

After Genetics: Huntington's disease, local data, global neuroscience

Jacqueline Hughes October 2010

**This thesis is submitted to the University of Wales in fulfilment of the requirements for the
degree of**

Doctor in Philosophy (Ph.D)

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In memory of Alan Brownhill, 1931 - 2006

After Genetics: Huntington's disease, local data, global neuroscience.

Summary

A 36 month ethnographic study of a research clinic with a combined role of disease management, included non participant observation of clinic routines, neurological consultations, motor and cognitive research testing of patients, plus interviews with patients, carers, clinicians, researchers and others associated with the clinic. A 'shift' of Huntington's disease into neurology was observed plus standardisation of research activities on an international scale. The clinic acts as a recruitment site for other experimental research. Research questions were – does a neurological instead of genetic framework make a difference to how the disease is regarded, and, what does research participation mean for patients and clinicians? A neurological framework appeared to encourage research participation because patients and carers considered it an opportunity for experimental treatment, including stem cell transplantation to the brain. Three analytic themes revealed: 'blurring' in operation of research and care, performances by all clinic actors linked to social and research expectations, plus the neurology framework increased patients' hopes in research aims. Sub themes included biomedicalisation, research translation, emotional work, research limitations, social benefits and transplant hope. Clinic researchers noticed tension in their dual research/care role, patients and carers noticed they were given time but little practical care.

Key words: clinic ethnography, blurring research and care, global research, huntington's, neuroscience, limitations of research, creation of hope.

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Chapter One

Introduction

In 2001, Stephen James received a letter from a doctor who had treated his late, estranged father in a nursing home. Stephen's parents had divorced several years ago, and the letter asked Stephen as the next of kin, to contact the doctor because there were aspects of his father's death that required discussion. Stephen, Jonathan his brother, and Carol their mother travelled to the hospital in another part of Britain from their home in the south of England. When they were settled in the consulting room, the doctor delivered the news which was to change all their lives; their late father had died of Huntington's disease. Carol and her sons were not aware of the disease, or what this might mean. The two sons were in their early twenties, fit and healthy, enjoying life. Jonathan had had some mental health problems which he received treatment for, but apart from that their future was bright. When they arrived home, they looked on the internet and found the awful details of Huntington's disease and what it meant to be 'at risk'. The two young men reacted very differently, Jonathan withdrew and did not want to know anything about it, but Stephen decided he would have a genetic test to find out if he too would develop the disease. The family went through the protocol of genetic counselling, but Jonathan remained withdrawn. After a few months, Stephen's test was done and the result was due, but Jonathan had gone missing. Stephen attended the clinic for his results on January 30th to find that he too would develop the fatal disease which had killed his father. On January 31st Jonathan's body was found washed up on a beach in the south of England. It was not clear if he had intended to take his own life, and an open verdict was recorded.

Carol is never sure what to tell people when they ask casually, 'Do you have children?' She sometimes replies that she has two sons but one of them has HD, and the well-meaning response can typically be along the lines of 'Oh I'm sorry, but at least its only one'. If she says she had two sons, but lost one, she typically hears 'Oh I'm sorry but at least you have the other one'.

(Reproduced from fieldnotes 2009, J Hughes)

Sag Harbor, Monday, June 30, 1806.

It has become our duty to publish the following melancholy circumstance, which took place at Easthampton a few weeks since:- Capt. David Hedges returned to his home in the evening, and found Mrs Hedges ironing clothes, and apparently in health- he retired to bed and left her at that employment, but on awakening in the morning she was not to be found. After considerable search and enquiry, her footsteps were traced from the house thro' fields of grain to the shore; and there is every reason to believe she has precipitated herself into the surf which washes the south shore. Mrs Hedges was about 40 years of age, and was much esteemed by her neighbors. This extraordinary step is attributed to her extreme dread of the disorder called *St. Vitus dance*, with which she began to be affected, and which her mother now has to a great degree. From some arrangements of her clothing it appears she had for some time contemplated her melancholy end.

Reported in the Suffolk Gazette, (East Hampton, New York) June 30, 1806 (after Wexler, 2008, p.3)

These stories of tragic early death, are separated by almost two hundred years, but they have much in common. The challenge of Huntington's disease remains for many families to this day, because there is no treatment or cure for a hereditary disease which takes away the ability to think and move independently in the middle years of life. The disease is rare, affecting possibly seven in one hundred thousand of the UK population according to official figures (Novak & Tabrizi, 2010), although recent reporting suggests this figure may be an underestimate (Spinney, 2010). There are approximately 6,300 affected people in touch with the Huntington's Disease Association in the UK, with at least a further 12,600 considered 'at risk' (Huntington's Disease Association, 2010).

Huntington's disease is dominantly inherited, which means that every child of an affected adult has a fifty percent chance of inheriting the gene and developing the disease themselves in adulthood. Death usually occurs fifteen to twenty years after onset of the abnormal motor symptoms and is often caused by complications of the effects of the disease. The disease usually affects adults in middle life, although it is known there is some variability in age of onset and severity of symptoms (Kremer: 2002, cited in Bates et al, 2002, p.29).

Huntington's is a neurodegenerative disease which progresses in several stages, and becomes visible due to involuntary twitching and jerking movements of the face, limbs and body (usually leading to complete disability), loss of cognition and insight and often dementia. However, the neuro-degeneration is known to begin long before any clinical signs of the movement disorder occur, and relatives often notice mood, personality or small physical changes in the affected person for some years prior to clinical diagnosis. This was clearly reported by relatives interviewed for this project, who recognized very small signs in their family members which indicated that they would indeed go on to develop the disease. A typical example was that of Edward, who told of his realisation that his wife was affected.

Jacki: OK, alright then. OK. Erm ... So, Anna had her test. She got her result, erm, but for you there were already signs because you'd seen ...

Edward: I knew it would be positive, yeah.

Jacki: Right. OK. Do you mind telling me what sort of sign you saw or

Edward: It's bizarre this is really, erm, when we first got married, when she used to sleep her hand used to go like that - but I just thought 'oh it's a bit of nerves' in the night, it didn't click in to me at all, until the scenario started to happen with the brother and I could see there was an issue there and once that started and the sister started to get diagnosed I thought well, that explains some things then. And like, if we'd be out or something and she'd go to clap her hands or something, she'd clap her hands in a strange way. You know, ... little symptoms.

(Interview 10, Edward, husband & carer)

Edward had noticed some different types of movements in the years before Anna had a genetic test, and he had put them to the back of his mind. However, when Anna's brother and sister began to develop physical signs and were later diagnosed, Edward realised that Anna too was affected. Anna did not realise and Edward did not tell her. This was also a typical reaction of family members who 'knew', they kept the information to themselves and the patient was eventually clinically diagnosed.

Huntington's disease provides a well documented example of how a disease is subject to changes in both social and scientific positioning over several years.

The present position of HD is in neuroscience, and the vastly increased amount of neurological research taking place on a global scale has raised hope and speculation for a treatment or cure among the Huntington's disease community. This recent shift from a primarily genetic framework forms the subject of this thesis, which will show the effects of the change in disease position and category for patients, families, scientists and clinicians.

From the earliest known records of the disease in the Middle Ages and continuing throughout most of the 20th century, Huntington's disease has been feared and hidden by many families, passed off as something different entirely, or erroneously believed to be the result of physical or emotional trauma. At various times in history it was considered to be witchcraft, and for the greater part of the twentieth century it was considered socially unacceptable to the point of eugenics by clinicians who would counsel people at risk not to marry or to be sterilized. For over a hundred years, it has been the subject of intense medical, biomedical, and genetic research by scientists and clinicians.

Alice Wexler (2008) tells a detailed story of the making of Huntington's disease in America, from the earliest times of its recognition to the present day. She has experienced Huntington's disease from both sides of medicine, her mother died of the disease, and her late father Milton Wexler, and her sister Nancy Wexler have been involved for much of their lives in scientific work towards understanding and ultimately, treatment for the disease. Importantly, Alice Wexler's account is a social history rather than a biological one, and she explains the way the disease has been regarded by patients, families and scientists and how different ideas at different times have created the atmosphere around the disease. Huntington's disease has been used as a 'model' disease for over one hundred years, firstly as an inherited disease of families, then as a neurological disorder, later as a 'single gene' disorder, a movement disorder, then as a psycho-social genetic disease and more recently as a fatal neurodegenerative disease.

However, the current situation in terms of neurological research and public awareness of the disease, mirrors the situation in the 1880's, when social, medical and scientific circumstances around the disease began to change (Wexler, 2008, p.96), and neurology became an important speciality of medicine. Wexler writes that the treatment of head injuries after the American Civil War in the 1860's had assisted the development of neurology in America, whilst in Europe this had happened ten years earlier. Young US clinicians travelled to Germany to be trained in laboratory work about the brain and then returned to the US with

their new knowledge, establishing medical practices for the treatment of nervous disorders. There was an expansion of asylums, a certain amount of biological reductionism, and along with the theory of evolution came authoritarianism and the ideas of eugenics (Wexler, 2008).

The medical profession in the late nineteenth and early twentieth centuries were complicit in eugenic strategies in the name of public health, and advised sterilization to whole families who were at risk of Huntington's disease, amongst other conditions which may cause 'feeble-mindedness'. Unemployment and poverty were considered to be the results of allowing unfit people to reproduce and social incapacity, rather than the effects of low wages and bad housing conditions (Porter, 1997, pp 639-640). This highly stigmatizing attitude prevailed in both the US and Europe, and the idea of populations free from 'mental defects', and the 'dregs of the community' was taken up by both right and left wing organizations in the early twentieth century according to Porter (1997). It was also taken to the extreme by the Weimar Republic and Third Reich in Germany from 1933 until 1945 (Bidiss, 1997, pp342-346) and included both those people affected with Huntington's disease and non-symptomatic relatives at risk. This was not the overt intention of the German race-hygiene laws as they were written, but became a popular interpretation of them as the following quote from Panse (1942) illustrates. He recorded that he had reported "all choreic cases, and moreover all suspicious cases and finally all not yet choreic sibs and offspring as being at risk to the health authorities". There is no doubt that killings took place of all types of people considered to be defective or risky, despite the 1933 'Law for the Prevention of Hereditarily Diseased Progeny' providing only for affected persons to be sterilized compulsorily (Harper, 1992; Muller-Hill, 1988).

According to Wexler (2010), it was not until the 1970's that stigma surrounding Huntington's disease in the US began to dissipate, as patients and scientists took part in the USA Commission for the Control of Huntington's disease. Public testimonies began to tell of the way people were still trying to live their lives in the shadow of eugenics, and continuing eugenic attitudes in the medical profession and wider society were exposed. One of the leading advocates for change was Marjorie Guthrie, the widow of songwriter Woody Guthrie, who had died with the disease in 1967 (Wexler 2008, p.53). This was an early example of the modern phenomenon of a celebrity admitting to a serious disease, which has since become a way of raising public awareness about various stigmatizing conditions (not all genetic in origin or indeed fatal), and in some cases this can contribute to reducing the stigma around disease.

The current placing of Huntington's disease within neuroscience is a move away from the disease's previous position in genetics. Within/prior to this project, the first indication I had of this difference was from the genetic medical notes of people in affected families. I was working on a research project which required me to create anonymised family pedigrees for use by mathematical colleagues who were researching insurance life tables. It was evident that a change had occurred in the way the files and accounts of hospital consultations for Huntington's disease were recorded. The change in recording was partly due to the change in emphasis from genetics to neuroscience, and I became interested in how this disease was represented at various times, and what might have an influence on the representation. In the older, archived medical files of people who were diagnosed with Huntington's up to forty years ago, there was a distinct format of longhand notes and copies of letters and artefacts, such as family pedigree diagrams, laboratory reports, post mortem reports, letters between family members and clinicians and many more examples, which covered the whole business of living with a chronic degenerative condition, and what this meant for the whole family.

The more recent files were far less descriptive, less 'social' in their detail, and more focussed on the individual patient and their disease progression. There was more evidence of technology, of various scientific testing, and standardized recording of motor functions and cognition. I have mentioned that this was only due in part to the change in positioning of the disease in neuroscience instead of genetics, and there are several other factors which must be considered. These include the styles of medical work which dictate recording practices, and the particular interests of the people who make the recordings in files. In addition, there have been legislative developments concerning the collection and storage of data and personal medical records, which all contribute to the changes in file content. However, those early files from the 1960s, 1970s and part of the 1980s contained important sociological records of disease, which have largely disappeared in more recent file recordings, because they are no longer collected as important information. Tibben (2002) asserts that there is a difference between the genetic and neurological approach to the patient and this is evidently an influence on the filed materials and records. As he puts it:

"There are important quantitative and qualitative differences between the way in which neurologists and geneticists attend to the patient. The neurologist is considering a neurological problem, the side effects of which may be given some

attention but are subsidiary. The geneticist offers more time and pays attention to the meaning of the disease and risks for patients and their families. There is a broader discussion of the impact of the disease on all life issues. This might explain why individuals with initial symptoms may prefer to visit a geneticist instead of a neurologist. A second explanation is that individuals with early signs of the disease may prefer a genetic test instead of a diagnostic test. Being identified as a gene carrier may confirm long-existing fears but allows the admission of being a patient to be delayed.” (Tibben, 2002, in Bates, Harper, Jones, 2002, p 224).

The move from genetics to neurology is therefore a clinical and practical difference in how Huntington’s disease is framed for the patients, families and clinicians. The purpose of this study is to show how this neurological framing operates in the research clinic by means of ethnographic observation, and what this can reveal about the purposes of clinics, and the expectations of patients, families and researchers.

The relationship between society and disease is an important feature of post industrial modern life, because as Lipton puts it “There is a set of expectations surrounding health and the body prevailing in western societies: we expect to feel well, without pain or disability, long after middle-age.... all surgery and medical treatment to be successful. And for the majority of people, these expectations are indeed met, serving to reinforce them even more strongly” (Lipton, 2003).

Kleinman theorized that diseases have social courses as well as biological ones (1995, p.151, see Wexler 2008, p.22 for a discussion of this), and in this thesis, I will show that the latest clinical patient-based research into Huntington’s disease, has further changed the social course of the disease, by placing the disease more firmly in a neurological category rather than a genetic one. In addition, the current research has taken on global proportions, with the collection of epidemiological data from national populations of Huntington’s disease patients all over the world to create a huge central data base. The clinic and the extended global project, will retain the patients’ data and human tissue samples indefinitely, for use by consortia of scientists to develop treatments, and eventually, it is hoped, a cure.

In the last two decades, new biomedical technological breakthroughs have been made, firstly by locating the site of the gene responsible for Huntington’s on chromosome 4, and then

developing a reliable genetic test which can be predictive or confirmatory of the disease (Huntington's Disease Collaborative Research Group, 1993). Secondly, approximately ten years ago, a very small group of UK and French Huntington's patients' received neuro-surgery to replace some of their damaged brain cells with brain cells from aborted foetuses (Bachoud-Lévi et al, 2006). The experiment was moderately successful, and encouraged further multi-million dollar global investment in Huntington's research, initially to develop drugs which will delay the disease progression. Eventually, scientists and patients hope that regenerative brain cell or stem cell transplantation will be available for this disease and other neurological conditions such as Parkinson's disease, motor neurone disease, and Alzheimer's disease.

Neurological research has increased dramatically following the creation of the Huntington's predictive genetic test in 1994, and this, plus the experimental neuro-surgery has precipitated a huge wave of optimism from patients, support groups, scientists and clinicians that a cure will be found. This has served to change the identity of Huntington's disease, from being a fatal genetic disease, to that of a neurological disease which many are convinced will eventually be treatable via drugs and/or regenerative medicine. However, the capacity of stem cells to produce neurological 'cures' is far from certain, and there are several scientific papers which express cautiousness following experimental transplants with animal and human subjects (Dunnett & Rosser, 2004; Keene et al, 2007; Cicchetti et al, 2009; Keene et al, 2009).

Almost in tandem with the longer term outcomes reported on the neural graft technology, the UK Government recently announced an All Party Parliamentary Group on Huntington's disease, to try to ascertain the true level of prevalence in the population (Spinney, 2010). This government initiative was stimulated by several factors; firstly, there is a discrepancy between the prevalence figures used by the UK government and the number of affected people who seek advice from the Huntington's Disease Association. The government prevalence ratio usually quoted is seven per 100,000, (Rawlins, cited in Spinney, 2010), but if the HDA figures of 6,300 are used, this works out at a prevalence ratio of just over twelve per 100,000.

The 6,300 people known to be seeking advice from the HDA is also considered to be less than the number of people who are actually affected (Spinney, 2010). In addition, the number of people who are pre-symptomatic in the general population is usually considered to be twice the number of those known to be affected. Add to this the fact that manifestation of

signs of HD are not in evidence usually until mid life, approximately between the ages of thirty five and fifty five, and there is the prospect of a large increase in the need for care and services for HD sufferers in the near future, which requires planning and resourcing. The disease may initially progress fairly slowly, people cope well in the early stages and continue to lead fulfilling and independent lives. However, increasing cognitive decline and physical disability affect almost all patients in the later stages, and assistance is required for everyday living. This has a huge impact on the economic, social and personal relations of patients and their families. Current care provision for patients is very uneven across the UK, with by far the greatest number of affected people receiving voluntary twenty-four hour care from family members. Those patients who have no relatives available to care for them, or for whom care has become too complex, are usually in nursing homes and may suffer from a lack of expertise in dealing with their condition.

The prevalence of Huntington's disease has always been a difficult figure to estimate. The development of a genetic test in 1994 was thought to be the technological breakthrough required to accurately check the prevalence rate, but it has not had that effect. Genetic testing is always voluntary, and restricted for those over eighteen. The majority of people at risk of Huntington's disease prefer not to know their status and reject the idea of a predictive test, the UK rate of predictive testing being stable somewhere between 17 and 20% of the at-risk population (Binedell et al, 1998). In the absence of any available treatment, this is understandable. It is thought that the prevalence rates in the UK population could be at least twice the rate of seven in one hundred thousand (Spinney, 2010). One reason for this is that HD has been so stigmatized in society, science and medicine, that some families to this day, continue to hide the true nature of the 'family ills'. Incomplete data from death certificates, and cases of misdiagnosis (particularly of Parkinson's disease which has some similar features), also add to the underestimation of disease prevalence. Collaborative research work will begin soon to try to determine a more accurate prevalence rate, and the Huntington's Disease Association of England and Wales is anticipating a 'feedback' effect, if higher prevalence rates are confirmed (Spinney, 2010, p.761). The 'feedback' effect means that once people consider the disease to be more common, even more individuals will come forward to acknowledge their illness. It may also have the effect of increasing the demand for genetic testing.

Sociologically, it is important to examine the trajectory of the enormous research programme dedicated to finding a treatment for HD, and to document how this is achieved on a local level from the day to day research clinic activity, which provides much of the data from patients and families. There are huge amounts of private and charitable funding being made available to take research further into the development of orphan drugs to halt the progress of the disease, and HD is used as a model for other neurological disorders (Hague et al, 2005). The science community consider that finding a treatment for HD will inevitably lead to treatments for other diseases such as Alzheimer's and Parkinson's disease 'in the not too distant future' (Hague et al 2005).

In the thesis, I present the findings of an ethnographic study at a clinic where patients take part in HD research. The important question is - how does framing the disease as neurological instead of genetic, (with the possibilities for future stem cell innovation), change the relationships between the patients, families and clinicians on one hand and the disease on the other? A second research question is, what does research participation mean for the patients, families and researchers? The possible futures of stem cell hopes have dominated medical research for the last few years and have been instrumental in raising awareness of genetics and regenerative medicine in all parts of society, stimulating a new phenomenon of 'stem cell tourism' (Campbell, 2010), which has brought warnings from leading UK scientists that there is yet a lack of reliable evidence underpinning treatments.

The clinic in this study, officially has a combined function of research and clinical management of the disease, but there are few, if any resources within the NHS at this site to accommodate the care needs of patients. However, the patients and families are both highly supportive of the research projects and at the same time desperately seeking assistance with the complex care issues of their everyday lives. The move away from genetics to neurology, together with the optimism regarding stem cells, has meant a higher public profile for the Huntington's disease research in general, and a lowering of the barriers and stigma surrounding the disease. However, there are also a few disadvantages related to services for patients. In the field of clinical genetics, a comprehensive service for families had developed over the last thirty years, incorporating counselling, genetic testing, care, family communication, home visits with a high level of support for carers and a high degree of co-operation with other social organizations involved with the patients' needs. Those issues cannot be dealt with by the

neurological research team who have to pass their concerns outside the clinic, often outside the hospital, sometimes back to GP's and primary care, or to other organizations, many of whom have little or no expertise with this disease. This is not only a result of the Huntington's research migration to neuroscience, it is also a feature of the present social care system which has pushed many chronically ill people outside the NHS framework, into the market-led environment of private care firms and packages, where expertise is often sacrificed in favour of lower costs and less skilled workers.

The project has provided data from ethnographic clinic observations, informal and semi-structured interviews, and a small amount of archival work. The data show that Huntington's disease has recently undergone a shift in category from genetics to neurology, together with a global emphasis on research unlike anything before. In this respect, the creation of global, anonymised data sets of brain disease progression (to be retained indefinitely) contributes both to the shift to neurology and the promise of a future cure. The current neurological framework around Huntington's disease has created the opportunity for several developments to combine and increase the hope for a treatment or cure in both patients and researchers. The developments which combine at this time are the social acceptance of foetal and stem cells as a functional material in experimental regenerative medicine, the western view of medicine as a means of restoring health (Lupton, 2003) and the increased acceptance of the biomedicalisation of the brain as separate from personhood or body (Clarke et al, 2003). These developments are viewed in the thesis via three main analytical themes, namely 1) the blurring of the roles of research and clinical care to recruit and retain research participants, 2) the performances enacted in the clinic which enable or disable the research, and 3) the utilisation of hope by patients, families and researchers in order to maintain engagement with research goals.

The remaining six chapters of the thesis follow a traditional style, and discuss the details of the planning of the study, the practical completion of fieldwork and the subsequent thematic analysis and recent related research. A brief outline of what can be found in each chapter is as follows. Chapter two considers the qualitative methodology used in the study from the planning stage, and covers the data collection and analysis. There were several methods of data collection, including non-participant observation

fieldwork of clinics, meetings and an information event in the community. In addition, there were many interviews of different types, for example chance encounters and conversations both in and around the clinic and the hospital, as well as planned semi-structured interviews with patients, carers, researchers and clinicians. What was particularly noticeable about the planned interviews, was the willingness of all the participants to give detailed and candid accounts of their part in the clinic work, whether or not they related this to the wider aspects of Huntington's research.

The literature concerning three emerging themes is the subject of Chapter three. Each theme has its own section in this chapter. It was made clear by the researchers from the outset of fieldwork that the clinic had a dual purpose of research and disease management, although I found these were not easily separated in initial observations. The movement between the two purposes was fluid and importantly, it was fully accepted by patients and carers, and this allowed me to identify the first theme as a 'blurring of the boundaries' between care and research. In contemporary western medicine, the practicalities of treatment usually take precedence in clinical consultation and there is a plethora of regulation surrounding research activities which are often considered to be completely separate. The 'blurring' of the two activities in the same clinic brings forward new questions about the regulation of research and the primacy of care, and also how to prioritise, when there is no standard medical treatment available, apart from experimental research. The second theme also occurred as a result of observation and interviews, and this was the existence of 'performance' as a part of the clinic, as a part of the disease, and as a part of the research. The literature here considers the different ways that this disease has been considered both in medical and social terms, over a long period of time, and how the latest phase of performing Huntington's as neuroscience, allows further translation of the technology surrounding it. The last theme of the literature chapter concerns the revival of hope in a cure or treatment, and the maintenance of future promise on both sides of the clinic encounter. The hope and promise factors are vital in keeping the research agenda visible, and there is evidence it helps patients and carers to make sense of the disease in some way. They may become socially visible and active in meaningful ways, and their participation acts

as a metaphorical slowing down of the patients' relentless physical and mental decline. In order to engender patient participation, certain criteria must be fulfilled on a societal level, there needs to be acceptance of particular 'translations' around the disease and I discuss this in relation to Ludwick Fleck's theory of 'thought collectives'. Fleck's ideas were of the early twentieth century, but there are interesting parallels with the current neuro-scientific ideology surrounding Huntington's disease.

Chapter four is an account of the first theme, 'blurring the boundaries of care and research' and how this was made visible in the data both from observations and interviews. The discussion moves from the identification of care and research, to the creation of research recruitment via this clinic and how this is of integral importance to the research strategy on a much wider international scale. Research is no longer a sideline activity, a curiosity to be pursued in spare moments, it has become the primary driver of the clinic and this is a change in emphasis of clinical medical work. This chapter also examines briefly, the types of clinic work that become secondary when the primary thrust of activity is research.

The fifth chapter concerns the second theme of the data analysis, that of 'performance, practice and experience' in the clinic and outside it. There are well established theorists of social performance such as Garfinkel and Goffman, there is the concept of the Parsonian 'sick-role', the existence of surveillance medicine as highlighted by Armstrong, the ceremony of the clinic as written by Strong, and the ideological position of Foucault regarding self surveillance which can undoubtedly be applied to most areas of clinical medicine and research in the twenty-first century. These concepts, forms of performance and ideologies are extracted from the data and shown to be part of the practice and experience of all the clinic actors, both when they are in the clinic and when they speak about the clinic. This illustrates the strength of individual expectations of clinic performance, and how people appear to naturally take on certain expected roles within the research enterprise. In addition, there is the role of the clinic research data itself to consider. This information, garnered from individual

patient 'stories' told as 'symptoms of illness', is standardized, collated and recorded to form anonymous micro-performances as part of global data collection.

Chapter six considers the third theme, the role of hope in the clinic. The utilization of hope is managed in the clinic environment in several ways, again by all the clinic actors. The hope and expectations of some patients for a recovery or treatment, requires skilful handling by the clinicians and researchers, yet it must also be preserved in order to maintain the research project. The researchers themselves need to feel justified that their work is worthwhile and will produce benefits for patients. Despite the justification for the research, progress is difficult to assess accurately, and this opens up areas of limitations, where in individual cases not all hope is justified, not all patients are suitable for research, and data collection may be discontinued. Such is the irony of mass research; it uses individual hope to facilitate research, and at the same time reduces individual data to anonymous particles of an unknown person, an unknown illness, a virtual model in effect.

In the final chapter of the thesis, I discuss the relevance of the three themes in relation to the local data collection and the global research project. The Huntington's disease neuro-scientific research project is based on the collection and analysis of massive data sets of particular disease symptoms, with the expectation that either stem cell technology or drug development will become a practical treatment to either slow down or halt the disease progression. This is considered a certainty by some researchers and clinicians, with the caveat 'if we continue to collect enough data we will find a treatment or cure, but we don't know when'.

During the neuro-scientific research process other social effects are apparent, and these are relevant to the two main research questions of the thesis. Firstly, I asked how does framing the disease as neurological instead of genetic, change the relationships between the patients, families and clinicians on the one hand, and the patients, families and the disease on the other? The discussion includes the social effects of the blurring between care and research, the shaping of the disease into a standardized, and reproducible model via regulation of the data, and the 'normalisation'

of research practices into clinical care, which replicates ideas about very early hospital medicine of the eighteenth century. Yet, within the boundaries of the clinic, the actors' performances become linked to the research objectives on both sides of the encounter, and biomedicalisation becomes a given. This effectively answers the second research question, which was, what does research participation at this clinic mean for the patients, carers and researchers? For the majority of patients and carers, it means a chance to participate socially in something meaningful, and potentially benefit from experimental treatment, and for the researchers, it signifies the current prime position of neuro-science in our society, juxtaposed with the normalization of this research as a mundane day to day activity which extends beyond the laboratory into the clinic.

Overall, the thesis shows how large scale international research is achieved in the local clinic, and the part played by local recruitment in advancing much larger research objectives. There are also local effects of the research agenda in terms of the regulation and standardisation of clinical practices which feed into the research at certain points, effectively recording what can be measured. Research and care are jointly overseen up to a point, the clinic operation itself is funded from research grants rather than the National Health Service. However, the majority of care provision and practice is also from outside the National Health Service with the co-operation of family carers, social services and charitable organizations. The ethnographic data show how the research activity is centrally co-ordinated and well funded, and in contrast, care is a much more individually difficult and piecemeal activity, with no central funding or regulation. This may indicate movement of NHS priorities away from the direct care of 'untreatable patients', where outcomes cannot be measured in more positive terms. Further research in this respect to try to establish the extent of research funded clinics in the UK (and the policies which underpin them), may have important long term policy implications given that until a treatment or cure is widely available, the number of Huntington's patients is likely to increase.

Chapter Two

Methodology

Introduction

This chapter will explain the methodology used in this study of the Huntington's disease research clinic and the researchers, patients and carers. The research design was informed by the research questions set out in the introduction to this thesis. Using several methods, I was able to gather rich and detailed data of the clinic and the lives of the patients and carers, plus the ways of working by researchers in the clinic. The range of methods used were participant observation, semi structured interviews, informal interviews, and a small amount of documentary analysis. The intention of this chapter is to describe these methods in detail along with their relative merits and disadvantages for this particular study. The use of multi-method research design has been documented as providing robustness in data collection and analysis (Janesick, 1994). In addition, I will draw attention to the ethical approval process and the access to the researchers and patients at the clinic, and access to patients and carers in their homes.

I will also discuss the various methods of gatekeeping which were in place around Huntington's disease itself, the clinic activities and the patients. The data collection was dependant on obtaining access to several different research sites such as the clinic, the administration of the research centre, the patients and carers both in the clinic and in their homes, and academic research seminars. I will also explain my gradually changing role in the research clinic and how I came to recognise this.

In addition, it is important to explain that the qualitative methods utilised in this study were chosen because they would provide the least disruptive method of seeing the clinic activity at first hand, and gave the opportunity to meet patients, carers and researchers during the clinic. Ethnographic methods were used in order to gain a greater understanding of the meaning of the research activity for all the actors involved. I begin by explaining the use of ethnographic methods in medical sociological research.

Ethnographic Methods

Ethnography as a method of social enquiry has existed for over one hundred and fifty years, as a useful and adaptable data gathering tool in many different settings (Atkinson et al, 2007, p.175). The whole point of ethnography is to produce accounts that “are grounded in a commitment to the first-hand experience and exploration of a particular social or cultural setting on the basis of (though not exclusively by) participant observation” (Atkinson et al, 2007, p.4). In this section, I will concentrate on how researchers of settings concerned with health and illness have utilised ethnography to produce vivid accounts via their ‘immersing participation in the lives of others’ (Bloor, 2001). Medicine and health constitutes a fairly large proportion of ethnography as a broad field, although as Bloor also points out, compared to the volume of medical research which takes place, these ethnographic health studies remain marginal (2001, p184).

Ethnography as a method of collecting and creating data from the field is not objective. It is a way of seeing the world or cultural space in appreciation of the meanings ascribed by the participants (Wolcott, 1999). Even this appreciation can be open to interpretation at several levels, that of the researcher collecting data, that of the creation of a field note, and that of the reader of any eventual text. For these reasons alone, it is not suggested that the ethnography described here is generalisable across all similar clinics. The purpose of the ethnographic observations and interviews in this study was to document and understand what participation in this research clinic could mean from different perspectives, the patients, their families, the clinicians, and researchers. In addressing the debates around ethnography and perhaps qualitative methods as opposed to quantitative inquiry, it is observed that ethnography has never been so popular in the social sciences and at the same time, subject to intense scrutiny (Atkinson & Hammersley:1994, cited in Denzin and Lincoln, 1994, p.249).

Other ethnographic clinic studies have covered various aspects of the clinical encounter, for example Strong (1979) examined the ways in which clinic interactions between parents and clinicians were achieved, by the use of various language devices and categorising of parents and roles. In his account, Strong (1979, p.16) explained that some of his methods lead to a concentration on the extreme or unusual and he was not trying to portray these occasions as wholly representative of the clinic. He clarified that the abnormal case was usually more helpful in the demonstration of certain rules and practices. This point is relevant to my study; I would not wish to claim that all the excerpts are typical of every Huntington’s research clinic, however, they are representative of my sociological

interpretation of the clinic functions I observed. Strong makes us aware that in displaying the difference between overt and covert actions of staff, it may have appeared as if his study was an exposé of medical staff. He explained that he did not have the same symmetrical data for the parents in the clinic, and because the staff were usually in control of the interaction, 'their covert actions are of greater analytical significance than those of the parents' (1979, p.17). This is an important methodological point worth making. In the study of the Huntington's disease research clinic, where tension sometimes occurred between the dual activities of research and disease management, it is not my intention to show any member of the staff or research team in a poor light, but to show evidence of the way the tension unfolds in the clinic and the strategies employed by various people to reduce or remove the tension.

Other medical ethnographic studies of note include MacIntyre (1977), Pettinari (1988) and Konrad (2005). MacIntyre studied ante-natal care of fifty first time mothers in Aberdeen, the study added important information about the assumptions made in the care of pregnant women and the subsequent feminist debates about the nature of managed childbirth. Pettinari (1988) studied the talk and the written records of surgery trainees in the operating theatre, and successfully showed the development of their recording styles from the beginning of training through to the end, as well as noting the difference in their spoken and written discourses. The elements of uncertainty observed in the spoken 'medical talk' were eliminated from the written records of operations, which showed confidence and competence in the surgical procedures. Monica Konrad's study of predictive genetic testing, charts the progress of technology and the subsequent altering of the relationship between ethics and medicine required in order to accommodate the implications of the new technologies.

Well known studies of clinicians in training such as Becker (1961) in the USA, and Atkinson (1981) in the UK, opened up the sociological knowledge of how people become doctors, and explained the informal rules and traditions which lead to their socialisation in the role. Becker studied the attitudes of student doctors, and the Atkinson study concentrated on the practical application of hospital bedside medicine. The Atkinson study was unique because it examined in detail the first clinical experiences of junior doctors, their first encounters with real patients in hospital beds as opposed to text books. The study revealed that the bedside encounter is by no means a simple exercise in either teaching students, presenting information or moving a patient's medical case along. Rather it is a complex interaction between the teacher and the student, and as Atkinson describes it "Students must generate displays of ability, under the scrutiny of their clinical teachers, and present a 'front'

of competence. In the context of such encounters, and through social accomplishments, medical knowledge is transmitted in a dramatic fashion” (Atkinson, 1981, p.10).

In order to address the research questions previously detailed in the introduction, concerning the meaning of clinical neurological research on Huntington’s disease to the patients, families and researchers, I decided that ethnographic methods would be the most likely route to uncovering the reasons for research participation and continued support and how these were brought about. It was my intention to gain an understanding of the place of experimental research in the lives of a group of terminally ill people for whom no treatment or cure exists. I also wanted to know what being a member of the research team meant for the clinic workers, and how this fitted into the work of a large research university and teaching hospital. The data analysis is informed by a grounded theory approach (Glaser and Strauss 1967) which provided a flexible, developmental approach to understanding the clinic research process and its impact on patients, carers and researchers.

In some respects, this project could be described as a study of how the illness of Huntington’s disease is viewed in the clinic, and how research work is achieved in the clinic. Ethnography would also allow a view into the world of the chronically ill long term patient and their care system. A study by Strauss and Corbin (1988) described the way that chronic illness was now the main feature of the demands on the US healthcare system and yet the prevailing organisation remained that of acute care. It occurred to me that Huntington’s disease patients and families in the UK health system may have had experiences which would also show the emphasis on provision of acute care facilities, due to the long term nature of the disease and the task of caring being performed by the family in many cases.

