequent data (Cronbach and Meehl 1955).

As no similar measure to the newly developed family quality of life instrument exists, two *a priori* hypotheses were devised to attempt to establish a relationship between the instrument and another measure. The first hypothesis devised was that *the impact of illness on family member's QoL is correlated to the family member's overall QoL*. During this stage of recruitment, family members were asked to complete both the WHOQOL-BREF, a generic quality of life measure, and the newly developed measure. The scores from the newly developed measure and the WHOQOL-BREF were then compared using Spearman's correlation coefficient, as the newly developed measure has ordinal scaling and the WHOQOL-BREF has interval scaling. The hypothesis would be accepted if the correlation were to be negative (as the two measures are scored in different directions). Correlations would also be expected to be seen between the separate domains of the two questionnaires.

Secondly, it was hypothesised that *the impact of illness on family member's QoL is correlated to the health of the patient,* in this case, the global health (GH) score. The relationship between disease severity and impact on the family has already been proven in several individual specialties (Balkrishnan et al. 2003; Reiter-Purtill et al. 2008; Zashikhina and Hagglof 2009). The instrument score and the GH score were correlated using Spearman's correlation coefficient, as both data sets are ordinal (Pallant 2005).

Sensitivity to change

It is important for any quality of life measure to be able to demonstrate change over time, or sensitivity to change (Lohr 2002). For the instrument, this means demonstrating a difference in family quality of life as a response to an improvement or decline in a patient's health. In this study, a preliminary investigation of sensitivity to change was carried out with five family members of patients from five of the core medical specialties. After a period of 2-3 months, the 25 family members were sent another copy of the instrument and the global health score. This time period was chosen as it was felt that it was long enough for the patient's health to

have changed in many of the wide range of conditions sampled, and was decided upon with the help and advice of experts. Those family members who indicated that their relative's condition had changed in severity by two or more points on the global health score were included in the analysis, which compared their original scores during the third round of recruitment to their scores after 2-3 months. The participants were divided into two groups- "improved" (where the QoL score had decreased) or "worsened" (where the QoL score had increased). The two groups were compared using the Wilcoxon Signed rank test to assess the within-group difference, the Mann-Whitney U test to interpret the difference in the scores between the two groups and the Spearman's rank correlation coefficient to determine the correlation between the changes in the QoL score and change in global health score. The sensitivity to change study was only a preliminary investigation with small numbers due to the time constraints of the project. This will be explored further in future work.

Regression analysis

A multiple regression analysis was carried out to identify some of the possible factors influencing family QoL. The choice of regression (standard, hierarchical, stepwise) was influenced by the results of the correlation tests carried out during the validation. The total QoL score was set as the dependent variable, and nine other demographic factors were set as the independent variables. These included health of the patient (GH score), family member age, family member gender, patient age, patient gender, relationship between family member and patient, patient's disease duration, family member's education level and family member's total annual household income.

DATA ANALYSIS

Data was analysed during the study using a variety of software packages and statistical techniques. Qualitative analysis was completed using NVivo (version 9), which quantified the common themes identified and calculated the number of themes mentioned by each participant. The Predictive Analytics SoftWare Statistics (PASW version 18, formerly known as SPSS) was used for the statistical analysis of all of the data, including calculating the frequency and percentage of each category of each variable, the mean, median, standard deviation and minimum and maximum. PASW was also used to carry out parametric tests on the quantitative data. The use of parametric and non-parametric tests has been widely debated in the area of quality of life instrument development. Parametric tests (for example factor analysis, ANOVA and structural equation modeling), which assume normality of data are commonly used in instrument design, and have been used successfully for many years (Norman 2010). Furthermore, Norman (2010) describes these methods as "versatile, powerful and comprehensive". Due to the large sample size, the fact that ANOVA has been shown to be robust for data with non-normal distributions (Pearson 1931) and the fact that

the results of parametric and non-parametric tests are often similar (Norman 2010), parametric tests were chosen to compare individual groups (gender, age groups, specialty etc.) and the family quality of life data. Spearman's rank correlation coefficient was chosen as it is the recommended test to examine the relationship between sets of ordinal data (Pallant 2005). Throughout, a probability of $p<0.05$ was considered to be significant. PASW was also used for the factor analysis of the questionnaire, where an orthogonal Varimax rotation was selected as it is the most widely used rotation method, thought to produce the clearest results (Fabrigar et al. 1999). PASW was also used to plot a Scree plot, and produce structure and component correlation matrices. The software RUMM2030 was used to perform the Rasch analysis, including producing all tables (item and person fit, principal components, residual correlations), graphs (category probability curves, equating item subsets, item characteristic curves, person-item distribution, threshold map) and statistical analysis (paired t-tests, ANOVA, mean, median, maximum and minimum). Microsoft Office Excel 2007 was also used alongside RUMM2030 to carry out a binomial test, and was also used to build many of the graphs presented in the Results chapters.

CHAPTER 3

Development of a Generic Family Quality of Life Instrument: Qualitative Phase

INTRODUCTION

In order to develop a quality of life measure for use in family members of patients, it was important to gather in-depth and high quality data about the ways that illness affects family members of patients. The aim of this part of the study was to collect in-depth information about the impact of illness on family members, which would serve as both the basis for a new measure, and as a valuable piece of qualitative work in its own right. Several methods for collecting this data were considered and this qualitative phase of the study was designed to underpin the constructs for further development of the measure, and to ensure that all aspects of this impact across medicine were captured. This information was then used to develop the items to be included in the new quality of life measure. Although this is a subject which has been covered in previous literature in individual disease areas, no other studies or bodies of work exist looking at the family impact of illness over a range of different diseases. Therefore, it was decided that original data from family members should be the source of the questionnaire items. This is especially important in the area of quality of life outcome research where the information gathered on which to base measurement tools should be subjective, and not measured or dictated by a clinician or observer (Streiner and Norman 2008).

METHODS

Individual interviews

Individual semi-structured interviews with family members of patients were chosen as the method of qualitative data collection. The aim of these interviews was to collect high quality and in-depth data about how illness affects family members of patients. To reduce interviewer bias, all interviews were carried out with the same interviewer. The interviews were semi-structured, with broad opening questions, subsequent follow on questions and then closed questions to encourage family members to give examples and expand on answers where possible. The opening question in each interview was *"Can you tell me about any ways your life has been affected by having an unwell relative?"*. At the end of each interview this question was repeated again to give the family member a chance to talk about any areas that had not been already covered. Each topic that the family member brought up was covered in detail in the first part of the interview and then an interview guide, based on the previous literature looking at the impact of illness on families (Ferrario et al. 2004; Poston et al. 2003; Turnbull et al. 2004), was used to discuss any topics which had not already been covered. The interview guide contained some themes and questions relating to how illness affects family members. The interview guide was used as a prompt but was not heavily relied upon as the interviewer wanted to keep an open mind about potential themes and topics in

this new area of research. Using a semi-structured interview helps the participants to feel more relaxed and more conversational, encouraging them to open up and talk in more depth about sensitive issues.

Inclusion and exclusion criteria

The inclusion and exclusion criteria for family members and patients for qualitative data collection were as follows:

Inclusion criteria for family members

- Age 16 years and above
- Able to understand and read English
- Be an immediate family member, or partner living with, or caring for, a patient diagnosed with one or more medical conditions under one of the selected specialities (see chapter 2)
- Able to give written informed consent

Inclusion criteria for patients

- Able to understand and read English (if over 16 years old)
- Diagnosed with one or more medical conditions under the selected specialities (see chapter 2)
- Able to give written informed consent (if over 16 years old; if patient is under 16 years old, parent/guardian must be able to give written, informed consent)
- Attending clinic with a family member, or being a hospital inpatient.

Exclusion criteria for family members

- Age under 16 years
- Unable to understand and read English
- Not considered by the patient as a family member
- Unable to give written informed consent
- Having a severe handicap or disability which prevents them participating in an interview or filling out a questionnaire.

Exclusion criteria for patients

- Unable to understand and read English (unless under 16 years old)
- Unable to give written informed consent (unless under 16 years old)

Sampling and recruitment procedure

Patients were recruited for the study in conjunction with clinicians from 26 specialties at the University Hospital of Wales, Cardiff, University Hospital Llandough, Gabalfa Clinic, Velindre Cancer Centre, in General Practice or at the participant's home. The selection of specialties and the clinicians' involvement is explained fully under in Chapter 2.

Purposive sampling (Barbour 2001) was used, where the clinicians selected at least five patients who were suffering from different conditions and were representative of typical patients in their specialty. It was considered important by the investigators to consider how the convenience sample in the study would differ from a randomly selected sample, so the interviewer met with each of the clinicians prior to recruitment to explain the importance of recruiting a representative sample from each specialty. Although this mostly included chronic patients, patients who were awaiting surgery or had undergone surgery were also included. Patients suffering from more than one medical condition were not excluded from the study as it was felt that including patients with co-morbidities would more closely reflect reality. Instead, the interviewer used the interview guide to focus the family member on the effects of the principal diagnosis: any extra information added to the richness and variety of the data.

One family member of each of the patients identified was then approached and asked to take part in the study. A variety of family members were approached. Written informed consent was taken from both the patient and family member before the interview began. Each interview took place in a private room without the patient present, unless the patient was under 10 years of age and the parent was being interviewed. Each interview was audio recorded and transcribed verbatim.

In order to obtain the highest quality data and reduce the chance of interviewer bias, the interviewer was trained for the task by attending a social research interviewing course and studied the art of interviewing, including the use of language, the set up of the interview room, and methods of analysis (Kvale and Brinkmann 2009).

Demographics

As well as collecting in depth information about the ways that illness can affect family members of patients, demographic details were also collected from family members and patients before the interviews. This was in order to allow the population interviewed to be

described, and at later stages in the study, comparisons were able to be made between different demographic categories. The data could also be used to assess the spread of patients and family members, and investigate the extent of the variety of conditions that were sampled. Demographics collected from the patient included: age, gender, ethnicity, primary diagnosis, other diagnoses, disease duration and occupation. Demographics collected from the family member included: age, gender, ethnicity, relationship to patient, and socioeconomic data including occupation, highest level of education and annual household income.

Pilot interviews

Three trial interviews were carried out with family members of patients prior to the beginning of the study. The results from these interviews were not included in the analysis, patients and family members were fully informed of the nature of the interviews and written informed consent was taken. The trial interviews gave the interviewer a chance to practise using the interview guide and make any necessary changes, as well as practicing using the audio equipment. The trial interviews also allowed the interviewer to rehearse the introduction to the study and practice using the consent and demographic forms. This allowed the interviewer to feel more relaxed and comfortable going into the first interview.

Sample size consideration

The sample size calculation for the interviews in this study was based around two considerations. The first was ensuring that the interviews contained data from family members across all 26 specialties, and as family members from some specialties were recruited before others, this meant that interviews had to be continued until a roughly equal number of family members had been sampled from all specialties. Given the time frame and the practicalities of the study, the interviewer aimed to recruit a minimum of five family members from each specialty, giving a total predicted sample size of 130 interviews. Secondly, the saturation point of the data was taken into consideration; a common way of determining sample size in qualitative research (Guest et al. 2006). The saturation point is the point at which no new themes arise, meaning that the participants are talking about common themes and a high level of confidence can be achieved that themes which are important and relevant to family members have been identified. In this study, interviews were continued well past this point to ensure confidence in the data, and to be sure that relevant themes from all specialties had been covered. This is discussed further in the "Discussion" section of this chapter.

RESULTS

Sociodemographic characteristics of the study participants

140 family members of patients were approached and asked to take part in the study. Seven family members declined to take part due to time constraints. This gave a total of 133 family members of patients recruited for interviews across the 26 specialties. Most family members were White British (93%), female (61%), the partner or spouse of the patient (56%) or the parent (22%). The mean patient disease duration was 8.9 years (range 1 month to 60 years), and the mean patient age was 54.7 years (range 3- 97 years). The demographics of the family members interviewed can be found in Table 3.1. As the sample covered a large range of specialties and illnesses and was designed as an exploratory qualitative study with a restricted time frame, the sample was stratified by medical specialty but not by sociodemographic variables. A large-scale study in individual patient and family member groups has been suggested as a piece of further work. Patients suffered from one or more of 144 diseases or types of surgery (Appendix E). The shortest interview lasted just 2 minutes and 25 seconds and the longest lasted for 60 minutes. The total length of the digital recordings of the 133 interviews was 23 hours, 6 minutes and 52 seconds. The mean interview length was 12 minutes. There was no missing data in this part of the study as the demographic questionnaires were completed by the interviewer.

Saturation point

The saturation point was reached at interview number 40, after which no new themes emerged, and family members were reporting very similar issues. Interviews were continued past this point to include data from all specialties, in case new or unexpected themes emerged. As there were many specialties, the interview number was continued to 133, giving confidence that all themes had been identified. At least 5 participants were sampled from each specialty apart from infectious diseases, general practice and urology where 6 were selected. Between interview numbers 40 and 133, the participants gave different examples, adding to the richness of the data.

Thematic analysis

The content of the 133 interviews was transcribed and then coded using NVivo 9 software. The process of coding the data followed that outlined by Braun and Clarke (2006), and included familiarising oneself with the data, generating initial codes, searching for themes, reviewing themes, and then defining and naming themes. In many quality of life studies, coding frames are developed based on the content of the interviews before coding begins (Gabriel and Bowling 2004). It was felt that as this area of research had not been explored

previously, a rigid coding frame would be too restrictive. Instead, before coding began, the interviewer identified ten main themes from the content of the transcripts and these themes were used as a starting point for coding. A flexible approach was used for the coding (Braun and Clarke 2006), and subthemes/codes were coded under the ten main theme headings.

Many of the subthemes identified from the coding related to one another, but each was placed under the most suitable theme heading and given appropriate labels to identify them. Quotes and examples from family members to represent each theme and subtheme were also identified from the text. Several steps were taken to verify the results of the coding and ensure reliable and high quality data (Singer et al. 1999):

a. The coding process was repeated twice using the NVivo software.
b. The coding process was also repeated manually by the interviewer, who wrote coding notes in the margins of the transcripts.
c. The coding and themes identified were discussed in detail with the other investigators (AYF, SS and MKB) and a consensus was reached.

The prevalence of each theme and subtheme was recorded to identify the most common areas of family members lives affected by illness.

Themes identified

Ten themes and 196 subthemes/codes were identified from the interviews with 133 family members. These themes represent the areas of family members' lives which are affected by patients' illness. The themes and subthemes are represented in Table 3.2 along with the percentage of family members who were affected by each theme. The main themes included: emotional impact (mentioned by 92% of subjects), daily activities (91%), family relationships (69%), sleep and health (67%), holidays (62%), support and medical care (61%), work and study (52%), financial impact (51%), social life (37%), and time planning (14%), as represented in Figure 3.1.

The mean number of themes mentioned by participants was 6 (median= 6, S.D= 2.03, range 0-10 [max=10]). The mean number of themes mentioned by family members from each of the 26 specialties was also calculated. Family members of haematology and genetics patients were affected by the most number of themes, and family members of gynaecology and diabetes patients were least affected (Table 3.3).

Table 3.1: Demographics of family members recruited for interview (n=133)

Total number of family members (n)	133
Gender	
Males	39%
Females	61%
Age (years)	
Mean	56.1
Median	56
Range	21-85
Relationship to patient	
Spouse/partner	56%
Parent	22%
Child	15%
Niece/nephew	1%
Grandparent	2%
Sibling	2%
Grandchild	1%
Cousin	1%
Educational level	
Less than secondary school	11%
Secondary school	34%
A levels/college course	27%
University degree	17%
Masters/doctoral degree	6%
Prefer not to say	5%
Ethnicity	
White British	93%
Mixed	2%
Asian or Asian British	3%
Black or Black British	2%
Combined annual household income	
Less than £10,000	13%
£11,000-£20,000	26%
£21,000-£30,000	23%
£31,000-£40,000	8%
£41,000-£50,000	6%
£51,000-£60,000	5%
£61,000-£70,000	4%
£71,000-£80,000	1%
£81,000-£90,000	2%
£91,000-£100,000	0%
Over £100,000	1%
Prefer not to say	11%
Age of patients (years)	
Mean	54.7
Median	61
Range	3-97
Patients' disease duration (years)	
Mean	8.9
Median	5.5
Range	1 month - 60 years

Table 3.2: Identified themes and subthemes from 133 interviews representing 26 medical specialties

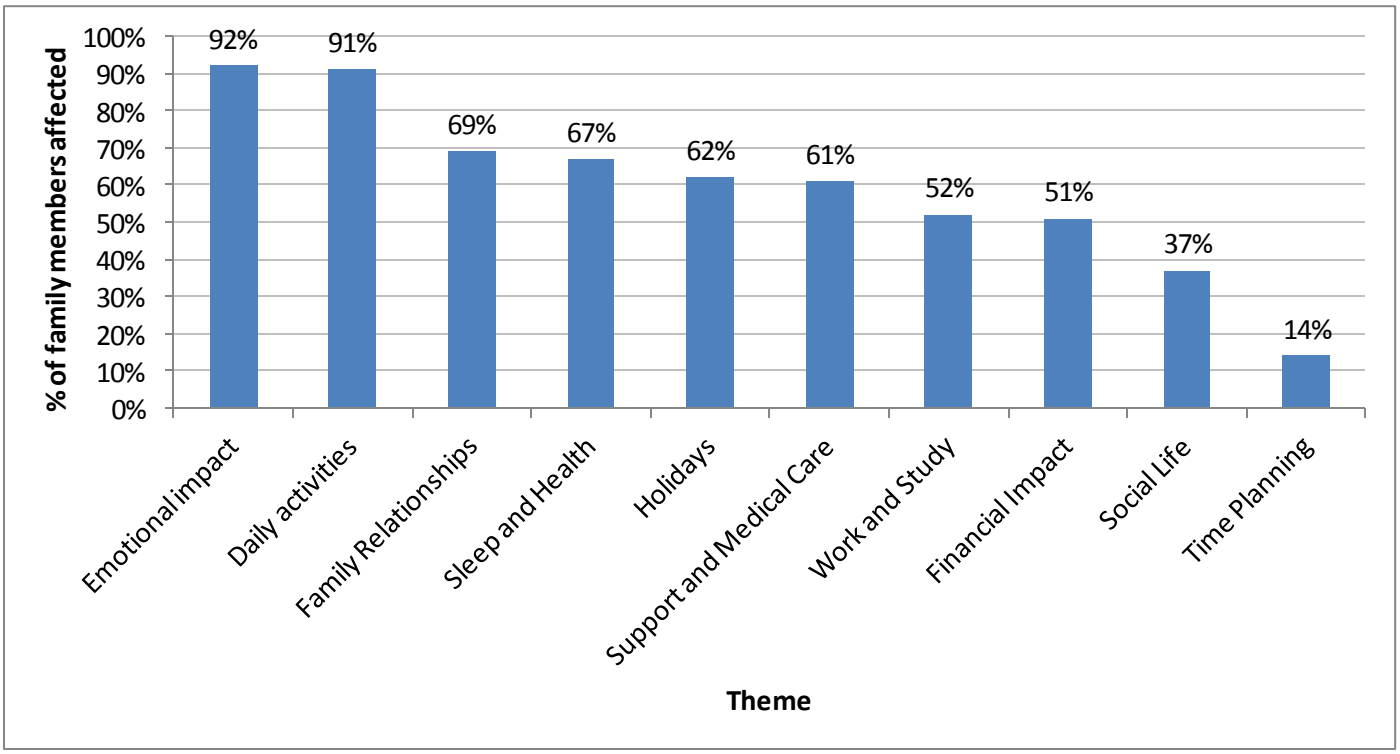
Theme (percentage of family members affected)	Subthemes/codes (percentage of family members affected)
Emotional impact (92%)	Accepting what has happened (6.0%) Affected behaviour or personality (9.8%) Aggression (0.8%) Angry (15.0%) Annoyed (6.8%) Anxious (5.3%) Can't cope (3.0%) Changing emotions (0.8%) Concern (6.0%) Confusion (2.3%) Crying (8.3%) Depressed (5.3%) Devastated (3.0%) Developed a tougher attitude (0.8%) Disappointed (1.5%) Disbelief (0.8%) Distress (3.0%) Fear (3.0%) Fed up (0.8%) Feeling down (1.5%) Feeling more emotional (6.8%) Feeling sorry for self (0.8%) Feeling tense (0.8%) Feels sorry for patient (5.3%) Frustration (27.1%) Grief (1.5%) Guilt (14.3%) Have to think about patient more (4.5%) Have to think ahead more (0.8%) Helpless (5.3%) Impatient (0.8%) In back of mind (10.5%) Increased responsibility (4.5%) Irritable (1.5%) Isolated (0.8%) Jealous (0.8%) Life on hold (1.5%) Life turned upside down (0.8%) Lonely (4.5%) Lost own life (3.0%) Missing patient when in hospital (1.5%) No one to talk to about feelings (20.3%) Panic (3.8%) Pretending everything is ok (6.0%) Psychological effects (1.5%) Relying on others and lack of control (1.5%) Rollercoaster (3.0%) Sadness (6.8%) Scared (1.5%) Shock (9.8%) Sick and tired (0.8%) Stressed (18.8%)

	Torturous (0.8%) Try to laugh about it (3.0%) Upset (22.6%) Why us (4.5%) Wishing patient was well (2.3%) Won't get life back (0.8%) Worry (35.3%)
Daily activities (91%)	Avoid situations where alcohol is present (1.5%) Can't ask patient to help around house (1.5%) Can't physically deal with patient (2.3%) Caring for patient (37.6%) Change of lifestyle (6.8%) Child care issues (3.8%) Coping around the house (6.0%) Difficult to do shopping (1.5%) Difficulty with patient's schooling (1.5%) Don't go out late at night (1.5%) Eating different diet (8.3%) Eating out (7.5%) Family activities (8.3%) Has to be a translator for patient (0.8%) Have to learn how to live differently (2.3%) Have to limit activities (3.0%) Have to move house (1.5%) Have to prioritise activities more (0.8%) Hobbies (34.6%) Increased housework (47.4%) Loss of freedom (4.5%) Making excuses not to visit patient (0.8%) No alone time (6.0%) No time for housework (2.3%) Not eating (1.5%) Patient moved into their house (0.8%) Plan mealtimes differently (0.8%) Retirement plans affected daily (4.5%) Stopped going out (8.3%) Takes up time (6.8%) Taking one day at a time (1.5%) Telephoning patient to check on them (0.8%) Thinks patient is lazy (0.8%) Thinks patient tries to do too much (3.0%) Travel (18.0%) Visiting patient in hospital (10.5%)
Family relationships (69%)	Argue more (24.1%) Become more of a carer (18.8%) Can't have children (0.8%) Difficult being single parent (2.3%) Doesn't like being away from patient (25.6%) Doesn't understand how patient feels (0.8%) Family torn apart (0.8%) Feels like patient has no one else to help them (5.3%) Feels that patient won't listen to them (1.5%) Hard to communicate with patient (1.5%) Has to look after patient's children (3.0%) Hates the patient (0.8%) Having to explain to children (2.3%) Made relationship uncomfortable (0.8%)

	Not spending enough time with family (4.5%) Other family members affected (14.3%) Other family members don't understand (2.3%) Patient thinks relative doesn't want to help them (0.8%) Playing more than one role (2.3%) Pressure from patient (3.8%) Relationship problems (3.8%) Sexual relationship (21.0%) Spend too much time together (4.5%) Stuck in the middle (0.8%) Supposed to care for each other (4.5%) Trying to get patient to go to school (0.8%) Want to spend as much time with patient as possible (1.5%) Wants patient to be normal (0.8%) Wants patient to be sectioned (0.8%) Wants to leave home because of patient (2.3%) Wishes patient was dead (0.8%) Wishes patient would talk more (0.8%) Worrying about how other family members are feeling (2.3%)
Sleep and health (67%)	Sleep affected (66.2%) Health affected (6.0%)
Holidays (62%)	Anticipating problems (6.0%) Arranging holidays (2.3%) Avoiding sun (3.0%) Cancelled holiday (9.0%) Change type of accommodation (3.8%) Changing holiday activities (10.5%) Didn't enjoy holiday (2.3%) Family problems caused by going on holiday (0.8%) Not going on holiday (30.8%) Problems with insurance (0.8%) Problems with medication on holiday (2.3%) Problems with patient when away (9.8%) Shorter holiday (3.0%) Travel (13.5%)
Support and medical care (61%)	Difficult to talk about disease (6.0%) Don't like asking for support (3.0%) Family support (20.3%) Knowledge of disease (15%) Medical care (39.1%) People don’t understand (8.3%) Support decreased in general (2.3%) Support from friends (16.5%) Support groups (6.0%) Welfare support (4.5%)
Work and study (52%)	Affects studying (3.8%) Can’t work (1.5%) Exams affected (0.8%) Good to forget about problems (3.8%) Had to change hours (6.8%) Had to give up job (9.0%) Having to juggle work (3.8%) Leaving work early (4.5%) Make up hours missed at work (0.8%) Not taking all holidays (1.5%)

	Taking time off for appointments (16.5%) Taking time off to look after patient (11.3%) Work not understanding (25.6%) Worrying about future work (3.0%)
Financial impact (51%)	Buying more (26.3%) Cancelling holiday (2.3%) Can't afford house modification (1.5%) Desperation for money (0.8%) Having to give up work (3.8%) Having to think more carefully about finances (2.3%) Hospital parking (5.3%) Medication (1.5%) More expensive holidays (2.3%) More washing (1.5%) Petrol (9.8%) Private healthcare (2.3%) Started gambling (0.8%) Supporting patient (2.3%) Travel to hospital (6.8%) Unable to spend money (0.8%)
Social life (37%)	Can't afford social life now (1.5%) Changing friends (0.8%) Come home early from social activities (2.3%) Decreased (12.0%) Friends' reactions (3.8%) Given up alcohol so don't socialise (0.8%) Have to cancel social activities (4.5%) Have to think more carefully about social life (4.5%) Only go out locally (3.0%) Seeing family less (3.8%) Seeing friends less (9.0%) Strangers' reactions (8.3%)
Time planning (14%)	No subthemes

Figure 3.1: The percentage of family members affected by each of the ten themes



Description and discussion of the 10 themes

Emotional impact

One hundred and twenty two (92%) of the family members interviewed were affected emotionally by the patient's illness. The most commonly mentioned emotions included worry (35%), frustration (27%), anger (15%) and guilt (14%).

Worry was often reported when the family members were thinking about the future, or the death of the patient. One family member whose wife had been diagnosed with lymphoma explained how his emotional state had changed completely with the worry of the disease reoccurring: *"you go through sort of like fear, anger, the whole... everything...the life that you had, you'll never have back because in the back of your mind there's always that worry of "Is it going to come back?""*

Frustration was a common emotion felt by family members of patients in the study. The husband of a patient with leukaemia described *"I do get very frustrated with it, because I look at her and think "I wish I could do something", but there's nothing I can do, I'm so reliant on other people...it's a man thing isn't it? We like to have a degree of control...and feel we are doing the right thing by our family".* Others described the changes they had to make to their lives as a result of having an unwell relative as being frustrating: *"We've been married a long time and we've done things together and all of a sudden life changes completely, you can no longer do many things...it's been quite frustrating.*"

One man described his mix of frustration and jealousy towards the family guide dog as a result of his partner's Leber Optic Neuropathy: *"sometimes I get frustrated cos you think, before she had the dog she was stuck in the house and she got the dog to get some independence. The dog means more to her than me because the dog will go out in the rain with her and I won't".*

Most of the anger felt by family members was directed at the situation they have found themselves in, with many expressing a "why us?" attitude. Others felt anger directed at others, for example the patient's medical team: *"I get angry sometimes, especially when I see her coming home and they're [the doctors] saying they won't give her another treatment and she's sitting there in floods of tears".*

Table 3.3: The mean and median number of themes mentioned by family members of patients in each medical specialty (in descending order)

Specialty	Mean number of themes mentioned by family members	Median number of themes mentioned by family members
Haematology	8	9
Neurology	8	8
Genetics	7	9
General practice	7	8
Oncology	7	7
Cardiology	7	7
Mental health	7	6
Colorectal surgery	6	6
Paediatric endocrinology	6	7
Elderly	6	7
Orthopaedics	6	6
Rheumatology	6	6
Gastroenterology	6	7
Renal	6	6
Urology	6	6
Chronic pain	6	6
ENT	6	6
Respiratory	6	6
Infectious diseases	5	5
Dental surgery	5	5
Dermatology	5	6
Post stroke	5	6
Wound healing	5	5
Gynaecology	5	4
Ophthalmology	4	3
Diabetes	4	4

Feelings of guilt were reported by family members for a number of different reasons. Some family members felt guilty for not doing enough for the patient, for example not having time to take over the household chores, or help the patient with washing or dressing. Most reports of guilt came from family members who felt guilty for enjoying themselves when the patient is unwell, or for getting on with their daily lives when the patient is unable to. The 67 year old wife of a patient with uncontrolled hypertension explained how she loved her part time job in a charity shop, but felt guilty about working: *"It makes me feel guilty that I'm still working because I keep thinking, especially [the patient] collapsed, we should make the most of the time we've got, but then if I'm stuck in the house with him then we just get on each others nerves"*. One family member whose husband has bipolar disorder described feelings of guilt as she feels embarrassed by his condition and lies to friends and work colleagues, in case they think her husband is "crazy". Two family members, both mothers of patients, explained that they felt they could have done more to prevent the patient from becoming unwell, and

this lead to feelings of guilt. One mother, whose child suffers from the rare genetic condition DiGeorge syndrome, felt guilty for not taking folic acid during her pregnancy, despite the fact that she had been told it would not have made a difference.

Less commonly reported psychological effects included feeling upset, annoyed, helpless, stressed and lonely. Twenty seven (20%) of the family members found it difficult to find someone to talk to about these feelings, often keeping their feelings to themselves. One family member described her difficulty in finding someone to talk to about her feelings: *"It's nice to have somebody to talk to, you know, if they just pop in...and stay for a cup of tea...when you first get the diagnosis everybody comes around and everybody phones but then it peters off and no one bothers".* Another, whose son has diabetes, said *"You just work your feelings out in your head, you don't want to show the kids that you're stressed because it just makes them worse."* Some participants reported that the stress of having to hide their emotions in order to provide support for the patient caused a change in their personality or behaviour, giving examples of becoming more withdrawn, more angry and more anxious. Others described feeling "helpless", as they feel there is nothing they can do to help the patient. The husband of a patient with Crohn's disease said: *"It was quite difficult not being able to help her and obviously she was in pain quite a lot. It makes you feel useless because you can't do anything apart from pass some encouraging words or sympathise but you can't change it".*

Feeling sympathy towards the patient, often described as "feeling sorry for" the patient was frequently described when family members talked about providing support. Others talked about finding it difficult to accept that their relative was unwell, particularly when the patient's symptoms were debilitating or severe. One mother of a four year old child with developmental delay and learning difficulties explained; *"From an emotional point of view...it's obviously a very difficult thing to get used to. The fact you have a child you think will be OK and you have hopes for him, and to find that completely turned on its head, and the fact he's probably going to need care 24/7 for the rest of his life...that impacts the whole family".*

As well as talking in depth about these emotional effects, family members also talked about some of the physical effects these emotions had, most commonly crying. The grandmother of a patient with muscular dystrophy explained how she had cried every day for six months after the diagnosis was made, and just thinking about the diagnosis still made her cry 17 years later. Other family members said they felt more emotional in general, and would react to everyday situations differently to before. Some family members also reported feeling lonely, saying "*I feel lonely because he [the patient] sleeps all the time",* and *"He [the patient] lies in*

bed and is looked after and no one cares about me". Stress was another subject brought up frequently by family members.

For some family members, the emotional effects of having an unwell relative are so severe that they compare their feelings to that of bereavement. One family member said "*It's almost...like a bereavement. You have aspirations for them...expect them to get married, have their own children, careers...and suddenly someone tells you actually your child is really disabled...it's almost like having a child die".*

Daily activities

The negative effect on day-to-day living as a result of having an unwell relative was reported by 121 (91%) of the family members interviewed. For 51 (38%) of the family members this involved aspects of caring for the unwell relative, including helping with dressing, personal hygiene needs, assisting with mobility and providing food. Many family members reported feeling a burden from caring for the patient, which often left them feeling like they had no freedom and no time to enjoy their own interests. The wife of a patient with prostate cancer said *"I wait on him, he just sits in his chair...I have to do his night bag [catheter bag] and everything and just have to be there really. If you go down the road or anything, you're thinking "oh, I've got to get back and make his tea" because he doesn't even make a cup of tea now, he can't".* Another family member discussed the difficult issues with caring for her daughter and assisting with her personal hygiene during psychotic episodes: *"When an episode would start she would wet herself, she would wee on the floor. When she was menstruating...I would have to help her. Otherwise she would just come downstairs covered in blood...So literally...it was a case of twenty four, seven".*

Forty seven (35%) of family members reported their hobbies or pastimes being affected and described a complete change in lifestyle as a result. Some described hobbies or activities they enjoyed with the patient, which they could no longer do together: *"We used to like climbing hills and mountains and she [the patient] can't. It's a bit distressing that she can't share in all of my enjoyments"*. Most family members who reported their hobbies being affected had to give them up, or spend less time on their own interests. The husband of a patient with vulvar neoplasia said: *"I ride a motorcycle and obviously that's had to go in the garage whilst she's been ill because I can't leave her".* Another family member described: *"I was doing my keep fit and going to different classes and stuff but all that came to a stop because I couldn't enjoy it any more. When I'm out on the bike I'm thinking about other stuff and when I'm walking I'm thinking "No, I should be doing something else"*. The hobbies and activities family members described as being affected included sports (swimming, shooting, rugby, fishing, going to the gym), activities (walking the dog, going to the theatre, going bowling), going to church and attending language classes.

Eleven (8%) of the participants described having to drastically change their diet as a result of the patient's illness, either to encourage the patient to eat a particular diet, or because they did not have the time to cook food or eat out. The wife of a patient with diabetes said *"We now have to have regular meals. There are a number of things we choose not to attend because we know the food is either slow in coming or doesn't come...it's vital to know what you're eating and you know it's there before you have your injections"*. One husband described the impact of his wife's diet restrictions*: "She's got coeliac disease which means...she has to have everything bought in; pizzas, bread, everything. She can't eat anything we would normally eat so I've got to make everything fresh for her which is a big expense as well".* Other family member described the impact of the patient's medical condition on eating out at restaurants, for example the husband of a patient with Crohn's disease not knowing exactly what ingredients are in the food, so being afraid to eat out in case the patient's condition was aggravated.

Sixty three (47%) of the family members described an increase in the amount of housework they had to do as a result of having an unwell family member. This was often a direct result of symptoms of an illness, for example having to do more washing when dermatology patients are applying creams, or the husband of a patient with leukaemia having to do more housework due to the side effects of the patient's treatment making her too tired to contribute. One family member described having to do her unwell mother's housework as well as her own: *"It's a case of hoovering, washing the dishes, going to the post office, sorting the paperwork. It's an awful lot and they [parents] don't realise...I'm sure they don't...it just takes up an awful lot to time...it's just time we could be spending doing other things"*. The wife of a patient with a pituitary adenoma and sciatia said *"Cleaning and things like that...I mean, I do that because he's not well enough...even as far as cutting the grass and things which he used to do before and he can't do any more. Cooking, I do all things like that, whereas years ago he would do that...it's much easier then. Now I guess it's difficult for him to do and he doesn't feel like doing it...his mood can sometimes be quite low".* Common examples of increased housework included washing, cooking, cleaning, making beds and gardening. Some family members had to pay for a cleaner as they felt they could not cope with the housework on their own, and many described how they were struggling with the amount of work they had to do.

Twenty four (18%) found that their everyday travel was affected, most commonly because the patient was no longer able to use their usual form of transport. The daughter of a patient with chronic kidney disease described the family's difficulty in getting the patient to hospital appointments: *"My brother brings us in but then he has to get a day off work to come because she can't travel by bus because she can't stand at bus stops, she can't physically wait the twenty minutes or so on a bus stop, and we'd have to catch two buses out here and*

two back". Others described how they could no longer travel to places or go on days out: *"My wife could not possibly travel. I mean, I can't get her down the drive...it takes two men to get her down the steps. In fact, we don't go out now."* Some family members reported that the patient could no longer drive as a result of their medical conditions, and so it made it a lot harder for them to get around, especially if the family member was unable to drive. Many family members travelling with patients found it difficult, and often impossible, to use public transport.

The family members' time was often taken up visiting the patient in hospital or attending medical appointments. One mother said *"It was busy because I was in [hospital] with her daily after finishing work, trying to find someone else to pick up the little one after school because she was too bad for me to take the little one into hospital to see her"*. This impact was also seen with family members of surgical patients, making long journeys every day to see the patient for weeks after major surgery. One family member described how her son was transferred to another hospital in Manchester, when she was living in Cardiff at the time, and she visited him at least once a week for the five months he was there. The impact of having to come to hospital for treatment was also felt by family members, particularly in dialysis patients where family members accompanied patients to their appointments three times a week.

Other daily issues included problems with child care, often relating to family members having to look after children as well as take on additional responsibilities. Others had to employ child minders or ask friends or family to look after their children as they were too busy to do so. The daughter of an elderly patient with osteoporosis described how she felt she had lost her freedom*: "I have reverted back to how it was when I was bringing up my children. Part of you feels to a certain extent that you are housebound...Sometimes I get frustrated that I can't just go out like I once did and leave post-it notes to say where I am for the rest of the family"*

Family relationships

Relationships amongst the family members were affected in 92 (69%) cases, with increased levels of stress and tension in households.

The most common explanation for a change in family relationships was as a result of the family member feeling that they could not leave the patient, and had to be there to care for them as much as they could (26% of family members). Naturally, this led to family members feeling they spent too much time with the patient, often at the expense of spending time with other members of the family. This was especially true with mothers of patients who had other well children. One mother described the impact of caring for her unwell child on the rest of her children: *"You get wound up and wound up and then the slightest thing...it's hard for*

those two [other children] sometimes because they think that we're sort of favouring her in some ways, helping her more, doing more for her and that kind of stuff. You try to explain but sometimes there are times when there's just no patience left and everybody's shouting". Other family members described not being able to spend time with their children, for example helping them with their homework. Some family members in this study also spent a lot of time worrying about how their other relatives were feeling and whether they were affected by the patient's illness, particularly worrying about the effect on children.

Not surprisingly, 32 (24%) of the family members interviewed reported more arguments in the family as a result of the patient's illness. Family members reported that arguments often arose from disagreements about responsibilities. A mother of two sons with Attention Deficit Hyperactivity Disorder (ADHD) said: *"My other half couldn't cope with what was happening so I was left to deal with everything. I dealt with all the claims for the boys, I had to deal with the doctors and all the appointments and his excuse was "I'm working, I'm working"".* Many family members described strained relationships within the household, sometimes as a result of the patient's changing mood affecting others in the family: *"Mood swings, tempers, arguing all the time, screaming, yelling. It affects us all you know. One minute she can be really pleasant to us and the next minute she's screaming down at us and we're shouting saying really horrible nasty things".* In some cases, the arguments stemmed from family members feeling pressure from the patient to look after them, and sometimes feeling unappreciated for what they do.

The partners and spouses of patients acting as a carer found the change of role in the relationship challenging, and many reported a negative effect on their sex life as a result. Others reported a decline in their sexual relationship due to the patient's physical condition, for example a lack of mobility. The wife of one patient described *"Well, we have no sexual activity at all. There's been nothing for the last five years and that has affected me because I was always a very touchy-feely person...now he's totally switched off from it...I'm looking at the man I love and I want to be cuddled. I want affection and he can't show any affection".* For some family members the side effects of the patient's treatment affected their sex life, for example tiredness or low testosterone levels. One family member described how she no longer found her husband attractive as he had put on weight due to his condition. Although many family members described feeling frustrated at the way their sex life was affected, they also understood what the patient was going through, and described how their lack of sex life did not bother them as much as some of the other issues they were facing at the time.

In one case, a mother described a feeling of hate towards her diabetic teenage daughter because of the way her illness has affected the family, saying that she sometimes wishes her daughter was dead: *"I want to walk out and not come home again, give her up, pass her onto*

someone else, and there have been times when I feel I can't deal with her, I can't and she won't listen to me. She hates me, I hate her, I can't talk to her. You know, if somebody said to me "yeah but she'll be gone one day" and I know it's terrible to say, but I think "yeah, I'll get a rest then". Three family members interviewed said they had thought about leaving home, one family member described her family as being "torn apart", and another described his mother's illness as the reason for his marriage breakdown: *" I've also had some personal difficulty in my marriage...stemmed from the fact that we are a close family in many ways and my sister and I were trying to support my mum and dad to the highest degree and I got the balance wrong, and as a result my wife didn't feel wanted, then someone else came along".*

Sleep and Health

Eighty seven of the family members (67%) reported a negative impact on their sleep and health as a result of having an ill relative. The two main reasons for loss of sleep were worry (32%) and having to wake in the night to help the patient (38%). Many family members were unable to sleep well as they would lie awake thinking about the patient, or what would happen in the future. One family member said *"I'm on antidepressants, I can't sleep without them...other than that I'm awake hours worrying. I'm so wound up".* Others worried that the patient would become unwell, or even die in their sleep, and so would wake up to check on them, with several family members describing how they would listen out for the patient breathing to make sure that they were alive. Other family members lost sleep as they had to physically get up in the night to help the patient, for example with personal hygiene needs, or to administer medication. The father of a patient with epilepsy described how he would wake up early to check on his son: *"I don't sleep too good at night because of his epilepsy...if [patient name] is in the shower, I know it sounds daft because he's nineteen years old, but if he's in the shower or the bath I make a point of being upstairs because I've got this thing that he could be in the bath and have a fit"*. The wife of a patient with bipolar disorder described the pattern of her sleep loss: "*As soon as he wakes up, my room is next door to him and I always listen...if he does not sleep he falls ill and that's one big sign...so when he does not sleep, I don't sleep myself*". Other less commonly mentioned reasons for sleep loss included staying up late to research about the illness on the internet or being unable to sleep in their usual bed, for example having to sleep on the settee whilst the patient is unwell. Many family members also described the effect of sleep loss on their lives the following day, often affecting them at work, or putting strain on their relationships. One family member even described feeling guilty for sleeping, in case that patient needed her in the night.

Some family members described a decline in their own health, and several had been diagnosed with depression. The mother of a 19 year old patient with schizo-affective disorder described how trying to look after her other daughter was affecting her sleep: *"I was living off*

an hour, or a maximum of two or three hours sleep a night and this was prolonged for eighteen months and in the end sleeping tablets weren't even working...even antidepressants don't help...just total anxiety all the time, the panic attacks I was having because I had to juggle everything". Some felt that the stress of the patient's illness worsened their own medical conditions, and brought on symptoms. Often, the family member would ignore their own medical condition or the symptoms they were suffering to concentrate on the patient. Others expressed concern about what would happen to the patient if they became unwell and could no longer look after the patient. Some family members even described feeling close to breaking down.

Holidays

A variety of problems associated with going on holiday were reported by eighty two (62%) of the family members in this study. The most common problem was not being able to go on holiday at all (31%), mainly because the patient was too unwell, but also because of the timing of regular hospital appointments and worrying about food abroad. The wife of a patient suffering from a number of medical problems including urinary retention, neuropathy and diabetes, described their reasons for not taking holidays: "*We used to go but we don't go anywhere now...because my husband has a back problem and had an operation so he finds it difficult to go very far with regards to driving and he prefers to sleep in his own bed now*". Another family member described having to split her family into two and take two separate holidays; one which was suitable for the patient and one which was fun for the rest of the family. Another reason given for not going on holiday was the family members themselves being too tired to go as a result of looking after the patient. Some family members described how they could no longer afford to go on holiday, often because of the patient or family member being unable to work. Family members also talked about being unable to go on holiday on their own, or with friends, because they felt they could not leave the patient at home. One family member described the impact her husband's illness has on the holidays she used to enjoy with friends: *"The last holiday we went on was in 2005...I've not left him at all to go on holiday myself...maybe now in the next few months now that he's getting better maybe I can think of that but I can't see myself going on holiday because...this condition is very unpredictable, so it affected my holidays as I just don't go on holiday".*

Nineteen (14%) of the family members experienced problems with travel, which affected planning holidays or sometimes prevented them going on future holidays. Most of the problems with travelling related to the patient finding the travel difficult, for example having a lack of mobility, or the need to be near a toilet. Most of the problems arose from travelling long distances on aeroplanes, often restricting the types of holiday the family decided to take in the future. The mother of a child with a duplex kidney system described her problems with

travelling on holiday: *"Getting on a plane where you know your child will disturb other passengers and where she needs the toilet lots and she's up and down the alleyway...it's that embarrassment and fear*". Other family members described problems with disabled traveller services, for example the wife of an elderly patient with co-morbidities including Raynaud's and osteoarthritis who booked a mobility buggy at the airport: *"It was bucketing down and I was trying to get [patient name] into a wheelchair, two cases and two hand luggages. He fell in the car park and the driver didn't seem bothered at all. We didn't understand why he didn't come up and look for us. Instead of that we had to walk around looking for him"*. One family member described the expense of buying different suitcases each time they went on holiday, trying to find one which was comfortable for the patient to use. The impact of travel restrictions on other members of the family was also felt: *"Flying is not an option for us, certainly at the minute because [patient name] requires so much equipment. It would be impossible but not being able to access certain types of places...impacts on [sibling name], our eldest son because we're not able to do all the big theme parks*".

Twelve (9%) family members interviewed reported having to cancel holidays they had already booked, often losing money, and many had problems obtaining medical insurance for the patient to be able to travel. The husband of a patient who had recent surgery for talonavicular arthritis described how they had to cancel a holiday at the last minute: *"She was on a stand by thing and they called her in to have the operation and I got in touch with the insurance company and there was no chance. So we lost all the money".* The mother of a 29 year old patient with colitis explained how her daughter's condition affected her planned holiday on two occasions: *"Well we were going to Austria, me and my husband, and then [patient name] was admitted so we cancelled it and then we thought we could go later on in the year so we rescheduled but then [patient name] took a turn for the worse so we cancelled it completely then, so we never went".*

Many family members described having to change the type of holiday they take because of the patient's condition. For some, they had to change the type of accommodation they stayed in, for example staying in a ground floor room in a hotel. Others stopped going to sunny holiday destinations because of the patient's condition. The mother of a patient with eczema said "*I've often thought about going on holiday abroad but because of his skin condition we can't. He can't go out in the sun cos it flairs up worse in the sun, so we can't go on holidays abroad".* Some family members felt that they had to choose holiday destinations closer to home, to avoid long distance travel, and many changed the type of holiday they go on, because of the patient. The wife of an elderly patient with sarcoidosis explained their altered holiday plans: *"When we say change our type of holidays, we used to go abroad a lot, now to go abroad the insurance is so astronomical that its ridiculous and we won't be able to*

get there unless we pay more for the insurance than the holiday. So we're sticking to caravanning and staying in this country". Many family members worried about problems with future holidays because of the unstable symptoms associated with some medical conditions. The family member of a patient with Crohn's disease said: *"We're going to America so there's no way I should have chosen that. We booked this back in May, was before this flared up again so there's no way I would have gone that far away had I known that this was brewing up. Well we probably wouldn't have gone away to be honest, I mean we might have gone locally somewhere but we wouldn't have made a big commitment to go abroad"*. Other holiday problems included family members reporting not enjoying their holidays because of worry about the patient, or describing problems relating to the patient's medical condition whilst they were abroad.

Support and medical care

Eighty one (61%) of the family members of patients described the effects of lack of support from friends and other family members. They often felt that other people didn't understand what they were going through and many found it difficult to talk about the patient's illness, often through embarrassment or a lack of knowledge about the condition. One family member, whose mother suffered from angina, described her frustration at the lack of support she received from the rest of her family: *"Half the time people don't want to know...I've got a shed load of brothers and sisters and none of them visit. You feel that they are selfish and they load it all on you"*. The husband of a haematology patient described how he could no longer rely on support from his friends*: "I suppose it's a man thing really but all my mates disappeared, you turn your back and all your friends are gone. The nurses offered me a support structure but I said "no, my friends [will help]". I named some people, but unfortunately they all just vanished for six months, so I dealt with everything myself...basically they vanished"*.

Another family member described how she didn't like talking about the patient's condition: *"Talking about it is difficult, very, very difficult because when you talk about it you've got to confront the reality of it, but you don't talk about it and sort of go into...it's not denial, but it's like...when people know about it, that's difficult"*. The family members who were embarrassed to talk about the patient's condition were often when the condition had a stigma attached, for example mental health conditions, or infectious diseases such as HIV. The family member of a patient with HIV described the effect of keeping the illness a secret from the rest of the family: *"Nobody apart from medical people know...we made a decision that we weren't going to tell anybody, it's the safest way. But that has had a bit of an effect on the children as they have to keep the secret".* One family member described how he told the pastor of his church

in confidence about his daughter's diagnosis of HIV, but that the pastor told other members of the church and this affected people's attitude towards the family.

Often, the family members needed to remind patients to take medication and several described being affected by issues relating to the patient's medical care including the timing of hospital appointments and not being given enough information about the patient's condition. The mother of a 17 year old patient with acne explained: *"He's had to have a routine with his skin and being the age that he is, a teenager, you want to think they're grown up and adult, but actually they're still very childlike and you have to remind him to take the tablets and cream his skin...every day. It adds a little bit extra to your day having those extra things on your mind- it can be quite stressful. Also you feel you have to remind him because you're the mum and you know he will forget, but then he resents you reminding him".* Family members also described feeling nervous about hospital appointments, feeling like they do not know what is happening to the patient and finding it difficult to deal with the symptoms of the disease, particularly when the disease was unpredictable or when symptoms were severe. The mother of an adult patient with epilepsy described how she had to accompany the patient at all times when she left the house, in case she had a fit.

Many family members said that they did not like asking for support from family, friends or welfare agencies. One mother described how she felt uncomfortable with asking for benefits she was entitled to: *"The shame of having to realise we are entitled to certain benefits like free school meals...but we don't take up that option because there's still that shame thing around it for us".* Others who did ask for support from welfare agencies often found it difficult to get hold of. The partner of a patient suffering from sleep apnoea described their struggles with obtaining benefits: *"Because the Social for him, have turned around and told him basically to naff off and that there's nothing wrong with him. They're not giving him the money that other people who have similar diagnosis and they've had every single penny given to them".*

Work and study

Sixty nine (52%) of the family members described how their own work or study was affected as a result of having an unwell relative. Commonly, family members had to take time off work to look after the patient or attend medical appointments with them. One family member described the effect of his wife's illness on his work: *"Well the number of hospital appointments we've got to go to...I'm lucky the way I work, I've got flexi time but I know it does cause issues...I suppose my staff see me supposedly swanning off early, I think they understand but I still feel, you know, another form of guilt for that as well".* Many family members described using up their annual holiday entitlement for hospital appointments and to look after the patient, leaving them with no holiday left for relaxation. Several family

members talked about saving up their holiday entitlement in case the patient became unwell and they needed to take time off to look after them. In some of these cases, the family members ended up losing out on taking holidays. Others talked about the difficulties they had with colleagues and bosses understanding their situation and allowing them to take time off work, particularly when they worked as part of a team. As well as taking time off work, some family members talked about having to change the hours they work or even change their job or career. One family member said: *"Our expectations were that we would continue in our careers and I would do some further study...now my husband doesn't work at all and I gave up the career I was in and started working part time in a job...I gave up a job with a very good salary and my husband gave up full time work".* Another described how her son's bipolar disorder affected her future career: *"Since I got my doctorate, my PhD, I was thinking about going abroad to get an international job or at least to volunteer...but I cannot take him behind me all the time. I am concerned about him, you know being stable. I will always not be happy, always thinking "what is happening, what is happening", so that has limited me"*.

In 12 (9%) cases the family member gave up their job completely. This had a huge financial impact on the families. The husband of a patient with severe depression said: *"I just didn't have the time [to work]. There are so many appointments to go to and obviously my wife needed care, it got very difficult to carry on [with work] really. Work were ok at the start, they tried to help although they weren't really that helpful in terms of having time off...we had to get a bit fussy and it was difficult as my wife needed me at home"*. Often the decision to give up work came from feelings of guilt by the family member that their relative was at home by themselves with no one to look after them. Most family members accepted the fact they had to give up work, but others regretted it. One family member said *"I would say [I gave up work] because she needed me more at home but I enjoyed my work far better than being at home".* As well as giving up work, some family members decided to take an early retirement to be able to care for the patient. Often in the situations when family members gave up work this was compounded by the patient being unable to work as well, causing huge financial problems for some.

For those family members who were able to continue with work as normal, many described having to juggle their work with their family life, and having to return home from work in the evenings to look after children and do all of the housework. Others felt they could not do their jobs properly as they were constantly thinking about the patient, worrying or feeling tired. For some family members, the source of future work was a constant worry. One family member talked about her plans for work in the future: *"Hopefully a job placement would be understanding that if you had to go...you can go. I don't know how a work placement would be...I haven't really crossed that path yet, but when it comes to it, that's something I will have to investigate more"*.

As well as work being affected, family members who were studying also reported their studies being affected. One family member described how she lost interest in her studies after her son became ill and so gave it up. Others had to postpone their studies to look after the patient, or due to emotional stress. One family member described exams being affected as he could not study effectively for them. Another said: *"There were other things...after my husband had his diagnosis...there were some studies I was thinking of pursuing but after that I sort of put it aside and said "let's see how this goes", but in the middle of all that, trying to cope with all the pressures, I've just put it aside".*

Financial impact

The financial impact of illness on the family (reported by 68 family members; 51%) was great, with 35 (26%) participants reporting having to spend money on items relating to the patient's illness, for example mobility aids or clothing. The mother of a teenage daughter with diabetes described her financial problems: *"I can honestly say that it's so much harder to buy healthier than it is to buy junk food...and being unemployed and not working...you basically take the cheaper choice...for her to eat a full healthy diet is impossible...I couldn't afford to do it*". Other extra expenses reported by family members included buying an extra refrigerator to store medication, buying expensive skincare products for the patient, and making alterations to the family house, for example installing a stair-lift. Some family members reported that they required these extra items but could not afford them, so had to live without. The wife of a patient with multiple sclerosis explained: *"You live to your means and there are things that we need to have done in the house, for instance the en suite bathroom has got quite a deep shower tray so on bad days [patient name] finds it difficult to step up into the shower without catching his foot, and so the bathroom needs work".*

Some family members reported extra expenses relating to the patient's condition, for example the cost of having to do more washing. The mother of a child with urinary problems said: *"the washing machine would break down every six months because we were using it so much because of her health needs...it's the costs associated with her needs"*. Others reported the costs of travel to the hospital with the patient for appointments, or to visit them in hospital. These extra costs often arose from patients being unable to use their usual forms of transport, so had to pay for a taxi, for example. One family member described the impact of her mother's illness on her finances: *"Mum's got a mobility car but I drive the car. I'm the only driver and the responsibility is down to me to pay for all the fuel. I can't afford to do it...she says "I'll give you something towards the petrol" and she'll give me five pounds when I've already spent twenty pounds so it leaves me short of money".* Those family members who talked about the costs of hospital visits also commonly talked about the cost of hospital parking, and one family member described how she bought a new car which was more

efficient to try to save money when travelling to hospital three times a week for her husband's dialysis. The husband of a patient with psoriasis talked about the costs of her treatment: *"Travelling to and from here every day...[patient name] goes through a tank of petrol, especially coming here every day for the sun bed treatment. Then there's clothes especially when she was using that tar stuff. I mean that stained her clothes so I mean virtually every year she had to have a new wardrobe when she was using that stuff".* Those family members who lived in England also reported the high cost of prescriptions. This was not applicable to those in Wales where prescriptions are free. In some cases, the extra costs associated with the patient's illness were unexpected and often hidden. The elderly mother of a female patient with colitis explained how she often looked after her daughter's children whilst she was unwell, but didn't want to put any burden on the patient by asking for money, so bought new sets of clothes for the children herself, including their school uniform.