It is possible that some of my research questions may have been answered by a survey or questionnaire, but repeated observations and participation in the clinic allowed me to appreciate the way in which the clinic was performed as both routine and mundane for researchers, and at the same time as exceptional and hopeful for families and patients. Wolcott (1999) puts forward a helpful question which could address many settings and I found this to be a useful approach in the clinic and in interviews; he asked ‘what do people in this setting have to know and do to make this system work?’ As Wolcott also suggested, it is not enough for the ethnographer to simply try to observe everything, and he helpfully pointed out that is not usually very successful. Instead there must be the start of an idea from the ethnographer of what would be of interest to them in this setting (1999, p.70). For my study of the Huntington’s disease clinic, my ‘hunch’ was concerned with the activities of research

and care happening together in the same place, and the meaning of the research clinic for all those involved.

Researching a marginalised population

Huntington's disease often remains undiscovered until middle life, when a diagnosis is life changing for most people and their families to put it mildly (Konrad, 2005). There are disturbing physical signs and symptoms, cognitive and behavioural changes, with no medical treatment or cure available. The disease is fatal, genetic, rare, dominantly inherited and has a highly penetrant effect, which means everyone who has a positive genetic test usually develops the disease at some point. Family secrecy and avoidance were ever present in those early accounts of the disease in the archived files, and the social effects of the loss of insight, cognition, and physical control of the body were explained at length.

Huntington's disease is often used as an example of a 'genetic disease with fatal consequences' in both medical and social terms (for an early description see Spillane & Phillips, 1937), and the feelings it evokes are often 'worst case scenario' genetics (Cox & McKellin, 1999, p.632). In the insurance business, it is the only genetic disease for which testing must be disclosed under certain circumstances, and in this it stands apart from other genetic diseases (DOH, 2005). In this respect, ethnography of the research clinic was that of a community marginalised by not only a health issue, but also the incurable status of the disease, and the absence of any possible avoidance of it by self imposed health behaviours. This contrasts sharply with late modern ideas of healthism, where the body is considered to be under surveillance, management and control of its owner (Lupton, 1997, p.103).

The Clinic Setting

The research clinic was held in a new research centre building on the main teaching hospital site, it was spacious and quiet, unlike many of the other outpatient clinics where noise, equipment and people jostled in the same space. Both the patients and the research staff seemed to appreciate the new space and what it signified about the patients' and the importance of the research.

R: Yeah, and certainly our patients have come from us having clinics in genetics where it was always more difficult, the rooms weren't really set up for a neurology clinic Well, we were very, very welcome until more recently when there was a sort of change of management.... But also it's a slightly dingier building, the light isn't so good you know – I do think it makes a big difference actually. And the other thing the patients have always said is that they actually, funnily enough, they like to talk to each other while they're waiting, they find that a really positive aspect, erm, which is interesting, erm, and I think it makes them feel less lonely, you know ...

(Extract from Interview with Researcher 4)

This short but succinct explanation of the advantages of the new location also discloses the researcher's knowledge about the participants' situation of loneliness, and the perceived added bonus of chatting to other patients and families in the waiting room.

This was also confirmed by interview data from families who noticed the difference too in some cases.

C: Since they've moved, it's lovely. It's lovely. I got to be fair, the receptionists – whoever they are, it's not always the same one – they make [us] very welcome, you know, ask if you want tea or coffee...It was over in Genetics Research ... I loved it over there. Because he was going so long, from when (he) got diagnosed.

So they all knew us and it was lovely but in this, it's not like a hospital ... They're lovely there and ... it is David isn't it? When we go in and they always speak to Dave. You know, they don't say 'would David like a cup of tea?', they ask David direct, 'would you like a cup of tea or coffee'. They're lovely.

(C = carer) (Extract from Interview 5 Patient and Carer)

C: I mean one of my biggest bugbears with the clinic for ages was when it was in the Genetic part, we couldn't put posters up. I mean, I was desperately trying to set up a support group in my area

and yet at the very clinic where HD was, I couldn't put up a poster because they wouldn't allow things on the wall. It's different now, there's things in this one but, I mean for a long time – and Valerie said the same, you just weren't allowed to put things on the wall and you just kind of felt ...

(C =carer) (Extract from Interview 3 with carer)

Although the two extracts above are different in their tone and their reasons for appreciation of the new location, what they have in common is the feeling that somehow, the clinic attendees are considered important, and that the primary concern of the clinic is the patients.

Having said that, the experience of coming to a research clinic once or twice a year often took the form of a 'special event' for patients and families, even though to the researchers it was a mundane, routine process. This became one area of particular observation, a social event with different personal meanings and emphasis for the participants (Atkinson & Hammersley:1989, p.32). However, this was not always an accurate or full picture. Further patient and carer interview data gave a different view, that of the stress involved for patients and carers in preparing for a clinic visit, making the journey, waiting around and participating in the non-invasive research, which they sometimes found unsettling because it involved awareness of increasing disability.

Although this is one 'local' clinic rather than the only centre for HD research, patients travel from other areas of the UK to visit, as well as the local population who have visited the same hospital site (but a different building) for many years. The senior clinician explained to me quite early in the study that the increase in requests to attend this particular clinic is related to the clinic's link to work on invasive neurological research in particular. There have been very few of these experimental procedures with HD patients in the UK to date. At the present time, the experimental procedures to replace HD patients' brain cells with those from aborted human embryos have been suspended until work on a new laboratory is complete. The senior clinician also considered that when this experimental work resumes, there is likely to be another increase in requests to visit this clinic, from patients around the UK. The clinic site is therefore somewhat unique; it has a representational value to the patients and families who are keen to obtain some treatment even if experimental, and it is one local data collection point in a large multi-national research programme. The research for a treatment or cure for HD, has taken on global proportions in the twenty-first century. There are over 130 research sites worldwide, and more than 25 in the UK. Measurable data of all

types is collected, including DNA, blood, urine, cognitive testing scores, filmed neurological responses and the Universal Huntington's Disease Research Scale (UHDRS). Data are centrally stored in different national repositories for use by the global research community in the quest for a treatment or cure. Some of the blood, urine and DNA samples go to Italy (by courier post) for storage, the numerical scales of disease progression are entered onto an international database which is managed from Germany, and the filmed recordings of neurological response tests are sent to the USA. The local clinic where I observed is part of the European Huntington's disease Network, which has the European Registry Project as its main objective. This project collects epidemiological data as described above, from thousands of patients across Europe. What makes this particular local clinic so attractive to patients and families in the UK, is that it is located close to the university laboratories where the neurological cell transplantation is being researched as experimental neuroscience, and the researchers in the clinic are part of the university research team.

The next part of this chapter will give details of the data collection process for the thesis, most of which took place in the clinic setting.

Data Collection

The observation data collection took place over eighteen months in the clinic, and most of the informal interviews were also collected during this time. Recorded interviews with clinic and NHS staff took place during the second twelve months of the project, and patient and carer interviews were the last set to be completed in the third twelve month period of the project.

The following tables give a numerical breakdown of the ethnographic data collection in terms of different types of observations, recorded semi-structured interviews and informal interviews.

Table 1: Ethnographic data collection – Observations - field notes

Type of observation	Number of sets of field notes
Clinic and research consultations	28
Pre-clinic meetings researchers	3
Post clinic debriefing, researchers and admin staff	4
Genetics and neuroscience seminars	3
Support group day	1
Total	39

Table 2: Ethnographic data collection – Recorded Interviews and informal interview

Type of interview	Numerical breakdown sets of field notes and transcripts
Informal interviews patients & carers (at clinic)	11 (field notes)
Informal interviews patients & carers (Support Group Day)	5 (field notes)
Informal with NHS staff, researchers, & others	3 (field notes)
Recorded clinicians & researchers	6 (transcripts)
Recorded other NHS staff	5 (transcripts)
Recorded patients	4 (transcripts)
Recorded carers	7 (transcripts)
Total	41

The arrangements for observations and interviews were initiated by letter; patients and carers could ‘opt-in’ if they wished. I had negotiated with the senior researcher and the clinic secretary so that details of my research project would be sent out with the clinic appointment letters. I supplied invitation letters, information sheets, consent forms and pre paid envelopes, and these accompanied the clinic appointments (see appendices). If someone agreed to be observed and meet me in the clinic, the idea was that they would send the signed consent form back to me and I would attend their clinic session. In practice, no one sent the consent

forms back to me, they sent them to the clinic, or to the genetics department, or brought them on the day. I never received them in time to plan an appointment based observation schedule. I quickly realised that my organised system was not going to work, and the alternative was to attend the entire clinics as much as possible and observe who ever turned up and agreed to it. Though this seemed to be a rather tenuous arrangement at first, it worked very well, and I felt more easily embedded as a regular clinic attendee who was there from the start to the finish of the clinic day. The added bonus was that I was able to experience clinic days as reasonably complete events, and I was eventually aware of the multiple activities happening simultaneously in different areas, not just the sections I was observing.

Fieldnotes

My main method of data collection in the clinic was via field notes. I found I wrote different types depending on where I was in the clinic. Most of the obvious writing took place in the neurological consultations, where I seemed to write furiously to keep up with the dialogue and activities. Other notes were made almost at the same time as observation or very soon after, and these were a little more reflexive and expressive. Different types of field notes are discussed by Emerson, Fretz & Shaw (2007, cited in Atkinson et al, 2007, p.352). They are selective forms of representation and therefore are only partial accounts which include 'interpretation and sense-making' (Emerson, Fretz & Shaw, 2007, p.353)

I began naively, noting physical spaces, people's activities within the space, and I was not at all sure I was doing the right things to get to know the clinic. I was very aware of the clinic talk, and this was easier for me to note, describe and possibly deconstruct, than the actions and taken for granted-ness of the site and the patient, clinician, hospital area. It took time for me to look at the site with full appreciation of the strangeness, and I was keen to fit in when I first arrived. I did not want to have the role of the stranger and I was very aware of being a social scientist, and not a bio-scientist as the other researchers were. I began by feeling it was necessary to mention to patients and families, that taking part in my research would not necessarily bring any improvements in their care, it was more a study about how the clinic worked.

Wolcott (1999: p.120) talks about different ways of approaching ethnography, either by 'shopping around' or looking at other introductory texts to find a suitable structure to begin with. My own approach began in what I thought was an organised way, I had copies of consent forms, stamped return post envelopes, lists of researchers, directions to the clinic. I

was open about my purpose there, and introduced myself to as many people as I could. I asked lots of questions about the clinic, and tried to make friends with the researchers. This had less effect than I hoped, and I suspect kept me as an outsider for longer, underlined my qualitative research as different to everyone else's quantitative projects, and possibly annoyed people with my enthusiasm to be included as a natural part of the clinic. I realised later that I was 'shopping around', just as Walcott described, trying to find something to hang onto as a research area, rather than gradually taking in the way the clinic worked and its effect on people.

After an observation day, I would write up the field notes again, sometimes more fully, and try to work out what had been happening, if I had not already noted it down. At the beginning of the observations this seemed to be very mechanical, and I often did not know immediately what I was seeing (Rock, 2001 & 2007). In between the lines of field notes, I would often write 'my note' or 'note to self' and then make a reference to a particular thought, theory or similarity with other notes I had taken, or an area of medical sociology I had read about. The purpose of this was that I should not forget what related thoughts had occurred to me as I was observing and as I reviewed the clinic. In effect I was writing down my mental notes too (Lofland & Lofland: 1995, p.90)

My field notes were my accounts of the activities of the clinic, and of as many actors as I could possibly observe. It became more focussed and rewarding as I became more embedded in the clinic. It was not a problem to make the field notes, but keeping the data under control was challenging. At the beginning of my clinic visits, I was conscious of trying to be unobtrusive to both patients and researchers, and I felt awkward with my 'outsiderness'. During later observations, I found I was concentrating on the main themes I had identified, and was intent on the interactions, often leaning forward and watching closely. My notes were more focussed on the themes of blurring between research and disease management, clinic performance, and the utilisation of hope.

I was able to attend two neuroscience and a genetic research seminars within the local research community, to hear how the bio-scientific neurology of Huntington's disease was presented to other bio-scientists. Field notes were taken on these three occasions. I also observed a Support Group day attended by patients and families, the HDA Regional Care Adviser, social workers and carers, with presentations given by some of the clinic researchers and by some family members. I made extensive field notes on this occasion and managed to informally interview several family members, a voluntary worker and a social carer.

Ethnographic experiences in the clinic

In conducting a clinic ethnography, it was evident that this would involve some delicate unpacking of the accepted positivist rationale of early 21st century Western medicine, and I was a little nervous. What might ethnography show, that would be useful to patients or clinicians? There was a sense that I should be making a contribution to the workings of the clinic somehow, but also an understanding that I should not interfere with clinic work or get in the way. The preoccupation with evidence based (and usually quantitative) research in medicine, meant that my work was in sharp contrast to the neurology clinic researchers. I had no closed sets of categories to look for. One definition of ethnography as a form of social research explains several features of the method;

1. A strong emphasis on exploring the nature of particular social phenomena, rather than setting out to test hypotheses about them.
2. A tendency to work primarily with unstructured data that have not been coded at the point of data collection in terms of a closed set of analytic categories.
3. Investigation of a small number of cases, perhaps just one case in detail.
4. Analysis of data that involves explicit interpretation of the meanings and functions of human actions, the product of which mainly takes the form of verbal descriptions and explanations, with quantification and statistical analysis playing a subordinate role at most.

(Extract from Atkinson & Hammersley, 1994 cited in Denzin & Lincoln 1994, p.248)

Bloor (2001) explains clearly that the ‘epistemological basis for ethnography is hermeneutics with its concern for immersive understanding’, and this was something I was aiming for by becoming embedded in the research clinic (p.179). However, Bloor continues his consideration of the role of ethnographies, with the assertion that critics of ‘immersion’ find this approach to be an essentialism, a search for the unattainable, and warns that medical ethnographies ‘cannot tell eager medical professionals what their reticent patients ‘really’

feel, think, and aspire to: there is no final authentic reality awaiting ethnographic evaluation' (also p.179).

Walcott (1999: p.50) explains his ambition to be the 'fly on the wall' when observing, and 'not to be seen as the evaluator', and I remember a similar feeling. I was probably over-sensitive to this because the LREC chairman had named my study as an evaluation, to my disappointment.

I wanted to be accepted in the clinic as a fellow researcher by the other researchers, who were mostly psychologists and bioscientists who used quantitative statistical methods. My initial visits showed that I was welcomed by the senior clinician, researchers and some staff, but viewed with some uncertainty by the psychologists and some NHS staff. Their view seemed to be that they knew what the clinic was about, and so what did I *really* want to look at which involved hanging around and making lots of notes? Similar experiences are discussed by Hammersley and Atkinson, who note;

"Gatekeepers, sponsors and the like, (indeed most of the people who act as hosts to the research) will operate in terms of expectations about the ethnographer's identity and intentions. As the examples of Hansen and Barrett make clear, these can have serious implications for the amount and nature of the data collected. Many hosts have highly inaccurate and lurid, expectations of the research enterprise, especially of ethnographic work" (Hammersley and Atkinson, 2007, p.60).

The well established procedures of the clinic were a place for me to begin to look and ask questions, which I suspect was irritating at times for the clinic researchers, even though everyone was exceptionally well-mannered towards me. I felt self-conscious and was aware that I was trying to appear normal, friendly and non-threatening (Hammersley & Atkinson: 2007, p.70). I was not sure if I should appear as 'entering the field with fresh-eyes', or in the 'role of convert', I probably tried both at various times (Lofland & Lofland, 1995). There was certainly a feeling of discomfort at different times, as I struggled with my own role in the clinic.

During an early introductory conversation with the lead neuroscientist on the Huntington's research clinic project, I had become aware of an organisational shift from 'management of Huntington's disease' to 'research and management' of the disease. It occurred to me that this might throw up some interesting things but I had no idea what they might turn out to be. Until this point, I had aimed the majority of my study at the examination of the archived medical records, interviews with families, and some clinic observations to compare old archived files with new ways of recording disease progression. However, the changes in organisation of the Huntington's disease clinics, was an opportunistic stimulus to which ethnography was a distinctly advantageous and flexible approach. (Atkinson & Hammersley: 1989, p.24)

Interviewing clinic professionals, patients and carers

In addition to the ethnographic observations, I used both informal and semi-structured interviews, visited patients and carers in their homes, and attended neuroscience research seminars and talks. The purpose of doing some interviewing was to check if my observations had been accurately transformed into field notes from the perspective of the different participants, and to fill in the gaps in my knowledge about the clinic and its effects.

The informal interviews (from encounters with patients, carers and all types of medical staff during the observations) were also in the form of field notes, often almost verbatim accounts of conversations. I had not anticipated collecting this type of data during observations, but it proved to be a worthwhile source. Wolcott (1999: p52) considers this to be one of the most important areas of data sourcing because it is part of the everyday nature of fieldwork. During clinic observations days, whilst making field notes, I would also sometimes carry out informal interviews with patients and families if the opportunity arose. I originally considered these informal interviews to be another type of field notes, but because they involved exchanges of dialogue between myself and others, sometimes for several minutes, informal interviews seems a more appropriate term according to Walcot (1999).

I made detailed notes concerning 16 informal interviews with patients, family members, carers, and 6 with researchers and NHS staff. With hindsight, it would have been a good idea to have digitally recorded these exchanges, but the truth is, I did not recognise them as a form of interview at the time. It may also have inhibited the naturally occurring conversation to request that we begin again and record the conversations which were informal. Recording

can be a barrier in some informal situations where the participant or interviewee is not expecting it (Rubin & Rubin, 2005).

Semi-structured recorded interviews with 11 clinic personnel also began during the first year of the study. These interviews were with researchers, elite professionals in the Clinical Genetics and Neurology departments of the hospital, and Department of Bioscience in the university, plus other professionals and administrators associated with this clinic from both inside and outside the NHS. I began this aspect of the data collection to strengthen the observational data, and to elicit the professional researchers' views on the clinic activities. It was also a strategy to engage the researchers and workers, and obtain a trust relationship, which, as I have previously mentioned, was not automatic with everyone I came into contact with (Bosk: 2008). Although there were themes I wanted to explore with researchers, I also hoped that many topics would be covered during 'naturally occurring talk' (Hammersley & Atkinson, 2007).

I had initially planned to have some interview content as part of the project data and intended that this should provide triangulation with the observation and file data, plus it would give the opportunity to ask further questions. It would also give the opportunity to check my understanding of what I had observed in the clinic with researchers, patients and carers, plus compare the families' perceptions with what the files had recorded.

The interviews with researchers highlighted themes of hope, uncertainty, the long term research view, tension between research and providing care, and the wider picture of global research.

R: Exactly, so whether the patient ends up getting a consultation which is largely clinical need or largely research depends on how they are on the day when they ...

R: It is an open-ended thing so for that clinic it's not a matter of ... there aren't any targets. So the idea is that everybody in Europe, who's part of this, collects data on as many of the patients they happen to see in clinic as they possibly can. So, erm ... and also the other element that actually I didn't mention is if the patients are on that registry database it makes it much easier for us to recruit them to other trials ... (Extract from semi structured Interview Researcher 5)

R= Clinic Researcher

In the extract, the researcher explains how the decision to do research work or a clinical consultation, depends on how the patient seems to be at the clinic appointment. There are no

recruitment targets, however, the implication is that all patients who are seen here are potential research participants. In addition, the researcher was explaining the importance of recruitment of patients to the main study and how all other recruitment hinged on this.

The final stage of the fieldwork was 10 semi-structured recorded interviews with patients, family and other carers, in the third year of study. These interviews took place in the patients and carers homes, and on one occasion at a workplace. In most cases, the home based interviews took place some months after clinic observations, and after I had previously met the participants either at the clinic or at the family support group day. Establishing rapport was extremely important, the patients and carers have such difficult lives that giving their time to my project was a commitment on their part, and something I wanted to acknowledge. It was noticeable that the people who responded with the most enthusiasm in the clinic and agreed to be interviewed at a later date, and seemed very keen that I should visit them at home, did not respond to later written requests for interviews. I have given some thought to this, and there are several possibilities, including the possibility that people attending the clinic found it easier to agree so that they are considered co-operative. It is also possible that the home situation and the patient's condition had changed between the first suggestion of a home interview and when the people were contacted. Approaching people whom I had met at the family day, or through local support groups was a much more successful recruiting strategy, and led to other recommendations for interviews in one or two cases.

Rubin and Rubin (2005) consider the establishment of rapport as vital to the building of trust, the flow of the conversation and the consideration of what subjects the participants think are important to discuss. When I visited people in their homes or in the workplace, I checked by telephone the day before to ask if the appointment was still convenient. This allowed for some rapport building before we met again. In one instance it was particularly useful, because finding the address was not straightforward, and heavy snow made my journey longer than anticipated. Several phone calls to the carer during the journey helped me greatly with directions, and gave the carer and myself some common ground to ease us into the interview- bad weather. Patients and carers interviews revealed themes of hope for the future of research, coping with HD in society, effects of caring in the home and on the family, and specific living knowledge of the disease.

Sue: It's just trying to get new people. It's hard to get people to come to the support group, that's the way they want it, they want to keep it in their family. And yet it's so open now, everybody knows about HD.

Jacki: Would you say that that's changed then? You said 'and now it's so open' so that sort of tells me that you appreciate that there's been a change in how the disease is perhaps looked at. Do you think that that's right?

Sue: Yes, but I'm always doing awareness I am, trying as much as I can to get people ... giving leaflets and things to the Doctor's surgery and the Dentist.

(Extract from Interview with Patient 4 and carer/family)

Sue = Carer, Dave = Patient, Jacki = Interviewer.

In this extract, Sue was explaining that people are reluctant to come to specially held support groups even though they may benefit from this. I drew attention to Sue's reference to the 'open' nature of knowledge about Huntington's disease and checked that I understood what she meant by this. Sue confirmed that she performed 'awareness' in her daily social life, and pinpointed that making professionals aware is also part of this. As the carer for her husband, Sue considered it her responsibility, to inform others about Huntington's disease, and yet pointed to the way that families can perpetuate the hidden nature of the disease by not attending support groups. She also made a contrast between her own openness and the reluctance of others.

The third source of data in the project was from personal genetic files of deceased patients, many of these gave details of families who struggled to cope with the disease, and preferred to hide the evidence of the family illness from the outside world, just as Sue had described.

Personal medical genetics files

The original plan for the thesis project involved an archival study of many personal medical genetics files, however, there had been difficulty in gaining ethical approval to view the files of living people due to the logistical problems of gaining informed consent. Given the difficulties of accessing enough files to make a sensible archival study, I redesigned the study to have the main focus as the clinic ethnography. The access to a few files was helpful

because it provided a contrast to the current system of recording patient data. I therefore arranged to view some personal genetics files of deceased patients because this was the least problematic type of access to arrange in terms of ethics and consent. At the beginning of the project, I had enquired about creating a list of files where the person was deceased. This was not possible because there was no system for identifying a list of files of deceased people separately from anyone else. A helpful colleague in the Genetics Department, found a way to assist me by looking in old work diaries, where unofficial lists of deceased people had been made. This proved to be the only way to discover a quantity of files apart from examining every file in the archive. It appeared to indicate that priority lies with the bioscience of Huntington's disease, and not records of patients' deaths, in effect, a disembodied knowledge from the patient themselves.

Analysis of data

Analysis of data was continuous throughout the study from the initial observations and interviews, through to summaries of field notes, and transcripts of recorded interviews. The main themes which were apparent from the field notes were firstly, the tension between the two areas of clinical care and scientific research, in effect a blurring of boundaries between two separate disciplines or activities, (which the researchers acknowledged but could do little to change) and secondly, the raised hope and searching for a treatment on the part of the patients and families. There were also themes of 'performance' in the clinic, for example the 'ideal patient' and carer, and the understanding but very 'focussed' researcher. The international nature of the clinic research was an overarching framework of all the clinic activity, and there were many positive references to the large amounts of global research by researchers. Other themes encountered in the ethnographic field notes were, the sense of occasion felt by patients and carers coming to clinic, and their explicit roles as patients, carers and family members in this setting. This contrasted with the role of the researchers who were keen to put people at ease, empathise and encourage, whilst carrying out the repetitive mundane research work of the clinic.

The field notes often showed a contrast between the research being carried out and the patients and carer's search for assistance and potential treatments. In addition, the global circulation of research data hung in the background, in huge contrast to the local collection of information, the care issues, the assessment of patients, the 'front' and 'back' stage areas of the clinic, and the performances of 'good' patients, 'good' carers and professional clinicians

and researchers. There was also a sub-text of tension between ‘providing care’ for people (clinical) and who should make this possible, and doing legitimate research (scientific activity).

Coming back to these detailed notes time and again, I was eventually able to pick out different layers of meaning which were not immediately obvious to me at the time of observation. One such area was the translation of individual patient and carer perspectives into numerical scales and anonymised data. This was happening during every research process I observed, but only long after observations had ceased was I able to perceive this change. Prior to the realisation of an existing translation, I had considered it to be note taking and ‘marking’ of patient’s scores, just mundane, normal clinic work. Discussions with my supervisors also caused me to think about the context in which the data was collected and the positioning of the clinic participants. As I carried out more observations this became easier to do. There was the clinic process to understand, and the actions of the patients, carers and clinicians to observe, and the meaning of the disease in this context to appreciate. Charmaz (2006: p.34) asserts that grounded theory works best if the researcher gathers data and proceeds to analyse it, because nuances of meaning and process are less easily missed. There will be many areas I did miss, simply because I must have attended to some things and not others (Atkinson:1990, p.9, cited in Emerson, Fretz & Shaw 2007, p.354). This is often used by positivist critics of both ethnography and grounded theory approaches because it produces a less generalisable picture of events and processes. However, ethnographic accounts by their nature are only partial representations, although they can achieve a degree of robustness from the collection of other data such as interviews and documentary evidence, described as corroboration by Walcott (1999).

In the interviews with researchers and professionals, the tension between care and research was always present, even though I was hearing official accounts of research programmes and aims. It was evident that researchers recognised the intent of patients who came seeking a ‘cure’ and left with nothing in particular. The massive global research effort was emphasised as justification for the continuing of research, and there was recognition of the prominence of this research centre and the amount of expertise to hand from the nearby bio-science research laboratories. There is also a long and continuing fascination on the part of bioscience to find a treatment and eventually a cure for Huntington’s disease, an achievement which will bring great kudos to those who achieve it. The patients and carers interviews were indicative of hope for the future cure of the disease and a recognition that most of what happened in the clinic was ‘monitoring’ of patients. There was also clear

recognition of the prominence of this research site in terms of expertise and access to potential stem cell transplants. There were many claims of altruism, and talk of the impact of the disease on lives. In addition there was talk of the impact of the clinic on families, both positive and negative, and the experience of time passing without a solution being found for individual illness.

The use of a grounded theory approach allowed the raising of categories to come from the data in all the methods employed, observation, interview and archive work (Lepper: 2000). The early coding of field notes and interview data produced many different analytical categories, at least 25 altogether, with some of them overlapping. An example of this was a category of 'Big Science vs. Clinical Management', from the observation data, and 'Raising of hope and expectation' in the interview data. These two areas were linked by the process of the clinic and its dual purpose, although it is possible to treat them entirely separately. This was indicative of what was emerging from the collected data, and represented what I had seen in the clinic, created from the ideas and concepts which occurred to me whilst I had observed. Their basis was not in undisputable facts, but in my own prior knowledge and influences (Glaser & Strauss: 1992, p.90). Occasionally there were interesting developments which underlined some part of theory and yet countered initial findings. For example, during clinic observations I perceived an element of performance on the part of patients and carers, due to entering a site where they were expected to fulfil certain roles as patient and carer, not only that, roles as 'ideal patient and carer' were employed to facilitate research involvement. The positive mindset surrounding clinic attendance led me to think of how the clinic was used as an 'event' in otherwise routinised, difficult lives. However, while checking on this aspect during interviews with patients and carers, there was more revealed about the tension caused by preparation for a clinic visit, the anxiety of travelling for patients, the tiring nature of the visit and the disruption to biography caused by undertaking research activities. This was only one element of complexity which existed in almost every category I created from the data, and led me to think of the research clinic as almost the embodiment of a chimera—something new created from the essence of different existing organisms.

Research ethics, access to files and redesigning the study.

Before I could officially begin to observe any documents or people, I had to apply for and receive approval from the NHS Local Research Ethics Committee. It is worth outlining this process because it is a crucial part of any study involving research recruitment of NHS

patients. It was also an important turning point at the beginning of the study which altered the main emphasis from an ethnographic archival study, to an ethnographic participant observation enquiry of a live clinic in action.

Any study involving NHS patients is required to have ethical approval from the Local Research Ethics Committee (LREC), and this is a necessary process but daunting in its bureaucratic procedures for a new researcher. Since the time of application for the HD study, the procedures have again been revised.

In the case of the Huntington's disease research study, I had originally intended to investigate archived material which exists in old medical genetics files of previous patients. This was considered a risky enterprise by the LREC and I subsequently changed the study design to an observation based project for the reasons I will explain here.

Having written a study protocol, I was required to hold an Honorary NHS contract which bound me to follow NHS rules of patient confidentiality, and this extended to contacting my prospective participants for all matters involving files, observations and interviews. The Research Ethics Committee approval was dependant on full compliance with NHS confidentiality regulations, of which written evidence was required. This was in the form of information sheets, consent forms and details of my prospective interview guideline (see appendices).

The project had received approval from the NHS trust Research and Development committee, which also acted as University Peer and Risk Review committee. This stated clearly that no data which would identify any patient must be recorded in any research documentation, and was subject to satisfying the first principle of the Data Protection Act. I had to demonstrate clearly that no rules of confidentiality of medical records would be compromised in this study. In order to do, this my application stated that the historical records would consist of those files relating to people who were deceased or with whom there had been no contact for at least fifteen years. All data would be anonymised and/or changed to avoid identification. This was to satisfy Part IV, Exemptions, section 33 of the Data Protection Act principles 1 and 2 (Cardiff University, 1998). More recent files for comparison were to be accessed with individual full informed consent obtained via initial contact with health professionals. Full informed consent would also be sought from current clinic patients prior to observation and interview. The Research Ethics application contained the research protocol, and details of Research and Development Committee approval with attention paid to the issue of access to medical notes.

My previous research work in collaboration with the Genetics department had informed most of my knowledge of the older large family files. These files from the Institute of Medical Genetics are guarded as precious and highly confidential, they are stored securely in a place known as 'the dungeon' with two electronically coded doors between them and outsiders. Outsiders are defined as anyone who has no right of entry to the electronically coded areas, and insiders are a few people who work in Medical Genetics. This includes secretaries and administrators, genetic counsellors and clinicians, and researchers with permission from the Genetics department. The files for Huntington's disease were kept here and were distinctive from other conditions by the colour of the file case. Although the condition is rare, there were more Huntington's files in this location than any other files, and some of the contents date back to the early 1960's and possibly earlier. Prior (2003) explains that the file accounts of patients also create a basis for social action, a justification of treatment or referral. There was a great deal of theorising in the archived files about familial relationships, because at the time the files were created, a research team was attempting to establish the extent of Huntington's disease in a particular geographical area. Berg & Bowker (1996) refer to this construction via medical records as being a central element in the re-writing of the patient's body. This showed as a definite contrast to the current files, which showed more coded evidence of medical examination and measurement related to neurological research, and designed to be read by neuroscientists, rather than the difficult social positioning of patients and families displayed in the older files.

Once my research ethics application had been accepted into the committee system, I was given an appointment to meet the Ethics Committee and answer questions about the study. The session began for me when I was invited into the committee room, and the Chair introduced my project to the members as 'an evaluation of the clinic service to HD patients'. The committee was made up of at least twelve members plus the Chair, this was twice as many as I had expected and I was quite nervous. I attempted to explain that my project was to be an ethnography of the historical files and the clinic rather than evaluation. The Chair responded with 'so it's an evaluation really, whatever you might want to call it'. However, evaluation was not the main problem for the Research Ethics Committee. Their members were most concerned about why I wanted to look at securely held, confidential medical genetics files, and how I would get informed consent from everyone who was named in the files before I could look at them. This was a rather difficult question, because if you cannot look in the files, you cannot know who is named in them. In research situations such as this, social researchers often seek co-operation from clinical staff, who will send information

letters and consent forms to people they have identified as possibly being responsive to the invitation to participate. The patients and family members may then 'opt-in' if they wish, and until they do there is no direct contact with the researcher. This arrangement is usually satisfactory in terms of patient confidentiality, but has several disadvantages. It can make recruitment a lengthy and sporadic process. It is also dependant on the clinical staff having the time and the inclination to carry out this task as a favour aside from their other work, and the resulting recruitment will reflect their perceptions of 'suitable patients'. However, there were no consent issues involved if people had died, looking at their files was acceptable practice.

Senior clinicians in the Genetics department were supportive of my project and I hoped this would assist in calming the concerns of the ethics committee. One committee member asked me if I thought it was right to go looking into people's files, and asked how I would feel if my family medical records were made available in that way. This was an important point that I obviously needed to take account of more seriously; my benign feelings were not necessarily reflective of the general public's attitudes to medical confidentiality. I had to agree that full informed consent to access files, although desirable, was not practical and probably unachievable. I attempted to justify my request for access in terms of the sociological value of looking at this disease from another perspective. I had also tried to work along the lines that all information would be anonymised and that the files were important historical documents. Direct contact to patients from the Genetics department appeared to be the correct solution. However, this was a path fraught with difficulty for the reasons previously stated, it was a lot to expect and rely on from NHS staff, and I was concerned it would make recruitment extremely slow. In addition, not everyone in the files was in contact with the Genetics service or wanted to be, I knew this from my previous research work. Despite my assurances of complete anonymity for all parties, concealment of identities, changes to locations and other details, the ethics committee members remained unconvinced of the need for access to files without full informed consent.

Several changes were made to the study in order to comply with conditions of ethical approval. I would only look at files of people who were deceased, as this posed no problems of consent. The study was redesigned to become an ethnographic account of the current research clinic plus interviews with clinic participants. In the event, looking at files proved to be a very minor part of the study, because the opportunity to observe the research clinic in action became the main focus. Access to the research clinic was simpler to manage within the terms of NHS patient confidentiality, and the LREC did not view 'evaluation' or

‘ethnography’ as problematic, providing both patients and researchers gave informed consent. The opportunity to observe the clinic became central to my study instead of peripheral, with additional material provided by ethnographic interviews, various other observations and a few file studies of now deceased patients.

Initially I felt some disappointment, because I considered it was important to use the rich data from files to make a record of the social interactions which helped medicine to create a disease and establish a service to patients. I remain convinced that such studies of older medical records can explain a great deal about the sociology of disease, research and treatment, as well as changes in technologies, ethics and attitudes towards medicine.

With hindsight, I now had full access to an interesting empirical study of a critical period in genetics and neuroscience.

Gatekeeping of the disease, the patients and the clinic

I had worked on a previous research project and knew several clinicians and workers in the Genetics department which allowed me to discover the structures and gatekeepers which required negotiation. There were gatekeepers for access to clinic researchers, access to patients, and access to information, and they were in the Genetics department, in the research clinic, and in the hospital more generally. Each had their own area of protectionism, guarding patients, files and places from the taint of un-informed consent.

One of the most important gatekeepers for the whole study was a senior research geneticist who facilitated access to the senior researcher, the neuroscientist who made consultations at the research clinic. I had requested a meeting to explain my early ideas for observing, and was keen to meet the person before I began to attend the clinic. I was invited to discuss my study and our meeting took place in the research scientist’s office, with the research geneticist making the introductions. Before I began, there was a short discussion between the two senior researchers about funding for the Genetics department and the implications for the research clinic. It seemed that budget cuts were looming, and the suggestion had been made at some level of hospital management, that disease management of HD should be combined with the neuroscience research clinic. Instead of attending separate clinics, the voluntarily researched patients and the disease managed patients would all follow the same pathway through the research clinic.

This appeared to be problematic for the research team, and was something I began to appreciate and attend to during observations. In effect, I was beginning to observe at a time

when the boundaries between the two separate activities of patient management and clinical research were becoming blurred.

Specialist nurse

Another important gatekeeper was a specialist nurse for HD, who arranged for patients to attend the research clinic. I maintained close contact because this was my route to knowing when clinics would take place and which patients I would be able to contact to ask about participation. This worked very well initially, but the specialist nurse moved to a different job after a couple of months of my involvement with the clinic. However, the nurse was instrumental in arranging my first visits to the clinic and showing me around, and in the weeks following her departure this proved to have been time well spent as I found my own way around the people and the processes.

A major feature of Huntington's disease in the national health system is the amount of gatekeeping which operates both officially and unofficially. An example of official gatekeeping was that patient files were kept very securely and were separate from any other medical file a patient may have in the hospital. A specific procedure was followed to take files to the clinic building for the research clinic and to return them to the genetics file store. Unofficial gatekeeping took place in the clinic when it was decided who would sit in and observe which consultations, if there were several clinic visitors. Another unofficial gatekeeping activity concerned non-symptomatic patients arriving for clinic appointments. They were often protected from seeing other more disabled patients in the waiting area, by means of careful shuffling of appointment times, or active intervention by alert NHS staff. I observed this happening during a research clinic and made field notes. During the clinic preparation time, Paula, a nurse specialist, had talked to the Professor about a new patient coming to clinic, and I was aware that some minor shuffling of times had been done.

I remembered Paula had said to Prof James earlier on to get this patient in for consultation quickly, as there were some badly affected people in the waiting room, and it might not be very nice for her to be out there with them. I wondered about that, protecting the patient from seeing other patients, and preventing them from a view of their future perhaps. But not everyone is

affected in the same way, although maybe you don't think like that if you are going to be affected. (Field notes March 2007)

Paula's negotiation of the appointment showed sensitivity to the new patient, and awareness of how they might be feeling when arriving at the clinic for the first time. It was also part of presenting the clinic as a benign environment, somewhere a patient may consider visiting again, perhaps to take part in research.

Other official gatekeeping and coding activities were already well established before the clinic moved to the research building, for example the visibility of the files. The genetic files for Huntington's disease were a different colour to those for other diseases, creating an unspoken code within the genetics clinics and genetics department concerning the movement of those particular files. NHS staff constantly reminded researchers from outside (such as myself) of Data Protection requirements, and the impossibility of removing files from the building or of 'cold contacting' current patients without a health care worker as intermediary. Gatekeeping of this type was aimed at preserving the privacy of patients from anyone who was not directly involved in their care (Mueller 1999 study refers)

In order to develop trust between myself, genetics staff, clinic and research staff, I attempted to keep my activities very transparent in and around the research clinic and the genetics department. I distributed information sheets about the project to all staff I encountered, and sought informed consent from everyone I talked to and interviewed (see Appendix for copies/details of information and consent forms). I also made myself known to reception staff whenever I entered the clinic or genetics department, signed in and out as a visitor and wore an identification badge when requested to do so.

The more senior clinicians and researchers were relaxed from the very beginning of my study about issues of patient privacy or confidentiality, but junior clinical staff, reception staff and some NHS staff were highly sensitive both to my presence in the clinic, and to other people arriving ad hoc, during my earliest months there. On one occasion some months into the research observation, a different NHS nurse was working as the research centre receptionist and was wary of allowing me into the clinic. As I approached the desk to sign in he asked if I had an appointment, I explained my observer role briefly and showed my university identity card. However, he decided I should wear a visitor's badge too, and proceeded to create a handwritten one which he put into a clear plastic holder with a clip attached. This caused a certain amount of comment from the clinic researchers when they

saw my new badge. At this stage, I was some months along in my clinic visits, and the researchers could barely contain their amusement.

The process of first obtaining informed consent from patients and carers had been arranged with the senior researcher and the research clinic administrator who sent out appointment letters to patients as I have previously described. However, becoming embedded in the clinic was a much better arrangement which enabled me to take advantage of being on the spot and make observations whenever the opportunity arose. Early in the study, I had to contact clinic researchers weekly to enquire if the clinic would take place, and after several months I began to receive occasional emails with lists of future clinic dates from one researcher.

Renegotiating access to individual patients was sometimes successful, and at other times it was not. One successful renegotiation happened by chance when I approached a couple in the clinic waiting room to ask them about a possible interview. As I began to talk to them, I recognised them from some months before, when they had declined to be observed. I was fully expecting to be turned down, but to my surprise, they agreed there and then to do a recorded interview with me in the waiting room.

A less successful attempt at renegotiating access happened towards the end of a clinic day when most activities had finished but cognitive testing had not. I had observed several neurological consultations and wanted to observe the psychology tests but I had not yet managed this. The timing of the psychology testing often coincided with my observations of neurological consultations, and initially the psychologists were wary of my clinic presence and unsure of my purpose. Early in the study they sometimes referred my observation requests back to the senior researcher even though I had ethical approval and informed consent from the patient. However, the following extract shows the unpredictable nature of field research and I was unable to use the data.

I asked if I could observe their cognitive tests today- they said yes, and would I like to consent the patient so I could use the data?- Yes! But the patient had been there ages today and had consented to 2 other studies, was tired, didn't want to go through consent process again. Let me sit in theCognitive language test - words and pictures, lists of things, and numbers to remember forwards and backwards, naming things, thinking of words to describe things.They will let me look at the blank tests another day- might ask me to do control part. They've got 15

affected, 15 pre-symptomatic, and 15 controls in the whole study.

Apparently there are very subtle language changes early on some French research has found.

(Field note 9c, 2007)

The psychologist delivered the 'no-consent' message to me from the patient, and so I sat in during testing but did not make notes. I had noticed the junior researchers and NHS staff went to great lengths to explain and clarify consent issues to patients whenever I was observing, and it took some time. On this occasion, it suggested to me that the consent process was normally lengthy for patients and perhaps inconvenient. This was mentioned to me by a researcher at another time, it was an informal confirmation that patients find the informed consent process onerous at times.

Informal interview between patients with Researcher.

Discussed ethics with Reschr. She says ethical requirements become burdensome to patients. Explaining all info sheet and consent form puts them off- they think they are signing up to more serious things than they actually are. (Field note June 2007)

The researcher was expressing a personal view about the consent process and how this could become a barrier for recruiting patients to research studies. It was not clear whether this experience was gained prior to this research clinic or during it. However, because the main focus of this particular clinic was research, recruitment was naturally important and obstacles were unhelpful.

Conclusion

In this methods chapter, I have explained the development of my research methods from obtaining ethical approval for the observations of neurological research clinics, through to how I collected and analysed data from the observations, interviews and personal genetic medical files. I have also commented on my own position in the research clinic and how this changed over the period of clinic observations. The next chapter of the thesis will review the relevant literature based on the three analytical themes I have taken from the project data, namely, the blurring of boundaries between research and care, the performances enacted in the clinic, and the utilisation of hope to assist the neurological research.

Chapter Three

Literature review

Introduction

There are two constant themes in the background of the thesis; the re-categorisation of Huntington's disease as a neurological problem rather than a genetic disease, and, the international global scientific research effort to find interventionist treatments, with the corresponding multi-national collections of local patient data for scientific use. The study of the Huntington's disease local clinic has revealed three further themes which are dependant on the new disease categorisation and the research operation, and these are as follows:

- 1) There is 'blurring' between the two activities of clinical care and disease research which is becoming a regular feature in some areas of medicine, especially clinical trials. This creates tensions between the research and clinical activities, and changes the care practices for patients.
- 2) The clinic routine and the nature of the disease, encourage certain types of performances and practices from clinic participants, which have an influence on the way the disease is viewed socially, medically and scientifically. Performances of particular types are invoked by patients and carers in order to try to obtain treatment, and researchers perform moral work in the clinic in response to these.
- 3) The creation and maintenance of hope in future treatment is one of the pervasive effects of the research clinic. The hope is produced in the environment of bioscientific advancement, and it also serves to encourage and maintain research participation from patients, enhance the clinic reputation and scientific capital, and create challenges for the clinicians who carry out the research to manage the expectations of patients and families.

I will discuss each theme in relation to the disease ‘movement’ into neuroscience and the global research effort, drawing on relevant and current literature.

The first section will discuss the blurring effect and the subsequent tensions created for research and clinical when these two formerly separate activities are pushed into a combined clinic framework. The combination of clinical care and research is not altogether a ‘new’ phenomenon, the earliest medical research began in the UK during the scientific revolution of the seventeenth century (Porter, 1999) however, the individual medical specialisms and conferences which heralded the routine of experimentation in medicine, occurred in the mid nineteenth century. For a period of time in the early twentieth century there was little ethical governance, but from the 1964 Declaration of Helsinki onwards, there has been an explosion of regulatory procedures which have served to keep the practices of medical care and medical research apart. Since the mid twentieth century, there have been increasing attempts to mark the borders of what constitutes clinical care and what is research in the exchanges between clinicians and patients. This has taken the form of special rules which govern when and where someone can be included in medical research programmes, and a host of regulations governing the activities of clinicians and researchers in different settings. This differentiation which began as an ethical process in ancient medicine, has been deemed necessary to prevent conflicts of interest occurring when clinicians who treat patients also engage in research, sometimes with the same patients. (See for example The Declaration of Helsinki (WMA, 1964), Universal Declaration of Bioethics and Human Rights (2005). In this chapter I will concentrate on the theme of ‘blurring’ of research and care as it has re-appeared in early twenty-first century medicine, and review some of the developments (and their effects) in clinical consultations where research may also be present.

The second section of the literature concerns the clinic as a place where the actors’ performances are related to the ways in which they understand their roles, and the roles of others. This is related to the practices of the clinic and the expectations held by both the researchers and the patients and carers. I will link this with the growth of biomedicalisation, and the expansion of medical technology, which is considered to

have begun in earnest in the mid twentieth century (Clarke et al 2003). This section will also review the role of the creation and use of patient's medical notes in presenting a picture of disease, specialised testing including genetic testing, and the impact of change in health care organisation on clinic practices.

The third section will discuss the theme of hope in relation to the biomedicalisation project, in particular the literature concerning future hope for stem cell technology. Discussions on translational work have revived in the last decade with the sequencing of the human genome, the growth of biotechnology to industrial levels of production, the need for a constant supply of large numbers of research subjects, and access to large amounts of data. The involvement of patients' organisations in research has added to the shaping of the drive towards translation of 'future technological promise' into treatments, a situation that Novas frames as 'political economy of hope'(2006). The requirements of large scale research projects such as Euro-HD, render the clinic a fertile source of human research subject material, with increased expectations of greater and faster translation into clinical treatments via news media, health promotion, project websites on the internet, and many other forms of global information management. In contrast to the global activity which stimulates the hopes and expectations of the Huntington's research community, the local clinic is considered by patients as a key site for the chance of treatment, and is actively used as a place of participation. The increasing demand for clinics for many newly identified and as yet untreatable and incurable conditions, provides a potentially richly accessible source of research material, and effectively 'blurs the boundaries' between clinical care and research activity. I will discuss this area first.

'Blurring' boundaries of care and research

Ethical traditions

Medical care and medical research are theoretically separate in current Western medical practice for ethical reasons, although this has not always been the case. A brief look at the development of ethical traditions and medical history reveals the Paris hospitals of

the late eighteenth century had been used as ‘scientific machines for investigating diseases and for teaching..’ (Porter, 1999) and at that time research and care were often the same thing.

In most medical traditions, there have long been accepted professional practices to deliver medical treatment on the basis of a code of ethics, and these were often but not always, derived from the ancient civilisations such as Greece, hence the Hippocratic oath (Veatch, 2000). However, it was during the nineteenth century that a code of medical ethics was formalised by the American Medical Association in its founding year of 1847. The code was based on the work of Thomas Percival, (born 1740) an English physician, and Veatch (2000) claims this code had more to do with the Scottish enlightenment values than simply ‘benefit-harm’ per the Hippocratic stance.

Further advances and changes to the nature of ethical codes followed, the most relevant for the thesis being the Declaration of Helsinki (1964), which refers to medical experimentation with human subjects and was recently amended in 2008.

In the early twentieth century, further moves were made towards separating the activities of research and clinical care, and to make each one a distinct area of expertise, with the informed consent of the patient or research subject (Porter, 1999). The reasons for the separation are concerned with the rights of human research subjects, and the ethics surrounding medical treatment, and the possibility of conflicts of interest by practising clinicians.

The separation and current merging of research and care

The majority of clinicians and scientists practising in the mid twentieth century have long accepted that there may be a conflict of interest in clinical and research encounters which is why the culture of professional ethical rules was developed; it is to ensure that liberties are not taken with regard to patient’s free will and choice about medical trials, and that patient’s clinical care is not compromised by their decisions about whether to co-operate in medical research or not. It also protects the researcher from later claims of health damage or wrongful treatment because the patients must be informed properly at the outset, hence ‘informed consent’.

Most of the ethical concerns about research reflect clinical trials of drugs or surgical procedures, or at the very least procedures which have a potentially measurable impact upon a research subject or a patient's or condition. It is necessary to indicate that in the study of the Huntington's research clinic, there is very little being 'done to patients' for the purpose of research apart from psychological testing, movement tests, and data gathering, and in this respect, the blurring of research and clinical care is a slightly different area to capture. What the epidemiological data is creating in the Huntington's research clinic, is a population ready for experimental medicine, and a standard form of the disease which is suitable for the purposes of neuroscience.

However, it is possible to view the global data gathering and epidemiological studies of Huntington's disease as a research instrument or 'trial' and apply similar distinctions to it as one might a drug or procedure. Until relatively recently (perhaps the last decade or less), these borders between care and research were rarely crossed except in unusual circumstances, life and death situations for example, and always (in theory) with the full informed consent of the patient or their next of kin. Western medicine has created a whole culture and technology around 'informed consent', 'experimental treatment', 'clinical trial', 'research ethics', and so on (Veatch, 2000).

However, in the latter part of the twentieth century and the beginning of the twenty-first, it has come to light that the separated areas of research and care are beginning to merge again in some areas of medicine. Literature has begun to appear on the 'blurring of boundaries' between research procedures and clinical care, (Clarke et al 2003, Easter et al, 2006, Hallowell et al, 2009). Boundaries which were created in the first place for reasons of protection of the individual patient and researcher as already described, and usually in consultation with interested parties, (such as specialist patient groups for example) have become barriers to the requirements of industrial scale scientific research. This is very apparent in the UK system of the National Health Service which provides medical care free at the point of need, and the separated clinical research activities, which require explicit consent from patients and enshrine the principle that a patient's medical treatment will not be affected if research participation does not go ahead. The reality in western scientific research is that it is difficult to find

enough of the right type of patients to successfully conduct clinical trials, (Petryna, 2009) and I will return to this in the discussion chapter 7.

Reference to the existence of blurring of the different types of scientific and medical work in the late twentieth century was made several years before the work of Clarke et al on biomedicalisation (2003), but in a slightly different context. Elston (1997) wrote about the physical locations of University College Hospital and University College London buildings, connected by subterranean passageways that remained out of public view.

“However, the subterranean metaphor was not intended to imply that the relationship between ‘science’ and healthcare was necessarily one of science underpinning health care; that is, a relationship in which ‘pure’ knowledge is created by scientists in laboratories, developed into technologies and then applied by clinical or public health practitioners to lay people’s health problems. To the patient, the dependency may often look more like the converse: relatively lowly laboratory technicians use routine technologies to serve powerful clinicians. Information, knowledge and technologies move in both directions between practice and the laboratory. The boundaries between different kinds of laboratories may be permeable, the distinction between science and healthcare practice blurred.”
(Elston, 1997, p.3)

The proximity of research laboratories to sites of research appears to have an influence on the types of collaborations and the working relationships between laboratory scientists and clinic scientists. In the case of the Huntington’s clinic some of the laboratory researchers also took part in and helped to create the regular research clinics. This aspect of the work did not form part of my research questions, but it may be a useful area to observe. The clinic workers did express some discomfort with patient testing on occasions, but the research held priority the majority of the time. The subterranean metaphor employed by Elston was issued in the introduction to a collection of writing, which included a study of blurring of care and research by Mueller (1997)

Mueller discussed a site of AIDS research where the dilemmas occurred between clinicians and nurses. The dilemmas were concerned with the status of patient-volunteers, and the different ways the status was viewed by clinicians and nurses. The clinicians had research reputations and career funding to consider in the light of potential recruitment, successful research and published outcomes. They viewed the patient-volunteers in a more objective way than the nurses who were more concerned with the care of the patients as individuals. Further disagreements became evident when patients wanted to switch to a study they believed was 'better'. The nurses questioned if this was true, and if so, why were the nurses being encouraged to retain patients in the 'old, less helpful study'? These differences in approach illustrate the problems of trying to align the requirements of recruitment and 'gold standard' research, with the long held ideal of what is in the patient's best interest. Care issues also became a point of difference in the Mueller study, because nurses appreciated the extent of patient-volunteers' illness (they saw them regularly), and they realised the effort required to participate in research and the toll on everyday life. Nurses were reported to want to extend general care into the research practices, but the clinician investigators were concerned with the increased costs, and drew research boundaries very narrowly around clinical trial practice. The study in question was American, and therefore further complications arose with medical insurance costs, uninsured patients, and the possible exclusion of the latter from the research trials. Attempts to reconcile the professional operational differences at the AIDS clinic site involved a team of consultants who interviewed the research teams, and discovered discontent with how the research protocols were operated on both sides, nurses and scientists. The consultants suggested the formalisation of professional roles and duties in the research protocol, and sets of policy guidelines. However, as Mueller reported, the policies and guidelines gave preference to the scientific and clinical research basis of the protocol and less emphasis and importance to the general care roles, so in effect, they reproduced the institutionalised patterns of scientific power and inequality.

The study indicated that nurses were more in touch with the patients and built up relationships with them, knowing their everyday lives and knowing the person, whereas physician investigators were slightly removed from this level of awareness, and so they

could more easily detach themselves from any contentious care issues. Mueller did not intend to study the operation of clinical research, her aim was to explore the intraprofessional relations and interaction in the clinical research environment, but she recognised the importance of observing clinical research in the light of the AIDS activism which had some small degree of influence over physician decisions. However, her study concluded that the organisational structures of health care were very effective in maintaining medical discretion and autonomy, over intraprofessional relations.

The study has relevance for the Huntington's research clinic study, because similar intraprofessional activity occurred there. The research team was led by a senior researcher (neuroscientist) who had honorary NHS consultant status. There were also specialist nurses in research nurse roles, and non clinical researchers such as the psychologists and the physiotherapist. The research centre staff in the Huntington's study were NHS employees rather than researchers, some of whom had nursing backgrounds and this may have accounted for some of the differences in approach to the use of the research centre which I refer to elsewhere in the thesis.

Patient expectations of the clinic in the Huntington's study also involved a certain amount of care, due to the unusual dual role of the clinic, and the research staff recognised this; the difference in the Huntington's study was that the research staff did not appear to hold different views from each other; they recognised that care should be given but considered it should come from elsewhere, within the NHS for example. On this there was a similarity with the Mueller physician-investigators who wanted to preserve the research budget for that purpose alone.

More recent important studies of the blurring of research and care include Clarke et al (2003) and the biomedicalisation of health. This important paper mapped the increasing role of science and technology in health care, and pointed to the inclusion of research as an element of the biomedicalisation project, together with self care and self surveillance.

The biomedicalisation of medicine

Clarke et al (2003) consider the changes in practical medicine from medicalisation to biomedicalisation of health care from a historical point of view. They indicate that the project of biomedicalisation organisation is much broader than that of medicalisation:

“Biomedicalisation is characterised by its greater organisational and institutional reach through the meso level innovations made possible by computer and information settings, including computer-based research and record keeping. The scope of biomedicalisation processes is thus much broader, and includes conceptual and clinical expansions through the commodification of health, the elaboration of risk and surveillance, and innovative clinical applications of drugs, diagnostic tests and treatment procedures. This includes the production of new social forms through “dividing practices”, that specify population segments such as risk groups (Rose 1994)” (Clarke et al, 2003 p.165).

Risk groups identified during the medicalisation project included for example, the unborn child, the pregnant woman, the elderly, the young, and those without access to clean water and healthy living conditions. Hogg (1999) claims that the public health message was taken over by medicine, from the very effective sanitation reformers of the nineteenth century who were city planners and engineers. Medicine not only espoused the values of cleanliness and improved living and working conditions, but it changed the focus to that of clinical interventions, screening and immunisation.

In biomedicalisation, the scope for intervention and surveillance is that much greater due to the power of information or ‘network society’ (Castells, 2000), the awareness of ‘risk society’ (Beck, 1992), and the huge expansion of biomedical research and work, including

‘new diagnostics, treatments, and procedures from bioengineering, genomics, proteomics, new computer-based visualisation technologies, computer assisted drug developments, evidence based medicine, telemedicine and telehealth, and so on’. (Clarke, 2003).

Risk groups identified in the era of biomedicalisation may include healthy people with a predisposition to certain forms of ill health as identified by their genetic profile, plus or minus their environment, their occupation, or their lifestyle. This would seem to match the criteria for the Huntington’s research clinic, where both healthy ‘at risk’

people are seen, and also people who are in the process of disease progression. The responsibility and emphasis of biomedicalisation towards self-care extends to all individuals, via various mechanisms to promote the achievement of optimum health and happiness through personal conduct including self-regulation, this approach being termed 'governmentality' (Clarke, 2003, p.165, citing the Foucauldian concept). In the Huntington's research clinic, the governmentality also extended to carers, when moral work was performed in the clinic by the researchers, and I will discuss the moral work within the theme of performance later in the thesis. Routine surveillance and testing such as in the Huntington's research clinic, therefore has resonance with the biomedicalisation project, which may include, in general social terms, both healthy people and the chronically sick on a far broader scale than before, with many people self referring due to perceptions of risk, and the cultural belief in the expanding predictive power of medicine. The application of biomedicalisation in the Huntington's research clinic has contributed to the evolution of a clinic where routine surveillance is utilised to create new knowledge on an international and global scale, again resonating with Clarke et al's theory (2003).

Routine research testing in clinical consultations

Clarke et al (2003) make the point that the increased capacity and reduced size of technology have led to the appearance of biotechnology on the desk, in the home and in the workplace. Home testing kits for blood sugar levels, blood pressure, allergies, and sometimes genetic predispositions are easily available and feed into the extensions of governmentality around health. It has become normal to be 'tested', even on routine visits to clinics and surgeries, there are myriad tests available to clinicians for quick results, so much so, that some medical establishments carry out routine testing or screening of patients who may attend for other reasons. The normalisation of testing is a well accepted part of medical participation, thus in the research clinic it can contribute to the blurring of care and research, with patients not being completely aware of the basis of their testing, and assuming it is all the same thing. This was certainly the case

in the HD research clinic, where non interventionist (non-drug trial) research took place.

There are few studies of the impact of routine research testing, it is often assumed to be harmless, but the case of genetic testing is slightly different due to the potential impact of the results. However, a US study found that neurologists used the relevance of genetics to the patients' condition as a sorting mechanism for the routine genetic testing (Browner & Preloran, 2010). Cost benefits were implicit in this study, so the neurologists' decisions were based on what routine testing would provide that was worth the cost to poor people. The authors observed the patient consultations and interviewed patients and clinicians, in a three year study. The sites of data collection varied from specialised centres to local clinics, and the participants were recruited via neurologists, who gave access to their clinics and patients. The point that the neurologists in the study made, concerned the relevance of genetics to the patient's condition or prognosis. They described the treatment of the patient being the same whether or not there was a genetic test carried out, and the prognosis being the same, yet they also acknowledged that 'knowledge was power', and patients could benefit from knowing the cause of their distress. On the benefit of knowledge, the clinicians were divided into two groups, one group believed that providing information which could not be acted upon, produced fatalism and passivity. The other group were more convinced that any diagnosis keeps hope alive, and that the information therefore has a value and contributes to patients' empowerment, in agreement with some of the patients. Again, this is representative of the blurring of research and care, or the blurring of the traditional values ascribed to them.

Clinicians were keen to keep hope alive for patients in the Browner and Preloran study (2010), despite their own feeling that to be unable to cure meant they were doing less than their best. Here we could write- 'less than their best.. in the patient's interest', to reflect the values of medical care rather than research. There is a parallel effect here with the Huntington's research clinic, where researchers carefully kept hope alive in order to maintain clinic recruitment. This presents a clear picture of the tension created for clinicians and researchers between knowing what causes a disease and being able to treat it, and being in a position to research a disease but not to treat it. The

researchers in the Huntington's clinic remarked that they had difficult feelings sometimes when faced with a very ill patient and a set of research exercises. This appeared to arise due to the 'emotion work' and 'feeling rules' which govern the clinical research situation (Hochschild, 1979). However, the Huntington's researchers did not hold dual care roles officially, apart from the lead clinician who had an honorary clinical care role. Hallowell et al (2009) do not indicate if they investigated feelings of conflict by researchers who did not hold an official healthcare role¹, i.e. were only involved in research. This may be due to the accepted practice of healthcare professionals conducting research because they have very direct access to patients. Alternatively, it may have been because the project was seeking to discover issues arising from the dual role held by researchers in their study.

It is also evident that scientists and clinicians sometimes find the balance between research and care is a difficult area to negotiate when they have to pick their way around the layers of rules and bureaucracy in order to carry out very routine and essentially non harmful applications. This can apply even within the research teams as the Mueller study illustrated (1999), where shades of opinion differ concerning rights of patients, the primacy of the research enterprise, and the priorities of the different research actors.

Hallowell et al (2009) examined the routinisation of research in clinical encounters in a cancer genetics clinic, with reference to routine genetic testing for BRCA genes in populations of women at high risk of breast cancer. The findings show that patients had difficulty in separating what was clinical care from what was done for research purposes. This was especially so when the procedures were concerned with data collection and epidemiology; this has a parallel in the Huntington's clinic study. Hallowell et al also discussed the strategies employed by researchers to create time and space between the two seemingly separate activities of research and care, effectively recreating boundaries between the two activities. This allowed them to manage what they experienced as a conflict of interest generated by the different roles they

¹ Only one lead researcher in my study had an honorary contract plus responsibility for clinical care. Those with only hon. contracts and no role in clinical care also experienced tension in the research role because they had to research people who were seriously ill. They had different ways of managing it.

themselves occupied within the clinic. Hallowell et al (2009) also make reference to the literature on patients' expectations from, and their understandings of, research, which are often at odds with research rationale and are confounded with the aims of clinical practice (Applebaum & Roth, 1982; Applebaum et al, 1987; Featherstone & Donovan, 2002, Lidz et al, 2004, Ponder et al, 2008). Hallowell and the authors of the paper assert that the literature on the subject of research and clinical care, suggests that this relationship is often ambiguous and potentially contested. However, they suggest that the perception of research having a therapeutic intent does not mean a misconception exactly, but indicates something of the construction of patient-clinician relationship.² It would appear from the Huntington's study that there is something to this, because participants held their relationship with the clinic researchers in great esteem and had built social bonds over several years of attendance.

Hallowell et al (2009) suggest scientific autonomy is protected by shielding researchers from outside interferences, such as care for instance. This suggestion echoes the point made by Mueller (1997) about the guidelines and policies which were enacted to avoid the intraprofessional differences in the AIDS research; in the end they protected the 'science view'. This according to Gieryn (1983), was a form of boundary work which ring-fenced the research activities and thus supported the pursuit of professional [research] goals.

Routine research is viewed as care

Easter et al (2006) report that research personnel are 'likely to be seen and see themselves as caregivers'. Their participants were physician-investigators, nurse-study co-ordinators, and patient-subjects, therefore all of them had experienced their dual roles in clinical and research domains. The authors hypothesised that the conflated relationship between the two areas can lead to the research being seen "erroneously- as individualised treatment", and they cite bioethics and social science literature as reaching similar hypotheses (Bamberg & Budwig, 1992; Brody & Miller, 2003; Churchill, 1980; Grunberg & Cefalu, 2003). Easter et al (2006) found that most patients

² This is an important rejoinder to Applebaum & Lidz.

considered the care they received in research as better than the care given by their usual health providers, in terms of clinical activities, psychosocial support, expressions of concern and close personal relationships with study team members³.

The researchers in the Easter et al study were sometimes conflicted between research and care, but mostly considered 'care' to be an integral part of research, and they considered that some of the questions posed about care versus research were 'unfair'. Easter et al (2006, p.708) speculate that "some of the phenomena we observed were strategies for resolving a role conflict, [and cited these authors]; Taylor 1992, Biddle 1986, Merton 1957". The authors of the study also express some concern that the 'special care' received via research may influence people to join and remain in studies. The paper concludes with the assertion that the moral frameworks for 'care' and 'research' are practised and received in 'similarity positions' citing (Brody and Miller: 2003, p.330). Importantly, Easter et al, make a recommendation that the ethical debates around the 'danger of confusing research with treatment' take account of the respondents' descriptions of the care they receive, and this would seem to be in accordance with a move towards more patient and consumer involvement in evaluation than previously.

The perception of research care as individualised medical care occurs in some cases (Easter et al, 2006), and this also happens in the provision of individual test results from research. This is a contested area between those researchers who want to provide results in those circumstances, and those who do not (Miller et al, 2008). Miller and her colleagues interviewed key informants in a Canadian cancer genetics research based testing programme. This was prior to the creation of the publicly funded clinical service in 2000. Their findings and conclusions were 1) that the communication of individual results made the research practices *seem* [italics in original] like clinical services for the respondents. 2) Whilst the respondents valued the way that research enabled a type of clinical access, the respondents considered this quasi-clinical service inadequate. (This is in contrast to the findings of Easter et al (2006) where research 'care' was perceived as better than that from normal care providers) 3) The respondents

³ In my study this was translated as having access to specialist treatment by elite professionals at the top of the research hierarchy and in the forefront of new knowledge.

recognised that their experiences were influenced by research imperatives, but interpreted the significance of this in different ways. Miller et al (2008) claim that the 'hybrid' state of research and clinical care in their study produced both inadequate care and insufficiently ethical research practices. They also suggest this raises questions about whether research should be made to serve clinical purposes, and suggest the need for further debate on the issue of individual research results (Miller et al, 2008).

The debates about the perception and operation of research and care have taken place gradually over the last ten years but appear to be increasing lately. The constant search for more research patients is one of the drivers of these debates, and one clinician recently suggested via a BBC webpage that care should be made conditional on patients taking part in research (Mathur, 2009). This position overturns the current ethical framework which confirms that medical care is not conditional on research participation.

Performance, practice and expectation

The research clinic as a site of performance, practice and expectation, is an interpretation which owes much to Erving Goffman and *The Presentation of Self in Everyday Life* (1959). Although arguably the research clinic is not simply 'everyday life', there are elements of performance and routine which render it mundane and ordinary. The interaction of the neurologist and the patients, sets the scene for further recruitment to research projects, and the clinic patient-participants and their carers create performances in the hope of gaining treatment, and in the hope of presenting themselves favourably in the research clinic environment. Goffman explained the importance of the setting (p.33), which can be assembled for a short time, or hired by someone with the money to do so. The virtual clinic in the HD study becomes a physical entity via such assemblage for one day each week or fortnight in a fairly new and distinctive setting compared to traditional medical settings; it lends itself to this imagined reality.

Goffman refers to the selection of 'front' that individuals need to select in order to conform to established social roles (p.36) and how this can cause difficulties if it does not fit the role exactly. Goffman's hypothesis is that the selection of 'suitable front' for a new task is usually from already well established fronts, but there can be a rather imprecise fit. This appears to be a relevant indication of the position of the clinic researchers, who are in the setting of care, but their roles do not provide this. The use of signage is also discussed by Goffman, to indicate the social role or variances from it. HD clinic researchers had the signage of their clothing; they did not dress in white coats or nurse uniforms and carry stethoscopes but ordinary casual dress indicating the absence of medical care. Dramatization of the role was also important to Goffman, and the HD clinic revealed difficulties with the dramatisation of the research role as opposed to the care role which was being sought by patients and carers.

Performance in the clinic was also enacted by patients and carers who attempted to idealise their roles, their activities and the way they were perceived by the researchers. This was not a coincidence, the clinic participants knew that their presentations held sway for the outcomes of the clinic visit, and they would participate according to the social norms in the hope of gaining a benefit in the form of treatment. There was also the performing of moral support of research by attending and acknowledging the research programme. To discuss the sub theme of moral support for research, a little later in the chapter I will consider the extension of the patients' and carers' roles into the research governance as portrayed by Novas; this falls within the theme of hope being created and maintained in the clinic.

Then and now: the different ways the disease is thought of

The clinic performances from all the participants are built around what they know to be the norms and expectations of hospitals, clinics, research, and the respective roles of themselves and others in these settings. The move to neurology has helped to shape new ways to think about HD, not only for the researchers in their direct engagement with the bioscientific objectives of the research, but also new ways to think about the disease for the patients and families. The comparison of 'then' and 'now' is made

easier for this study of the clinic by the existence of the old archived files which give a picture of an entirely different disease from that observed in the current research clinic. When they are examined retrospectively, medical records and files can portray what mattered at the time of the clinical consultation. There are references to what tests were made, what questions were asked, what information was given, how the patient responded to the consultation and to any subsequent treatment. Berg (1996) considers that the medical record is an important part of the focus for sociological research because,

‘the modern patients’ body Foucault has so aptly described is produced through embodied materially heterogenous work- and the medical record plays a crucial role in this production’. Berg extends this further ‘As organisational infrastructure, the medical record affords the interplay and coordination of divergent worlds. Seen in this light, as a site where multiple stories about patients and about organisations are at stake (including the interoperability between these stories), the medical record becomes highly relevant both analytically and politically’. (Berg, 1996)

Berg portrays the reading and writing work of medical records as artefacts which mediate the social relations that act and work through them (Berg, 1997). He displays the work of the medical record in three different situations via a series of vignettes from field work, putting forward the view that these other activities give life to the medical record because without them the record would not have any relevance. Firstly, the record is examined as it appears at the beginning of a clinical encounter between doctor and patient and in the process of medical decision making.

The second example shows how the medical record mediates the medical work in hospital wards, Berg explains how the writing of orders in the medical record transforms a patient into a ‘body manageable’ for the hospital routines. Thirdly, he illustrates how the practice of reading and writing produces particular renderings of patient histories (Berg, 1997). Berg’s point is that the medical records do not merely reproduce the clinical encounter, they are a constitutive part of, and ‘form a crucial site

in the sociotechnical organisation of medical work'. If they can be perceived this way, records can reveal changes in the sociotechnical organisation of medical work, such as that of clinics, wards and laboratories. In observing clinic work, it is possible to view the activities where the record is employed, referred to and re-created in the clinic and how these activities may be mediated by the recordings. In the first section where Berg considers the role of reading and writing the record in producing 'doctoring', he puts forward that we tend to concentrate on the intellectual qualities involved in studying to become a doctor, when in fact, tasks are often highly embodied activities involving the immediate surroundings. Continually returning to paperwork is one example of achieving cognition in the clinical encounter, it is part of a selective process of the talk and the written information which eventually produce a plan of action (Berg, 1996, p 505). Berg describes this process at length and in detail, as only one part of how the medical record achieves the transformation of the patient's condition from a non diagnosed state, into 'a manageable problem'. This is highly relevant to the HD research clinic where the performance of changing the type of information recorded has a long term effect on the organisation of the disease.