Often, as mentioned under the theme "Holidays", family members were affected financially when planning holidays. This included the high cost of travel insurance for the patient, having to cancel holidays and losing money, and going on more expensive holidays. One family member described: *"Obviously we can't get decent insurance and on several occasions we've had to cancel holidays...for example we arranged to go to Rome for the [rugby] international...she was taken ill again and had to come in here [the hospital] and I lost the money from that"*. Another theme already discussed which relates to financial impact is "Work and Study". Both family members and patients having to reduce their working hours, or give up work completely had a huge financial impact on the family. The husband of a patient with chronic pain described the impact of his wife having to give up work: *"The ability for her to work, the financial implications, having to think constantly about what you're going to do and how you're going to do things, be it shopping, decorating or even the most mundane things".* As a result of the financial impact reported by the family members in the study, many felt that they had to think more carefully about the way they spent their money. For one family member, he felt frustrated as, because of his wife's illness, he no longer got the opportunity to spend his money as he would like, for example on holidays or days out. In contrast, the partner of a patient who had been recently diagnosed with HIV described how he had developed a "live for the day" attitude since the diagnosis, and had lost between £40,000 and £50,000 gambling over the previous two months.

Social life

Lack of money and feeling the need to leave social events early to look after the patient were amongst the many reasons for the impact on social life reported by the family members (49 family members; 37% affected). The mother of a child described how her child's ADHD had an impact on her social life: "*We have to pay babysitters seven pounds an hour to get*

anyone to babysit and we don't have any friends and family who do it on a regular basis. I suppose we don't like to ask because we know what a handful she is, so it's financially difficult to have a social life because we're paying a lot of money...but by about ten o'clock we're just too tired to do much anyway". Others described feeling that they had to come home from social events early to look after the patient, for example to put them to bed. Many family members described a general decrease in their social life, and gave a variety of reasons for this. The main reason given was that family members did not feel comfortable being away from the patient for a long time, or that the patient was too unwell to join in with social activities. One family member said: *"Because he doesn't feel like it, because he's tired, and because sometimes it's just easier to stay in...I mean we do try and make an effort now and then... but it's not regular, not even once a month now.* Another said *"My social life has been curtailed...It's not so much that my wife wants me to be near her at all times, a lot more I don't like being away from her for any length of time in case anything crops up"*. Often, family members described variations in their levels of social activity depending on the health of the patient. Some family members reported cancelling planned social activities as the patient was too unwell to go, or the family member felt they could not leave them. Others only socialised locally so that they were not far away if the patient became unwell.

Eleven (8%) were concerned about how strangers would react to their relative's medical condition, especially when the condition was visible, for example relatives of dermatology and wound healing patients. The wife of a patient with multiple myeloma described her concerns: *"we used to go out, once or twice a week, or have a meal or something but now we can't do it because with the treatment he doesn't eat properly so we think why go out and pay all that money if he's not going to eat it, so he won't go, and he's lost a lot of weight so we don't want people to see him".* Family members reported feeling uncomfortable and angry about strangers staring at the patients and sometimes making negative comments. They often felt this was because of ignorance about the patient's condition, for example thinking eczema was contagious. Some family members also felt embarrassed to go out of the house with the patient, or felt that the patient was too embarrassed to go out. As well as the reaction from strangers, some family members reported a negative reaction from their family and friends. One family member reported how her friends found it very difficult to understand her husband's diabetes, and despite telling them that he would need to eat regularly and at set times, many would ignore her requests when hosting meals or planning days out. Another family member described how other parents reacted to her daughter's diagnosis of epilepsy: *"She used to go out for tea to different children's houses during the week...then they found out about her epilepsy and she was dropped like a hot potato by the parents. She was no longer invited out for tea or anything. I found that hurtful because she was well...one mother said to me that she had a healthy daughter and didn't want her to pick it up, or catch*

it from [patient name]". Family members also reported having to plan their social activities more carefully, often planning their social lives around the patient.

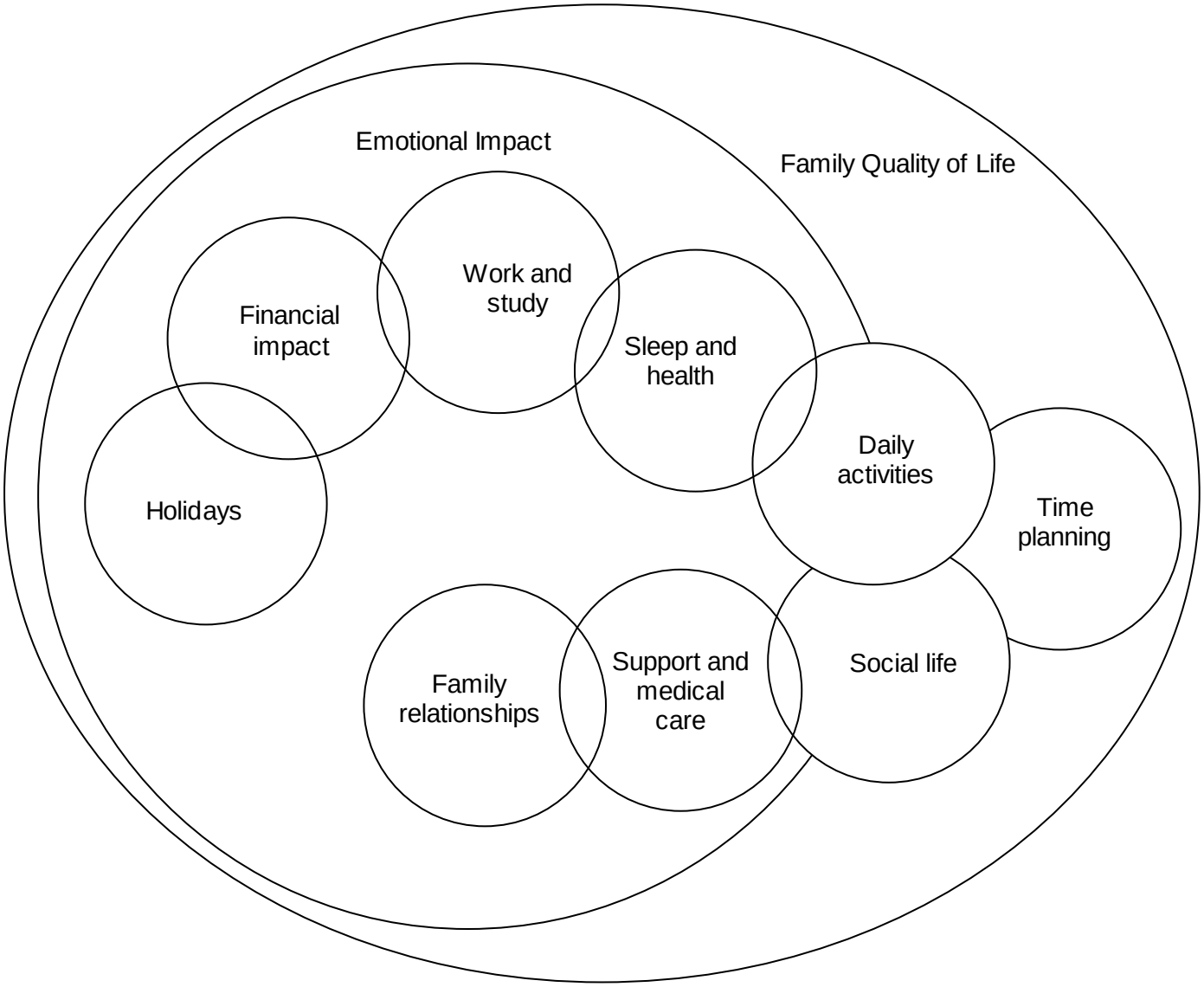
Time planning

Nineteen (14%) of the participants talked about difficulty in being able to plan their time effectively because of the patient's illness. Reasons included having to attend medical appointments at short notice and the unpredictability and worsening of patient's symptoms. One family member said: *"I get really frustrated and a bit angry, it's very unfair of me but I don't seem to be able to plan anything anymore. You know, if someone says "Would you like to come?" and I say "I'll let you know", because I know damn well that I'll probably have to let them down if I say I'm going".* Other family members said that they no longer plan things months or years in advance, such as holidays, and many reported that they had lost any spontaneity in their lives as everything had to be planned to precision. Many family members described how a lot of time and effort had to go into planning days out, and as a result some did not feel they could be bothered to make plans. One family member said *"We never plan anything anymore because if you plan something, something will always go wrong. If he [the patient] is alright then we just get up and go".* Examples of activities which family members found difficult to plan included holidays, days out, social life, transport to hospital appointments, family activities, shopping and daily activities, and visiting other members of the family. Several family members commented that they felt they had lost their freedom.

Relationships between themes

Although each of the themes and subthemes in the study measure individual concepts, relationships were identified by family members between many of the themes. In particular, the theme "emotional impact" was related to all of the other nine themes, apart from "time planning". In turn, the theme "time planning" was related to "daily activities" and "social life". Figure 3.2 shows the relationships between the ten themes identified. This shows that an impact on one area of a family member's life may be related to an impact on another. For example, family members reported that a lack of sleep (sleep and health) as a result of feeling worried (emotional impact) during the night affected their work the next day (work and study). A lack of support from other family members (support and medical care) caused tensions in the household (family relationships) and stress (emotional impact) for the family member caring for the patient. These relationships reflect the multidimensional concept of quality of life, and demonstrate the widespread and varied knock on effects of illness on family members.

Figure 3.2: A model of family quality of life informed by the identified themes, reflecting between themes the relationships from the family members' perspective



Positive effects

As well as the negative effects on family members' quality of life reported above, a small number of positive effects were also identified by family members during the interviews. Seventeen (12.8%) of the family members' interviews managed to identify one positive effect of the patient's illness on their life, and these positive effects were only identified under the "emotional impact" and "family relationships" themes. In these positive examples, family members described relationships within the family improving as a result of the patient's illness, and often they described becoming closer as a family or pulling together to support the patient through their illness. One family member said: "*Whilst some would say "I'm not putting up with this anymore" and walk out, you think you can't, can you? Have to battle*

through, sort of thing. So I suppose it's making our relationship stronger cos it's making us work through things. But there are times when it does affect us". Other family members described overcoming the "challenge" of the patient's illness and making them realise how precious their life is. The majority of these positive comments were found in the "family relationships" theme, and eight of the ten themes contained no positive comments from family members at all. This shows an overwhelming negative impact of illness over the majority of areas of family members' lives.

One of the purposes of developing a generic family instrument is to identify the areas of family members' lives that are impacted in a negative way so that support can be put into place to improve their lives. Including positive items in the instrument would have not represented a true picture of the impact of illness on families, as the positive effects were so limited and rarely reported. To be able to use the instrument in disease assessment, all of the items must be measuring the same, negative, trait and be scored in the same direction (Ware et al. 1995). Adding positive items into the instrument has the potential to cancel out, or dilute the negative effects reported by family members. The inclusion of positively worded items in a quality of life instrument may also compromise patient confidence in the instrument, as the majority of family members interviewed would struggle to identify *any* positive effects of the disease. Therefore, no positively worded items were included in the instrument.

Informal feedback

As well as talking in great detail about the ways their lives have been affected by having an unwell relative, many of the family members also commented on the study, and how they felt about being asked about the impact of the patient's illness. Many of the family members explained that they had never been asked about how the patient's illness affects them before, and others had not ever stopped to think about it. Several of the family members became emotional during the interviews and were often surprised about how emotional they felt. Many of the family members expressed how much they benefited from having talked about these issues, and had not discussed them with anyone else before. There were no negative comments from the family members regarding this study, and many of them commented about how they thought the study was a good idea, and how it might benefit them. Some of the family members said that they would now talk to others more, particularly other members of their family as a result of being interviewed.

Instrument development

The interview transcripts and themes identified were used to develop a quality of life measure for use in family members of patients. The study team gave detailed consideration

to the type of measure, the length, design, structure, layout and scale (Streiner and Norman 2008). Throughout the instrument development process, thought was given by the study team as to the intended use(s) of the new measure and development decisions were made accordingly. For example, the completion time of the measure and ease of use for completion in a clinical setting, and the comprehensive yet simple language used in the measure, which would be understood by all, for example for use in a disease education setting.

The type of measure was considered by the study team, and it was decided that a simple, self-administered questionnaire would be developed. Using a self-administered instrument could potentially help to eliminate interviewer bias, and places less burden on the administrator. The measure would be generic, as the themes identified cover family members of patients from 26 different specialties. In order for the measure to be user-friendly, it was decided that the questionnaire should be short in length, and should take no more than five minutes to complete (tested at a later stage in the study). The instrument should have more than ten items to allow a minimum of one item from each theme identified, but should not have more than 50 items as this could put an increased burden on the participant and professional time especially if it is to be used in clinical settings. It was decided that the items would be constructed as statements rather than questions, as most of the data gathered from family members is made up of statements and examples. Using personal statements, for example using the term "I", would help the family members to identify with the items more easily. It was also decided that the instrument would have a polytomous response scale, so that family members would be able to express their level of agreement with each item, or in this case, the extent to which each item affects their life. The study team decided that each item should be short and concise in length and simple language should be used to make the instrument user friendly and quick to complete. Using short sentences and simple language also reduces the reading ability of the measure, so that it is able to be understood by the majority of the population. A maximum of around twelve words for each item was therefore decided by the study team.

Item generation

The interview transcripts and coding were consulted during the item generation. The study team decided that all ten of the themes, and any of the 196 sub-themes mentioned by greater than 5% of family members interviewed, would be considered for inclusion as items in the new instrument. The aim of this cut off point was to exclude any "outliers" or unusual subthemes which would not be applicable to the generic population, but ensure that the most commonly mentioned subthemes were retained. However, any age, gender and specialty specific sub-themes were retained. or each of the subthemes mentioned by over 5% of

participants, a statement was developed to reflect the content of the theme. The original wording used by the family members was carefully considered, and any commonly used words were inserted into the items. Each item was phrased in a clear and concise way, and the universality of each item was also considered during this development phase; items were designed to be applicable to all ages, relationships to the patient and both genders, and specialty-specific wording was not chosen.

It was decided that the theme "emotional impact" would be represented by statements relating to each of the sub-themes. Those mentioned by over 5% of family members were developed into items and then many were combined or eliminated as they overlapped with others. For example the sub-theme "feeling more emotional" was eliminated as it was included within all of the other sub-themes, which each expressed a different emotion. Most family members who mentioned this theme also gave examples of these emotions. The sub-themes "angry" and "annoyed" were combined to form one item "I feel angry". In the same way, the item "I feel sad" was developed to include subthemes "sad", "depressed", "crying" and "upset". "Worry" formed its own item "I feel worried", as it was the most prevalent emotion and linked with many of the other themes. The sub-theme "pretending everything is ok" was not included as it was unclear as to whether this was a positive or a negative aspect.

Under the theme "daily activities", two of the subthemes relating to eating were combined to form one item "my eating habits are changed". This item covered both "eating out" and "eating a different diet". The sub-theme "caring for patient" formed one item, and the word "burden" was included, as it was a key word used by family members during the interviews. The sub-theme "travel" became one item, and after studying the interview transcripts, the word "every day" was added to the item, as it would distinguish the item from the holiday theme. The sub-themes "takes up time" and "visiting patient in hospital" were combined to form one item. "No alone time" was used to form its own item, with the wording taken from the family member interviews; "it is hard to find time for myself". The sub-theme "stopped going out" was combined with several of the similar sub-themes under "social life" and formed the item "I need to stay at home", which covers both the social and day-to-day aspects of the theme. The item "my leisure activities are affected" was added to represent the examples the family members gave of activities they chose to do in their spare time.

For the theme "family relationships", the subtheme "become more of a carer" was eliminated as it is already covered by the "caring for patient" item. It was decided that the subthemes "argue" and "other family members affected" would be kept as two separate items, and original transcripts were referred back to for wording of these items. The sub-theme "sexual relationship" was retained and developed into the item "my sex life is affected". The original interview transcripts were consulted during the development of this item, as there was

concern amongst the study team that the item could be only applicable to partners or spouses of patients. However, the interviews showed that, whilst this was often the case, there were also examples of parents of child patients whose sex lives were affected, or sexual relationships with family members' partners where the family member and the patient are siblings, for example. Therefore, the item was retained.

Under the theme "sleep and health", three items were developed. The first item was developed from the sub-theme "sleep affected", and the second from the subtheme "health affected". The word "well-being" was also included, as it was one of the key words mentioned by family members under this theme. Thirdly, the item "I feel tired" was developed, as it was reported by many of the family members under the sub-theme "sleep affected", but also in relation to "work and study" and "emotional impact" themes.

As there were so many individual examples mentioned by family members under the theme "holidays", the subthemes were combined to form one item. This is because many of the examples given by family members were considered by the study team to be disease-specific. The one item "I experience problems with holidays" includes all of the relevant examples and sub-themes. Also, the study team identified that not all families would take holidays, and so it was felt that including more than one question under this theme would affect the universality of the items.

Several of the sub-themes were removed from the theme "support and medical care" as when they were converted to items, it was unclear as to whether they should be positively or negatively worded. The sub-theme "support from friends" was felt not to be universal by the study team, and was eliminated. The sub-theme "people don't understand" was combined with "Difficult to talk about disease", and after consulting the interview transcripts, two separate items were formed, which also represented people's reactions to the patient's illness; "I worry about strangers' reactions to my family member's condition" and "I find it difficult to talk about my family member's condition".

The theme "work and study" was made up of several quite specific sub-themes. Therefore, they were combined to form one item, "my work or study is affected". The study team considered that it was appropriate to form only one item, as not all of the family members were employed, so having a single item improved universality and reduced the number of specific items. This was also the case for the theme "financial impact", where one universal item; "my family expenses have increased" was developed to represent the individual, and often specific sub-themes. By far the most commonly mentioned sub-theme under "financial impact" was "buying more", which is well represented by this single item.

Under the theme “social life”, the sub-themes “social life decreased” and “seeing friends less” were considered similar and combined to form a single item. The sub-theme “strangers’ reactions” was already covered by the item “I worry about strangers’ reactions to my family member’s condition”, designed to combine this sub-theme with the sub-theme “People don’t understand”. The theme “time planning” was represented by one item, as many of the examples given by family members were similar and were all encompassed by the item “I find it hard to plan my time and activities”. The word “activities” was added as during the interviews family members gave many specific examples of the activities they now found difficult to plan.

The lengths of the 30 items generated were analysed. Only two of the items were over twelve words in length, containing 13 and 15 words. These items were retained as the wording of both of the items was still considered clear and concise. The items were scanned for ambiguity or complex language by the study team, and none was found.

Addition of utility questions – the rationale

During the interviews, it was noted that many of the family members gave examples of the time they spent looking after the patient, or put numbers to the amount of sleep they lost per night. Being able to measure this information could potentially be a surrogate indicator of the extent of the effect of illness on the family members. It would also add a semi-objective element to the measure and the change could be assessed over time. The interview transcripts were consulted, and three different areas which could be measured by time were identified; hours of sleep lost per night, hours spent caring for the patient and hours spent doing housework. These three themes had already been proven to be relevant to the impact of illness on the family, but adding a utility element to the instrument could increase its potential for use in economic evaluations (Torrance 1987). Each of the three areas were developed into a utility question, asking the family member to specify the number of hours per day they spent caring for the patient, doing housework and how many hours of sleep they lost per night. These three questions were included as a separate part of the developmental instrument.

Instructions and layout

The instrument was laid out over three A4 pages. The first page contained the three utility questions and a set of simple instructions for the user. These instructions were also repeated briefly at the top of the second page, and the 30 items were spread over pages two and three. The instructions reminded family members that this instrument is relating to them and how they feel, not how the patient feels, and asked them to tick a box for each statement. At

the end of the instrument, the words “Thank you. Please check that you have answered every question” were added to help reduce missing data.

Scaling

After considering the possible options for the instrument response scale, a Likert scale was selected as the most suitable for this instrument. This was because the family members in the interviews reported different levels of impact relating to each theme. For example, some reported that their social life was greatly affected across all areas, and others reported that their social life was affected, but just slightly, and gave one example. Therefore, the study team thought it important for the family members to be able to differentiate between these levels of impact, or intensity, when using the instrument. Likert scales allow logical, ordinal progression along the response options, and would allow the family members to subjectively express the extent of the impact of each theme. As some family members during the interviews had reported no impact for some of the themes, and others had reported a high impact, a bi-polar Likert scale (two ends with extreme values) was selected to give the family members a wide selection of response options, with the option of a “not at all” category. Using the guidelines set out by Streiner and Norman (2008) for designing measurement scales, a 5-point Likert scale, labelled as Extremely, A lot, Moderately, A little and Not at all, was developed. The number of response options (“steps”) was carefully considered. An odd number gave the user the option of choosing a middle, or neutral option, and therefore a 5 or 7 step scale was considered. When an instrument is being designed to sum the individual scores to form a total score (as this instrument may have in the future), having a 5-point scale does not result in a significant loss of information (Streiner and Norman 2008), and so for ease of use, and to be user-friendly, a 5-point scale was initially selected. Having a 5-point scale allows for a large range of total scores and therefore can increase the potential for demonstrating responsiveness and sensitivity of the scale. All five points on the scale were labelled to reduce bias towards labelled or unlabelled boxes when a mixture are used (Streiner and Norman 2008). As the aim of the instrument is to measure the user’s perception of frequency/ agreement, it does not matter that vague quantifiers are used to label the response options (e.g. “A lot”)(Streiner and Norman 2008). Although every effort was made by the study team to select adjectives for labels which represent equal intervals, the scale cannot be assumed to be interval (Streiner and Norman 2008) until further tests, such as Rasch analysis, are carried out. In addition to the five response options, the option “Not relevant” was also added to help identify any items which family members do not feel are relevant to them, and to aid with the later analysis of the questionnaire, for example when selecting items to remove.

Recall period

The choice of recall period for any quality of life measure is very important. The ability of users to recall events is vastly overestimated by instrument developers (Norquist et al.), and as this new instrument will be used in family members of patients with a wide variety of illnesses, the disease symptoms cannot be used as a guide for recall period. As the response to questions is likely to be influenced by the patient's health status at the time, and longer recall periods can increase patient burden, a shorter recall period is preferred (U.S. Department of Health and Human Services Food and Drug Administration 2009). As this instrument will also be used to demonstrate sensitivity to change over time later in the development, a short recall period will improve the accuracy of the data, as there will be less change of overlap of recall periods. Therefore, an immediate recall period was chosen for this generic family quality of life instrument. The phrase "at the moment" was chosen as it was used commonly by family members during the interviews. The inclusion of an immediate recall period will allow the measure to be used in family members of patients whose illnesses fluctuate on a day to day basis, as well as those whose symptoms occur less frequently, improving the generic properties of the measure. The information and literature used to decide the recall period was based around patient reported measures (Acaster et al. 2012; Norquist et al. 2012) and there is no similar guidance available for family measures. Therefore, the recall period will be assessed as part of the content validity by family members at a later stage of the study.

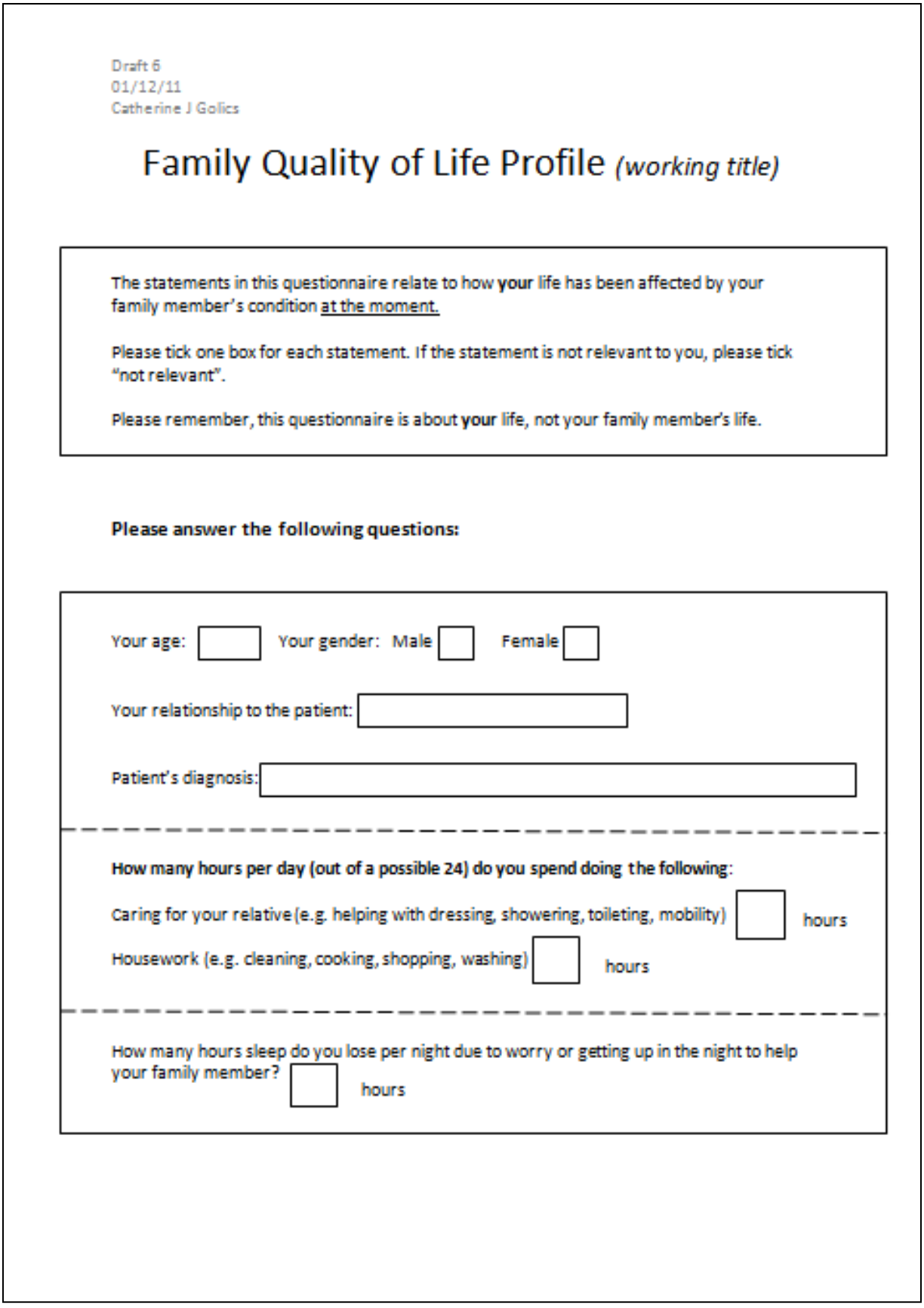
Naming of the new generic family quality of life instrument

After much deliberation between the study team, the new generic family quality of life instrument was named the Family Reported Outcome Measure (FROM). This name complements the commonly used term Patient Reported Outcome (PRO) Measures, which refers to a group of instruments, including quality of life instruments, which give a subjective insight into patient's opinions about their own health (Dawson et al. 2010). As the new instrument is offering the first insight into measuring family quality of life from the family member's point of view on a generic level, the study team felt that this wide and all-encompassing name reflected the wide use potential of the new measure. The name is distinctive, forms a simple acronym, and is easily identified as being a subjective reported outcome measure. Copyright of the measure and its name was established under the copyright law of England and Wales and also recorded at the United States Copyright Office at the Library of Congress, thereby establishing copyright worldwide through the US copyright agreements with many other countries.

Thirty items with a 5-point Likert scale have been developed for inclusion in the preliminary version of the FROM (Figure 3.3). Before this preliminary version is finalised, and developed

into the “developmental version”, the items will be subjected to a content validity study where family members and experts will be asked their opinions on the proposed preliminary questionnaire as a whole. The original working title of the preliminary version, the Family Quality of Life Profile was preserved for the next stage of the study.

Figure 3.3: The 30-item preliminary version of the FROM



Draft 6
01/12/11
Catherine J Golics

Family Quality of Life Profile *(working title)*

The statements in this questionnaire relate to how **your** life has been affected by your family member's condition at the moment.

Please tick one box for each statement. If the statement is not relevant to you, please tick "not relevant".

Please remember, this questionnaire is about **your** life, not your family member's life.

Please answer the following questions:

Your age: ☐ Your gender: Male ☐ Female ☐

Your relationship to the patient: ☐

Patient's diagnosis: ☐

How many hours per day (out of a possible 24) do you spend doing the following:

Caring for your relative (e.g. helping with dressing, showering, toileting, mobility) ☐ hours

Housework (e.g. cleaning, cooking, shopping, washing) ☐ hours

How many hours sleep do you lose per night due to worry or getting up in the night to help your family member? ☐ hours

Subject code:.....
Subject initials:.....

Score:

Family Quality of Life Profile *(working title)*

The following statements relate to how **your** life has been affected by your family member's condition <u>at the moment.</u>

Please tick a box for each statement. If the statement is not relevant to you, please tick "not relevant".

		Extremely	A lot	Moderately	A little	Not at all	Not relevant
1.	I feel worried	☐	☐	☐	☐	☐	☐
2.	I feel angry	☐	☐	☐	☐	☐	☐
3.	I feel guilty	☐	☐	☐	☐	☐	☐
4.	I feel sad	☐	☐	☐	☐	☐	☐
5.	I feel frustrated	☐	☐	☐	☐	☐	☐
6.	I feel tired	☐	☐	☐	☐	☐	☐
7.	My behaviour or personality is affected	☐	☐	☐	☐	☐	☐
8.	I feel I have no one to talk to about my thoughts	☐	☐	☐	☐	☐	☐
9.	I feel a burden of caring for my family member	☐	☐	☐	☐	☐	☐
10.	My housework has increased	☐	☐	☐	☐	☐	☐
11.	My eating habits are changed	☐	☐	☐	☐	☐	☐
12.	My family activities are affected	☐	☐	☐	☐	☐	☐
13.	My leisure activities are affected	☐	☐	☐	☐	☐	☐
14.	My hobbies are affected	☐	☐	☐	☐	☐	☐
		___	___				___

Please turn to the next page

		Extremely	A lot	Moderately	A little	Not at all	Not relevant
15.	It is hard to find time for myself	☐	☐	☐	☐	☐	☐
16.	I need to stay at home	☐	☐	☐	☐	☐	☐
17.	My every day travel is difficult	☐	☐	☐	☐	☐	☐
18.	My time is taken up visiting my family member in hospital or attending medical appointments	☐	☐	☐	☐	☐	☐
19.	My sex life is affected	☐	☐	☐	☐	☐	☐
20.	I argue with my family member	☐	☐	☐	☐	☐	☐
21.	My family expenses have increased	☐	☐	☐	☐	☐	☐
22.	I experience problems with holidays	☐	☐	☐	☐	☐	☐
23.	I find it hard to plan my time and activities	☐	☐	☐	☐	☐	☐
24.	My own health or well-being is affected because of my family member's condition	☐	☐	☐	☐	☐	☐
25.	My sleep is affected	☐	☐	☐	☐	☐	☐
26.	My social life is affected	☐	☐	☐	☐	☐	☐
27.	I worry about strangers' reactions to my family member's condition	☐	☐	☐	☐	☐	☐
28.	I find it difficult to talk about my family member's condition.	☐	☐	☐	☐	☐	☐
29.	My work or study is affected	☐	☐	☐	☐	☐	☐
30.	My relationships with other family members are affected	☐	☐	☐	☐	☐	☐

Thank you

Please check that you have answered every question

DISCUSSION

The impact of a patients' illness on families is widespread and profound and family members are affected in multiple ways across all medical specialties. This stage of the study has identified the major ways in which family lives can be affected by disease and the commonality of issues across all diseases. The impact on the family member is largely independent of the condition the patient suffers from. This is the first study to identify the similar experiences of family members of patients across the whole of medicine, and the unique findings are relevant to all healthcare professionals. Even in those specialties where family members are least affected, they still reported being affected by three of the ten themes, proving that the family impact of disease needs to be considered and addressed in all areas of medicine. Many family members also said how grateful they were to talk about the subject, that they had never been asked about it before and voiced the lack of support they had received in dealing with the effects of the patient's illness.

The percentage of prevalence have been reported for each theme but it is important to note that these percentages should be viewed as guides to each of themes, and not an attempt to formally quantify them. This extension of the theme descriptors is helpful in terms of communicating the importance of these issues. Reporting the percentages of family members affected by each theme adds to the depth and interest of the data, especially as this is a new subject which has not been investigated in this way before, and as there were a large number of interviews. As the interview guide used was semi-structured, and reviewed after the pilot interviews, all family members were given the chance to report on the key themes. The in-depth interviews with family members of patients from a wide range of specialities provides a solid platform for the development of a new generic family quality of life measure, whilst serving as a valuable piece of qualitative research on its own.

Whilst the saturation point was identified as interview number 40, interviews continued well past saturation, as a decision was made to include a minimum of five family members from each specialty. In this situation, there is a disjunction between imposing a saturation point and the decision to include family members from all disease areas. Extending the interview number so far beyond saturation resulted in a large volume of data. Although no new themes emerged between interview number 40 and 133, new examples of each theme were given by family members, adding to the richness of the data. The main implication of continuing so far beyond the saturation point was the time required to transcribe and analyse such a large volume of data, where much of the data could be considered redundant. However, as the qualitative part of the study is an important piece of work in itself, and the new measure was designed to be used in a wide variety of illnesses, it was felt that it was more important to ensure that all specialties were covered in the qualitative work.

One of the difficulties faced when developing the preliminary instrument was deciding upon the recall period for the measure. This was particularly difficult given the number of specialties and disease areas covered and the difference in symptom frequency and type between the different diseases. Using the term “at the moment” was decided as the most suitable option, but it is important to note the impact of using an immediate recall period on the interpretation of results. It is likely that the current mood of the family member could have an impact on the way that they answer the questions and therefore the overall score of the measure. For example, a pre-existing negative mood could intensify the emotions felt by the family member, particularly in a clinical environment. In turn, a pre-existing positive mood, for example if the patient is given good news during the medical consultation could influence the way the family member responds to items in the measure in a positive way; playing down or minimising the negative impact of the illness on their life. Therefore, when interpreting scores from the measure it is important to bear in mind the influence of the current mood of the family member and possible influences on the score. Administering the measure on a number of separate occasions in different environments could help determine the extent of the influence of mood.

SUMMARY

- This chapter provides information on the ways that family members are affected by illness.

- For the qualitative data collection, semi structured interviews with family members of patients were chosen as the method of data collection.

- Sample size in the study was determined by the inclusion of at least five family members from each of the 26 specialties in the study.

- Saturation point was calculated after the interviews were complete.

- Three trial interviews were conducted prior to the start of the study.

- 133 family members of patients from 26 specialties were recruited and interviewed.

- The content of the 133 interviews was transcribed verbatim and coded using NVivo9 qualitative software.

- Ten themes and 196 sub-themes/codes were identified from the interview transcripts.

- The ten main themes included: emotional impact (mentioned by 92% of subjects), daily activities (91%), family relationships (69%), sleep and health (67%), holidays (62%), support and medical care (61%), work and study (52%), financial impact (51%), social life (37%), and time planning (14%).

- The mean number of themes mentioned by participants was six (median= 6, S.D.= 2.03, range 0-10 [max=10]).

- Family members of haematology and genetics patients were affected by the most number of themes, and family members of gynaecology and diabetes patients were affected by the least number of themes.

- Quotes and examples from family members were used to illustrate each theme.

- Relationships between themes were identified and a diagram model of family quality of life was produced.

- Sub-themes mentioned by >5% of family members were developed into questionnaire items. Some items were merged or eliminated.

- Three utility questions were also created.

- The items were formed into a proposed preliminary instrument with a 5-point bi-polar Likert scale.

- The instrument was named the Family Reported Outcome Measure (FROM).

CHAPTER 4

Evaluation of the Content Validity of the Family Reported Outcome Measure (FROM)

INTRODUCTION

Content validity checking helps to ensure that a scale has enough items and covers each of the domains measured (Streiner and Norman 2008). It is important that the items of an instrument are both relevant to, and representative of, the target population or construct, and obtaining good content validity can increase the probability of obtaining high construct validity in later stage validation tests (Haynes et al. 1995).

Content validity involves both qualitative and quantitative methods. The most common method used is gaining the opinion of multiple "expert" judges using formalised scales to assess relevance, representativeness, specificity, and clarity. Using such scales can help to identify any items which need refining or omitting during questionnaire development (Haynes et al. 1995). After potential items were developed from the themes that emerged from interview transcripts and formed into a questionnaire (the preliminary version), the content validity of the items was assessed using a panel of judges (Appendix N). The content validity panel was an important step in the development of the developmental version of the Family Reported Outcome Measure (FROM) and gave the investigators a clear idea of whether the items that had been developed were relevant, phrased clearly and were complete in their wording. The questionnaire instructions, layout and scaling were also critically considered by the panel.

Throughout this chapter, the terms "content validity panel", "panel of experts" and "panel of judges" are used interchangeably, and all refer to the individuals who make up the panel carrying out the content validation. The first version of the FROM made up from the themes from the qualitative phase is referred to as the "preliminary version", the second version of the FROM made up from the results of this content validation is referred to as the "developmental version" and the third version, after item reduction is the "final" version, or the "FROM-16".

METHODS

The content validity of FROM was carried out in two parts; qualitative and quantitative assessments. In the qualitative part of the process, members of a content validity panel were asked to comment on the preliminary version of the FROM as part of a semi-structured focus group set up. In the quantitative part, the panel members, and other experts, were asked to complete a formalised scale to assess each item, and the instrument as a whole. The results from the qualitative and quantitative parts of the process were then used to make changes to the items to ensure that they were relevant to, and representative of, family members of patients. The procedures for both parts of the process are described separately below, followed by the combined results.

PART 1: QUALITATIVE ASSESSMENT

Selection of the content validity panels

The 30 items developed for inclusion in the preliminary version of the FROM were subjected to content validation by a panel of judges. When selecting this panel, it is important to consider the instrument's purpose (Cook and Beckman 2006), and therefore family members were invited to assess the content validity, as well as clinical and academic experts. It was decided by the investigators that a separate panel meeting would be held for family members, to encourage a more intimate and relaxed environment where they would feel more comfortable talking about personal issues. The recommended number of members of a content validity panel varies between 3 and 20 (Grant and Davis 1997).

The first content validity panel (Panel A) was made up of consultants, specialist nurses and academic experts in the field of quality of life assessment. All consultants involved in the study across the 26 specialties were invited to attend the lunchtime meeting, and were asked to identify a clinical nurse specialist from their area who would be also willing to attend. Three academic experts in the field of quality of life were also invited to take part (the co-supervisors of this research study). The Panel A discussion, which lasted one hour, took place in a board room within the University Hospital of Wales and was digitally recorded. Ten of the 26 invited consultants attended the meeting, along with one clinical research fellow, six specialist nurses, one genetics counsellor and three academic experts. Table 4.1 shows the background of the members of Panel A.

The second content validity panel (Panel B) was made up of family members of patients. During the interview stage of the study, family members were asked whether they would be interested in attending the panel meeting, and six agreed. These family members were contacted via telephone and then by post inviting them to the panel meeting, which took place immediately after Panel A meeting in a board room within the hospital, and was also digitally recorded. Three of the six invited family members were able to attend: they were related to patients from general practice, mental health and genetics specialties. Although the numbers for panel B were low, further content validation was carried out with family members during the validation stage of the study.

Table 4.1: The background of the members of content validity panel A

Specialty	Number of panel members	Profession
Ophthalmology	1	Consultant
Neurology	1	Consultant
Infectious Diseases	1	Consultant
Dermatology	4	Consultant Academic expert Academic expert Specialist nurse
Paediatric Endocrinology	1	Consultant
Chronic Pain	2	Consultant Specialist nurse
Wound Healing	1	Research fellow
Cardiology	2	Consultant Specialist nurse
ENT	1	Consultant
Gynaecology	1	Consultant
Urology	1	Consultant
Colorectal Surgery	1	Consultant
Genetics	1	Genetics counsellor
Dental surgery	1	Specialist nurse
Gastroenterology	1	Specialist nurse
Pharmacoepidemiology	1	Academic expert

Procedure

The two content validity panels were held in a private room around a large table to facilitate discussion between panel members. The meeting was chaired by CJG, whose role was to remind panel members about the purpose of the meeting, to present discussion topics, encourage discussion between panel members, and control the group dynamics so that each panel member had the chance to talk and discussion of irrelevant topics was kept to a minimum. In both panels, members were asked to discuss the preliminary questionnaire layout and design, discuss each item in turn, and discuss six potential problems identified by the investigators during the development of the preliminary version of the FROM. Panel A were asked to give their expert clinical opinions, having worked closely with both patients and families, and were asked to consider the details of the preliminary FROM in relation to the illnesses covered by their specialty. Clinical expertise is considered an important attribute for panel members when developing a clinically relevant measure (Grant and Davis 1997). In turn, Panel B were asked to give their personal opinions about how the preliminary FROM related to their experiences as family members of patients with chronic conditions. The results of both panels were then combined and changes were made to the items of the preliminary version of the FROM accordingly. If there was any disagreement between panels over decisions, the investigators would refer back to the qualitative interview transcripts to assist with final decisions.

Each of the 30 items was discussed in turn by both panels. Topics for discussion for each item were influenced by those outlined by Haynes et al. (1995): item relevance, representativeness, specificity and clarity. After each item was discussed in turn, the panel members were asked their opinions on six issues identified by the investigators during item development:

1. Should the FROM contain frequent reminders throughout the questionnaire, reminding the family member that the questionnaire relates to them and not the patient? Several family members talked during the interviews about how the patient's life was affected instead of theirs and had to be reminded regularly to talk about themselves.
2. Should the items containing "leisure activities" and "hobbies" be split to form two separate items? This was debated by the investigators who could not come to a consensus during item development.
3. Should the term "condition", "disease" or "illness" be used in the FROM?
4. Should the term "relative" or "family member" be used in the FROM?
5. Should the order of response categories go from "not at all" to "extremely", or the other way around?
6. Are there any obvious potential translation problems? The FROM has the potential to be translated into different languages after publication and the panel were asked to consider whether they could see any obvious potential language or cultural problems.

PART 2: QUANTITATIVE ASSESSMENT

As a multi-method approach is recommended during content validation (Haynes et al. 1995), quantitative assessment was also carried out. Content validation is often supported by agreement statistics between the panel members (Futrell 1995) and this gives a statistical basis for item removal or rewording.

Procedure

The consultants, nurses, academic experts, family members and other healthcare professionals who were invited to attend the content validity panels meetings were also sent a copy of the preliminary version of the FROM and a questionnaire feedback form (Appendix M). Those who were attending the content validity panels were asked to bring along their completed forms, and those who were unable to take part in the panels were asked to complete their forms and return them to the investigators before the panel meetings. The comments on these completed forms were then used to form some of the discussion items during the panels.

Participants were asked to rate each of the 30 items from the preliminary FROM on a four point Likert scale (1= strongly disagree, 2= disagree, 3= agree, 4= strongly agree) for each of the following criteria:

Language clarity: the sentences and wording should be clear, understandable, straightforward and simple. Phrases and wording should be unambiguous and jargon free and should be understood by someone with a reading ability of 12 years.

Completeness: the sentences should be complete, not broken and should end appropriately.

Relevance: each item should be relevant to the subject area and target population.

Scaling: the scoring system of the FROM is a 5-point adjectival scale. Panel members should rate the scaling system as to whether the response options fit the question, or not.

The four criteria were developed on the basis of recommendations for structural elements in content validity (Ferketich 1991; Grant and Davis 1997; Lynn 1986). Under each rating scale, space was provided for comments or suggestions for change.

RESULTS

The results of the content validity are presented in two parts: quantitative and qualitative. The quantitative results are those from the questionnaire feedback forms rating scales. The qualitative results are the comments made on the questionnaire feedback forms ("written feedback") and the discussions between the members of both panels ("expert panel" and "family member panel"). The changes made to the items of the preliminary FROM to form the developmental version of the questionnaire are then discussed, where both the qualitative and quantitative results are taken into account.

Part I - Quantitative Assessment

The results for the quantitative assessment were based on the returned questionnaire feedback forms (n=23). The results for each of the four rating criteria; language clarity, relevance, completeness and scaling were analysed (Table 4.2). Each of the 23 judges rated all 30 items, so this gave a total number of ratings of 690. Items which had performed poorly under each of the criteria were identified. These were items where ≥ 4 of the 23 judges had selected "disagree" or "strongly disagree" for the criterion listed. The disagreement with these items was considered when deciding which items to amend or retain as a result of content validity assessment.

There was also a small amount of missing data (less than 10% for each question) present in the sample, which is reported at each stage below. Studying the layout of the questions given and the areas where missing data arose, it became apparent that a few of the respondents had not noticed the final page of questions as it was printed on double-sided paper, so had not answered them. With the way this part of the study was conducted, this did not cause a problem as the questions and responses were discussed during the panel meetings, and areas where missing data were present were discussed further and in more detail to identify whether the missing data was random or selective.

Language clarity

When the judges were asked whether the items showed good language clarity, there were 511 ratings (74%) for strongly agreed, 90 (13%) for agreed, 41 (6%) for disagreed and only 6 (1%) for strongly disagreed. Six percent of ratings were missing and therefore were not included in the analysis. The judges' median rating for language clarity was 4 ("Strongly agree"). For language clarity, five items were identified as poorly performing items, with judges disagreeing or strongly disagreeing with the language clarity; item 9 "I feel a burden of caring for my family member" (n=7), item 12 "my family activities are affected" (n=5), item 16 "I need to stay at home" (n=5), item 7 "My behaviour or personality is affected" (n=4), and item 20 "I argue with my family member" (n=4).

Completeness

When the judges were asked whether the items were complete in their wording, there were 531 ratings (77%) for strongly agreed, 90 (13%) for agreed, 21 (3%) for disagreed and only 6 (1%) for strongly disagreed. Six percent of answers were missing and so were not included in the analysis. The judges' median rating for language clarity was 4 ("Strongly agree"). For completeness, only one item was identified as having disagreement, item 16 "I need to stay at home", with 4 of the judges disagreeing that the item wording was complete.

Relevance

When the judges were asked whether the items were relevant to family members of patients, there were 559 ratings (81%) for strongly agreed, 69 (10%) for agreed, 14 (2%) for disagreed and no judges strongly disagreed. Seven percent of answers were missing and therefore were not included in the analysis. The median judges' rating for language clarity was 4 ("Strongly agree"). For relevance, only item 17 "my everyday travel is difficult" was identified as a problem item, with 5 of the judges disagreeing with the relevance of the item.

Scaling

When the judges were asked whether the items fitted well with the response options, there were 497 ratings (72%) for strongly agreed, 90 (13%) for agreed, 48 (7%) for disagreed and only 6 (1%) for strongly disagreed. Seven percent of answers were missing and therefore were not included in the analysis. The median judges' rating for language clarity was 4 ("Strongly agree"). For scaling, two problem items were identified, where judges disagreed that the items matched well with the response options. These were items 8 "I feel I have no one to talk to about my thoughts" (n=5) and 16 "I need to stay at home" (n=4).

The overall level of disagreement with the four criteria was 5%, with 1% of judges showing strong disagreement (Table 4.2), meaning that 95% of the judges thought that the items were written clearly, were complete, relevant to family members and fitted well with the response options. The item that judges most strongly disagreed with the criteria was item 16 "I need to stay at home", which showed disagreement with regards to language clarity, completeness and scaling.

Table 4.2: The judges' ratings (n=23) of the 30 items across four criteria

Judges' response option	Judges' ratings of the 30 items against the four criteria (%)			
	Language clarity	Completeness	Relevance	Scaling
Strongly agree	511(74)	531(77)	559(81)	497(72)
Agree	90(13)	90(13)	69(10)	90(13)
Disagree	41(6)	21(3)	14(2)	48(7)
Strongly disagree	6(1)	6(1)	0(0)	6(1)
Missing	42(6)	42(6)	48(7)	49(7)

Content validity index

As an additional test of content validity, the content validity index (CVI) was applied to the quantitative data (Lynn 1986). The CVI has mainly been used in nursing research, for determination of content validity in the development of multi-item scales rated by multiple judges, and has been recommended over traditional methods, such as kappa, due to its ease of calculation and understandability (Polit et al. 2007). The CVI, which requires a minimum of three expert raters, can be calculated on an item level (I-CVI) and scale level (S-CVI). *The item content validity index* (I-CVI) is calculated as a level of agreement between judges for each individual item. It is calculated by the number of experts giving a positive rating ("agree" or "strongly agree" in the case of the FROM) divided by the number of experts (n=23 for the preliminary FROM), therefore calculating the "proportion of judges in agreement about relevance" (Polit and Beck 2006; Polit et al. 2007). The minimum acceptable I-CVI value for items varies depending upon the number of judges involved. Lynn (1986) gives a minimum acceptable value of 0.80 for ten judges but does not give values for a higher

number of judges. However, the author states that the minimum acceptable value decreases as the number of judges increases (Lynn 1986). For this study, a minimum value of 0.80 was used. Table 4.3 shows the I-CVI values for the 30 items. It can be seen that three of the items fall below the minimum acceptable value of 0.8; item 9 (0.78), item 16 (0.76) and item 17 (0.79). This reflects the earlier results of the four rating criteria, where items 9, 16 and 17 also showed problems with one or more of the criteria. These results will be considered when changes are made to the items of the preliminary FROM to form the developmental version of the questionnaire.

The *scale content validity index* (S-CVI) is defined as "the proportion of total items judged content valid" (Lynn 1986). In the case of the preliminary FROM items, this would be those scored "agree" or "strongly agree" and is calculated by the average of the I-CVIs (Polit et al. 2007). The minimum acceptable value varies depending upon the number of judges involved, but for more than three judges the minimum acceptable S-CVI value is 0.8 (Lynn 1986). The S-CVI for the FROM items was calculated as 0.88, which is an acceptable value to suggest that the scale is content valid.

Test of agreement

The 30 items of the preliminary version of the FROM were rated on a 4-point ordinal scale for four different criteria by 23 judges. It is important to establish the reliability (or the interrater reliability) between the ratings given. This will determine whether the ratings given are consistent between judges, and whether the data produced by the judges' ratings can be relied upon. The most commonly used measures of agreement are kappa coefficient and intraclass correlation coefficient (ICC). Kappa coefficient is more appropriate when the data collected are nominal, whereas interval/ordinal data are best assessed using ICC, therefore ICC was chosen for the quantitative data (Elwyn et al. 2003; Futrell 1995). The intraclass correlation coefficient (ICC) analysis of absolute agreement between all 23 judges showed an ICC of 0.97 ($p \leq 0.001$, CI= 0.94 to 0.99), indicating a high level of agreement between the 23 judges and supporting the content validity of the items chosen for the developmental version of the FROM.

Part II - Qualitative Assessment

The results from the questionnaire feedback forms ("written feedback") and the discussions between the members of both panels ("expert panel" and "family member panel") were reviewed and are presented in Appendix N. The members of both panels discussed each item in detail, along with the proposed questionnaire layout and the six potential problems identified by the investigators during the development of the preliminary version of the FROM.

Table 4.3: The I-CVI values for the 30 items of the preliminary version of the FROM

Item number	I-CVI value	Item number	I-CVI value
1	0.96	16	0.76
2	0.94	17	0.79
3	0.88	18	0.89
4	0.90	19	0.90
5	0.89	20	0.82
6	0.86	21	0.86
7	0.88	22	0.83
8	0.93	23	0.88
9	0.78	24	0.90
10	0.93	25	0.90
11	0.93	26	0.89
12	0.83	27	0.93
13	0.90	28	0.93
14	0.87	29	0.91
15	0.94	30	0.89

The FROM Items

As well as voicing their opinions, the panel members made suggestions for changes to the items. In summary, both panels (referred to hereafter as "the panels") suggested that the grey shading was misleading and should be shaded in the same way for each item. They also suggested that item 7 (My behaviour or personality is affected) measures two different concepts and should be split into two items. The panels disliked the wording of items 8 (I feel I have no one to talk to about my thoughts), 9 (I feel a burden of caring for my family member) and 11 (My eating habits are changed), and they did not think that the wording of these items fitted well with the response categories. The expert panel thought that there was an overlap between items 13 (My leisure activities are affected) and 14 (My hobbies are affected), but the family member panel felt that they could distinguish between the two easily. The panels also disliked the wording of item 16 (I need to stay at home) and the expert panel also disliked the wording of item 17 (My every day travel is difficult). Both panels thought the wording of item 22 (I experience problems with holidays) could be improved, and made suggestions for alternative wording. For item 28 (I worry about strangers' reactions to my family member's condition), the expert panel felt the word "strangers" was not appropriate and should be changed. The expert panel also commented on the layout of the questionnaire, suggesting changes to the instructions and making general comments about the project and the development of the questionnaire.

The FROM Potential Problems

The two panels also discussed the six potential problems identified by the investigators during the development of the FROM:

1. *Should the FROM contain frequent reminders throughout the questionnaire, reminding the family member that the questionnaire relates to them and not the patient? Several family members talked during the interviews about how the patient's life was affected instead of theirs and had to be reminded regularly to talk about themselves.*

The written and verbal feedback from both panels concluded that it was not necessary to have reminders for every item, and that a reminder should be placed at the top of each page.

2. *Should the items containing "leisure activities" and "hobbies" be split to be two separate items? This was debated by the investigators who could not come to a consensus during item development.*

The results of the written feedback and the expert panels felt that there was a large overlap between the two items (13 and 14), and that one should be dropped. However, one member of the expert panel pointed out that it depended upon the individual's interpretation of the two terms. Therefore, the family member panel were asked about their views on the two items, and felt that they should remain separate as they thought of leisure activities and hobbies as two different things.

3. *Should the term "condition", "disease" or "illness" be used in the FROM?*

Both panels and the written feedback agreed that the term "condition" was the best wording to use in the FROM.

4. *Should the term "relative" or "family member" be used in the FROM?*

The expert panels and the written feedback had mixed opinions about this and did not come to a consensus, however the family member panel felt that the term "relative" was too distant and that "family member" should be used instead.

5. *Should the order of response categories go from "not at all" to "extremely", or would the opposite be more appropriate?*

Several members of the expert panel felt that the order of the response categories should be reversed, starting with "not at all" and ending with "extremely", with "not relevant" being the final category. One family member from Panel B felt that the ordering of the categories was acceptable, but the others did not have an opinion on this subject. Some members of the expert panel also suggested having three response options instead of five, but they were informed that this will be decided as a result of item reduction in the subsequent stages of development of the FROM.

6. *Are there any obvious translational problems? The FROM has the potential to be translated into different languages after publication and the panel were asked to consider whether they could see any obvious potential language or cultural problems.*

Both panels commented that the language used in the items was pitched at the correct level, and the expert panel identified several potential translational issues. These included the fact that some cultures may feel that caring for an unwell family member is a duty, and that the family member is not entitled to feel any negative emotion. Potential problems with translating the response option labels were also identified.

Changes to the Preliminary version of the FROM Resulting from the Content Validation

The changes to the preliminary version of the FROM as a result of the content validity panels are summarised in Table 4.4.

The panel's views were that using grey shading on alternate items emphasised these items more than others. Consequently the grey shading was altered so the space in between each item was shaded. This meant that the same emphasis was placed on each item. The font type and size remained unchanged, as the panels felt that they were acceptable. There were mixed responses from the panel members about the use of examples in the items, and so no examples were added as the investigators felt that examples in items may include response restriction and leading the family member to respond in a certain way. Item 6 (I feel tired) remained unchanged; the expert panel asked whether the question meant physically or emotionally tired, but the investigators came to the conclusion that it did not matter which, and that both were applicable to the item. Item 7 (My behaviour or personality is affected) was split into two separate items as both the family members and experts felt that two separate concepts were being measured. This item also scored lowly in the written feedback for language clarity, and therefore an improvement was made by splitting the item. Item 8 (I feel I have no one to talk to about my thoughts) scored poorly during the written feedback for scaling, and this was reflected by both panels. Therefore, the wording of the item was changed to "It is difficult to find someone to talk to about my thoughts", as it was considered that this version of the item would be a better fit with the response options. Item 9 (I feel a burden of caring for my family member) had a low I-CVI and scored poorly on both language clarity and scaling during quantitative feedback. The two panels also debated about the item and some of the expert panel disliked the term "burden". The family member panel were split in their opinion of the word "burden", however as it originated from the original interviews with family members, "burden" was retained but the item was rephrased to read "I feel that caring for my family member is a burden" to improve the language clarity.

Table 4.4: Changes made to the preliminary version of the FROM as a result of content validation

Preliminary FROM	Developmental FROM after content validity panel meetings
Grey shading used for alternate items	Grey shading used between every item
No examples were used in the items	Unchanged
Item 1: I feel worried	Unchanged
Item 2: I feel angry	Unchanged
Item 3: I feel guilty	Unchanged
Item 4: I feel sad	Unchanged
Item 5: I feel frustrated	Unchanged
Item 6: I feel tired	Unchanged
Item 7: My behaviour or personality is affected	Split into two items: 1. My behaviour is affected and 2. My personality is affected
Item 8: I feel I have no one to talk to about my thoughts	It is difficult to find someone to talk to about my thoughts
Item 9: I feel a burden of caring for my family member	I feel that caring for my family member is a burden
Item 10: My housework has increased	My housework is increased
Item 11: My eating habits are changed	My eating habits are affected
Item 12: My family activities are affected	Unchanged
Item 13: My leisure activities are affected	Unchanged
Item 14: My hobbies are affected	Unchanged
Item 15: It is hard to find time for myself	Unchanged
Item 16: I need to stay at home	I feel the need to stay at home
Item 17: My every day travel is difficult	My every day travel is affected
Item 18: My time is taken up visiting my family member in hospital or attending medical appointments	Unchanged
Item 19: My sex life is affected	Unchanged
Item 20: I argue with my family member	Unchanged
Item 21: My family expenses have increased	My family expenses are increased
Item 22: I experience problems with holidays	I experience problems with going on holiday
Item 23: I find it hard to plan my time and activities	Unchanged
Item 24: My own health or well-being is affected because of my family member's condition	Unchanged
Item 25: My sleep is affected	Unchanged
Item 26: My social life is affected	Unchanged
Item 27: I worry about strangers' reactions to my family member's condition	I worry about peoples' reactions to my family member's condition
Item 28: I find it difficult to talk about my family member's condition.	Unchanged
Item 29: My work or study is affected	Unchanged
Item 30: My relationships with other family members are affected	Unchanged
Opening statements containing the phrase "has been"	Opening statements replaced with the word "is"
Opening statements containing the word "tick"	Opening statements containing the word "mark"
Use of the term "family member"	Unchanged
Use of the term "condition"	Unchanged
No reminder at the top of each page of the questionnaire	A statement at the top of each page of the questionnaire reminding respondents that the questions relate to them and not to the patient.
Response categories ordered "extremely" to "not at all"	Response categories ordered "not at all" to "extremely"

The tense of item 10 (My housework has increased) was altered to match the other items, instructions and response options which are written in the present tense. Item 11 (My eating habits are changed) was changed from “changed” to “affected”, as the expert panel and written feedback were unsure as to whether this item was relating to positive or negative changes, and they felt that the wording did not fit well with the response options.

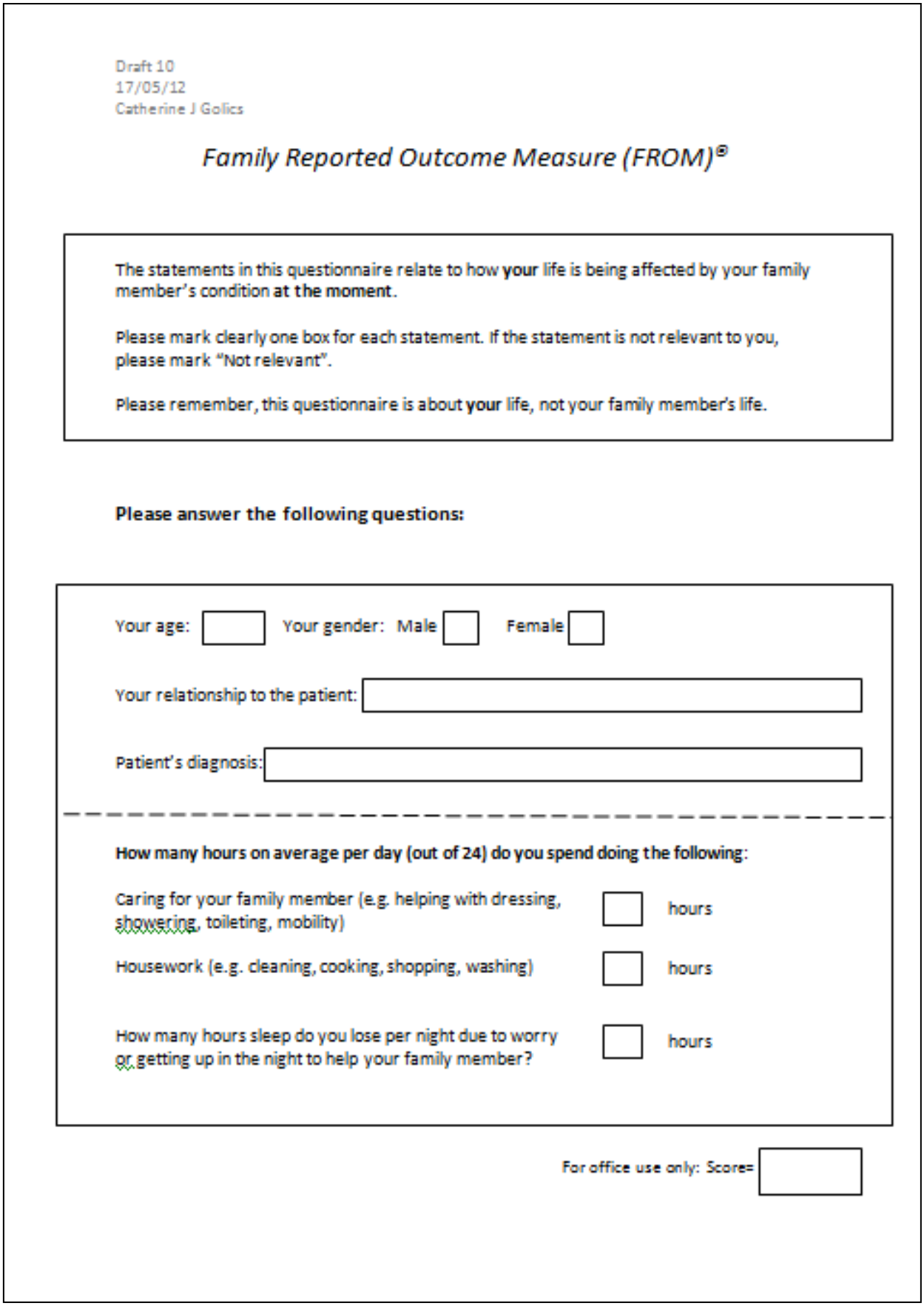
Although there was much debate between the expert panel regarding the wording of items 12 (My family activities are affected), 13 (My leisure activities are affected) and 14 (My hobbies are affected), and whether there was a difference between the item concepts, the family member panel felt that they could distinguish between them and that they were valuable, and so all three items were retained and unchanged in their wording. Item 16 (I need to stay at home) was changed to “I feel the need to stay at home”, as quantitative and panel feedback suggested that the item was incomplete and showed poor language clarity. Item 17 (My every day travel is difficult) was changed to “My every day travel is affected”, to improve the language clarity. Although some of the members of the expert panel felt that this item was not relevant to family members of patients in their individual specialty (reflected by a low quantitative feedback score for “relevance”), the family member panel disagreed and felt that the item was relevant to many respondents and understood its meaning. Therefore, the item was retained. Although the investigators were concerned that the wording of item 19 (My sex life is affected) could be too blunt or explicit, the panels both disagreed and so the wording was left unchanged.

The wording of item 20 (I argue with my family member) was also unchanged, as although it was rated poorly with regard to language clarity during the quantitative feedback, a consensus for change could not be reached between the two panels. The tense of item 21 (My family expenses have increased) was changed to match the other items, instructions and response options which are written in the present tense. Item 22 (I experience problems with holidays) was debated by both panels, as the expert panel did not know what type of holiday the item was referring to, and thought it was too vague. However, looking back to the interview transcripts and the origin of this item, it encompasses a huge variety of problems and many different types of holiday, so it was felt to be acceptable as the respondent would interpret the item in their own way. Furthermore, the family member panel endorsed the item as being relevant, and suggested changing the wording to “I experience problems with going on holiday” to improve the clarity. Written feedback for item 27 (I worry about strangers’ reactions to my family member’s condition) suggested changing the word “strangers” to “peoples”, as it could relate to people the respondent knows. The tense of the instructions at the beginning of the questionnaire was also changed to better reflect the immediate recall period and the tense of the item wording. As a response to comments from the expert panel, the instructions were changed from “tick a box” to “mark a box” to allow respondents to

indicate their answers with a cross if they desired. The statement “Remember, all of these statements relate to how your life is being affected by your family member’s condition at the moment” was added to the top of each page of the questionnaire as a result of feedback from both panels. The order of the response categories was reversed after feedback from the expert panel, and the use of the category “not relevant” was retained as the investigators felt it would be helpful to identify any mis-fitting or irrelevant items at a later stage. The use of “not relevant” will be readdressed later in the further development of the FROM. Although the utility questions received mixed feedback from both panels, they were retained as they had not yet been trialled in a population of family members, and the issue was readdressed later in the FROM development.

The final changes identified during the qualitative and quantitative content validity were implemented to form the developmental version of the FROM (Figure 4.1). The 30 items were increased to 31 items, and the response categories remained the same. The developmental version of the FROM was then taken forward to the validation stage for item reduction.

Figure 4.1: The 31-item developmental version of the FROM



Draft 10
17/05/12
Catherine J Golics

Family Reported Outcome Measure (FROM)®

The statements in this questionnaire relate to how **your** life is being affected by your family member's condition **at the moment**.

Please mark clearly one box for each statement. If the statement is not relevant to you, please mark "Not relevant".

Please remember, this questionnaire is about **your** life, not your family member's life.

Please answer the following questions:

Your age: ☐ Your gender: Male ☐ Female ☐

Your relationship to the patient: ☐

Patient's diagnosis: ☐

How many hours on average per day (out of 24) do you spend doing the following:

Caring for your family member (e.g. helping with dressing, showering, toileting, mobility) ☐ hours

Housework (e.g. cleaning, cooking, shopping, washing) ☐ hours

How many hours sleep do you lose per night due to worry or getting up in the night to help your family member? ☐ hours

For office use only: Score= ☐

Family Reported Outcome Measure (FROM)©

The following statements relate to how **your** life is being affected by your family member's condition **at the moment.**

Please mark clearly a box for each statement. If the statement is not relevant to you, please mark "Not relevant".

		Not at all	A little	Moderately	A lot	Extremely	Not relevant
1.	I feel worried	☐	☐	☐	☐	☐	☐
2.	I feel angry	☐	☐	☐	☐	☐	☐
3.	I feel guilty	☐	☐	☐	☐	☐	☐
4.	I feel sad	☐	☐	☐	☐	☐	☐
5.	I feel frustrated	☐	☐	☐	☐	☐	☐
6.	I feel tired	☐	☐	☐	☐	☐	☐
7.	My behaviour is affected	☐	☐	☐	☐	☐	☐
8.	**It is difficult to find someone to talk to about my thoughts**	☐	☐	☐	☐	☐	☐
9.	**I feel that caring for my family member is a burden**	☐	☐	☐	☐	☐	☐
10.	My housework is increased	☐	☐	☐	☐	☐	☐
11.	My eating habits are affected	☐	☐	☐	☐	☐	☐
12.	My family activities are affected	☐	☐	☐	☐	☐	☐
13.	My leisure activities are affected	☐	☐	☐	☐	☐	☐
14.	My hobbies are affected	☐	☐	☐	☐	☐	☐
15.	My personality is affected	☐	☐	☐	☐	☐	☐

Please turn to the next page

Remember, all of these statements relate to how **your** life is being affected by your family member's condition **at the moment**.

		Not at all	A little	Moderately	A lot	Extremely	Not relevant
16.	It is hard to find time for myself	☐	☐	☐	☐	☐	☐
17.	I feel the need to stay at home	☐	☐	☐	☐	☐	☐
18.	My every day travel is affected	☐	☐	☐	☐	☐	☐
19.	My time is taken up visiting my family member in hospital or attending medical appointments	☐	☐	☐	☐	☐	☐
20.	My sex life is affected	☐	☐	☐	☐	☐	☐
21.	I argue with my family member	☐	☐	☐	☐	☐	☐
22.	My family expenses are increased	☐	☐	☐	☐	☐	☐
23.	I experience problems with going on holiday	☐	☐	☐	☐	☐	☐
24.	I find it hard to plan my time and activities	☐	☐	☐	☐	☐	☐
25.	My own health or well-being is affected because of my family member's condition	☐	☐	☐	☐	☐	☐
26.	My sleep is affected	☐	☐	☐	☐	☐	☐
27.	My social life is affected	☐	☐	☐	☐	☐	☐
28.	I worry about peoples' reactions to my family member's condition	☐	☐	☐	☐	☐	☐
29.	I find it difficult to talk about my family member's condition.	☐	☐	☐	☐	☐	☐
30.	My work or study is affected	☐	☐	☐	☐	☐	☐
31.	My relationships with other family members are affected	☐	☐	☐	☐	☐	☐

Please check that you have answered every question. Thank you

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DISCUSSION

The content validation of the preliminary version of the FROM was both an essential and enriching process. The design of the content validation process met the recommended criteria reported in the literature. For example, both qualitative and quantitative techniques were used (Haynes et al. 1995), and the minimum recommended number of 3 panel judges was exceeded (Grant and Davis 1997). Recommended structural elements for content validity were covered by assessing the scale for its language clarity, completeness, relevance and scaling. Both the qualitative and quantitative feedback was drawn from a large number of judges from mixed backgrounds (family members, clinicians, nurses, research fellow, genetics counsellor, academic experts), helping to increase confidence that the changes made to the questionnaire and the items were representative of the general population of family members, and were clinically relevant across a range of specialties.

The inclusion of nurses as members of Panel A proved vital as they were able to offer a unique insight into the lives of their patients and their families and provide examples from their experiences of working closely with family units. In turn, the academic experts on the panel played a vital role in answering questions from panel members about both quality of life and questionnaire development. The discussion between the members of panel A was both lively and detailed, and although many of the panel members discussed the issues raised in relation to their own speciality, they could also appreciate the generic nature of the FROM, and seemed to bear this is mind when recommending changes. If consensus was not reached between panel members on decisions taken, the investigators referred back to the earlier interview transcripts with family members and used these to make final decisions. In turn, with some issues where disagreement was seen between panel A and panel B, the recommendation from panel B was often taken, as they were representative of the target population. In contrast to panel A, panel B had a more friendly and relaxed atmosphere. This informality was created by the investigators during the introduction speech, and continued throughout the panel meeting. This allowed the family members on the panel to talk more openly about personal issues, and all the panel members felt comfortable enough to explain to the rest of the panel about how the items related to their own personal experiences, and gave examples. The relaxed panel environment was extended to the refreshment break, where the family members offered support to each other in relation to the issues raised in the panel meetings. Discussion of the item wording was of particular importance in panel B, as the language level of the FROM had to be targeted towards the intended audience, including using words that the family members understood and felt comfortable with. Both of the panel discussions were vital steps in the content validation of the FROM and enriched the quality of the questionnaire.

The use of quantitative feedback data to identify content problems amongst the proposed items was also successful. The percentage of judges who agreed and disagreed with each of the four criteria were identified, and the I-CVI was used to identify poorly rated items. These items were then improved following the recommendations from the panels. The majority of judges agreed that the preliminary FROM items were relevant, complete, contained clear language and fitted well with the response options. The agreement between panel members was tested and found to be high, giving confidence that the developmental version of the FROM shows high content validity. The panel discussions, statistics and changes made to form the developmental FROM provide a solid basis for the FROM to proceed to the item reduction stage.

SUMMARY

- Qualitative and quantitative methods were used for content validation of the preliminary FROM.
- Two panels formed the qualitative part of the content validation process. The panels consisted of experts (n=21) and family members (n=3). The written feedback from questionnaire feedback forms (n=23) was also used for qualitative data analysis.
- For quantitative analysis, the panel members, and other invited family members and experts were asked to rate the preliminary FROM items using a 4-point Likert scale on four criteria: language clarity, completeness, relevance and scaling.
- 95% of the judges thought that the items were written clearly, were complete, relevant to family members and fitted well with the response options.
- The scale content validity index was 0.88 suggesting that the content validity for the scale was high.
- The test of agreement between the written feedback from judges was measured using ICC and the result was 0.97 ($p \leq 0.001$, CI= 0.94 to 0.99), indicating a high level of agreement between the 23 judges.
- The two panels discussed the individual items, the questionnaire layout and wording and some of the specific issues raised by the investigators.
- The results from the two panel discussions and the written feedback were recorded in a structured way in order to be compared and contrasted.

- Changes to the measure were made as a result of qualitative and quantitative feedback; one item was split to form two items, and changes were made to item and instruction wording. These changes formed the developmental FROM.
- The developmental version of the FROM was created ready for item reduction.

CHAPTER 5

Item reduction of the Family Reported Outcome Measure (FROM) using Rasch and factor analysis

INTRODUCTION

The developmental version of the FROM, with changes suggested by the content validity panel, was tested in a second field study of family members for item reduction. The results from the 31-item developmental questionnaire were then analysed using both item response theory and factor analysis. The three utility questions were also analysed and the results reported in this chapter.

METHODS

Study Population

The study population for the validation of the FROM was made up of family members of patients across 26 medical and surgical specialties at the University Hospital of Wales, University Hospital Llandough, Velindre Hospital and General Practice. During recruitment for this stage of the study, it was aimed to recruit equal numbers from each of the 26 specialties, and decided that a minimum of five family members from each specialty would be approached for recruitment.

Rasch analysis

In this chapter, the process of Rasch analysis and the subsequent changes made to the FROM are explained as a stepwise process, explained in Figure 5.1 (see Chapter 2 for details of Item Response Theory and Rasch model).

RESULTS

Demographic Characteristics of the Study Participants

245 family members were approached to take part in the study and four declined to participate due to time pressures. One subject was eliminated due to incomplete answers on the questionnaires. The final validation was carried out using data from 240 family members of patients, from 26 specialties shown in Table 5.1. All 240 family members were asked to complete the developmental version of the FROM. The demographic characteristics of the family members are shown in Table 5.2. Most participants were female (67%), Caucasian (96%), the spouse or partner of the patient (50%), the child (21%) or the parent (18%). The remaining 11% were made up of a variety of other relatives including siblings and grandparents. The mean age of family members was 53 years (range 16-90) and the mean age of patients was 53 years (range 1-91). The mean patient disease duration was 103 months (8 years and 7 months). Disease duration ranged from one month to 63 years.

Figure 5.1: The 13 steps used for Rasch analysis

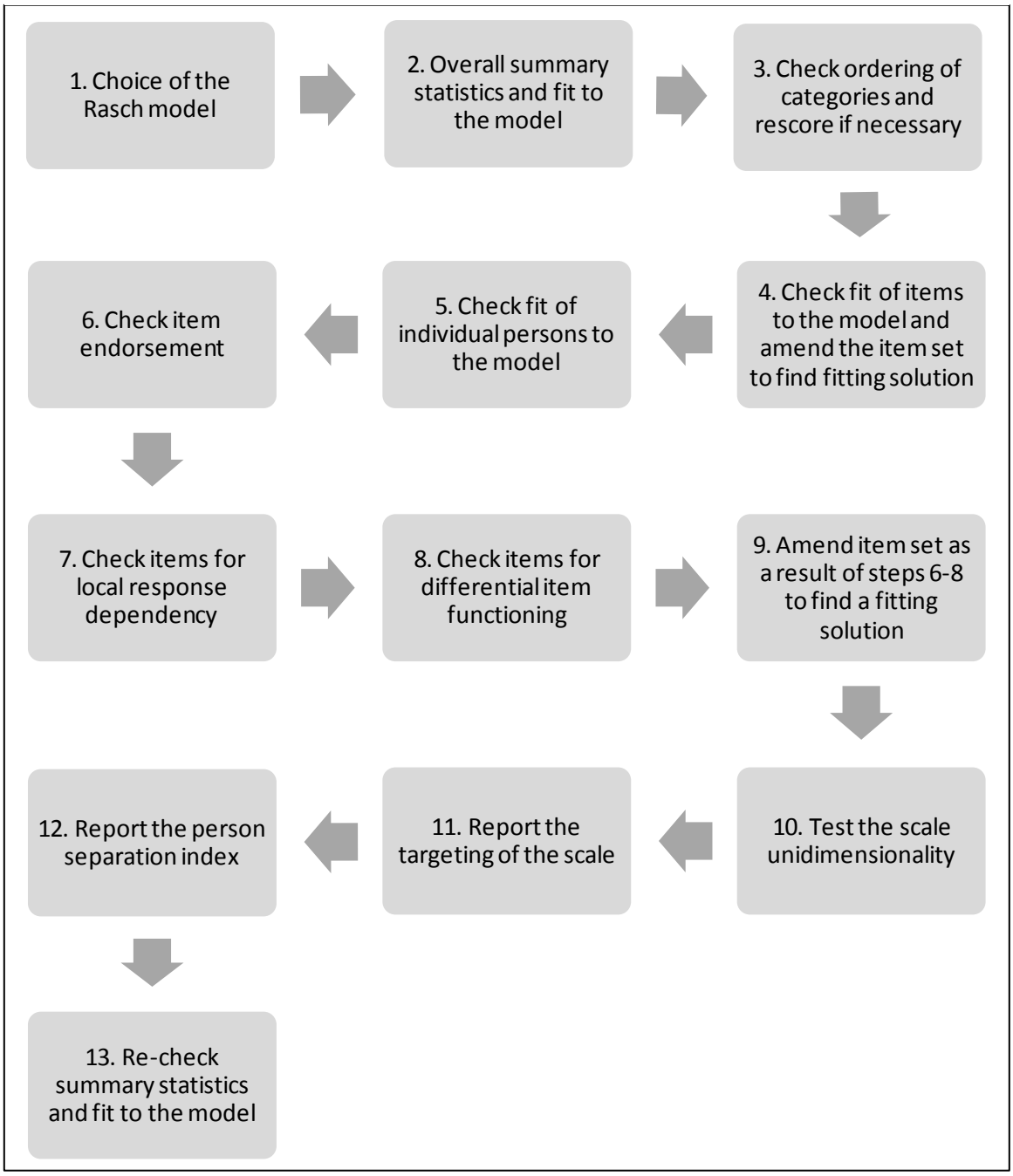


Table 5.1: The 26 specialties included in the item reduction stage of the study

Cardiology	Infectious Diseases
Care of the Elderly	Mental Health
Chronic Pain	Neurology
Colorectal Surgery	Oncology
Dental Surgery	Ophthalmology
Dermatology & Paediatric Dermatology	Orthopaedics & Paediatric Orthopaedics
Ear, Nose and Throat	Paediatric Endocrinology
Endocrinology	Post-stroke
Gastroenterology	Renal & Renal transplant
General Practice	Respiratory
Genetics	Rheumatology
Gynaecology	Urology
Haematology	Wound Healing

Table 5.2: Demographics of the family members during the item reduction stage

Total number of family members	240
Male	80
Female	160
Mean age of family members (range)	53 (16-90) years
Relationship to patient	
Spouse/partner	50%
Parent	18%
Child	21%
Other[a]	11%
Ethnic origin	
Caucasian	96%
Asian/Asian British	2%
Afro-Caribbean	2%
Total number of patients	240
Male	114
Female	126
Mean age of patients (range)	53 (1-91) years
Mean patient disease duration (range)	103 (1-756) months

[a] Other includes nephew, grandparent, sibling and grandchild.

Applicability and Practicality of FROM

During this stage of the study, family members were invited to comment on the 31-item developmental version of the FROM, in particular on the item wording, any items which were not relevant, the three utility questions, and the construct of the questionnaire as a whole. These comments also informed some of the decisions which were made during the item reduction. Table 5.3 shows the comments from family members, both positive and negative, at this stage of the study. The comments were informal remarks made by the family members rather than responses to structured questions.

Table 5.3: Comments regarding the developmental 31-item FROM from family members of patients during the item reduction phase

- I find it difficult to fill in the box with patient's diagnosis.
- It's hard to fill in the hours per day questions. Do I put a "0" if I don't do it?
- I'm stuck on the question about holidays. I can't actually go on holiday at all, so I put "a little"?
- Do I fill it out as me or as the patient?
- I would prefer time frame responses, such as sometimes, a lot, all of the time.
- I'm stuck on the utility questions. I can't distinguish the time I spend waking up due to my own medical problems and my wife's.
- The questions are well worded and not too long.
- It needs to be made more clear where the end is, as you turn over the page expecting it to finish.
- What do I do if I don't have any hobbies?
- For the utility questions, if I don't spend any time doing these things then what do I write?
- I didn't notice "not relevant" as I didn't read the response options properly before I answered.
- I don't talk to anyone about my feelings so how do I answer that question? I didn't notice the "not relevant" box.
- I can't quantify the hours on the questions on the front page.
- Most of the questions are not relevant to me.
- The expenses question is really good and relevant as I now have to drive my wife to appointments every week.
- "It is difficult to talk to someone about my thoughts". I don't know how to answer this one with the response options given as the questions don't fit them.
- I can't answer the utility questions- it's impossible to quantify this.
- A lot of the questions overlap and its like asking the same questions over again.
- I do the accounts for my relative, does that count for the "caring" utility question?
- "I feel tired". Well I feel tired anyway, but if this is relating to my wife then it needs a "because of..." after it.
- "My every day travel is affected" is a very odd question- what does it mean?
- I found some of the questions difficult to answer because I hadn't really thought about them before.
- "My personality is affected". I think someone else should comment on this, I can't really say.
- What do I do after the first page?
- The most relevant utility question is the sleep one.
- I would like to see a "general comments" box so I can add more of my thoughts.
- I find it hard to write the patient's diagnosis.
- Do I put my details on the front page?
- The "every day travel" question is odd.
- Does travel insurance count for the holiday question?
- How would I know if my behaviour or personality has changed? You should be asking her this, not me.
- The questions cover everything well.
- I don't feel like "burden" is the right word.
- The question about sex life isn't relevant to me.
- The questionnaire is quick and easy to fill in.
- I missed out the first question as there wasn't much space between the top and the first question.
- Does question 10 mean the work I do normally do around the house has doubled?
- I think there's a question missing. There should be a question about worrying about other members of the family being affected. I worry about my mother having to look

after my father.
- I think for question 19 the two parts of the question should be swapped around so that medical appointments comes first as its more relevant.
- I don't understand the question about burden. What does it mean? Does it mean that it affects other areas of my life or that I find it difficult?
- I think the questions reflect my thoughts really well and you seem to have covered everything. I couldn't have written them better myself.
- The questions you are asking are spot on.
- Does "stay at home" mean my home or theirs?
- "My personality is affected". I've not got a clue, you'd have to ask someone else.
- What do you mean by family activities?
- That questionnaire will do well, the questions are really good.
- Those questions are really spot on.
- Whoever constructed that questionnaire did a really good job.
- The main thing that distressed me was not finding out what was wrong with my wife for a long time, and no question covers that.

Utility questions

The developmental version of the FROM included three utility questions, designed to quantify the effects of the patient's illness on three different areas of the family member's life. The three utility questions were:

How many hours on average per day (out of 24) do you spend doing the following:

1. *Caring for your family member (e.g. helping with dressing, showering, toileting, mobility)*
2. *Housework (e.g. cleaning, cooking, shopping, washing)*
3. *How many hours sleep do you lose per night due to worry or getting up in the night to help your family member?*

For each of the three questions, there was missing data, as many of the family members reported that the utility questions were very difficult to complete. The number of valid responses (n) and the response rate (r) is given for each question. The mean number of hours per day spent caring for the patient (n=207, r=86%) was 4.5 (range 0-24). The mean number of hours spent doing housework (n=214, r=89%) was 4.9 (range 0-24). The mean number of hours sleep lost per night (n=206, r=86%) was 1.5 (range 0-8).

Family members found these three concepts very difficult to quantify, as seen in the patient feedback in Table 5.3. For each of these questions there was also a large amount of missing data. In order to produce a simple, easy to use measure, the burden on subjects needs to be low, and it was felt that these utility questions increased the burden on subjects. Furthermore, as the answers are given in numbers, it is difficult to compare them to the Likert-style items, and they would not be able to be incorporated in a total score. It was therefore decided that these three questions should be dropped from the FROM altogether.

However they provide interesting information, particularly the amount of time family members lose from their day providing care to the patient.

Missing data

Missing data for the utility questions has been discussed above, but there was also missing data seen in the questionnaire item responses. The amount of missing data varied between items, with the lowest missing data percentage of 0.4% and the highest of 17%. The amount of missing data can be used to analyse the acceptability of items, and can suggest that some items need to be re-worded or clarified if a large volume of missing data is identified. In this case, the high missing data percentages for some items suggests that re-wording should take place. In this stage of the study, missing data was analysed alongside the percentage of responses for each response category (Table 5.5) when selecting items to remove or re-word. It is also important to consider how the missing data is handled. RUMM2030 gives the option to create data sets with complete data records only, or includes all cases but removes missing values case-wise. Both options were selected during the Rasch analysis, depending upon the test being used.

Sample size

The consideration of sample size during Rasch analysis is extremely important, as item fit statistics can be highly sensitive to sample size when using polytomous data (Smith et al. 2008). In general, to be able make discriminations between people, as large a sample size as possible is ideal (Streiner and Norman 2008). In order to be confident (99% sure) that an item calibration is within 1 stable logit, a sample size of 50 is required and in order to be within ½ a stable logit, the sample size requirement rises to 150 (Linacre 1999). The ideal sample size to produce a statistically stable measure given by Linacre (1999) is up to 250 subjects. In this study, the sample size was 240, meaning that it is high enough to be confident that a statistically stable measure will be produced.

Scoring of the developmental version of the FROM

The developmental version of the FROM has 31 items, each scored on a 5-point Likert scale, with a 6th response option of “not relevant” (scored 0). Each item in the developmental version of the FROM was scored: “Not at all”= 0, “A little”= 1, “Moderately”= 2, “A lot”=3, “Extremely” =4. This gave a possible score range of 0-124.

Rasch analysis

Rasch analysis was carried out on the developmental 31-item version of the FROM. Table 5.4 shows the 31 items along with their item numbers for reference. The process of Rasch analysis followed that shown in Figure 5.1.

Step 1. Choice of the Rasch model

As the FROM has more than two response categories, the polytomous Rasch model was chosen, as opposed to the dichotomous model. There are two versions of the polytomous model; the Andrich Rating Scale Model (Andrich 1978) and the Masters Partial Credit Model (sometimes called the Unrestricted model) (Masters 1982). The Partial Credit Model places no restrictions on the threshold parameters, whereas the Rating Scale Model, which produces a higher degree of specificity, dictates that all thresholds must be equally spaced across the trait for all of the items. In other words, the Rating Scale Model expects the distance between the thresholds separating each response category (for example, between "Moderately" and "A lot") to be the same across all of the items (Tennant and Conaghan 2007).

RUMM2030 uses a Likelihood-Ratio Test to determine which model should be used on a set of data. In the case of the FROM data, the Partial Credit Model was chosen, as the result of the Likelihood-Ratio Test showed that the data did not fit the Rating Scale Model ($p<0.05$).

Step 2. Overall summary statistics and fit to the model

The next stage in the Rasch analysis process was checking the overall summary statistics of the FROM and how well it fits to the model without any adjustment (Figure 5.2). The person and item fit statistics are transformed by RUMM2030 to an approximate Z-score representing standardised normal distribution. The fit residual mean value for all 31 items is -0.22 with a standard deviation of 1.97. The fit residual mean would be expected to be closer to 0, and the standard deviation would be expected to be much closer to 1 to give adequate fit to the model. The misfit is supported by a significant chi squared interaction of 271 (degrees of freedom= 93) and $p\leq 0.01$, showing a lack of invariance of item difficulty across the scale, meaning that the scale cannot differentiate between the different groups; from those whose quality of life is affected greatly to those who are not affected at all. A chi squared probability value of above 0.05 would suggest that there is no significant deviation between the observed data and what is expected from the model (Bland and Altman 1995; Tennant 2011). This could be caused by misfit to model expectations of respondents or items (as explained by the summary statistics), or both.

Table 5.4: The 31 items of the developmental version of the FROM

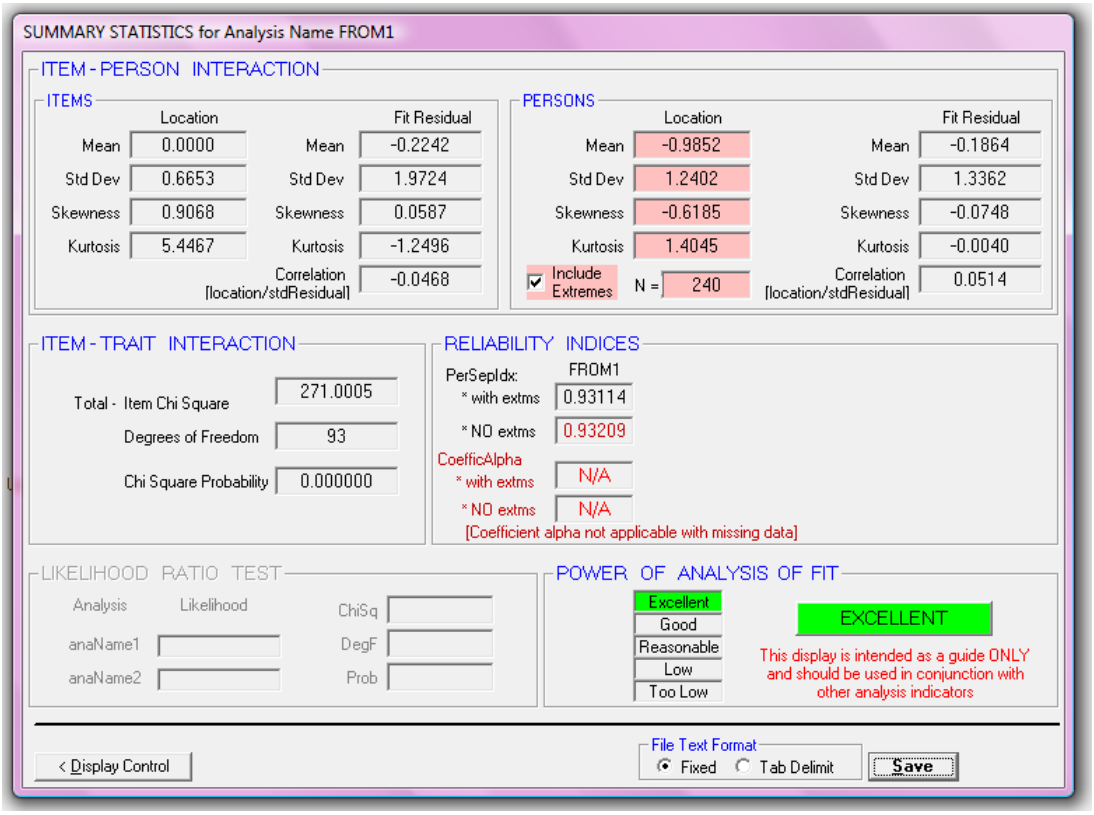
Item number	Item
1	I feel worried
2	I feel angry
3	I feel guilty
4	I feel sad
5	I feel frustrated
6	I feel tired
7	My behaviour is affected
8	It is difficult to find someone to talk to about my thoughts
9	I feel that caring for my family member is a burden
10	My housework is increased
11	My eating habits are affected
12	My family activities are affected
13	My leisure activities are affected
14	My hobbies are affected
15	My personality is affected
16	It is hard to find time for myself
17	I feel the need to stay at home
18	My every day travel is affected
19	My time is taken up visiting my family member in hospital or attending medical appointments
20	My sex life is affected
21	I argue with my family member
22	My family expenses are increased
23	I experience problems with going on holiday
24	I find it hard to plan my time and activities
25	My own health or well-being is affected because of my family member's condition
26	My sleep is affected
27	My social life is affected
28	I worry about people's reactions to my family member's condition
29	I find it difficult to talk about my family member's condition
30	My work or study is affected
31	My relationships with other family members are affected

The residual mean value (0= perfect fit to model) for persons was -0.19 with a standard deviation of 1.34 (1= perfect fit to model), indicating no serious misfit amongst the respondents in the sample. It is also worth noting that at this stage the mean person location value was -0.99 (0= perfect fit to model), meaning that in general the family members were of a slightly lower "ability" than the FROM (their quality of life was affected at the lower end of what the items and responses allowed for), and that family members scored lowly on the scale. The 31-item developmental version therefore failed to fit the Rasch model, and improvements to the scale construct are required.

Step 3. Check order of categories and rescore if necessary

In a polytomous data model, checking the threshold ordering is very important when considering fit to the model. As a person ability increases (in the case of the FROM, as the quality of life of the family member is affected more greatly), then they should be more likely to obtain a higher score. This progression should be logical, so as the QoL is affected more, the family member is more likely to score a 20, than a 19, for example. The term "threshold" means "the point between two response categories where either response is equally probable...for example the probability of scoring a 0 on the item or scoring a 1 is 50/50" (Pallant and Tennant 2007).

Figure 5.2: Summary statistics for the 31-item developmental version of the FROM



In RUMM2030, threshold ordering is checked using threshold maps, which identify graphically whether each item is progressing in a logical order. Disordered thresholds can indicate that subjects find it difficult to discriminate between response options, often when there are too many response options or when the labelling of the response options is confusing (Pallant and Tennant 2007). The common response to disordered threshold is to collapse the number of response categories, which often improves fit to the model (Pallant and Tennant 2007; Pesudovs and Noble 2005; Streiner and Norman 2008; Zhu et al. 1997).

The threshold map in RUM2030 was checked for the FROM, to identify any disordered items. Figure 5.3 shows that 24 of the 31 items were disordered, as marked by "**" on the map. Next, the category probability curves for each item were looked at. Figure 5.4 shows an example of an ordered category probability curve for Item 1, where it can be seen that responses to this item fall in a logical, progressive order. On the other hand, Figure 5.5 shows an example of a disordered probability curve for item 4, where it can be seen that even at the point where the probability of scoring a 2 is highest (at person location -1), it is still more likely that a 1 will be scored instead.

Figure 5.3: The threshold map for the 31-item developmental version of the FROM

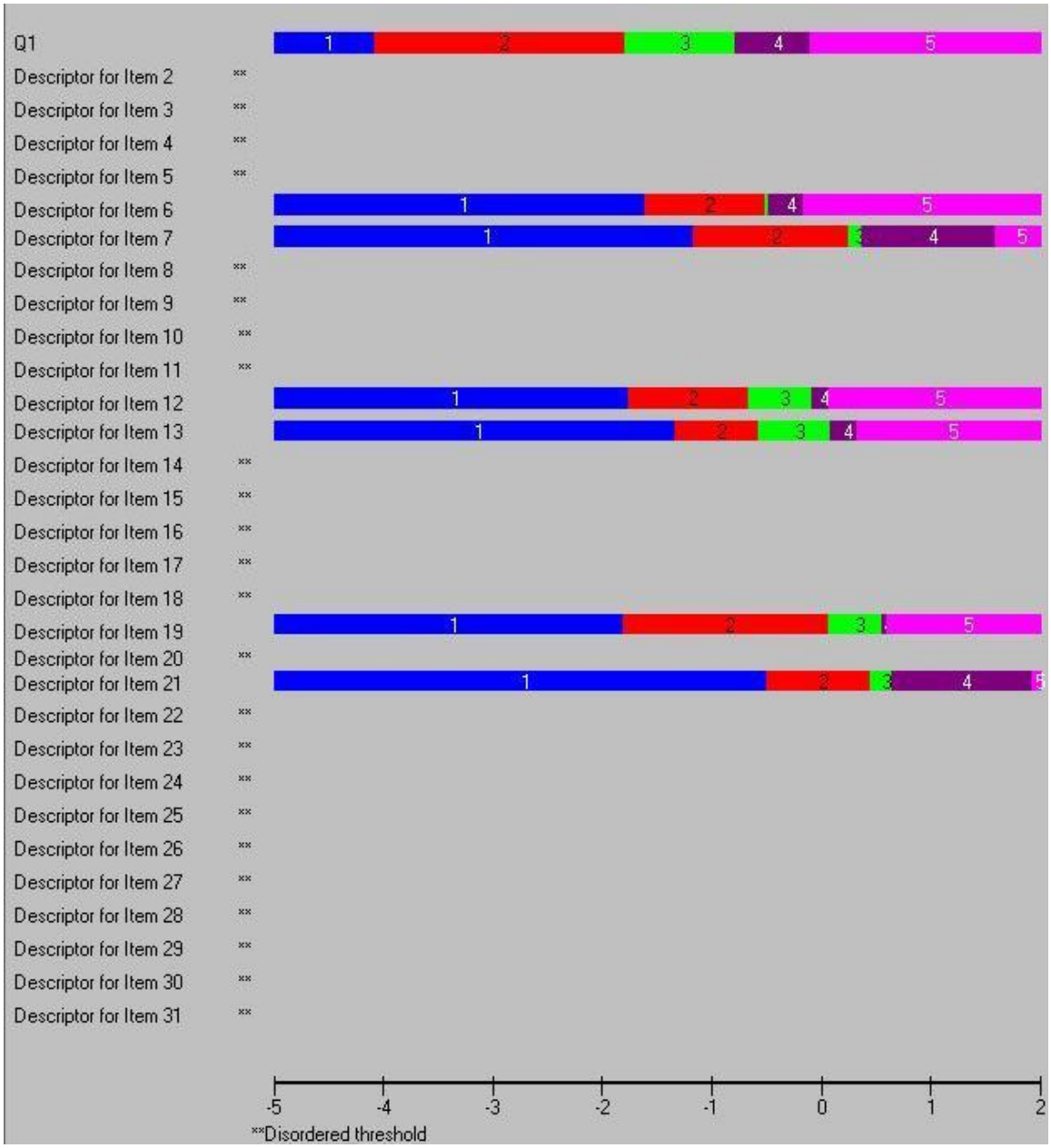


Figure 5.4: The ordered category probability curve for item 1

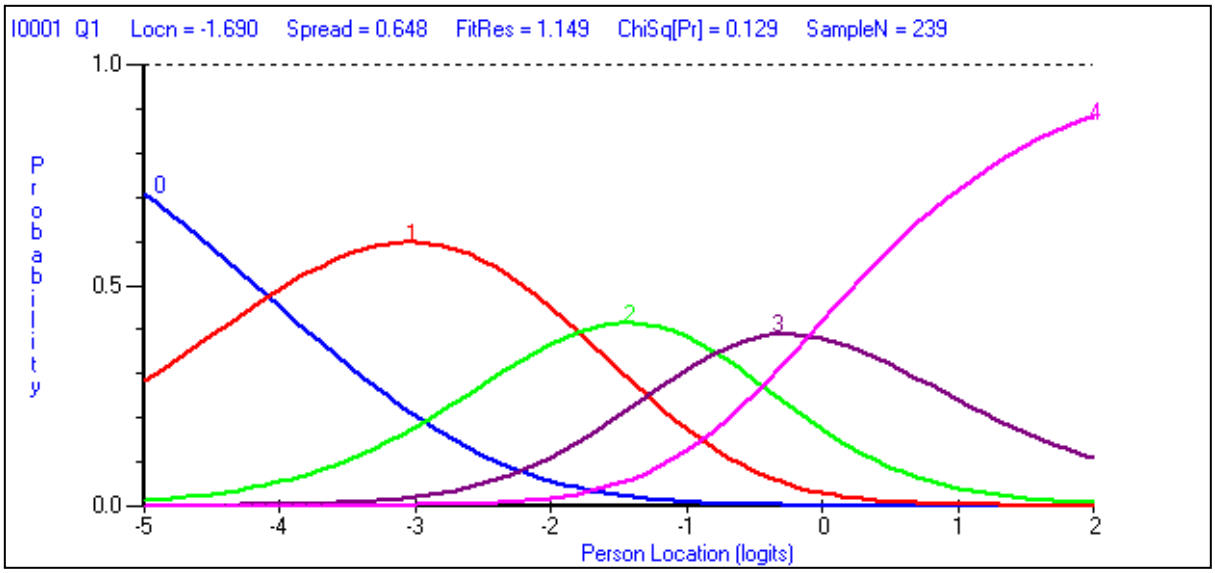
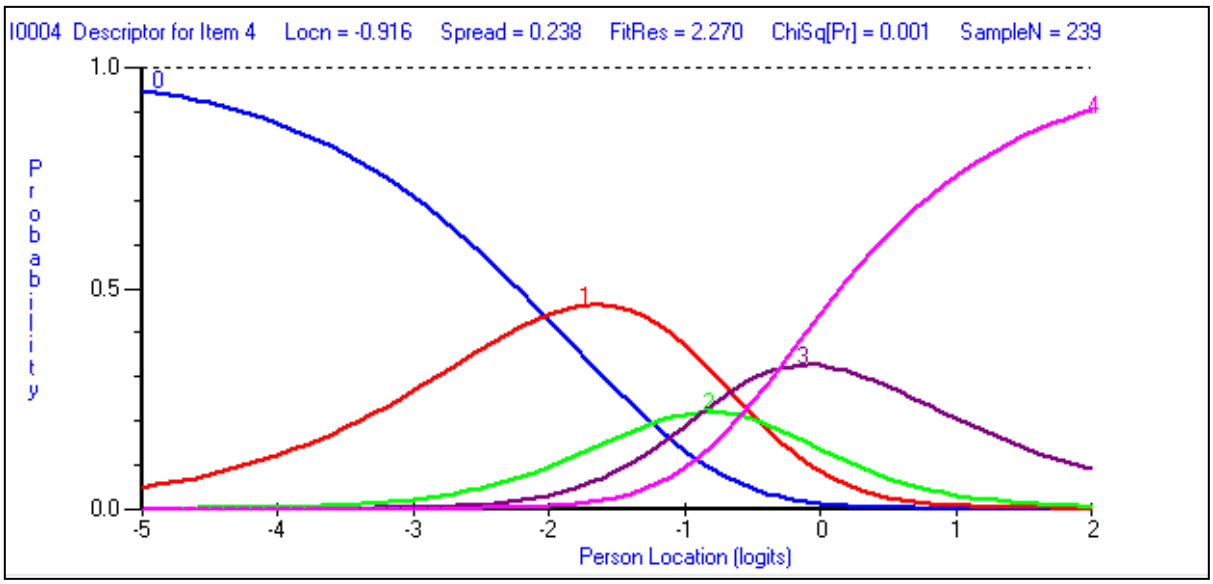


Figure 5.5: The disordered category probability curve for item 4



It was decided that the response categories of the FROM would be collapsed in order to obtain threshold ordering across all items and improve fit to the Rasch model. Previously, two main approaches to collapsing the categories have been taken. Firstly, collapsing the categories individually for each item in turn, and therefore having a different scoring system for each item (Chien and Bond 2009; Lamoureux et al. 2006). Secondly, categories can be collapsed for the questionnaire as a whole, giving the same scoring system for each item (Lamoureux et al. 2008; Penta et al. 1998). In the case of the FROM, it was decided that the categories would be collapsed for the questionnaire as a whole, giving a uniform scoring

system. There were several reasons for this. Firstly, the majority of items were disordered and would need categories collapsing. Collapsing categories in the same way for all of the items would make the FROM easier for investigators and clinicians to score, as they would not be required to use a score sheet. It was felt that this would enable the FROM to be more widely used in a clinical setting, as it would also reduce the time burden on the clinician. Furthermore, having the same scale scoring for each item would avoid putting a greater emphasis on certain items where the subject can potentially score more highly if different scoring is used (Uhlig et al. 2007). This was felt to be especially important in the area of quality of life measurement, where the concept is subjective to the family member, and not a physical attribute, for example in a disability index.

In order to decide which categories to collapse, the category response proportions were examined for each item, along with the category probability curves. Table 5.5 shows the proportions of responses in each scoring category, and it can be seen clearly that categories 3 and 4 have few responses across most items. In particular, these categories contain less than 10% of responses for many items, and were therefore identified as low-scoring categories, suggesting that individually, they are not relevant for many family members. For all but three of the items, combining categories 3 and 4 would result in a new category with percentage scores of over 10%.This was also reflected in the category probability curves for disordered items. Categories 3 and 4 ("A lot" and "Extremely") were collapsed to form one category, and the categories were rescored, as shown in the "Rescore 1" column in Table 5.6. Collapsing these two categories also made sense from a conceptual point of view, as it can be seen how subjects could find it difficult to distinguish between the wording of the two categories.

Collapsing categories 3 and 4 did not improve the overall model fit. The chi squared probability value remained at 0, and mean item fit residual worsened to -0.35 (standard deviation 1.95). Only one of the disordered items benefitted from the rescore, meaning that 24 of the 31 items were still disordered. Therefore, the category probability curves were examined again, and it was decided that categories 1 and 2 ("A little" and "Moderately") would also be collapsed to form one category. From a conceptual point of view, this would enable the subjects to more clearly distinguish between whether an event occurs "Not at all", "A little/Moderately" or "A lot/Extremely". The column "Rescore 2" in Table 5.6 shows how the categories were rescored, giving a scoring system of 01122.

Table 5.5: The category response percentages for the 31-item developmental FROM

	Percentage response for each category				
Statement	**0**	**1**	**2**	**3**	**4**
Item 1	3	20	27	27	23
Item 2	47	25	12	6	9
Item 3	53	24	9	6	8
Item 4	17	26	15	20	22
Item 5	21	28	15	16	20
Item 6	25	23	16	17	19
Item 7	39	33	15	10	4
Item 8	51	18	12	9	10
Item 9	76	13	8	3	0
Item 10	47	18	16	13	6
Item 11	52	28	10	5	5
Item 12	25	26	20	15	14
Item 13	32	24	20	13	11
Item 14	47	22	12	11	9
Item 15	47	31	10	8	4
Item 16	41	28	10	14	7
Item 17	36	25	14	18	6
Item 18	55	24	8	8	4
Item 19	27	39	19	9	6
Item 20	58	14	9	7	12
Item 21	51	29	12	6	2
Item 22	37	26	19	8	10
Item 23	37	18	13	16	17
Item 24	41	27	11	15	7
Item 25	47	3	11	7	5
Item 26	34	3	12	13	11
Item 27	39	27	13	11	9
Item 28	63	19	9	6	3
Item 29	69	20	4	4	3
Item 30	60	22	10	4	4
Item 31	64	19	9	4	4

Collapsing the five response categories into three meant that all 31 items were now ordered (Figure 5.6). The summary statistics showed that the chi squared probability value remained at 0, and mean item fit residual worsened to -0.4 (standard deviation 1.88). Collapsing response categories did not improve the fit to the model, but as the thresholds are now ordered and the response categories are working correctly, further tests to improve fit to the Rasch model can now be carried out. It is important to note that creating fewer categories can impact the future responsiveness and sensitivity of the measure, as there is a narrower range of possible scores.

Table 5.6: The rescore values for each response category of the 31-item developmental FROM

Category (response option)	Original score	Rescore 1	Rescore 2
Not at all/not relevant	0	0	0
A little	1	1	1
Moderately	2	2	1
A lot	3	3	2
Extremely	4	3	2

Step 4. Check fit of items to the model and amend the item set to find fitting solution

As well as looking at the overall fit, it is important to look at the fit statistics for individual items in order to identify those items which are misfitting and therefore not performing well. The fit residuals (the divergence of each person from the model) for each item were examined. As the data approximate a normal distribution, a mean of 0 and a standard deviation of 1 would be expected. Any items with a fit residual of greater than +/- 2.5 (Mavranezouli et al.; Pallant and Tennant 2007) are a cause for concern as they represent a misfit to the Rasch model. High negative residuals suggest that items are not adding anything new to the scale (Mills et al. 2009; Tennant 2011) and can be considered for removal. In the case of the FROM, the misfitting items were identified as item 27 (fit residual -3.57), item 24 (-3.51), item 13 (-3.01), item 28 (3.25), item 3 (3.06) and item 10 (2.57). These items were removed from the FROM one at a time, starting with the most mis-fitting item (item 27). After each item removal, the overall fit statistics of the measure were evaluated (Table 5.7). Consequently, removing all six of the mis-fitting items improved the fit to the Rasch model (Table 5.7), bringing the standard deviations closer to one (analysis number 6). However, the chi squared probability value remained at 0, suggesting that there is still further work to be carried out to finalise the FROM.

Step 5. Check fit of individual persons to the model

As well as checking the fit of individual items, it is important to check the fit of the individual subjects to the Rasch model. This can identify mis-fitting respondents who may be skewing the analysis. Similar to item mis-fit, individual persons are expected to fall within a fit residual of +/- 2.5. Those with a high negative fit residual could be answering too perfectly, and trying to answer the items as they think they should be (Tennant 2011), and low negative fit residual could mean that mis-fitting persons could be showing a bizarre response pattern (Tennant and Conaghan 2007). Removing mis-fitting persons from the analysis can often have a significant impact on the fit of the measure, however removing these persons could raise questions about the construct validity of health-related scales, especially those which are being newly developed, as the mis-fitting persons may represent an important aspect of health, for example an unreported co-morbidity (Pallant and Tennant 2007). In the case of the FROM, 11 respondents were identified as having fit residuals greater than +/- 2. Removing these respondents from the analysis improved the overall fit to the model slightly, bringing the mean item and person fit residuals down by 0.02 and 0.04 respectively. However, as the fit to the model was not greatly improved, and the removal of the mis-fitting respondents could have a negative effect on the construct of the FROM, it was decided that these respondents would be included in the further analysis.

Step 6. Check item endorsement

The endorsement for each item was examined using the category response frequencies in RUMM2030. Items which had a low endorsement level (a high percentage of respondents scoring 0) were considered for removal as they were not seen as important or relevant for a large number of family members. Conversely, items with a high endorsement level (a high percentage of respondents scoring 2) were viewed as being important and relevant to a large percentage of family members, and contribute greatly towards the impact on their quality of life. The two items which showed a lower level of endorsement compared to the others were items 9 and 29. The scores for item 9 were: 0=169, 1=46, 2=6. This item was a very important concept during the interview stage, but the wording of the statement was criticised by family members during their feedback. Therefore, it was decided that the concept behind this item should be retained in the FROM, but the wording of the item should be reconsidered. Item 29 was also lowly endorsed: 0=156, 1=55, 2=15. This item measures a similar concept to item 8, which shows higher endorsement, and therefore should be considered for removal. The endorsement level of individual items was considered during the later stages of Rasch analysis, when deciding on item retention and removal.