The reading and writing also provided a mechanism for controlling the doctor-patient interaction and were used instrumentally to shape the path of the encounter, the timing of responses, the breaks in talk, and the movement between stages of the consultation (Berg, 1996). Berg asserts in conclusion that the medical record can be seen as a type of organisational memory (cites Bowker, 1994), and as such cannot exist without a mechanism for forgetting. This selectivity is what makes the record work as it does in medicine, and observing what type of things are selected and deselected can explain a great deal about the type of work which takes place. Berg even refers to some restrictions on unstructured information leading to the limitation of social history information (1996, p.520). This is in contrast to the archived records of the Huntington's patients, which are extremely rich in social history detail, an aspect which was considered to make the records 'bad' by some bioscientists (Garfinkel & Bittner, 1967; Heath, 1982; Heath, 1996). During a seminar with some bioscientists in the course of this research project, I was asked why I was so fascinated by such 'bad examples' of medical records which were described as 'esoteric' and not truly

representative of 'good' medical records. The question itself revealed the preferences and 'cleansed' nature of the disease in bioscience where the brain activity is under scrutiny rather than the patient, the family and aspects of living everyday life.

It is unclear whether Berg considers the medical record as a technology in its own right, but what is certain is that the medical record can display the uses of technology in the clinic and the surrounding environment in detail, and can assert 'where medicine is at', in its production of the record of encounters over the years. This is particularly true over time, where a medical record may be an almost complete life history, or several histories where whole families were often involved in the case of genetic medicine.

Biomedicalisation as a means of extending the technology of the clinic

Clarke et al (2003) identify five key processes which,

“both engender biomedicalisation and are produced through it:

- 1) the political economic reconstitution of the vast sector of biomedicine
- 2) the focus on health itself and the elaboration of risk and surveillance biomedicines
- 3) the increasingly technological and scientific nature of biomedicine
- 4) transformations in how biomedical knowledges are produced, distributed and consumed, and in medical information management, and
- 5) transformations of bodies to include new properties and the production of new individual and collective technoscientific identities”.(p.161)

Each of these areas is dealt with separately by Clarke et al, who consider that biomedicalisation is the result of techno-scientific changes in biomedicine which are transforming the twenty-first century as much as, if not more than, medicalisation transformed the twentieth century. The main difference for the authors is the 'enmeshment' of technoscience with the human and non human body, and the intensity and scale of the transformations.

As well as the pointing towards the mass organisation of infrastructures to support and further develop biomedicalisation, Clarke et al underline the moral responsibility for self health and knowledge which is dominant in the twenty-first century, and which feeds a whole new extension of the industry of biomedicine, and creates new identities within society based on such things as categories of risk, carriers of genes, and tissue and organ compatibilities. This is such an overall concept that the authors term it ‘Biomedical TechnoService Complex Inc.’, and consider it a new social formation, something that is co-produced alongside the biomedical advances. According to Clarke et al, this makes their argument historical and not programmatic. I am unsure what this means exactly; that the new biomedical social form being co-produced was incidental, a quirk of circumstances, and not planned perhaps?

The authors identify the mid 1980’s as a time when the technoscientific innovations and new social forms began to have an impact on biomedicine from the inside to the outside. They claim that the industrial revolution framework was still evident when we developed ‘big technology’ and ‘big science’ such as electrification of transport links and the atom bomb. They also claim that beyond this, the technoscience revolution made ‘big science’ much more accessible to the individual, with big technology available on the desk top, in a small box, or even in the body. This shift is defined as being from enhanced control over external nature and the world around us, to transformations of an internal (biological nature) nature, often ‘transforming life itself’. They make the link between medicalisation being co-constituted of modernity, and biomedicalisation being constitutive of post modernity. The table of this shift is reproduced here for clarity. (Clarke et al, 2003)

Table 3. from p168-169 American Sociological Review, vol 68, 2 April 2003.

Table 3. The Shift from Medicalization to Biomedicalization

Medicalisation	Biomedicalisation
Control	Transformation
Institutional expansion of professional medical jurisdiction into new domains	Expansion also through technoscientific transformations of biomedical organizations, infrastructure, knowledges, and clinical treatments

Economics:	<i>The US Biomedical TechnoService Complex Inc</i>
Foundation and statefunded (usually NIH), Biomedical, scientific and clinical research with accessible/public results	Also increasing privatization of research including university, Industry collaborations with increased privatization, And commodification of research results as proprietary knowledge
Increased economic organization, rationalization, corporatisation, nationalisation	Also increased economic privatization, devolution, transnationalisation/globalisation
Physician dominated organisations	Managed care system-dominated organisations
Stratification largely through the dual tendencies of selective medicalisation and selective exclusion from care based on ability to pay	Stratification also through stratified rationalization, new population dividing practices, and new assemblages for surveillance and treatment based on new technoscientific identities
<i>The Focus on Health, Risk And Surveillance</i>	
Works through a paradigm of definition, diagnosis, (through screening and testing), classification and treatment of illness and diseases	Works also through a paradigm of definition, diagnosis, (through screening and testing), classification and treatment of risks and commodification of health and lifestyles
Health policy as problem solving	Health policy as problem defining
Disease conceptualized at level of organs, cells	Risks and diseases conceptualized at the level of genes, molecules, proteins
<i>The Technoscientisation of Biomedicine</i>	
Highly localized infrastructures with idiosyncratic physician, clinic, and hospital records of patients (Photocopy & fax are major innovations)	Increasingly integrated infrastructures with widely dispersed access to highly standardized, digitized patient medical records, insurance information processing, and storage
Individual/case based medicine with local (usually office based) control over patient information	Outcomes evidence based medicine with use of decision support technologies and computerized data banks in managed care systems
Medical science and technological interventions, (e.g. antibiotics, chemotherapy, radiation, dialysis, transplantations, new reproductive technologies)	Biomedical technoscientific transformations (e.g. molecularisation, biotechnologies, geneticisation, nanoscience, bioengineering, chemo-prevention, genetic engineering, and cloning)
New medical specialities based on body parts and processes, and disease processes (e.g. cardiology, gynaecology, oncology), assumed to be universal across populations and practice settings	New medical specialities based on assemblages- loci of practice and knowledge of accompanying distinctive populations and genres of sciences and technologies (e.g. emergency medicine, hospitalists, prison medicine)
<i>Transformations of Information, and the Production and Distribution of Knowledges</i>	

Professional control over specialized medical knowledge production and distribution, with highly restricted access (usually limited to medical professionals)	Heterogenous production of multiple genres of information/knowledge regarding health, illness, disease and medicine widely accessible in bookstores and electronically by Internet etc.
Largely top down medical professional- initiated interventions	Also heterogeneously initiated interventions (e.g. new actors include health social movements, consumers, Internet users, pharmaceutical corporations, advertisements, websites)
<i>Transformations of Bodies And Identities</i>	
Normalisation	Customisation
Universal taylorised bodies; one size fits all medical devices/technologies and drugs; superficially (including cosmetically) modified bodies	Individualised bodies; niche marketed and individualised drugs and devices/technologies; customized, tailored and fundamentally transformed bodies
From badness to sickness; stigmatisation of conditions and diseases	Also new technoscientifically based individual and collective identities

Clarke et al consider that the table is a historical cumulative representation to be read from left to right and not an 'either/ or'. They point out that the transformation from medicalised to biomedicalised is not wholly even, both aspects can occur at the same time, and the shifts are of emphasis rather than complete changes. This seems to negate the previous point about the links between the two different regimes as being of 'modernity' and 'post modernity'. In effect, the industrialisation of biomedicine in terms of its scale and scope could be interpreted as being of modernity, rather than post modernity.

The only difficulty with Clarke's table and theoretical position is that chronic, disabling, terminal conditions are not specifically mentioned, and these play an important role in some of the changes to medicine's repertoire of the early twenty-first century. The position of biomedicalisation as shown or described here suffers a little like that of Parson's- there is no 'category' for those who will not recover. Clarke et al describe the imperative of moral responsibility to properly manage one's chronic illness (cites Strauss, Corbin et al, 1984), and this is the closest the theory gets to accommodating the chronic long term illness patients. Although the authors do not explicitly claim that in chronic illness this is an extension of governmentality in the Foucauldian sense, it would seem to be implied.

Another area where biomedicalisation may have had influence is on the carers of the chronically ill, where they consider themselves to be accountable for the patient at an almost professional nursing care level, in a further extension of governmentality.

Biomedicalisation, chronic illness and biographical disruption

Biographical disruption was revealed in the HD clinic throughout the 'clinic talk', when patients or carers would perform the biographical accounts of the patient's illness. The disruption continues and increases over the patient's remaining years of life, and often there is disruption to the carers' lives and those of other family members.

A thorough discussion of chronic illness, its place and meaning in contemporary society points out that there is a relationship between the changing demographics of modern societies, age, chronic illness and disability (Bury, 1997). It is ironic that the levels of disability increase with age, yet population age increases with the improvement of health care in earlier life, plus the advances in treatment of infectious diseases and the expansion of additional care for conditions such as stroke and arthritis. Bury points out that there is also a shift from cure to care, with the separation of social and welfare care from medical treatment, and specialist medical services replacing what was formerly known as 'geriatric care' (1997). This has major implications for the policy and provision of suitable care in contemporary society, the majority of people who require long term care are older chronically sick people (Bury, 1997, p115). However, this leaves a gap where younger chronically sick people may not 'slot in', and there has been criticism of the labels and research around disability issues for younger people, together with a paucity of state provision in the UK. Conversely, the maintenance and improvement of one's own health has gained momentum, and public health issues tend to look at the consequences of diseases, rather than finding explicit causes as in previous eras. Bury explains that since the 1970's more explicitly sociological work has been carried out on the meanings of disability both in the UK and the USA (Blaxter, 1976; Strauss, 1975, both cited in Bury, 1997, p.121). Both the US and UK studies found that the most intractable problems for the chronically ill were found in wider social issues linked with their medical conditions.

Three areas were identified, 1) biographical disruption, (Bury, 1982) in part due to the modern culture of expectation of long life and health, 2) the impact of treatment where relevant, and 3) long term adaptation and management of illness into normal life (Bury, 1997). These three areas remain pertinent to the biomedicalisation agenda thirty years on, and there is no sign of a concerted effort to streamline health policy and care provision for people in these situations, which is patchy. If anything, faith in science appears to have increased, with each major breakthrough in biomedical knowledge being hailed as the answer to all chronic and fatal illness. This creates an added imperative for the chronically ill person to seek out the 'right treatment', and be proactive in pursuit of it. For conditions with no treatment or cure yet available, this also leaves a gap in the biomedicalisation agenda, which is waiting to be filled with something which will restore personal identities from the biographical disruption of something like Huntington's disease.

A more recent research project considered the biographical disruption of not chronic illness but a terminal condition, Motor Neurone Disease (Locock et al, 2009). This progressive neurological condition takes sufferers through loss of mobility, speech and breathing capacity, and death usually occurs within five years. The biographical disruption applied to chronic illness is re-interpreted by the authors as biographical abruption, taking into account the patients' views that a diagnosis means life as they have known it, is over. There is no lengthy time frame in which to create a repaired biography although some patients did attempt to restore normality and control (Locock et al, 2009). However, the time frame of Huntington's disease is variable from diagnosis or first symptoms to the terminal stage; it can be relatively few years, or in some cases it may be twenty years plus. This further deepens the uncertainty of biography in these cases, and underlines the continuing uncertainty of biomedicine which does not yet have a solution. In earlier work on biographical disruption, Bury (1982) noted that this uncertainty did not lead to disillusionment with medicine, although the limits of medicine at the time were appreciated by patients. This is also a continuing feature of biomedicalisation in the twenty first century, with the appearance of hope as a by-product of biomedical advances such as DNA sequencing and stem cell technology (Martin et al, 2008).

Performing the technology of chronic illness monitoring

In his work on surveillance medicine, (Armstrong, 1995) argues that the spaces for illness were becoming re-mapped to include problematisation of normality, the redrawing of the relationship between symptom, sign and illness, and the resiting of illness outside the corporeal space of the body. These features are evident in the epidemiological research programmes surrounding the HD clinics, and the surveillance project now extends further in creating global communities of data gathering and sharing in order to further the meanings extracted from surveillance of identified groups of chronically ill people. In the HD research clinic, this monitoring has extended to include people who are healthy, but who carry the gene for Huntington's disease, effectively turning them into patients before there are any outward signs of the condition. There is some consideration in the bioscientific research that although outward signs of the disease are not obvious until the middle years of life (approximately between the ages of 35 and 55), the neuronal degeneration may begin at birth or soon after. This may eventually bring about some form of very early detection regime in those thought to be at risk; the earliest way to detect the disease is pre-implantation genetic diagnosis, and this is often used in families where there is a risk of HD. Recent developments in the investigations of an accurate population incidence of HD have speculated that the true rate in the general population could be twice as high as previously thought, perhaps 14 or 15 per 100,000 people. Together with the notion that the brain degeneration begins shortly after birth, would this instigate blanket screening of neonates? If so, it would be highly contentious given the present policy of not performing predictive testing for HD under the age of eighteen years old in non symptomatic cases. Would a screening programme for neuronal degeneration change the status of testing for children and young people to disease testing instead of predictive testing, and create patients during the thirty or so years of what would have been normal life, had no testing been carried out? All these matters are brought forward by the increase of surveillance medicine in this particular environment of biomedicine.

Armstrong describes the progress and development of Hospital Medicine, and Foucault's account of the new spatialisation of illness, as being a type of cognitive mapping of symptoms noticed and reported by patients. Added to this experiential information was the skilled clinicians' observance of signs during clinical examination which then led to the knowledge of the underlying disease (1995). This element of clinical practice has remained the same with the added refinement of new technologies to look beyond the clinicians gaze, into the body, into cells, molecules and genetic material. Armstrong also refers to Jewson's argument (1976) that the addition of a Laboratory medicine model at the end of the nineteenth century, further objectified the patient body and obscured the identity of the patient. This appears to line up with the views of Mueller's physicians in the 1997 study, that the information is a representation, objectified and in no way linked to the identity of the individual person/patient, and therefore there is no moral obligation to inform or seek consent to share data. In the HD research this is one possible interpretation of the extended use of the large global collections of data which are partially anonymised at the point of collection by means of a bar-coded identity link. The data is then objectified further when it is stored in mass international repositories and retrieved as simply data at any future date; it is completely disembodied from the person to whom it relates. This has already occurred in the HD research as one researcher told me. We were discussing the publication of research based on data from the international data base, and if this gave the researchers any sense of international involvement. The reply was that it was impossible to tell if any of the local data had been used, and this was a little disappointing for the researchers. The use of disembodied standardised information may be the very best use of data in order for science to develop treatment for rare and fatal diseases, however, it risks creating a model disease that no one recognises as a 'real' life representation. In order to consider what the influences are which determine how diseases are modelled and thought of, it may be relevant to briefly consider the work of an early sociologist of medicine, Ludwik Fleck, who wrote in 1935 that it was necessary to create a 'thought collective' in order to bring about acceptance of scientific facts.

Ludwik Fleck and Thought Collectives

Ludwik Fleck's little known 1935 publication has become better known since Thomas Kuhn mentioned it in his 1968 work. At the time of publication 'Genesis and Development of a Scientific Fact' (Fleck:1935), was not considered to be worthy of major review or comment. Fleck's work had created a synthesis of the sociology, history and philosophy of science, but according to later translators of the work, it may have been thought of as 'another historical case study in medicine' (Trenn:1979, in Fleck (1979) preface p.xvii). dimension.

Fleck used the template of the science which uncovered the cause of syphilis as bacteriological and established the Wasserman test, to show that 'scientific facts', are socially created, and not the products of single discoveries by individuals. One of the fundamental components that Fleck considers to be vital for the emergence of a scientific fact, is the existence of a 'thought collective' and a 'thought style'.⁴

Kuhn wrote that on re reading Fleck he realised there were areas he had not understood and several he still found fault with, including the idea of 'thought collectives'. However, the fundamental idea remained, that scientific facts are created by a complex process of social consolidation. In the Preface to the 1979 book, Trenn (1979) explained how interest in Fleck's work came much later than the interest in Karl Popper for example, who published at almost the same time.

Ilana Lowy on Ludwik Fleck

The literature on transmission of medical knowledge has a long history but has not appeared until relatively recent times in medical sociology according to Lowy's paper on Fleck, a Polish-Jewish physician (1988). She cites Fleck (1935) as being unusual for his time in that his contribution was an early development of the idea that scientific facts are constructed, rather than the prevailing positivist view of the 1920's and

⁴ This is what Kuhn finds misleading and a source of tension in the text. The extrapolation of individual thought style/collective onto a group who all possess it/ same thing. There is an alternative interpretation of Fleck though..

1930's, that scientific facts lie waiting to be discovered. Lowy describes his work as being virtually unnoticed, and only rediscovered after publication of *The Structure of Scientific Revolutions* (Kuhn, 1962), where Kuhn credits Fleck's 1935 work "Genesis and Development of a Scientific Fact" as a major influence on his own thought.

According to Lowy, Fleck, an immunologist and bacteriologist, had strong sociological interests throughout his medical training, and he was convinced that the social process was instrumental in the development of science. He explained the emergence of scientific facts via "distinct 'thought collectives', each composed of individuals who share a specific 'thought style'. He also used a great number of medical examples in his work, and yet seemed to have been considered as more of a generalist in the field of the sociology of science (Lowy, 1988). Lowy takes this up and analyses his views, selecting ideas which may be of interest to medical sociologists today. In this pursuit, Lowy acknowledged that her presentation was not completely faithful to Fleck's original intentions, but asserted that his ideas are of relevance to "studies of interactions between fundamental research and its practical applications in general, and more specifically to studies of the development of medical knowledge" (Lowy, 1988, p.139). The organisation of 'thought collectives', was not without disadvantages for Fleck, the training required to enable a specialist to 'see' disease correctly within the specialist domain, necessarily left out the ability to 'see' in all other medical specialities and domains. A contemporary equivalent situation would explain the need for inter and multi-departmental work to treat and cure complex diseases.

Fleck also referred to the way that society constructs disease as 'ideal, fictitious pictures, known as morbid units', and he thought it was impossible to reduce complexity and individual pathology to physics, chemistry or biology. He considered that it was impossible to elaborate a global medical theory which allowed for the understanding of all the observed phenomena (surrounding a disease?), and therefore it made it impossible to have a single approach to disease.

This idea of the complexity and subtle differences in disease pathology making a single approach impossible, are important when it is considered that there is currently a global approach to research on Huntington's disease. The shift in emphasis from genetics to neuroscience for Huntington's is connected with the global multi-research

approach, the translation of scientific facts and the emergence of new ideas and ways of categorising. This is the result of multi-cultural research co-operation, both nationally and internationally, with communities of different 'thought collectives' (Fleck, 1935) working together on some level, including, neuroscience specialists, epidemiologists, psychologists, geneticists, bioscientists, neurosurgeons, towards treatment or cure. Fleck could have been describing this when he wrote in 1935:

"The intercollective communication of ideas always results in a shift or a change in the currency of thought (...). The change in thought style, that is the change in readiness for directed perception offers new possibilities of discovery and creates new facts". (Fleck, 1935, 109-110, cited in Lowy, 1988). Lowy's final point about Fleck is that many of his ideas can be tested with sociological methods, and Kuhn (1979) considered that 'Fleck opens avenues for empirical research'. It is possible to apply some of these ideas to the Huntington's research clinic and the social and scientific developments around it; Fleck considered that the formation of medical knowledge was dynamic, time dependant and involved society as a whole, it was not limited to medical experts (Lowy, 1988).

It is important to say that Fleck has his detractors. There is always another point of view, and Hedfors (2007) provides a thorough account of alternative theories to account for Fleck's apparently unusual position at the time his monograph was published. Hedfors contests that Fleck's bioscientific methodology was rather suspect even in terms of the difficulties of science during both pre-war and war years, and contests the apparent acceptance of other sociologists of science that Fleck was an important and competent scientist in his own field. This is not the main point of mentioning Fleck and so I will curtail the discussion here, except to say that this appears to be a rather polemic view of Fleck which asserts that because his science was average at best, he has no real value even in social studies of science, and may indeed have had seriously flawed motives when he was involved in the Nazi typhus experiments in Buchenwald (Hedfors, 2006).

Fleck's account of translation

However, Hedfors (2006) also has her critics, who strongly dispute her account of Fleck's motives and activities (Amsterdamska et al, 2008). This multi authored paper includes several scholars of Fleck, including Lowy, Bonah and Schnelle, who have all produced highly detailed accounts themselves, and who refute completely Hedfors' suggestions of Fleck's full and willing co-operation with Nazi experimentation.

Bonah (2002) explains in great detail the relevance of Fleck's theory of thought collectives, and places it in the context of 1930's Germany and two crises affecting modern medicine at that time. Fleck and a contemporary Julius Moses, were writing in the wake of the Lubeck BCG crisis of 1930, but came up with different answers to the perceived dilemma of medicine- that of the opposition between a patient's wellbeing, and the production and application of scientific knowledge. Moses, according to Bonah, found the 'system' of medicine to be at fault and the subsequent decreasing confidence in the medical profession and research was the second crisis. His solution was to put forward guidelines for the ethical conduct of research and practice; Fleck's pursuit was to propose a deeper, more democratic understanding of science and knowledge by means of detailed examination of how science functions and establishes facts. Bonah cites Fleck as follows: "Popular exoteric knowledge stems from specialised esoteric knowledge. Owing to simplification, vividness, and absolute certainty it appears secure, more rounded and more firmly joined together". (Fleck, 1935, p.113)

What Bonah is claiming, is that Fleck could appreciate the social process which elicited facts as scientific, in addition to the extension of communication which transmitted and translated those scientific facts outside the thought collective, which inevitably led to transformation of meaning. This may have relevance for the transformation of Huntington's disease from a genetic incurable illness to a set of neurological symptoms. With this new definition of HD, increased hope in the power of technology now and in the future is transmitted outside the scientific environment and is taken up by patients and families, and I will now look at the theme of the creation of hope in technological research via the promised futures of medical stem cell technologies, or translational research.

Current views of translational research

More contemporary work on translational research often uses the term ‘bench to bedside’, (Lowy, 1996; Wainwright et al, 2006; Rubin, 2008), and also includes the reverse trip from bedside to bench (Martin et al 2008). In Lowy’s book, this phrase ‘bench to bedside’ is in the title, and indicates the multi-dimensional team of clinicians and scientists who together attempted a clinical trial for a cancer drug interleukin-2, between the years 1987 and 1992. Lowy explains there could be “two diametrically opposed accounts” of this clinical trial; she uses the metaphors of ‘black and white accounts’.

The white account would extol the successes of the trial, and line up with the trial organisers views, and would present the project as a major contribution to cancer therapy, and a breakthrough in terms of co-operative working between clinicians and scientists. It did lead to some remissions in some selected patients, and although IL-2 was not an efficient ‘cure’, it would be presented as a significant contribution to the cure. This account would be key to having fostered mutual understanding of the two professional group’s different concerns, and paved the way for increased preclinical and clinical research at the institute, plus development of projects modelled on this IL-2 trial in other western countries, particularly the US. The professional benefits from this major success would be considered appropriate reward for the diligent work and organisational skills in making such a venture successful. Interestingly, Lowy put forward an alternative ‘black’ account which looked rather more deeply at the results of the IL-2 trial. This, she asserted,

“would reject the organisers’ analytic categories and viewpoints. The exemplary collaboration among clinicians, scientists and industrialists claimed by the organisers would be framed instead in terms of self-interest and self-promotion that increased the profits of industrialists and the glory and power of selected physicians and scientists; the Cancer IL-2 trial was objectively shaped by personal ambition, however sincerely the major actors may have believed that they were motivated solely by scientific

curiosity and the desire to improve the lot of cancer patient. One should always ask who benefits and who loses in a given clinical experiment. Moreover, a dispassionate look at the results of the IL-2 trial in the years 1987-1992 reveals a striking gap between the significant professional gains enjoyed by some of the organisers and the limited benefits to the great majority of patients. The existence of such a gap should make us wary of accepting the actors' statements and declared beliefs at face value".

(Lowy, 1996, pp 9-10)

This warning from Lowy serves as a reminder of the various lenses through which clinical trials and research of any kind may be viewed. It also acts as a reminder to consider sociologically, the social, economic and political frameworks in which research is made possible in late modern society.

Recent sociology of science perspectives on the nature of research translation are often concerned with the utility of laboratory discoveries and the 'promise' of their availability as treatments for example (Wainwright et al, 2003; Martin et al, 2008) and I will return to the discussion of this now.

Hope created by future promise

The existence and operation of the Huntington's research clinic is itself viewed as a source of hope and benefit by patients and families. The involvement of groups outside medicine in support of research has increased the imperative and momentum of research in some cases (Novas, 2006). The discussion for Novas centres on the activism involved in patients' organisations which become central to the research process and encourage the biovalue of patients' blood and tissue, in a manner which he terms 'political economy of hope'. He also charts the types of involvement which tend to lead to increased activism, from gaining disease knowledge, support group formation, then fund raising and in some cases, influencing the direction and acceptance of research proposals. Novas considers that this 'political economy of hope' is a contributor to the 'transformation of the field of contemporary biopolitics'.

This certainly has links with Fleck's view that new knowledge is a social process created by all of society and not just scientists. Novas claims that,

‘through becoming involved in the promotion of health and in the generation of biovalue, patients’ associations contribute to the elaboration of norms relating to how biomedical research should be conducted and how its therapeutic and economic benefits should be distributed’ .

(Novas, 2006, p.291)

There is also the issue of shaping the social norms in relation to research, by means of becoming very knowledgeable about the disease, and then seeking to match this knowledge with relevant research applications which are already underway. The influence of the patient groups can be considerable if they find that there is no research underway or if the research is focussed differently to the patients’ priorities. Novas describes the activities of PXE as a case study where the patients organisation founded the biobank of tissue and blood, in order to facilitate the research (2006, p.296). This is not the case in the Huntington’s research, the instigation of the multinational research has come from a non profit making organisation, but the model of activity is very similar- it is a preparation ground for data banks ready for experimental medicine. Having said that, it is possible that the non profit making organisation has its roots in patient activism. The organisation CHDI, is fairly circumspect and does not give details of its donors, but does give details of its employees and management structure. Novas’ concluding points concern the fact that the hopes of patients and families act as attempts to shape the future health of populations, and in being so focused on the future, abandon the current needs of the people presently affected with the illness (Stockdale & Terry 2002, cited in Novas, 2006, p.302).

Hope in the promise of future stem cell technology was something very evident in the HD research clinic and several authors have discussed the relative optimism and uncertainty associated with this. Ever since the stem cell stories began to appear, the possibilities for replacement tissue, organs and body parts have become part of the expected future for many people whether they are ill or not. The stem cell has become

the provider of new organs to prevent the shortfall in transplants, it has become the replacement body part, the bio-engineering of the cell to repair the body processes, there is a great deal of hope attached to the name, despite the very few processes which actually take place. The examples of such technology are still so rare and experimental as to be very news worthy, yet this does not seem to dampen the public's enthusiasm. The phenomenon of the use of hope by science is the subject of Nik Brown's work on *Reconnecting Regimes of Truth and Hope* (2005). Brown has identified that there is a change in the public consumption of science because instead of reliance on what is known now, interest and importance is centred on the future of biotechnologies which are as yet unrealised. This has resonance with the hopes expressed by the HD clinic participants, the majority of whom wanted to have the chance of stem cell transplantation to delay or prevent the progression of Huntington's disease. Brown indicates a broad rebranding of the bio-industries towards a more romantic future view of life. Brown claims this has been instrumental in changing the politics of the biosciences, and that this has influenced areas of biomedicine especially in the development of cord blood banking. As an example, cord blood banking contains all the elements of romantic future promise, and it encourages parents to plan for an unknown future, 'just in case', something ill befalls their new child.

Hope as a motivator has also been utilised by psychosocial medicine in oncology as a measurable indicator of how cancer patients can mobilise themselves in overcoming the disease. Hope is being constructed here as an ontology, something that exists and has influence, (Herth, 1992, cited in Brown, 2005). There is even a name for the entity-oncohope. This has not been translated directly or purposely onto the neurological research framework for Huntington's disease via the research clinics, but its time may arrive. Patients and families do lay claim to the importance and existence of hope as a motivator in their daily struggles with the disease. Brown makes an important point about the utilisation of hope in oncology, in that it transfers responsibility and has a disciplinary effect, and this was in some subtle ways present in the HD research families' discourses. It is probably too soon to assess properly whether or not 'neuro-hope' has any disciplinary effect on Huntington's patients and carers, but it would seem

to be at least present from the hopeful discourses presented both in the clinic and outside it.

Conclusion

This chapter has discussed the three themes emerging from the research clinic data, the blurring between research and care in the clinic, the performances enacted in the clinic which are connected with research and medical practice and patient experience of the disease, and finally the use and placing of hope in the research context. The themes are part of the current sociological literature, and recent research which seek to reveal some of the social mechanisms underlying current bioscientific research. In addition, the re-categorisation and movement of Huntington's disease into neurology instead of remaining in genetics, has added an extra point of discussion to the chapter because this concerns how disease is organised and for whom, and what purpose. A further development of Huntington's research has been the move to global scale data collection in readiness for experimental medicine, and this has influenced the way Huntington's disease is presented and thought of in neurology.

The themes also bring forward some important questions concerning how medicine and research are accomplished in the twenty-first century; for instance, does the 'blurred boundary' between care and research indicate that care is no longer the primary driver of a combined clinic which is funded by research? In terms of the second theme of performance and expectations, does the existence of the research clinic add to expectations of treatment and cure which surround the patients' visits and is this officially acknowledged as an effect of the neuro-scientific research? Does the clinic add weight to the expectations of the researchers about how quickly translation into treatments may be achieved? Finally, how do the researchers prioritise and combine the day to day research activity with the often unexpected 'emotion work' and 'feeling rules' (Hochschild, 1979) which often surface during patient consultations? The management of patient hopes appears to be an important factor in maintaining research momentum and building capacity. The themes and associated questions reveal the prevailing social structures of medicine and science, and illustrate the pace of change in

research, where almost all existing guidelines for patient care trail in the wake of the bio-scientific project.

The next Chapter, four, is the first of three data analysis chapters and discusses the analysis of the data in relation to the theme of ‘blurring’ of research and care.

Chapter Four

Blurring the boundaries of care and research

Introduction

In this chapter I discuss the issue of ‘blurring’ which occurred in the research clinic due to the simultaneous activities of clinical care and research. There are suggestions that the two activities are so separate, have such different priorities and aims, that there are distinct differences between research ethics and the ethics of clinical practice (Miller & Brody, 2002). It is undoubtedly important to ensure that patients are fully informed as to what their research data may be used for, and aware of when they are being medically treated and when they are taking part in research (see Declaration of Helsinki, 1964), for all the reasons of medical misconduct that caused the ethical treatment regulations to be created (Lemmens & Miller, 2002).

Enshrined in the ethics of research participation are the notions that all research participation must be voluntary, that no particular treatment benefit should be expected from research especially in random controlled trials, and that patients may exercise choice at any time about their participation. However, when clinical care and research take place simultaneously, there are areas of tension present due to the conflicting aims of the two activities. However, it is debatable whether the two areas are as different as some authors seem to think. Freedman (1987) argued for a position of ‘clinical equipoise’ which enabled clinician-researchers to reconcile their therapeutic obligations with commitment to scientific standards. Lemmens and Miller (2002) advised against setting research and clinical care against each other in a ‘Jekyll and Hyde’ approach. This chapter will show how the blurring between research and care occurs in the research clinic, and some of the ensuing tensions for researchers, patients and carers. I will use an example of a particular type of recruitment into an allied project, to illuminate some of the advantages and disadvantages of the mixed research and care approach, and to make visible the operation of the clinic. However, before this I will give some background information about the movement of the disease to neuroscience from genetics, and the routine of a clinic day. Both of these, the positioning of the disease in a different specialism, and the research clinic routine, were important factors in creating the conditions for blurring to occur.

From Genetics to Neuroscience

The beginning of the fieldwork coincided with the reorganisation of the Genetics department and the re-allocation of Huntington's disease management patients to the Huntington's Neuroscience Research clinics.

The previous arrangements for the management of Huntington's patients had evolved over thirty years, during which time, specialist clinicians and nurse counsellors had built up supportive networks with patients and families. This included contact by phone for advice on day to day issues, home visits if necessary, and a safe environment for information and worry sharing. All management of the disease would be dealt with in future by the Neuroscience research team. The Genetics department would concentrate on genetic testing of people at risk, and counselling of patients and families. Huntington's patients and families in need of disease management in terms of drugs to mitigate effects, or advice on day to day matters would be offered the neuroscience HD research (and now management) clinic rather than the genetics service. The only difficulty with this strategy was that the neuroscience research department was not created to manage the condition and support patients and families; it was created around the research and there were no additional resources available. This left something of a 'gap' in the services to patients and families.

What was already in place, however, was the research clinic where the disease progression was being observed in a similar way as it would have been in a management clinic, using similar neurological tests and cognitive assessments. It was evident that this appeared to be a 'doubling up' of similar practices, even though the basic aims of each practice, research and disease management, were completely different. The thought of no clinic at all for the Huntington's patients was considered the worst outcome by many, including the disease support groups, clinicians, and the Huntington's Disease Association which is a charity for Huntington's families. The neurological research received funding from an outside organisation CHDI Inc., in the USA, to become part of a European wide project to collect epidemiological data on thousands of Huntington's disease patients in many countries. The move of Huntington's disease into neurology may have proved to be of some benefit to the research project, but there was no provision for disease management. This was a difficult issue, research does not usually provide medical care, this would usually have come from the NHS. However, there is no standard treatment for the disease, simply palliative drug treatment in some cases, but by no means all. Disease progression results in

death after fifteen to twenty years. In addition, patients needed to be able to 'opt in' to research participation voluntarily.

The following paragraph is an extract from my field notes of a meeting between the lead researcher, a genetics colleague and myself at the start of the project.

(Extract from field notes, late 2006)

Whilst we waited for the Professor in her office we had coffee, she had been delayed in a meeting. She arrived and apologised profusely, explaining to us that she had been in an unscheduled meeting about the clinic. A letter had been discussed, which she handed to the Genetics colleague. She explained that some people thought that it was not appropriate for Geneticists to be dealing with the management of HD via their clinics, and so with very little notice and no resources or arrangements, the management of the condition would pass to the Neuroscience research clinic. The brief letter was the only official communication she had had about the situation.

The effect of this is that affected HD patients who normally attended the Genetics based management clinic were referred to see the neurologist about their condition, and asked if they wish to take part in research. It was explained that the procedure would effectively be the same as if they were coming to a management clinic, but the information gathered would be entered on a database for research purposes, if they agreed to this. The basic European Registry Project is a longitudinal epidemiological study, which collects data according to the Universal Huntington's Disease Rating Scale (UHDRS). The aim is to build on the accumulated data to give a picture of the 'European type(s)' of HD. I have elaborated on this project in more detail in the discussion chapter, but as well as being an epidemiological study, the European Registry Project is a gateway into other projects including drug trials and potentially invasive brain surgery with transplanted foetal stem cells.