Figure 5.6: The threshold map for the 31-item developmental version of the FROM after collapsing response categories

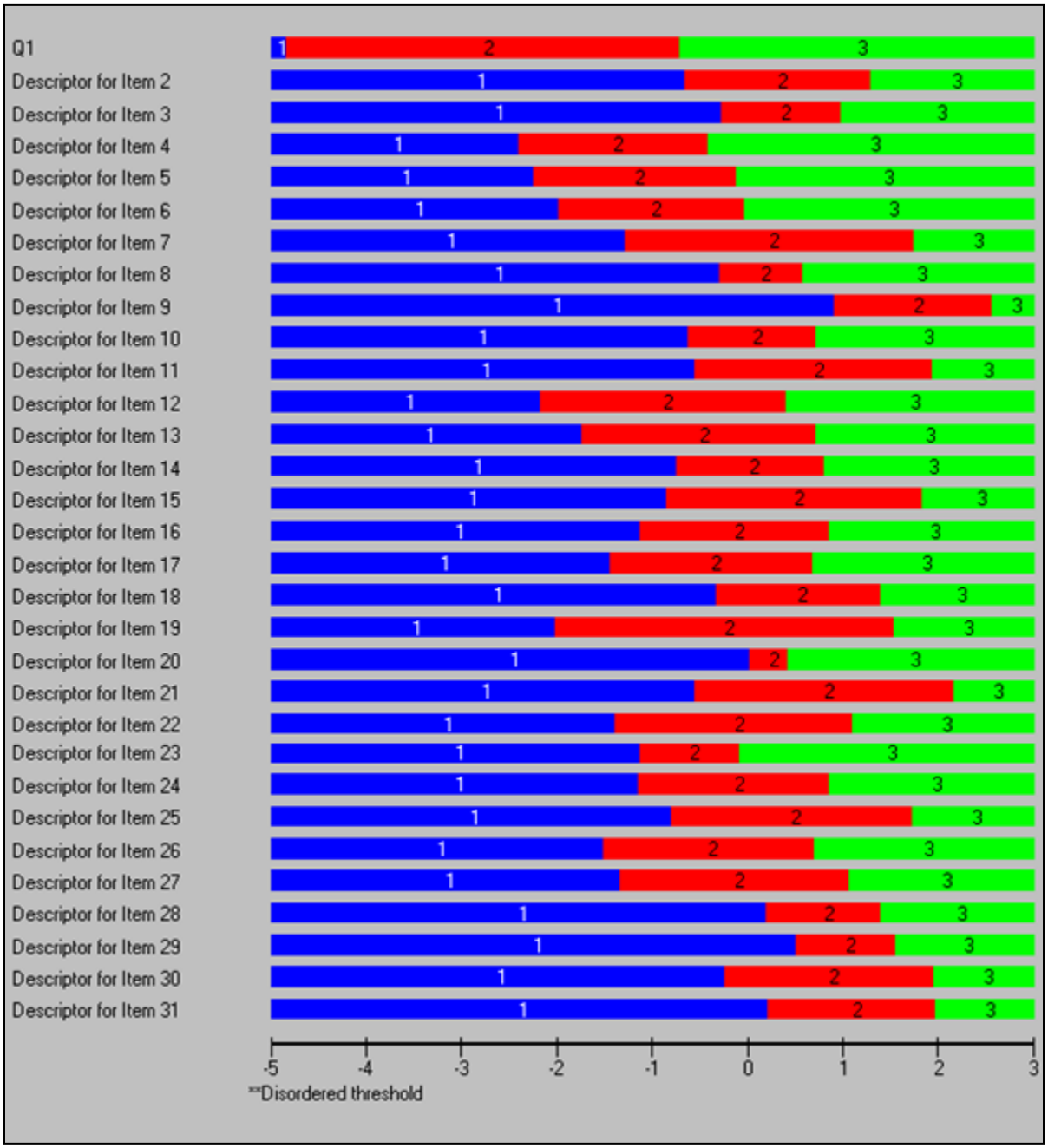


Table 5.7: The step by step removal of mis-fitting items in the FROM

Analysis number	Description	# items in scale	Chi^2 p value	Item fit residual mean (S.D)	Person fit residual mean (S.D)
1	Developmental version of FROM with categories collapsed	31	0	-0.4 (1.88)	-0.2 (1.35)
2	Remove items 27 and 24	29	0	-0.33 (1.68)	-0.2 (1.29)
3	Remove items 27, 24 and 13	28	0	-0.3 (1.59)	-0.19 (1.26)
4	Remove items 27, 24, 13 and 28	27	0	-0.33 (1.52)	-0.2 (1.23)
5	Remove items 27, 24, 13, 28 and 3	26	0	-0.35 (1.39)	-0.22 (1.20)
6	Remove items 27,24,13,28,3 and 10	25	0	-0.37 (1.37)	-0.22 (1.19)

Step 7. Check items for local response dependency

One source of possible misfit within a scale is local response dependency. The Rasch model assumes local independence; once the main factor (in this case the Rasch factor) is removed, there is no correlation between items (Streiner and Norman 2008). This means that a person's response to one item will have no bearing on their response to another item in the scale. Removing local dependence can help to improve fit to the Rasch model, often by eliminating or combining items which appear to measure the same attribute and show high correlation. In RUMM2030, local response dependency is assessed by looking at the residual correlations between items (Kahler and Strong 2006). Potentially problematic dependency between items can be identified between items if the residual correlation is between 0.2 and 0.3 above the average of all of the item residual correlations (Hansen et al. 2012). There are other ways of carrying out checks for local response dependency, including directly correlating the item responses (Streiner and Norman 2008), but RUMM2030 produces a correlation matrix of the fit residuals for each item. The average residual correlations of the FROM items was -0.039, therefore correlations between items of above 0.16 were identified. Table 5.8 shows the items which were identified as having high residual correlations and the solution decided by the investigators for each. To decide on a potential solution, the wording of the items was considered, along with the level of endorsement and the feedback from family members regarding the items. The prevalence of each item concept during the interview stage was also considered.

Step 8. Check items for differential item functioning

Differential item functioning (DIF) is another important potential source of misfit in the data. Items are classed as showing DIF when different groups of respondents (for example male and female) within the same sample respond differently to an individual item (Pallant and

Tennant 2007). This can mean that an item is biased towards one particular group of people. Although there is no consensus regarding the best way to measure DIF, running an analysis of variance is the most common way (Streiner and Norman 2008), and also the method used by RUMM2030. Two types of DIF exist; uniform and non uniform DIF. Uniform DIF is seen when each group of respondents shows a consistent systematic difference in their responses to an item, across the whole range of the attribute being measured (Teresi et al. 2000), identified by a significant analysis of variance test. Non uniform DIF is seen when differences in groups vary across levels of an attribute, and there is non-uniformity in the differences between groups (Tennant and Conaghan 2007). Uniform DIF can cancel out in a measure, for example if there is one item biased more towards females and one towards males in the same measure (Tennant and Pallant 2006a). If the uniform DIF does not cancel out, the problem item(s) can be split, for example by gender, and scored separately(Lundgren-Nilsson et al. 2005). Non-uniform DIF is more difficult to deal with, and items sometimes have to be rewritten or removed from the scale (Pallant and Tennant 2007). However, as emphasised by Streiner and Norman (2008), the impact and significance of the DIF must also be considered in relation to the concept being measured; it is important to consider whether the presence of DIF in the measure is problematic, and it is important to look further than the statistics.

The presence of DIF in the FROM was identified using one way analysis of variance (ANOVA) carried out in RUMM2030. The respondents are split into roughly equal class intervals depending upon their FROM score. RUMM2030 produces tables for the ANOVA tests for each item, and these are examined for significant differences ($p<0.05$). Item characteristic curves were also produced for each item using RUMM2030, to show the effects of DIF graphically. The different groups analysed for the FROM were age and gender. Relationship to patient and medical specialty were also considered, but the numbers in these groups were so small that no reliable conclusion could be made. It was also decided that the presence of DIF by specialty was not problematic, and could be expected; family members of patients from different specialties would be expected to answer differently and be biased towards certain questions. For example, family members of patients from gynaecology or urology may score more highly on an item regarding their sex life due to the nature of the patient's symptoms. DIF by relationship to patient was also not seen as problematic; closer relations to patients (e.g. spouses) may score more highly on some items than more distant relatives. DIF becomes more of a problem when using Rasch analysis for measurement in education or in validating disability scores where bias would be problematic.

Table 5.8: The local dependency identified within the FROM and potential solutions

Items involved	Potential solution
Items 1, 2, 4 and 5	These four items are all measuring a similar attribute (emotions), so some local dependency could be expected. However, all of these items show good fit to the model. The endorsement level for all of the items was high, so all should be retained.
Items 7 and 15	The two items are measuring similar attributes, and this was noted by the family members during this stage of recruitment. The two items were given poor feedback by family members, and so will have to be reworded if one is retained. Item 7 is more highly endorsed so should be kept over item 15.
Items 12 and 14	These two items have similar wording, and only one should be retained. Item 12 is worded more clearly, as several family members felt that, as adults, they no longer have "hobbies". The wording of item 14 fits well with the theme of the FROM.
Items 14 and 16	Removed item 14
Items 14 and 18	Removed item 14
Items 16 and 18	It is difficult to see from a conceptual point of view why these two items are showing correlation as they measure two very different things. Therefore retain both.
Items 12 and 23	Although it can be seen from a conceptual point of view why there is a correlation between these two items (both involve doing things together as a family), the theme of holidays was highly prevalent during the interview stage, and is clearly very important to family members so both items should be retained.
Items 14 and 30	Removed item 14
Items 20 and 23	Both items emerged as important concepts during the interview stage of the study and are measuring different things, so both should be retained.
Items 6, 25 and 26	These three items are all measuring similar concepts so do not need to all be retained. Item 26 is the most highly endorsed, and items 5 and 26 measure two overlapping concepts. These items need to be investigated further.

DIF was identified in twelve of the FROM items (Table 5.9). The item characteristic curves were then examined to identify which group the item was biased towards when uniform DIF was shown. Figure 5.7 shows an example of uniform DIF by age seen in item 14, and Figure 5.8 shows an example of non-uniform DIF by gender in item 15, where the lines on the graph can clearly be seen to be crossing, demonstrating non-uniformity.

Step 9. Amend item set as a result of steps 6-8 to find a fitting solution

Six of the mis-fitting items from the developmental 31-item version of the FROM were removed during step 4 of the Rasch analysis. The results of the tests for item endorsement, local response dependency and DIF showed that further item reduction is required for the FROM to fit the Rasch model. Removal of the items was carried out in a stepwise process with one problem item being removed at a time, and the effect on the summary statistics was

stated after each stage of item removal (Table 5.10). The reasons behind each stage of removal are stated below.

Figure 5.7: The item characteristic curve for item 14 showing uniform DIF by family member age

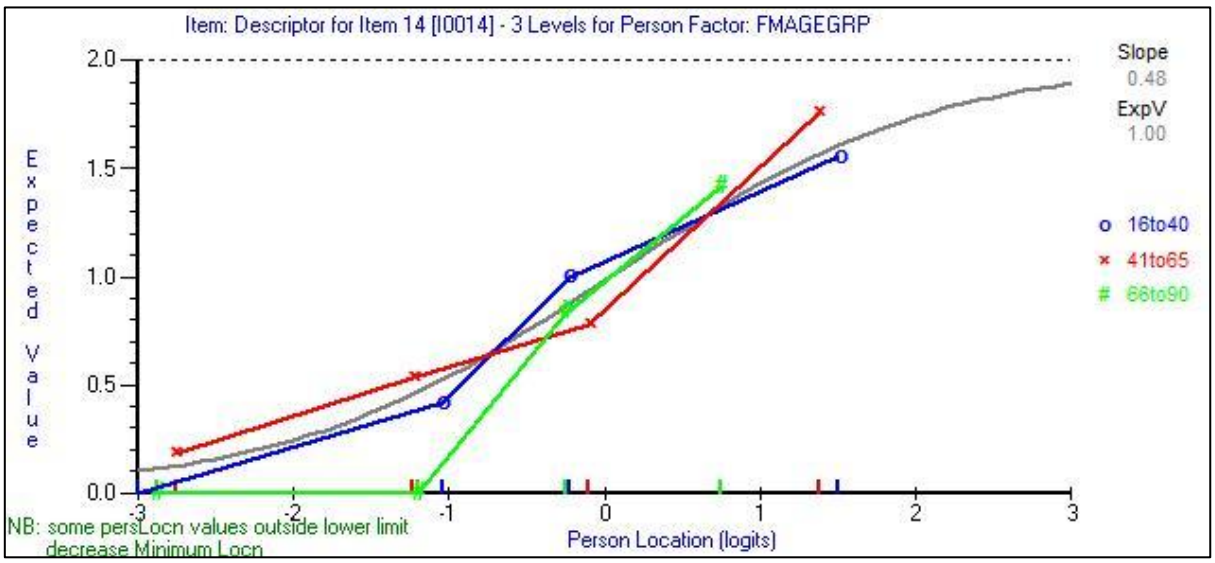


Figure 5.8: The item characteristic curve for item 15 showing non-uniform DIF by family member gender

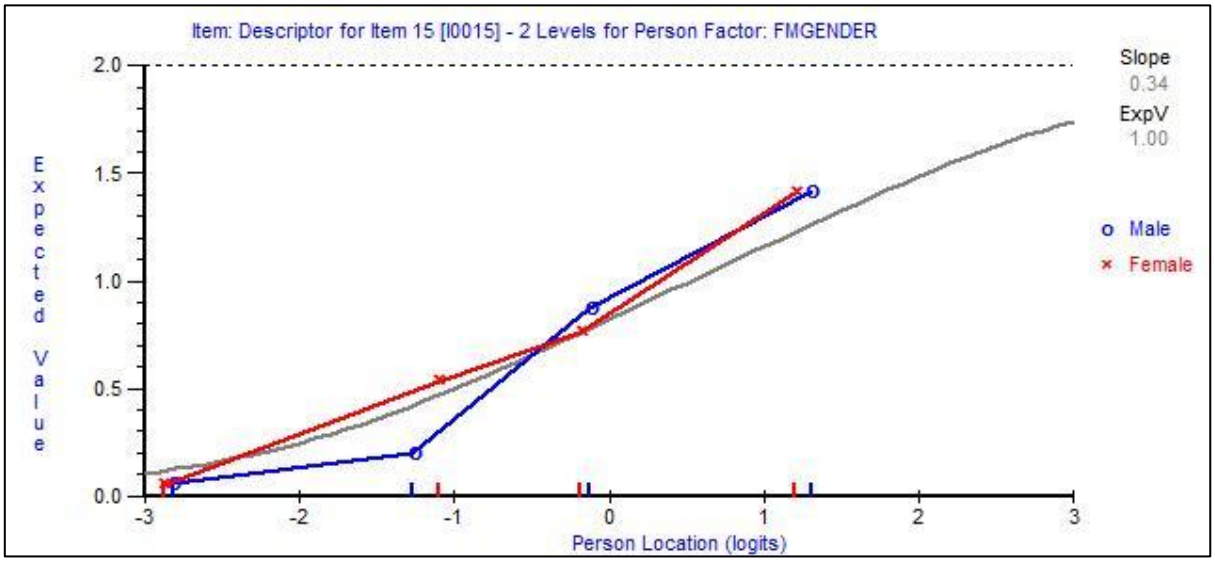


Table 5.9: Uniform and non-uniform DIF identified in the items of the 31-item developmental FROM

Item	Uniform DIF by family member age	Non-uniform DIF by family member age	Uniform DIF by family member gender	Non-uniform DIF by family member gender
1	No	No	No	No
2	No	No	No	No
4	No	No	Yes (p=0.034) Female higher	No
5	No	No	No	No
6	No	No	Yes (p=0.011) Female higher	No
7	No	No	No	No
8	No	No	No	No
9	No	No	No	No
11	No	No	No	No
12	No	No	Yes (p=0.028) Male higher	No
14	Yes (p=0.015) Younger higher	No	No	No
15	No	No	No	Yes (p=0.03) Female higher
16	No	No	Yes (p=0.040) Female higher	No
17	No	No	No	No
18	No	No	No	No
19	No	No	No	No
20	No	No	Yes (p=0.009) Male higher	Yes (p=0.003)
21	No	No	No	No
22	No	No	Yes (p=0.001) Male higher	No
23	Yes (p=0.0009) Older higher	No	Yes (p=0.0098) Male higher	No
25	No	Yes (p=0.044)	No	No
26	No	No	Yes (p=0.017) Female higher	No
29	No	No	No	No
30	Yes (p=0.008) Younger higher	No	No	No
31	No	No	No	No

ANALYSIS 5

Analysis 5 included the removal of the six mis-fitting items during step 4 of Rasch analysis, and lists the statistics that need to be improved to improve fit to the Rasch model. The chi^2 value should be above 0.05 for the measure to fit the Rasch model (Tennant and Conaghan 2007). The fit residual means for items and persons were already close to 0, but a reduction in these values would be an improvement, and the standard deviations should be closer to 1.

ANALYSIS 6

For the next step of item removal, five further items were removed. Items 6 and 25 were removed as they showed local dependency with item 26, which was retained as it was thought to be the most clearly worded item, and the most favoured by family members. Item 14 was removed as a result of local dependency with several other items. Family members expressed that it was a badly worded item. Items 7 and 15 showed local dependency, and both were given poor ratings by family members. At this stage, both were removed. The removal of these five items improved the fit statistics, reducing the fit residual means and bringing the standard deviations closer to 1.

ANALYSIS 7

Items 8 and 29 showed local dependency, so they either needed to be combined into one item or one item removed. As each item should measure one concept, item 29 was deleted and item 8 was retained. The two items are describing a similar concept and have similar wording. Removal of item 29 worsened the fit statistics slightly, but not significantly enough to cause concern. The chi^2 p value was also brought into an acceptable range (p=0.15).

ANALYSIS 8

Analysis 8 mirrored analysis 7, and item 8 was deleted whilst item 29 was retained. This worsened the fit statistics further, and worsened the chi^2 p value once again (p=0.02).

ANALYSIS 9

During analysis 9, both items 8 and 29 were deleted. This was carried out to ensure that keeping either item 8 or item 29 was beneficial to the FROM from a Rasch point of view. Removal of both items worsened the fit statistics and the chi^2 p value remained below 0.05 (p=0.03). Therefore, as a result of analyses 7-9, and from a conceptual point of view, it was decided that item 8 should be retained and item 29 deleted.

ANALYSIS 10

During analysis 6, both items 7 and 15 were deleted. These items showed local dependency, low endorsement and were rated poorly by family members. To ensure that deleting both items was the correct decision, item 7 was added back into the FROM, as it was the more highly endorsed item of the two. This addition worsened the fit statistics slightly, and the chi^2 p value fell (p=0.01). Therefore, the removal of both items 7 and 15 was deemed to be the correct decision.

ANALYSIS 11

During analyses 11-15, three items which had been retained in the FROM up until this point, but which had potential problems were removed one by one to look at the effect they had on

the fit statistics. In analysis 11, item 19 was deleted as it was an item which received poor rating from family members and had a low endorsement. It was also felt that the concept of time was covered by item 16, which was retained. Deleting item 19 did not make a large difference to the fit residual mean values, but it improved the standard deviations and increased the chi^2 p value (p=0.17), in comparison to analysis 7 which was the most recent analysis deemed to have the best combination of items.

ANALYSIS 12

In analysis 12, item 21 was deleted as it was lowly endorsed and the concept of relationships with other family members was covered by item 31 which was retained. Removal of item 21 whilst retaining item 19 did not have a great effect on the fit statistics; the fit residual values for item fit worsened slightly, but improved slightly for person fit. Therefore, item 21 was tested further during analysis 14.

ANALYSIS 13

Item 17 was deleted during analysis 13 as it was the most mis-fitting item left in the FROM (fit residual value of -1.81). The chi^2 p value was still not significant, so item 17 was deleted the concept of having to stay at home was also covered by other items, such as the emotional-based items. Item 17 was also largely mis-fitting in comparison to the other items, so it may have been skewing the analysis. The removal of item 17 further improved the fit statistics and increased the chi^2 p value to 0.04. Therefore, it was decided that item 17 should be removed from the FROM.

ANALYSIS 14

In analysis 14, items 17, 19 and 21 were deleted. Items 17 and 19 had already been identified for deletion, and removing item 21, which was identified as an ambiguous item improved the fit statistics and brought the chi^2 p value up to 0.18. Therefore, analysis 14 was decided as the basis of the final set of 16 items for the FROM as the combination of items produced excellent fit statistics whilst reflecting the earlier qualitative themes.

Dealing with DIF

Eight of the 16 items retained during the item removal were identified during step 8 as having differential item functioning (DIF). As mentioned in step 8, items with uniform DIF can either be split into each of the groups in question (for example having different scoring systems for males and females for an item) or DIF can cancel out in a measure at test level (Tennant and Pallant 2006a) and the person estimates would not be affected. In RUMM2030, items with DIF are grouped into a subtest to see if the DIF cancels out. Table 5.11 shows that the items showing uniform DIF have the potential to cancel out, as there are roughly equal numbers of

items biased towards males and females. A subtest in RUMM2030 was carried out which showed that both DIF by gender and DIF by age cancelled out in the measure as a whole (p=0.6 and p=0.8 respectively). Only one item (item 20) showed non-uniform DIF. The wording of this item had already been discussed in detail during the content validity panel and by the family members during their feedback, and all family members felt that the direct wording of the question, using the term “sex life”, as opposed to “intimate relationships” was much clearer to them. Therefore, the item was retained in the FROM as the concept was very important to family members during the interview stage, especially for partners and spouses, and the preferred wording was used. To encourage family members to answer personal questions such as this item, it was made clear to participants that privacy and confidentiality was maintained during all parts of the study.

Table 5.10: The stepwise process of item removal during Rasch analysis

Analysis number	Description	# items in scale	Chi^2 p value	Item fit residual mean (S.D.)	Person fit residual mean (S.D.)
5	Remove items 27,24,13,28,3 and 10	25	0	-0.37 (1.37)	-0.22 (1.19)
6	Remove items 27,24,13,28,3,10,6,7,14,15 and 25	20	0.0099	-0.27 (1.16)	-0.2 (1.10)
7	Remove items 27,24,13,28,3,10,6,7,14,15,25 and 29	19	0.146	-0.28 (1.18)	-0.22 (1.07)
8	Remove items 27,24,13,28,3,10,6,7,14,15,25 and 8	19	0.017	-0.32 (1.2)	-0.2 (1.1)
9	Remove items 27,24,13,28,3,10,6,7,14,15,25,8 and 29	18	0.025	-0.34 (1.15)	-0.21 (1.03)
10	Remove items 27,24,13,28,3,10,6,14,15,25 and 29	20	0.006	-0.30 (1.2)	-0.2 (1.08)
11	Remove items 27,24,13,28,3,10,6,7,14,15,25,8,29 and 19	18	0.17	-0.33 (1.14)	-0.21 (1.04)
12	Remove items 27,24,13,28,3,10,6,7,14,15,25,8,29 and 21	18	0.076	-0.31 (1.19)	-0.2 (1.05)
13	Remove items 27,24,13,28,3,10,6,7,14,15,25,8,29 and 17	18	0.036	-0.28 (1.1)	-0.2 (1.04)
14	Remove items 27,24,13,28,3,10,6,7,14,15,25,8,29, 19,17 and 21	16	0.18	-0.36 (1.07)	-0.22 (1.01)

Step 10. Test the scale unidimensionality

One of the assumptions of Rasch analysis is unidimensionality, and this can be tested using RUMM2030. If the scale is unidimensional, the scores for each item can be summed to produce a total score. The unidimensionality of the scale and factor identification will be tested further using factor analysis, but carrying out this test in RUMM2030 gives an idea as

to whether the scale is unidimensional, using an independent t-test comparison of person locations estimated using two different subsets of items taken from the scale (Smith 2002). In a similar method to local dependency, the residuals (which remain when the Rasch factor is removed) are examined. The items are correlated with the first remaining factor (the first component). The items which correlate positively are grouped into one subtest, and those which correlate negatively are grouped into another (Table 5.12). Separate person estimates are then derived for the two subtests, and an independent t-test is carried out on the two subtests. For unidimensionality to be accepted, less than 5% of the t-tests should be significant at the p=0.05 level (Smith 2000). In the case of the FROM, 25 (9.66%) of the 236 t-tests were significant, suggesting that the scale is not unidimensional. The deviating results from the t-tests were found to be significant when 95% confidence intervals from a binomial distribution were applied and the lower bound confidence interval did not overlap the 5% value (confidence intervals: 8% to 13%), which confirms that the unidimensionality of the FROM is not acceptable (Tennant and Pallant 2006b). This will be investigated further during factor analysis.

Step 11. Report the targeting of the scale

Figure 5.9 shows the targeting of the FROM. The top section of the graph represents the family members and their ability levels (how much their QoL has been affected) and the bottom part represents the item locations and distribution. The outliers can be seen at either end of the scale. The mean location score for items is set at 0, meaning that the targeting of family members can be assessed using the mean person location value, which in the case of the FROM was -0.622. For a well-located measure, the mean person location value should be around zero, and the small negative value seen in the case of the FROM suggests that the sample as a whole was located at a lower level than the items (Tennant and Conaghan 2007). However, the deviation from 0 was not large and the sample consisted of patients with a diverse range of illnesses ranging by disease type, duration and severity, so was not directed towards those patients who are extremely unwell, or who have particularly affected relatives. There was no statistical difference (evaluated using one way analysis of variance in RUMM2030) between the location, or ability, of family members of different genders (p=0.97) and age groups (p=0.22).

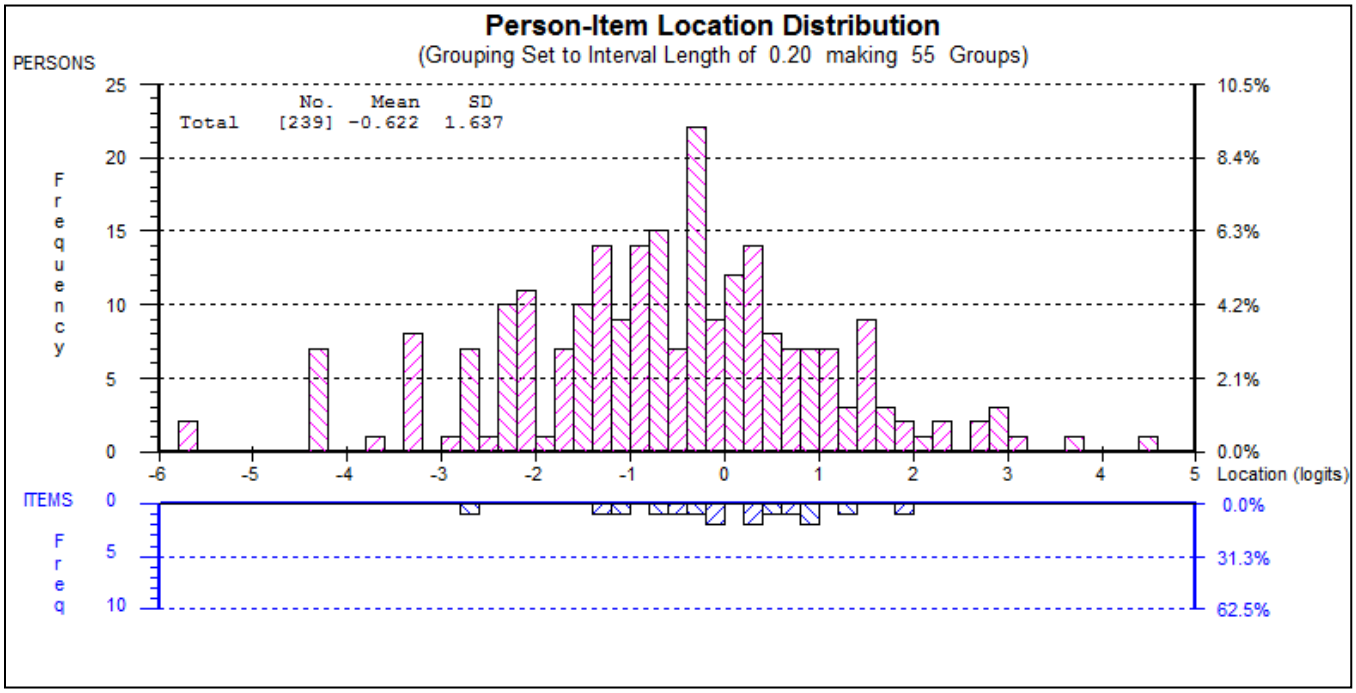
Table 5.11: Uniform and non-uniform DIF identified in the items of the 16-item final FROM

Item	Uniform DIF by family member age	Non-uniform DIF by family member age	Uniform DIF by family member gender	Non-uniform DIF by family member gender
1	No	No	No	No
2	No	No	No	No
4	No	No	Yes (p=0.008) Female higher	No
5	No	No	No	No
8	No	No	No	No
9	No	No	No	No
11	No	No	No	No
12	No	No	Yes (p=0.043) Male higher	No
16	No	No	Yes(p=0.018) Female higher	No
18	No	No	No	No
20	No	No	Yes (p=0.011) Male higher	Yes (p=0.003)
22	No	No	Yes (p=0.002) Male higher	No
23	Yes (p=0.0008) Older higher	No	Yes (p=0.03) Male higher	No
26	No	No	Yes (p=0.005) Female higher	No
30	Yes (p=0.01) Younger higher	No	No	No
31	No	No	No	No

Table 5.12: Loading of items onto the first component extracted during principal component analysis of the FROM

Item	Loading onto first principal component
4	0.736
5	0.558
2	0.540
1	0.473
8	0.146
31	0.021
9	0.011
26	*-0.073*
12	*-0.118*
20	*-0.179*
11	*-0.225*
22	*-0.394*
23	*-0.323*
30	*-0.347*
16	*-0.475*
18	*-0.543*

Figure 5.9: The person-item location distribution of the FROM



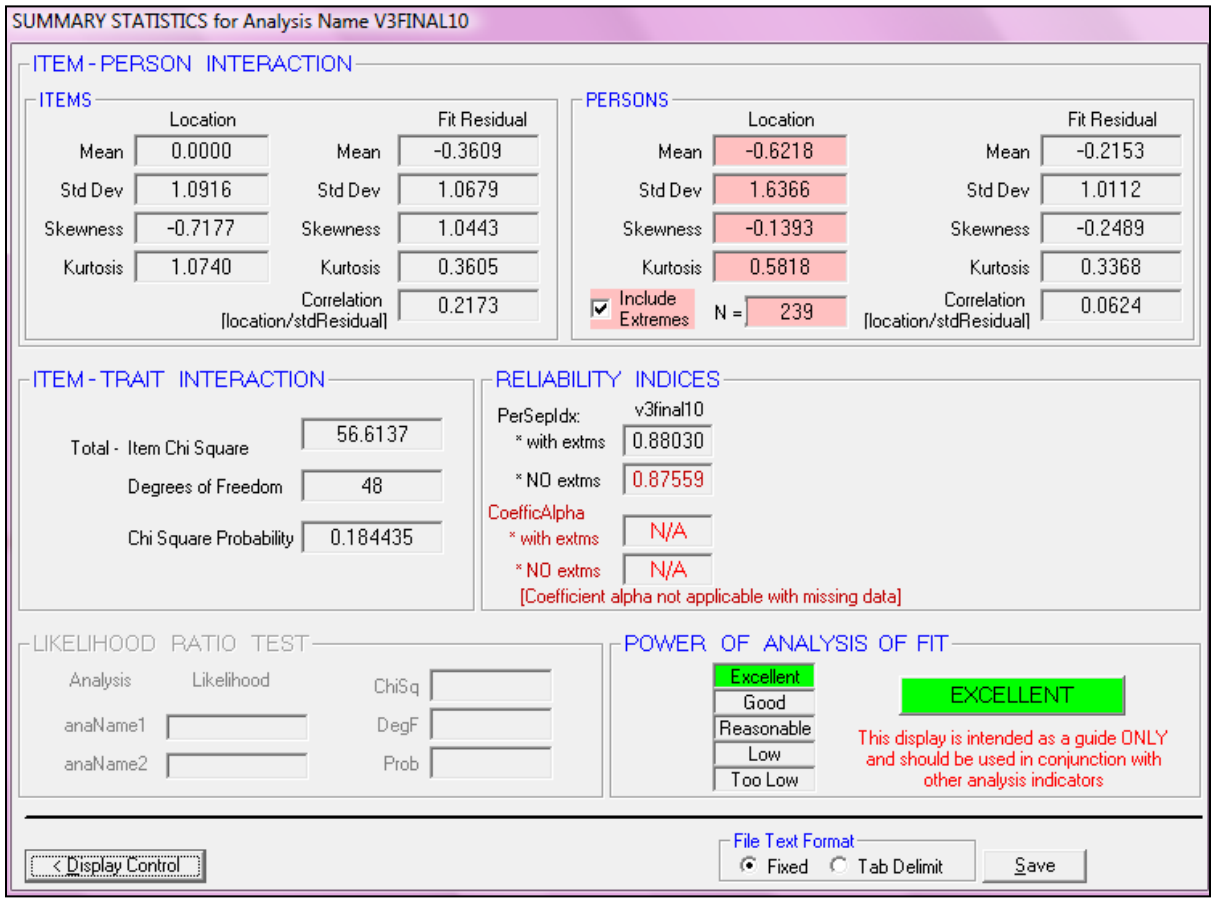
Step 12. Report the person separation index

The person separation index (PSI) is reported as a measure of reliability of the scale equivalent to Cronbach's alpha (Cronbach 1951). The PSI indicates whether the measure can discriminate between different groups of respondents. The minimum acceptable level of the PSI is 0.7, meaning that the measure can significantly distinguish between 2 different groups of respondents. The PSI of the FROM was calculated at 0.88 (rounded to 0.9), meaning that the measure can significantly distinguish between 4 different groups of respondents (Fisher 1992). A high PSI is also an indicator that the fit statistics can be relied upon, as there is less error surrounding them (Tennant 2011).

Step 13. Re-check summary statistics and fit to the model

The summary statistics for the final version of the 16-item FROM demonstrated that it has a good fit to the Rasch model (Figure 5.10). The fit residual mean value for items was -0.36 with a standard deviation of 1.07. The chi squared interaction value is 56.6 (degrees of freedom= 48) and p= 0.18. The residual mean value for persons was -0.22 with a standard deviation of 1.01.

Figure 5.10: The summary statistics for final 16-item version of the FROM



FACTOR ANALYSIS

The Rasch analysis of the FROM suggested that the measure is made up more than one dimension, or factor. Therefore factor analysis, a component of classical test theory was used to identify these factors and assign items to each. Factor analysis can also be used to reduce items (Floyd and Widaman 1995) in a scale. The statistical methods of factor analysis are used to identify relationships between groups of items with similar themes. In the case of the FROM, Rasch analysis was used as the primary method of item reduction as it has many advantages over factor analysis (Streiner and Norman 2008). The suitability of the remaining 16 items of the FROM were also assessed using factor analysis to strengthen the argument for the reduction of 31 items to 16. Factor analysis was used to help finalise the scoring of the final version of the FROM, and to decide whether the items in the FROM are uni or multi dimensional.

There are two types of factor analysis; exploratory factor analysis (EFA) and confirmatory factor analysis (CFA). EFA is used when little is known about the correlation between a set of items, often when developing new measures as a way to identify the factor structure of the

new measure, or to reduce items (Fayers and Hand 1997). On the other hand, CFA is used to test a hypothesis or theory, usually when the factor structure of a measure is known (Pallant 2005). In addition, CFA has been recognised as having little potential value in quality of life research (Fayers and Hand 1997). Therefore, EFA was selected as the method for factor analysis of the FROM.

Reliability of the FROM

The reliability of the 16-item FROM was measured before the factor analysis was carried out. This can help identify items which are considered weak, and should be removed from the scale. If all of the items perform strongly, it can be taken as a sign that the Rasch analysis produced a reliable and strong version of the FROM. The internal consistency, measured using Cronbach's alpha (Cronbach 1951) was 0.91, indicating good reliability of the scale (Pallant 2005). Weak items which should be removed from a scale are those with a low item-total correlation. The definition of a low correlation varies between 0.2 (Kline 1986) and 0.4 (Juniper et al. 1997), and all of the 16 FROM items were shown to have an item-total correlation of above 0.4 (range 0.4-0.7), which indicates that they are all strong discriminative items and should be retained. As a secondary measure, each item was then deleted from the scale in turn, and the Cronbach's alpha remained high (0.90-0.91) each time. The further removal of items did not improve the reliability of the FROM, and in most cases the reliability was reduced. This gives further confidence that the FROM contains a strong and appropriate set of items.

Tests for sample adequacy

Gorsuch (1983) suggested guidelines for the sample size of subjects for factor analysis, and these were consulted in relation to the FROM. A minimum of five to ten cases for each item should be used as a sample size for factor analysis, and for the FROM, 240 subjects were used for a 16-item measure. Bartlett's test of sphericity (Bartlett 1954) was applied to prove that the variables in the correlation matrix are correlated. It should be significant at $p<0.05$ to reject the hypothesis that the variables are uncorrelated (Pallant 2005). For the FROM, Bartlett's test of sphericity was significant ($p<0.001$). A second test of sample adequacy was also applied, the Kaiser-Meyer-Olkin (KMO) (Kaiser 1974), where a value of >0.6 should be observed for the sample to be considered adequate for factor analysis (Kaiser 1974). The FROM showed a KMO of 0.91, which is considered "marvellous" (KMO >0.9) by Kasier (1974).

Exploratory factor analysis

Exploratory factor analysis was carried out using PASW statistical software. Factors were identified using a combination of several methods. Those factors with Eigenvalues ≥1 were

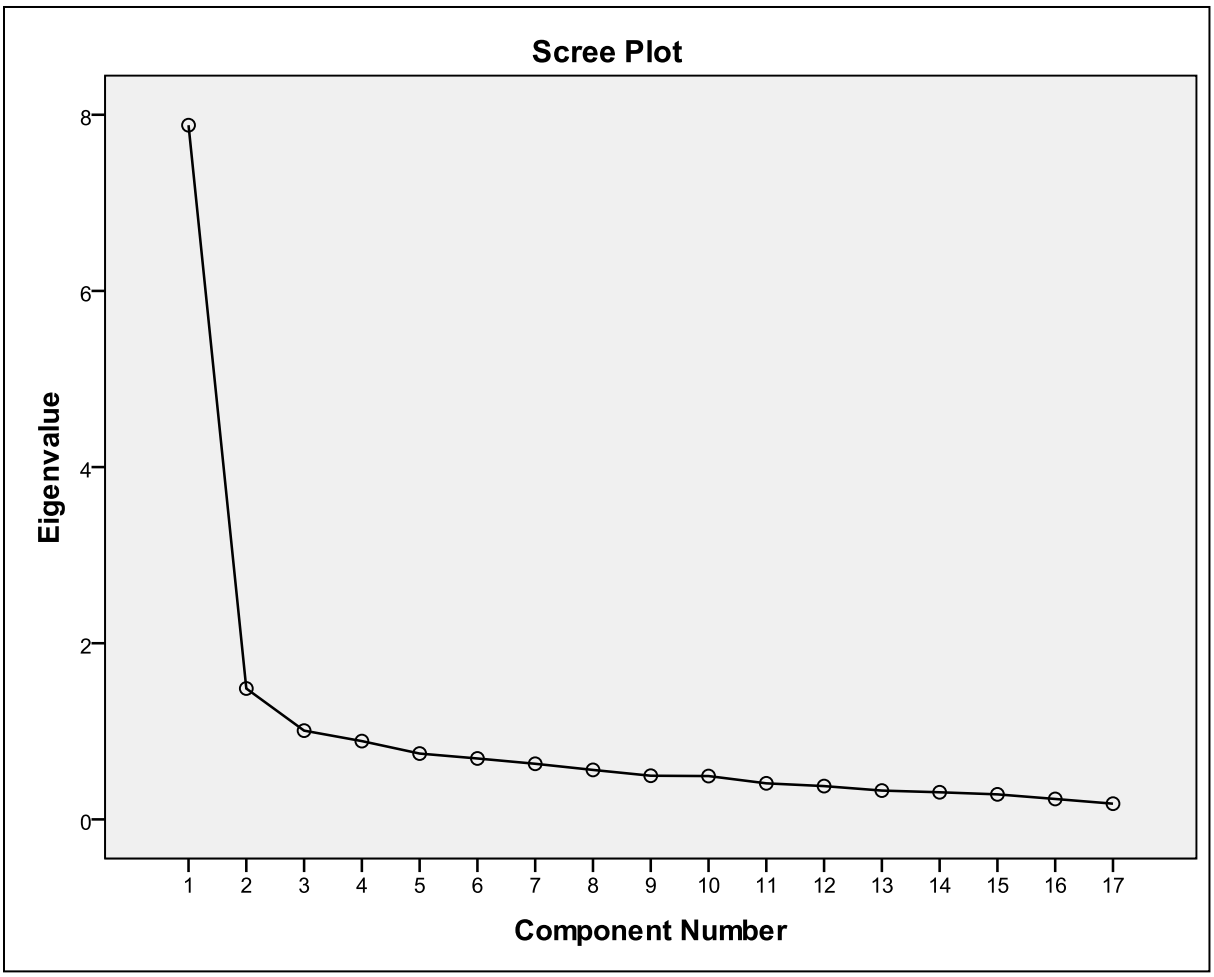
retained for further analysis, following Kaiser's criterion rule (Pallant 2005). The principle component analysis for the 16-item FROM (Table 5.13) revealed three factors with Eigenvalues ≥1, which together explained 61% of the variance, higher than the minimum recommended 50% value for a stable factor solution (Streiner 1994). One of these factors was very close to the Kaiser's criterion cut off point with an Eigenvalue of 1.008.

Cattell's scree plot (Cattell 1966) was drawn for the FROM (Figure 5.11). The scree plot shows a sharp drop after the first and second factors, and then the line becomes flat, suggesting the first two factors account for most of the variance. It is recommended that any factors above the "elbow" of the graph (factors 1 and 2 in the case of the FROM) are retained or considered for further analysis (Cattell 1966). This supports the results of the principal component analysis, as the third factor was on a borderline value for retention. Therefore, two factors of the FROM are taken forward for factor rotation.

Table 5.13: The principal component analysis for the 16-item FROM

Component	Initial Eigenvalues			Extraction Sums of Squared Loadings		
	Total	% of Variance	Cumulative %	Total	% of Variance	Cumulative %
1	7.881	46.361	46.361	7.881	46.361	46.361
2	1.486	8.741	55.102	1.486	8.741	55.102
3	1.008	5.928	61.030			
4	.889	5.228	66.258			
5	.747	4.397	70.655			
6	.692	4.068	74.723			
7	.631	3.714	78.437			
8	.562	3.304	81.741			
9	.496	2.916	84.656			
10	.492	2.894	87.550			
11	.409	2.408	89.958			
12	.378	2.223	92.181			
13	.327	1.925	94.106			
14	.308	1.811	95.916			
15	.284	1.671	97.587			
16	.232	1.364	98.951			
17	.178	1.049	100.000			

Figure 5.11: Scree plot to show the variance in the components of the FROM



Factor rotation

Factor rotation was carried out to identify the patterns of item loading onto each of the two factors. Varimax rotation was chosen, which is often regarded as the most conceptually clear and the most commonly used orthogonal rotation method and does not require the factors to be correlated to one another (Fabrigar et al. 1999). After the Varimax rotation technique was applied, the structure matrix (Table 5.14) showed the loading of items onto two clear factors. All items loaded above the minimum threshold value of 0.4 (Finch 2006). Items were assigned the factor to which they had the highest loading. It was decided that as the loading values on both factors for item 9 were very similar (0.40 and 0.36), this item would be placed on the factor which best fits with the concept of the item.

Table 5.14: The structure matrix of the FROM showing the loading of each item onto the two factors extracted*

	Component	
	1	2
Q1		.676
Q2	.316	.706
Q4		.849
Q5		.770
Q8	.364	.534
Q9	.406	.364
Q11	.631	.307
Q12	.641	.417
Q16	.715	.446
Q18	.763	
Q20	.686	
Q22	.594	.383
Q23	.728	
Q26	.641	.357
Q30	.745	
Q31	.534	.410

*Extraction Method: Principal Component Analysis. Rotation Method: Varimax with Kaiser Normalization.

Final version of the FROM

The final version of the FROM was developed as a result of the family member feedback, Rasch and factor analysis, the final version of the FROM (Figure 5.12) was developed. Each of the changes made to develop the final version is outlined below.

Layout and design of the FROM

The developmental version of the FROM was three A4 pages long. Now that the number of items and response categories have been reduced, the FROM was redesigned to fit onto one side of one sheet of A4 paper. This is more practical for use in a clinical situation, as it reduces responder burden and reduces the chance of pages getting mislaid during photocopying or printing. The font size of the instructions and items was reduced by one font size, but this made very little visual difference to the measure. The grey lines separating items were narrowed in order for the items to fit onto one page. Feedback from family members also showed that they found having to turn the page over to answer more questions confusing and unclear, so reducing the FROM to one page helped eliminate this

problem. The use of large individual tick boxes for each response category was maintained as it was felt to be clear and had not caused confusion when completed by the family members.

Statements and instructions

The statements and instructions on the FROM were condensed and made clearer for the final version. The investigators felt that there was no need to have the instructions repeated twice on the questionnaire, and so they were included just once, at the top of the page. The bold type face was retained, to emphasise the recall period chosen, and the fact that the FROM is relating to the family member's life and not the patient's. Rather than keeping the extra statement during the introduction, "please remember, this questionnaire is about **your** life, not your family member's life", the phrase "Because of my family member's condition..." was added before each set of items, to draw the subject's attention back to the fact that the FROM is about their life, as several family members reported being confused about this. The demographic questions were retained; age, gender, relationship to patient and patient's diagnosis, although several family members found it difficult to complete the patient's diagnosis section. This is a question which could potentially be completed by the investigator or clinician. The final instruction, "Please check you have answered every question. Thank you" was retained as it clearly worded and may help to prevent missing data.

Utility questions

As discussed earlier in this chapter, the three utility questions from the developmental version of the FROM were deleted as they caused too much responder burden and were not comparable to the other 16 items.

Items

As a result of Rasch analysis, 16 items were retained for the final version of the FROM. 15 of the 16 items had not caused any problems to family members when completing the developmental version, so the wording remained the same. However, the wording of item 9 was given negative feedback by family members, particularly the use of the word "burden". Family members were reluctant to answer this item as, although they regarded caring for their family member as being "difficult" at times, they did not feel it was a "burden". This was reflected in the low endorsement statistics for this item. It was felt that item 9 measures an important concept, and one that was very prevalent during the interview stage, so the item was retained but the wording was changed. When family members gave feedback about this item, and during the interview transcripts, they often used the word "difficult", so this was included in the item and "burden" was removed. The item was re-worded to read "Caring for my family member is difficult".

Domains

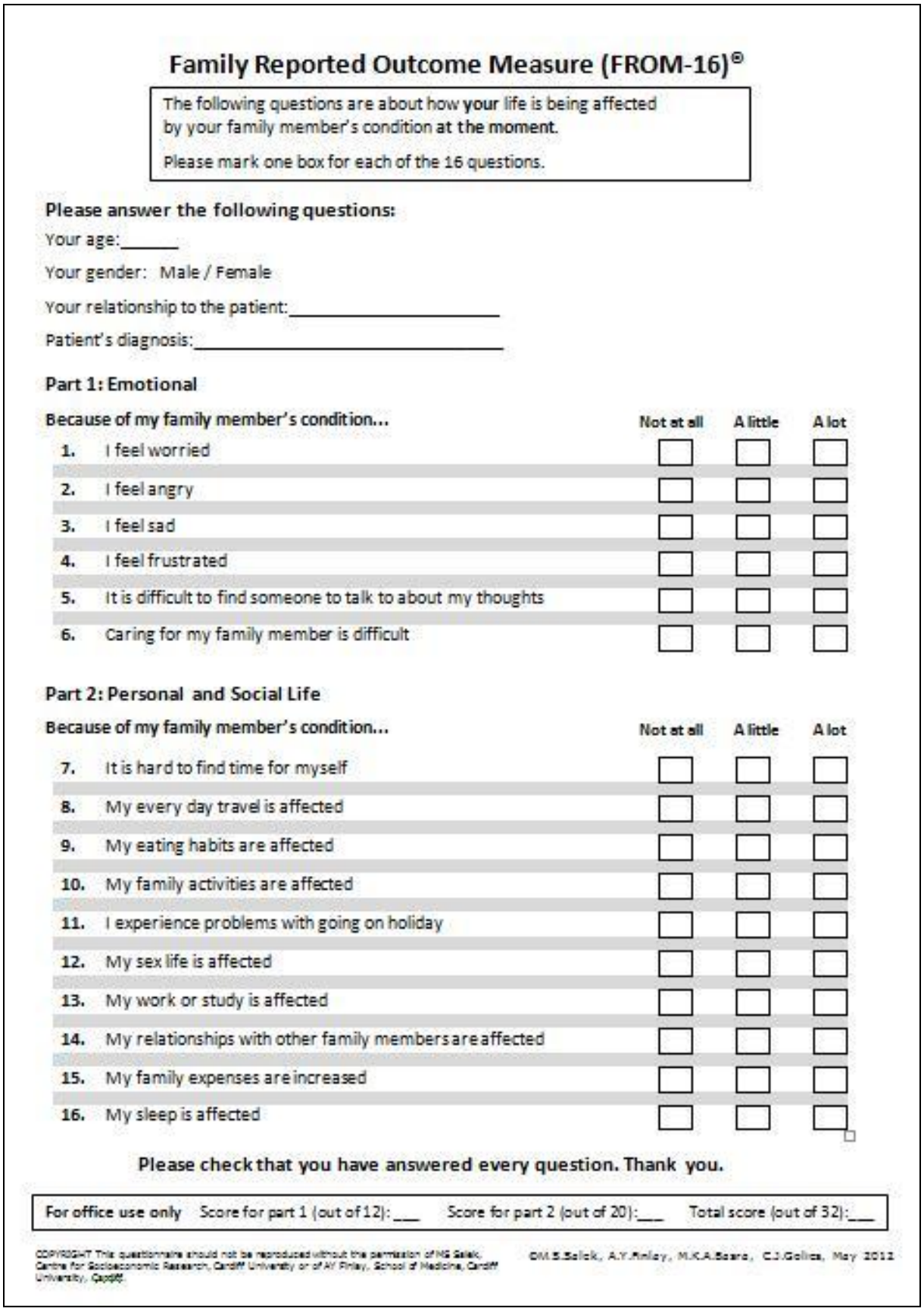
Factor analysis of the 16-item FROM revealed two factors. All of the 16 items loaded clearly and strongly onto one factor, apart from item 9 which loaded strongly onto both. The items loaded onto the two factors in a logical way according to their theme or construct; all of the similar items loaded onto one factor. The first factor, which was labelled “Emotional” loaded five items: Worried, angry, sad, frustrated, and difficult to talk to someone. The second factor, “Personal and Social Life” loaded 10 items: Hard to find time for self, travel, eating habits, family activities, holidays, sex life, work or study, relationships with other family members, family expenses and sleep. The emotional or psychological based items loaded clearly onto one factor, whilst the more social and daily aspects loaded onto the other. The naming of these factors is discussed further in the General Discussion chapter. Item 9 loaded strongly onto both factors and it was decided that it should be placed on the factor where it best fits the concept. Looking back to the interviews where the theme first emerged, family members described not just the physical impact of caring for the patient, but put a greater emphasis on the emotional effect this had on them and their relationship with the patient. Many family members associated emotions such as sadness and frustration with the caring aspects. This is reflected in the new wording of the item “Caring for my family member is difficult”, as it encompasses both the physical side (the caring) and the emotional side (the difficulty). The investigators felt that this item fitted better in the “Emotional” factor, which brought the item total for this factor to six items.

Scoring

The scoring for the final version of the FROM was amended, as the response categories were collapsed from five to three. The new categories were renamed as “not at all” (scoring 0), “A little” (scoring 1) and “a lot” (scoring 2). The naming of the new categories was felt to be balanced, and the names of the categories were easily distinguishable from one another, for example a clear difference can be seen between “A little” and “A lot”. The new scoring system gives the final version of the FROM a total score of 32, and total domain scores of 12 (six items in domain 1) and 20 (10 items in domain 2) respectively. An updated scoring box was added to the bottom of the FROM with space for the scores for each domain and the total score.

The 31-item developmental version of the FROM was successfully reduced to a 16-item final version (FROM-16), which fits well to the Rasch model and displays two clear factors (Figure 5.12). The final version of the FROM then underwent further validation, which will be described in Chapter 6.

Figure 5.12: The final 16-item version of the FROM (FROM-16)



Family Reported Outcome Measure (FROM-16)©

The following questions are about how **your** life is being affected by your family member's condition **at the moment.**

Please mark one box for each of the 16 questions.

Please answer the following questions:

Your age:_____

Your gender: Male / Female

Your relationship to the patient:_____

Patient's diagnosis:_____

Part 1: Emotional

Because of my family member's condition...	**Not at all**	**A little**	**A lot**
1. I feel worried	☐	☐	☐
2. I feel angry	☐	☐	☐
3. I feel sad	☐	☐	☐
4. I feel frustrated	☐	☐	☐
5. It is difficult to find someone to talk to about my thoughts	☐	☐	☐
6. Caring for my family member is difficult	☐	☐	☐

Part 2: Personal and Social Life

Because of my family member's condition...	**Not at all**	**A little**	**A lot**
7. It is hard to find time for myself	☐	☐	☐
8. My every day travel is affected	☐	☐	☐
9. My eating habits are affected	☐	☐	☐
10. My family activities are affected	☐	☐	☐
11. I experience problems with going on holiday	☐	☐	☐
12. My sex life is affected	☐	☐	☐
13. My work or study is affected	☐	☐	☐
14. My relationships with other family members are affected	☐	☐	☐
15. My family expenses are increased	☐	☐	☐
16. My sleep is affected	☐	☐	☐

Please check that you have answered every question. Thank you.

For office use only Score for part 1 (out of 12):___ Score for part 2 (out of 20):___ Total score (out of 32):___

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DISCUSSION

Item reduction of the 16-item FROM was carried out using a combination of traditional and modern approaches. The use of item response theory (Rasch analysis) indicated which items did not fit the statistical model desired for a Likert-scale instrument. Rather than relying solely on the statistical approach and removing "mis-fitting" items, the content validity data was referred back to in order to identify whether the poorly performing items were the same according to both statistics and family member feedback. These two forms of information were combined when deciding upon changes to be made to the developmental version of the FROM. Combining the two types of information helped to ensure that the resulting FROM measure was both statistically valid for use in the intended population and reflected the views and opinions of family members, as determined by the qualitative interview content and the content validity feedback. The types of changes made to the developmental FROM included collapsing response categories, re-wording items and removing items. The "problem" items identified by the Rasch analysis were also those which had been rated poorly by family members and experts during the content validity panels, and there were very few situations where conflicts arose between the Rasch analysis and family member feedback. To ensure that the final version of the FROM contained the most representative and statistically-sound combination of items, each problem item was removed in a stepwise process and the summary statistics of the measure were referred back to each time. At each stage, the investigator was aware of the influence of her judgment and opinion of each item, and a conservative approach was taken when deciding to remove items. At all stages, the results of the Rasch analysis was discussed with the study team, who provided their opinions on the decisions taken.

Factor analysis was used to confirm the results of the Rasch analysis and to identify the two factors in the FROM. All items strongly loaded onto one factor (item 9 loaded strongly onto both), supporting the fact that the most relevant items had been retained during Rasch analysis. Furthermore, items were grouped into two factors which reflected the themes identified during the earlier qualitative phase. The careful combination of Rasch and factor analysis, along with the information from the content validity panels and qualitative interviews helped to ensure that the final version of the FROM contains the items which are most relevant to family members, and that they are able to express their own experiences and opinions accurately and fully using the FROM-16.

SUMMARY

- The developmental version of the FROM was given to 140 family members to complete.

- The family members provided informal comments as to the content of the developmental FROM which assisted in the item reduction process.

- The three utility questions were dropped from the FROM due to poor feedback and low response rate.

- Rasch analysis was performed using a 13-step process.

- The Partial Credit Rasch model was selected.

- The overall summary statistics for the developmental version of the FROM showed a poor fit to the Rasch model (fit residual mean for items (S.D)= -0.22 (1.97), persons (S.D)= -0.19(1.34), $\chi 2$ p=0).

- The categories were collapsed for the questionnaire as a whole, giving a uniform scoring system and making the FROM score much easier for investigators and clinicians to calculate.

- Six items were found to be mis-fitting with fit residuals greater than +/- 2.5, and these were removed one at a time.

- Two items showed a low level of endorsement: items 9 (I feel the burden of caring for my family member) and 29 (My work or study is affected) and both were considered for removal.

- Items were removed or combined in a stepwise process, with the overall fit statistics consulted at each stage.

- 16 items were retained and tested for differential item functioning (DIF) by age and gender. Eight of the 16 items showed uniform DIF by either gender or age, or both. Both DIF by gender and age cancelled out at test level (p=0.6 and p=0.8 respectively).

- Only item 20 (My sex life is affected) showed non-uniform DIF, but it was retained as it was identified as a very important and frequently occurring theme by family members during the interview stage.

- The summary statistics for the final version of the 16-item FROM indicate that it has a good fit to the Rasch model. The mean (S.D) fit residual values were -0.36 (1.07) for items and -0.22 (1.01) for persons (Total $\chi^2 = 56.6$, df = 48, p = 0.18).

- Factor analysis was performed on the 16-item measure.

- The principle component analysis for the 16-item FROM revealed three factors with Eigenvalues ≥1, which together explained 61% of the variance.

- All items loaded above the minimum threshold value of 0.4. Items were assigned the factor to which they had the highest loading.

- The items loaded onto the two factors in a logical way according to their theme or construct.

- The first factor, “Emotional”, loaded five items: worried, angry, sad, frustrated, and difficult to talk to someone. The second factor, “Personal and Social Life” loaded 10 items: hard to find time for self, travel, eating habits, family activities, holidays, sex life, work or study, relationships with other family members, family expenses and sleep. Item 9 loaded highly onto both factors but its concept matched better to the “Emotional” factor, which bought the factor total to six items.

- The final version, the FROM-16 was then finalised and deemed ready for further validation.

CHAPTER 6

Evaluation of the psychometric properties of the 16-item Family Reported Outcome Measure (FROM-16)

INTRODUCTION

The previous chapters have proven that the FROM-16 shows high content validity. Before the measure can be used to collect data, it is important to ensure that it is measuring the concepts it is intended to measure and that it is accurate and reliable. As outlined in chapter 2, the FROM-16 should score highly in terms of reliability, validity and sensitivity to change, and this chapter outlines the tests which the FROM-16 was put through in order to demonstrate this. The chapter also looks at the scoring distribution of the FROM-16 and whether the scores vary by factors such as age or gender.

For the duration of this chapter, the term FROM refers to the 16-item final version of the instrument, FROM-16.

METHODS

Study population

The study population for the validation of the FROM was made up of family members of patients across 25 medical and surgical specialties at the University Hospital of Wales, University Hospital Llandough, Velindre Hospital and General Practice in Cardiff. The earlier stages of the study were carried out with patients from 26 specialties, but recruitment problems in the mental health specialty meant that it had to be dropped from this stage of the study. During recruitment for this stage of the study, the investigator aimed to recruit equal numbers from each of the 25 specialties, and it was decided that a minimum of five family members from each specialty would be approached for recruitment.

Recruitment of family members

During this stage of the study, family members and patients were recruited from outpatient clinics and wards. Patients were first approached by the consultant, and then the investigator to gain verbal consent to discussing the study. Patients and their family members were then taken to a private room and given the study information leaflet (Appendix O-R) to read. They were given the opportunity to ask any questions, and were then both asked to sign the consent form. At this stage, family members were also asked to consent to email or postal follow up, although they were still eligible to take part in the study if they declined. Demographic details were then collected from the patients and their family members. The patients were then asked to leave the room so that they did not influence their family member's responses to the questionnaires. The family member was asked to complete the FROM, which was timed. They were then asked to rate the patient's health on

a scale of 0-10 (global health score) and complete written feedback about the FROM. They were also asked to complete the WHOQOL-BREF questionnaire.

After 1-2 weeks, all 120 family members were contacted via post or email and asked to complete the FROM and the global health score again. They were provided with a freepost envelope, and a second follow up was sent a week later if no reply was given.

After 2 months, 25 of the family members were contacted again via post or email and asked to complete the FROM and the global health score again. They were provided with a freepost envelope, and a second follow up was sent a week later if no reply was given.

FROM scoring

The final version of the FROM comprises of 16 items with three response options for each, ranging from Not At All (scoring 0), A little (scoring 1) and A Lot (scoring 2). The lowest possible score of the FROM is 0 and the highest is 32. The higher the total score, the greater the effect on the family member's quality of life. The items in the FROM are divided into two parts (domains): Emotional (comprising of 6 questions, maximum score of 12) and Personal and Social Life (comprising of 10 questions, maximum score of 20). For the purposes of validation, the FROM score for each family member was reported as both two separate domains, and as one complete score.

RESULTS

Demographic characteristics of the study participants

131 family members were approached to take part in the study and nine declined to participate of whom seven did not have the time, and two for personal reasons. In addition, one family member decided to withdraw from the study before the questionnaires were complete, due to personal reasons and another subject was excluded due to incomplete answers on the questionnaires. The final validation was carried out using data from 120 family members of patients, from 25 specialties (Table 6.1). All 120 family members were asked to complete the FROM, the WHOQOL-BREF (a generic quality of life measure) and rate the patient's health from 0-10 (0 meaning the poorest health possible and 10 meaning the best health possible). Most participants were female (n=79, 66%) Caucasian (n=125, 94%), the spouse or partner of the patient (n=69, 52%), the parent (n=32, 24%) or the child (n=25, 19%) of the patient (Table 6.2). The remaining 5% were made up of a variety of other relatives. The mean age of family members was 54 years (range 17-85) and the mean age of patients was 52 years (range 1-90). The mean patient disease duration was 83 months (6 years and 11 months) ranging from one month to 40 years.

Table 6.1: The 25 specialties included in the validation stage of the study.

Cardiology	Infectious Diseases
Care of the Elderly	Neurology
Chronic Pain	Oncology
Colorectal Surgery	Ophthalmology
Dental Surgery	Orthopaedics & Paediatric Orthopaedics
Dermatology & Paediatric Dermatology	Paediatric Endocrinology
Ear, Nose and Throat	Post-stroke
Endocrinology	Renal & Renal transplant
Gastroenterology	Respiratory
General Practice	Rheumatology
Genetics	Urology
Gynaecology	Wound Healing
Haematology	

The FROM affirmation responses

Scores of 1 ("a little") and 2 ("a lot") were considered as affirmation responses for each item (Table 6.3). This indicated that the family member found this item to be relevant and applicable to them, rather than selecting "not at all". The most highly affirmed items were "I feel worried" and "I feel sad" and the lowest were "my eating habits are affected" and "my work or study is affected".

The FROM score

Total scores for the FROM (0-32) ranged from 1 to 32, median = 11.50, mean = 12.28 and SD = 7.47. There was no floor effect (scoring 0), and only one subject scored 32 showing a minimal ceiling effect. The mean total domain scores were 5.6 (Emotional) and 6.7 (Personal and Social Life). Table 6.4 and Figures 6.1 and 6.2 show the distribution of FROM total scores.

The majority of items (items 1,3,5,7,8,9,10,14,15 and 16) contained no missing data across the 120 family members who completed the measure. The remaining items contained only one or two cases of missing data, except for item 13 (work or study) where four family members had missing data. This still only represents 3% of missing data for this item, but suggests that the item may not be as acceptable to respondents as the other items. Looking at the content of the items and referring back to the qualitative work, this missing data may have arisen by family members not knowing how to answer the item if they are not currently working or study (for example if they are retired). This can be tested in future work when the FROM is further validated in individual disease areas, through cognitive debriefing interviews which assess the respondents understanding of the item wording and concepts. The low amount of missing data in the FROM overall, suggests that the measure is both acceptable to family members and that the scale used is feasible.

Table 6.2: Demographics of the family members during the validation stage

Total number of family members	120
Male	41
Female	79
Family members age (years)	
Mean	54
Median	53
Range	17-85
Relationship to patient	
Spouse/partner	52%
Parent	24%
Child	19%
Other[a]	5%
Family member level of education	
Less than secondary school/GCSEs/O Levels	4%
Completed secondary school/GCSEs/O Levels	40%
A Levels/college course	25%
University degree	13%
Masters degree	8%
Doctoral degree	2%
Prefer not to say	2%
Missing	6%
Family member household income per year	
Less than £10,000	11%
£10,000 - £20,000	26%
£21,000 - £30,000	15%
£31,000 - £40,000	13%
£41,000 - £50,000	7%
£51,000 - £60,000	3%
£61,000 - £70,000	4%
£71,000 - £80,000	1%
Greater than £81,000	4%
Prefer not to say	10%
Missing	6%
Ethnic origin	
Caucasian	94%
Asian/Asian British	3%
Afro-Caribbean	2%
Total number of patients	120
Male	55
Female	65
Patients' age (years)	
Mean	52
Median	57
Range	1-90
Disease duration (months)	
Mean	83
Median	48
Range	1-480

[a] Other includes nephew, grandparent, sibling and grandchild.

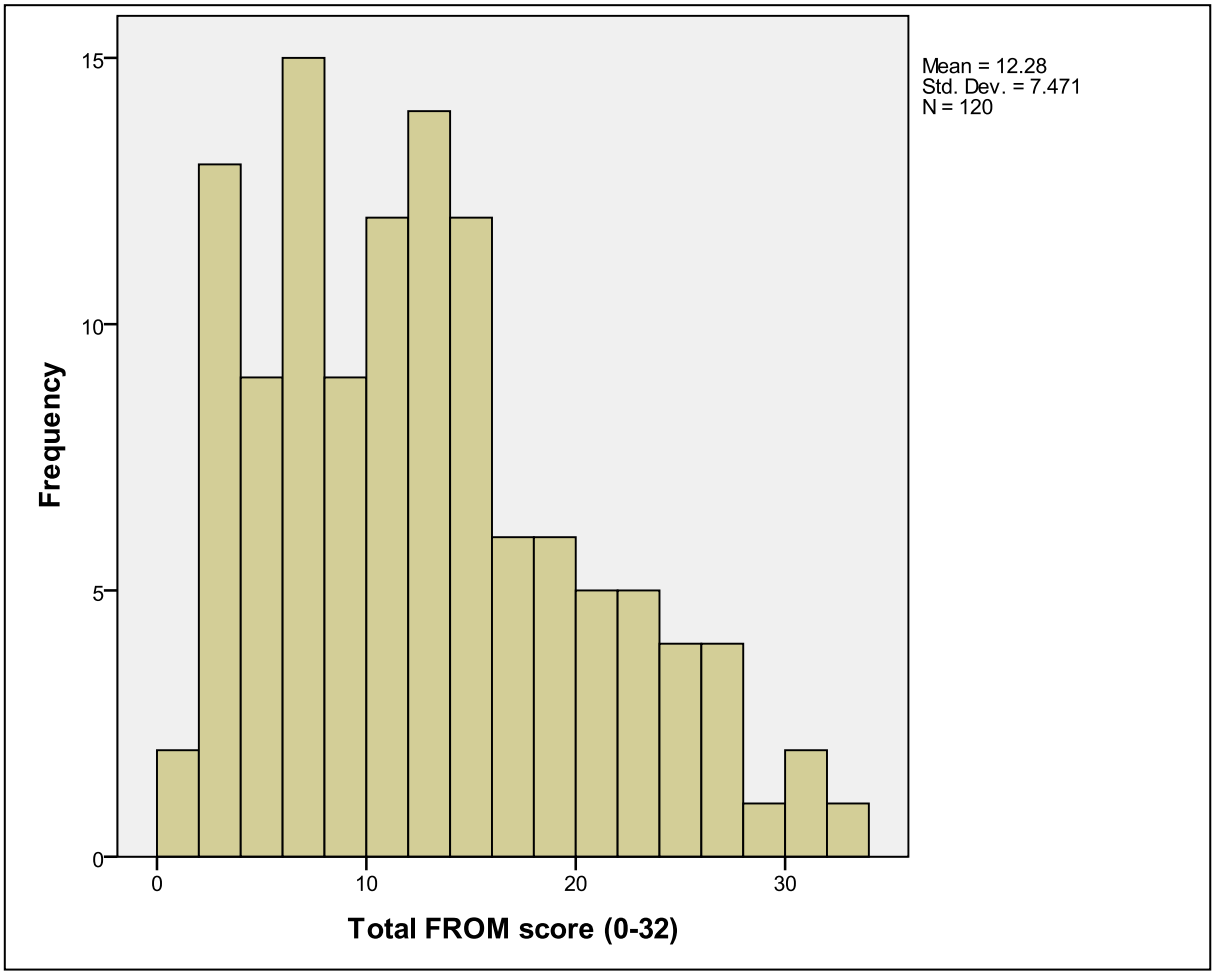
Table 6.3: The affirmation percentage for each FROM item (n=120)

FROM Items	Number of family members affirming to item (%)
Part 1 Worried	115 (95.1)
Angry	48 (39.6)
Sad	97 (80.2)
Frustrated	95 (78.5)
Talking about thoughts	55 (45.5)
Difficulty caring	72 (59.6)
Part 2 Time for self	62 (51.3)
Travel	52 (42.9)
Eating habits	41 (33.9)
Family activities	87 (71.9)
Holiday	63 (52.1)
Sex life	51 (42.1)
Work or study	47 (38.8)
Family relationships	55 (45.5)
Family expenses	70 (57.9)
Sleep	77 (63.6)

Table 6.4: Frequency of the total FROM scores (n=120)

Score	Frequency	Percent	Cumulative Percent
0	0	0	0
1	2	1.7	1.7
2	8	6.7	8.3
3	5	4.2	12.5
4	2	1.7	14.2
5	7	5.8	20.0
6	3	2.5	22.5
7	12	10.0	32.5
8	4	3.3	35.8
9	5	4.2	40.0
10	10	8.3	48.3
11	2	1.7	50.0
12	9	7.5	57.5
13	5	4.2	61.7
14	5	4.2	65.8
15	7	5.8	71.7
16	3	2.5	74.2
17	3	2.5	76.7
18	3	2.5	79.2
19	3	2.5	81.7
20	3	2.5	84.2
21	2	1.7	85.8
22	1	.8	86.7
23	4	3.3	90.0
24	4	3.3	93.3
26	4	3.3	96.7
29	1	.8	97.5
31	2	1.7	99.2
32	1	.8	100.0
Total	120	100.0	

Figure 6.1: Histogram to illustrate total FROM score and frequency



The scoring for each individual item was also looked at. The most highly scoring items based on their mean score (with a possible range of 0-2) were feeling worried (1.42), feeling sad (1.13), feeling frustrated (1.17), family activities being affected (0.93) and effect on sleep (0.90). The mean scores for each of the 16 items are shown in Figure 6.3.

Comparisons of the FROM scores for sociodemographic characteristics of family members and patients

Family member gender

Although the FROM mean total score varied slightly between males (11.83) and females (12.52), there was no significant difference between their scores (p=0.63). For individual items, there was no significant difference in the mean scores for males and females (p= 0.24-0.95), except for one item, “effect on sleep” (Table 6.5 and Figure 6.4). For this item, the mean individual item score for females was significantly higher than that of males (M= 0.63, F= 1.04, p<0.05). For the two domains (Part 1 and Part 2) of the FROM, there was no

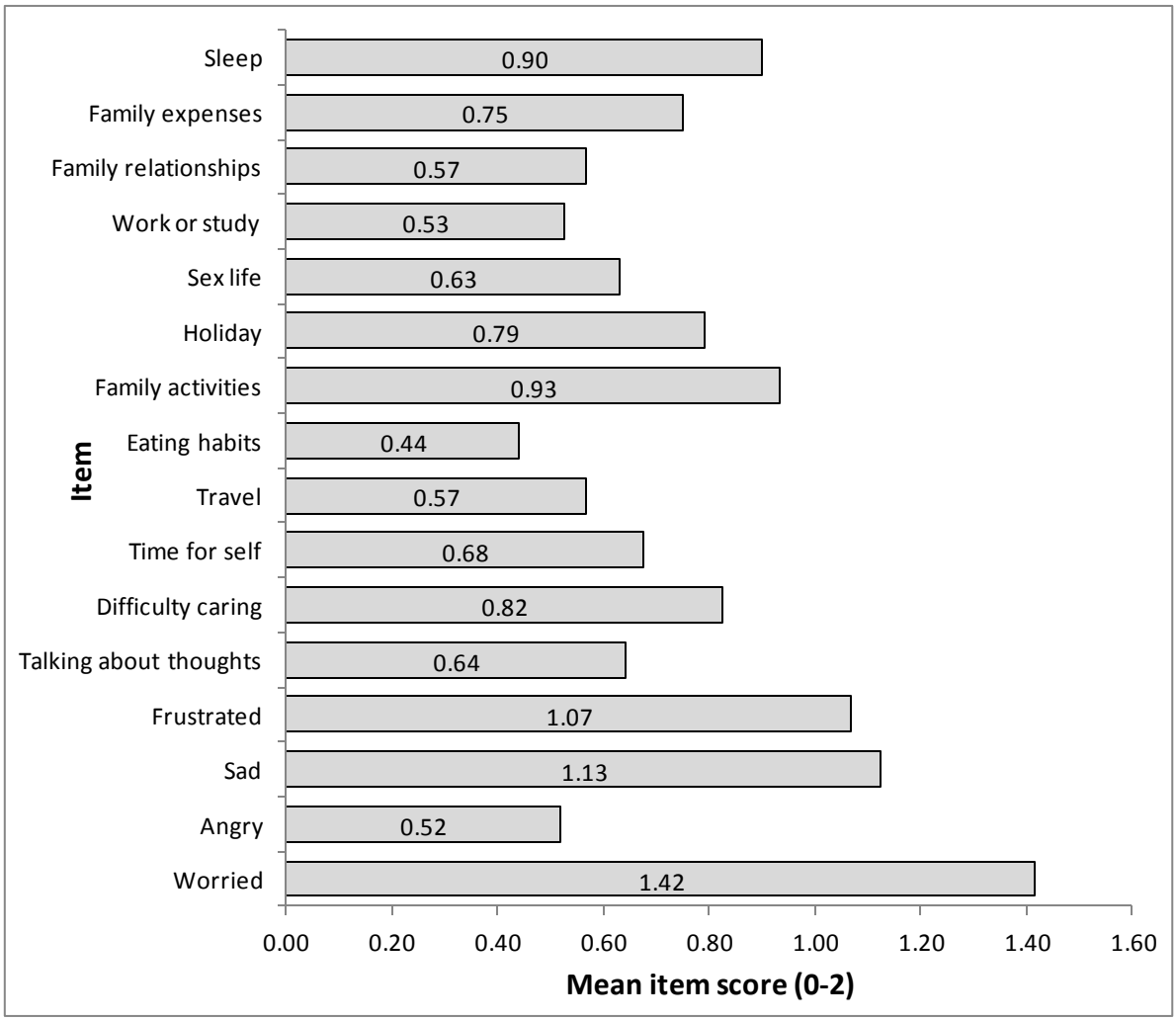
significant difference in the scores for males and females in each domain (Part 1 p= 0.95, Part 2 p= 0.37) (Table 6.5).

Figure 6.2: A box and whisker plot to illustrate distribution of the FROM total score (n=120)

Patient gender

Both the total FROM scores and the individual items scores were also compared for the gender of the related patients. The FROM mean total score of family members of male patients (12.73) was slightly higher than that of female patients (11.91), but this difference was not significant (p= 0.55) (Table 6.6). There was no significant difference in individual item scores for family members of male and female patients (p= 0.083-0.979) (Table 6.6). For the two domains (Part 1 and Part 2) of the FROM, there was no significant difference in the scores for family members of male and female patients in each domain (Part 1 p= 0.84, Part 2 p= 0.42) (Table 6.6).

Figure 6.3: The mean scores for individual items of the FROM (n=120)



Family member age

The FROM mean score for family members' age was calculated by dividing the family members up into six age groups. Group 1=16-30 years, group 2=31-40 years, group 3=41-50 years, group 4=51-60 years, group 5=61-70 years, group 6≥ 71 years. The FROM mean total scores across the six groups were calculated (Table 6.7) and, as there were six groups, a one-way analysis of variance (ANOVA) was carried out to compare the mean scores. This was followed by a post-hoc test to identify which age groups, if any, differed most by their mean.

Table 6.5: Comparison of family members' mean total and individual item scores for their own gender.

FROM item	Family member gender	N (family members)	Mean	SD	p value
Worried	Male	41	1.37	0.58	0.49
	Female	79	1.44	0.57	
Angry	Male	41	0.56	0.71	0.61
	Female	77	0.49	0.68	
Sad	Male	41	1.15	0.65	0.81
	Female	79	1.11	0.73	
Frustrated	Male	41	1.07	0.72	0.95
	Female	77	1.06	0.66	
Talking about thoughts	Male	41	0.59	0.71	0.57
	Female	79	0.67	0.81	
Difficulty caring	Male	41	0.93	0.76	0.29
	Female	78	0.77	0.77	
Time for self	Male	41	0.59	0.71	0.35
	Female	79	0.72	0.77	
Travel	Male	41	0.61	0.80	0.64
	Female	79	0.54	0.69	
Eating habits	Male	41	0.37	0.66	0.38
	Female	79	0.48	0.68	
Family activities	Male	41	0.90	0.66	0.27
	Female	79	0.95	0.71	
Holiday	Male	40	0.85	0.83	0.58
	Female	79	0.76	0.85	
Sex life	Male	40	0.68	0.86	0.67
	Female	79	0.61	0.78	
Work or study	Male	40	0.43	0.59	0.34
	Female	76	0.58	0.77	
Family relationships	Male	41	0.51	0.64	0.54
	Female	79	0.59	0.73	
Family expenses	Male	41	0.66	0.66	0.33
	Female	79	0.80	0.77	
Sleep	Male	41	0.63	0.66	0.008
	Female	79	1.04	0.82	
FROM Score	Male	41	11.83	6.96	0.63
	Female	79	12.52	7.76	
FROM Part 1 Score	Male	41	5.66	2.99	0.84
	Female	79	5.54	3.01	
FROM Part 2 Score	Male	41	6.17	4.85	0.42
	Female	79	6.97	5.37	

Figure 6.4: Mean FROM score for individual items for male and female participants

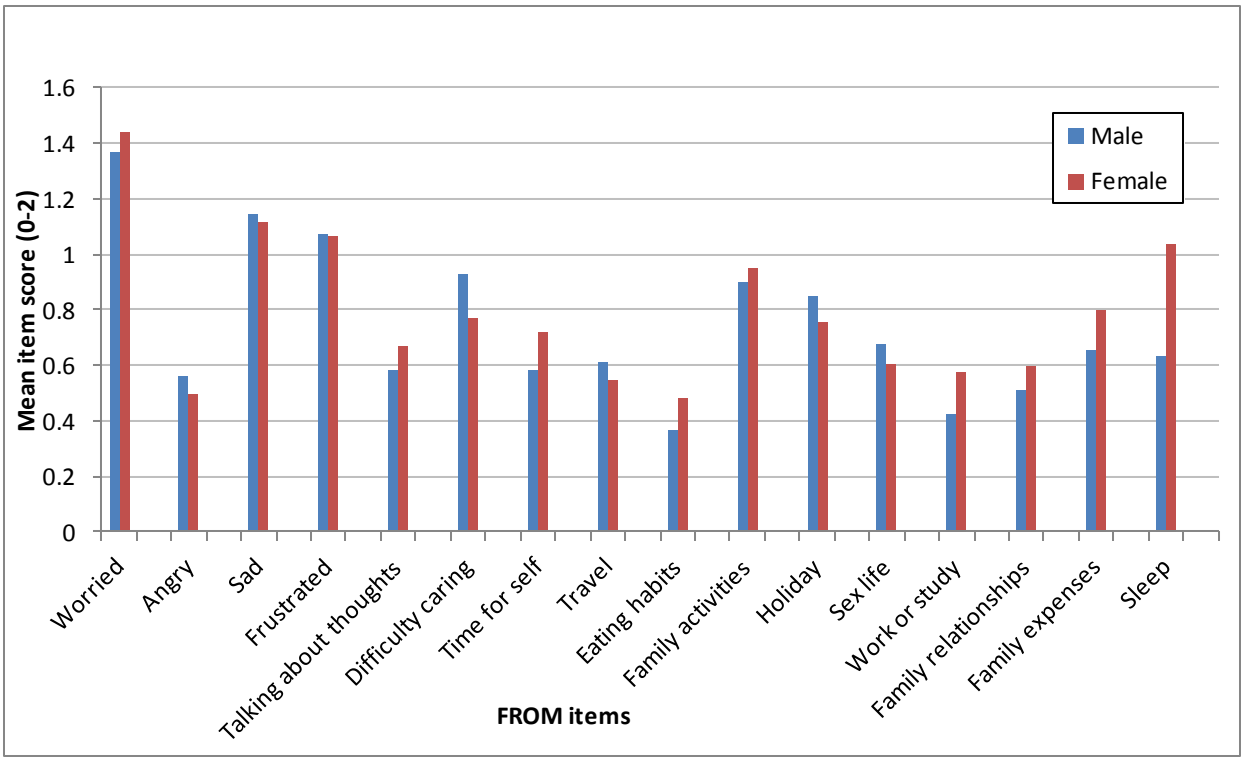


Figure 6.5 shows that the age group who experienced greatest impact on their life was Group 4 (51-60 years) with a mean FROM score of 14.93, and the group who experienced least impact was Group 2 (31-40 years) with a mean FROM score of 9.6. Results of the ANOVA test showed that there was no significant difference in the mean FROM total score across the six age groups (p=0.33) (Table 6.7).

An ANOVA test was also carried out on the scores (0-2) for the individual items of the FROM, to investigate whether there was a significant difference in mean score for the individual items across the six age groups. There was a significant difference between family member age groups for three of the FROM items: Holidays (p=0.045), Sex life (p=0.045) and Work and Study (p=0.022). The Levene Test for Homogeneity of Variances was carried out to test the equality of the variances, as the ANOVA test assumes equal variance. The item "Holidays" (p=0.327) was found to have a non-significant Levene F statistic value, meaning that equality of variance can be assumed, and the ANOVA test value can be taken as correct. The items "Sex Life" (p=0.005) and "Work and Study" (p<0.001) were found to have a significant Levene F statistic significance value (p<0.05), and the difference in mean item score between age groups was calculated using Welch's t-test (assuming unequal variance). The Welch test showed that the item "Work and Study" showed significant differences in item score between the six age groups (p= 0.031), but the difference in score for age groups for the item "Sex Life" was not significant (p=0.053). The only other item to show evidence of

unequal variance (Levene F statistic value of p=0.018), was the item “Worried”, which went on to show no significant difference in the mean score across the six age groups as a result of the Welch test (p=0.178).

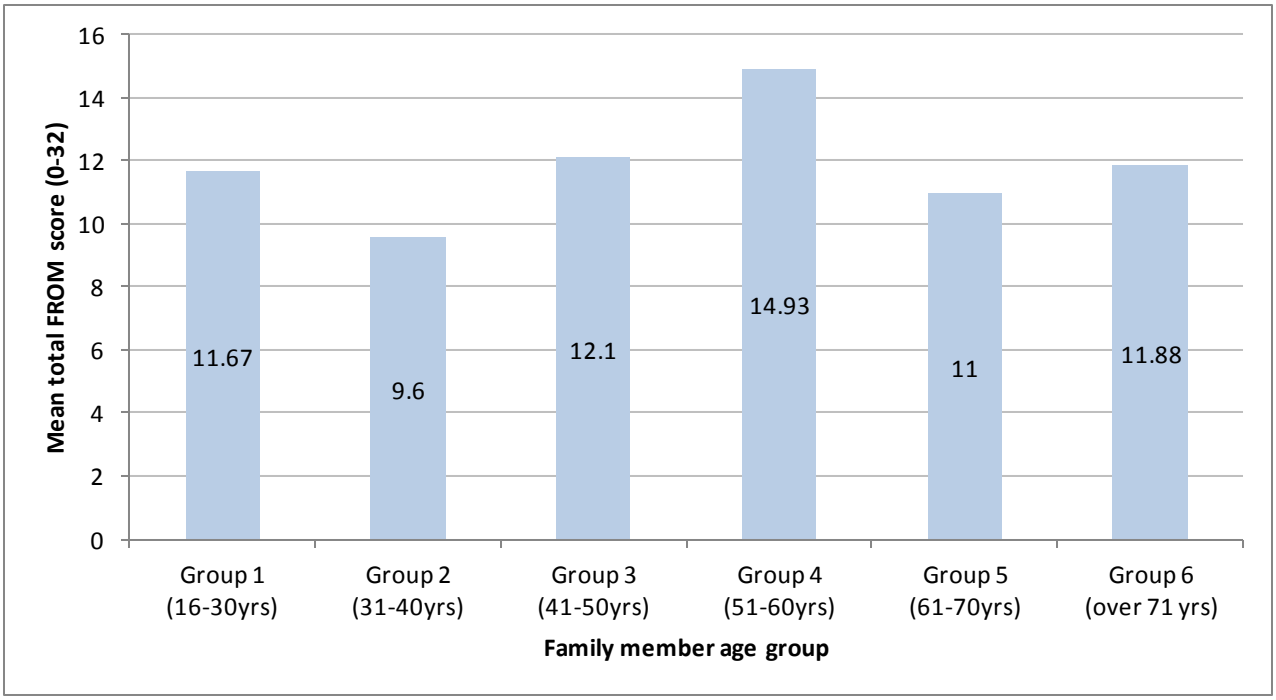
Table 6.6: Family members’ mean total and individual item scores for patients’ gender.

FROM item	Patient gender	N (patients)	Mean	SD	p value
Worried	Male	55	1.42	0.60	0.98
	Female	65	1.42	0.56	
Angry	Male	53	0.45	0.67	0.36
	Female	65	0.57	0.71	
Sad	Male	55	1.04	0.69	0.21
	Female	65	1.20	0.71	
Frustrated	Male	53	1.08	0.68	0.91
	Female	65	1.06	0.68	
Talking about thoughts	Male	55	0.67	0.80	0.69
	Female	65	0.62	0.76	
Difficulty caring	Male	54	0.83	0.75	0.90
	Female	65	0.82	0.79	
Time for self	Male	55	0.80	0.78	0.10
	Female	65	0.57	0.71	
Travel	Male	55	0.58	0.71	0.84
	Female	65	0.55	0.75	
Eating habits	Male	55	0.49	0.69	0.46
	Female	65	0.40	0.66	
Family activities	Male	55	1.02	0.68	0.22
	Female	65	0.86	0.70	
Holiday	Male	55	0.75	0.82	0.60
	Female	64	0.83	0.87	
Sex life	Male	54	0.72	0.81	0.26
	Female	65	0.55	0.79	
Work or study	Male	53	0.62	0.77	0.18
	Female	63	0.44	0.67	
Family relationships	Male	55	0.53	0.72	0.57
	Female	65	0.60	0.68	
Family expenses	Male	55	0.78	0.76	0.67
	Female	65	0.72	0.72	
Sleep	Male	55	1.04	0.82	0.08
	Female	65	0.78	.76	
FROM Score	Male	55	12.73	7.61	0.55
	Female	65	11.91	7.39	
FROM Part 1 Score	Male	55	5.56	3.07	0.95
	Female	65	5.60	2.94	
FROM Part 2 Score	Male	55	7.16	5.35	0.37
	Female	65	6.31	5.06	

Table 6.7: The FROM mean total scores for each category of family member's age

Family member age group	Number of family members	Mean FROM total score	Std. Deviation	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
16-30	12	11.7	9.4	5.7	17.7	1	29
31-40	10	9.6	4.4	6.5	12.7	2	15
41-50	29	12.1	6.5	9.6	14.6	3	26
51-60	29	14.9	8.4	11.7	18.1	2	31
61-70	24	11.0	7.6	7.8	14.2	1	32
Over 71	16	11.9	6.8	8.2	15.5	2	26
Total	120	12.3	7.5	10.9	13.6	1	32

Figure 6.5: The FROM mean scores for each age category of family members



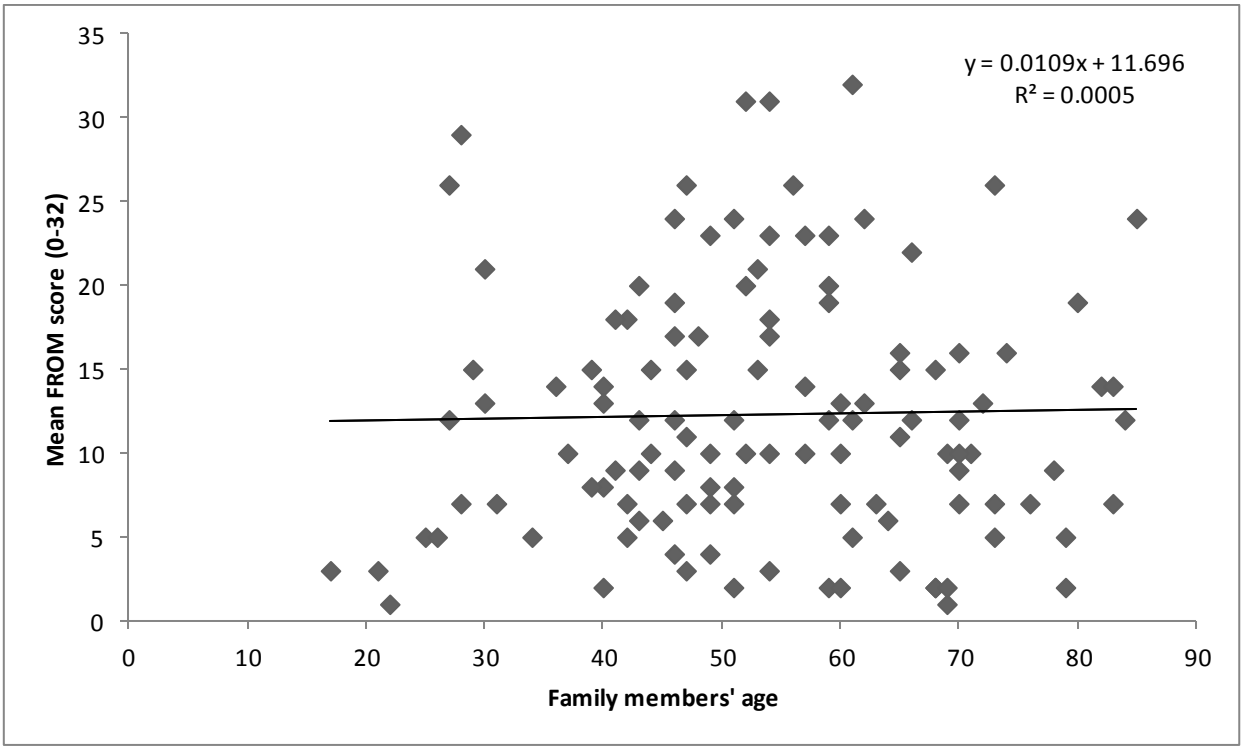
Therefore, the items "Holiday" and "Work and Study" which were identified to have significant differences in their mean scores by age underwent a Tukey post-hoc analysis. For the item "Holiday", the main difference was between age groups 2 and 4, and for "Work and Study", the main difference was between age groups 4 and 5. This latter observation may be explained by the fact that the majority of the subjects in group 6 (over the age of 71) were retired, and therefore the patient's illness would have less of an effect on their work life.

The mean score of the two domains of the FROM ("Emotional" and "Personal and Social Life") were also analysed according to family member age using the ANOVA test. The

"Emotional" domain was found to have homogeneity of variances (p=0.80) and no significant difference in the mean score across the six age groups (p=0.89), however the "Personal and Social Life" domain was found to have unequal variances (p=0.03) and a statistically significant Welch test value (p=0.04). This suggests that the emotional effects on family members of patients are similar across all ages, but the effects on personal and social life differ by age, with the main differences between group 5 (51-60yrs) and all of the other groups.

Spearman's Rank Order Correlation coefficient showed that there was no significant correlation between the family members' age and the total FROM score (r= 0.02, p= 0.80). This shows that family members of all ages are affected by illness, and that neither older or younger family members are likely to be affected more greatly. Figure 6.6 shows the relationship between family members' age and total FROM score.

Figure 6.6: The relationship between family members' age and FROM total score



Patient age

The FROM mean score for patients' age was also calculated by dividing the patients up into eight age groups. Group 1= 0-10 years, group 2= 11-20 years, group 3= 21-30 years, group 4= 31-40 years, group 5= 41-50 years, group 6= 51-60 years, group 7= 61-70 group 8 ≥ 71 years. The mean total FROM scores across the six groups were calculated (Table 6.8 and Figure 6.7).

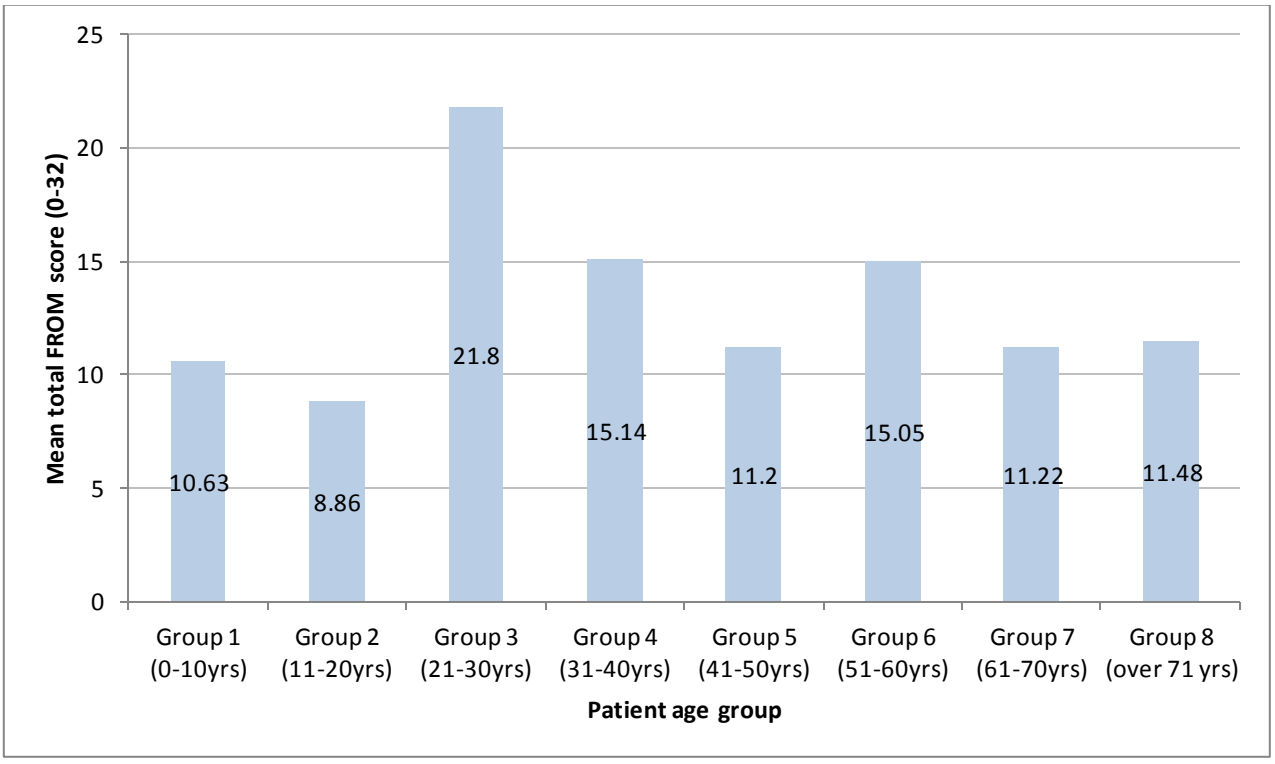
Table 6.8: The mean total FROM scores for each patient age category

Patient age group	Number of patients	Mean FROM total score	Std. Deviation	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
0-10	8	10.6	10.4	2.0	19.3	1	29
11-20	14	8.9	5.3	5.8	11.9	2	21
21-30	5	21.8	7.9	12.1	31.6	13	32
31-40	7	15.1	7.3	8.4	21.9	2	23
41-50	15	11.2	6.8	7.4	15.0	3	26
51-60	20	15.1	8.9	10.9	19.2	2	31
61-70	18	11.2	5.5	8.5	14.0	2	24
Over 71	33	11.5	6.5	9.2	13.8	2	26
Total	120	12.3	7.5	11.0	13.6	1	32

Figure 6.7 shows that the most affected family members were those with patients in Group 3 (21-30 years) with a mean FROM score of 21.8, and the family members who experienced least impact were those with patients in group 2 (11-20 years) with a mean FROM score of 8.86. Results of the ANOVA test showed that there was a significant difference in the FROM mean total score across the eight age groups (p=0.019). The post-hoc analysis showed that the main difference was found between group 2 and group 3 (p= 0.016). The mean score of individual items of the FROM were then compared according to patients' age. Nine of the 16 items showed equal variance, so the ANOVA test was performed. The other seven showed unequal variance, so were analysed using Welch's t-test. Of the 16 items, four showed significant differences between the patient age groups. These items were "Worried" (p= 0.008), "Talking about thoughts" (p= 0.006), "Eating habits" (p= 0.008) and "Family expenses" (p= 0.029).

The mean score of the two domains of the FROM ("Emotional" and "Personal and Social Life") were also analysed according to patient age using the ANOVA test. The "Emotional" domain was found to have homogeneity of variances (p=0.217) and a significant difference in the mean score across the eight age groups (p=0.004), however the "Personal and Social Life" domain was found to have unequal variances (p=0.018) and a non significant Welch test value (p=0.313). This suggests that the effects on family members' personal and social lives are not likely to differ depending on the age of the patient, but that family members with certain patient age groups are more likely to suffer from emotional effects. Family members with patients in group 3 (21-30 yrs) were more likely to suffer emotional effects (mean "Emotional" domain score= 9.6 out of 12), than those with patients in groups 1 (age 0-10yrs, mean score= 4.38 out of 12) and 8 (age ≥ 71, mean score= 5.03 out of 12).

Figure 6.7: The FROM mean scores for each patient age category



Spearman's Rank Order Correlation coefficient showed that there was no significant correlation between the patients' age and the total FROM score (r= 0.01, p= 0.88). This shows that family members of patients of all ages are affected by illness, and that neither family members of older or younger patients are likely to be affected more. Figure 6.8 shows the relationship between patients' age and total FROM score.

Family member type

In order to compare the mean total FROM scores between the different relatives of patients, family members were divided into five groups. Group 1= Partner (unmarried), Group 2= Spouse, Group 3= Child, Group 4= Parent, Group 5= Other (made up of more distant relatives including grandparents, grandchildren, nieces and nephews). The FROM mean total score was then calculated for each of the five groups. Table 6.9 shows that the highest mean total FROM scores were found in unmarried partners (13.45), closely followed by spouses (12.69) and parents (12.66). The lowest mean scores were found in the category "Other" which included more distant relatives. The results of an ANOVA test found that there was not a significant difference between the FROM mean total score with respect to the relationship between the patient and the family member (p=0.15).

Figure 6.8: The relationship between patients' age and total FROM score

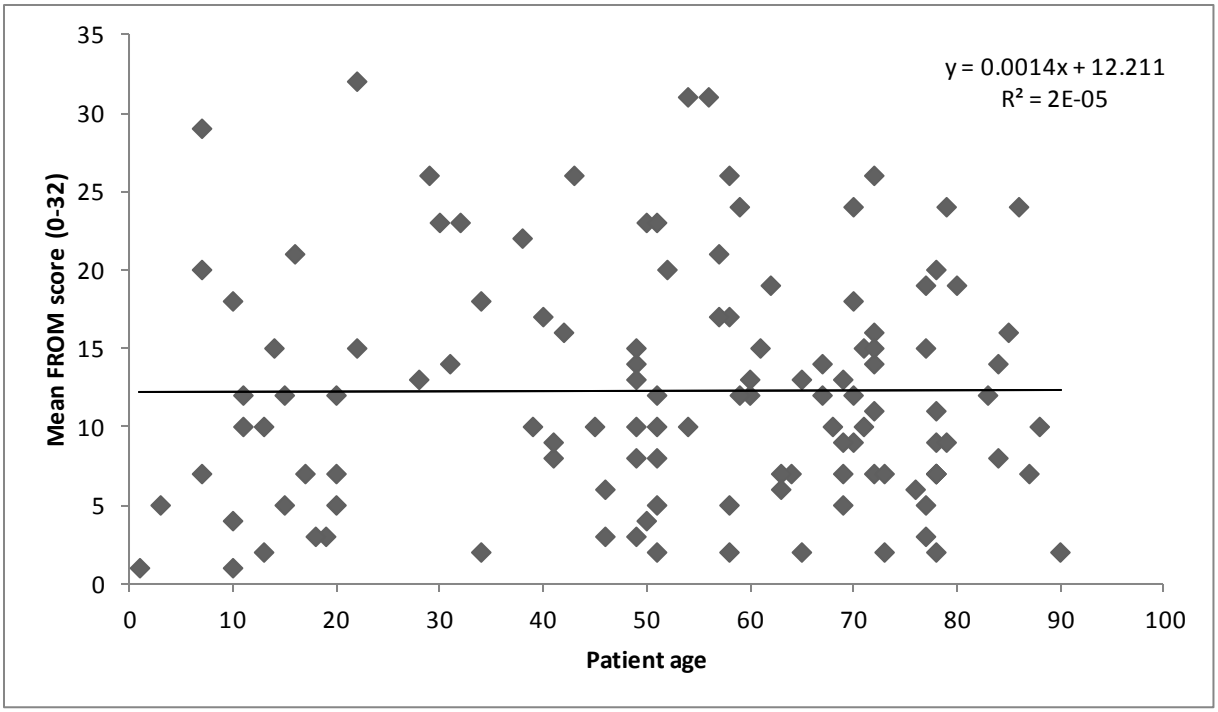


Table 6.9: The FROM mean total scores for each family member relationship category

Relationship to patient	N.o of family members	Mean FROM total score	S.D.	95%Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
Partner	11	13.5	6.3	9.2	17.7	4	23
Spouse	52	12.7	8.1	10.4	15.0	2	31
Child	24	12.0	5.8	9.5	14.4	2	24
Parent	29	12.7	7.9	9.7	15.7	1	32
Other	4	3.0	1.6	0.4	5.6	1	5
Total	120	12.3	7.5	11.0	13.6	1	32

Medical specialty

Although the numbers of family members recruited from each medical specialty was small (around 5 from each), the mean total FROM scores were still calculated for each specialty. Although the numbers are too small to draw any firm conclusions, this test gives a preliminary idea of which family members are most greatly affected across the different disease areas. Only one family member was sampled from the genetics specialty at this stage of the study, and so this subject was excluded for this analysis (number of family members=119, number of specialties= 24) (Table 6.10). The highest mean scores were

found in family members of neurology patients (19.8), oncology patients (17.6), haematology patients (16.6) and chronic pain patients (16.6) (Figure 6.9). The lowest mean scores were found in family members of ophthalmology patients (4.25) and orthopaedics (5.80) (Figure 6.9). As expected, the variances were found to be significantly unequal (Levene statistic p=0.028), and Welch's t-test showed that there were significant differences in the mean values between specialties (p=0.01). This difference was expected, as illnesses from different specialties result in different symptoms, have different treatment courses and vary in severity, all of which can affect family members in different ways.

The mean total FROM scores for family members of patients from each of the 24 specialties were also compared with the total scores for each of the two domains of the FROM ("Emotional" and "Personal and Social Life"). Table 6.11 shows the top five and bottom five specialties with regard to the mean total score (0-32), Part 1 (0-12) and Part 2 (0-20). This shows that family members of patients from neurology, chronic pain, oncology and haematology are likely to be affected both emotionally and socially as a result of the patient's illness. Although family members of patients from urology score in the top five for Part 1 "Emotional", they do not appear in Part 2 "Personal and social life", suggesting that they are more likely to be affected emotionally than socially. Although general practice scored close to the mid-point for total FROM score (15th out of 24), it scored lowly for Part 1, suggesting that family members are less affected emotionally, possibly because of the less urgent nature of long term conditions followed up in general practice. Family members of patients from both ophthalmology and orthopaedics scored lowly on both domains and the total FROM score, suggesting that family members are least affected by these conditions.

Number of patient co-morbidities

The number of co-morbidities the patient suffered from was correlated with the total FROM score. The aim of this test was to assess whether the number of different illnesses the patient has affects the quality of life of the family member. The correlation between number of co-morbidities and FROM score was 0.25 (p= 0.007), suggesting a weak positive correlation, and a weak association between the number of patient co-morbidities and the FROM score. This suggests that family members of patients with a large number of illnesses can be more greatly affected, but that this association only happens some of the time.

Table 6.10: The FROM mean total scores for family members of patients from each of the 24 specialties

Specialty	N.o of family members	Mean FROM total score	S.D.	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
Haematology	5.0	16.6	8.9	5.6	27.6	6.0	24.0
General practice	5.0	11.0	2.6	7.7	14.3	7.0	14.0
Oncology	5.0	17.6	5.9	10.2	25.0	8.0	24.0
Cardiology	5.0	9.8	4.8	3.8	15.8	5.0	15.0
Neurology	5.0	19.8	3.4	15.6	24.1	15.0	23.0
Colorectal surgery	5.0	11.6	6.5	3.5	19.7	7.0	23.0
Paediatric Endocrinology	5.0	9.8	4.0	4.9	14.7	5.0	15.0
Elderly	5.0	9.4	9.9	-2.9	21.7	2.0	26.0
Orthopaedics	5.0	5.8	7.1	-3.0	14.6	1.0	18.0
Rheumatology	5.0	11.4	11.6	-3.0	25.8	2.0	26.0
Gastroenterology	5.0	14.4	7.2	5.4	23.4	7.0	26.0
Renal	5.0	11.4	7.6	1.9	20.9	2.0	23.0
Urology	5.0	14.0	3.8	9.3	18.7	9.0	19.0
Chronic Pain	5.0	16.6	11.9	1.8	31.4	5.0	32.0
ENT	5.0	13.6	10.2	1.0	26.2	5.0	31.0
Respiratory	5.0	12.4	6.8	4.0	20.9	5.0	21.0
Infectious Diseases	5.0	10.0	6.2	2.3	17.8	2.0	17.0
Dental surgery	5.0	10.4	6.3	2.5	18.3	3.0	20.0
Dermatology	5.0	10.2	11.0	-3.4	23.8	2.0	29.0
Post-stroke	5.0	11.8	6.7	3.5	20.1	3.0	19.0
Wound healing	5.0	14.2	7.6	4.8	23.6	3.0	24.0
Ophthalmology	4.0	4.3	2.6	0.1	8.4	2.0	7.0
Diabetes	5.0	12.6	2.3	9.7	15.5	10.0	15.0
Gynaecology	5.0	10.8	3.4	6.6	15.1	7.0	16.0
Total	119.0	12.1	7.3	10.8	13.5	1.0	32.0

Figure 6.9: The FROM mean total score for family members of patients across 24 different specialties

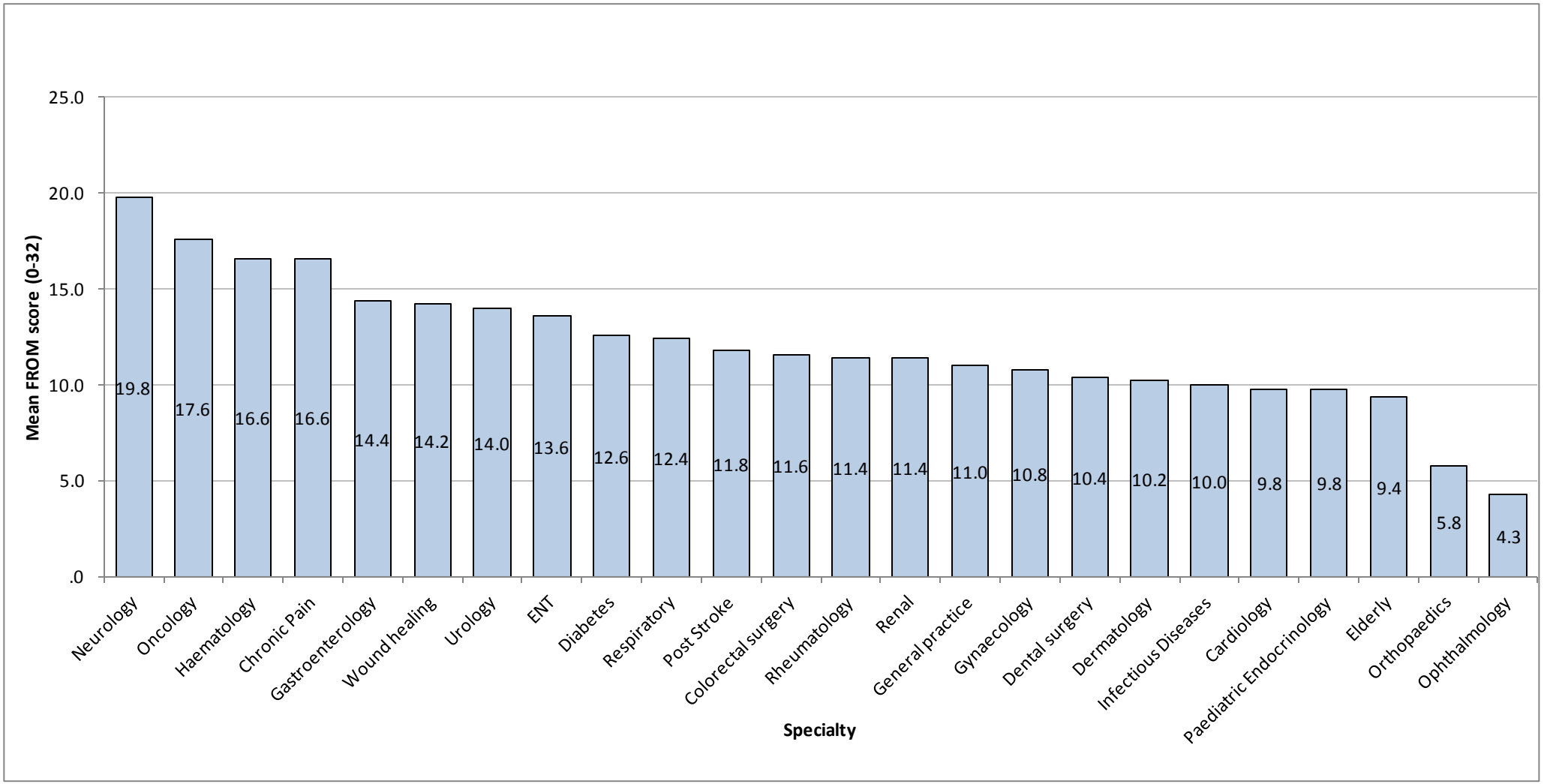


Table 6.11: The top five and bottom five specialties with regard to mean total FROM score and total score for each domain

	Mean total FROM score (0-32)		Mean total Part 1 score (0-12)		Mean total Part 2 score (0-20)	
Top 5 specialties	Neurology	19.8	Neurology	8.0	Neurology	11.8
	Oncology	17.6	Chronic Pain	7.6	Oncology	10.8
	Haematology	16.6	Haematology	7.0	Haematology	9.6
	Chronic Pain	16.6	Wound Healing	7.0	Chronic Pain	9.0
	Gastroenterology	14.2[a]	Urology	6.8[b]	Gastroenterology	8.8
	Wound Healing	14.2[a]	Oncology	6.8[b]		
Bottom 5 specialties	Ophthalmology	4.3	Ophthalmology	3.0	Ophthalmology	1.3
	Orthopaedics	5.8	Orthopaedics	3.2	Orthopaedics	2.6
	Elderly	9.4	Paediatric endocrinology	4.2	Infectious diseases	3.8
	Paediatric endocrinology	9.8	General Practice	4.2	Cardiology	4.6
	Cardiology	9.8	Diabetes	4.4	Gynaecology	4.8[c]
					Elderly	4.8[c]

[a] Both Paediatric endocrinology and Cardiology had the same mean total FROM score so are both listed in position 5

[b] Both Urology and Oncology had the same mean score for Part 1 so are both listed in position 5

[c] Both Gynaecology and Elderly had the same mean score for Part 1 so are both listed in position 5

Socioeconomic status and FROM scores

Socioeconomic data was collected from all subjects during this stage of the study. Tests were carried out on this data to investigate whether annual household income or level of education influenced the total FROM score.