The movement of Huntington's disease from genetics into neuroscience, paved the way for the dual role clinic to take place, and brought the hitherto separate practices of research and care into the same clinic environment, simultaneously. Another possibly unforeseen effect of this, is the earlier creation of 'patients'. Tibben (2002) has written about the fundamental differences between a neurological consultation and a genetic one. People at

risk may prefer to see a geneticist because before they are clinically diagnosed they remain 'at risk' gene carriers. Tibben considers this is preferable to some people because it allows the admission of being a patient to be delayed. When all persons at risk are seen in the neurology research clinic, they are all patients, even though some of them may be pre-symptomatic. There is a separate research programme available for pre-symptomatic people, Predict-HD, which is designed to seek out the very earliest signs and symptoms of disease in the pre-symptomatic population. Previously, there was reliance on the geneticist's clinical opinion, and in some cases, this would have delayed the status of 'affected patient' for some considerable time allowing the person to continue their life unaffected by diagnosis for the time being. The next section will discuss the clinic routines, the main neurological consultation, and recruitment into other studies at the clinic.

Routine, consultation and recruitment

About an hour or so before the first patient was due to arrive, the various researchers who worked at the clinic together with the neurologist gathered in the neurologists consulting room for a pre-clinic meeting. This included psychologists, perhaps a physiotherapist, a neurology research nurse, perhaps a Huntington's research nurse, with myself as the observer, and the HD charity support worker who had been invited to attend the clinic. This was where information was shared about patients, those who are expected on the day, and additionally those who will not be attending further clinics, either because they have become terminally ill or died.

The neurologist usually looked at the files on her desk and explained something about each patient who was due to attend. The patient's last visit may have been discussed, and other relevant topics such as how long ago it was since they were seen, what issues they brought to the clinic last time, how they were advised regarding drugs and care, which research projects they are enrolled in, and importantly if they might appear to be suitable for further recruitment today. The recruitment was not ad hoc, it was guided by the knowledge of the researchers about the patients, and decisions were made concerning who would approach people.

The other researchers will add their own pieces of information concerning the day's patients sometimes this is about the research, and sometimes about levels of co-operation. These are pieces of shared tacit knowledge of patients and the illness; but it is not clear exactly how this informs the application of the tests administered in research. It appears to

forewarn researchers about the level of the tests to be taken, especially where information is shared about patients not wanting to take part in certain parts of the research, such as the cognitive testing. The extent and level of cognitive and movement testing depended a great deal on the disease stage of the patient; carers reported that the testing 'got less' over successive clinic visits as the patients became more disabled, until it was 'just a chat'.

Researchers also shared information about the personal attributes of patients and the level of support patients had from their carers. This appeared to have some indirect influence on recruitment to other studies. An example from the field notes illustrates this form of talk.

Extract from field notes 2007:

"...the next one is falling a lot. Balance ok, very choreic, still working. Has been resistant to medication, but would be good to get him into the physio study 6 months before he takes the meds. Ring at my request, ask if they want another visit pre-12 month recall. He's a very conscientious man, he won't take sick leave to visit the clinic, he takes holiday."

The above extract is descriptive of the patient's physical problems, his attitude to work, and his resistance to medication, perhaps indicating that the person is trying to maintain 'normal life' for as long as possible. I discuss this aspect further in a later section of this chapter, the moral and sentimental work done by researchers which added to the descriptions and identification of patients. However, the extract clearly shows how the recruitment of this patient is being managed together with his clinical care. The suggested order of the 'physio study' and the medication perhaps 6 months later, was an indication of the tacit planning and organising of recruitment in this case. The recruitment was influenced by the clinical position of the patient because he was not yet taking palliative drugs.

An important aspect of the pre-clinic meeting is the organisation of the potential research subjects from the list of the day's patients. Researchers often request recruitment of new patients for their individual projects in this meeting, asking if the neurologist thinks certain people may be suitable, and if so, can they be asked about further recruitment during the initial neurological consultation. There is brief discussion about who will see which patients first, the neurologist or one of the other researchers. There is sometimes a discussion about how recruitment is progressing in the different projects and trials, and

information is shared about changes to tests or procedures, and the central database. Then the researchers disband, and wait for the patients to start arriving.

The first patient to arrive would normally see the neurologist first, and this allowed further recruitment to the other studies. The neurologist would question the patient and carer about day to day living, using a standardised measurement instrument, the Unified Huntington's Disease Rating Scale or UHDRS. The scale was created by the Huntington's Study Group of America, and has gone through several updates according to the increasing knowledge of the disease and brain function. The current version of the UHDRS is considered to be a highly reliable and consistent measure of the main physical and cognitive problems associated with HD (HSG, 1996). Topics within the UHDRS include, motor, cognitive and behavioural assessment, independence scale, functional assessment and total functional capacity. During the questioning about their capabilities and daily lives, patients and carers can demonstrate their ability to cope and also highlight difficulties. Each consultation is therefore both standardised and personalised, and this contributes to how families feel about the clinic, they consider that they are understood by the best experts in the field.

Creation of 'research friendly' neurological HD

The UHDRS is used to guide the neurological consultation and provide information for Professor James to record in a quantitative manner. Special documents were designed to make it simple and straightforward to record the information during the patient consultation. There are future plans to make this an online activity during the consultation, with the direct input of data to the database. In effect, this creates a standardised form of the disease which can be measured against other individuals, populations, and previous visits. The data is available to the researchers in the 'home group' immediately, and it can be accessed by other European research groups with permission via the research project's co-ordinators. The data is managed by the neurologist who decides how to transcribe the information the patient gives, into research data via the written or digital recording of the results. This is usually performed in the clinic during the consultation, and is occasionally completed after the patient has left the room. The following extract from my field notes illustrates how this is achieved via 'clinic talk'.

Extract 1, observation 24, field notes 2008.

A married couple I had spoken to in the waiting room, entered Prof James's room. They appeared to be lively, sociable and interested in research, having talked to me about their family tree and artefacts from their family. The patient was a retired electrical engineer and his wife was a retired teacher. I went to sit behind Prof James at one side of the room, my view was of the side of Prof James, and facing the patient and his wife. I began to make notes as the consultation began.

(not a recording).

Prof James: How are things?

John: Mm yes...

Amy: The movements are much worse.

(At this point a discussion was initiated by the HD support worker, about support in the area where they live.)

Amy: That's very very helpful, thank you.

Prof James: Now, drugs?

Amy: (gets them from her bag) Mr. Smith in Hertford prescribed them (passes them) - he hasn't taken any of them because Helen James, the HDA rep says there are side effects, like increased depression.

Prof James: I would be inclined to prescribe Olanzapine, we can start low, it might be enough? (voice rises in encouragement)

Amy: That's what the HDA person said.

Prof James: Well, I agree with her, we'll try that.

Amy: As for food, I use the pressure cooker, its all soft, no roasts. There's been such a problem with his dentures and implants, its hard to get a dentist and to get him there.

Prof James: We have dentists here who see HD patients, its just a matter of referring you really.

Amy: That's stunning, its absolutely great, thanks so much.

During this first part of the consultation, the ‘talk’ has been of support and treatment, in effect the clinical management of the disease. The neurologist has also offered an alternative drug treatment and the services of an NHS specialist dentist, both as part of clinical treatment. There were relatively few clinical studies published up to 2006 of the use of this drug in Huntington’s disease, and the authors of a report urged clinicians to publish more on this (and other drugs used in HD) to increase the primary literature base (Bevan and Paton, 2006).

Returning to the consultation, the neurologist moved onto the Unified Huntington’s Disease Rating Scale and activities of daily life.

Prof James: OK, so that’s your teeth, your swallowing, what can you do?

John: I still vacuum and lawn mowing, I still shower and dress.

Amy: He used to do ‘It’s a Knockout’¹ you know, we were five times in Europe, we videoed everything, it shows how HD is such a cruel disease.

John: I still do all the accounts, the spreadsheets and that you know.

Prof James: That is fantastic, well done

Amy: Well I have the internet side, he does the rest, do you want my email?

Prof James: Excellent, that’s very useful. OK. Good. (Starts to fill in blue form for Registry project)

Amy: I know about Data Protection, you have to be careful but I’m quite happy for contact on email.

Prof James: OK can I examine you?

(Short examination in the main room, patient is in wheelchair)

Prof James: The movements are not *too* bad- underlying the involuntary movements- they are not bad at all.

Amy: Keeping going is very important.

¹ This was a popular TV show in the late 1970’s and 80’s. Teams from different towns across Europe competed in bizarre sporting and physical tasks, involving balancing, climbing, and getting in and out of water. It required skill, fitness, co-ordination and a very good sense of humour.

Prof James: This is a very good time to start the new
medicine, (then a very firm announcement)
You WILL benefit.

The exchange of information here was concerned with the completion of the blue report form in the file, this indicated the patient was enrolled in the Registry project. Amy, John's wife, was keen to give information about the previous capabilities of her husband. This is a form of re-asserting the patient's biography which was often utilised by carers to different effects. Here, Amy was telling that John is or was, a strong, healthy person with a sense of teamwork and community spirit, and that he still made a valued contribution. Amy also showed that she was a capable carer, she was up to date, she understood the complexity of the disease, that John needed to 'keep going' with the activities, and she suggested email to have more immediate contact with the clinic. The Professor meanwhile, reassured John that the medication will be of benefit, unlike the previously prescribed drug which he and Amy had rejected due to the possible side effects.

Most of the information for the research was gathered from the neurological examination and recorded on the blue registry project forms, whereas the helpful interjections from Amy were of no consequence in this exchange. The neurological information fitted into the disease recording framework around the consultation. This is how specific signs and symptoms were harnessed to assist the research project, by adding to recruitment and forming a standardised repertoire. The result will be a model of disease which fits with the measurable criteria and symptoms experienced by most individuals. The responses that John made to the examination either by speech, movement or understanding were converted into numerically scaled information and recorded as facts about his condition. Later these 'scores' were entered onto a database which is administrated from Germany; there are over one hundred and thirty research centres in Europe entering patient data from clinics. The local researchers in this clinic will be able to access their own patients' data immediately, and can apply to access some or all of the other centres data for comparison studies. This is the basic data (with a few alterations for the project purpose) which is made internationally available, the data base being a repository of disease epidemiology. The local clinic is collecting data for use in much larger scale research..

The standardisation of the disease assessment and measurement goes further in that all UK centres are co-ordinated so that data is entered uniformly and correctly. In addition,

training DVD's are distributed to clinicians to show how the tests and assessments must be carried out, to remove variation in operation and interpretation as much as possible.

At the end of the neurological consultation, the patient and carer are usually directed to one of the other researchers, physiotherapist, psychologist or research nurse to complete further assessments for the research. The next section will discuss one of these other studies and the procedures involved, and how these are converted to data.

Recruitment to other studies

Entry into all the allied research projects is facilitated in most cases by the neurologist making a direct face to face personal invitation to patients. This may be related to a particular difficulty the patient has encountered, it is therefore directly connecting the research to that aspect of the patient's clinical care. One such study was concerned with the frequency of falling and general mobility, recruitment was based on the patient (or carer) reporting of falls in the main neurological consultation. The aim of the study was to a) estimate the frequency of falls in people with Huntington's disease and b) make a preliminary assessment of tools appropriate for assessing the risk of falling (Busse, Wiles & Rosser, 2009). It appears that prior to the recently published research, information about falls in HD was mostly anecdotal and had not been studied in depth.

The researcher who carried out the falls study was one step removed from selecting participants, relying on the neurologist Professor James' filtering for suitable candidates. Each of the allied researchers has a minimum amount of control over the patients put forward for their individual projects, although as previously mentioned pre-clinic discussions are often used to share information and allocate patients.

I was able to negotiate a few detailed observations of the procedure of the mobility and falls study, which also took place in the clinic. It was evident during these observations that very specific data was required from the patients. Thus the precise type of numerical data about falls was obtained from the source - either a patient or carer's first hand accounts. The researcher had to apply a numerical value to the accounts of falling, and gain the agreement of the patient about this value. Patients were not always prepared for this and it was sometimes a problem which appeared to inhibit the research a little.

The researcher was a physiotherapist and had a set routine of questioning and examination. The patient was required to give definite answers and the physiotherapist recorded the answers in numerical form. The patient also undertook a series of physical tasks

and the results were recorded in either numerical or scale form. The researcher also filmed patients carrying out walking and sitting tasks, to assist with further analysis of the effect of the disease on balance and co-ordination abilities.

Creating ‘hard data’

The field notes from the observation illustrate how the legitimacy of accurate data measurement is a constant theme made visible in the study of patient falls. The observation of the research routine began in a side ward of the research centre. I was not the only observer, a physiotherapy research student watched too. We sat by the windows and tried to keep out of the way. George, the patient was a tall, slim, smart man, with grey curly hair. He was casually well-dressed, able to walk and understood what was going on. Ethel, his wife sat opposite him, she was fair haired, a bit plump and looked very nervous, a small table between them. Beth, the physiotherapist sat at right angles between them. The following extract is from my field notes.

Extract 1 from field notes, observation 6

Explanations were given about the research and consent issues which took about five minutes, then Beth offered the couple some tea, and went off to make it. They both seemed very pleased with the offer. [*In the field notes, I had noted I was not fully aware of what the physiotherapy research was about at this point.] Beth came back, tea was being drunk and she began to question George. ‘How often do you trip?’ George said he didn’t trip. Ethel said ‘You tripped up the steps coming in here’. He mumbled something. Beth asked him again, ‘How often? Every day? How many times?’. George tried to answer but couldn’t seem to decide on a reasonable estimate. Ethel started to count and calculate on her fingers, and said ‘ If he’s out, he trips more’. Beth asked him ‘Four times a day? More or less than that?’. George looked at Ethel, who was holding her tea and shaking her head a bit. ‘Well what does she think?’ he asked.

‘I want to know what you think,’ said Beth, ‘is it everyday or not?’ I thought Beth appeared a bit irritated by how things were going, she spoke very deliberately and wasn’t smiling much. The sequence of questioning continued with George not very sure of the precise answers which seemed to be required. He kept looking at Ethel who did some head shaking and rolling of her eyes. Beth

repeated that its what George thinks himself that matters, and that he didn't need to look at Ethel for the answers.

From this extract, it can be seen that the physiotherapist required precise numbers to be given, or at least a good estimate from George who was required to be aware of his frequency of tripping. However, it did not seem as if he had counted or noted his occasions of falling prior to this visit to the research centre and it was a puzzle for him. Ethel was able to estimate the number of falls, but her information was also imprecise “*If he's out he trips more*”. George attempted to enlist Ethel's help because he finds answering in the required framework impossible. However Beth the researcher wants his answer specifically, “*I want to know what you think*”.

My field notes recorded that I wondered at this point, if the study was about perceptions of falling. I was unaware at the time of the observation that the criterion for inclusion in the study was two or more falls a year. One fall a year was not a qualifier, therefore it was important that the qualifying conditions be met early in the process. This was an example of the transformation of patient perception into hard numerical form, eventually creating scientific statistical data. Due to the close boundaries of ‘non-faller’ versus ‘faller’, it was essential for the researcher to establish the legitimacy of the research participants into these categories.

However, it appeared as though the patient would have no prior knowledge of criteria and in this case was unable to be precise. The criteria had not been mentioned in the pre-clinic meetings or neurological consultations I had observed, hence my misinterpretation of the study as something to do with perception. In order to create hard data from the patient's experiences of falling, Beth attempted to establish the data accuracy. She is asking “Are you a ‘faller’ for my study or not?” There are two other accounts of falling given here, one from Ethel, the patient's wife and one from the referring clinician, but neither of these are precise either, so the researcher needs to establish legitimacy for the data. Lack of precise answers was also pathologised as part of Huntington's disease for some patients; loss of cognition was also one of the areas of measurement in the research and was usually measured via a language test. Levels of emotion were also used as indicators of normal or abnormal functioning but it was not clear how these were measured. I will go on to discuss this in the next section with reference to an example of a patient's emotion about the disease.

Disease pathology and emotion

A lack of specificity from a patient, as shown in the previous section, could be pathologised in the research as being ‘part of Huntington’s disease’. This was considered problematic for the patient rather than the researcher. The researcher’s view of what stage of disease had reached was prioritised, for example, a loss of perception, or someone behaving in a demanding way may have been rationalised as ‘in denial’ or as a sign of disease progression.

During one observation, a social carer explained that Laura’s child had been removed from her care and now lived with her father. Laura expressed anger and sorrow, and wanted to discuss genetic testing of her child. She was immediately discouraged and prevented from pursuing this discussion, in line with current ethical practices around genetic testing of children.

Extract 3 from field notes, observation 15

Laura: I know I’m going to die aren’t I? (Begins to cry) I think a lot.

Carer: She has been better but now misses her daughter a lot- she was taken to live with her father 17 months ago. She’s 6, and she’s very happy.

Laura: I miss her (crying).

Carer: The father brings the daughter to see her Mum.

Laura: I’m very angry, f’ing taken her away from me. F’ing nothing wrong.

Prof James: What about other ‘downs’?

Carer: Mostly to do with Jenny.

(several exchanges about drugs here)

Laura: Yes (pause) You know my Jenny? Will she have HD? I want to know, I’m worried about her.

Prof James: We don’t know– she will have to decide when she’s 18.

Laura: I got my father’s gene-will she get it?

Prof James: (ignores question and examines patient’s limbs).

The movements aren’t too bad, they used to be worse. We might be able to decrease the medication eventually. (Laura

leaves to go to bathroom) [From here, neurologist was speaking privately to the social services carer]

She will decline more, there's nothing we can do to slow the condition. The changes are what we'd expect at this stage of the disease. She has retained a good sense of humour, she's very lively. But she's struggling with change, and she will do from now on.

Laura's concern for her daughter was not pursued. Her disease was considered to be progressing, her neurological responses were prioritised and measured for the research. The 'struggle with change' was forecast to be a permanent feature.

Carer: Jenny is becoming more distant and I think this upsets her, I'm sure she knows.

(Prof James recruits patient into physiotherapy falls study)

Carer: There was [sic] about 6 visits in one year to a special court about her daughter, then a big deterioration - could that have caused it? She's had it ten years. Well, that's since diagnosis. She told me ages ago when she was young she couldn't carry a tray of glasses.

Prof James: Laura was in denial for so long. She wouldn't accept anything. Often people tell us that stressful events precede declines etc, but we don't really know

During the observation I felt sympathy for Laura's worry about her child, the agony of wondering if the disease would appear in the daughter. It was only later whilst reviewing the field notes that I realised that the patient's anger and sorrow had been pathologised into 'denial' and 'stage of disease'. In contrast the carer asked if anger, sorrow and deterioration were reactions to the stress of a difficult court case. These are two quite different perspectives of the patient's condition at the same time. However, the difficult personal circumstances have no bearing on the recording for the research. Laura's troubles were 'disease' data.

Care issues passed outside the clinic boundary

Difficult situations for patients could not always be dealt with by the research clinic and had to be passed to outside services. Prior to my observations, there had been a psychiatrist in the research clinic but due to other work commitments, the collaboration ceased. The remaining researchers acknowledged they were unable to carry out accurate psychiatric assessments, and could not offer advice or support in this respect to the patients and carers. They reflected this was not ideal for patients, and not always a comfortable position for themselves. The only recourse in these cases was for the lead researcher, the neurologist, to refer patients to community psychiatric services in the person's home area.

One particular observation illustrated how these difficulties were met. Sophie had HD and attended the clinic with Nigel her husband. At the end of the consultation, Nigel mentioned Sophie's struggle to deal with her declining abilities and how this made her feel.

Extract 4 from observation 26, field notes.

Prof James: Anything you want to ask?

Sophie: Not really, can't think of anything

Nigel: She mentions suicide, but she's not happy for me to say it.

Prof James: How often do you think about it?

Sophie: Well, I don't know, its just something there sometimes.

Nigel: She saw her Dad, she doesn't want that for herself

Sophie: (irritated) I don't want you looking after me.

Nigel: But I will - I don't want you to go, not like that

Prof James: (To both) Maybe its difficult to separate the depression from the disease or events? Maybe we can increase the citalopram? (To Sophie) You have to think how suicide would affect Nigel. You seem quite down from last time I saw you. You could see the local psychiatrist, I'd be happy with that, because some GP's have experience with psychiatric drugs. Have we got the GP's details?

Nigel: Its changed twice, now it's a lady, I can't remember her name.

Prof James: You must phone our secretary with this information. This is the priority.

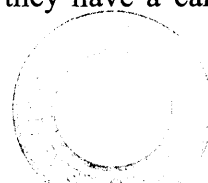
Nigel: It has been a difficult time the last 12 months.

Prof James: Well it's a miserable condition. Good. Just wait outside now, and I'll check with the physio, I'm not sure where she is.

Several things happened in this consultation, after the signal for it to end when Professor James asked Sophie if there were any questions. Different perspectives of Sophie were presented, the initial one by Sophie, then her husband, then Sophie again, and later the clinician. Professor James negotiated the disagreement between the couple, and questioned further. Sophie was irritated, this was an important matter of autonomy for her. However, Prof James issued a moral objection to the effect of her potential suicide on Nigel, a diagnosis of a worse depression since the last visit, and suggested a psychiatric appointment. However, the priority from the consultation was notification of GP details. The personal troubles raised by the husband were placed outside the clinic, with the GP or the community psychiatrist. So the matter of possible suicide will not be explored here, although it will be recorded. The transfer of the problem to the GP was a matter of importance so that assistance could be given. Some of the problems of living with HD cannot be dealt with in this clinic, although if a problem is mentioned, it must be dealt with. This represents a particular challenge for a combined research and management clinic for such a serious condition. The two functions of disease management and research were evident in the consultation, however, the neurologist is constantly negotiating which function will take priority, and how the research aims can be satisfied in the clinical practice of care. This represents a merging of care and research, or blurring of the roles, which cannot be planned for.

Moral and Sentimental work

The moral and sentimental work took place in meetings before and after the clinics. Professor James and the researchers discussed which patients would attend, but not all patients responded to the clinic invitation. The Professor explained to me the various reasons this occurred. People who have Huntington's disease can have problems with cognition which affects their ability to organise their lives. Unless they have a carer or someone to



assist them and remind them, they may not manage to keep appointments or find their way to the clinic. This is especially true of people who live alone and whilst it is important to maintain independence for as long as possible, it does mean they may not always be able to access helpful services.

In the clinic meetings there may be reference to the caring or family situation, and whether other family members are being seen in the research clinic, or if it would be possible to recruit them. There may also be an identification conversation like this,

‘is he the one who thought he had ants crawling on him?’.

(Field notes, pre-clinic meeting May 2007)

Family members and carers are also discussed in the clinic meetings and their attributes may be mentioned as in this example:

‘His wife is a brilliant carer, she really has a handle for how the disease is’.

‘Yes she’s a nurse’.

‘Ah well that explains it’ .

(Field notes, researchers talk, back stage July 2007).

Researchers noticed and expressed opinions about the patients, and their biographies, and made comparisons between types of patient and types of carer. These features also appear in the research tests, consultations, and after-clinic talk as forms of reassurance, support and occasionally disapproval (Featherstone et al, 2006, p. 113).

Moral and sentimental work did not appear to have any specific purpose, even though it was shared knowledge. It seemed to be used as a device to assist the ‘sorting’ of patients and carers into categories and to seek agreement from the rest of the research team. It was also a social device within the researchers group which enabled them to share thoughts, and test their opinions.

What is left out?

The use of the Universal Huntington’s Disease Rating Scale (UHDRS) in the main consultation has two main purposes for the clinician/researcher. It will give a picture of the disease progression in the patient since the last rating, and it provides some measurable data

for the European Registry database. However, from the patient and carer's angle, it is asking about how they live with this disease in normal day to day situations. For example, 'can you wash, dress, eat independently?' What the rating scale cannot do is provide any respite from symptoms, arrangements for relief care, explain if a problem is caused by the disease or not, or take account of how the patient and carer feel about their situation. Some of the areas which are left out, are ways of coping or not coping, the worry of day to day problems such as choking or falling, the increasingly narrow world of sufferers and carers, and the risks for other family members.

In harnessing the UHDRS, patients and families are asked about the day to day activities which progressively become more difficult with advancing disease. The impression is given that they are attending an expert service which has full understanding and awareness of their daily troubles, from the general issues of movement disorder, down to fine details of what and how they manage to eat, and how much and when they sleep. It is fair to say that the research clinicians do acknowledge all this detail, but they use only parts of it - the parts which can translate into measurable quantity for the research. The rich descriptions of holidays, former achievements, current diet and fears for the future are sidelined for numerical scores and likert scales of movement, cognition and function. The areas which remain untouched by the research exercise itself are dealt with by services other than research, albeit there is sometimes a helpful referral to smooth the way. The research has become the service to the patients in this case, a redrawing of disease to fit into the research categories. This is not to say that symptoms and problems are ignored or unnoticed, but as there is very little treatment available, they often hang in mid air, until a suggestion is made for re-organisation of self-help or outside intervention from a GP or a community psychiatrist. The complexity of the disease is reduced into separate measurable categories, the personal is not dealt with or is directed towards the HDA representative, and the unruliness of living with the HD body and mind becomes measured and represented by controlled, numerical data.

It is ironic that the UHDRS is considered to be so good and reliable (the 'gold standard') as a measurement of disease precisely because previous clinicians did include all the fine detail in their reports of the illness. What is now included for measurement is a matter for the research designers of the European Registry study.

This has enabled the questioning to be developed to ask about the things that can be measured in terms of categories of brain function; these become the 'legitimate' form of the disease because they represent areas of potential intervention. These relate to day to day

living but are limited to the most basic motor and cognitive functions. However, the research cannot cover all patients in the HD population, because, patients cannot access research unless they are able to travel to the hospital and research centre. This is a contrast to previous NHS arrangements of support/surveillance type visits to the family homes of people diagnosed. Therefore, as people become more disabled, their data ‘drops’ out of sections of the research, and eventually they stop attending. Non attenders do not figure in the research at all. Overall, the study is taking account of only a section of the HD population at a particular time in their disease state, those who attend the clinic, and are able to do the research. This is thought to be about fifty percent of affected people. The created model of HD will be based on this fifty percent of people, it will therefore show early and perhaps middle stage HD. It will be a partial view, intended to assist the design of early interventions, and will not provide any information for the later stages of HD.

Creating, using and enhancing expertise

The expertise of Professor James and the research team was recognised outside the clinic by the community of HD sufferers and families at risk, who were keen to look at the project websites, and eager for news of breakthroughs. The enhancement of expertise was fostered within the research consultations by the researchers who encouraged research participation with references to current research taking place in the laboratories of the nearby university, promissory language of possible trials, and potential treatments in future years. The promissory language was general rather than specific and also represents the researchers own keenness for breakthrough treatments to legitimise the clinic work.

Several factors contribute to the normalisation of research in the disease consultation (the accuracy of the UHDRS in tapping into daily life, and the enthusiasm for research, the further recruitment to other studies), especially where there is no approved or available treatment. This phenomenon has been noticed by other research (Petryna, 2009), although in a slightly different context. Petryna noted the effect of global research on populations who would not normally have access to medical treatment in Eastern Europe and Brazil for example. Facets of this situation include the patient’s expectation of expertise, the hope for novel treatments, and a lowering of barriers between the patient and the clinician. The barriers of medical superiority in normal doctor-patient relationships have been under pressure for some time in the developed world, due to the open availability of medical

knowledge to the general population and a more distributed responsibility for the body and health. In the case of the 'research and care package', barriers are lowered due to the active search for more knowledge with the patient as co-researcher. The mutually beneficial relationship where patient co-operation and data are exchanged for a potential treatment has long been acknowledged, (Porter, 1999) but there are stronger meanings to this when the disease is incurable. When there are no successful treatments, the only possibility is the promissory effect of a clinical trial or experimental surgery.

Meaning and status of the clinic

Patients and carers often described their clinic visit as an opportunity for 'just a chat' with the Professor and the researchers. The idea of the visit being 'just a chat', indicated the relaxed atmosphere in the clinic, the familiarity of the routine and the type of non-invasive examination. Yet, the dual purpose of the clinic was somehow acknowledged. One of the first patient and carer interviews illustrated this. In addition to the routinised surveillance there was the need to keep in touch with experts and feel involved in something. There was acknowledgement of the incurable nature of the disease and the regular contribution of data.

Ken :. on and off every six months to a year we've come down and paid a visit for about half an hour to an hour..., its mainly only a chit chat.

Jacki : But it keeps you in touch doesn't it?

Ken: Its basically that yeah, yeah, because there's no cure for Huntington's, its just a case of them collecting data really.

(Interview 1, October 2007, Husband, main carer)

There was no discomfort with this explanation, it appeared to be an accepted routine for patients and carers who were familiar with the arrangement. The research was 'collecting data' and appeared of no consequence to this person. Another carer also expressed a familiar and comfortable attitude to the clinic:

Edward :- I would have gone there whatever time they gave me to be honest, you just fall into line with things like that don't you?...a bit of waiting time basically and in to see Sarah, do the motor tests

as you call them, general chat, 'how are things going'. She [Anna] used to say 'everything's alright, it's fine'.
(Interview 10, Carer, 2009)

This view of the importance of attending the clinic for Edward and his late wife ('I would have gone there any time'), was combined with the idea of regular contact with the clinic professionals on a friendly co-operative basis, and a 'chat' appeared again. Edward also made use of Professor James' first name, as if talking about a friend or someone well known to them. This suggests an equality of purpose in the clinic, and a mutual understanding of the importance of regular contact. There is a more equal and less deferential stance to the medical authority of the clinician, in conjunction with the acceptance of the clinic as routine, and a 'falling into line' with the appointments system as expected behaviour for patients and families. In some cases, clinic attendance also drew comparisons with other hospital clinics.

Informal interview, Carer 9, February 2007.

"You see, we like coming here, its so nice, everyone is, from the porters right up to the Professor. Its not like that in our local hospital you know, its being run down"

This carer and his wife had travelled from another part of the UK, and he indicated his appreciation of the way people are treated here, and extended the compliment to the whole hospital. The specialised clinic also confers status on those who attend as patients, they feel valued at all levels in the organisation, which is a progressing enterprise in contrast to the description of the local hospital as 'being run down'. The patient and carer considered the care and facilities were better here than nearer to home, which justified their long journey and gave them control over the care.

The meaning of research for patients and families

The patients and carers had a rather sketchy idea of research most of the time and their own role in it, often describing the visit as a 'check up', 'just a chat', or 'monitoring' the condition. Most patients and carers had never taken part in a randomised controlled drug trial at this clinic, which was what they associated with research participation. This sometimes led them to say that they were not involved in research really, they had never

been asked to take part, even though they had volunteered for ‘anything and everything that was going’. This made me question what does ‘being in research’ for the clinic participants? It appeared that research for this cohort meant having some kind of intervention, whether drug based or surgical. The following data extract displays the uncertainty around the meaning of research in this clinic.

Interview 2, Enid, Mother, 2008

Jacki: Can you remember the first time you were asked about research in the clinic? Or before you went to the clinic?

Enid: Erm, well in the clinic yes. We signed up for any research because you grab at any straw don’t you, you know, and at that time, going back quite a few years now, there was the stem cell wasn’t it? To the brain ? They’d done two operations, how many years ago – about six years ago.

Jacki : Yeah, five or six.

Enid: Yeah. And I mean we thought ... but of course it’s not that is it, you know, it’s a long way off isn’t it.

Jacki: Mmm, it is at the moment.

Enid: Yeah. So, we thought it would have been ... I mean I’m silly, no, because Alan was lucky with his heart transplant, I thought oh Alice is going to be lucky with hers you know but ... keep our fingers crossed, you never know do you.

This carer focussed on the idea that research in the clinic may be a surgical experience, because she has had experience of another child who had a heart transplant. The possibility of stem cell transplant surgery for Huntington’s disease was seen as normal, beneficial and available via this clinic. Patients and carers were aware of disease symptom monitoring and some testing which consisted of movement assessments, cognitive tests and sometimes physiotherapy questions and exercises, but these were usually viewed as monitoring the disease progression in the individual rather than contributing directly to research.

Jacki: Hm. But if we took it away from what’s outside the clinic, when you know that you’re going to the clinic for example, and perhaps Stephen has come up from home and he’s stayed with you for a day or so and then you go off to the clinic – and whether you

go in with him or not ... you know, what do you think about where you're going? Do you ... erm, what impression do you have about what you're going to get when you're there?

Carol: I think my impression is that ... sometimes I just feel it's a monitoring situation and that, er ... I suppose because of the nature of HD, again, what can be done? Erm, there isn't an awful lot that can be done except perhaps to tell you that movement's got worse or ... and then what do you do then? There's a black hole then isn't there, there's no answer. So, it's a monitoring ... but it is a point of contact. It's all there is to be honest.....

(Interview 6, Mother, 2008)

In this extract, the movement tests, psychological testing and cognitive tests were all viewed as monitoring the disease, and as a way of calculating how the patient was, rather than being specifically research exercises.

A clinic focussed on the patient

This particular clinic was considered by patients and carers to be 'better' than the previous clinic based in the Genetics Department. despite being difficult to find from within the hospital. The extract shows how one carer coped with this.

Interview 10, Edward, Husband carer.

Jacki: OK. When the clinic moved from the Institute of Medical Genetics into the...

Edward : Main hospital, it's a nightmare to find!

Jacki: It is, it's a little bit hidden away And I think whichever way you approach it, you've either got to come through the maze of the hospital or through the maze of the one way car parking and road system haven't you ?

Edward: Mm, I ended up with all the porters in the end and just said (laughs) 'tell me where I'm going', they took me up there in the end.

Jacki: Yeah. That's not a bad strategy I think. Erm, did you notice

any ... was there anything that struck you about being in that research centre as opposed to being in the Genetics Department? Was there anything particularly different?

Edward: I think it was more welcoming. Once you actually got in there. Getting there was a pain but when you went in there it was much brighter and ...

Jacki: Do you mean the surroundings?

Edward: Yes, the surroundings?

Jacki: And the people?

Edward: I think the people were much the same to be honest.

Jacki: Yeah. But it's a nicer place to be?

Edward: Yeah, it was nice and airy and modern. It felt less 'clinty'.

Jacki: So, did the environment have a positive effect on your visits?

Edward: Yeah, I thought so, yeah. I thought so.

Researchers also considered that this site was a better place for the clinic than the previous location, for several reasons. This researcher explained some of the differences.

Interview, Researcher 3

Researcher: Yeah, and certainly our patients have come from us having clinics in genetics where it was always more difficult, the rooms weren't really set up for a neurology clinicBut also it's a slightly dingier building, the light isn't so good you know – I do think it makes a big difference actually. And the other thing the patients have always said is that they actually, funnily enough, they like *to talk to each other* while they're waiting, they find that a really positive aspect, erm, which is interesting, erm, and I think it makes them feel less lonely, you know ...

There was acknowledgement from carers that the clinical focus was very much on the patients, even though aspects of the care situation, quality of life, family and difficulties, were frequently mentioned in the interviews and in the clinics. The patients' neurological condition and care situation were clearly thought to be dealt with separately in the minds of

patients and carers, who expressed ideas about limited NHS time and resources - for example 'obviously the focus should be the patient' (Interview 3, Mother)

Interview 3, Carol, Mother, 2008.

"So that's what I wanted to filter past ... some of that past, but you can't. There isn't the time. There isn't the opportunity. So I think perhaps often carers/family members, regarding the clinic, probably have things to say, or they'd like to say, or they'd like an opinion on, that is as it stands, it's sometimes impossible to do.....

Because obviously the main focus should be the patient. And you can't ... I'm not criticising that and I know the amount of work and pressure ... you're looking at time and you know it's not there".

This comment fits into the idea that medical resources and time are limited and should be applied only to those in greatest need. It embraces the twentieth century idea of health care as scarce, expensive and focused on individual need only. It also raises the issue of support and advice for the wider family and carers, a topic which does not have a legitimate place in this research and management clinic.