The family members' level of education was divided into five groups. Group 1= Less than secondary school education, group 2= Secondary school education, group 3= A Levels/college course, group 4= University degree, group 5= Masters degree or above. Those family members who had chosen "Prefer not to say" or had missing values for education level were eliminated (n=110). Table 6.12 shows the mean total FROM score for each education level group. Although the family members in groups "A levels/college" and "Masters degree or above" had the highest mean total score (13.63 and 12.82 respectively), there was no trend towards higher scores depending on education level, and an ANOVA test showed that the difference in mean score between the groups was not significant (p=0.29).

Table 6.12: The mean total FROM score for family members according to level of education

Family member education level	Number of family members	Mean FROM total score	Std. Deviation	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
Less than secondary education	5.0	7.0	2.8	3.5	10.5	4.0	10.0
Secondary school education	48.0	12.3	7.5	10.1	14.5	1.0	29.0
A levels/college	30.0	13.6	8.2	10.6	16.7	2.0	32.0
University degree	16.0	10.1	7.2	6.2	13.9	1.0	24.0
Masters degree or above	11.0	12.8	5.4	9.2	16.4	7.0	23.0
Total	110.0	12.1	7.4	10.7	13.5	1.0	32.0

The family members' total household income per year was divided into eight groups. Group 1= Less than £10,000, group 2= £11,000-£20,000, group 3= £21,000-£30,000, group 4=£31,000-£40,000, group 5=£41,000-£50,000, group 6=£51,000-£60,000, group 7=£61,000-£70,000, group 8= over £71,000. Those family members who had chosen "Prefer not to say" or had missing values for household were eliminated (n=100). Table 6.13 shows the mean total FROM score for each household income level group. Although the family members in groups "£21,000-£30,000" and "£31,000-£40,000" had the highest mean total score (13.78 and 13.88 respectively), there was no trend towards higher scores depending on education level, and an ANOVA test showed that the difference in mean score between the groups was not significant (p=0.48).

Patient disease duration and FROM scores

The total FROM score was correlated with the patient's disease duration to investigate whether the length of the patient's illness has an effect on the quality of life of the family member. Spearman's Rank Order Correlation coefficient showed that there was a poor, non significant correlation between the patient disease duration and the total FROM score (r= 0.143, p= 0.131). This shows that the quality of life of family members is not associated with the duration of the patient's illness (this may differ for individual, or groups of diseases), and that in this varied patient population, the impact on the family member does not increase as the disease duration progresses.

Table 6.13: The mean total FROM score for family members according to total household income per year

Family member household income per year	N.o. of family members	Mean FROM total score	S.D.	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
Less than £10,000	13.0	11.9	7.2	7.5	16.2	1.0	29.0
£11,000 to £20,000	31.0	12.4	7.6	9.6	15.2	2.0	26.0
£21,000 to £30,000	18.0	13.8	9.0	9.3	18.3	2.0	31.0
£31,000-£40,000	16.0	13.9	7.1	10.1	17.7	2.0	32.0
£41,000-£50,000	8.0	7.4	3.7	4.3	10.4	1.0	12.0
£51,000-£60,000	4.0	12.0	6.1	2.4	21.6	6.0	19.0
£61,000-£70,000	5.0	8.4	3.8	3.6	13.2	5.0	15.0
£71,000 and over	5.0	13.2	7.9	3.5	23.0	4.0	24.0
Total	100.0	12.2	7.4	10.8	13.7	1.0	32.0

The patient's disease duration was then rescored into six groups. Group 1= 1-12 months, group 2= 13-24 months, group 3= 25-36 months, group 4= 37-72 months, group 5= 73-120 months and group 6= over 121 months. The results of an ANOVA test showed that there was not a significant difference between the family member mean total FROM score across the six groups (p=0.54). Table 6.14 shows that the disease duration group with the highest mean total FROM score is group 4 (37-72 months) with a mean of 14.56.

Table 6.14: The mean total FROM score for family members according to patient disease duration

Patient disease duration (months)	N.o. of family members	Mean FROM total score	S.D.	95% Confidence Interval for Mean		Minimum	Maximum
				Lower Bound	Upper Bound		
1-12	22.0	10.5	7.2	1.5	7.3	13.7	1.0
13-24	15.0	11.3	7.9	2.1	6.9	15.7	2.0
25-36	15.0	11.1	5.8	1.5	7.9	14.3	2.0
37-72	16.0	14.6	8.5	2.1	10.1	19.1	5.0
73-120	26.0	13.5	7.4	1.5	10.5	16.5	3.0
over 121	26.0	12.5	7.8	1.5	9.4	15.7	1.0
Total	120.0	12.3	7.5	0.7	10.9	13.6	1.0

Scores of the two FROM domains

The FROM is made up of two domains (Part 1: six questions, scored 0-12 and Part 2: 10 questions, scored 0-20). The total scores for each of the two domains were found to have a positive strong correlation ($r= 0.62$ $p<0.001$) using Spearman's Rank Correlation Coefficient. This means that family members who score highly in one domain are also likely to score highly in the other, suggesting that the two domains, "Emotional" and "Personal and Social Life" are related.

Reliability

The reliability of the FROM was assessed in terms of internal consistency and test-retest reliability.

Internal consistency

The internal consistency of the FROM was measured to determine the extent to which all of the individual items measure the same attribute. Internal consistency of the FROM using Cronbach's alpha coefficient was 0.91 suggesting high internal consistency (Table 6.15). This was not improved by deleting individual items (0.90-0.91) demonstrating that all of the items contribute to the total FROM score (Table 6.16). The two domains also showed high internal consistency (Emotional= 0.80, Personal and Social Life=0.89).The scale variance also remained within a relatively narrow range (49.4-54.4) when individual items were deleted, which is consistent with a high Cronbach's alpha.

Table 6.15: Internal consistency (Cronbach's α) of the 16 items of the FROM

Cronbach's alpha	Cronbach's alpha based on standardized items	N of Items
0.911	0.910	16

Table 6.16: Item- total statistics of the FROM

FROM item	Scale mean if item deleted	Scale variance if item deleted	Corrected item-total correlation	Cronbach's alpha if item deleted
Worried	10.72	54.39	0.38	0.91
Angry	11.62	52.44	0.51	0.91
Sad	10.99	52.80	0.46	0.91
Frustrated	11.09	52.41	0.52	0.91
Talking about thoughts	11.5	50.20	0.66	0.90
Difficulty caring	11.34	50.13	0.67	0.90
Time for self	11.45	49.88	0.72	0.90
Travel	11.56	51.11	0.60	0.91
Eating habits	11.71	51.77	0.60	0.91
Family activities	11.21	50.70	0.69	0.90
Holiday	11.36	49.41	0.66	0.90
Sex life	11.55	51.31	0.53	0.91
Work or study	11.58	51.51	0.57	0.91
Family relationships	11.59	51.13	0.65	0.90
Family expenses	11.39	51.49	0.58	0.91
Sleep	11.25	49.82	0.68	0.90

Test-retest reliability

The test-retest method was used to assess whether the FROM produces more or less the same results when it is administered to stable subjects on two different occasions separated by a period of time. All 121 family members were followed up via post or email and asked to complete a hard copy or an online version of the FROM a second time after a time period of 7 to 14 days from when they were first recruited. Family members were also asked to complete a Global Health (GH) Score, which asked them to rate the patient's health on a score of 0-10, both during initial recruitment and for the 7-14 day follow up. In accordance with the study design, those family members of patients whose GH score had changed by more than one point (suggesting that their health has changed, and they are therefore not stable) were eliminated from this stage of the study. One further subject was eliminated due to incomplete questionnaire responses.

A total of 74 (61.2%) family members returned the second set of completed questionnaires 7-14 days after first recruitment. 23 of these were eliminated due to a change in GH score of more than one point. The test-retest was based on data from 52 family members (43.0%) in whom the health status of the patient had not changed (as assessed by the family member). The ICC value for the total FROM score was 0.93 which suggests that the scale is able to show reproducible results in stable subjects.

For individual items, the ICC values ranged from 0.59 to 0.88 and the ICC values for the total scores for the two domains of the FROM were 0.86 and 0.93 (Table 6.17). These values indicate a strong agreement between scores at the two time intervals. Figure 6.10 shows the mean scores for each of the items at initial recruitment and follow up, demonstrating similar mean scores on the two occasions. Although the ICC is the recommended statistical method for the test-retest investigation (Streiner and Norman 2008) a paired sample t-test was also carried out to support the results of the ICC. The paired samples correlated strongly (r=0.87 p<0.001) and there was no significant difference (p=0.51). Figure 6.11 shows the correlation between the initial and the follow up FROM scores.

Table 6.17: Intraclass correlation coefficient values for individual items, total scores for domains and total FROM score.

FROM item	ICC	95% CI		p value
		Lower	Upper	
Worry	0.65	0.88	0.96	<0.001
Angry	0.76	0.58	0.86	<0.001
Sad	0.72	0.51	0.84	<0.001
Frustrated	0.75	0.56	0.86	<0.001
Talking about thoughts	0.86	0.75	0.92	<0.001
Difficulty caring	0.76	0.58	0.86	<0.001
Time for self	0.82	0.69	0.9	<0.001
Travel	0.78	0.63	0.88	<0.001
Eating habits	0.89	0.81	0.94	<0.001
Family activities	0.82	0.68	0.9	<0.001
Holiday	0.85	0.74	0.92	<0.001
Sex life	0.84	0.71	0.91	<0.001
Work or study	0.91	0.84	0.95	<0.001
Family relationships	0.59	0.28	0.76	0.001
Family expenses	0.76	0.59	0.86	<0.001
Sleep	0.85	0.74	0.91	<0.001
Part 1	0.86	0.77	0.92	<0.001
Part 2	0.93	0.88	0.96	<0.001
Total	0.93	0.88	0.96	<0.001

Figure 6.10: The relationship between the initial FROM score and the follow up FROM score 7-14 days later for family members of stable patients

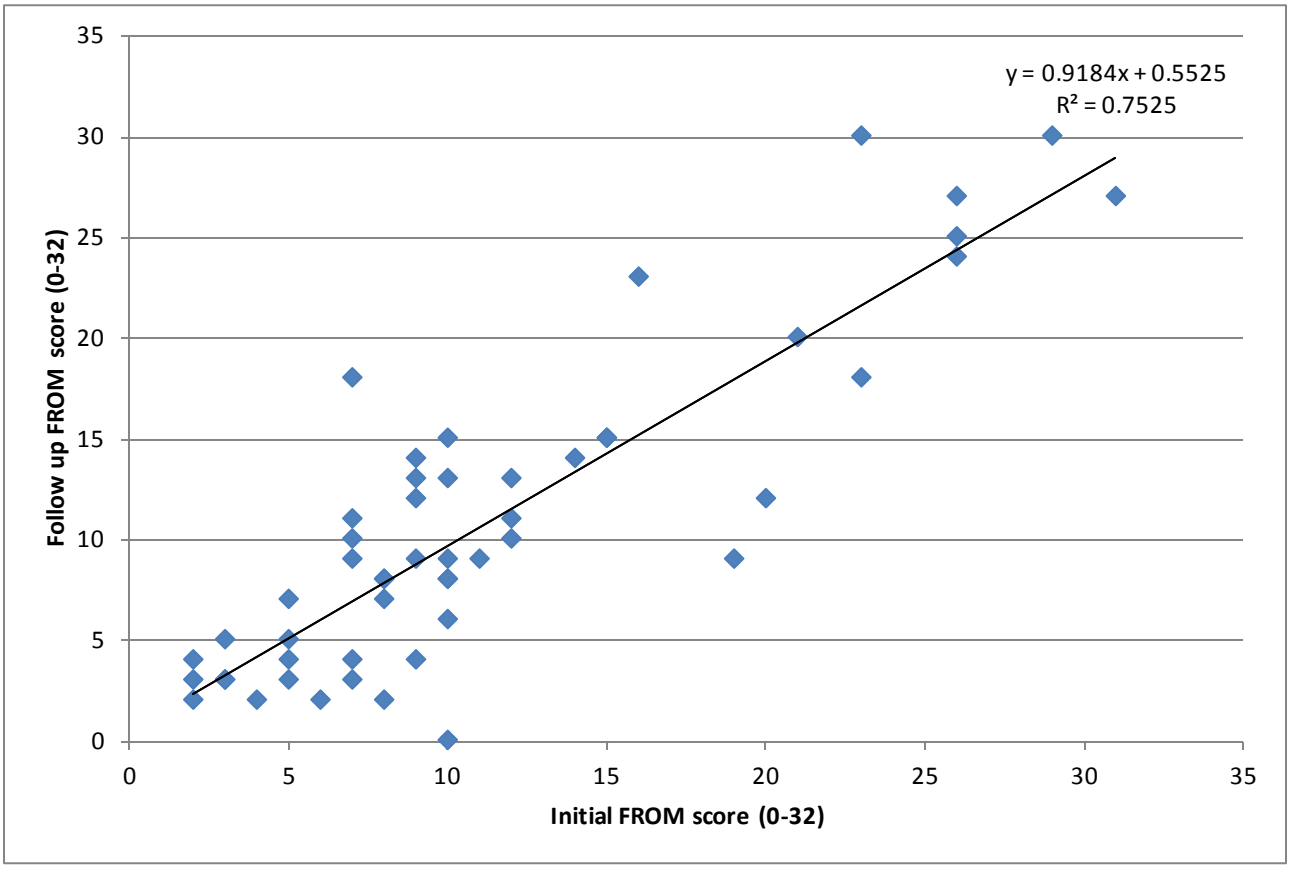
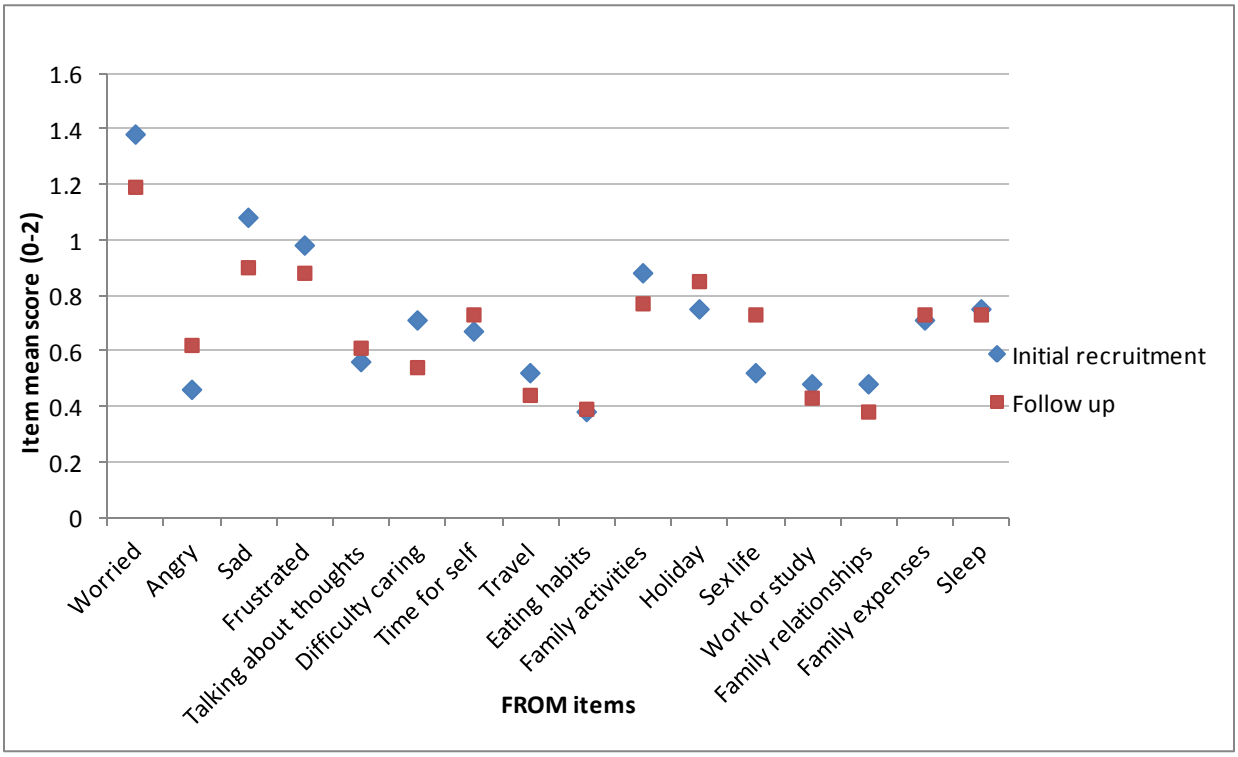


Figure 6.11: Mean scores for the individual items of the FROM at initial recruitment and follow up after 7-14 days



Face validity and practicality

Although content validity was carried out during the item reduction stage of development, a test of face validity was carried out during the validation stage, to ensure that the changes made to the FROM during item reduction did not compromise the validity of the instrument. During initial recruitment, all 120 family members were asked to complete four questions about the face validity and practicality of the FROM using "yes" or "no" tick boxes. For the final question, family members were also asked to explain their answer if they ticked "no".

Question 1: *Is the questionnaire easy to complete?*

> 120 (99.2%) family members answered "yes" and 1 family member (0.8%) answered "no".

Question 2: *Are the response options for the questions straight-forward?*

> 120 (100%) family members answered "yes".

Question 3: *Are the instructions and statements clear?*

> 118 (99.2%) family members answered "yes" and 1 family member (0.8%) answered "no". (n=119 as one missing value was eliminated)

Question 4: *Do the questions cover all the areas of your life which have been affected by your family member's illness?*

> 104 (87.4%) family members answered "yes" and 15 (12.6%) family members answered "no". (n=119 as one missing value was eliminated).

Those family members who had answered "no" for questions 4 (12.6%), were asked to comment below the questions about what they thought was missing from the questionnaire. Seven family members made comments, and their answers are shown in Table 6.18 alongside the investigators comments for each. No new areas of life were suggested which were not covered by the FROM, so therefore no changes were made as a result of the family members comments, as justified in Table 6.18.

The high levels of agreement for the four questions suggest that the FROM has high face validity, is practical and easy to use.

Table 6.18: Comments from family members regarding the face validity and practicality of the FROM and corresponding comments from the study investigator.

Family member comments	Comments/justification from investigator
More detail	The items are designed to be simple and widely applicable so adding more detail to them would potentially affect the validity
Although it mentions added expense it fails to mention the added stress of being the sole provider and caring for someone with a disability as well as raising children and maintaining the home	These aspects are covered by a number of items in the FROM (several of the "Emotional" items, family activities, work and family expenses) and would not be applicable to the majority of family members (as reflected during the qualitative interviews) so would not justify an additional item.
Because we have children living at home and they have been affected by [patient's name] illness I think they would benefit from counseling	This comment does not relate directly to the questionnaire content
My son is very forgetful which is a worry as he no longer lives around the corner from me	This aspect comes under Item 1 "I feel worried", and is an example of this theme
Other aspects affecting other things not addressed i.e. other family illness/caring responsibilities and own illness	The questionnaire is designed to assess the QoL of one family member as a result of one patient's illness and not the family as a whole. Caring is covered by item 6 "Caring for my family member is difficult"
How it affects work	This is covered in item 13 "My work or study is affected"
My wife also has health problems	The questionnaire is designed to assess the QoL of one family member as a result of one patient's illness and not the family as a whole

As a further test of practicality, the family members were timed as they completed the FROM. The mean completion time (n=108) of the FROM was 115 seconds (1 minute and 55 seconds) with a range of between 55 seconds and 272 seconds (4 minutes 32 seconds). The standard deviation was 54.1 seconds. A positive correlation between the FROM timing and the family member's age was also found using Pearson's Coefficient ($r=0.44$, $p<0.001$)

Readability and item length

The FROM was designed to be easy to read and be able to be completed by any adult family member. This was tested by looking at the length of items, and the instrument Flesch readability score. The Flesch readability score for the FROM was 64.7. Documents with a Flesch readability score of between 60 and 70 are given a "standard" reading ease (Flesch 1948) which can be understood by most people. The mean length of the 16 items was 5.6 words (range= 3-12 words), demonstrating that the items are concise and easy to read.

Sensitivity to change

Family members from five of the core specialties were included in the sensitivity to change study. The sensitivity to change study was designed to assess whether the FROM scores change with a change in the patient's health. This part of the study was designed to assess

whether the FROM scores can detect change. Participants included five family members from colorectal surgery, respiratory, renal, dermatology and rheumatology. The 25 family members were sent a copy of the FROM along with a global health score (scored 0-10) after a period of 2-3 months after initial recruitment. The family members were followed up using their initial method of choice, either by post or email. In accordance with the study design, those family members of patients whose GH score had changed by more than one point (suggesting that their health has changed) were included in this stage of the study. A second reminder was sent out to those who had not returned the questionnaires after 14 days.

A total of 14 (56%) family members returned the second set of completed questionnaires 2-3 months after recruitment. Only two out of the 14 responses showed a change of more than two points on the GH score and were therefore eligible for the analysis. The other 12 participants showed no change (4 out of 14) or a change of less than two GH score points (8 out of 14) and so were excluded from the analysis.

The number of valid responses from this test was too low to carry out any statistical analysis or draw any conclusions regarding the sensitivity to change of the FROM. This is discussed further in the Discussion chapter of this thesis. However, as the majority of participants showed a stable health state after 2-3 months (a change of less than 2 points on the GH score), an additional test-retest reliability study was carried out on this data.

Additional test-retest study

The additional test-retest was based on data from 12 family members in whom the health status of the patient had not changed after 2-3 months (as assessed by the family member). The ICC value for the total FROM score was 0.94 which suggests that the scale is able to show reproducible results in stable subjects, even after 2-3 months with no change in health state.

Construct validity

The construct validity of the FROM was assessed by testing two *a priori* hypotheses about the construct of the scale.

Hypothesis 1: The impact of illness on family member's QoL is correlated to the family member's overall QoL.

To test the first hypothesis, the scores of the FROM were compared with the scores of a generic quality of life measure, the WHOQOL-BREF, which was also administered to family members during initial recruitment. The attributes of the two measures are compared in Table 6.19.

Table 6.19: The attributes of the FROM and the WHOQOL-BREF

	The FROM	The WHOQOL-BREF (Skevington et al. 2004; World Health Organisation 1996)
Concept measured	The impact of a patient's illness on their family member	A subject's subjective quality of life
Target population	Family members (age ≥ 16) of patients (of any age)	Adults
Number of items	16	26
Possible total score range	0-32	1-130[a]
Number of domains	2 ("Emotional" and "Personal and Social Life")	4 ("Physical health", "Psychological", "Social relationships" and "Environment")
Scoring format	Scores are added to form total score[b]	Scores are added to form total score
Scoring direction	Higher score= lower QoL	Higher score= higher QoL

[a] The WHOQOL-BREF has several different scoring systems allowing the raw score to be converted to a transferred score and compared to the WHOQOL-100. For the purposes of this test, the raw score (1-130) is used.

[b] The two domain scores for the FROM are usually reported separately (as they form two factors), however for simplicity in this study (as there are no direct clinical implications), the scores of the two domains are added to form a total score.

Data from family members were analysed for correlation. One subject was removed due to incomplete WHOQOL-BREF score (n=119). Table 6.20 shows the descriptive statistics for each of the questionnaires.

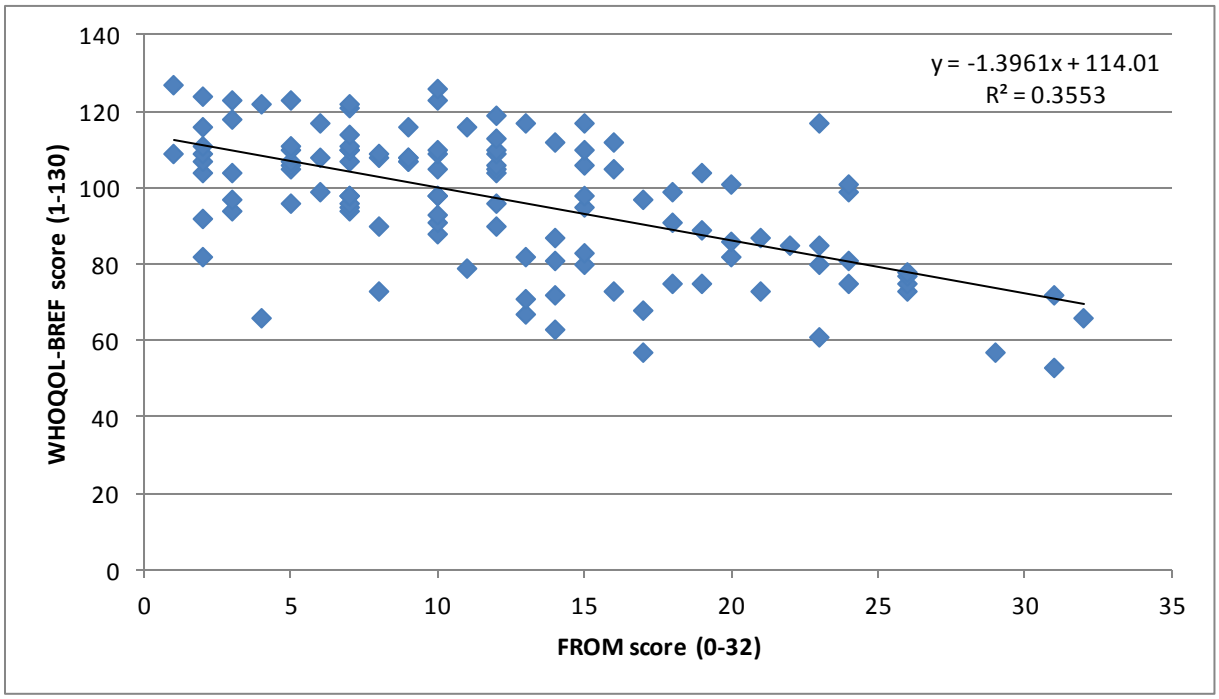
Table 6.20: Descriptive statistics of the FROM and the WHOQOL-BREF

	FROM score	WHOQOL-BREF score
N	119	119
Mean	12.28	96.87
Std. error of mean	0.69	1.61
95% confidence interval for mean	Lower bound = 10.92 Upper bound =13.64	Lower bound= 93.68 Upper bound= 100.06
Standard deviation	7.50	17.57
Minimum	1	53
Maximum	32	127
Range	31	74
Median	11.0	99.0
Interquartile range	10	28
Skewness	0.62	-0.48
Std. error of skewness	0.22	0.22

Using Spearman's rank correlation coefficient, a correlation was found between the FROM scores and the WHOQOL-BREF scores ($r= -0.55$, $p<0.001$). The correlation is negative due to the different scoring directions of the two questionnaires. This shows that a family member with a high FROM score is likely to have a low WHOQOL-BREF score, indicating that the impact of illness on family members of patients is correlated to their overall QoL. This

correlation is shown in Figure 6.12, showing a large range (1-32) of FROM scores, but that most of the WHOQOL-BREF scores were in the top half of the scale (above 65). This data confirms the first construct validity hypothesis.

Figure 6.12: The relationship between the FROM and the WHOQOL-BREF scores



Correlations were also assessed for the two domains of the FROM and the four domains of the WHOQOL (Table 6.21). The strongest correlation (r= -0.57) was found between the FROM domain "Personal and Social Life" and the WHOQOL-BREF domain "Social relationships".

Table 6.21: The correlations between the domains of the FROM and the WHOQOL-BREF

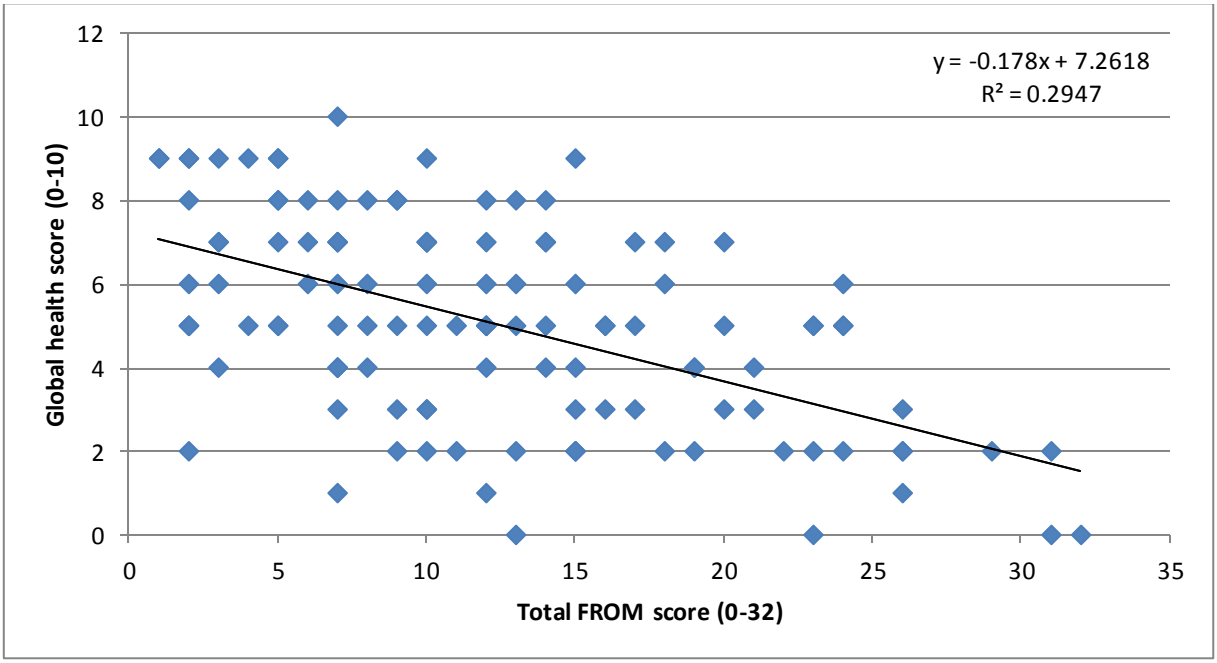
FROM / WHOQOL-BREF	Domain 1 "Emotional"	Domain 2 "Personal and Social Life"
Domain 1 "Physical health"	-0.36	-0.44
Domain 2 "Psychological"	-0.43	-0.50
Domain 3 "Social relationships"	-0.44	-0.57
Domain 4 "Environment"	-0.38	-0.48

Hypothesis 2: The impact of illness on family member's QoL is correlated to the health of the patient

The second hypothesis was tested by comparing the FROM score with the global health (GH) score. At the time of initial recruitment, family members were asked to rate the patient's

health on a scale of 0-10. The GH score representing the family member's assessment of the patient's health ranged from 0-10 with a mean of 5.1 (SD=2.5). The GH score was found to correlate negatively with the total FROM score using Spearman's rank correlation coefficient (r= -0.51, p<0.001). The two domains also showed a moderate correlation with the GHS (Emotional r= -0.48, p<0.001;Personal and Social Life r=-0.45, p<0.001). This shows that the lower a patient's health (as perceived by the family member) the greater the impact on the family member's quality of life (Figure 6.13). The large degree of scatter on either side of the trendline shows the variability of correlation at the individual level.

Figure 6.13: The relationship between the total FROM score and the Global health score (family members' assessment of patients' health)



The scores of each of the 16 FROM items were also compared to the GH score (Table 6.22). The correlation was significant for all of the items (r= -0.21-0.46). The items showing the strongest correlation with the GH score were those concerning holiday (r=-0.46), difficulty caring (r=-0.45), and time for self (r=-0.44). This suggests that as the patient's health deteriorates, the family member experiences more problems with going on holiday, finds caring for the patient more difficult and has less time for themselves. The two items showing the weakest correlation were those concerning sex life (r=-0.21), and family expenses (r=-0.26), suggesting that these two aspects are less affected by the health of the patient.

Table 6.22: Correlations between the individual items of the FROM and the global health score

FROM item	r	p
Worried	-0.35	<0.001
Angry	-0.28	0.002
Sad	-0.30	0.001
Frustrated	-0.30	0.001
Talking about thoughts	-0.28	0.002
Difficulty caring	-0.45	<0.001
Time for self	-0.44	<0.001
Travel	-0.37	<0.001
Eating habits	-0.30	0.001
Family activities	-0.42	<0.001
Holiday	-0.46	<0.001
Sex Life	-0.21	0.02
Work or study	-0.39	<0.001
Family relationships	-0.28	0.02
Family expenses	-0.26	0.04
Sleep	-0.29	0.02

Multiple regression analysis

A multiple regression analysis was carried out to explore some of the factors influencing the family members' QoL. Health of the patient (assessed by the family member) has already been identified as one of these influencing factors during the testing of the second hypothesis, therefore a hierarchical regression analysis was chosen. The total FROM score was set as the dependent variable, and independent variables were health of the patient (GH score), family member age, family member gender, patient age, patient gender, relationship between family member and patient, patient's disease duration, family member's education level and family member's total annual household income. These nine variables were entered into the analysis in the order given, and the summary results are shown in Table 6.23(a,b). Table 6.23(a) shows that the correlation between the set of independent variables and the dependent variable (FROM score) was moderate (R=0.54 for model 9). The results of the regression analysis showed that the GH score was the main predictor, explaining 25% of the variance. It was the only variable to cause a significant change in the R^2 value (change= 0.25, p<0.001). The second highest change in R^2 can be seen when patient age is added (change=0.03, p=0.06). Family member age, family member gender, patient age, patient gender, relationship between family member and patient, patient's disease duration, family member's education level and family member's total annual household income only added 4% to the variance. From Table 6.23(b), it can be seen that the GH score was the variable with the most significant association with the total FROM score (β=-1.65, p<0.001) and therefore the main predictor. This reflects the results found in testing the second

hypothesis. The only other significant predictor in this model was patient's age, although the β value was small (β=-0.07, p=0.04).

Table 6.23(a and b): Results of the hierarchical multiple regression analysis for the total FROM scores

(a)

Model	R	R^2	Adjusted R^2	Std. error of the estimate	Change Statistics				
					R^2 change	F change	df1	df2	Sig. F change
1 (a)	.502	.252	.245	6.446	.252	35.650	1	106	.000
2 (b)	.502	.252	.237	6.476	.000	.008	1	105	.931
3 (c)	.505	.255	.234	6.492	.004	.492	1	104	.485
4 (d)	.530	.281	.253	6.411	.026	3.658	1	103	.059
5 (e)	.535	.287	.252	6.415	.006	.846	1	102	.360
6 (f)	.540	.292	.250	6.423	.005	.768	1	101	.383
7 (g)	.540	.292	.243	6.455	.000	.001	1	100	.971
8 (h)	.541	.293	.236	6.482	.001	.153	1	99	.697
9 (i)	.542	.294	.229	6.511	.001	.113	1	98	.737

a. Predictors: (Constant), Global Health Score
b. Predictors: (Constant), Global Health Score, Family Member Age
c. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender
d. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender, Patient Age
e. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender, Patient Age, Patient Gender
f. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender, Patient Age, Patient Gender, Relationship
g. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender, Patient Age, Patient Gender, Relationship, Disease Duration
h. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender, Patient Age, Patient Gender, Relationship, Disease Duration, Family Member education level
i. Predictors: (Constant), Global Health Score, Family Member Age, Family Member Gender, Patient Age, Patient Gender, Relationship, Disease Duration, Family Member education level, Family Member household income

(b)

Variables (predictors)	Unstandardised coefficients		Standardised coefficients		
	B	Std. Error	Beta	t	Sig.
(Constant)	24.062	5.573		4.318	.000
Global health score	-1.650	.271	-.541	-6.101	.000
Family member age	.052	.052	.105	.995	.322
Family member gender	.459	1.572	.029	.292	.771
Patient age	-.070	.033	-.228	-2.088	.039
Patient gender	-1.133	1.443	-.077	-.785	.434
Relationship	-.389	.446	-.085	-.873	.385
Disease duration	.000	.007	.000	-.001	.999
Family member education level	-.279	.586	-.044	-.476	.635
Family member household income	.100	.297	.031	.336	.737

DISCUSSION

This study indicates that the FROM-16 is reliable and valid in family members of patients from a variety of specialties and diseases. Although the numbers of family members recruited from each specialty was small, the mean total FROM scores were still calculated for each specialty. This gives a preliminary idea of in which specialties family members are most affected, though disease specific data will be needed before any clear conclusions can be drawn.

The reliability of the FROM was demonstrated by a high Cronbach's alpha (0.91) and high ICC (0.93) for test-retest reliability. The validity of the FROM was successfully proven using two a priori hypothesis. The use of the global health score proved successful in the FROM validation and previous studies have found that family members are able to accurately assess patient disease severity (Housman et al. 2002) further increasing the reliability of this result. The high correlation seen between the FROM and the WHOQOL-BREF suggests that the impact on a family member's quality of life as a result of the patient's illness contributes greatly to their general QoL, even with other potential external influences.

SUMMARY

- The study population for the validation of the FROM was made up of 120 family members of patients across 25 medical and surgical specialties.

- The family member was asked to complete the FROM, which was timed. They were then asked to rate the patient's health on a scale of 0-10 (global health score) and complete written feedback about the FROM. They were also asked to complete the WHOQOL-BREF questionnaire.

- After 1-2 weeks, all family members were asked to complete the FROM and the global health score again.

- After 2 months, 25 of the family members were contacted again and asked to complete the FROM and the global health score again.

- Total scores for the FROM (0-32) ranged from 1 to 32, median = 11.50, mean = 12.28 and SD = 7.47. There was no floor effect (scoring 0), and only one subject scored 32 showing a minimal ceiling effect.

- Although the FROM mean total score varied slightly between males (11.83) and females (12.52), there was no significant difference between their scores (p=0.63).

- Spearman's Rank Order Correlation coefficient showed that there was no significant correlation between the family members' age and the total FROM score (r= 0.02, p= 0.80).

- Spearman's Rank Order Correlation coefficient showed that there was a poor, non significant correlation between the patient disease duration and the total FROM score (r= 0.143, p= 0.131).

- Internal consistency of the FROM using Cronbach's alpha coefficient was 0.91 suggesting high internal consistency.

- The test-retest ICC value for the total FROM score was 0.93 which suggests that the scale is able to show reproducible results in stable subjects.

- The mean completion time (n=108) of the FROM was 115 seconds (1 minute and 55 seconds) with a range of between 55 seconds and 272 seconds (4 minutes 32 seconds).

- The number of valid responses for the sensitivity to change was too low to carry out any statistical analysis or draw any conclusions regarding the sensitivity to change of the FROM.

- Using Spearman's rank correlation coefficient, a correlation was found between the FROM scores and the WHOQOL-BREF scores ($r= -0.55$, $p<0.001$).

- The GH score was found to correlate negatively with the total FROM score using Spearman's rank correlation coefficient ($r= -0.51$, $p<0.001$).

- The results of the regression analysis showed that the GH score was the main predictor, explaining 25% of the variance.

CHAPTER 7

General Discussion

Family quality of life and the impact of illness on family members of patients are subjects which have been explored and studied in several areas of medicine, but little information is available in the literature about the impact of illness on families across medicine as a whole. One of the key objectives of this study was to explore the existing research available in the area of family quality of life, and identify gaps which need addressing. Therefore, the structured literature review, which was first carried out at the beginning of the study and then updated over the following two years formed an important part of this study (Golics et al. 2013). Health, psychological and social databases were used to identify papers to review during the literature search, and the study team, including the investigator and supervisors identified the key search terms used during the search. Ideally, a full systematic review of the literature would have been carried out, but given the time constraints of the study this was not possible as effective systematic reviews rely on adequate time and resources (Jones 2004). In the case of this study, the structured review identified a gap in the literature, but published procedures required for systematic reviews were not followed. Instead, every effort was made by the investigator to make the review both reproducible and accountable. This included reporting the number of articles identified, screened and reviewed from each of the searches and reporting the date of each search and the key terms used. This information was then used to update the review on a six-monthly basis.

The literature review highlighted the lack of information about the impact of illness on families across the whole of medicine. Although the numbers of articles found were reasonably high for most of the search terms used, after review the majority of articles were found to be unrelated to family quality of life. Many disease-specific studies looking at family impact were identified from the review, and several disease-specific measures were also found. Very little information was found about the generic impact of illness on family members, and no instrument currently exists to measure this impact. Instruments containing the term “family” were identified and at first glance these appeared to measure the generic impact on family members, but although they had been given very generic-sounding names, all of these measures were designed for specific populations, usually family members of paediatric patients. The results of the literature review confirmed that the impact of illness on family members is often underestimated and has not been fully explored in most medical specialties. In those areas where the impact had been explored, it was found to be severe and widespread. It was therefore felt that a generic family quality of life study would produce interesting and valuable information to compare to, and expand the current available literature. There was not enough information found in the current literature to suggest whether the areas of impact on family members of patients are the same across all areas of medicine. It is important to note that during the two years after the literature review was first carried out, the only new information identified was disease-specific and that no generic,

multi-speciality study was found. The information found in the literature review was compared to the qualitative results of this study, as explained later in this chapter.

Qualitative work and study design

The semi-structured interviews with family members of patients resulted in detailed and extensive information about the family impact of illness. Although many of the family members interviewed became emotional during the interviews and found it difficult to talk about their experiences, the rapport built between the family member and the interviewer helped family members to open up and share information that many claimed not have shared with anyone else before, and was therefore uncovered for the first time in this study. The willingness of the family members to open up and share information when questioned on their own in a private room also suggests that the FROM-16 measure may be best completed individually by family members, without patients or professionals directly observing or being able to influence answers. The use of a semi-structured interview guide helped to direct family members towards relevant areas and topics without introducing bias or asking leading questions. The questioning style used during the interviews was to begin with open questions and follow them up with closed questions to encourage the participants to provide examples. The use of the general open question at the start and end of each interview proved successful, as many family members were prompted to talk about subjects they had forgotten or had been overlooked by the interviewer. In hindsight, the choice of one-to-one interviews over focus groups was considered to be the correct choice, as the detail of information provided by the family members was more sensitive than originally predicted and family members may not have felt comfortable enough to discuss these impacts in front of others, especially when they are not used to discussing them themselves.

One of the strengths of the qualitative work in this study was the large number of interviews carried out with family members of patients suffering from a variety of illnesses. The inclusion of family members of patients with co-morbidities provided more realistic and diverse information. On reflection, excluding family members of patients with more than one illness would have produced a measure with a much narrower use potential, as patients commonly suffer from more than one medical condition (Gabriel and Michaud 2009; Guh et al. 2009). Since it was important to gather information from a wide range of diseases during the qualitative phase, the interviewer directed the family member to talk about the primary condition, as identified by the clinic they were attending. This also proved successful and added to the richness of the data.

The determination of saturation of information from the interviews in this study was different from other qualitative studies. Usually, interviews are completed in batches, analysed and

assessed for saturation (the point after which no new information arises). The saturation point therefore determines the total interview number required (Guest et al. 2006). However, in this study it was important to interview a cross-section of family members and interview equal numbers from each speciality. Since there were 26 specialties involved in the study, due to the practical time constraints it was impossible to interview one from each speciality in turn, then a second, third etc. Therefore, it was decided that a minimum of five family members from each specialty would be interviewed regardless of whether saturation was reached. This ensured a cross-section of family members and a variety of medical conditions from each specialty. Therefore, although the continuation of interviews from number 40 (saturation point) to number 133 seems excessive, it ensured that the views of family members from all 26 specialties were considered.

The use of a pilot study of five family members helped the interviewer to practice using the interview guide and questioning style. Since informed consent was taken from the patient and family member, there was a significant amount of paperwork to be completed before the interview, so the pilot study helped to ensure that this procedure went smoothly during the study interviews. The qualitative themes resulting from the study were similar to those identified across previous studies during the literature review. However, the extent of the impact and the emotional examples given by family members were beyond what was initially anticipated by the study team. These themes are discussed below. It is often not considered appropriate to report the percentages of participants mentioning each theme in qualitative research (Kitzinger 1995). This is especially true where a non-structured interview technique is used, and participants are asked different questions in each interview. However, it is felt that in this study, reporting the percentages of family members affected by each theme adds to the depth and interest of the data, especially as this is a new population which has not been investigated in this way before, and as there were a large number of interviews. As the interview guide used was semi-structured, and reviewed after the pilot interviews, all family members were given the chance to report on the key themes. It is important to note that the percentages attached to the qualitative report were not used to place a greater emphasis on certain themes in the final questionnaire, and all were considered in this context equally. The percentages reported came in useful when deciding which items to include in the FROM (a 5% cut off point was used), but all items mentioned by over 5% of the family members were considered as equally important for inclusion. The 5% cut off level allowed the large volumes of data from 133 interviews to be managed and controlled so that the most relevant and common themes were included in the instrument.

Family quality of life themes

The emotional impact of illness on the family was one of the main findings from the qualitative part of this study. Many other disease specific studies also found a large emotional impact, including family members of patients with multiple sclerosis (Aymerich et al. 2009; Bowen et al. 2011), where high levels of anxiety and frustration were reported. Emotional impact was the most highly reported theme in both this study (92% of family members affected), and in a study with family members of dermatology patients (98%), where similar emotions including worry, frustration and stress were reported (Basra and Finlay 2007). In this study, 20% of family members felt they had no one to talk to about the way they were feeling, which was also noted by Davis et al (Davis et al. 2009) who found that family members were unable to share their true feelings with others. The great emotional impact felt by family members in this study is consistent with that of a study of spouses of patients with prostate cancer (Kornblith et al. 1994), who were found to experience a greater emotional impact than patients themselves.

Problems with daily activities were reported by family members of patients in this study. Other studies have also identified this aspect, including going shopping and hobbies such as walking (Eghlileb et al. 2007). Daily activities of family members were shown by Goldbeck (2006) to be affected immediately after the diagnosis, as well as in chronic conditions, reflecting the results of this study where patient disease durations ranged between one month and 60 years. Siblings of patients with a disability have also reported a decrease in family activities (Opperman and Alant 2003), as seen in the family members in this study, often preferring to spend time on their own. Changing their daily activities and routine as a result of the patient's condition meant that some family members had to completely alter the way they lived their lives, and this also impacted on others around them such as other members of the family, friends or work colleagues. Often, family members reported spending less time on things they enjoy (e.g. hobbies and family activities) and more time on activities relating to the patient (e.g. caring and attending medical appointments). A large number of family members reported spending more time caring for the patient, for example helping with washing, bathing and dressing. Most of these family members were not registered carers, but many were caring for the patients on a full time basis. These family members represent hidden carers; those who are caring for patients but not receiving any financial support, and often do not realise they may be classed as "carers" (Olsen et al. 2005). These results support the fact that this study is looking at "family members", as opposed to "family caregivers" or "carers". Most of the family members experiencing an impact on their lives would not have been eligible for the study if they were required to be carers, and many of the important issues surrounding the lives of the family members would have been overlooked. When designing studies involving family members it is important to consider whether it is the

impact on the family or family member, or the impact on the caregiver which is to be measured, and how the “carer” status will be assessed.

Several of the partners and spouses of patients interviewed in this study reported marital and relationship problems as a result of the patient’s illness. Parents of unwell children have also been shown to report low marital satisfaction and depressive symptoms directly related to the severity of the child’s condition (Berge et al. 2006; Golics et al. 2013). Many family members found it difficult to accept their changing role in the relationship with the patient; children having to look after their parents, and spouses having to act as carers. This was also reflected in a study by Boeije and Van Doorne-Huiskes (2003) who explored the difficulties in the changing relationships between spouses and patients with multiple sclerosis, and in a study of family members of advanced heart failure patients (Aldred et al. 2005). The negative impact of patient’s illness on family member’s sex life was an important theme in this study, and one which was also identified in a study of family members of patients with overactive bladder (Coyne et al. 2010) and prostate cancer (Kornblith et al. 1994). A lack of family cohesion was also found to be a predictor of depression in families of patients with HIV (Demi et al. 1997), reflecting the relationship identified in this study between the themes family relationships and emotional impact.

Another finding of this study was the impact of illness on the sleep and health of the family member, particularly the effects of stress and worry at night. The physical effects of having to get up in the night to help the patient were also reported. Sleep was also shown to be a major problem in family members of patients with bladder disorders (Coyne et al. 2010) and parents of children with cerebral palsy (Davis et al. 2009). The extent of the impact of illness on the family members’ health was evident in this study, with family members reporting diagnoses of depression and existing illnesses worsening. This study identifies family members as a hidden “patient” group, with an apparent “ripple effect” of illness; one patient being unwell has the potential to create several more “patients” in the family (Lieberman and Fisher 1995). This can then magnify problems with finances and family relationships, in a vicious cycle. This hidden burden has a potentially large financial impact on the health care system that could potentially be reduced with appropriate family support. This also suggests that clinicians should consider not just the health of the patient during consultations, but also the potential health problems of the family members as a result. This would be particularly applicable in primary care settings where general practitioners often provide care to more than one member of a family. Additionally, clinicians should consider the health status of relatives and the support available to the family as possible causes or trigger factors when making a diagnosis of depression.

Many of the problems faced by family members of patients in this study were similar to those reported by family members of patients with psoriasis (Eghlileb et al. 2007), including stopping going on holiday abroad, restricted activity on holiday, or feeling worried or uncomfortable whilst away on holiday. The frequency of dialysis appointments for patients with chronic renal failure also causes problems for families trying to organise holidays (Reynolds et al. 1988). Typically, people use holidays as a method of relaxation, and from this study it was evident that many families of patients were either unable to go on holiday or experienced problems when they did go away. This meant that they were not getting the relaxation or escapism they needed, which could potentially increase the levels of stress they feel on a daily basis. Not only were their levels of stress increasing and their lives being affected on a daily basis, they were not able to experience the break from every day routine which most people take for granted. Many of the family members also found that the services available to them when they were on holiday, for example mobility services, were inadequate and difficult to acquire. These are services which need reviewing or improving if family members are to have any chance of enjoying holidays.

Many of the family members in this study felt uncomfortable asking for the support they required, something which is reflected in a study by Brown et al. (2003) who found that many families of paediatric patients with intellectual disability were dissatisfied with the amount of support they received from others. This study also found that families did not have time to seek out social support services, linking to the “Time planning” theme found with the family members interviewed. Caregivers of cancer patients also reported problems with medical care, similar to those found in this study including difficulty getting help from professional organisations and problems co-ordinating different healthcare professionals (Osse et al. 2006). “Disability-related support” was identified as an important factor in the family quality of life of children with disabilities (Summers et al. 2005). With such difficulty in accessing funding and support available to them, many family members claim carer benefits, especially if they are unable to work. If appropriate social support was provided, many of the family members may have been able to continue working, reducing the burden on government financial resources.

This study found that many family members had problems with their work or study as a result of the patient’s condition, with a minority (9%) having to give up work altogether. This was similar to findings in other studies. For example, caregivers (5%) of breast cancer patients had to give up work completely (Grunfeld et al. 2004) and many also used up their holidays to care for the patient. For family members of patients with multiple sclerosis, it was not just the strain of loss of work they felt, but also the problems with adjusting to the loss of income as a result, and even just the threat of lost income caused financial stress (De Judicibus and McCabe 2007).

Family members in this study described the financial impact of having to travel to hospital appointments, or to visit the patient in hospital. This particular financial aspect was also identified in families of children with thalassaemia major (Clarke et al. 2009) and children with chronic illnesses in general (Gannoni and Shute 2010). Many of the other financial issues mentioned specifically by family members across all specialties in this study were also identified in family members of children with chronic diseases (Gannoni and Shute 2010), including the cost of food items and employment.

The impact of illness on the social life of family members was an issue for family members in this study, particularly when they felt restricted to looking after the patient. Social well-being was identified as an important issue for family quality of life in family members of children with disabilities (Poston et al. 2003), who reported impact on their own friendships. As a result of a restricted social life, and having to cancel planned activities, family members of patients with advanced heart failure feel isolated (Aldred et al. 2005) and stop seeing their friends and family. The Impact on Family Scale (Williams et al. 2006), to assess the impact of paediatric chronic illness on the family, also contains several items relating to social impact, including seeing friends and family less, changing plans at the last minute and having little desire to socialise. These are also all examples given by family members in this study.

Time planning was a significant problem for family members in this study as a result of having an unwell relative. This was also reflected in a study by Basra and Finlay (Basra and Finlay 2007) who found that family members of dermatology patients also had trouble planning their time around caring for the patient. In turn, families of patients with chronic pain have been shown to cope better with the situation if family rituals and routines are unchanged (Bush and Pargament 1997) something which family members in this study found very difficult to do.

Although several positive themes were also found during the interviews (for example, the illness bringing the family closer together), these were not included as items in the FROM-16, as the intention of the instrument was to measure the negative impact. Equally, themes relating to areas of life which cannot be changed were not selected as individual items, as the measure has been designed to be used to assess change in family quality of life as a result of specific interventions. For example, during the development process, the item “I have had to give up work/retire early” was incorporated into a general item about work, as retiring is something which, although not impossible, could often not be easily changed as a result of an intervention.

Quantitative work and study design

The quantitative stage of the study was carried out in 25 of the 26 specialities originally involved in the study during the qualitative phase. The speciality which was excluded from the quantitative analysis was mental health, as the outpatient community clinics proved very difficult to recruit from in the time available. The community-based clinics only saw a handful of patients a day and often family members did not attend with patients or did not want to take part in research studies. A second consultant clinician was drafted in to help with recruitment efforts but recruitment was unsuccessful. Although this means that the FROM-16 did not involve family members of patients from mental health in its initial validation, several of the patients with other conditions had mental health co-morbidities including depression and anxiety. Further speciality-specific validation of FROM will be required in the future.

The development of the final version of the FROM-16 was carried out using a combination of content validity panel feedback, Rasch and factor analysis. This three-way approach combined the traditional classical test theory approach, which has been heavily criticised (Prieto et al. 2003), with the modern approach of Rasch analysis which is becoming more commonly used for item reduction, and the qualitative voice of the family members and experts on the content validity panel. Although the use of Rasch analysis for item reduction is not routinely used alone, the use of a pure statistical method to remove items from a measure was recently criticised at the 2012 International Society for Quality of Life Research conference during the Industry Advisory Committee symposium (2012), where it was felt that the "patient voice" would be lost. This had already been taken into consideration during the design of this study, where a combination of methods was used for item reduction. As expected when three methods were used, there was some tension between the results produced by each method. The first tension arose between members of both content validity panels (expert and family members), where different members of the two panels had opposing views about some items. This highlighted the importance of the focus group discussion for the cognitive debriefing of the FROM, which allowed these issues to be debated between the members of each panel until a consensus was reached. Generally, when the family member panel disagreed with the expert panel, the view of the family member panel was taken, as the instrument was designed for their use. The qualitative and quantitative feedback was drawn from a large number of judges from mixed backgrounds, increasing confidence that changes made to the items were representative of the general population of family members, and were clinically relevant across a range of specialties. The second tension arose between the results of the Rasch analysis and the content validity panel results. Great care was taken during the Rasch analysis to refer back to the concept elicitation interviews and to ensure that the views of the family members were represented in the measure. This included taking into account the endorsement rate of each item and

considering the reasons why the item could be performing badly according to the Rasch model before removal or re-wording.

The most challenging decision made as a result of the Rasch analysis was the collapsing of the five response categories down to three and deciding upon uniform scaling across all items. This goes against the most common practice of collapsing response categories in different ways for different items (Lamoureux et al. 2008). However, the aim of the FROM was to be an easy to use and easy to score instrument, and it was strongly felt that having a complex non-uniform scoring system would jeopardise the usability of the FROM. It was also thought that non-uniform scoring would put an unwanted emphasis on certain items. This decision was made with the views of the family members in mind and was an example of where the statistical methods and reality of the proposed use did not mirror each other. In the process of factor analysis, another example of tension between the methods was seen, where wording of the qualitative interviews was consulted when deciding upon on which factor to place a highly cross-loading item. It was therefore felt that the item reduction of the FROM was executed in the most robust way possible, considering both the statistical and scientific methods alongside the human voice of the content validity panels. This careful balance and step by step approach to item reduction was considered one of the strong points of the FROM-16 development and was reflected by the successful validation of the measure.

The naming of the measure and the two domains was an important part in the development of the FROM-16. The name “Family Reported Outcome Measure” and the acronym “FROM” are instantly recognisable and self-explanatory. They also differ from the existing family measure names, and fit well alongside the area of patient reported outcomes (PROMs), suggesting that the measure is defining a new concept, which is different from PROMs, but is of similar importance and significance. The memorable and innovative name “FROM” will aid in its promotion and publication. The two FROM domains were named “emotional” and “personal and social life”. These names were chosen to reflect the content of the items contained within them, but following discussions regarding naming, an interesting observation was made. When describing patients’ health-related quality of life, three domains are often considered: physical, psychological and social (Phillips 2008). The FROM-16 contains the psychological and social domains, but lacks a physical domain. This observation makes sense when assessing family quality of life, as the FROM-16 is not designed to measure the physical problems of the family members themselves; the physical domain would be relevant to the unwell patient, but less relevant (or irrelevant) to the healthy family member. The FROM-16 should be scored as two individual domain scores, as the results of the Rasch and factor analysis proved that a total score was inappropriate and the measure was made up of two different constructs. As with this study, in future work the total score of the FROM-16 could be used for comparison with other measures, but the individual domains

should also be assessed separately. For clinical purposes, and when assessing family members on an individual basis, the two domains should be scored separately.

Although the numbers of family members recruited from each specialty was small, the mean total FROM scores were still calculated for each specialty. This gives a preliminary idea of in which specialties family members are most affected, though disease-specific data will be needed before any clear conclusions can be drawn. The disease areas where family members scored most highly on the FROM were oncology, neurology, haematology and chronic pain, where the diseases tend to be very disabling, with few cures and long-term treatments, often with severe side-effects. On the other hand, ophthalmology, where conditions tend to have a gradual onset, and orthopaedics, where many illnesses are cured operatively produced the lowest FROM scores.

The reliability of the FROM was demonstrated by a high Cronbach's alpha (0.91) and high ICC (0.93) for test-retest reliability. The minimum acceptable value for α varies between authors (Clark and Watson 1995; Heppner et al. 1992), and it has been suggested that for a measure to be used clinically, the α should be above 0.9 (Ponterotto and Ruckdeschel 2007), a criterion met by the FROM. As the items came directly from the family members interviewed and used language taken from the interview transcripts, it was expected that the reliability would be high, as the family members would be able to relate to the items and have no trouble understanding them or relating them to their own experiences. The choice of time interval for follow up in a test-retest analysis was an important consideration (Pallant and Tennant 2007). A retest interval of between two and 14 days is usually considered acceptable (Streiner and Norman 2008), so an interval of 7-14 days was considered appropriate for this study. The use of postal and email follow-up, freepost return envelopes and a second round of follow up for non-responders proved successful, and produced a moderately high response rate.

Positive results were seen from both construct validity tests. Ideally, an existing measure measuring the same, or similar concept would have been used to assess the construct validity. However, as already identified, no such measure exists. One option would have been to make alterations to an existing measure in order to use it as a comparator for content validity, but as it was unclear whether the results of this study would be similar to previous studies of family members of paediatric patients, or carers, this approach was considered inappropriate. Instead, two *a priori* hypotheses were tested in relation to the concept of the FROM-16. For the first hypothesis, the scores of the FROM were compared with the scores of a generic quality of life measure, the WHOQOL-BREF. The WHOQOL-BREF was chosen as it is widely used and has strong psychometric properties (Skevington et al. 2004), so it would be robust enough to rely on to measure the intended concept. The

correlation between the two measure scores was -0.55 ($p<0.001$). This correlation is within the range expected by the study team, as the relationship between family and patient quality of life has been already demonstrated with similar correlations in disease-specific studies (Basra et al. 2007; Kornblith et al. 1994; Weitzenkamp et al. 1997). It was also unknown how strongly the concepts of family quality of life and patient quality of life would be correlated in most of the disease areas before the study, so the study team did not expect a very high correlation. Considering this proven link between patient quality of life and family member quality of life, it would be interesting to see whether providing support for the family member and improving their quality of life would improve the quality of life, and even the health of the patient (Rees et al. 2001).

During the construct validity testing, many of the family members commented about how much easier the FROM-16 was to complete, in comparison with the WHOQOL-BREF. Family members struggled with the wording and meaning of some of the items of the WHOQOL-BREF, for example questioning what was meant by the item “How healthy is my physical environment?”. This often meant that the family member spent longer than anticipated with the investigator during this stage of the study, and often assistance was required from the investigator to complete the WHOQOL-BREF. Although the completion of the instrument was not timed in this study, it has been suggested that the WHOQOL-BREF can be completed by healthy individuals in under five minutes (Skevington et al. 2004). However, this was not consistent with the family members in this study, who often took longer than five minutes, and in some cases up to 15 minutes to complete the WHOQOL-BREF. Whilst not officially timed, an estimate of time taken to complete the measure was made by the investigator, in relation to the total time the family member spent with the investigator. It is possible that the family member population in this study and in the WHO study (Skevington et al. 2004) varied in terms of demographics, explaining the difference in completion time. However, using a different, more simple and easy to use generic measure may have increased the speed of data collection and put less burden on the family members completing the measures.

The second hypothesis assessing whether the impact of illness on family member’s QoL is correlated to the health of the patient, using the global health score, resulted in a correlation of -0.51 ($p<0.001$). This is a relationship which has been widely demonstrated in previous disease-specific studies (Balkrishnan et al. 2003; Reiter-Purtill et al. 2008; Zashikhina and Hagglof 2009), and so the correlation was expected to be stronger in this study. This moderate correlation either suggests that family members are unable to accurately predict the health of the patient across all specialties, in contradiction to previous evidence (Housman et al. 2002), or that the family member’s life is not impacted more greatly when the patient’s health is low. This finding may suggest that family members are affected by a

patient's illness regardless of the severity of that illness, and that their quality of life is not reduced further when the patient's illness is more severe.

The lack of sensitivity to change data produced from the study is considered to be a limitation and is also discussed later in this chapter. Sensitivity to change is an ongoing process, often completed over a course of several months or years, and involving intervention studies, where a change in patient disease state is likely, or expected. The time limitations of this study meant that only a preliminary sensitivity to change study was designed, with 25 family members of patients recruited. The patients were not subjected to an intervention, and the results from the sensitivity to change study found that only two of the family members had reported a change in the patient's health state and were therefore eligible for the analysis. This meant that the numbers were too small to analyse, so no sensitivity to change data resulted. This could have been avoided by tying the sensitivity to change study in with a clinical intervention, or change in treatment. This is planned as a future study.

Evaluation of the FROM-16

The newly developed FROM-16 instrument is designed for "real life" use. Throughout the development of the measure, great emphasis was put upon the usability and simplicity of the FROM-16. With a growing number of quality of life instruments being developed and several disease-speciality family measures already in existence, the FROM-16 must be able to show advantages over existing measures, and be attractive to researchers and clinicians to use on a regular basis and not purely as a research tool. The title, instructions, items, response options and scoring of the measure are all contained on one A4 page. This reduces the chance of sections of the FROM-16 being lost during photocopying or pages being separated, and means that the completed FROM-16 can be filed alongside other A4 clinical notes. The single-page measure is also more appealing to family members, as the measure looks short and concise, and lack of completion from not noticing a second page is eliminated. The number of items in the FROM-16 is an additional factor in assisting its usability, as only 16 items are needed to capture the extent of the family impact of illness. During the design of the FROM-16 it was decided that the language used would be simple and the items should be readable by family members of all backgrounds and reading abilities. One of the ways this was implemented during the design process was by choosing not to include examples or lengthy explanations in the FROM-16 items. High readability was proven by the mean item length (5.6 words) and the Flesch readability score of 64.7. These factors lead to a short mean completion time of two minutes for the FROM (n=108). This short completion times gives the FROM-16 an advantage over other longer quality of life measures and will place less burden on the questionnaire administrator as the FROM-16 can be completed and scored easily and quickly. The quantitative content validity statistics can

be used to further evaluate whether the FROM-16 is successful in its design aim to be simple and easy to complete yet inclusive. All family members thought that the FROM was easy to complete, the response options were straightforward and the instructions were clear. The feasibility and acceptability of the measure were reinforced by the low volume of missing data in the cohort completing the final version of the measure. Furthermore a great majority thought that the FROM-16 items covered all areas of their life which had been affected. This demonstrates that family members see the FROM as user friendly and have few problems with completion.

As well as that of being user friendly, there are additional advantages to producing a concise and simple measure. In the future, the measure may be required to be translated into different languages. The simplicity and shortness of the items will allow for ease of accuracy of translation and the simple response option labels will help to reduce ambiguity during translation. This increases the international use potential of the FROM-16. Although the FROM-16 has proven high readability and simplicity, it also has the potential to be made even more user-friendly. For example, pictures or cartoons could be added next to each item to aid visualisation of each item for those with low literacy skills, and to help focus the mind for each item.

When evaluating the properties of the FROM-16, it is important to compare it to existing measures available to identify its strengths and weaknesses. The FROM-16 is shorter than the existing family measures, including the Impact-on-Family Scale (Stein and Riessman 1980), the Beach Center Family Quality of Life Scale (Hoffman et al. 2006), the Family Quality of Life Survey (Issacs et al. 2007) and the Family Strain Questionnaire (Ferrario et al. 2004), and has a shorter completion time. Of the four measures mentioned, the Impact-on-Family Scale (IOF) has the shortest completion time of 10 minutes compared to the time of 2 minutes for the FROM-16. The IOF also has the shortest number of items; 24 compared to the 16 items of the FROM-16. The Family Strain Questionnaire Short Form, developed in 2010, contains 30 items and can be completed in five minutes (Vidotto et al. 2010). The FROM-16 also has the advantage over existing measures that it was developed from interviews with family members of patients rather than from existing literature, so contains items derived directly from the words of family members themselves. This is an advantage over existing measures developed from literature and expert opinion (Ferrario et al. 2004; Issacs et al. 2007). The main advantage of the FROM-16 over other “family” measures is that it contains items designed for any family member of any patient with any disease. This is unique to the FROM-16 as the other measures identified all specify disease populations. For example, the existing measures have been designed for use in family members of paediatric patients (Hoffman et al. 2006; Stein and Riessman 1980) or caregivers of patients (Ferrario et al. 2004; Issacs et al. 2007). Although the measures have been designed for different

populations, similar domains and item themes to the FROM-16 can be seen. Both the FROM-16 and the other four measures identified contain domains and items relating to an emotional impact. Other overlapping themes include family relationships (Ferrario et al. 2004; Hoffman et al. 2006; Issacs et al. 2007) and financial impact (Issacs et al. 2007; Stein and Riessman 1980). Interestingly, the majority of the domains covered in the previous family measures are also covered in the FROM items. For example, the Family Strain Questionnaire contains a domain specifically called “thoughts about death”(Ferrario et al. 2004). Although this is not covered directly as an item in the FROM-16, there are several items relating to emotional impact which could be used to express this feeling, for example items about feeling worried or sad. One potential disadvantage of the FROM-16 over other measures is that it is restricted to self-administration use only. The Impact-on-Family Scale has the option to be interviewer administered if the family member has poor literacy skills (Stein and Riessman 1980). The FROM-16 has not been validated for interviewer administration and although self-administration would be the preferred methods of administration, there may be some benefit in carrying out a separate validation study in those with low literacy skills. A gap analysis study involving the FROM-16 and the four existing family measures would also be an interesting and beneficial future study, to understand the novel, specific themes which have been uncovered during the development of the FROM-16.

Although it is difficult to make direct comparisons between the FROM-16 and disease-specific family measures, many similarities were seen between themes and items in the disease-specific family measures. The Overactive Bladder Family Impact Measure (Coyne et al. 2010) contains items about difficulties with travel, social activities, caring for the patient, frustration and sleep, which are also contained in the FROM-16. In turn, the Family Dermatology Life Quality Index (Basra et al. 2007) contains items about emotional impact, relationships, social life, burden of care and work which are also contained in the FROM-16. The majority of items contained in disease-specific measures are covered by the comprehensive, generic items of the FROM-16. For example, the Quality of Life in Life Threatening Illness - Family Carer Version (Cohen et al. 2006) uses the term “distressed” for one of its emotional items. It is likely that this would be covered by the FROM-16 items of feeling sad, worried or frustrated. As “distressed” is such an extreme and emotive word, it would be expected that a family member would select the “A lot” response option to convey their experiences using the FROM-16. When making comparisons between the disease specific measures and the FROM-16, it is also important to bear in mind the target population. The Quality of Life in Life Threatening Illness - Family Carer Version (Cohen et al. 2006) is designed for carers of cancer patients and so contains items such as “Over the past two days (48 hours) I wondered if the place [the patient] was staying (home, hospital, other) was the right place to be”. Items like this may be specifically carer-related and

therefore not always applicable to the family members in this study, as many of them did not see themselves as caring for the patient, but were still affected by their illness. This all-inclusive aspect of the FROM-16, designed for family members who are not necessarily carers allows much broader use across family members compared to other measures.

In order to further assess whether the FROM-16 is likely to be appropriate for use in family members of patients with a wide range of illnesses, includes relevant themes and has the potential to be used as an alternative to existing measures, the content of the FROM-16 was compared with two of the identified disease-specific family measures. On reviewing the 10 items in the Family Dermatology Life Quality Index (Basra et al. 2007), 90% were covered by items in the FROM-16. The only item not included related to an increase in housework. In some situations, this may be covered by the FROM-16 item "Caring for my family member is difficult". On reviewing the 19 items in the Overactive Bladder- Family Impact Measure (Coyne et al. 2010), 95% were covered by items in the FROM-16. The only item not covered specified a feeling of guilt. Other items were approximated by the FROM-16, for example feeling annoyed (FROM-16 item containing the term "angry"), or impatient (FROM-16 item containing the term "frustrated"). This gives assurance that it is likely that FROM may be used appropriately in families with a sufferer of these diseases, though this of course will eventually have to be specifically tested in a prospective study. Concerning those items not covered by the FROM-16, these are all themes which were identified during the qualitative analysis in this study, and were developed as items in the developmental version of the FROM-16. It may be that different versions of the FROM are required for different specialties, but this is unknown without further testing.

In previous work looking at the impact of illness on family members, and on those closest to the patient, this impact has been referred to as a "secondary impact". In one dermatology study, the close social group around the patient affected by the impact of the patients' illness was described as the "Greater Patient concept" (Basra and Finlay 2007), an analogy to a city with a nucleus and surrounding suburbs. The authors of this study suggested that this term could be applied to specialties other than their own. Throughout the course of this study, these terms to describe the impact of illness on family members were considered in relation to the themes and experiences described by the participants. It was felt that the term "secondary impact" was generally unsuitable when describing the impact of illness on family members. Although the impact is "secondary" to the patient and their illness (it affects someone other than the patient themselves), the impact is a "primary" impact, as it is being described by the family members and relates directly to the way that they feel and the ways their lives are affected. The impact can be seen as "secondary" to the patient, but it is important to consider it is "primary" to the family member as it being reported directly by themselves. This terminology should be considered when designing and reporting studies

using the FROM, and whether the focus of the study is primarily on family members, or is a secondary outcome of a patient-related study. As the term “secondary impact” was felt not to always be appropriate to the impact of illness on family members, the term “family impact” was felt to be more focused and offer more clarity.

At all stages of the study the family members gave feedback about the importance of the study, the study design and the end product, the FROM-16. The comments made by family members were positive and encouraging. One of the most common comments made by family members was regarding the high importance and relevance of the study, and about how they had not had the chance to talk about the impact on themselves before. Family members talked about how infrequently they were asked about their own feelings, both by their own family and friends and also by healthcare professionals. This meant that their feelings often stayed bottled up inside and as a result built up rather than being resolved. During the interviews, many family members had to be reminded, or re-directed to talk about the impact on themselves, and not the patient, as they were so used to talking about the patient and how they were affected. Many found it difficult to talk about their feelings, as this was the first time they had been asked about the impact on themselves, and a surprisingly high number of participants became visibly emotional, especially during the qualitative interviews. Although all were given the option to withdraw from the interview when they became upset, all participants insisted on continuing, suggesting that they understood the importance of the study and were both willing and keen to participate. The past experience and training undertaken by the interviewer helped to ensure that participants felt comfortable during the interview, and this was proven by the rich, in-depth personal information obtained. Family members also recognised that other members of their own family were affected and could benefit from this study, and many expressed how they wished that all of their family could be interviewed individually. During the validation stage of the study, many family members commented informally about the items in the FROM-16, particularly how relevant they were to them, and how all of the important areas of their life had been included in the measure. One particularly memorable quote from a participant was:

“This questionnaire is spot on. How did you come up with the questions? It’s covered everything, it’s like you have jumped into my head. My wife and I had an honest discussion the other day about how her illness has affected me and these were just the kind of things we talked about”.

As well as the informal feedback from family members, there was informal feedback and involvement from patients. Many of the patients involved in the study encouraged their family members to take part and felt that it was important that their views were heard. Many said that they recognised the impact that their illness was having on their family member(s)

and the lack of interest in the area, and they thought the family member should be offered more support. This was echoed by the collaborating consultants in the study, who strongly recognised the importance of being able to measure the impact of illness on family members of patients. As this area of research is not widely recognised and has been largely overlooked in the past, it was expected that the consultants may have had reservations about the study or not recognise its potential relevance to their clinical practice. Involving the consultants so closely in the recruitment of participants and designing of the FROM-16 gave the investigator a real-life insight into their opinions and reactions to the study. From the initial invitation meeting with each consultant, through to the dissemination of the study results, many expressed their personal interest in the study and suggested ways they would like to use the FROM-16 in the future. At each stage of the study, the clinicians were interested in the results obtained and offered their input and suggestions with regard to the development of the FROM-16. The overwhelming positive and enthusiastic feedback from both family members and clinicians was unexpected, but reflected and strengthened the views of the study team regarding the importance of the study. The interest in the study from healthcare professionals is also encouraging for the future use of the FROM-16 and the development of the research area.

Potential scope and usage

Access and future use of the FROM

As the first generic family quality of life measure, the FROM has the potential to be used in a wide variety of situations. These range from being used clinically to identify areas of family members' lives which are affected, to being used as a secondary endpoint in a clinical trial of a new drug. When choosing to use the FROM, it is important for the investigator to identify their goals for using the measure, to familiarise themselves with the scoring of the measure and to plan how the results will be used to meet their goals. It is planned that permission will usually be given to any researcher who wishes to use the FROM, and that it will be easily accessible to researchers through a dedicated website. Clinicians or patients who wish to use the FROM for routine clinical use will be able to do so directly, without seeking permission, by downloading the FROM from the website. Members of the FROM study team will be available to guide researchers on the use of the FROM, and a manual will be available detailing the patient and family member populations used in the development of the FROM, the use and scoring of the FROM and information regarding the validation of the measure. Researchers will be encouraged to use the FROM both as a single tool, and alongside other measures as part of larger projects.

Clinical trials

Due to the robust development of the FROM and its extensive validation, the measure has the potential to be used in clinical trials of pharmaceuticals sponsored by the industry, for example as a secondary endpoint. As well as demonstrating that a new drug treatment or device benefits a patient in terms of symptom control or quality of life improvement, pharmaceutical companies will now be able to show that a drug treatment has an impact on the lives of family members of patients. Although no specific guidance is offered by regulatory agencies with regard to the measurement of impact of illness on family members, the FDA have issued guidance relating to the use of patient reported outcome measures in trials (FDA 2009). This guidance advises that such measures should have demonstrated validation including the implementation of several of the psychometric tests carried out during the validation of the FROM, including test re-test and construct validity. In order to be used in a clinical trial, the FROM will need to have demonstrated responsiveness to change, something which has not been tested during this study, but will need to be addressed in future work. The FDA also recommends that patient reported outcome measures are re-validated in the intended population within the trial. In particular, re-validation of content validity is important, through the process of cognitive debriefing, as there remains a possibility that new concepts that are not covered by the FROM could emerge for specific diseases, though the extensive and excessive interviewing beyond saturation in the qualitative study make this unlikely. Although the reporting of family quality of life is a new concept within clinical trials, FROM could have a potential role in assessing treatments which through their effect have a potential impact on the family. It is as yet unknown whether regulatory agencies such as the FDA or EMA might in the future accept label claims relating to the impact of a treatment on families of patients as well as on patients themselves, however the simplicity of the FROM allows for the possibility of the measure to be incorporated into a trial as an additional measure with minimal cost or time burden. The FDA already recognises the influence of the family in the field of patient reported outcomes, as their 2009 guidance states that PRO instrument items can be developed from “literature reviews, transcripts from focus groups, or interviews with patients, clinicians, family members, researchers, or other sources”(FDA 2009). Interest in, and use of, the FROM in the commercial and industrial world has the potential to influence regulatory agencies to issue specific guidance on the use of family measures in trials.

Service provision

The FROM also has the potential to be used in service provision and treatment regulation. Health Technology Assessment (HTA) authorities such as the National Institute for Health and Clinical Excellence (NICE) in the UK have shown an increasing interest in using patient reported outcome measures in development of treatment algorithms and guidelines (Doward

et al. 2010). The FROM has the potential to be used alongside patient measures in service provision, particularly when a new treatment has a possible benefit to families. One example of the potential use of the FROM in service provision could be, say, in the context of an expensive treatment with a once weekly hospital-based administration, when a current thrice-weekly treatment has shown to have an impact on both the patient and the family members' lives. The inclusion of a family-based measure, particularly in diseases where families are highly impacted or involved in care and treatment, would encourage assessing clinicians to take a more family-centred approach to treatment and service provision.

The development of the FROM-16 also provides the opportunity for the exploration of utility data relating to family members of patients. Patient health states relating to different FROM-16 scores could be developed and the utility values for family members of patients with different diseases could be calculated using the standard gamble or time trade-off (TTO) methods. The attributes for each health state could be developed for family members using the items of the FROM-16 and the qualitative data from this study. Utilities for family members could be calculated and attached to the FROM-16. The FROM-16 utility data could be then used as an additional tool in service provision, for example in the evaluation of pharmaceuticals by NICE, alongside patient utilities (Brazier 2008).

Disease education and support services

Due to the comprehensive nature of the FROM and the items being derived from the experiences of family members themselves, the measure has the potential to be used in disease education programmes. Although disease education programmes exist for patients, few support programmes are available specifically for families. In this study, we have identified the great need for support for families of patients with chronic illness, and both the qualitative results and themes and the FROM itself could be used as the basis for setting up a generic national family support group. A few support groups exist in the UK for families of patients with specific diseases, for example Rethink who provide support to family members and patients with mental illness (http://www.rethink.org/index.html). Many other disease-specific support and education websites focus on "carers" and so may be unsuitable for, or seem inaccessible to, family members. There are extensive online resources available for patients, including websites with directories of patient agencies, but no such website exists for family members of patients. The qualitative themes identified in this study could be used as a basis for planning the areas of support provided by a generic family support service, and the FROM has the potential to be used as a screening tool to identify areas of concern for family members participating in such a support group. As many of the family members interviewed in this study expressed relief at being able to open up and talk about the impact

on their lives, and expressed feeling “alone”, access to a support group and being able to meet others in similar positions may also have a positive impact on family members’ lives.

In addition to being used as a guide to establishing family support services, the FROM also has the potential to be included in existing services to improve understanding of the family impact of disease. For example, the FROM could be used in existing patient education programmes, to indicate that the service providers are interested in taking into account the views of the patient’s family, and to tailor some of their course material towards the secondary impact of disease. The FROM could be given to family members of patients in advance of the course and the results used to inform the course leaders of the family themes deemed important to cover during the programme. Support could then be offered in relation to these themes in particular. This use of the FROM has the potential to improve communication between patients and members of their family, and to encourage them to talk about and address issues which are affecting both family members and patients. One of the major themes identified in this study was “Family Relationships”, and using the FROM in educational programmes with family members and patients could help to address some of the components of this theme, including communication between the patient and family member and arguments and tensions within the family. The items in the FROM provide family members and patients with starting points for discussions and common terminology to use when talking about the impact of disease.

Support and care for family members could also be provided through established community-based patient programmes such as the Expert Patients Programme (http://www.expertpatients.co.uk/). This programme already has elements of family-related support, such as a specific parent programme and the use of software designed to help patients and family members communicate about their experiences of living with a disease. The use of a generic family measure like the FROM could help to assess the impact of the illness on families and identify specific areas where they require support. The short completion time and ease of scoring mean that the FROM could easily be incorporated into existing programmes. The FROM could be used to identify any change in family quality of life before, after or during the programme and could be used as a tool to evaluate the effectiveness of such programmes on families as well as patients. The standardised scoring of the FROM and the lack of weighted items means that it could also be used to compare the effectiveness of two or more different support or education programmes, and be used in advertising claims for such groups.

Research

Being the first measure of its kind, the FROM has many uses and roles within research. The existence of a generic family measure now allows for the field of secondary impact of

disease and family impact to be explored more widely. Comparisons can be made between the family impact of different diseases, different treatments for the same diseases and the impact on different family members within one family. Findings from disease-specific family studies can be compared to findings in the same population using the FROM. The relationship between patient and family quality of life will be able to be further explored using the FROM as a key tool in this research area. As well as individual research studies, larger population and social studies looking at the impact of illness on family members will now be possible to be carried out using the FROM as the primary research tool. For example, the impact of illness on family members of patients could be compared between geographical areas, NHS trusts, or GP practices, looking at both the extent of the impact (total FROM domain scores) and the areas of most concern (endorsement percentages of individual items). The development of the FROM opens up a whole new field of healthcare research, one which has previously been largely untouched and unable to be measured. The potential impact on medical and social research is considerable, with the FROM being able to play a key role in many types of studies.

Clinical use

The FROM has the potential to be used widely in clinical environments. The use of the FROM in clinical practice could have many of the same previously reported advantages of using patient reported outcomes in clinical practice (Greenhalgh et al. 2005). The use of the FROM before or during consultations with the patient and family member(s) could encourage a more family-centred approach to care. In clinical practice, the FROM could be used as a tool in multi-disciplinary team meetings to improve communication between healthcare professionals, encouraging them to discuss and act on decisions regarding patient care and treatment which have an impact on family members, and consider this potential impact when starting new treatments. As well as improving communication between healthcare professionals, use of the FROM empowers the patient to raise family issues during consultations and to make the clinician aware of the impact of their disease on their family. In the same way that previous research has suggested that partners attend medical consultations with patients to be given the chance to discuss issues relating to the family (Rees et al. 2001), it may also be of benefit for other family members to be invited to attend consultations with the patient, with the FROM being used as a tool to identify issues for discussion.

Currently, if a clinician wanted to measure the impact of illness on a patient's family, they would have to use a disease-specific measure, if an appropriate one existed. There is a lack of evidence of the routine use of disease-specific measures in clinical practice, and many authors of these measures are very vague about their potential for future use, for example

suggesting use in “research and clinical settings”(Coyne et al. 2010). Furthermore, there is no evidence that these existing measures are comparable in their scoring, so the clinician would be limited to measuring the impact of one disease. From this study, it has been hypothesised that the FROM could be used across all disease areas, as a generic measure. It can be used to compare the impact of illness on the family across the whole of medicine, for example when measuring the impact of a patient with more than one illness. This is particularly useful in general practice, where a general practitioner may wish to evaluate the impact of several illnesses on family members of patients. The FROM is therefore more flexible in its scope of use than existing disease-specific family measures and may replace the use of them in many clinical areas.

The FROM can be used to address a number of goals in clinical practice, many of which complement the goals and uses of patient reported outcomes in clinical practice (Greenhalgh et al. 2005; Snyder et al. 2011). The FROM could be used as a secondary measure when assessing the improvement or worsening of a disease, or when comparing or assessing treatments. It gives the clinician extra useful and relevant information to work with alongside the patient’s clinical and QoL data when making decisions regarding patient care. As well as being used as an assistive monitoring tool, the FROM could also be used as a screening tool in clinical practice to detect potential patient problems. Used routinely, the FROM could identify problems with the patient’s management or treatment which are not necessarily having a large impact on the patient, but have an impact on the family. With its short completion time, using the FROM routinely would cause minimal burden to the clinician or family member.

Challenges

In general, clinicians feel it is appropriate to discuss quality of life issues with patients and see quality of life information as important (Greenhalgh et al. 2005). However, these findings are only in relation to patient data, and not data concerning the impact of illness on family members. The use of patient reported outcome measures in clinical practice is a relatively new area, and one which is still in early stages of exploration and use in many disease areas. Will the introduction of the concept of measuring family quality of life in clinical practice be too ambitious given the lack of routine collection of patient data in clinical practice? The area of family impact of disease is one which will be new to the majority of clinicians as until a concept can be measured, it has a minimal profile or “existence” with more importance in the scientific world. The results of this study and the publication of the FROM could subtly alter the mindset of the medical profession in terms of placing the patient in a more family-centred context. As well as altering the mindset of the medical profession, this study is also likely to produce data which is a big challenge to the profession, as the

impact of illness on families has been shown to be great but largely overlooked. At the moment, the impact of illness on family members is generally ignored, and this study challenges the medical profession to respond to this significant issue and identify ways to support family members of patients.

As well as changing the mindset of the medical profession in general, there may be a need for individual healthcare professionals to alter their practice and their way of thinking, and to accept the fact that the family burden of disease is so great. Whilst this poses a hypothetical challenge to individuals, the positive and enthusiastic response to the study by healthcare professionals involved is encouraging. The majority of healthcare professionals recognised the importance of the study results and the need for a generic family outcome measure, particularly in specialties where family measures do not already exist.

From the experiences of the study team, it is apparent that healthcare professionals are already routinely thinking about how illness affects families, with many clinicians providing examples specific to their speciality during the focus group discussions. In particular, the nurses involved recognised the great impact of illness on family members through working closely with patients in their home environment. The challenge posed by the results of this study is how to bridge the gap between the knowledge and ideas of the healthcare professionals and using this information, along with the FROM, to inform and improve clinical practice.

Study implications

- The literature review presented in this thesis identified that no previous study or measure exists to assess the impact of illness on family members of patients over a wide range of conditions. The literature review will help future researchers understand the gaps in the area and an overview of the previous related work.

- The qualitative interviews with family members of patients from 26 specialties identified 10 key family quality of life themes. These themes may be used in the future development of this area of research, and direct researchers towards the areas of importance for family members. In addition, this could inform development of an organised generic family support group.

- This study highlights the importance of using an appropriate definition of “family” when carrying out research in the area, and the impact that disease has on family members of patients who are not necessarily carers.

- This study highlights the extent of the impact on family members, which has been previously immeasurable in most specialties. It identifies a hidden area of healthcare which needs to be explored and studied further.

- This study provides a comprehensive and robust way of measuring the impact of illness on family members of patients.

- The results of this study demonstrate a relationship between patient quality of life and family member quality of life. This relationship has not been shown in a generic context before.

- The results of this study demonstrate a relationship between the health of the patient and the family member quality of life, as seen before in individual disease areas, but never in a generic context.

- Preliminary information about which specialties are most affected has been provided, and this can be investigated further by future research.

- This study has shown how both Rasch and factor analysis can successfully complement each other in the development of an outcome measure. This is an approach which can be explored further in future studies.

- This study provides information which challenges the mindset of the medical profession to develop strategies to address the unmet need identified.

- The measure produced in this study (the FROM) has implications for use in clinical practice, research, disease education and support, clinical trials and service provision.

Limitations

Although the study was designed to the highest possible standards, there were still a number of limitations:

- Although the literature search carried out during the study was structured it was not strictly systematic (according to PRISMA guidelines), meaning that some papers could have been missed during the search. It is difficult to assess the likelihood of this happening, and if there were no time constraints associated with the study a

systematic review would have been carried out. This would have ensured that no existing publications were missed during the literature search.

- The majority of participants were White British. It is possible that culture and ethnicity could influence the way family members are affected by illness, and some themes could be of greater importance to specific cultural groups. This could be determined by further sampling of family members of patients from different ethnic backgrounds. It is also important to be aware of potential cross-cultural issues with the FROM-16, as translation has been suggested as potential future work. The assessment of quality of life in different cultures depends upon a variety of factors including cultural based illnesses, perception and response to illness and religious and social behaviours (Bullinger 1997). It is likely that there will be specific cultures where the items of the FROM-16 are not applicable to family members, particularly those which are very different to the western society in terms of views on illness and ways of life. As well as the aspects of life the FROM-16 covers, it is important to consider the concept and definition of "family" when assessing the cross-cultural suitability of the FROM-16. For example, in traditional Indian cultures where women are often considered subordinate to men, and elderly family members often remain with their families (Mullatti 1995), many of the items of the FROM-16 may be irrelevant. For example, the female family members may not go out to work, or have knowledge of the household financial situation. In turn, when an elderly member of the family is unwell, the other family members may feel that it is their duty to care for them, and many of the emotional questions in the "Emotional" domain of the FROM-16 may be irrelevant, or even offensive to individuals of this culture. Further testing and content validity studies using the FROM-16 in different cultures are required, to measure the extent of its cross- cultural relevance.

- The median age of family members during the qualitative and quantitative stages of the study were 56 years and 53 years respectively. Although the age range of family members was wide, ranging from 18 years old to participants in their 90s, the median age was high, suggesting that the sample was not representative of the full age range. However the number of patients interviewed who were aged 18 – 40 was 24. This could result in some of the items in the FROM being less relevant to younger family members. This could be determined by carrying out a large scale study with equal numbers of family members across all age categories.

- Several of the items in the FROM contain the word "impact", but do not specify whether this impact is positive or negative. As discussed in chapter 3, the aim of the FROM was to measure only the negative impact of illness on family members of

patients, as the positive effects were so rarely seen during the interviews. Using the word “impact” suggests that the items can be answered positively or negatively, but the scaling chosen does not permit positive responses to be recorded; only negative and neutral. This is a limitation of the measure, as it may be seen as confusing for some family members and affect the way that they respond to items. This may have been avoided by wording all of the items in a clearly negative way, or allowing for positive scoring. As the positive effects were so rarely seen during the interviews, the most suitable option would have been to word the items negatively. This may have improved the clarity of the measure.

- Another limitation is that not all medical conditions from each specialty were represented fully, both in the qualitative and quantitative phases of the study due to time restrictions of the study. Physicians were asked to select patients with different conditions best representing their specialty. This expert knowledge and the large total number of interviews carried out beyond the saturation point helped to ensure a representative sample. However, there is still a chance that selection bias may have occurred during the identification and recruitment of participants as purposive sampling was used (Tongco 2007). This method of sampling was chosen by the investigators as it provides a cross section of rich, detailed data and in the case of this study, it ensured that a wide variety of patient medical conditions were sampled. Although we presume that the FROM is generalisable with respect to family members of patients with all diseases, this should be demonstrated definitely for individual diseases or disease areas.

- The small numbers of patients and family members collected from each speciality could also be considered a limitation of this study. However, the qualitative interviews proved that there were a limited number of ways family members lives are affected, and commonality was seen across the specialties. Furthermore, during the face validity testing, 87.4% of family members felt that the FROM covered all areas of their life which had been affected, and the majority of the feedback comments provided to explain this were concerned with individuals’ examples of items, which were covered more broadly (e.g. “There is no item to cover the fact that I am worried whenever my son leaves the house”). This is also reflected in the fact that although the interview saturation point was number 40, interviews were continued to 133.

- The majority of family members in the study were sampled from patients in secondary care settings including hospital wards and outpatient clinics. Only one of the 26 specialties in the study was based in general practice. It is possible that additional

themes specific to family members of patients in primary care settings would have been revealed if a larger sample was taken from primary care. This limitation can be addressed in the future by carrying out a detailed content validation study in family members of primary care patients.

- Due to the small sample size and the lack of intervention or change in patient disease state, no reliable sensitivity to change data emerged from this study. This is the only psychometric data lacking for the FROM, and the lack of evidence of responsiveness to change limits the potential use of the FROM as it is currently presented. It is possible that reducing the number of response categories from five to three during the Rasch analysis decreased the responsiveness and sensitivity (and therefore power) of the measure as this produced a smaller range of possible total scores and differences between family member quality of life scores are less prominent and subtle differences in scoring may be unable to identify In order to be used in many of the ways proposed by the study team, the FROM must undergo further sensitivity to change studies to ensure its reliability for use in interventional research. This is proposed as a future study.

- Although the FROM has been designed to be used in any family member of a patient, a high proportion of the family members sampled in this study were partners. Although parents, siblings and children were also represented, this could have had an impact on the themes identified, and they could be considered to be more orientated towards partners of patients. A future large-scale study involving a sample of many different family members would determine the extent of the potential partner bias in the study.

- During the psychometric testing, a web-based version of the FROM was used. This web-based tool was not tested for its equivalence to the paper version of the FROM. This could have potentially introduced bias into the study for those family members who used the web-based tool. This is a limitation which can be corrected in future work, though there is no reason to think that the paper and web versions will not be equivalent.

- The FROM and the WHOQOL-BREF were used as comparators in the construct validity stage of the study. However, these two measures have different recall periods (immediate and last two weeks), which may have affected the answers given by participants. The influence of this on the construct validity is unknown but expected to be minimal.

- One possible limitation of the FROM-16 is that the closeness of the relationship of the family member to the patient could distort their perception of the disease impact. This could result in exaggeration or underestimation of certain areas of impact and skew the resultant measure score. However, as the impact on the family member is subjective, this bias could prove negligible. The extent or certainty of this impact has not previously been demonstrated, but the availability of the FROM-16 now allows the possibility of specific studies to address this.

Future work

- A large-scale multi-specialty study using the FROM is planned. A larger number of family members of patients from each specialty or disease area will be sampled, and further information regarding the scoring distribution and properties of the FROM will be collected. During this study, a greater number of patient conditions will be sampled, and the larger sample size will enable further testing of the psychometric properties of the FROM. The summary data from such a study could be used as a baseline or comparator for further studies, or when validating the measure in specific diseases. A large-scale study will provide information as to which items on the FROM are most highly endorsed by family members, and the areas where family members need more support. Further comparisons will be made with family members of patients between different specialties, and specialties where family members are most greatly affected will be identified.

- The original 31 items of the developmental version of the FROM could be re-visited, alongside the qualitative data to produce several different versions of the FROM. These could vary by length and intended purpose of use; for example, a short version could be produced with one or two summary items from each of the two FROM domains. Computer adaptive testing (CAT) could be used to produce a dynamic version of the FROM with items varying depending upon relevance to the patient's disease. New versions of the FROM will need to be psychometrically re-validated to the same robust standards as the original FROM-16 measure. Producing a number of different versions of the FROM will increase its use potential and practicality, but could result in confusion for potential users.

- In this study, only one family member from each patient was sampled. Future studies could include using the FROM as a tool to compare the effects of a patient's illness on different members of the family, and identify those members of the family who are

most affected. Separate content validation could be carried out for the FROM within different relationship groups, for example parents, children or siblings. Items of the FROM which are most relevant for each relationship group could be identified and provide information as to the areas of support needed for each family member of a patient.

- Although only small numbers of secondary relatives were included in this study, the effects of illness on more distant relatives (for example cousins and grandparents) could be studied. This could be compared to the results for closer family members, to determine the distance that the family impact of disease travels as family members less close to the patient are introduced. Other comparison groups within families could include those who live with the patient and those who do not (although this study suggests that there will be little difference). Families where more than one member is unwell could also be compared to families where there is only one patient, to assess the effect on the well family members.

- The FROM could be compared with other existing family measures. A high correlation would be expected to be seen between the scores of the FROM and the scores of other family quality of life measures, due to the similar effects seen on family members from different specialties in this study. One interesting piece of future work would be to carry out a gap analysis between the FROM and other existing family measures, to identify those concepts where the FROM differs from other measures, and to further demonstrate the need for a generic family measure.

- As an additional output to the FROM, a conceptual framework and model of family quality of life could be designed to represent the themes identified during the qualitative work and their relationship to one another. The themes covered by the FROM and existing family quality of life instruments could also be included in the model.

- The difference between the impact of illness on family members and carers could be assessed using the FROM and one of the existing measures for carers of patients. This would determine whether there is as much of a difference between the impact on family members and carers as was hypothesised in this study.

- A study involving a control group should be carried out with the FROM. The control group could be made up of family members of healthy individuals, or patients who have been cured of a disease. The inclusion of a control group would help to prove

that the impact on family members is a result of the patient's illness with minimal influence from other external factors, therefore strengthening the reliability of the FROM.

- During further development and studies involving the FROM, it is important to establish population normative values ("norms") or the measure. This could include norm values for family members of a healthy population, or norm values for different disease areas or family members. These norm values can then be used as benchmarks for FROM scores and for comparison between different disease areas.

- This study has identified areas of daily activity where family members are affected and require support. In future work, any existing family support groups could be identified and the areas of support they provide to families could be compared to the themes included in the FROM. Additional areas where support for families is needed could be identified and included in these support groups. The information gathered through the development and validation of the FROM could be used as a basis of content for a new family support group or internet network.

- At the moment, the only information provided by the total FROM score for each domain is that a higher score is equivalent to a lower quality of life. Banding of scores in a future study would aid with score interpretation and the impact that different total scores have on family members' lives. These score bands could be used to identify improvement or decline in family member's quality of life, or could be compared to the score banding for generic patient measures.

- The results of the sensitivity to change study were inadequate to provide evidence as to the responsiveness of the FROM. Therefore a future large-scale interventional study will be required to collect sensitivity to change data. The possibility of using the FROM in a treatment intervention study will also be explored.

- In order for the FROM to be accessible and easy to use, an electronic version of the measure should be developed and validated. This would enable the FROM to be used on electronic devices such as computers, tablets or smart phones. The web-based measure used in the psychometric testing study should also be tested for its equivalence to the FROM to increase the reliability of the test-retest data.

- A large-scale study using the FROM in a primary care setting should be carried out to identify any differences between the impact of illness on family members in primary and secondary care which were not identified during the present study.

- The FROM should be validated in individual disease areas and specialties for future disease-specific studies. The psychometric validation procedures outlined in this thesis provide guidance on how future disease-specific validation studies should be completed. Of particular importance is the content validation and cognitive debriefing of the FROM in individual disease areas.

- This study showed a correlation between family quality of life and patient quality of life using the FROM and the generic patient measure, the WHOQOL-BREF. Future studies could include correlating the FROM with disease-specific patient measures, to further improve the construct validity of the FROM.

- One of the many proposed uses of the FROM is in clinical settings. It is important to consider how the measure will be integrated into the clinical practice setting and how results will be used to influence practice (Snyder et al. 2011). Therefore, it would be useful to carry out a feasibility study with clinicians and use the results of this study to influence the future recommendations for clinical use of the FROM.

- Although the FROM is designed mainly for use with family members of patients with chronic diseases, future studies could be carried out to identify the areas of a patient's specific disease progression where family members are most likely to be affected, and therefore require support. For example, the FROM could be administered when the patient first experiences symptoms, immediately after diagnosis, during different types of treatment, or before and after surgery.

- Several of the specialties included in this study were paediatric specialties. Although similar family quality of life themes were seen across all areas of medicine, an interesting future study could be to carry out a paediatric-specific study with different family members of child patients, and compare the impact of child and adult patients on family members.

Conclusion

This study provides new and detailed information into the ways that family members lives are affected by patients' illness. Many of the specialties and disease areas included in this study have been previously overlooked in terms of family impact, and the results from this study have helped to understand both the individual areas of family members lives which are impacted, and also the extent of the impact. The qualitative results have proven that the impact of illness on family members is widespread and profound across all disease areas, particularly the emotional impact felt by those closest to the patient. This study has also shown that family members do not receive enough support to cope with the issues that they face, and that these issues are very rarely addressed by healthcare professionals.

Previously, the term "secondary impact" has been used when describing the impact of illness on family members, but this study has shown that this impact is far greater than a secondary impact, and that the term "family impact" is more comprehensive and offers more clarity. The most important outcome of this study is the Family Reported Outcome Measure (FROM), as the concept of family quality of life is now possible to be measured, quantified and brought to the attention of healthcare professionals and researchers. This unique instrument is simple, easy to complete and score, and this study has shown evidence of its reliability and validity. The FROM has the potential to be used in many different settings, including clinical settings, clinical trials, research and disease education. The results from this study, and the development of the FROM provide a platform for further research into family quality of life.

Although the numbers sampled from each of the 26 specialties in this study were small, a preliminary comparison between specialties was made, and the data showed that family members of patients from certain specialties were affected more than others. The study also provides further evidence that the patient's quality of life correlates with that of the family member, and that the patient's health state correlates with the family member's quality of life. These findings should be considered by clinicians when making decisions regarding patient care and treatment, and how the family are affected by decisions made. It is hoped that the results from this study will help the medical profession to focus towards a key area of healthcare which is often overlooked, and give the family members a stronger role in patient care, encouraging shared decision-making. The views and needs of family members should be taken into account during medical consultations, and new strategies should be put in place to provide support to family members as well as patients.

This thesis described in detail a new, exciting and highly relevant area of medicine. It challenges the way that healthcare professionals view patients and families, and provides a more detailed and comprehensive picture of the areas of family members' lives which are affected, and the similarities between medical specialties. It challenges healthcare

professionals to think beyond the patient, and whilst healthcare is often described as “patient centred”, this study proves that the impact on the family is a significant issue which needs to be considered and addressed by all healthcare professionals.

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PUBLICATIONS AND PRESENTATIONS

Conferences- Keynote speeches

Golics CJ, Basra MKA, Salek MS, Finlay AY. The Impact of Disease on the Lives of Family Members. 14th Congress of the European Society for Dermatology and Psychiatry, March 2011. Zaragoza, Spain.

Conferences- Oral presentations

Golics CJ, Basra MKA, Salek MS, Finlay AY. A qualitative inquiry into the effects of disease on the family and the partner- "The Greater Patient". International Society for Quality of Life Research (ISOQOL) 18th Annual Conference, October 2011. Denver, USA.

Conferences- Poster presentations

Golics CJ, Basra MKA, Salek MS, Finlay AY .The Development and Content Validation of a Novel Generic Family Quality of Life Instrument: the Family Reported Outcome Measure. International Society for Quality of Life Research (ISOQOL) 19th Annual Conference, October 2012. Budapest, Hungary

Golics CJ, Basra MKA, Salek MS, Finlay AY. The Family Reported Outcome Measure©: initial validation of a generic QoL measure for family members of patients with chronic medical conditions. 42nd Annual Meeting of the European Society for Dermatological Research, September 2012. Venice, Italy.

Golics CJ, Basra MKA, Salek MS, Finlay AY. Diseases profoundly affect the quality of life of family members of patients. 41st Annual Meeting of the European Society for Dermatological Research, September 2011. Barcelona, Spain.

Journal articles

Golics CJ, Basra MKA, Finlay AY, Salek MS. The development and validation of the Family Reported Outcome Measure (FROM-16)© to assess the impact of disease on partner or family member (submitted).

Golics CJ, Basra MKA, Finlay AY, Salek MS. The impact of patients' disease on family quality of life: an experience from 26 specialties. Int J Gen Med (in press).

Golics CJ, Basra MKA, Salek MS, Finlay AY. The impact of disease on family members: a critical aspect of medical care. JRSM. 2012 (in press).

Abstracts

Golics CJ, Basra MKA, Salek MS, Finlay AY. The development and content validation of a novel generic family quality of life instrument: the family reported outcome measure. Quality of Life Research 2012; 21:S1-132.

Golics CJ, Basra MKA, Salek MS, Finlay AY. A qualitative inquiry into the effects of disease on the family and the partner- "The Greater Patient". Quality of Life Research 2012; 20:S1-106.

Golics CJ, Basra MKA, Salek MS, Finlay AY. Diseases profoundly affect the quality of life of family members of patients. Journal of Investigative Dermatology.2011;131:S1-S115.

Golics CJ, Basra MKA, Salek MS & Finlay AY. The impact of disease on the lives of family members. Acta Dermato-Venereologica 2011; 91(2): 219-220.

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APPENDICES

APPENDICES CONTENTS

Appendix A

GIG CYMRU NHS WALES
Canolfan Gwasanaethau Busnes
Business Services Centre

South East Wales Research Ethics Committee Panel C
Direct Line: 02920 376823/376822
Facsimile: 02920 376835
Email: Carl.phillips@bsc.wales.nhs.uk

Miss Catherine Golics
PhD Student
Cardiff University
Centre for Socioeconomic Research
Redwood Building
King Edward VII Avenue, Cardiff
CF10 3NB

21 May 2010

Dear Miss Golics

Study Title:	**The Conceptualisation, Development, Validation and Practical Application of a generic Family Quality of Life Measure**
REC reference number:	**10/WSE03/12**
Protocol number:	**10**

Thank you for your letter of the 17 May 2010, responding to the Committee's request for further information on the above research, and for submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation [as revised], subject to the conditions specified below.

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

- Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

- For NHS research sites only, management permission for research ("R&D approval") should be obtained from the relevant care organisation(s) in accordance with NHS research governance arrangements. Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk. Where the only involvement of the NHS organisation is as a Participant Identification Centre, management permission for research is not required but the R&D office should be notified of the study. Guidance should be sought from the R&D office where necessary.

- Sponsors are not required to notify the Committee of approvals from host organisations.

- It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The final list of documents reviewed and apprcved by the Committee is as follows:

Document	*Version*	*Date*
Covering Letter		08 April 2010
REC application	IRAS Version 2.5	09 April 2010
Protocol	10	19 May 2010
Investigator CV	CJ Golics	01 February 2010
Participant Information Sheet: Stage 2 Family Member	3	17 May 2010
Participant Information Sheet: Stage 2 - Patient	3	17 March 2010
Participant Information Sheet: Stage 4 - Family Member	3	17 May 2010
Participant Information Sheet: Stage 4 - Patient	3	19 May 2010
Participant Information Sheet: Stage 5 - Family Member	3	17 May 2010
Evidence of insurance or indemnity	Cardiff University	30 July 2009
Letter from Sponsor	Cardiff University	02 February 2010
CV	Professor Salek	01 February 2010
Letter from Cardiff University		08 April 2010
Supporting Letter from University of Glamorgan		30 March 2010
Protocol Flow Chart	1	01 February 2010
Proposed Interview Schedule	1	03 March 2010
Participant Information Sheet: Stage 5 - Patient	3	17 May 2010
Participant Information Sheet: Stage 6 - Family Member	3	17 May 2010
Participant Information Sheet: Stage 6 - Patient	3	17 May 2010

Participant Information Sheet: All Stages Patient - Child Age 6-10	3	17 May 2010
Participant Information Sheet: All stages Patient - Child Age 11-15	3	17 May 2010
Participant Consent Form: Stages 2,4,5 Family Member	2	08 March 2010
Participant Consent Form: Stage 2,4,5 Patient	2	08 March 2010
Participant Consent Form: Stage 6 - Family Member	2	08 March 2010
Participant Consent Form: Stage 6 - Patient	2	08 March 2010
Participant Consent Form: Stage 2,4,5,6 - Patient child	2	08 March 2010
WHOQOL		
Letter from Velindre Risk Review Committee		31 March 2010
Response to Request for Further Information		17 May 2010

After ethical review

Now that you have completed the application process please visit the National Research Ethics Service website > After Review

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

10/WSE03/12	**Please quote this number on all correspondence**

Yours sincerely

Mrs J Jenkins
Chair, Panel C
South East Wales Research Ethics Committees

Appendix B

GIG CYMRU NHS WALES
Bwrdd Iechyd Prifysgol Caerdydd a'r Fro
Cardiff and Vale University Health Board

Ysbyty Athrofaol Cymru
University Hospital of Wales

Heath Park,
Cardiff, CF14 4XW
Phone 029 2074 7747
Fax 029 2074 3838
Minicom 029 2074 3632

Parc Y Mynydd Bychan,
Caerdydd, CF14 4XW
Ffôn 029 2074 7747
Ffacs 029 2074 3838
Minicom 029 2074 3632

Eich cyf/Your ref
Ein cyf/Our ref
Welsh Health Telephone Network 1872
Direct line/Llinell uniongyrchol

Tel: 029 20746986
Fax: 029 20745311
CAV_Research.Development@wales.nhs.uk

From: Professor JI Bisson
R&D Director
R&D Office, 2nd Floor TB2
University Hospital of Wales
Cardiff
CF14 4XW

05 July 2010

Dr Richard Moore
Consultant Nephrologist
University Hospital of Wales
Heath Park
Cardiff
CF14 4XW

Dear Dr Moore

Project ID : 10/CMC/4769 : The Conceptualisation, Development, Validation and Practical Application of a Generic Family Quality of Life Measure.

Thank you for your recent correspondence addressing the points raised about the above project and supplying the following revised documents:

Document	Version/Serial number	Date
Letter responding to CaRRs comments		15-6-10

Your response and revised documents were reviewed on 05/07/10 by the Cardiff and Vale Research Review Service (CaRRS).

I am pleased to inform you that the Panel now has no objection to your proposal. You have informed us that Cardiff University has agreed to act as Sponsor under the Research Governance Framework for Health and Social Care.

I understand that you have already obtained:

- Favourable opinion from the NHS Research Ethics Committee

Bwrdd Iechyd Prifysgol Caerdydd a'r Fro yw enw gweithredol Bwrdd Iechyd Lleol Prifysgol Caerdydd a'r Fro
Cardiff and Vale University Health Board is the operational name of Cardiff and Vale University Local Health Board

POSITIVE ABOUT DISABLED PEOPLE
UHW129X

- Honorary research contracts/letters of access for members of the research team where required

Cardiff & Vale UHB is therefore happy for the project to begin.

May I take this opportunity to wish you success with the project, and to remind you that as Principal Investigator you are required to:

- Ensure that all members of the research team undertake the project in accordance with ICH-GCP and adhere to the protocol as approved by the Research Ethics Committee
- Inform the R&D office if any external or additional funding is awarded for this project in the future
- Inform the R&D office of any amendments relating to the protocol, including personnel changes and amendments to the actual or anticipated start and end dates
- Complete any documentation sent to you by the R&D office or University Research and Commercial Division regarding this project
- Ensure that adverse event reporting is in accordance with the UHB adopted Cardiff and Vale NHS Trust Policy and Procedure for Reporting Research-Related Adverse Events (refs 164 & 174) and Incident Reporting and Investigation (ref 108)
- Ensure that the research complies with the Data Protection Act 1998
- Ensure that arrangements for continued storage or use of human tissue samples at the end of the approved research project comply with the Human Tissue Act 2004 (for further information please contact Sharon Orton, HTA Coordinator OrtonS@cf.ac.uk).

If you require any further information or assistance, please do not hesitate to contact staff in the R&D Office.

Yours sincerely,

Professor Jonathan I Bisson
Chair of the Cardiff and Vale Research Review Service (CaRRS)

CC Chris Shaw, Research and Commercial Division, Cardiff University
Miss Catherine Golics

Appendix C

Correspondence to: Mrs Sarah Townsend, Research and Development Manager, Velindre NHS Trust, 3rd Floor, 14 Cathedral Road, Cardiff, CF11 9LH
Email: Sarah.Townsend@wales.nhs.uk
Tel: 02920 196529/Fax: 029 20344695

Professor Sam Salek
Welsh School of Pharmacy
Cardiff University
Redwood Building
King Edward VII Avenue
Heath Park
Cardiff
CF10 3XF

13th July 2010

Dear Professor Salek

2010/VCC/0012: The Conceptualisation, Development, Validation and Practical Application of a generic Family Quality of Life Measure

Thank you for your letter dated 18th May 2010, in which you responded to the issues raised by the reviewer on the 25th March 2010. Your response was forwarded to the original reviewer as per Trust procedures, who have confirmed the responses as satisfactory.

I am therefore pleased to take Chair's action to approve this project on behalf of the Research Risk Review Committee (RRRC) .This decision will be reported for ratification at the next RRRC on 26th August 2010.

Approval lapses if the project does not commence within 12 months of Trust approval. The Committee reserve the right to information on the progress of the project at any time and should receive a progress report six monthly and a written report on completion. On completion of the project please inform the R&D office.

All correspondence relating to this project should be forwarded to Sarah Townsend, R&D Manager, R&D Office, 3rd floor, 14 Cathedral Road, Cardiff CF11 9LJ ext 6529.