The clinic ethos

The HD research and management clinic was not a centre of complete care for the patients and the wider family: genetics, risk to family members, and counselling is not the focus – rather, this is squarely on observation and recording of patient signs and symptoms, the collection of large amounts of data which is anonymised and shared with a growing European network. However, empathy and professionalism was delivered in a manner much appreciated and endorsed by patients and carers. Additionally, the ethos of the clinic as a whole made an impact on patients and families. The general reception of patients, the offer of tea or coffee with biscuits, the demeanour of the receptionist, and the calm consideration of research staff during the visit meant that patients and families were extremely appreciative of this time and attention.

Interview 5, Angela carer, and David, patient.

Jacki: So you were saying that it's nice since they've moved – and where was it before?

Angela: It was over in Medical Genetics Research ... I loved it over there.

Jacki: Did you?

Angela: Because he was going so long.

Jacki: Yeah? How long were you going there?

Angela: Oh gosh, it's been thirteen years.

David: Oh yes. Thirteen.

Angela: Yeah, yeah, from when David got diagnosed. So they all knew us and it was lovely but in this, it's not like a hospital ... they're lovely there and ... it is nice David isn't it, when we go in and they always speak to Dave. You know, they don't say 'would David like a cup of tea?', they ask David direct, 'would you like a cup of tea or coffee'. They're lovely.

Interviewer: Is that different then to other places you've been or other places you go?

Angela: Yes, yes. People seem to ask me.

David: Its different.

Angela: 'Can he drink?' That's what they usually say 'Can he drink?'

Jacki: They don't ask David direct?

Angela: No. No. And they do down there, they're very good. Yeah, even right to... the receptionist, right up to the Professor. They all, you know, ask David – it's lovely.

The high standard of personal regard for patients and carers did bring benefits for the researchers, it encouraged continued research participation and communicated something of the nature of the research. In addition, it was viewed as a special event by patients and carers who do not experience this self esteem in other settings. The repeated visits over many years often lead to a more personal relationship with clinic staff and researchers than would be the norm in shorter term research projects, and this is also appreciated on both sides of the research. Opportunities to ask about research did present themselves from time to time, and

patients were keen to be informed of progress. However, there was also an underlying realisation that progress is slow, and research may not be of direct benefit to the current clinic patients.

Disease progression, treatment stasis

Many people in the study had been involved with Huntington's disease for all or a great part of their lives, having had an affected parent, sibling, partner or child with the condition. Siblings who cared for their affected family members may have first seen one of their parents deteriorate over many years, then found themselves caring for a sibling for another twenty years. In one case, a wife cared for her husband for over twenty years and now cares for her adult child with the condition. The number of life years involved in just one family can easily reach a hundred, with some carers now in their own retirement years.

This was the case with the following interviewee who had experienced her mother with the disease, and later her sister. There was a gradual realisation that the research testing was reducing in successive visits, and then clinic visits became infrequent due to increasing disability.

Jacki: So, was that ... did you still attend the clinics with her then?

Theresa: As long as she wanted to go, yes. Yeah. And, er, they did, they just got further and further apart because ...

Jacki: How do you mean they got further and further – the appointments?

Theresa: The appointments, yes ... and ... and once she became more infirm and it was difficult to go ... I guess they just sort of fizzled out.

Jacki: Right. OK. And what did you do when you saw the clinician? What did they do? You said it was very routine

Theresa: Yes, well, they would just ask how she'd been and, er, whether there were any more symptoms, that kind of thing.

Jacki: Yeah. Yeah. And did she cope OK with all that?

Theresa: Yes. Yes, she did.

Jacki: Because you said you thought there was no change, you went there and nothing had seemed to progress ...

Theresa: Yes, it was, yeah.

Jacki: And did she notice that or ... do you think she felt they were, as you said

before, keeping a weather eye sort of thing? did she express anything like that?

Theresa: No, no she didn't. She just ... I ... she just wanted to do anything that would help her – or even help the clinicians or anybody. She would contribute and she would co-operate with anything.

(Interview 8, Theresa, Carer for sister)

Although Theresa recognized and maintained the altruism of her sister, there is also the indication that if there was something to be gained, her sister was keen to try. I was aware that Theresa was pausing and she seemed sad and quieter. This was one of those occasions where I wondered if I had asked something which had opened up thoughts about the future. Her sister was now completely incapacitated and without speech. She had survived for twenty three years and Theresa organised a team of round the clock carers.

The slow research progress was mentioned by patients and carers, who gave rough calculations of 'disease years' that people had been involved with the disease, research and the clinic. In the next extract one carer describes how she thought of the disease in relation to her family and her own life.

Interview 3, Enid and Alan, carers for Alice

Jacki: Well ... well as long as they feel that they're getting the care and the attention that they need, you know.

Enid: But, erm, to my way of thinking that.. the medication hasn't progressed since my husband had it.

Jacki: Really?

Enid: No. Because – the actual medication is just one tablet!

Alan: Well all that medication does is ...

Enid: I'm talking Alan.

Alan: Sorry.

Enid: It's just to pacify her, to calm her down at night – so it's nothing for the complaint. At all.

Jacki: Right. And what sort of medication would you think would be helpful – I mean I'm just plucking that question out of the air, if you thought there was something that would help her, what do you think it would be?

Enid: Well ... to control movements, to control ...

Alan: Balance.

Enid: Balance. Balance really is the whatsername, you know. Because ...

Alan: I thought there was something out for balance like?

Enid: No there's nothing for balance, nothing at all. Well not that I ... no there isn't, that one tablet is just to calm them down. And he only has that, he has the full strength one at night, so there's nothing really that controls their balance, they've got nothing for that. Because that all comes from the brain doesn't it?

Jacki: Yes, Yeah. OK.

Enid: So, I don't think that's progressed in – what 35 years?

Another person interviewed had cared for their partner with HD until their unexpected death a year before. In this interview, the duality of meaning attached to research is demonstrated. Edward fully supports research and is very active in fundraising activities for the HD Association, yet finds there is little or no progress, and no feedback despite the continued 'promise' of the research programme.

Edward: When you first find out about it, the first thing you want to know is how long is it going to take to get cured? And I still haven't got any closer to a date of that over ten years. It doesn't seem to be progressing in terms of – no doubt it is but it's not being communicated if it is. In terms of ... probably because they can't give you false hope, which I understand, I understand that. But it doesn't appear to me to be a huge amount of progress – there might be loads but we're not knowing, we're not being fed that information. I'm not getting ... I don't think I'm any closer to knowing there's a cure or an effective management by drugs of HD. ...I don't know if we're any closer, that's my perception of it. Obviously there's a lot of work going on. Yeah, so I can see that there's definitely obviously progress being made but there's so many issues, ethical issues I suppose and things which appear to be holding it back. That's my impression of it.

(Edward Interview 10, Carer)

In this extract, there is an expression of the desire to know some kind of timeframe to find a cure, and it is starkly contrasted with the fact that there is no difference in the timeframe even after ten years. Additionally this carer mentioned the amount of work going on as being 'obvious', and progress as being 'obvious', yet expressed the frustration of not knowing precisely what and how much progress. He also mentioned ethical issues and 'things which appear to be holding it back', indicating that he appreciated the complexity of the research world.

The need to protect patients' feelings

The clinic has a role of encouraging and enabling research participation, and managing (as well as possible) the progression of Huntington's disease in patients. In the interviews and observations it was evident that patients, families and carers were greatly encouraged by research participation, despite not being aware of important breakthroughs in research. Patients and carers noticed the changing patterns of consultation and research as the illness progressed. This included some tests being discontinued, others being altered, and the carers becoming more involved in answering for the patients.

Jacki: So, what sort of things did you do when you did the research?
Was it all sort of bloods and tissue things or was it tests or tick boxes or .. ?

Angela: Yeah. All like that...sometimes it's all according to what we have. ...like obviously in the beginning, they were a lot longer appointments because of him being tested and there was a lot more – he used to go and have bloods and different things but now we just review what's going on, how he's been. Obviously then I'm spoken to because for the medication part and ... how's he been and...

(Interview 4, Angela, wife and carer)

There were times when patients noticed the style of testing, became uncomfortable with it, and decided to withdraw from it. This was mentioned in relation to the cognitive testing research, which is an extra part of the European Registry project to collect longitudinal data from large numbers of patients.

Jacki:... but you weren't so keen on the other bit Sue, the psychological bit was it?

Ken: No, she didn't like that because they were asking her questions such as they were trying to catch her out...It's a little bit...something she didn't like.

(Interview 1, Ken, Carer, husband)

This was a feature mentioned frequently, the anxiety caused by psychological testing. There were times when patients knew they had not done well on a psychological or language test and the carers became aware that they had taken this on board. Sometimes carers withdrew from that part of the study, saying it was too unsettling and the patient was very upset when they got home, or they found it too tiring. This was the case in the above extract, the patient and her husband decided that she would not do that type of test again, but would continue coming to clinic. This illustrates the tacit nature of the knowledge about research which is held by patients and families. It does not feel like research at the clinic unless it becomes uncomfortable, then they may exercise the option to withdraw. They are not withdrawing from a potential treatment in this case, but rather avoiding the anxiety of testing, and the discomfort they feel with increasing incapacity.

Enid: ... I don't know but I think she came through that quite well really because ... I mean, I was witnessing her doing it and you had to say ...first of all you had to say the colour that was on the paper and then you had to say the word was, but you were looking at the colour and you'd think that's yellow but it wasn't yellow it was [red?] written there you know. But I think she done ... well, myself I did, I thought she done quite well on that you know. And she knew when she'd gone wrong kind of thing, you know.The colour one and the word one in so many seconds or minutes ... minutes, wasn't it? Words beginning with different letters you know. She didn't do so well on that, no she didn't do so well on that. I suppose it put her on the spot, you know.

(Interview 3, Carer, Enid)

In this extract, Enid explained the patients test performance as good initially, and later ‘she didn’t do so well on that’. However, in the middle of this, she expressed that ‘she knew when she’d gone wrong’, thus minimizing any idea of failure, and later ‘it put her on the spot’, indicated that she thought the test was slightly unfair, rather than her ability declining.

Researchers also noticed the patients’ discomfort during cognitive testing and used strategies for overcoming it and getting the test completed. This ranged from reassurance and encouragement to a focus on getting through the tests due to time pressure.

Jacki: Right, okay. So with your role in doing those cognitive tests then how do you find them when you run them? If you’re doing some cognitive testing for Registry for example?

Researcher: There’s several different elements. There’s sometimes people are quite motivated to do them and sometimes they’re not. So it’s always nicer to test somebody who’s happy to be tested. A lot of people particularly dread the cognitive tests because for those people with insight it’s quite easy to see that you’re not performing very well and it’s quite – it would be quite obvious to yourself doing the test that you’re not doing well I think.

(Later in the same interview)

Jacki: So I was – my next bit was what do they tell you about it? What do patients tell you about their research participation? But you say you don’t get much feedback, or you don’t actually request feedback?

Researcher: No. Sometimes during testing they’ll be frustrated like – especially with the PREDICT tests because they’re quite hard a lot of them. And so I’ll say – if someone appears to be struggling or getting annoyed, I’ll say this one is a bit frustrating or, it’s annoying or it’s really difficult, or something like that.

Jacki: You’re showing some empathy and saying it’s not you it’s the test?

Researcher: Yes. Yeah, and just stick with it and it’ll be over soon. And sometimes if they look like they’re tired we do have breaks and people are allowed to have breaks whenever they like, but I’ll say

there's only a couple more now and you've probably got about ten minutes left or there's three tests left, something like that.

Jacki: Yeah. So it's a little bit of encouragement but also the empathy and the sort of 'you're okay don't worry' sort of – ?

Researcher: And the other side of it as well is, with PREDICT some of the visits are three hours for cognitive. So, not only have you got to encourage them and everything and talk to them, obviously a bit like you would if you talked to anybody, and you've got to also draw the line between getting on with things, and then sitting and chatting. I mean you're on quite a tight schedule, you need to get through things, and so it's the balance between getting through things and not pushing and rushing somebody and letting them have time.

Jacki: And how does that work when they come because I find –

Researcher: There are sort of cues, the cues that you can give to say that you need to move on to the next thing. If someone's really, really, really talkative then it's difficult.

Jacki: And I think patients do actually like coming to that clinic from what I've seen. They're very motivated to come to the clinic aren't they? And they do want to share things with you. So I imagine that can be a bit tricky if they're very talkative and maybe they don't see that many other people.

Researcher : No, no. Or have anybody who can empathise with what's going on.
(Interview, Clinic researcher 3)

The researcher explained the difficulties in arranging long research sessions with patients, when there is a clear research schedule to achieve in a limited time. Additionally, it is recognised that how the patient feels on the day could influence their performance in the tests, and consequently whether they decide to remain in the research or not. Therefore recruitment can be affected after patients have signed up, if they feel the research testing is too uncomfortable. This is important in longitudinal research where several data sets are important components of the data base.

Conclusion

This chapter has discussed the data and analysis concerning one of the three cross-cutting themes of the thesis - 'Blurring' and tension between research aims and clinical management which occurs as a result of the dual role of the clinic. One of the most important tensions concerned the change in emphasis of the clinic from genetics to neurology and how this impacted on the dual role of research and disease management. The pattern of the clinic day was based upon the neurological research programme, which consisted of several projects being studied in one clinic and there was an effective rotation of research data collection. This programme assisted in the creation of a 'research friendly' form of Huntington's disease, which was standardised and routinised according to the procedures and demands of the data collection. During the data collection, the recollections and perceptions of the patients and carers were converted into standard, numerical or digital data for storage in a large international data base, which represented a global scale disembodiment of the disease.

There were both advantages and disadvantages for patients and researchers with the dual system of research and care. One advantage was that people felt very well cared for and understood because the research was very tightly focussed on the patients. However, there were important areas of care which could not be dealt with effectively, and these had to be passed outside the clinic to other organisations and agencies. This raised questions about what are the legitimate matters for the research and management clinic, to either approach, prioritise or pass on.

The meaning and status of the clinic and its research remained highly prioritised for the patients and carers, although they did not often make great distinctions between research and care, except when they felt uncomfortable with a research activity. Researchers tried to avoid patients 'dropping out' of tasks by means of confidence building and encouragement, and they realised there was also an important task to do in maintaining people's motivation to attend the clinic. The researchers however, acknowledged that having both care and research activities taking place, meant there are some areas which are difficult to negotiate because they are unable to advise on day to day matters, and they have a research agenda to comply with. The lack of practical research progress in producing treatments is quite frustrating to patients and carers, who notice that there is no real progress despite the 'hype' of genetics, and latterly the increased expectations of neurology and regenerative medicine with foetal stem cells.

Chapter Five

Performance, practice and experience

Introduction

The Huntington's research clinic is performed and created on both sides of the encounter, by the researchers and the researched. Once people enter the clinic (or prepare to enter) they become aware of their 'category' in this space and may behave according to their beliefs and expectations about clinics, hospitals, disease and research, or they may resist stereotypical roles and act according to some other set of personal guidelines. Whatever the guiding premise, the meanings of the clinic and the clinic activities to individuals are influential on the roles taken up within the clinic. Moving beyond the often quoted altruism of research participation, this chapter opens up the taken for granted status of patients and researchers, to reveal the meanings of taking part in a global research project.

The overarching theme is of 'Performance', but this could also be described as 'presentation' in the way Goffman intended (1959). The 'clinic routine' is not a one-off performance for any of the actors: repeat visits are made by patients and carers, and repeat performances are enacted by the clinicians and researchers for the variety of audiences they receive each fortnight. Goffman suggested that due to the repetition of these social roles, (plus occasionally the same audience in this case), 'a social relationship is likely to arise' (1959, p.27). This chapter will examine the aspects of performance or presentation by the clinic actors to gain understanding of the social relationships, and social meanings in the clinic environment.

Several sub themes are present, and these are organised in a 'loose' metaphor of a theatrical production, including sections entitled 'Setting the scene' and 'Film Camera Action', to 'Performing silent witness'. The director of the whole production (the neurologist) and her team of researchers create new knowledge from the performances of the clinic participants according to the script of the research programme. Additional material may be used in the production of the new knowledge; the personal and private troubles of affected patients may be considered useful to the script, or may be discarded as superfluous. The research script is in continuous development and is widely distributed to other theatres and directors, in preparation for the future major production of medical experimentation. In this performance, genetics is no longer in the limelight and the lead role has passed to neuroscience.

Setting the scene

The Huntington's disease research clinic takes place in the Special Research Centre of the University Medical Centre. The centre is oddly placed on the hospital campus and difficult to find for the first time visitor. There is not much signage to indicate its presence from the outside of the building, and the inside route from the main hospital reception is complicated with corridors, lifts and walkways to follow. My fieldnotes from my first visit gave an impression.

The entrance to the research centre is reached from inside the main hospital building, and the hospital post room is situated immediately outside the doors to the research centre. This struck me as rather odd because it makes the research centre difficult to find when arriving from inside the main hospital, and many patients are keen to tell how long they have been roaming the corridors trying to find the right place. The post room area at the entrance to the clinic is a well worn, busy working environment, there are often large mail cages left haphazardly in the corridor, adding to the 'industrialised-working' feel of this approach area before you reach the research centre. There is a small office opposite the post room where some of the ancillary staff congregate in the late afternoon when they are ready to go home. They gather around the doorway with their coats and bags, and are lively and noisy. There is another official entrance to the main building which houses the research centre, but access is via steps and unsuitable for patients in wheelchairs. The corridor access from this official main entrance goes right through to the post room entrance, it is used by some people who have nothing to do with the research, on clinic days various people often arrive in the corridor passing through on their way to somewhere else. (Extract from fieldnotes, Feb 2007)

The Huntington's research is not the only work that takes place in this centre, other research groups from within the University use the space on a sessional basis. Occasionally, they are around on HD research clinic days, but remain very much in the background. There are also permanent research centre staff in an office who oversee the running of the centre. I described the physical layout in fieldnotes when I first attended the clinic:

Most of the research rooms are placed along a corridor which runs ahead lengthways through the building from the reception and entrance. The facilities consist of several examination rooms & consulting rooms, a room which is like a small hospital ward, a staff kitchen for making tea and coffee, and a room with computing equipment for staff using the centre. There is a general reception area, plus a backroom to the reception, where the research centre staff do their own work, and an office occupied by the research centre manager and another person. These people keep very much to themselves during research clinics and are barely visible to patients. They occasionally come out of their rooms (they keep their doors open however), and walk along the corridor, or may leave the research centre via the post room entrance. They may acknowledge the researchers, but have little interaction with them. Occasionally they are approached and asked something by patients, and then they will alert the researchers. The research centre is quiet and efficient, with few signs of the more usual hospital activities. There are no in-patients or clinical staff in uniforms, no movement of trolleys or treatment paraphernalia, it's rather like the reception area of a large successful company. (Extract from fieldnotes Feb 2007)

In contrast to the rest of the hospital, the research centre is spacious, with large windows overlooking a new construction site. The lack of hospital equipment and staff in uniforms means it is much quieter than the rest of the hospital public areas and waiting rooms. Once I had explored the whole of the clinic, I thought the reception area was like the 'front stage' (Goffman, 1959), it had been created very differently to the rest of the hospital, and there were areas which were off limits to the public, the 'back stage' areas.

It is very obvious that the research centre is different to the rest of the hospital and it is in contrast to the post room area immediately outside. The décor is pale blue and calm, the waiting area is spacious, carpeted, it has comfortable chairs placed against the walls around the edges, there are often flowers on the curved reception desk. A receptionist (or sometimes a nurse fulfilling this role) waits behind the desk, seated, and is only just visible because the counter is chest high. The research centre is usually quiet, calm and completely different to the bustling noise of the rest of the hospital. When the first set of doors swing closed behind you, the quietness is immediately noticeable. (Extract from fieldnotes Feb 2007)

The research centre appears to give a good impression of the hospital and the urban area to people who travel long distances to attend. Clinic visitors also like the main concourse or meeting area of the hospital, which has coffee shops, a post office, a high street chemist, a

jeweller, a clothes shop, and a book and music store. The concourse and the outside garden areas are accessible for people in wheelchairs, so clinic visitors often use these for a break between research tasks and consultations. Participants often comment on the pleasant surroundings and say how calm and relaxed it is, and they draw comparisons with their local health services.

Clinic participants do not realise that this research building has a different financial and organisational basis than most NHS services or clinics. They typically say things like 'this is like a private hospital', 'we never feel rushed here', 'its so nice to come somewhere people are so helpful and friendly'. The spacious 'front of house' of the reception area is in contrast to the 'back stage' areas which are reached via a set of double doors to the left of the reception desk. Leading to the 'back stage' is a narrow corridor with several smaller rooms, mostly on the right of the corridor. Patients and carers only have access here when they are being escorted to a consulting room by a researcher. The first room is for neurology consultations, it has a small waiting area outside it with a couple of chairs and magazines on a small table.

A corridor runs to the left of the reception area leading to the research facilities. There are two sets of swing doors with glass panels in the corridor, the first doors separate the reception/waiting area from the rest of the centre, the second separate the room used by the neurologist and a couple of other consulting rooms, from the rest of the corridor. This second corridor section is narrow, there is just enough room for two people to pass, and accessed here, patients and staff toilets, kitchen, a larger consulting room with a hospital bed and storage cupboards. At the far end the corridor is broader again, and the largest 'back stage' room is here containing computers, desks and storage cabinets. This is used for research administration on clinic days only, but not exclusively by HD research staff. There are often other people in here working, they seem to stay quiet and don't interact with the HD research clinic staff. The clinic researchers, (the research nurses, the psychologists and the physiotherapist) congregate and wait for patients in this 'back stage room', which is the furthest away from the reception area. The neurologist visits this room, usually before the clinic, and occasionally during clinic between patients. This is usually to speak with one of the researchers, she doesn't stay long, she works in her own room, I have never seen her sit down in here and stay. All the other HD researchers tend to cluster around a group of 3 desks. (Extract from fieldnotes Feb 2007)

The researchers all have separate offices elsewhere in the university, the 'clinic' is created physically for a few hours each week or fortnight, and is a virtual centre at all other times. The temporary nature of the clinic is not immediately apparent. Someone mentioned bringing paperwork in a wheeled trolley, and there was a discussion about not leaving anything in the room at the end of the day.

They set out their own paperwork which they bring with them for clinic day, they may use a laptop they have brought with them, or they may discuss their projects and patients with each other. If there are other researchers from different projects using the room, they tend to sit in the other areas and there is virtually no communication between the HD clinic researchers and the other people, who are usually engrossed in their own work.

The exception to this congregation is the HD charity support worker, who usually sits in the reception area until the patients arrive. She approaches them and may sit to talk with them before they have their first consultation. She often makes tea for the patients and carers while they wait to be seen.

All other administration for the clinic takes place away from it: the letters of invitation to patients are sent from a secretary's office in the genetics department. They use genetics headed paper too! The researchers are based in different locations; in the neurology department of the hospital, or in the Bio-Science Department or the School of Psychology of the University. The patient files are kept in secure storage in the genetics department and obtained just for the clinic day, then returned to secure store. (Extract from fieldnotes March 2007)

The creation of the 'Clinic Day' also has a type of rehearsal, in the form of a pre-clinic meeting. This is where the patients are discussed and the potential for their enrolment in research is assessed informally, I have referred to this in more detail in Chapter four. The researchers will go to and from the 'back stage room' and reception area to see if anyone has arrived and if they are needed to recruit new patients, take care of the consent procedure, or perform particular research tests.

Access to the main performance

Gatekeeping was a major feature of the clinic and it occurred in several layers. There was a 'concealed dispute' over the legitimate use of the research centre in and the gatekeeping was used to try to maintain territorial influence. There was regular clinic attendance by the

charity support worker, I was made aware early in my observations that some research centre staff were unhappy with the arrangement because the person did not hold an Honorary NHS contract. Honorary contracts confer responsibility to act as if bound by NHS rules of confidentiality and respect for patients and data. Unfortunately, this was a source of tension for the research centre staff who had requested compliance with this arrangement. It was not clear if the support worker had been asked directly about it, but the research team were asked and did not pursue any action. This was another level of gatekeeping which produced strong tensions. One of the research team nurses was the main clinic gatekeeper, because she arranged for various observers to attend; her concerns were about the consulting room becoming 'too crowded'.

The charity support worker often comes to the clinic and she almost always sits in on the consultations. The researchers find this helpful, because she deals with non research issues, and the patients get another person to help with their problems, some are practical. For example, how to apply for a wheel chair, or get an increasingly disabled patient to accept that they need one, how to go about getting help from local authorities for support in and out of the home, how to get in touch with charities and agencies. The charity support worker knows many of the patients and families who come here - she often talks to them in the waiting room and she nearly always comes into the main consulting room. I have seen her open the patient's files on the desk and look inside. She may have been getting an address or phone number.

Her presence is a contentious point with the management of the research centre. There's no NHS contract, and this is often mentioned by the research centre staff. They expressed annoyance that no one takes any action. The centre is managed by a committee of mostly NHS people, and they decide on use of the centre. They act as a proxy ethics committee in some ways, maintaining administrative barriers to 'protect' patients in research. Another situation concerned some of the management, the designation of the clinic. Since the change to a combined management and research clinic, some people had said disease management was not a proper use of a research centre. I thought it seemed a bit of a 'cat and mouse' situation; the clinic was necessarily titled a research and management clinic because the previous management clinic in genetics had ceased. Yet the clinic did not have the full facilities or expertise to successfully manage every patient and had to pass some of that work to the charity support worker for instance. But for the research centre management, and to keep the location, the clinic was designated as primarily research. (Extract from field notes no 23. Feb 2008)

The dispute over the proper use of the research centre had spilled over into who was allowed to access the patients. The tension was really a tussle for control over who had clinics here and their purpose. This made visible the differences in performance from NHS based staff and research staff. The leading research scientist had no problems with observers such as myself, trainee Genetic Counsellors, or scientists, or nurses from other geographical areas or health organisations with prior agreement. However, the NHS staff who were uncomfortable with the clinic use and concerns about access to patients, performed a strong sense of responsibility to patients and confidentially, or at least they made this the visible point of their objections.

Gatekeeping also extended to my clinic attendance, but from a different direction. The clinic researchers expressed their discomfort as to why I was making a lot of notes when I was observing, and why couldn't I just record everything? The next section will discuss this in more detail.

The discussion with the researchers about what they and I were doing during clinics occurred in the 'back stage' room after I had observed a neurological consultation and had a short discussion with the researcher and a nurse observer from another part of the UK. One of the local researchers asked me what I was writing, and why do I write so much? I explained I was making notes on the ethnography of the clinic encounters, the consultations, and the research procedures to try to show how these things are achieved. She replied that she gets very nervous being observed in 'micro-detail'. Another researcher joined in the discussion and asked 'what are you doing with the data from the research testing sessions?' I replied that I will only use it with consent from the patients, and I asked if the two researchers will consent to an interview. They said they will consider it.

Then the second researcher presses me on why I take down so much detail of the tests. I explain that I am interested in the routinisation of the tests and how they are performed and received, and how they differ between people with symptoms and people without. One of the researchers then went on to say that having me observe has made her reflect on how she is perceived as a professional in terms of good, bad, indifferent. It has made her conscious that some of the questions and actions she asks of the patients may upset them. She gave an example of a question, 'Do you fall?', and actions they could not perform. The research instrument she uses (a questionnaire) is the best one available, but she is aware of its limitations for some patients, and its not as good as it could be. She agreed that I could use this material, but found the observations very difficult and uncomfortable because she felt she was being judged. She asks me why I don't record the sessions or, type straight into a laptop. I told her I did not have ethical approval for

recording, and I may not observe properly if I rely on audio tape or visual material.

This conversation really made me think hard about what I was doing in the clinic. Do I need to make so many notes- should I make it less obvious? Am I attending to the right things, am I aware of the social activity or am I judging researchers? Is my presence disruptive, am I a spare pair of hands empathising with everyone? I tell the researcher that I would probably feel the same about being observed and she does not have to consent. A while later she tells me that she does more home visits now because it's a better environment for the patients to do the questionnaire and tests, apart from the ten metre walk test. She also says another researcher had asked her about how she asks questions of the patients. This made her feel defensive, then worried that someone else could do it better, easier or kinder? I thought this was quite insightful of the possible effect of her research on the patients, and perhaps it had not been a topic for discussion before in the research team (although other effects of the research had been discussed). The researchers have left the back stage room, I am alone here writing up these notes when an alarm goes off- the alarm control is flashing the message:

'10 bed ward- AFFRAY!'. I moved out of the room into the corridor, but there is no sign of activity, affray or otherwise.

(Extract from fieldnotes October 2007)

Although the interaction with the two clinic researchers had begun in a slightly defensive way on both sides, it had led at least two of us to reflect on what and how we were doing things. One of my first thoughts about the research activities was the impact on the patients, yet this seemed to be a new experience for the researchers, or perhaps they had reconciled themselves to it somehow. My presence as an observer had disrupted the 'taken-for-granted' operation of the research in this case and caused some moments of discomfort for the researchers. The second researcher who had asked what I would do with the data, was more uncomfortable with my presence and note taking of patient responses, rather than being watched herself. It seemed as if she wanted to test my authenticity as a legitimate researcher by checking my intentions regarding the data.

My own reflections about my purpose in the clinic, gave me a route to consider the roles and legitimacies on display and how I might think about them. I followed up on my conversation with the researchers by mentioning it to the lead researcher, that there was some discomfort expressed with observations. She was not unduly worried, she explained she has a great deal of experience in being observed over many years and is comfortable with the procedure, but she remembered being conscious of observers in her early career. Later in the project when I had stopped the observations, she mentioned that she had a few

concerns at the beginning that my presence may affect the consultations in some way, but this had not happened at all, and she was content with the working arrangements.

There were other expressions of questioning legitimacy in the clinic, this time from the charity support worker, whose attendance had been questioned by the research centre staff. During this observation occasion, I found myself having a cup of coffee with the support worker in the clinic kitchen. There was a gap between patient consultations because someone had not attended.

Saw support worker, had coffee, said she hates coming to this clinic. I was surprised. Thinks its too hierarchical, spoke about her time as medical social worker in UK. I was surprised about hierarchy feelings too. Allegedly friendly domain but she says always consultants very much above the rest, she mentioned tensions within research team. Said she didn't think much had changed as far as consultants were concerned (I think they have changed, but didn't say). In genetics they are approachable- maybe its different in other hierarchy, say surgery etc. She's very chatty today, I liked her a bit more. Sometimes can be a bit forbidding, you never know what she thinks, get feeling I've done or said something terrible. But if she hates coming here that might be why she doesn't always look too approachable. Mentioned to Prof, all fine with me observing, worked out way of recruiting via her clinics, great help.

Noticed support worker not shy about looking in patients files even when consultant there, takes notes of names and addresses. She does know a lot of patients, and approaches them in waiting room before they see consultant. She sort of takes part in consultations too. Makes suggestions, tries to help. Will do interview with me, great.

(Extract fieldnotes, July 2007)

I was surprised at the suggestion of tension within the research team and I tried to probe a little but nothing more was forthcoming, just general comments about 'hierarchy'. Later in the project I interviewed the support worker and some of her concerns became clearer. They amounted to concern for the status and funding of the clinic and what was available as a service to the patients and families.

Genetic risk is sidelined

Genetic risk was hardly mentioned during the neurological consultations I observed. Occasionally the patients mentioned it, if they referred to another family member, or they mentioned which parent they inherited the disease from. However, it was rarely a major

point in any discussion, the focus was on the neurology and the data gathering, the ‘what is happening to the patient, in their brain, their body?’ rather than anything else. This was a re-organisation of the disease management work in the neurology research clinic.

I never saw the neurologist refer anyone for genetic counselling or have a discussion about inheritance and risk. However, this may simply mean that I never saw it happen (Bloor, 1997, p.40)

There were different occasions when the non-genetic focus became visible. It was sometimes in the consultation or allied research projects, and on one occasion in the waiting room. I was talking generally with patients and families and recruiting for observations in the consultation and interviews. People were often willing to talk in the waiting area and were open in their opinions about this clinic, which were usually positive. They described how they had come to attend this particular clinic, especially if they were regular attenders or had travelled long distances to get here.

The wife of one man spoke to me while the man was in one of the rooms having some kind of cognitive test with one of the psychologists. The patient’s wife told me they had not had a diagnosis yet from anyone about her husband’s illness. This was their first clinic visit and I was surprised, because I thought people referred into this clinic were definitely genetically tested and aware of their diagnosis. The patient’s wife told me they knew little or nothing of her husband’s parents or family, which meant there was no definite family history to include in making a diagnosis. They had been sent to this clinic by their local neurologist who had said he could not do anything for the patient.

The patient’s wife was very talkative and nervous, then she asked me about testing in the context of ‘is there a test he could have to get a diagnosis?’, and I was on the spot. Although I knew there was a test available, I was a clinic observer and not in a position to say if a test would be available or advisable. I said this would be a very good thing to raise with the neurologist during the consultation. I explained as carefully as I could, that not everyone has to have any tests, and many people like to give it a lot of thought. I was very hesitant and wondered if the lady noticed, but she seemed to be more anxious that her daughter could not come with them because she was on holiday. She was on the verge of tears. I was about to offer to get her a cup of tea, when her husband reappeared with the psychologist. Unable to observe couple’s consultation, arranged to observe something else. (Extract from field notes March 2007)

It looked as though the affected person was already doing a cognitive research project with one of the psychologists. How would that work, if there was no official diagnosis? Perhaps testing would provide a first visit benchmark of someone’s’ cognitive abilities.

This was also a management clinic, so perhaps the couple had come here for their diagnosis. I really wanted to see their consultation but was unable to do so, and their situation made me aware of the difficulties of having research and disease care management all together in one clinic. It would seem that the neurologist and the researchers would have to be highly sensitive to individual family situations and carefully negotiate how and when to reveal knowledge.

However, a couple of months later, the same couple came to clinic again, this time with their adult daughter, who had three children of her own. I spoke to them in the waiting room and they were keen to get information about the disease, the family, and how it might affect them. They agreed to an observation and later interviews. In the neurological consultation it was not clear if the mode and the implications of inheritance were evident to the family. They were fairly relaxed and talked to the neurologist about how inheritance might be, explanations were given of the 50% risk in auto-somal dominant conditions to the children, and the 25% risk to the grandchildren. The risk was not dwelt on at all, the affected man sat quietly throughout, his wife and daughter did the talking. They appeared to accept the idea of risk in a very practical, matter-of-fact way. There was no mention of genetic testing or counselling. As the family were leaving the consultation room, the daughter gave me her phone number to arrange an interview, she was extremely keen to take part.

After the consultation, the neurologist and I spoke a little about the case. I asked if she thought they understood what the effects and risks were. The neurologist thought that perhaps they were in denial about the situation. The neurologist asked if I had noticed the patient in the consultation. I said he seemed very quiet, and the neurologist said he seemed to not be taking part, and she suspected he doesn't really know what is going on, so he lets his wife and daughter do the discussion. From the discussion I had observed, there were several 'at risk' adult family members to consider, and I knew from previous work with the genetics department that efforts would normally be made to have contact with those family members, certainly the family's views would be sought. The purpose of contact would be to offer counselling and to discuss the best way forward to deal with the information, and perhaps to arrange to let the wider family know of any possible risk. However, this is not the role of a research clinic, especially a neurological one. This seemed to be a re-organisation of work around genetic risk information within the family; it was not foregrounded in the same way in neurology as it was in genetics. I decided to wait a little

while before I contacted the family for a possible interview, if the diagnosis was rather new, they may need time to work things out before I started to ask them questions.

Patient disappearance

Patients could 'disappear' from consultations if their clinic performance was in conflict with the research direction. On these occasions, patients' responses were sidelined in favour of some other explanation from the carer and this was accepted by the researchers. Patients kept to their own explanations despite this, they repeated their answers but did not challenge their carers strongly.