Yours sincerely

Sarah Townsend

Mrs. Diane Smith
Executive Director of Nursing and Quality and RRRC Chair

Appendix D

University of Glamorgan
Prifysgol Morgannwg
Faculty of Health, Sport and Science
Cyfadran Iechyd, Chwaraeon a Gwyddoniaeth

Ms.Catherine Golics
PhD Student,
Welsh School of Pharmacy
Redwood Building
King Edward VII Avenue
Cardiff CF10 3NB

Direct phone: 01443 483827
Fax: 01443 483831
Email: dcohen@glam.ac.uk

30 March 2010

Dear Catherine,

Conceptualisation, Development and Validation of a Generic Family Quality of Life Measure

Thank you for sending me your protocol for review.

The proposal addresses an important issue and is timely. It is increasingly recognised that illness can affect family as well as patients and development of a valid and reliable measure of family quality of life is overdue.

I found the proposal to be methodologically sound. You clearly understand the process needed to develop the measure and I have little doubt that if you get the co-operation from all whose input is needed that you will end up with a useful measure.

I also don't see the proposal as raising any particular ethical concerns. The standard approaches regarding obtaining consent, giving people time to think, maintaining anonymity, storage of data etc. all seem to be in place. Your methods of gaining information from patients and their families do not impose much burden.

On a very minor note

1) You are inconsistent in your spelling of specialty/speciality. Specialty is preferred but whichever you use, you should be consistent.

2) Your use of the word 'disease' in the introductory sections is fine, but I was uncomfortable with its use in relation to the study given the range of specialties covered. Might you consider something like 'condition' instead? Relatives of many patients in wound healing, orthopaedics, post-stroke, etc, may be put off by questions like "tell me about any ways your life has been affected by your family member's disease".

Yours sincerely

David Cohen
Professor

Professor/ Yr Athro Donna M Mead
Dean of Faculty/Deon y Cyfadran
University of Glamorgan/Prifysgol Morgannwg, Pontypridd, CF37 1DL, UK/DU
Tel/Ffôn 01443 483094 Fax/Ffacs 01443 483118
www.glam.ac.uk
Professor/Yr Athro David Halton Vice-Chancellor/Is-Ganghellor

Appendix E

Abdominal wound	Duplex kidney system	Neuropathic pain
Acne	Dysphagia	Non-hodgkin's lymphoma
Addisons disease	Eczema	Obesity
Adenomyeloneuropathy	Epilepsy	Ostearthritis
ADHD	Fibromyalgia	Osteonecrosis of the gums
Agenesis of corpus couosum	Folliculitis of the vulva	Osteoporosis
Alzheimers	Gallstones	Pancreatic transplant
Anaemia	Glaucoma	Pancreatitis
Angina	Global Development Delay	Paralysis of vocal chords
Aortic dilatation	GORD	Pituitary adenoma
Aortic Stenosis	Gout	Pneumonia
Asthma	Haemophilia	Polymyalgia rheumatica
Atherosclerosis	Haemophilic arthropathy	Primary biliary cirrhosis
Atopic dermatitis	Hayfever	Prostate cancer
Atrial fibrillation	Hearing loss	Pseudophakia
Auto-immune hepatitis	Heart bypass	Psoriasis
Benign tremor	Heart failure	Pulmonary embolism
Bi-polar disorder	Hepatitis C	Raynaud's
Bladder cancer	Hernia	Renal cancer
Bowel cancer	HIV	Retinal detachment
Brain tumour	Hypercholesterolemia	Rheumatoid arthritis
Breast cancer	Hypertension	Rosacea
Broken jaw	Hyperthyroidism	Sarcoidosis
Bronchiectasis	Hypothyroidism	Schizo-affective disorder
Bronchopulmonary aspergillosis	Idiopathic pulmonary fibrosis	Schizophrenia
Cancer of pharynx	Incontinence	Sciatica
Cardiomegaly	Irritable bowel syndrome	Sleep apnea
Cataracts	Ischaemic heart disease	Small bowel cancer
Cerebral palsy	Ischaemic nephropathy	Spinal surgery
Charcot-marie-tooth syndrome	Knee replacement	Splenic lymphoma
Chondromalacia patellae	Large granular lymphocyte leukaemia	Stomach ulcer
Chronic back pain	Learning difficulties	Stroke
Chronic hyperventilation	Leber optic atrophy	Talipes
Chronic kidney diease	Left ventricular failure	Talonavicular arthritis
Chronic pain	Leg ulcer	Thyroidtoxicosis
Chronic UTI	Leukaemia	Total
Coeliac disease	Lichen planus	Trigeminal nerve damage
Colitis	Lichen sclerosis	Turners syndrome
Conjunctivitis	Lupus	Upper GI bleed
COPD	Lymphodema	Urinary retention
Crohns disease	Lymphoma	Uterine cancer
Curvature of the spine	Macular degeneration	Vascular disease
Dementia	Microcephaly	Vertigo
Depression	Missing	Visual inattention
Diabetes	Motor neurone disease	Vulval intra-epithelial neoplasia
Diabetes type 1	Multiple sclerosis	Vulvodynia
Diabetes type 2	Muscular dystrophy	Wart on gum
Diabetic retionopathy	Myeloma	Wolff–Parkinson–White syndrome
Digeorge syndrome	Neuromyelitis optica	

Patient medical conditions, as reported by patients and confirmed by the content of their medical notes for the qualitative stage of the study.

Appendix F

Stage 2 Family Member

Information Leaflet
Version 3- 17/05/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

We are investigating the impact of illness on the quality of life of the patient's family.

Invitation

You are being invited to take part in this study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve.
Your relative has already agreed to take part in the study, and has given permission for you to be approached too.
Please take time to read the information carefully, and decide if you want to take part.
Please ask if you have any questions about the study, or if there is anything you find unclear.

What is the purpose of the study?

It is well known that any illness and medical condition can have a big impact on a patients' quality of life, in terms of physical discomfort, psychological distress and social problems.
Several studies have also looked at the effect of particular conditions on the patient's family members, and how their lives have changed as a result of living with an ill relative.
We intend to gather information on the quality of life of the family members of patients with a wide range of illnesses.
This information will then be used to develop a questionnaire to measure family quality of life.
Eventually, it is hoped that this information will lead to increased patient care, with the views and needs of the patient's family being further understood and taken into account when making treatment decisions.

Why have I been chosen?

You have a relative who has been diagnosed with a medical condition.
This means you can take part. There will be about 600 other people taking part in the study too.

Do I have to take part?

No, it is up to you whether you take part.
If you do decide to take part you will be asked to sign a consent form after reading this leaflet.
If you to decide to take part you can still withdraw at any time without giving a reason.
A decision to take part or not to take part or to withdraw will not affect the standard of care your relative will receive.

What will happen to me if I decide to take part?

The study will involve an informal interview/discussion- you will be invited to talk about the ways you think your life has been affected by your relative's medical condition.

You may decide to take part in the interview now. You can also choose to have the interview at your home, or come back to the hospital at a more convenient time. If you decide to come back to the hospital, your travel expenses will be reimbursed.

This interview will be audio-recorded, but only the study team will have access to these tapes.
There are no tests .You will not need to take any medication.

How long will it take?

It will take on average 30 minutes to complete the interview.

What are the benefits of taking part?

We cannot promise the study will help you but the information we get from this study may help improve the treatment of people, and support the families of people with the same condition that your relative has.

What are the possible risks of taking part?

We may ask you to talk about effects of your relatives condition which could make you feel emotional. If this does happen, you have the option to stop the interview at any point, without having to give a reason. We can also support you if this happens, by discussing the problem with your relative's care team if you both agree to this, or by directing you to support services or information.

What happens when the research stops?

The results will be used as part of a PhD thesis and will be published in a scientific journal. You will not be identified in any report or publication. You will be provided with a copy of this publication if you are interested.

What if there is a problem?
If there is a problem during the interview, you can ask for the interview to be stopped at any time. You will be asked whether you would like the information you have given us to be destroyed.
Any complaint about the way you have been dealt with during the will be addressed. Contact numbers for the study team are given at the end of this leaflet.

Will my taking part be kept confidential?

All the information which is collected from you will be kept strictly confidential. Each person involved is given a code number for confidentiality. Only the investigators will have access to your details that link with the code number. Your personal data will be kept for up to a year after the study has ended, The results of the study will not reveal your name or address.

Examples of how the information you give will be described in publications include:

> "A 30 year old female whose husband has been suffering from psoriasis for 10 years described feeling frustrated by his condition".
>
> "One male felt he could not continue with his job as a builder as a result of his father's heart failure"

Sometimes we may use direct quotes, for example:

> A 56 year old female described the effect of her sister's depression on her own social life: " I never go out any more, I worry about her too much".

Who has reviewed the project?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by South East Wales Research Ethics Committee.

Who is funding the project?

The project is being funded by Cardiff University.

What should I do if I have any complaints about the conduct?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions [029 20876017]. If you remain unhappy and wish to complain formally, you can do this using the NHS Complaints Procedure. Details can be obtained from Cardiff and Vale University Health Board website [http://www.wales.nhs.uk/sitesplus/864/page/40894].

If you have any complaint regarding any aspect, you can contact any of the following:

1. Prof S. Salek	Tel: 029 20876017
2. Cardiff and Vale University Health Board	Tel: 029 20743742
3. Cardiff University (sponsor)	Tel: 029 20879130

Contact for further information

If you have further questions, please feel free to contact one of the study team:

1. Prof. A.Y Finlay	Tel: 029 20744721
2. Catherine Golics (investigator)	Tel: 029 20 8760 17
3. Dr M.K.A Basra	Tel: 029 2074 5874
4. Prof. S. Salek	Tel: 029 20 8760 17

Thank you for taking time to read this and for your help.

Project Protocol Version 10- 17/05/10

Appendix G

Stage 2,4,5. Family member

Consent form

Version 2- 08/03/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

Code No:

	Please initial
I confirm that I have read the information sheet (**Version 3**) and understand the intent of the study. I also have had the opportunity to ask questions, and had these answered satisfactorily.	☐
I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason and without my relatives medical care being affected.	☐
I hereby, give written consent to participate in the study that involves me taking part in an interview/ completing a questionnaire about the effects of my relative's medical condition on my life quality. I understand that interviews will be audio-recorded.	☐
I agree to take part in the above study	☐

Name of participant .....

Date .....

Signature .....

Name of investigator .....

Date .....

Signature .....

Appendix H

Stage 2 Patient

Information Leaflet
Version 3 – 17/05/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

We are investigating the impact of illness on the quality of life of the patient's family.

Invitation

You are being invited to take part in this study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the information carefully, and decide if you want to take part. Please ask if you have any questions about the study, or if there is anything you find unclear.

What is the purpose of the study?

It is well known that any illness and medical conditions can have a big impact on a patients' quality of life, in terms of physical discomfort, psychological distress and social problems. Several studies have also looked at the effect of particular conditions on the patient's family members, and how their lives have changed as a result of living with an ill relative.
We intend to gather information on the quality of life of the family members of patients with a wide range of illnesses.
This information will then be used to develop a questionnaire to measure family quality of life. Eventually, it is hoped that this information will lead to increased patient care, with the views and needs of the patient's family being further understood and taken into account when making treatment decisions.

Why have I been chosen?

You have been diagnosed with a medical condition, and are attending a clinic with a member of your family, or a member of your family is visiting you in hospital.
This means you can take part.
There will be about 600 other people taking part in the study too.

Do I have to take part?

No, it is up to you whether you take part.
If you do decide to take part you will be asked to sign a consent form after reading this leaflet.
If you to decide to take part you can still withdraw at any time without giving a reason.
A decision not to take part or not to take part or to withdraw will not affect the standard of care you will receive.

What will happen to me if I decide to take part?

The study will involve an informal interview/discussion with your relative - they will be invited to talk about the ways their life has been affected by your medical condition.

This interview will be audio-recorded, but only the study team will have access to these tapes.

You will not need to take part in the interview.

There are no tests (e.g. blood tests, x-rays). You will not need to take any extra medication.

How long will it take?

It will take on average 30 minutes to complete the interview with your relative.

What are the benefits of taking part?

We cannot promise the study will help you but the information we get from the study may help improve the treatment of people, and support the families of people with the same condition that you have.

What are the possible risks of taking part?

We may ask your relative to talk about effects of your condition which could make them feel emotional. If this does happen, they have the option to stop the interview at any point, without having to give a reason. We can also support them if this happens, by discussing the problem with your care team if you both agree to this, or by directing them to support services or information.

What happens when the research stops?

The results will be used as part of a PhD thesis and will be published in a scientific journal. You will not be identified in any report or publication. You will be provided with a copy of this publication if you are interested.

What if there is a problem?

If there is a problem during the interview, your relative can ask for the interview to be stopped at any time. You will be asked whether you would like the information they have given us to be destroyed.

Any complaint about the way you have been dealt with during the will be addressed. Contact numbers for the study team are given at the end of this leaflet.

Will my taking part be kept confidential?

All the information which is collected from you will be kept strictly confidential. Each person involved is given a code number for confidentiality. Only the investigators will have access to your details that link with the code number. Your personal data will be kept for up to a year after the study has ended, The results of the study will not reveal your name or address.

Examples of how the information your relatives give will be described in publications include:

> "A 30 year old female whose husband has been suffering from psoriasis for 10 years described feeling frustrated by his condition".
>
> "One male felt he could not continue with his job as a builder as a result of his father's heart failure"

Sometimes we may use direct quotes, for example:

A 56 year old female described the effect of her sister's depression on her own social life: " I never go out any more, I worry about her too much".

Who has reviewed the project?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by South East Wales Research Ethics Committee.

Who is funding the project?

The project is being funded by Cardiff University.

What should I do if I have any complaints about the conduct?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions [029 20876017]. If you remain unhappy and wish to complain formally, you can do this using the NHS Complaints procedure. Details can be obtained from Cardiff and Vale University Health Board website [http://www.wales.nhs.uk/sitesplus/864/page/40894].

If you have any complaint regarding any aspect, you can contact any of the following:

1. Prof. S. Salek	Tel: 029 20876017
2. Cardiff and Vale University Health Board	Tel: 029 20743742
3. Cardiff University (sponsor)	Tel: 029 20879130

Contact for further information

If you have further questions, please feel free to contact one of the study team:

1. Prof. A.Y Finlay	Tel: 029 20744721
2. Catherine Golics (investigator)	Tel: 029 20 8760 17
3. Dr M.K.A Basra	Tel: 029 2074 5874
4. Prof. S. Salek	Tel: 029 20 8760 17

Thank you for taking time to read this and for your help.

Project Protocol Version 10- 17/05/10

Appendix I

Stage 2,4,5. Patient

Consent form

Version 2 – 08/03/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

Code No:

	Please initial
I confirm that I have read the information sheet (**Version 3)**) and understand the intent of the study. I also have had the opportunity to ask questions, and had these answered satisfactorily.	☐
I hereby give written consent that I have no objection if any of my family members wants to participate in the study. This involves the participant taking part in an interview or completing a questionnaire on the effects that my medical condition has on the life quality of my family member.	☐
I understand that sections of my medical notes may be looked at by study investigators where it is relevant to my family taking part in the research. I give permission for these individuals to have access to my medical records.	☐
I agree to take part in the above study	☐

Name of patient .....

Date .....

Signature .....

Name of investigator .....

Date .....

Signature .....

Appendix J

Family member & Patient Demographic Data Sheet

Version 1- 26/08/09

Study Title: **The conceptualisation, development, validation and practical application of a Generic Family Quality of Life measure.**

Family member Information

Code number	
Initials	
Age	
Gender	
Ethnic Origin	White / Mixed / Asian or Asian British / Black or Black British / Chinese / Other …….…….……..
Relation to patient	
Current Occupation	

Patient information

Name/ initials and code number	
Hospital number	
Age	
Gender	
Ethnic Origin	White / Mixed / Asian or Asian British / Black or Black British / Chinese / Other…….…….……..…….……..
Diagnosis	
Other diagnoses	
Disease duration	
Current Occupation	
Socioeconomic Code	

Appendix K

01/12/11

Dear .....

Thank you for your interest in being a part of our questionnaire feedback panel.

The information from the interview which I carried out with you last year, and the other 132 interviews I also carried out, has been used to design a questionnaire for family members of patients. **The questionnaire will be used to assess the impact of illness on families of patients**.

We are interested in getting feedback about the questionnaire. I would therefore like to invite you to be a part of a feedback panel which will involve an hour long informal discussion with myself, my project team and some other family members of patients.

Your attendance at this meeting will be greatly valued. It may not benefit you directly, but it is likely that it will help other family members in the future.

The details for the meeting are as follows:

Wednesday 14th December 2011
2pm-3pm
Library, 3rd floor, Glamorgan House, Heath Park, University Hospital of Wales, Cardiff, CF14 4XW.

Please let me know if you are able to attend by telephoning me on 07791 727553 or email golicscj@cardiff.ac.uk as soon as possible.

In the mean time, I enclose a copy of the questionnaire and a feedback form for you to complete and bring to the meeting.

If you have any problems or questions please do not hesitate to contact me.

Regards,

Catherine Golics
PhD Student, Cardiff University

Project supervisors: Professor Andrew Finlay, Professor Sam Salek and Dr Mohammad Basra

Appendix L

Family Quality of Life research meeting. Wed 14th Dec,12.30, Glamorgan House, UHW.

Dear Dr.....

I am emailing in relation to my PhD project, The Family Quality of Life Study.

You kindly allowed me to attend your clinics/visit patients on your ward/visit your patient's homes last year to carry out interviews with family members of patients.

The content from these interviews has now been used to form a questionnaire to measure the impact of disease on family members of patients.

A critical part of validation of the questionnaire is gaining your views on the content and I am emailing to invite you to attend a lunchtime discussion group to assess the content validity of the questionnaire.

The details for the discussion group are as follows:

Wednesday 14th December 2011
Dermatology Board Room, 3rd Floor, Glamorgan House (next to Medicentre), UHW.
12.30pm-1.30pm
Lunch will be provided

I am also keen to invite specialist nurses in order to gain their views. I would be grateful if you could provide me with the contact details for a nurse in your team who may be interested.

On your reply to this email I will email you a copy of the questionnaire and a brief pre-meeting feedback form to complete. I will also post a copy of these documents to you next week. Please bring these to the meeting.

If you are unable to attend the meeting it is still critical for this part of the study that we receive your feedback. Please complete the feedback form and send it back to me via email or internal mail (Room 2.51A, Redwood Building, Cathays Park) before Tuesday 13th December.

The meeting will only last one hour, lunch is provided and your attendance will be greatly valued.

Regards,

Miss Catherine Golics
Dr Mohammad Basra
Professor Andrew Finlay
Professor Sam Salek

Appendix M

Questionnaire feedback form- content validity

Name……

Thank you for agreeing to take part in the questionnaire feedback as part of the content validity. Each item on the questionnaire needs to be assessed for language clarity, completeness, relevance and scaling. The following definitions are provided to ensure standardisation and so that each person has the same understanding of these criteria.
Please rate each of the questionnaire items on the following:

A. **Language clarity**: the sentences and wording should be clear, understandable, straightforward and simple. Phrases and wording should be unambiguous and jargon free and should be understood by someone with a reading ability of 12 years.

B. **Completeness:** the sentences should be complete, not broken and should end appropriately.

C. **Relevance:** each item should be relevant to the subject area and target population.

D. **Scaling:** the scoring system is a 5 point adjectival scale. Panel members should rate the scaling system as to whether the response options fit the question, or not.

PLEASE NOTE- YOU DO NOT NEED TO COMPLETE THE QUESTIONNAIRE YOURSELF.

Item 1: I feel worried					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 2: I feel angry					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 3: I feel guilty					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 4: I feel sad					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 5: I feel frustrated					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 6: I feel tired					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 7: My behaviour or personality is affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 8: I feel I have no one to talk to about my thoughts					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 9: I feel a burden of caring for my family member					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 10: My housework has increased					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 11: My eating habits are changed					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 12: My family activities are affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 13: My leisure activities are affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 14: My hobbies are affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 15: It is hard to find time for myself					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 16: I need to stay at home					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 17: My every day travel is difficult					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 18: My time is taken up visiting my family member in hospital or attending medical appointments					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 19: My sex life is affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 20: I argue with my family member					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 21: My family expenses have increased					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 22: I experience problems with holidays					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 23: I find it hard to plan my time and activities					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 24: My own health or well-being is affected because of my family member's condition					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 25: My sleep is affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 26: My social life is affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 27: I worry about strangers' reactions to my family member's condition					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 28: I find it difficult to talk about my family member's condition					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 29: My work or study is affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Item 30: My relationships with other family members are affected					
	Strongly agree	**Agree**	**Disagree**	**Strongly disagree**	**Any comments:**
Language clarity	☐	☐	☐	☐	
Completeness	☐	☐	☐	☐	
Relevance	☐	☐	☐	☐	
Scaling	☐	☐	☐	☐	

Appendix N

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
General comments on layout	The questionnaire layout is very simple to use	The questionnaire is too busy for elderly people. Could space out the questions more to reduce chance or error.	Language clarity was very good
The use of grey shading in the questionnaire	I wonder if the coloured lines for every other question would make the individual feel that these questions are of some significance. Whether it would be better kept plain?	Grey shading is useful as it directs you to the line.	Would prefer to not have grey shading
		It doesn't make some items appear more important than others	Grey shading is good as splits up the questions
		Does it look better shaded or with dotted lines? Could shade every line as it's then easy to follow. I agree that we need to do something to guide people to the right response.	I think that having dotted lines wouldn't encourage people to write on them- would be a good idea
		Could you shade the items which are most important? That would get people to mark the items most important.	
		There could be potential problem with grey colouring when photocopying/printing.	
The font type and size		Font size is fine	Font size is fine
General comments about items	Generally the question wording is relevant and straight forward	You could use examples in the items	Use of examples? I think it would be useful to remind you of things like insurance and things you don't think of.
		Don't use "is affected", say if its negative or positive	Examples-I don't think it limits you as it's just examples
Item 1	Should be changed to "I am worried"	I am worried that the respondent will tick the same response option for all of the emotion questions	Clear and relevant
Item 2	Angry at what?		
	Should change to "I am"		
Item 3	Guilty about what?		
	Guilty about what? Is this disease specific?		

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
Item 4	No comments		
Item 5	Change to "I am"- it's clearer		
Item 6	In what way?	Does it matter if emotionally or physically tired?	
	Emotionally or physically? Or does it not matter?		
	Tired= exhaustion or tired= emotional/fed up? Does it matter?		
	Change to "I am"		
Item 7	Maybe ask two questions?		What if both are affected? They are two different things
	Behaviour and personality are possibly two different things? "extremely" does not seem to apply.		Two different things- personality would be the way I am thinking about things and behaviour would be the way I am behaving with other people.
	These are two statements. Personality is very abstract- stick to behaviour.		I think one is the way you think and one is the way I communicate with others- they are different
	Could be changed to "has been changed"		Should be split into two questions
	Affected negatively or positively? Often out of adversity emerges positive behaviour		
Item 8	Delete "I feel"	Not sure about response options, especially "extremely"	Is clear
	Scaling perhaps needs to be related to timings/frequency?	Response categories aren't appropriate for question	The response scale doesn't quite sound right - would be hard to work out which word fits.
			It's more of a yes or no question, not something you can quantify
			There are different people you can talk to about different things

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
Item 9	Needs to be rephrased, doesn't read right	Burden is a bad word	I don't like burden – it's too emotive. You could use the word "responsibility"
	Not terribly elegantly stated!	Not clear if the burden is the patient or family mental?	Burden makes you feel like you're blaming the patient. I don't regard it as a burden, I don't like the word
	? Grammar?	This is a good question	Burden could be relevant to some people
	Do not like the use of the word "burden"	Will people know what this means? Who is the burden?	Responsibility and commitment are suggestions of alternative words to use
	The term "burden" is a problem	Could say "it is a burden caring for my family member"	How about saying it's time consuming?
	I don't understand this statement. Does the patient's relative feel that they are a burden or that they are burdened by the patient?	Or "I feel burdened"	
	Say "caring for my family member is a burden to me"	Or "I feel caring for my family member is a burden"	
	Could the question be worded "I feel caring for my family member is a burden"	I understood what you were trying to get from that but I didn't like the way it was worded.	
	Re scaling: "extremely" doesn't fit. Is "tremendously" or "immensely" more appropriate or too emotive?	I think if the patient read this, they would feel from looking at the questions that they are a burden. I think they already feel a burden and seeing these questions in the extreme then it would feel a burden and that's what worries me about this. I don't want the patient to feel like this.	
		This question is a true representation of how people feel and its good that someone is thinking about them.	

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
		Patients are very aware of this issue and they know about the burden they place on the carer.	
Item 10	May not be relevant to all		It's straight forward
	"Extremely" doesn't fit		
Item 11	Less time or no appetite?	Should be eating habits have changed.	Yes, relevant
	Either "are" or "have" changed?	Is this negative or positive?	
	?"diets"		
	Eating habits HAVE changed.		
	Again, "extremely" doesn't fit		
Item 12	Positive or negative?	Is this negative or positive?	The term "family activities" is clear.
	Activities with my family are affected	How about "my family activities are adversely affected"?	Family activities is good as affects other children too
	Wording unclear. Need to define better	Does my family activities mean the same as my activities with my family?	I think it means you can't do things as a family, maybe because the one member is ill so cant join you.
	? the activities of the family as a whole or the relative's activity within the family?	Language clarity is poor (explained the origins of question), wording needs changing	I think it means you can't get involved in family activities
	Not sure of the differences/similarities between questions 12, 13 and 14	Needs the word "shared" i.e. "shared family activities".	I think it could mean if you're tired then you can't do things for as long a period of time as you'd like
		I think it's clear and can't see any other way it could be interpreted.	I think it's similar to social activities
		Needs clarity	
Item 13	Positive or negative?		I think 12 and 13 could be linked maybe
	What is the difference between leisure and hobby?		
Item 14	Positive or negative?	Big overlap of 13 and 14.	13 and 14 could be linked but leisure activities could be more social and hobbies are by yourself

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
	Items 13 and 14 overlap a bit, I'm not sure how relevant 14 is?	Depends on interpretation of hobbies and leisure activities	I think of leisure activities as something outside the house
	Overlap with 13	13 and 14 are repetitive	Is watching TV a leisure activity? Yes, I think it is
	13 and 14 very similar- drop one		Leisure and hobbies should be as 2 separate questions
	Do we need this as well as 13? Combine 13 and 14.		
	What is the difference between leisure and hobby?		
Item 15	It is relevant but it's difficult to quantify	Overlaps with 13 and 14 a bit	Clear and fits with response categories
Item 16	Why? Need to care?	Could say "I am forced to stay at home"	It's relevant but depends on people's situation and the illness
	The word "extremely" does not really fit	Or "I feel the need to stay at home"	I don't think "extremely" fits well here. "Very much" would be better.
	I am forced to stay at home?	Couldn't quite understand this item	
	Could change to "I feel the need to...". Not sure this question is relevant at all, what are you trying to answer with this question?	Can you say "I can't go out"?	
	How about "I can't go out"	Some people feel they can't leave and other people literally can't leave so I think to me there are 2 different issues.	
	Include a "duration" reference. Maybe "I need to stay at home more" or "I need to spend more time at home"	I feel the need to and I have to are two different questions	
	Would "I feel the need to stay at home" be more appropriate?		
Item 17	Why?	If they can go out and do something then their travel is probably not affected. Do you mean if you take the patient.....?	More relevant to parents of children I think.
	I struggle to understand	Could put examples in	Every day travel

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
	what you are getting at here? Mode of transport, duration, freedom to travel or destinations?	there to make clearer	means going out somewhere.
	Not sure what you are trying to say with this question? Is it relevant?	Not too relevant to some specialties	I think this will be more relevant depending on the age of the relative and patient
	This question may not obtain the info needed. I.e. if the person completing the questionnaire is still going to work they might do this without any impact. However, if this question is "my usual travel with my family member is difficult" a different, more relevant answer might be obtained	Is this with or without the relative?	
Item 18	This could be two questions		Fits response categories
	These are two different points and if the first is true then this will provide a different impact		Could be two questions
Item 19	Positive or negative?	Should not be phrased as "intimate relationships" as "sex life" is clearer and they actually mean different things - is it physical? Yes.	Wording is obvious but shouldn't use more subtle term
	A very important but often avoided subject		"intimate relationships" is different to sex life.
			Wouldn't be offended by this
Item 20	Argue more?	Does this mean the patient? Should this be clarified or does it mean they argue with everyone all the time as they are so worried about what is going on around them?	How about "I argue with family members" because if you're under stress you take it out on whoever is nearest
	Might be worth clarifying the "affected" and not other family members	Whole family or patient? The way it is worded implies one person	I think it depends on whether you're trying to find out the reaction to the person who's ill which is different from the general family
	Do you need to clarify if it's the ill family member?	Is this in comparison with before? Have they always argued	I think there should be a question to cover the fact that

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
		with their family member?	you also argue with other family members, maybe they don't appreciate it. This could overlap with question 30.
	With the disabled family member? Or all?	As a result of....?	
	Is this the family member with the illness?		
	Perhaps "more than usual" should be added to Q. Many family members argue anyway!		
Item 21	Not relevant to me What are family expenses?	It sounds good	I don't count things up like that
			I wouldn't think to include things like hospital parking when answering this question
			I think insurance would come under this question too
Item 22	Why?	Is this physically going on holiday, school holidays, Christmas? With the affected person?	Yes, very relevant
	You almost want a "because of" at the end of the sentence	It's not a complete question for me. At the end there should be a because....	I think of travel and things whilst on holiday
	I have problems planning a holiday	You might find that people will put "not relevant" when they mean they can't go anymore, but in a sense that is "extremely". We find that people do tick "not relevant". It comes down to individual interpretation.	I think of insurance as well
	Vague? Financial implications/lack of disabled facilities/carrying extra luggage/meds/insurance costs/decreased mobility of patient etc etc.	That's down to how you introduce the questionnaire. You need to know if they can remember when their family member was well. Maybe they didn't know that family member when they were well.	Potential problem with people ticking not at all/not relevant if they can't go at all (when they should be ticking extremely)
		With or without relative?	How about "problems with going on holiday" as holiday could

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
			just mean once you're there
Item 23	Too similar to other questions		I think it's relevant
	?repetitive		
	Say "it is hard to...."		
Item 24			I think it's relevant
Item 25		Is this negatively?	Links to item 24
Item 26	Too similar to other questions	Is this negatively?	Straight forward
	?repetitive	Maybe use "deteriorated" rather than "affected"	
Item 27	Is it different from question 1? (I feel worried). It's a negative reaction?	Is the issue worry or the reaction? (reaction). Not clear if it's a negative reaction.	Relevant.
	Don't like word "strangers". How about "I worry about how others will react to my family member's condition"	This is really applicable to a lot of specialties	
Item 28	Do you mean isolated?		Links to not having anyone to talk to
			Relevant
Item 29	Positive or negative?		Straightforward
Item 30	Any in particular?	Needs clarity	Related to question 20
The questionnaire instructions	Change opening statement to "The following statements relate to how your life is being affected at the moment by your relative's condition". At the moment it is confusing as has two separate tenses.	You're asking "has been affected" and then "at the moment", should it be "is affected"? We're talking about the present tense. Or "is being affected"	
	The opening statement could read .....relate to how your life is being affected by your family member's condition. (Do you need anything more i.e. at the moment?)	Could you put the name of the patient in the instructions e.g. "caring for X".	
	Use the word "mark" instead of "tick"	"At the moment" can be different from the last 2 or 3 weeks so is a time frame better than at the moment? If it's for a chronic condition then a time	

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		frame gives you more scope for capture but acute intervention might carry different meaning. Sometimes patients can misinterpret at the moment e.g. in pain clinics (before or after taking meds).	
		People's interpretation of any time frame will be different anyway so it doesn't matter which one you use. "At the moment" will probably give you the result you want.	
		Maybe you could put a time frame in brackets after "at the moment"	
		Putting a time frame e.g. a week would have a problem for some questions e.g. holidays, whereas present would be ok.	
		Use the word "mark" instead of "tick", as "tick" may be alien to some people. Could use "mark clearly".	
		Do you want people to reflect or answer straight away? Might need to give them some guidance about this.	
Use of the term "family member" or "relative"?	Participants may be put off by the use of the cold term "family member", who might be their much loved child, hence suggestion to change to "your relative's condition".	You say caring for your relative but I'm worried that in some cases they will have more than one.	Family member is best as not much difference
		Is it worth adding to the front page "is there more than one family member you are looking after" or another way of putting it.	I think family member is someone who is close to you now. Relative could be distant
		Could you put how many people they look after? E.g. a box for a	Could use close family member

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		number?	
		Either exclude people looking after more than one family member or include a space for them to specify how many people they look after.	
Use of the term "condition", "illness" or "disease"		Use condition	Not disease, condition is best
		Could have a problem with "condition" as there are interventions, operations etc.	
The language used in the items and potential translation problems	The language appears to be pitched at the right level	Should the questions all be negative? (yes)	The language appears to be pitched at the right level
		"I am" is easier to understand than "I feel" for translational purposes. Especially for the emotional questions. Possible idea.	
		Some cultures would think you are not entitled to these emotions as it's your duty.	
		Might be translational problems with "very much", "extremely" etc. In Indian only one word for these. Might be better with yes/no.	
The use of reminders throughout the questionnaire		I think reminders should go in	Just at the top of the page
		I think if you space out the questions and it goes onto the next page, so at the top of each page would be a reminder, not after each question as that might be confusing.	Don't need reminders on every question
The response categories	Extremely replaced by All the time	Restrictive. Maybe use only 3 categories - very much, moderately, not at all.	"Extremely" does not fit well. "Very much" would be better.
		Or, a lot, a little and not at all.	Order of response options is OK
		Could leave unlabelled boxes between the 3 response categories	

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		At the start of the questionnaire you should have a number scale, or show people the number (e.g. 1/10) that is equivalent to each response category. This will standardise it and have equal intervals. Or different scales for different items.	
		Or just give them the scale, numbers not any category labels, but reliability very low	
		Possible translation problems with response categories.	
		Extremely should be replaced	
		Time scale should be used	
		Would very much be better than extremely? Yes it would.	
		Order of categories- should be the other way around. Start negatively.	
		Makes sense to start with negatively- risk of getting it wrong	
		Generally don't like wording of scale throughout	
The use of the response category "not relevant"	Not sure that not relevant option is necessary as have not at all option.	Is there a difference between not relevant and not at all?	The use of not relevant is important to some questions
	I am not sure I or others will understand the difference between the categories of "not at all" and "not relevant".	Is "not relevant" a relevant category for the emotion questions? It could be given as a response option for some questions only.	
		"not relevant" should be removed completely (half the people agreed)	
		Could move not relevant to the end so it separates them.	

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
		Or put on separate line?	
The words "hobbies" and "leisure activities"	Items 13 and 14 overlap a bit, I'm not sure how relevant 14 is?		Items 13 and 14 overlap a bit, I'm not sure how relevant 14 is?
	13 overlaps with 14		13 overlaps with 14
	13 and 14 very similar- drop one		13 and 14 very similar - drop one
	Do we need this as well as 13? Combine 13 and 14.		Do we need this as well as 13? Combine 13 and 14.
The demographics and utility questions	Should the patient's age be included for ease of analysis i.e. to specifically break down data regarding family members of paediatrics/elderly patients etc?	I'm wondering why the two utility questions are separate on the front? (I explained it was just for demographic purposes).	Cant quantify "how many hours"- very difficult
	Regarding How many hours ….. doing housework. Some patients' carers might be extremely house proud and might not be able to do as much house work as they did previously because their relative is ill. Similarly, if a patient used to do the house-work and now their carer has taken over, (s)he will see an increase in the number of hours spent. Therefore is this information relevant. What is it telling you????	Is the patient's age relevant? It's different for different generations. Do you have the details of the patient anyway? Should it be included? (decided possibly no).	"How many hours" depends on the condition. Would be better to put "on average". The examples are not necessary, its a bit restrictive
		Housework is an important issue in some specialties, but is it relevant to all?	The examples on the "how many hours" is confusing. Think it suggests you should cross off the ones you don't do
		Losing sleep through worry and through getting up are two different things. Worry does not necessarily impact on life, whereas moving someone several times a night is totally different impact on your life than worry so I think it should be 2 questions, also "I feel worried" is also a question.	Impossible to answer.
General	Whilst I understand this	We are so focused on	

Discussion topic	Written feedback	Expert panel discussion	Family member panel discussion
comments about the project	is extremely valuable work, I am concerned that many patients already feel burdensome and this questionnaire might add to this feeling. As a patient, I would be interested to know what my carer is being asked to complete and how (s)he is responding. Relationships involving patients with chronic conditions, as you are no doubt aware, are under extreme stress. Secrecy surrounding the questionnaire would add to this. Openness on the other hand might exacerbate patients' feelings of guilt, worthlessness etc. Openness might also influence carers' responses if they are attempting to protect their family member's feelings. It's so very difficult to untangle caring ---- GOOD LUCK!	the patient and totally forget about the family members. It's not us being judgemental - these topics have all arisen from developmental work so they are very legitimate	

Appendix O

Stage 5 Family Member

Information Leaflet
Version 3-17/05/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

We are investigating the impact of illness on the quality of life of the patient's family.

Invitation
You are being invited to take part in this study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve.
Your relative has already agreed to take part in the study, and has given permission for you to be approached too.
Please take time to read the information carefully, and decide if you want to take part.
Please ask if you have any questions about the study, or if there is anything you find unclear.

What is the purpose of the study?

It is well known that any illness and medical conditions can have a big impact on a patients' quality of life, in terms of physical discomfort, psychological distress and social problems.
Several studies have also looked at the effect of particular conditions on the patient's family members, and how their lives have changed as a result of living with an ill relative.
We intend to gather information on the quality of life of the family members of patients with a wide range of illnesses.
This information will then be used to develop a questionnaire to measure family quality of life.
Eventually, it is hoped that this information will lead to increased patient care, with the views and needs of the patient's family being further understood and taken into account when making treatment decisions.

Why have I been chosen?

You have a relative who has been diagnosed with a medical condition.
This means you can take part. There will be about 600 other people taking part in the study too.

Do I have to take part?

No, it is up to you whether you take part.
If you do decide to take part you will be asked to sign a consent form after reading this leaflet.
If you to decide to take part you can still withdraw at any time without giving a reason.
A decision to take part or not to take part or to withdraw will not affect the standard of care your relative will receive.

What does it involve?

You will be asked to fill out a questionnaire about the ways your life has been affected by your relative's medical condition.

There are no tests. You will not need to take any medication.

How long will it take?

It will take on average 10 minutes to complete the questionnaire.

What are the benefits of taking part?

We cannot promise the study will help you but the information we get from the study may help improve the treatment of people, and support the families of people with the same condition that your relative has.

What are the possible risks of taking part?

This study involves questionnaires so there are no risks associated with it.

What happens when the research stops?

The results will be used as part of a PhD thesis and will be published in a scientific journal. You will not be identified in any report or publication. You will be provided with a copy of this publication if you are interested.

What if there is a problem?
Any complaint about the way you have been dealt with during the will be addressed. Contact numbers for the study team are given at the end of this leaflet.

Will my taking part be kept confidential?

All the information which is collected from you will be kept strictly confidential. Each person involved is given a code number for confidentiality. Only the investigators will have access to your details that link with the code number. The results of the study will not reveal your name or address.

Who has reviewed the project?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by South East Wales Research Ethics Committee.

Who is funding the project?

The project is being funded by Cardiff University.

What should I do if I have any complaints about the conduct?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions [029 20876017]. If you remain unhappy and wish to complain formally, you can do this using the NHS Complaints Procedure. Details can be obtained from Cardiff and Vale University Health Board website [http://www.wales.nhs.uk/sitesplus/864/page/40894].

If you have any complaint regarding any aspect, you can contact any of the following:

1. Prof. S. Salek	Tel: 029 20876017
2. Cardiff and Vale University Health Board	Tel: 029 20743742
3. Cardiff University (sponsor)	Tel: 029 20876017

Contact for further information

If you have further questions, please feel free to contact one of the study team:

1. Prof. A.Y Finlay	Tel: 029 20744721
2. Catherine Golics (investigator)	Tel: 029 20 8760 17
3. Dr M.K.A Basra	Tel: 029 2074 5874
4. Prof. S. Salek	Tel: 029 20 8760 17

Thank you for taking time to read this and for your help.

Project Protocol Version 10- 17/05/10

Appendix P

Stage 5 Patient

Information Leaflet
Version 3-17/05/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

We are investigating the impact of illness on the quality of life of the patient's family.

Invitation

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What is the purpose of the study?

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We intend to gather information on the quality of life of the family members of patients with a wide range of illnesses.
This information will then be used to develop a questionnaire to measure family quality of life. Eventually, it is hoped that this information will lead to increased patient care, with the views and needs of the patient's family being further understood and taken into account when making treatment decisions.

Why have I been chosen?

You have been diagnosed with a medical condition, and are attending a clinic with a member of your family, or a member of your family is visiting you in hospital.
This means you can take part. There will be about 600 other people taking part in the study too.

Do I have to take part?

No, it is up to you whether you take part.
If you do decide to take part you will be asked to sign a consent form after reading this leaflet.
If you to decide to take part you can still withdraw at any time without giving a reason.
A decision to take part or not to take part or to withdraw will not affect the standard of care you will receive.

What does it involve?

In this study, your relative will be asked to fill out a questionnaire about the ways their life has been affected by your medical condition.

You will not need to fill out a questionnaire.

There are no tests (e.g. blood tests, x-rays). You will not need to take any extra medication.

How long will it take?

It will take on average 10 minutes for your relative to complete the questionnaire.

What are the benefits of taking part?

We cannot promise the study will help you but the information we get from the study may help improve the treatment of people, and support the families of people with the same condition that you have.

What are the possible risks of taking part?

This study involves questionnaires so there are no risks associated with it.

What happens when the research stops?

The results will be used as part of a PhD thesis and will be published in a scientific journal. You will not be identified in any report or publication. You will be provided with a copy of this publication if you are interested.

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Project Protocol Version 10- 17/05/10

Appendix Q

Stage 6 Family Member

Information Leaflet
Version 3-17/05/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

We are investigating the impact of illness on the quality of life of the patient's family.

Invitation

You are being invited to take part in this study. Before you decide whether to take part, it is important for you to understand why the research is being done and what it will involve.
Your relative has already agreed to take part in the study, and has given permission for you to be approached too.
Please take time to read the information carefully, and decide if you want to take part.
Please ask if you have any questions about the study, or if there is anything you find unclear.

What is the purpose of the study?

It is well known that any illness and medical conditions can have a big impact on a patients' quality of life, in terms of physical discomfort, psychological distress and social problems.
Several studies have also looked at the effect of particular conditions on the patient's family members, and how their lives have changed as a result of living with an ill relative.
We intend to gather information on the quality of life of the family members of patients with a wide range of illnesses.
This information will then be used to develop a questionnaire to measure family quality of life.
Eventually, it is hoped that this information will lead to increased patient care, with the views and needs of the patient's family being further understood and taken into account when making treatment decisions.

Why have I been chosen?

You have a relative who has been diagnosed with a medical condition.
This means you can take part. There will be about 600 other people taking part in the study too.

Do I have to take part?

No, it is up to you whether you take part.
If you do decide to take part you will be asked to sign a consent form after reading this leaflet.
If you to decide to take part you can still withdraw at any time without giving a reason.
A decision to take part or not to take part or to withdraw will not affect the standard of care your relative will receive.

What does it involve?

In this study, you will be asked to fill out a questionnaire about the ways your life has been affected by your relative's medical condition.

Your relative will be asked to fill out a questionnaire about how their life has been affected by their medical condition. They will also be asked to rate their health on a scale.

We will ask if we can contact you and your relative by post, to send you a questionnaire after the study. This is optional. You can still take part in this part of the study if you do not want to be contacted by post.

There are no tests. You will not need to take any medication.

How long will it take?

It will take on average 10 minutes to complete the questionnaire.

What are the benefits of taking part?

We cannot promise the study will help you but the information we get from the study may help improve the treatment of people, and support the families of people with the same condition that your relative has.

What are the possible risks of taking part?

This study involves questionnaires so there are no risks associated with it.

What happens when the research stops?

The results will be used as part of a PhD thesis and will be published in a scientific journal. You will not be identified in any report or publication. You will be provided with a copy of this publication if you are interested.

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Who is funding the project?

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What should I do if I have any complaints about the conduct?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions [029 20876017]. If you remain unhappy and wish to complain formally, you can do this using the NHS Complaints

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Project Protocol Version 10- 17/05/10

Appendix R

Stage 6 Patient

Information Leaflet
Version 3 – 17/05/10

Study Title: The conceptualisation, development and validation of a generic Family Quality of Life measure.

We are investigating the impact of illness on the quality of life of the patient's family.

Invitation

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What is the purpose of the study?

It is well known that any illness and medical conditions can have a big impact on a patients' quality of life, in terms of physical discomfort, psychological distress and social problems. Several studies have also looked at the effect of particular conditions on the patient's family members, and how their lives have changed as a result of living with an ill relative.
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Why have I been chosen?

You have been diagnosed with a medical condition, and are attending a clinic with a member of your family, or a member of your family is visiting you in hospital.
This means you can take part. There will be about 600 other people taking part in the study too.

Do I have to take part?

No, it is up to you whether you take part.
If you do decide to take part you will be asked to sign a consent form after reading this leaflet.
If you to decide to take part you can still withdraw at any time without giving a reason.
A decision to take part or not to take part or to withdraw will not affect the standard of care you will receive.

What does it involve?

You will be asked to fill out a questionnaire about how your life has been affected by your medical condition. You will also be asked to rate your health on a scale.

In this study, your relative will be asked to fill out a questionnaire about the ways their life has been affected by your medical condition.

We will ask if we can contact you and your relative by post, to send you a questionnaire after the study. This is optional. You can still take part in this part of the study if you do not want to be contacted by post.

There are no tests (e.g. blood tests, x-rays). You will not need to take any extra medication.

How long will it take?

It will take on average 10 minutes to complete the questionnaire

What are the benefits of taking part?

We cannot promise the study will help you but the information we get from the study may help improve the treatment of people, and support the families of people with the same condition that you have.

What are the possible risks of taking part?

This study involves questionnaires so there are no risks associated with it.

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Project Protocol Version 10- 17/05/10

Appendix S

WHO/MSA/MNH/PSF/97.6
English only
Distr.: Limited

WHOQOL-BREF

PROGRAMME ON MENTAL HEALTH
WORLD HEALTH ORGANIZATION
GENEVA

For office use only

	Equations for computing domain scores	Raw score	Transformed scores*	
			4-20	0-100
Domain 1	(6-Q3) + (6-Q4) + Q10 + Q15 + Q16 + Q17 + Q18 ☐ + ☐ + ☐ + ☐ + ☐ + ☐ + ☐	=		
Domain 2	Q5 + Q6 + Q7 + Q11 + Q19 + (6-Q26) ☐ + ☐ + ☐ + ☐ + ☐ + ☐	=		
Domain 3	Q20 + Q21 + Q22 ☐ + ☐ + ☐	=		
Domain 4	Q8 + Q9 + Q12 + Q13 + Q14 + Q23 + Q24 + Q25 ☐ + ☐ + ☐ + ☐ + ☐ + ☐ + ☐ + ☐	=		

* Please see Table 4 on page 10 of the manual, for converting raw scores to transformed scores.

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I.D. number

ABOUT YOU

Before you begin we would like to ask you to answer a few general questions about yourself: by circling the correct answer or by filling in the space provided.

What is your **gender**? Male Female

What is you **date of birth**? _____ / _____ / _____
Day / Month / Year

What is the highest **education** you received?
None at all
Primary school
Secondary school
Tertiary

What is your **marital status**?
Single Separated
Married Divorced
Living as married Widowed

Are you currently **ill**? Yes No

If something is wrong with your health what do you think it is?_____illness/ problem

Instructions

This assessment asks how you feel about your quality of life, health, or other areas of your life. **Please answer all the questions.** If you are unsure about which response to give to a question, **please choose the one** that appears most appropriate. This can often be your first response.

Please keep in mind your standards, hopes, pleasures and concerns. We ask that you think about your life **in the last two weeks.** For example, thinking about the last two weeks, a question might ask:

		Not at all	Not much	Moderately	A great deal	Completely
	Do you get the kind of support from others that you need?	1	2	3	4	5

You should circle the number that best fits how much support you got from others over the last two weeks. So you would circle the number 4 if you got a great deal of support from others as follows.

		Not at all	Not much	Moderately	A great deal	Completely
	Do you get the kind of support from others that you need?	1	2	3	4	5

You would circle number 1 if you did not get any of the support that you needed from others in the last two weeks.

Please read each question, assess your feelings, and circle the number on the scale for each question that gives the best answer for you.

		Very poor	Poor	Neither poor nor good	Good	Very good
1(G1)	How would you rate your quality of life?	1	2	3	4	5

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
2 (G4)	How satisfied are you with your health?	1	2	3	4	5

The following questions ask about **how much** you have experienced certain things in the last two weeks.

		Not at all	A little	A moderate amount	Very much	An extreme amount
3 (F1.4)	To what extent do you feel that physical pain prevents you from doing what you need to do?	1	2	3	4	5
4(F11.3)	How much do you need any medical treatment to function in your daily life?	1	2	3	4	5
5(F4.1)	How much do you enjoy life?	1	2	3	4	5
6(F24.2)	To what extent do you feel your life to be meaningful?	1	2	3	4	5

		Not at all	A little	A moderate amount	Very much	Extremely
7(F5.3)	How well are you able to concentrate?	1	2	3	4	5
8 (F16.1)	How safe do you feel in your daily life?	1	2	3	4	5
9 (F22.1)	How healthy is your physical environment?	1	2	3	4	5

The following questions ask about **how completely** you experience or were able to do certain things in the last two weeks.

		Not at all	A little	Moderately	Mostly	Completely
10 (F2.1)	Do you have enough energy for everyday life?	1	2	3	4	5
11 (F7.1)	Are you able to accept your bodily appearance?	1	2	3	4	5
12 (F18.1)	Have you enough money to meet your needs?	1	2	3	4	5
13 (F20.1)	How available to you is the information that you need in your day-to-day life?	1	2	3	4	5
14 (F21.1)	To what extent do you have the opportunity for leisure activities?	1	2	3	4	5

		Very poor	Poor	Neither	Good	Very good

				poor nor good		
15 (F9.1)	How well are you able to get around?	1	2	3	4	5

The following questions ask you to say how **good or satisfied** you have felt about various aspects of your life over the last two weeks.

		Very dissatisfied	Dissatisfied	Neither satisfied nor dissatisfied	Satisfied	Very satisfied
16 (F3.3)	How satisfied are you with your sleep?	1	2	3	4	5
17 (F10.3)	How satisfied are you with your ability to perform your daily living activities?	1	2	3	4	5
18(F12.4)	How satisfied are you with your capacity for work?	1	2	3	4	5
19 (F6.3)	How satisfied are you with yourself?	1	2	3	4	5
20(F13.3)	How satisfied are you with your personal relationships?	1	2	3	4	5
21(F15.3)	How satisfied are you with your sex life?	1	2	3	4	5
22(F14.4)	How satisfied are you with the support you get from your friends?	1	2	3	4	5
23(F17.3)	How satisfied are you with the conditions of your living place?	1	2	3	4	5
24(F19.3)	How satisfied are you with your access to health services?	1	2	3	4	5
25(F23.3)	How satisfied are you with your transport?	1	2	3	4	5

The following question refers to **how often** you have felt or experienced certain things in the last two weeks.

		Never	Seldom	Quite often	Very often	Always
26 (F8.1)	How often do you have negative feelings such as blue mood, despair, anxiety, depression?	1	2	3	4	5

Did someone help you to fill out this form?.....

How long did it take to fill this form out?.....

Do you have any comments about the assessment?

.....

.....

THANK YOU FOR YOUR HELP

Appendix T

Family Reported Outcome Measure (FROM)© Feedback

Please answer the following questions about the FROM:

1. Is the questionnaire easy to complete?

   Yes ☐ No ☐

2. Are the response options for the questions straight forward?

   Yes ☐ No ☐

3. Are the instructions and statements clear?

   Yes ☐ No ☐

4. Do the questions cover all the areas of your life which have been affected by your family member's illness?

   Yes ☐ No ☐

If no, what has been missed?

……

……

……

Thank you

Appendix U

Date:

Dear Sir/Madam,

You were recently approached to take part in a Family Quality of Life research project. At the time you completed a questionnaire looking at the impact of your family member's condition on your life.

As part of the project we are sending participants another copy of our questionnaire to complete 1-2 months later. We would be grateful if you could complete the enclosed **questionnaire and the global health score** and return them to us in the stamp addressed envelope provided **as soon as possible**.

We cannot promise that the study will help you but the information that we get from this study may help improve the treatment of people, and support the families of, people with the same condition your relative has. In order for the study to be successful, it is important that we have a high number of questionnaires returned.

If you would prefer to complete the questionnaire online, please visit **https://www.surveys.cardiff.ac.uk/family** , or you have any questions, please contact Catherine on 02920 876017 or GolicsCJ@Cardiff.ac.uk.

Yours faithfully,

Family Reported Outcome Measure (FROM-16)©

The statements in this questionnaire relate to how **your** life is being affected by your family member's condition **at the moment.**
Please mark clearly one box for each statement.
Please remember, this questionnaire is about **your** life, not your family member's life.

Family Reported Outcome Measure (FROM)

5. Part 1: Emotions and Feelings

	Not at all	A little	A lot
a. I feel worried	○	○	○
b. I feel angry	○	○	○
c. I feel sad	○	○	○
d. I feel frustrated	○	○	○
e. It is difficult to find someone to talk to about my thoughts	○	○	○
f. Caring for my family member is difficult	○	○	○

6. Part 2: Personal and Social Life

	Not at all	A little	A lot
a. It is hard to find time for myself	○	○	○
b. My every day travel is affected	○	○	○
c. My eating habits are affected	○	○	○
d. My family activities are affected	○	○	○
e. I experience problems with going on holiday	○	○	○
f. My sex life is affected	○	○	○
g. My work or study is affected	○	○	○
h. My relationships with other family members are affected	○	○	○
i. My family expenses are increased	○	○	○
j. My sleep is affected	○	○	○

Appendix W

Family member interview guide

Introduction

- Remind participant that the information they give is confidential.
- Remind participant that the interview will be tape recorded.
- Remind participant that they can terminate the interview at any time.
- Tell the participant that the purpose of the interview is to find out how their lives have been affected by having a relative with a disease, and encourage them to answer questions as honestly as they can, giving examples when possible.

Opening question

- Can you tell me about any ways your life has been affected by your family member's condition?

Main interview questions (overview)

- Can you tell me how living with someone with your relative's condition makes you feel?
- Can you tell me what things in particular make you feel like this? Can you give examples?
- Do your activities change as a result of feeling like this? If so, how?
- How do you cope with feeling like this?
- Who do you talk to about feeling like this?
- Do you use any support services eg websites/counseling to help you with your feelings? If so, what do you use and why?
- How does your relative's condition affect your social life?
- Can you think of any social activities that you used to do which you can't now as a result of your relative's condition?
- What effect does your relative's condition have on your day to day activities?
- Does your relative's condition have any effect on your housework? If so, how?
- What effect does your relative's condition have on your friendships with others, both friends and strangers?
- Has your relative's condition affected any relationships in your family? If so, how?
- Do you feel that any of the family member's roles or responsibilities have changed as a result of your relatives condition? Can you explain how?
- Do you buy anything special of different as a result of your relative's condition? Can you explain what and why?
- Do you have any financial problems associated with your relative's condition? What are the cause of these?
- Does your relative's condition affect your job at all? If so, how?
- Has your relative's condition affected going on holiday at all? If so, how?
- Does your relative's condition affect your sleep? If so, why?
- Has your relative's condition affected your health at all? If so, how?
- Have you changed what you eat at all? If so, how?
- Do you have any support from people or groups? Can you tell me more?
- Has your sex life been affected at all? (partners only) If so, how?

Closure

- Is there anything else you can think of that you haven't told me?

- Is there anything else you would like to discuss?
- Thank you for your time.