On one occasion early in my observations, a patient came to clinic with her social services carer. Joan, the patient, was in her forties, had spent most of her life being cared for due to other medical conditions. Due to her increasing disability, her long term care was being re-assessed and the care home where she had spent many years was no longer suitable because she was using a wheelchair. Doorways and lifts were not wide enough, and this meant that parts of the care home became inaccessible for her. Most of the talk concerned the difficulties of finding a care home which could accommodate people with both physical and mental difficulties. Joan, had very bright blue eyes which darted around the consulting room, but she spoke very little and quietly. The discussion turned towards her mood, Joan very quietly said 'feel sad'. When the neurologist asked why that was, she replied even more quietly, 'the house'. The carer immediately gave an alternative explanation. Joan had been upset about moving at first but she had accepted it now, the reason she was sad today was because they had hoped to go to the coast for fish and chips after the clinic but there was not enough time.

There were several performance purposes visible from the observation. Firstly, the carer was an employee and was keen to show the professional skills of caring which included extending quality of life experiences to patients. This was common when professional carers accompanied patients to the clinic. Secondly, the carer had spent time explaining how well the housing problem was being dealt with and how the patient had adjusted. The 'fish and chips sadness' was a repair of the patient's 'house sadness'. Later that day in a post clinic exchange between the neurologist and another researcher, the consultation was discussed but with no mention of either the 'house sadness' or the 'fish and chip sadness'. The sadness reason whatever it may be, had no bearing on the clinic issues, and in any case it was being dealt with outside the clinic boundary.

A further example of patient disappearance in the consultation occurred in a lengthy consent procedure. It was clear that no consent issues were compromised, simply that the patient's response was not taken account of at one point.

This was a re-consenting process due to changes in the arrangements for storage of data, and the researcher who undertook this process was thorough and unambiguous in explanation of the new arrangements.

His wife is very assertive and lively, teases him in a friendly way 'Your attitude is so lax these days'. They talk about things at home while the researcher goes for an information sheet. The wife tells me she works and she had to get up earlier than for work to get here this morning. The talk changes between the couple, its about contact lenses, lifts in the car, taking medication early enough on prior evening, changing tone it's a bit tense, wife a bit critical of husband. The researcher returns, another change in research, questionnaire scoring reports has changed, needs to re-consent that too. Do they have any questions? Researcher goes through consent form, reads it aloud to patient and wife. But his wife says 'Yes', and signs form while husband reads information sheet. Researcher leaves to get a new pen. On return, asks about roads on the way, they discuss traffic in their home town, patient still tries to read information sheet. Researcher asks patient any questions, and reads out this consent form too. There are changes to the time of retention instead of fifteen years, tapes will be retained indefinitely. They move onto the research exercise.

Researcher: Anything changed since your last visit?

(Long pause, 10 -15 secs)

Patient: Tingling, pins and needles really.

Wife: Medication, you didn't bring it did you?

Researcher: Citalopram and venflouxine?

Wife: He still takes those and another night one.

Researcher: I'll ring you at home for that- no problem. So, I'll do your bloods, and the Professor will do the motor exam. I'll take you upstairs to do your questionnaires now.

We all moved to an office in another block of the hospital, in the Neurology department. The questionnaires are all about perceptions in the last week. The patient also does some tests with a psychologist in an adjacent office, this includes a paper and pencil test and a computer test. His wife does a questionnaire on her own of her perceptions of him, we all have tea and biscuits while the couple complete the questionnaires. I look at a blank copy, its like a mood scoring test.

(Extract from fieldnotes October 2007)

During the consenting process, the patient twice tried to read the information sheets and was sidelined when his wife quickly gave consent. However, the patient's position was more noticeable when he responded to the research exercise by saying 'Tingling, pins and

needles really', because this was not remarked on by anyone, or explored. The comment was passed over as if it was never spoken, and instead, the subject of medication is brought up by his wife and this line is followed through by the researcher. During this very thorough re-consenting process, and the research procedure (it took an hour and a half), the couple became a little irritated somehow. The wife began to be sharper with her husband as if she was impatient, and the patient became slightly less co-operative or confused. It was uncomfortable to watch and listen, and I had a feeling of wanting to reassure people, to try to smooth over the cracks. I remained silent and observing. There was no way of telling what the cause of the unease was, the length of time, the nature of the tests and what they might imply, being observed, or something else entirely. The research performance had priority and continued.

Increasing scientific capital

The consent for Euro-HD registry includes a type of 'catch-all', which means that the patients also agree to be contacted about other research projects in the future. So in effect, this is a shortcut to increasing the scientific capital of the research. There is a waiting audience for future developments in the research programmes which will need participants. There are some practical benefits for the scientists and a few consequences. The main benefit for the scientists and researchers is that a ready prepared database of suitable participants is at hand without having to go through further consent procedures in order to contact them. This has the effect of speeding up the recruitment of volunteers for new projects. In one particular instance of experimentation with bio-markers from this project, the reported outcomes highlighted the speedy recruitment as a great advantage over other types of research recruitment (Tabrizzi, et al 2009). This increases the power of the research and the scientists, because the research subjects are poised and ready to be utilised, there is no longer the lingering doubt of not having 'big numbers' to make it meaningful in a quantitative sense, or a possibility of not enough of the 'right' participants. This perpetuates the view that big science needs big numbers to 'prove itself' beyond doubt, and in as short a time as possible. Where future research funds are at stake, it may be that those centres who can say without doubt, 'we have people ready to test this now', will achieve the lion's share of research funding. It does mean however, that patients and families may have increased anticipation about being contacted and increased hopes of experimental treatments.

Performing consent

Researchers performed and achieved informed consent in a variety of ways. There was informed consent 'light', which was used to recruit into the main Euro-HD Registry project, where the minimum of information was given and it was presented as a formality with virtually no impact for the patient. In contrast, there was 'intense' informed consent which took place in an allied project, and lasted for over fifteen minutes, with full explanations and checks to ensure the patient and carer understood the implications. Yet another version was 'cheerful' informed consent, which was delivered as a script and was intended to be reassuring to the patient.

The 'light' version took place in the main neurological consultation during a patient's first clinic visit. I made some brief fieldnotes during the consultation, which was quite short, it only took about twenty minutes compared to the usual forty minutes.

Only two patients in clinic this week. First one is a man on his own, quite tall, retired here from South of England. He has noticeable choreic movements, but shows a very positive outlook. I thought he seemed a bit shy, but he says he is fine with me observing. A brief consult re neurology, the patient has no questions, he feels fit and healthy and has no problems. He has family nearby but doesn't rely on them, he is still driving, will have to stop soon. Agreed to be contacted about research projects, and have data collected. No paperwork done here re consent, presume one of the others does it. After this consultation he went off with one of the psychologists to do tests: put him down 'for all of it' he said, then 'anything to help anybody', usual altruistic type of response. (Extract from fieldnotes July 2007)

Later that day in same clinic;

Discussed ethics with researcher. She shares a personal opinion, ethical requirements sometimes becomes burdensome to patients. Explaining all the information sheet and consent forms puts them off- they think they are signing up to more serious things than they actually are. Very helpful re recruiting for my study, agreed to go through clinic lists and send my project info out with appointments. (Extract from fieldnotes July 2007)

There was a balance to strike in the process of consenting patients. Time was seen as an indicator of seriousness in this environment, so if consent took too long, it could seem to be something more serious than it really was. In addition, the routine-ness of this basic epidemiological research obscured the lengthy retention time of samples and data, and any

possible implications. Literature refers to the potential future difficulties with the present understanding of informed consent ethics in the case of genomic research (Mascalzoni et al, 2008), in neurological work (Glannon, 2006) and in some cases suggests potential solutions (Sharp, 2004). This clinic represented only the tip of a huge consent iceberg in experimental medicine. Researchers and bio ethicists are engaged in discussions about the future of the consent process and what might be possible ways forward (Mascalzoni et al, 2008). This aside however, I was grateful for the chance to contact patients before their clinic appointment; this was practical cooperation which I had hoped for. It also proved to be quite an effective recruitment device for my project after one or two small adjustments.

‘Happy’ informed consent was yet another style in the clinic. The performing of the consent process in a light hearted manner intended to deal effectively with an important but routine process and reassure the patient of the non-invasive nature of the testing. The researcher explained several aspects of the procedure.

I found this procedure a bit uncomfortable to watch like last time I saw it. The researcher is very cheerful, it’s not condescending, but it feels odd. The explanation about this test went like this:

‘In France, they have found some changes in language use of people in the very early stages of HD – we are doing this test to see if it’s the same in English. Ok?’ (well not very encouraging, but honest I suppose).

‘Any risks in this study were with the MRI scan, which is not happening at this hospital as we have no access to the scanner, so there are no risks to you with this test’.

What about the risk of feeling awful I wondered? I wondered if people know how they are doing, and if it upsets them to think they can’t do things, think of things, answer quickly enough. They must be exhausted, it’s a long test over an hour, and quite fast paced. I feel tired trying to listen and concentrate, they must feel that too?.

The patient started well, he was happy and cheerful, list words beginning with a letter, another letter, another, then he gets stuck on the last letter – ‘I keep thinking of animals’ he said. Matching symbols, shapes, colours, names. Numbers backwards, do two things at once, make a shopping list, copy hand movements, remember sentence, make a sentence, day, date, year, building where we are, point to and remember something. That sentence again, read words, non-verbs past tense (wonderful made up words I really wanted to laugh but I resisted this was a very difficult task), tring (Like in Oxfordshire? he asked, researcher not very amused), veed, pravel, ameed, poyn, theep, cless. Long lists of them, many many more. Then normal verbs, long lists of them, faster. I would love to interview this patient later.

The man stayed in a fairly good mood but did seem a little tired and irritated after about forty five minutes, he was sort of deflated, perhaps he knew he wasn't doing well at it? There were a few stalling points when he just repeated what the researcher said, she reminded him to say the past tense. The researcher didn't ask him if he wanted to stop, the test just continued. In a bit of a panic realised my car park time was well and truly up, so I had to make apologies and leave before the very end of the test. (Extract from fieldnotes August 2007).

The observation of the consent and the research testing in this example were at odds with each other in some way. The portrayal of the procedure during the consent process was light hearted and happy, yet the execution of the test was very serious and appeared rather intimidating. Of course this was only one example of the test, it may have been an isolated event. At the same time, some of the strange word constructions did sound highly amusing, but this was definitely not a shared amusement. Later research data confirmed to me that patients do indeed get upset with this form of testing, because they can tell when they make errors even if they cannot say the right thing. Some patients decide they will opt out of this testing in future visits. The effect of this on the research data is uncertain, I asked two researchers and got two different answers. One said it does not matter, all the data is useful. The other (who carries out some of the testing) said that it's a bit unhelpful because the tests are in sets, and incomplete sets mean it is difficult to assess.

Creating data

The classification and recording of data was one of the main clinic purposes, and this was carried out in several ways. In the main neurological consultation, questions were posed concerning the activities of daily living and quality of life. The answers were used to create 'scores' in a quantitative form, which would indicate the stage of disease the patient had reached.

The questions and the scoring are very specific, they form part of the standard scale for assessing the extent and progress of disability, the Unified Huntington's Disease Rating Scale (UHDRS). There are six sections or components

1. Motor Assessment
2. Cognitive Assessment
3. Behavioural Assessment
4. Independence Scale
5. Functional Assessment
6. Total Functional Capacity (TFC) (Shoulson & Fahn 1979, Shoulson, 1981)

Huntington's disease is described as having five stages which can be linked with the scores on the Total Functional Capacity (see 6 above, Shoulson & Fahn 1979, Shoulson, 1981, for full details see Nance & Westphal, 2002, p. 483). Briefly, in stages 1 and 2 patients are usually still able to work, drive or travel and enjoy leisure activities. In stage 3 there is a transition from independence to an increased need for supervision and assistance, and in stage 4 and 5, 24 hour supervision and full personal care are usually required. The UHDRS has been subjected to tests and is considered to be a highly reliable gauge of disease progression (Huntington's Study Group, 1996), it is also regularly updated to include other helpful indicators of clinical relevance.

All patients enrolled in the basic European Huntington's Disease Registry project are tested on these scales. In the UK there are currently over 1700 participants to this project (private correspondence EHDN, August 2010).

In the research clinic, the research work means creating this data from the neurological and other research consultations with patients, by means of anonymising patient records, completing questionnaires and scoring reports, doing psychological exercises concerning reasoning and verbal fluency, and then entering this data on a data base. The data base forms a European wide view of Huntington's disease, there are 138 study sites in 19 countries, with 26 study sites in the UK.

Anonymising patients

To anonymise the patient data, a numerical reference is applied to each participant's information, and this is in the form of a bar-code. One of my observations was in the 'back stage' room, where I witnessed the enrolment of a patient onto the European Registry Project.

Something of a ritual that happens is the bar-coding of samples and data, in order to render them semi-anonymous. The barcodes (like those from consumer goods etc) are pre-produced, and apply to a sort of 'kit', that is then registered on the computer as being for that particular patient. In the 'kit' (a sealed plastic bag) are sticky labels showing the barcode, and blood/urine sample containers.

The researchers talked about 'demogs' and what you need to start entering the data, they started by quoting someone's name, date of birth, address, then swiped a hand held scanner across a bar coded sheet, and stuck bar coded labels onto the sample phials, paper tests, and data sheets for the files.

The blood and urine samples, plus all the data with the bar-code ID, are sent off to different centres, in Italy, Germany and the US. The centres then process the samples, register them, and they become part of multi-national data sets for scientists to work on either now or in the future. The researchers in the back stage room also talked about how the couriers sometimes take the wrong packages, lose samples, and occasionally refuse to take blood samples. It was explained that the research team have no control over which couriers to use and some are definitely better than others. It appeared there was a feeling of anxiety and slight annoyance from the researchers that the courier system was not foolproof.

There is also the interesting aspect of a sort of 'league table' for the number of samples despatched to the Italian labs. One of the psychols was looking at it and commented to the others that this centre was top with 10 samples for the month, beating a much bigger research centre. They all seemed very pleased and a bit surprised.

At the beginning of the project, I was told by one of the researchers that originally the samples were going to be kept for 10 years, then it was extended to 15 years, and latterly it was extended to 'indefinitely'. Each occasion the time frame altered, the patients had to be re-consented. The researcher who told me this had carried out a renewed consent when I was observing once, it was quite lengthy, it took ages, the people were getting a bit irritated. It seemed serious for the researcher and a boring formality to the patient's wife. (Extract from field notes August 2007)

There is a complex international research infrastructure to the data collection in this research, the bar-coding kits are supplied in readiness for the collection and cataloguing of as much data as possible, biological, physiological and quantitative. The aim is to take the opportunity to collect as much data as possible from as many research centres as possible, in order to give greater numerical, statistical power to any eventual trials of new drugs or treatments. The samples and data will continue to be used long after the patients can no longer attend the clinic and provide further samples; the re-consenting process has allowed for data and samples to be kept indefinitely, in effect, a bio-bank and data bank which will provide research material for many years.

Despite the best efforts at central organisation of sample collection and delivery, this was outside the control of anyone performing the research. The samples and data were only useful if they reached their destination at the overseas laboratories, although the extent of any unreliability was not detailed, and it may not have been known. The feedback via a type of 'league table' giving the number of samples received from individual research centres, appeared to be slightly motivating to the clinic researchers. The physical distance

between the research being performed in the clinic, and the samples being utilised in laboratories elsewhere in Europe and the US gave an unusual perspective to the clinic. You could speculate that this gave the researchers a greater sense of involvement in a large global project, and some international recognition for their work.

Film, camera, action

There were two types of filming in the research clinic, one of which was of motor skills and was allied to the main Euro-HD project. I was told there is a training package in progress for this filming and all of the neurological movement tests, so that clinicians can uniformly carry out the tests to a particular formula and standard. Some clinicians have their own favourite way of carrying out seemingly standard tests. Standardising the set routines would allow comparison of movement differences across a larger group than the 'national' populations. The films were to be kept indefinitely in the USA and provided a regular observation of pre-symptomatic patients through to the early and middle stage of disease. I observed the filming twice. The neurologist carried out the testing whilst one of the other researchers operated a hand held DVD camera. The filming took place in a separate room near the 'back-stage' room and was very business like.

The neurologist began by asking the patient to follow a finger as it was passed in front of the patient's eyes. At this point the camera was so close to the patient's face it was almost touching it. The neurologist asked the patient to stick out their tongue and keep it out for a while, then to say certain words- 'no ifs ands or buts' and 'kitty kitty kitty'.

The patient was asked to walk along in a straight line, putting the feet close together in a 'heel-to-toe' type of movement. The neurologist then did another type of balance test, (possibly named retro-pulsion,) where the participant stands with their back to the clinician, who tries to pull them backwards by the shoulders whilst the patient resists. The whole test is filmed at close quarters and in great detail. It must have taken perhaps ten or fifteen minutes to complete the filming of the testing which did not seem to unsettle the patient at all. I did wonder about anonymity of the patient, because I had seen medical textbooks from thirty years previously, where photographs of patients had the eyes of the person covered by a black rectangle in an attempt to avoid identification. The filming which took place here had a fair amount of concentration on the eye area and face as part of the test. The completed films were sent to the USA and used to measure the movements and neurological responses in early and pre-manifest stages of the disease. Not everyone was

filmed however, patients who could not walk unaided, and those in the later stages of disease were not filmed, which pointed to the research efforts being concerned with early and pre-disease states.

A second type of filming occurred in the allied physiotherapy study, where patients were filmed walking in a straight line, turning in a small space and walking back towards the camera. The filming of walking usually took place in the corridor which ran the length of the research centre, which did seem rather public. Occasionally, other people using the corridor would have to wait until filming had finished before continuing their route. I observed this filming in detail several times, and on a few occasions, an accompanying carer would remark to me that the patient would never be able to do this at home, or ironically, that this was the best walking they had achieved for some time. The physiotherapy part of the study was aimed at uncovering the cause and extent of falls in HD patients, so there was often a direct benefit for those patients who could take part, where practical suggestions could be made to enable walking to be less hazardous.

Standardisation in the clinic

The research clinic is involved in the standardisation of the disease, the standardisation of data, and the standardisation of patients. This creates a different type of disease to that previously known in the genetics department. The purpose is to create a population ready for experimental medicine. The cognitive and neurological tests are not new, the same tests (or a combination of the same tests) took place during the time they were administered by the genetics department, however there was always some flexibility in the way the tests were performed and interpreted. This probably applied across all medical sites, and must have been noticed and problematised in the Huntington's Euro-HD research, leading to a move towards standardisations of data collection and data entry. I was made aware early in the fieldwork that different professionals will carry out and interpret the tests depending on their interest, expertise and experience. For example, during a discussion with the neurologist and a physiotherapist, I had enquired about a test known as the 'Luria tri-step test'. This name was familiar to me, I had seen it written in files many times, and heard people talk about it, but I did not know what it entailed. It was explained to me that the Luria tri-step test was a particular physical and cognitive test. My field notes explained how it is done:

The Professor took the patient into the side room for an examination, with the physio who is learning to do the movement tests. The tests are physical ones, to help to diagnose the patient, by showing if there are any physical effects which mark disease progression. I had quietly asked the physio about one test –a Luria-tri-step test, I've seen it written in the notes before, but never known what it is. She explained and showed me, it's a complex hand movement which starts with a fist, then open palm and side of hand, then open palm and hand flat. You have to show the patient the movement, without explaining, and see if they can do it. If they can't, you can say 'fist, side, flat', and you have to get them to keep doing it. I found it quite hard, it's not a natural movement and not easy to repeat correctly a few times in quick succession.

(Later in fieldnotes for same observation)

We talked about the Luria tri-step test again. Professor and the physio were saying that some people who had been thought to have the disease and had scores that were 'not good' on the Luria test, went on to have the predictive test but the test was negative. Other people who had a positive genetic test sometimes did well on the Luria test. They discussed how they scored the test too, and how it was difficult to get a clear cut result sometimes. It appeared that the operation of the test and its interpretation was variable. I asked about the continued relevance/adaptation of this test and similar tests when people are becoming more adapted to different movements like texting, and keyboards, and less physically adept in some cases due to lack of general physical activity. They didn't say much about that really, they seemed a little surprised at the question, maybe I don't understand the neurological implications? (Extract from fieldnotes, observation 2, March 2007)

The discussion between the neurologist and the physiotherapist was illuminating because they knew their own ways of carrying out the Luria test, and they also knew that other people did it differently. The test result is recorded on a score sheet as part of the written data, it was not filmed when I observed. The two researchers talked about the ways they did the test and the ways they scored it, and found there were slight differences even then, despite attempts having been made to standardise it. This means that the Luria tri-step test is open to a certain amount of interpretation, and professional judgement. However, it is only one part of a set of diagnostic tests and is not definitive in its own right.

In order to align all the data from different test centres in Europe, the Euro-HD project needs to be co-ordinated, and any variations in testing and data recording are required to be smoothed out. Standardisation across research centres and countries will enable data to be unified and sample size to be increased in the event of any drug trials. The bigger the sample size, the more power will be given to the results. Further examples

of standardisation in the way that data was recorded in files and the advantages were expressed by the researchers and clinicians.

I asked about the forms she was filling in, in the patient's file, it looked like she was ticking boxes and initialling. She said that there has been much greater standardisation with the way the forms are filled in during consultation and this helps her a great deal in picking up the patient at the next visit. It wasn't clear if this was wholly due to the research projects which are ongoing- maybe something to look at later.

She said it's much clearer how the person was last time due to the way things are recorded, so she has a better picture of what their progression is, much faster than searching through pages of longhand notes. (Extract from fieldnotes, observation 3, Feb 2007)

Neurologist procedure in clinic after consultation, dictates notes briefly for letter to file, then completes forms for HD assessment. Very standardised, no personal family social info recorded in file it seems. (But social things are here, in the talk with charity support worker or me).

(Extract from observation 9, August 2007)

The previous type of medical notes, similar to the ones I had seen in archived files, were longhand notes concerning the patient and often the family situation, together with various photocopied forms with varying amounts of test information recorded. The filing of the 'old style notes' was efficient, but not particularly ordered, one might have had to search for a specific piece of information among the many sheets of paper. The current version of a standard form with spaces for numerical 'scores', indicative words of patient condition, and assessment of numerical stage of disease, gave an instant snapshot of the areas most important to the neurologist in re-assessing the patient. The point about this change in recording style is not that it is clearer and more efficient, which it undoubtedly is, but that there is a whole list of signs symptoms, behaviours, and care needs which are not recorded. The disease has become confined to the body, more specifically the brain, rather than the person, the family and the home environment (Clarke et al, 2003). These other areas are no longer dealt with by a 'health service', they are moved on and passed to other outside agencies, such as the GP, social services, and the support worker for the charity. The focus is clearly on the brain function, the effect on the body, and the measurement and recording of these things in detailed, standardised forms. The previous version of Huntington's disease was a local family experience, often distressing with successive generations contacted, prepared for testing if necessary and supported by the NHS during many years of caring. The people were visible in the files, with their particular difficulties documented

and acknowledged. The latest version of the disease is a disembodied model, a neuro-biological process, counted and shaped to fit the requirements of a global research exercise. It has little to do with a health service. What is created by the standardisation of disease is a population in readiness for experimental medicine.

Performing within the clinic ceremony

The questioning by the neurologist during the main consultation encouraged the production of the 'expected' patient and carer roles in the clinic. This was because the Unified Huntington's Disease Rating Scale focused directly on the expected activities and roles of patients and carers in their environments outside the clinic. Patients and carers performed distinct roles in the clinic, based on their knowledge of the medical system, Huntington's disease, and their expectations of the clinic consultation, the accepted version of the 'ceremony of the clinic' (Strong, 1979). The roles taken by patients were often stoical, showing themselves to be still part of the social world, able to make decisions, and knowledgeable about their disease; the carers often took a role which showed them as highly competent carers, yet also defenders of the patient's independence and autonomy.

Tracy and Phil were having a follow up consultation at the clinic. Tracy has HD, Phil is her husband and carer. Phil had spoken to me in the waiting room about his wife's walking. It had deteriorated and she often looked unbalanced, but she thinks she is fine. Phil was worried that she cannot appreciate the change in herself, he noticed that people looked at her and he wondered if they thought she had been drinking.

(Extract from field notes Feb 2008, not a transcript.)

Prof James: How are you?

Tracy: Trying to make the most of life really

Prof James: I last saw you a year ago

Tracy: Yes near Christmas time

Prof James: How do you manage at home?

Tracy: Some I do, some he does

Phil: I do the hoovering

Prof James: How about finances?

Tracy: Work are still paying me, and they are good really

Prof James: And what about driving, have you stopped?

Phil: I do the driving, she does the shopping

Prof James: And things like laundry?

Tracy: Yes I do that.

Prof James: What about medication, what are you taking?

Tracy: Just anti-depressants

Prof James: Is it Citalopram?

Tracy: Yes
Prof James: How much?
Tracy: 20 milligrams a day
(Extract from field notes, Feb 2008)

This exchange gave the basis for the consultation. The questions formed part of the standard scale for assessing the extent and progress of disability (UHDRS). Phil was also taking part and answered some of the questions. As the main carer for Tracy he was key to her well-being, however, his own health had suffered and he had spent time in hospital.

Prof James: (*turns to husband*) And you've been ill too haven't you?
Phil: Yes, they removed my large intestine, because of ulcerative colitis, and I got a post operative infection, I was in ITU for 4 days. We were worried sick about each other, there was no time to arrange anything, the social care that Tracy needs.
Prof James: How did you cope?
Tracy: Well, I just got on you know, being very careful, microwaved meals and that, my mum's ill too, so it's not so good.
Phil: She has a twin sister who calls, but she works so she can't do a lot. I'm getting stronger though, and I work nights.
Prof James: Is there anyone coming in to help?
Phil & Tracy: No, not yet.
Prof James: Might be worth looking into early. What about falls- have you had any this year?
(Extract from field notes Feb 2008)

The first exchange between the couple and the neurologist showed elements of stoicism, but further probing reveals the fragility of the care situation outside the clinic and despite several other family members being close by, there is not an infallible network of help to rely on. Phil raises the issue of the couple not being able to care for each other and their mutual worry about that. The Professor then moves to ask in more detail specifically about falls.

Tracy: Minor falls, a couple of weeks ago
Phil: She fell twice in the lounge a couple of weeks ago
Prof James: So we could be looking at 20 falls a year?
Phil: Well, that includes stumbles, so, full falls, maybe 10
Prof James: How about your mood? How...
Tracy: Well with him being ill, it's quite hard, not being able to get about. It's starting to get back on track a bit.
Prof James:is there anyone helping you with that?
Tracy: I've been seeing the psychiatrist, but she's left- I'm seeing Pamela Jones at SCAT in Mayville
Prof James: Has the citalopram been helpful?
Tracy: Well its ok,
Prof James: Are you irritable?
Tracy: Yes I am

Phil: Well it's better since she's on the tablets, she doesn't bite my head off
(Extract fieldnotes Feb 2008)

There was a rough calculation here of the number of falls per year although there was no reason given at this stage. The neurologist returned to the UHDRS questioning the patient about swallowing, choking and sleeping. These are all areas where problems can arise in HD.

Prof James: Now can I examine you? Will you take your shoes and socks off please?

As the Tracy takes off the shoes with some difficulty, the consultant moves from behind the desk and towards the small examination room, the husband is watching his wife, who stands unsteadily and starts to move towards the examination room.

Phil: Catch her if she falls- it's not my responsibility now!
(Extract fieldnotes Feb 2008)

It was very telling that the husband was aware of his wife's unsteadiness and jokingly refuses responsibility at this point. After all the examples of being a good and aware carer, he willingly gave up the responsibility and announced it to the neurologist. This was a frequent occurrence in the consultations, as soon as the carer saw a chance to pass on the caring responsibility to the neurologist, they did so, even if only for a few minutes. This was a case of recognising medical authority in the clinic environment, plus a shedding of responsibility, in effect, taking a break from caring for a few minutes and it continued while Tracy and the neurologist were out of the room.

During his wife's examination, Phil talks to me, the charity support lady and the student, about his own illness, about how worried he was while he was in hospital and could not look after Tracy. He tells us that she drove to see him, and he was very worried about her driving ability.

Tracy and the consultant return to the room after the examination, and she puts on her shoes again, it's more difficult than taking them off but the Phil waits before offering to help. He fastens the shoes for her.

Prof R is filling in blue forms in the patient file, writing and ticking boxes. The husband replaces his wife's bag on the chair.

Prof R: Ok most important is the depression side of things, but we won't change the medications. Now about research, we have an academic physiotherapist here, she is trying to develop objective measures of physio at home and she can see you at home. If you're interested, she could see you and you do seem very suitable.

Phil: She does do some exercises

Tracy: Lifting my knees, legs etc.

Prof: Keep doing it its all good and won't affect the research.
(Extract fieldnotes Feb 2008)

Phil performed what many carers did in the clinic when the patient left the room, he expressed his own worries and concerns, visibly relaxed and allowed himself a few minutes of respite. This was noticeable in carers, their physical position changed, their voice changed, and in some cases there was slight irritation when the patient returned, but not in this case. Phil's worry about Tracy's driving ability appeared to be well founded because in a previous part of the consultation, the couple had told the neurologist that she was no longer driving. The neurologist completed the UHDRS assessment form (the blue form) for the research project. It became clear why the number of falls per year was queried; there was a research project concerning falls, and the patient was told she seemed 'very suitable'. Additionally, the patient was reassured that whatever exercise she does herself, it would not affect the physiotherapy research adversely. This emphasises the research priority of the clinic. The patient, carer and clinician were all performing their roles in accordance with their previous knowledge of clinics, and the 'expected' social roles and behaviours in the research clinic environment.

Not all patient and carer performances in the clinic were expressed in the 'expected' way in the clinic 'ceremony' (Strong, 1979). Early in the fieldwork period there was a consultation in which one of the family was considered uncooperative and this went against the accepted process of the clinic ceremony. The next section will discuss this as a form of moral work in the clinic.

Moral performances

There are reciprocal expectations which surround the clinic and all parties perform moral work which becomes visible when the clinic ceremony is disrupted, and the expectations are not fulfilled. The following extract illustrates how the moral work is achieved in the clinic interaction and where points of difference can be seen (Featherstone et al, 2006). There was sometimes more than one moral view which held legitimacy and it was not always evident how each party understood the other.

A family arrived, wife, husband and his son, a young man of around twenty years old in a wheelchair who had the juvenile form of the disease.

Extract 1 from field notes, observation 1.

Prof directed her initial talk to the patient, he often looked at his step-mother in a way that seemed to be asking for confirmation of the

question or help with an answer. The step mother did most of the answering about the young man. The father was a little bit defensive but smiling, sometimes not looking directly at his son or at Prof. He spoke fairly quietly, leaning back in his chair with his arms folded but most of the time he looked over Professors head, gazing out of the window. He said this would be the last visit to the clinic, it was getting too difficult to get his son in and out of the car. He said the local authority are not co-operating with arranging transport to get the young man to college, so he has stopped going. Professor asked about getting a suitable vehicle to transport him. The family replied that they have other children and the vehicle they had been offered wouldn't accommodate them all. The local authority wanted to take their own car in part exchange. They thought this was unfair because they wouldn't own the special vehicle but would have to give up their own transport in order to have it. Professor said that it was not possible to do home visits, and there was an uncomfortable pause which no one filled. The father didn't say much, the wife did most of the talking and she looked directly at the Professor, but occasionally the father would nod in agreement. The patient had difficulty responding, replies were often left behind, and the conversation had moved on. It felt quite tense when the transport issue and college were being discussed, the father had his arms folded and he looked out of the window again.

The atmosphere was very controlled in the consulting room, people spoke quietly and deliberately and there was some tension as the local authority was mentioned in connection with transport and college difficulties. The step mother filled most of the 'spaces' in the talk. There seemed an area of stalemate between the father and the consultant both saying there were things they could not do. It was a little challenging, and the clinician's suggestion about transport was turned down. The father had made his point about not coming to the clinic again, so in effect was planning his withdrawal of co-operation due to the transport difficulty.

Later in the consultation the patient, the stepmother and the Professor had moved into the side room for a motor examination. The father was left in the consulting room with me and a student observer.

Extract 2, field notes, observation 1

Pete, the father, started to talk to the student and I. He'd broken his leg very badly falling off a ladder, and he might have to have it amputated. He said the doctors had told him because his bones were so strong, when he'd fallen his ankle had been forced up into his calf bones and they had shattered. He talked about how painful it all was, and how he'd had a lot of morphine. He told us that while in hospital he had insisted that the physio's let him try to put his foot on the floor, because there had been no feeling in it, and the doctor said it

might have to come off. He said that the next morning after putting his foot on the ground and trying to put weight on it, the circulation returned and only then the medical staff started to try to save his leg. Pete said before this they just wanted to amputate. He said he'd talked to other people who'd had their leg amputated and it caused more problems than they had with a damaged leg. He was worried that he wouldn't be able to drive (he was a taxi driver). I remembered thinking that he was talking about himself a lot, and wondered why he did that at his son's hospital appointment. Then I thought maybe it was because he was nervous with 2 strange people in the room and it was a bit quiet. I wondered if he was worried about not working because he would be at home and might have to watch his son get worse and die.

Pete did what many carers did, he explained his own health problems in the consultation 'gap'. He told of the difficult experiences he had with medical staff, and thought they had not paid enough attention to saving his leg. He was concerned about the possibility of being worse off without his leg, and he was suggesting that the medical staff had got it wrong in his case, the inference being that they did not always know best. The talk about his son and transport had sparked some disagreement with the neurologist. He had a struggle with medical authority in this environment, about his own leg, his son, his car. After the consultation, the neurologist dictated her notes, and spoke about the family, including her interpretation of the patient's father.

Excerpt 3, field notes, observation 1.

As Prof dictated a letter after they left the room, she talked to me a bit about the family. Apparently they never bring the drugs even though they are always asked to beforehand. She explained that the father had been in denial about the son for a long time, but now he always arrives and says he won't be bringing his son again as it's getting too difficult. She thinks he's a difficult man. He doesn't get involved in his son's care, the entire burden falls on the step mum, she is stressed and has asked for counselling to help her prepare for the death of the young man, but there is a 9 mth wait. This will be too late. Prof thought it was a great shame they've stopped getting him to college, he was very welcome there and really enjoyed the interaction with other people his age, the staff were great and it gave the family (step-mum) a break. Prof thought that the young man had deteriorated quite a lot since the last visit and he may not have very long to live. She thinks it would be better if the father would try harder to make the most of the last part of the boy's life. She thinks that the father thinks all the agencies owe him everything and if he doesn't get it, he stops co-operating. The father lost his wife to Huntington's disease when the son was very young.

As I mentioned at the beginning of this section, the expectations of people in the clinic lead to moral work. The field notes show aspects of the moral work in the clinic, which had nothing to do with research or management of the disease, but included statements about how people could conduct themselves both within the clinic and dealing with the illness outside the clinic. Pete did not fit this ideal template and was described as a 'difficult man', who did not co-operate if he is requested to. In contrast, Liz, the stepmother was described as 'stressed' and the care of the step son as a 'burden' for her. It was evident in the consultation that the step mother played a more practical and visible caring role, and the father was positioned by the clinician as being outside this. The family had other children, teenagers from previous relationships and a young toddler together. The father was working as a taxi driver, and with a family of six to provide for, the parents must have both felt very pressurised.

It was possible that Pete placed himself at a slight distance from the situation as a way of coping rather than denial of the situation, and a way of retaining his place as a father. The son's illness was out of his control, the care of his son was taken on by his wife, he had to find a place somewhere, and he did this through working and making decisions. The decisions were not always exclusively about the care of the dying son, he was thinking of transport for the rest of the family too. He also thought of the assets to his family by his ownership of his vehicle which seemed important to him, and the impact of a hospital visit on all the family. Yet the hospital visit did not bring any relief in the situation, it was an exercise, a ritual, and it took both parents away from the care of their other children. I had also reacted by thinking the man talked about himself a lot at his son's medical appointment, yet where else was there a space for this? Looked at from Pete's position he did care, he was also proud and wanted to be strong for his family. He did not want his leg amputated because it would make him less able to be strong in working and less able to provide for everyone. He appeared uncooperative in the clinic environment where everything is focussed on the patient. Forgetting the drugs was inconvenient and perhaps careless, but it did not make much difference to the appointment. Likewise, coming to clinic was probably difficult and there was very little benefit for the family. Why should everyone have to re-arrange their lives for a day to accommodate a data collection?

I could see a point here. The description of the family and the perceived shortcomings of their care role, reveal the 'ideal type' of care that clinicians hope their patients will receive.

Ideally, it would include full co-operation with medical staff and their suggestions, and carers would be available at all times with few other commitments and preferably no other priorities. Carers are also expected to make extra effort to give patients enjoyable experiences in the last few years of life, often with little financial or practical support. These 'ideal type' behaviours are part of what society expects and perceives as humane 'good care', yet in the majority of cases they are provided by unpaid, untrained family members and close friends, who have to make sacrifices in their own lives in order to achieve these noble aims. There is some support from Social Services when people become very disabled, yet this often relies on the abilities of the carer to access the right people to begin the process of claiming. There are support organisations but these rely on charitable donations, and availability is patchy. It is clear that the work being done in the criticism or valorisation of carers is moral work, it amounts to the medical profession telling how people should live with disease (Nettleton, 1991; Turner, 2002; Featherstone et al, 2006). When faced with 'less than ideal' carers and patients, the researchers experienced difficult feelings and attempted to balance these somehow.

Another facet of this observation was the difference in the recording of the consultation. When I had viewed old files some years before this research, there was a large amount of social data recorded in longhand notes, concerning the wider family, often written in a similar way to what the clinician had verbally expressed to me about the father in this case. However, the current research has brought more standardisation of recording of patients and consultations, and is limited to the assessment of the scores on the UHDRS scales, in a quantitative format. Yet the moral work was still there in the talk around families and care, although it was not recorded for posterity in the patient's file. After the clinic observation and the talk about this family, I felt that the same patterns of censure and recommendation were apparent in the clinic as had been evident in the old style notes, but they were now placed in 'the talk' and not generally recoverable after the event.

Performing 'moral' support of research

Family carers discourses were concerned with several forms of moral responsibility; responsibility to preserve patient autonomy, to be knowledgeable about the disease, to be an expert carer, to seek treatment and support research. Although the issue of responsibility was hardly directly mentioned, it was implicit in the descriptions of the patients and carers about how they became involved in research. It appeared to be taken

for granted that research participation was a good thing, that a degree of altruism was involved, and that there was always a possibility the patient and family would benefit directly from a research project (Rubin, 2008). Benefit, in this case meant receiving some kind of experimental treatment which would alleviate disease symptoms, or slow down the disease progression. Yet within support for research there was an area of uncertainty regarding appropriate treatment via research, timing of potential research and treatments and the lack of support for the carer.

The mother of an adult man with HD explained her son's rationale for being involved in research and mentioned both a potential benefit to the son and for others in the future.

#... obviously read about research in general and what have you, and said that he'd be willing to enter onto the research. Erm, you know, he's told me – even it was too late for something to help him there might be something there that could help other people or in the future. So he expressed an interest in research and I think they put his name down for certain things, you know.

(Extract Interview 5, Carol, Mother)

This family had discussed the possibilities of research involvement, and the patient's mother explained the 'future thinking' and altruistic reasoning of her son. The phrase 'even if it was too late for something to help him', revealed that receiving some help for himself was an ideal outcome from the patient's point of view.

Located within the moral imperative to support research was the urgent need for care. The next family asked for care, when they could see their daughter's problem had been partly caused by a stay in hospital. They had volunteered for any research, but until this point had not been asked to participate in anything additional to the basic epidemiology of Euro-HD Registry.

We have never been asked though, I've never done ... we've never done research, only the physio, that's all - we've never had anything else and actually it was myself who said to Professor – because we could see the weakness in her bones, legs, could anything be done? And then of course that research programme on physio came in didn't it? And luckily we got.. [it] ... (Extract Interview 3, Enid, Mother)

The mother had asked for help and expressed gratitude that there had been a research programme available which was able to help her daughter. Later she was referred to a community physiotherapist to continue the support. This was not fully acknowledged as

'research' by the carer, it was deemed to be practical help with a physical need. The mother Enid referred to 'luck' quite frequently in her interview, she was quick to spot the problem and the hospital's oversight which had not put physiotherapy in place. It made me question whether this person would have had support if the research had not been there, and if the mother had not asked. Would the daughter's decline in strength have been considered to be part of Huntington's disease, despite it being avoidable? These types of questions recurred throughout the observation and interview processes, and revealed the low expectations people have of available treatments for rehabilitation. As I have mentioned elsewhere in the thesis, there is a care requirement to provide rehabilitation for people who are not expected to recover from major neurological problems, until the end of their lives. In this case, the research clinic played a vital role in giving a practical solution via research.

Interviewing Edward, a man whose wife had died the year before, he gave a description of unreserved support for a stem cell transplant procedure if it had been available. Anna, his wife had died unexpectedly, and the family had striven to keep life interesting and participatory for her throughout her illness. Edward portrayed a high level of support in practical terms from family and a network of friends, and this was unusual. Most carers and families struggle with minimal assistance and lack of financial resources, settling into an uneasy rhythm of periods of calm and crisis management. Even with the 'ideal care' and his support for the research, Edward also had some frustration with the lack of communication about progress.

Interviewer: So with the research that you saw there and you feel that perhaps things haven't moved that far, or it's not communicated that they've moved that far, despite there being a – hint if you like, that stem cells might provide some kind of treatment if not a cure, erm, what sort of view did you have – if you had one – of actually being in this research and ...?

Husband: We'd have done it.

I: You'd have done it?

H: Without question of a doubt. We talked about that. I said that to the neurologist.

I: Yeah? And what was it then that kept you focussed on that?

H: Kids.

I: Your children?

H: Somebody's kids, anybody's kids really. You know, because if you can ... OK, you've got to live with it but do you ... you know, if you can do anything to help somebody else not to have to live with it, that's the approach.

I: Yeah. And so it became something that you felt was important?

H: Very.

I: To the two of you?

H: Yeah. That's why, when they asked us about the research for the training issues, for the consultants, whatever level they were, doctors, we just said yeah, let's do it.

I: And did that take place over time then?

H: Yeah, it was a progressive thing because it was like when she was ... well, she could still walk when we started doing that. And they would just video her walk and video different motor actions, yeah.

(Extract Interview 10, Edward, Husband)

The family were keen to take part in any research even though the stem cell transplant had not materialised, and Edward had referred to the lack of progress of research over the ten years his wife had been deteriorating. His perspective was to do anything to help, to move the situation forward, even if there was no direct benefit to his wife as he had originally hoped. The participation in training for doctors was recognised as valuable to the research effort even though the husband's description was that it was 'just video her walk and video different motor actions'. This had satisfied the patient and carers' need to take part and make a contribution to the research efforts. There was a sense from patients and carers that the decision to be 'in research' was something taken for granted, a way of not missing out on the promised future (Brown and Kraft, 2006; Rubin, 2008), and if not directly benefitting themselves, ensuring a future treatment for other sufferers.

The prospect of patients hoping for some experimental treatment was not lost on the researchers either, who knew that the stem cell transplant programme was a major draw for their clinic recruitment, even though no transplants were taking place at the moment. They acknowledged that news of stem cell 'future promise' had an effect on recruitment at the clinic although they had tried to mitigate the effects of this very early in the research process.

Intvwr: And how much you ... how ... there's a few questions really ... how do you manage the ... any expectations ...

Prof: Yeah. Yeah.

Int: ... that the patients might have about potential treatments?

Prof: Yeah.

Int: Or ... and how knowledgeable do you think most of the patients are?

Prof: Yeah, that's a very interesting question. The expectation issue is a really important one so ... in a way we ... we had to face that very early on because we started looking ... we started trying to translate ... so in fact it isn't stem cells at the moment. Everyone calls it stem cells but it's actually the foetal tissue and hopefully that will pave the way to stem cell therapies but the transplant we started, you know, working towards

a clinical trial, erm, round about 1999 and at that time there was enormous expectation about what these transplants would do. So in fact a huge amount of our time was spent ... a lot of contact with the media, a lot of contact personally with patients actually lowering that expectation and you know, sort of trying to get out the message, this is a highly experimental therapy, works in rodents but it's a huge leap to humans, can't promise it'll, you know, obviously we wouldn't be doing it if we didn't think it had some prospect but you know very, very early days, we can't in any way guarantee it will *ever* be useful. And we ... we actually spent ... put a lot of time and effort into lowering expectations from that point of view which I feel has been ... was a good investment really because I think the worst thing we could have done was to hype up ... and of course the media tends to hype these things up for you.

(Extract Interview 4, Lead Researcher)

The researchers were in a difficult situation because they needed to keep going with the research recruitment, whilst acknowledging that the process would be lengthy and they could not give any time scale or definite answers. They also needed to show that the research could provide recruitment too, because future funding may depend on this. These performances were what Moreiro and Palladino (2005) referred to as 'mutual or parasitic relationships' (Brown, 2005) 'which have become the medium of political engagement within the biomedical domain' (Moreiro and Palladino, 2005).

Moral support for research was almost unquestioned by patients and carers, except for one interviewee who took a slightly different view and showed insight beyond the argument that 'all research is a good thing at all times'. Carol was not the main carer for her son who was still independent but she played a large role in helping him to adapt to the changes he was experiencing. The mother and son had obviously talked at length of their feelings about the disease and research, and found there were some difficult areas of decision making which they may have to face in the future.

Carol: ... I think I've stood back from it a bit more and realising that, you know, in an ideal world, of course, there'll one day be something for HD, even if it's not a cure, something that can alleviate more of the problems, or halt it. But realistically you have to think that might not happen. And it certainly might not happen in time for Stephen. So, we temper it if you know what I mean. I am interested. But I do find sometimes when you're reading about research, it's very technical..... Because parent/carer, is looking for what can be ... I've a friend who's relation died of cancer. They were diagnosed with cancer but there was all sorts of options – they died in the end Jacki right, but there were all sorts of options. And they clung to them. Sometimes with HD, there's

nothing. You've got the diagnosis, you've got the illness, there's a big nothing. You now have to struggle with it as best you can. And that's when you look for hope with research, is that there might be something to alleviate some more ... there might be a way of halting it, that's what you hope for. But realistically, when you're being cold and realistic, you think 'I don't think so'.

Jacki: What is it that makes you realistic? What is it that brings you to that point? As opposed to having ... I mean you mentioned before, I think you said 'false hope' and I wondered where that occurs with you? Where you think 'I need to be realistic'?

Carol: I think as you watch somebody deteriorating you become realistic. When you see the small things – it's not always the big things – when you see the small things going, then you have to be realistic in the fact that if somebody said tomorrow, there was something for HD, my first question would be well what will it do for him? Does it make it go away or does it just halt it? At what point do you halt it? You know there's a point here – in quite a cold and clinical thing – there's a point where the quality of life can be so ... and it's not that way with him at the moment but there is a point where the quality of life is so poor that you've got to ask that big question. Do you want to hold it back? You know – and Stephen and I have ... I'm not religious as in devout but I have a religion. He told me over Christmas, he no longer believes in any religion, he's told me what he wants for a funeral, he wants a humanistic funeral, erm, he doesn't want resuscitation....he's doing a....er...

Jacki: An advance directive?

Carol: Advance directive, which I actually agree with. But when you start doing those things you come up against the cold, hard facts – you begin to realise, once you watch Stephen and you see the small things going, you really do ask yourself, OK so if they found a cure now that's OK, Stephen's quality of life at the moment could be termed OK. Erm, but at what stage is the cure not going to work – I mean is it going to like reverse it or is it just going to hold it like it is? You have to live with reality as well and there are some stages in HD where to be quite frank with you I wouldn't like to see it [happen] to Stephen. I think the quality of life rather than quantity of life can be very important and not just for HD to be honest. There are other illnesses. I'm not for one minute saying you bump people off.

Jacki: No, no, not at all.

Carol: I'm not saying that. But ...Stephen also talks about assisted suicide and especially while he's still capable of doing it. And I don't ... I think those are facts, they're not... it's not what you want but you deal with reality. There's a point where his quality of life will be awful for him maybe and, erm, you deal with reality. So you hope for a

future but you deal on a day to day basis with facts and the facts are that Stephen can no longer write as well as he used to, he's dropping more things than he used to, he's falling more than he used to. He drops his food, he's having a problem with that. Those are facts. The cure, research, is something that floats over there. You don't want it ... not to stop – you want it to go on, particularly for other people; I'm talking future generations but there's a point as the illness progresses, that that gets farther away.

(Pause)

Carol: And you wish it well, you really do, but it's not relevant to what you're dealing with – you're dealing with the day to day life. That is, I'm sure, there are very dedicated bodies doing lots of things but they can't help you with what you're doing there in that day. They... it's somewhere else.

(Extract, Interview 5, Carol, Mother)

This insightful explanation of what the research meant to this mother and her son, went far beyond what I had expected to hear. This family appreciated the research but realised that it was not going to help them directly. The mother referred to the lived reality of the disease, with no treatment available experimental or otherwise. Carol and her son had discussed both quality of life and quality of death; no one else in the study was as frank. This was not faith in future promise for themselves, this was facing reality with stoicism, and realising the length of time and distance between current life and possible treatments of the future.

The people who do the research do not have to face the day to day trials of living with the effects of Huntington's disease, even though they may be sympathetic. The mother makes the important point that the research activities have little to do with the relentless progress of the disease and the struggle to keep going with normal activities of living. She emphasises that there is a point at which the research becomes irrelevant to the patient and their family. It may be important to discuss further the nature of voluntary participation in research when there are no other options for treatment or care, and why research seems to have more status and funding, than care for patients and families (Stockdale, 1999).

Performing expertise and responsibility

During the interviews carers performed a variety of expertise and responsibility actions. They showed their expertise and knowledge of disease, understanding of the patients' needs, and those of the wider family who may be at risk.

The next extract shows the expert care of a man who can no longer eat enough to keep his energy levels stable, and is being fed directly into the stomach by a pump system known as PEG (short for percutaneous endoscopic gastrostomy). The couple, Angela and David, were in the lounge of their home, David was sitting in a special chair with the feeding pump next to him. They had a new kitten who was more interested in exploring beneath furniture than sitting quietly with David. He understood everything, and replied when he could, Sandra often included him in the conversation and I followed her lead.

Angela: His feed has finished now. What's happened? I don't know what's going on. (Laughs) For some reason that overflowed.

Jacki: It's all quite complicated isn't it David really?

David: Very. Very.

Jacki: Do you get on OK with it though?

David: I do. I do. I've got used to it.

Angela: Yeah, you've got used to it now. No, I've only got to just flush some water through there now. When his feed finishes he has a bit of water flushed through it so it doesn't block. OK ... you're all done now.

(Extract , Interview 5- David and Angela)

David was content that I should stay in the room whilst Angela attended to the feed tube, there was little to see and minimum interruption of the interview. Angela explained more about food and how important the high calorie intake was to David.

Angela: Lovely, he's put on weight. Seems a different person altogether. He's not waking up in the night hungry. His speech has improved believe it or not – because I think he was so anxious in the morning, he'd wake up and he'd go 'food, food, food' and that was the first words that came into his mind that he was hungry and 'food'. But now he wakes up in the morning he's not – he's comfortable, he's calm and as you've seen he's speaking. Yeah. He knows what you're on about, he seems a different person since he's had the PEG.

Jacki: Yeah, that's good.

Angela: And he's going to ... if he puts on any more he'll come to weightwatchers with me aren't you. [Laughing].

Jacki: That might be little way off!

Angela: But no you're – David – doing well aren't you? You feel comfortable.

Jacki: Was that a hard decision? To ...

Angela: Erm, on David's point, yes because he actually made a living will and he said he never wanted a PEG.

Jacki: Right.

Angela: But his issues were ... we talked about it and the issues were that David didn't, in final stages, he didn't want a PEG to be inserted to keep him alive and just feed him – final stages. But, he understands by

having this PEG he can have a quality of life now. In the beginning when he made the living will he felt it was just to keep him alive and he didn't want to be kept alive by a tube, that was his wishes. Hard to do but that's what we decided. So he was quite happy with having the PEG – aren't you, now?

David: Oh yes, oh yes.

Jacki: You're obviously a lot more comfortable though aren't you with it so ...

Angela: Yeah, lovely. And he still has some food because I just liquidise it down. Like today, we're having a cooked dinner today – nice kidney beans out of the garden, Dad's garden, butter squash and creamed potatoes, you know, liquidise that and he loves it. Just a little taste, keep him going.

Jacki: Well it's part of life isn't it and you can retain that quality of life as you say and enjoy it ...

(Extract, Interview 5, David and Angela)

The PEG feeding system had made a huge difference to David's life and enabled him to take part in normal interaction again. The couple both appreciated the benefits that the PEG feeding had brought them, in allowing David to relax and not feel hungry all the time. The difference made to David and Angela's lives, by the use of PEG feeding was only possible because Angela had become adept at maintaining the feeding equipment and administering food, and she took on this responsibility willingly. She also included David at family mealtimes so they could continue to share normal food interaction. This was a case of lay expertise in the care of a seriously ill person, expertise which was more visible in everyday life than in the clinic (Prior, 2003; Bury et al, 2005).

There were several important issues involved in the conversation at this point which revealed the responsibilities involved in caring for someone seriously disabled, but who still had the facility to make choices about life. The question of the patient's nutritional well-being was also linked with his strong feelings about the end of his life. It was important that the PEG feeding did not compromise David's wishes regarding the 'living will', and Angela accepted David's views and worked within them. These sensitive skills of understanding and acceptance are in addition to the practical caring for a disabled body for twenty four hours every day.

Preserving patient autonomy

Carers usually sought to preserve the patients' autonomy but faced challenges about their timing of 'helpful' behaviour. The increasing physical disability of the patients was easier to deal with than the cognitive changes which took place quite subtly over a period of time.

'Taking over' was seen as one of the hardest things to do, and was sometimes done very reluctantly because the carers were acutely aware of removing responsibility from the patient. Cognitive decline was a particularly difficult issue for carers in terms of the clinical consultation. They recognised that the clinic was about the patient, and that there was no official route for carers' worries or concerns. Although this was accepted by the carers and was described 'as it should be', there were problems which affected the accuracy of information giving by patients to the researchers. There was often tension in the thought of expressing a different view or of compromising the patient's independence. Carers expressed major concerns about whether the patient gave the clinician a true picture of 'how things are', with the majority feeling that the patient was hiding the extent of their illness. This was a source of worry, because carers said they do not get a chance to speak to the clinician privately, and sometimes they had a different version of events to the one told by the patient.

Carol, the mother of Stephen explained how she tried to protect Stephen's autonomy and the trust relationship she has with him.

Carol: Yeah. Stephen would not necessarily agree with me. Probably if you had Stephen here I would not be saying some of this. Because I know it would raise a problem. Erm, I would long to be able to feed in some more information to the clinic and have their take on it but I'm aware that they could not then perhaps say it to Stephen because there'd only be one source it could have come from. It's almost like 'grassing' to me (laughs). That feeling of, whoa I've got to kind of keep this to myself and hope that somewhere along the way the clinic picks it up or ...

Jacki: So, would you ... how would you describe that because you know, er, I mean correct me if I'm wrong because I feel as if you are, in considering those things, perhaps trying to do several things at once which would be – preserve Stephen's independence, erm, preserve his privacy in the way that you might expect a doctor to preserve the privacy of a patient, and the other thing is preserving your relationship with him.

Carol: Yes. It's all in there. It would be so easy by certain actions putting a lot of that under threat ... I want Stephen to be – I think we've all come to the conclusion that Stephen needs to be independent for as long as is physically possible. Which is a great statement, isn't that a smashing statement, you know, just like that.
(Interview 6, Carol, Mother)

The carers marked a definite line between the medical needs of the patient being legitimate clinic business, and their own caring difficulties not having legitimacy in this forum. It appeared there was an area of 'no man's land' between the patient experience of reality as told by the patient in the clinic, and the realities experienced by carers and family members. The carers were wary of crossing boundaries in this area, for fear of compromising patient autonomy, and their relationship with the patient. In this carer's case, there was no identifiable 'space' found in the clinic consultation where the carer could speak confidentially to a researcher or the neurologist without the patient knowing. There was realisation that the patient was still in charge, but concern from the carer that the wrong picture was being given to the neurologist. Carol revealed that she thought sometimes she ought to keep things to herself, but also wanted the clinic visit to 'pick up on it' somehow. This has potential implications for management and care, because the clinician can only offer appropriate care if they have access to the full picture of the patient's condition.

The next extract illustrates how one carer did find a way to bring issues up during the consultation, but this caused him concern about 'taking over'.

Jacki: Yeah. And you also felt that [patient] didn't like to answer, as well as that being part of the disease.

Edward: Mm.

Jacki: So, latterly you were perhaps answering for her and a bit concerned that came over as you taking over but I'm sure it wasn't.

Edward: No, you're a hundred per cent right there. That's exactly how it felt because she would just nod and say 'yeah, that's fine'. She'd ask a question ... and sometimes it wasn't fine and I'd say that, 'no, it's not quite' because ... she'd say, 'how do you find your balance?', Sara would say, if you were talking about balance and she'd say 'fine' and I'd have to tell Sara that it's actually not fine, there's an issue there. And she'd be fine then when they'd start talking about it but she wouldn't say about it. Do you see ?

(Interview 10, Edward, Husband, main carer)

The husband explained two things in this extract, firstly that there was an issue with the disease progress because he felt his wife was 'masking' the extent of her problems by not answering things and saying 'fine', and that this was part of the disease. He showed his knowledge of the disease and of his wife and her difficulties. He also showed how he created a space to introduce a more accurate picture of his wife's disease progression, but that it caused him worry about taking away her independence in the consultation.

The implication is that the recording of disease progression and scoring on UHDRS is dependant on the accuracy of both versions of reality, that of the patient and of the carer, and also to an extent, what the researcher believes is the right version of events. With the further standardisation of testing and recording this could potentially be an area of difficulty/compromise in assessment.

Performing ‘the patient voice’

When a patient became very disabled and unable to speak easily for themselves, the carer often continued their participation by proxy, and this happened frequently in interviews. There were also instances of ‘telling by proxy’ when the patient was not present.

It was often the case that a carer would begin their account of the patient’s illness from the time before the disease was apparent with stories of patients’ lives, experiences and accomplishments. One occasion when this happened I heard a patient’s life story from his friend who attended the clinic with him. Mike, the patient, was in one of the consulting rooms taking a cognitive test, and Liz agreed to talk to me with his permission.

The woman introduced herself as Liz, she looked about thirty or so, and was casually dressed, and quietly spoken. She lived near her friend Mike, the patient, and had taken it on herself to be around, and ‘protect him’ from others in the neighbourhood who she felt were taking advantage of his disability. The patient, Mike, had movement problems and also liked to drink in the pub, a combination which brought a lot of misunderstanding about his condition from the people who lived near him. Liz had known Mike since childhood, they grew up living near each other. Other family members were no longer nearby and he was having difficulty coping alone. Liz told me that people from the pub had been going back to Mike’s home regularly, and they continued their drinking there until the early hours, some of them took drugs and ended up staying for days at a time. This caused problems with the home help Mike was supposed to have, sometimes the helpers could not clear up the house because of the other people there, at other times they could not get access at all. Money and other items had gone missing from Mike’s home.

Mike was not able to deal with things when he had been drinking, his friends from the pub were making his life more difficult to manage, and Liz was not sure if they knew about Mike having Huntington’s disease. Liz was training to be a social worker, and had taken the necessary steps to get Mike registered as a vulnerable person, because several times the police had been called to his home regarding the noise from the after hours drinking sessions, and they had not been

very helpful to him. Liz explained that now the local police knew the situation, they were supportive of Mike and had cleared people out of his house once or twice.

There were some family problems in the past with his father but Liz didn't know exactly what these were, and his brother, who never came to see Mike, appeared to have cut himself off from the family. Liz explained that she was a bit worried because having no next of kin nearby, it would eventually be difficult for Mike to express what he wanted or have someone speaking on his behalf. She had phoned the brother once when Mike's home situation was really difficult but the brother did not want to get involved and asked her not to phone again.
(Informal interview 11, clinic waiting room)

This extract shows the sometimes chaotic life of a person who was unable to protect themselves due to increasing physical and cognitive disability. Liz was being a responsible adult in caring for his social position, and protecting a vulnerable person. By telling Mike's story, she legitimised her position as the person who is making his life easier and more organised. Mike was fortunate in having a friend to look out for him and alert the right agencies when difficulties arose but this is not everyone's experience. Unfortunately there are people with HD who do not have friends or family to assist them like this. Being involved in the clinic, and having Liz to help him organise himself for visits meant that Mike was on the radar of those agencies and should receive care as his situation progresses. Although this care was not organised or provided by the clinic, there was the opportunity to be 'in the system' and benefit a little.

Performing as 'silent' witness

Family members explained that they had seen the onset of symptoms in the patient several years before diagnosis. In all these cases, they had not immediately discussed it with the patient, but had waited and kept the knowledge to themselves. Most of the family members who kept this knowledge had seen other family members with the disease and knew the progression and prognosis. In the next extract, a sister told me the story of her realisation that her own sister had HD. The sisters had suffered as children due to their mother having Huntington's disease, and when she had died some years before, they thought that was the end of the problem.

Jacki: Can you tell me a bit about that? How you saw some symptoms in your sister and you recognised it – did she as well? And what happened from there?

Theresa: I ... (sighs) ... I don't think she knew anything about the symptoms that I recognised which was because she was outside the door here and – with her husband – and she just went like that and that was enough. I knew ...

Jacki: Just bending forward, that slight ...

Theresa: Movement, yes I **knew** that she had it.

Jacki: Really?

Theresa: Yeah. It was **that** clear.

Jacki: And at that point did you know that it was hereditary, that it was genetic?

Theresa: (Sighs)

Jacki: Because you'd not wanted to know ...

Theresa: I did because ... it was becoming more into the public domain.

Jacki: Right.

Theresa: So I knew from that point of view – it wasn't from any active finding out myself.

Jacki: Mm. But you know, perhaps if you read something or you see something if you've already got some knowledge tucked away it kind of resonates with you a bit doesn't it?

Theresa: It does, it does, yeah.

Jacki: And you'd probably read a paragraph where someone else would skip over it.

Theresa: Yes, so by then I did know.

Jacki: Yeah. Yeah.

Theresa: I did.

Jacki: Was that quite hard for you?

Theresa: Oh, it was awful, it was ... I just ... stuck my head in the sand for about three years. Yeah. I didn't want to face it ... I just ... the thought of going through all that again, I just wanted to run away.

Jacki: Mmm. And how was your sister, did she have any recognition or were you able to discuss it with her or not?

Theresa: No, I didn't mention it to her at that stage but from then on, obviously I was looking. I was looking and the little things – like this kind of thing, you know.

Jacki: Just little movements.

Theresa: Little things like that. Yes. And, erm ... (sighs) I think in her heart, she knew. But I wasn't going to rock the boat by ... in case, if she didn't know I didn't want to put it on her. But I think that in her heart she did know.

Jacki: Mm. So what happened to make you both aware, that you were aware ... if you know what I mean?

Theresa: Well, she ... she took an overdose. She did it twice. And alongside this, she was ... she was married but it wasn't a satisfactory marriage and it was hard to tell ..what was getting to her was ... what may or may not have been in her mind.

(Interview 8, Theresa, Sister, main carer)

Theresa was the main carer for her sister who had HD, and she was very sad telling this story of how she had come to recognise the signs of the disease in her sister but had not

discussed it with her for over three years. She was protecting her younger sister from this awful information ‘in case she didn’t know, I didn’t want to put it on her’, and she carried this knowledge until events overtook them and her sister attempted suicide possibly for reasons unconnected with the disease. Theresa’s story hinged on the deep familial knowledge of gestures and movement which had caused the moment of recognition, and the protection of her sister for several years. Additionally, this was the story of her sister, how she came to have HD, and there were many more rich biographical details in this interview, family stories, personalities, and tragedies, all with the background of Huntington’s disease and its effects.

This interview was reminiscent of the type of story I had seen in the HD files when I first began to research family pedigrees for the insurance project a few years before. This was the type of story which was no longer recorded anywhere, a story which tells so much about what the disease meant to those affected and their families, and how it changed their lives forever.

Conclusion

This chapter has discussed the many types of performance that actors both inside and outside the clinic adopt, and the meanings these have for them. The social roles of patients, carers and researchers are interwoven with the creation of new knowledge about the disease. This knowledge which is currently being repositioned within the framework of neuroscience, is global in scope.

The neurological research version of the disease has now been reconfigured as something quite different to the lived experiences and lay practices of families who live with HD. The current disease version is something measurable in terms of body movements and cognitive function, and once there is a concentration of standard types of measurable symptoms, new HD populations will emerge from the central European data base which is effectively a Huntington’s disease bio-bank. These new populations will be the experimental groups for the testing of drugs and possibly regenerative medicine. Researchers meanwhile, are busy testing, measuring, gathering data and creating standards of the neurological forms of the disease in the emergent HD populations. The industrial scale of the research only becomes apparent when the data is anonymised, bar-coded and stored securely for later retrieval.

Patients and carers know that the chance of any experimental treatment lies in clinic participation and the recording of their research performances at regular clinic visits. This is an important motivator for continued participation, even though on one level they do realise that any benefit of treatment is more likely to occur in a future they will not share. However, this does not dissuade those who are inclined to join the clinic, for whom their strong belief in an eventual cure via this type of research, means they consider their own participation as a valuable and highly moral, social good.

This 'moral' performance also extends to the family carers who in consultations often display their expertise in being aware of the importance of research, and are keen to encourage other carers and patients to get involved. Some of these very vocal and active carers displayed an almost religious zeal for research participation, and were often heavily immersed in local support group activity too. It was evident that keen carers had formed opinions about what constituted ideal care, ideal treatment and ideal participation, and they found ways to perform these activities in the clinic, although there were occasional dissenters, such as Pete, the father of the young man. Very similar ideals were held by and encouraged by the clinic researchers, who could utilise this as a means of increasing the scientific capital produced by data from this clinic. The responsibility for the production of the research data (leading to scientific capital in this case), appeared to be readily taken up by patients and carers, in examples of self surveillance behaviours allied to the clinic objectives, which is a feature of the biomedicalisation agenda (Clarke et al, 2003).

However, the moral work which was a regular feature of clinic interaction was rendered invisible by the standardisation of data collection in the new disease model. Clinic researchers consider their own moral work as an integral part of their job and generally do not pay special attention to it. This was illustrated by the reactions of two researchers to my observations, when they said it made them feel slightly uncomfortable and more aware of how they interact with the patients. The performance of the patient's voice, the protection of the patient from social problems, the silent acceptance of the changes in the patient until clinical diagnosis, all these examples of the carers' moral and emotion work, do not appear in the research data, yet these have a profound effect on research translation because in many cases they enable translation to take place. This appears to have some resonance with the ideas of Ludwick Fleck (1935) as discussed in Chapter 3, where one interpretation of his work was an appreciation of the social processes which created scientific facts, and the extension of communication outside the thought collective which leads to transformation of meaning (Bonah, 2002). In the case of the clinic performances, the social processes which

create scientific facts include the reported self surveillance data and the 'performed' tests data which are converted to hard facts. The extension of communication outside the 'thought collective' (in this case outside the clinic and research environment via patients and carers) leads to a potential transformation of meaning, where experimental treatment could take place.

Chapter Six

Hope created and maintained

Introduction

The reframing of Huntington's disease as a set of neurological symptoms has been instrumental in creating an atmosphere of hope for treatment and cure around the disease.

The future, both imagined and discussed, is a key feature of patients', carers' and clinicians' discourse in and around the clinic. Patients and carers seek to derive benefit from attending the clinic and being part of a hopeful, forward looking trajectory. The prospect of foetal cell transplantation and later, stem cell transplantation has increased expectations in the Huntington's disease community.

It is important to understand how patients' and carers' hopes for the future treatment for HD are made visible in the clinic, and also to understand why patients and carers perceive Huntington's disease differently in neurology research. The previous genetics framework of HD identified the disease as 'hereditary and incurable'. I will discuss how the clinic has fuelled the hopes of patients, carers and clinicians for future treatments for Huntington's disease, and has begun to break down the view that there was nothing to be done.

Hope was evident in the clinic observations of the discourse from patients, carers, and clinicians, many of whom mentioned research and potential treatments during consultations. The hopes expressed by carers (and often by them as proxy for the patients) generally fell into two distinct categories. These were either 1) a return to a former capable state for their loved one, in one or more dimensions which had become difficult, for example eating or walking, or 2) they hoped the patient would be eligible to receive invasive surgical treatment to the brain with foetal cell implantation, despite the project being highly experimental and under suspension for the time being in the UK.

Researchers carefully utilised hope as a motivator for continued clinic attendance and support of research activities. They employed ambiguity and the rhetoric of hope as

a means of keeping the patients and carers engaged with the clinic activities, and aligning the patients and carers with the research aims and objectives, whilst maintaining an ethical position about research expectations. Researchers and clinicians were circumspect and predicted a longer trajectory for research success, but also expressed firm beliefs of success in stages. They were convinced of the need for large numbers of human samples, a strategy which they anticipated would primarily help to develop drugs to delay symptoms and slow the onset of disease. Foetal cell transplantation was a more distant horizon for researchers but they were very aware of the motivating effect of talk of potential future transplants on their patients.

I will show how the hopes of patients and carers were both managed and encouraged in the clinic by the researchers. I will also explain how the influence of the clinic goes far beyond the individual appointment and maintains the hopes of patients and carers between visits.

Managing hope and expectations in the clinic

When patients and carers attended the clinic, they were very keen to obtain more information about current research. They typically said things like ‘I’d really like to know exactly what research is going on, even if I don’t understand all of it’ (Carers 1, 5, 6 & 10), or they referred to press and media articles to open the subject for discussion. The response of the clinician or researcher depended greatly on the context, but on the whole research enquiries were dealt with very optimistically towards patients and family members. Patients and carers were reassured that they were ‘in the right place’ to hear research news, and that the research was moving quickly and would bring benefits eventually. The researchers themselves included their own and collective hope in conversations, emphasizing their desire to bring about treatment for people suffering.

To give an example, the husband of one patient mentioned that HD research was not often seen in the press. This prompted a broad but optimistic response from the clinician, who explained the research aims of this clinic, and the wider European context. She gave an indication of the scale of the European research project, and

balanced this with the rarity of the disease possibly being the reason for the lack of press attention.

This was building the carer's faith in the research clinic, and including him in the 'clinic research talk' even though he could not 'see' the research himself. By giving this inside information (and also warnings about premature news of progress), the clinician was managing the hopes of this couple. It was necessary to be both encouraging and measured in her approach.

Professor James gave a thumbnail sketch of the research position; she explained the post-genetic test, post-gene explosion of research. The huge number of laboratory based tests was mentioned, and that the aim was to find a drug to slow down the progression of the disease. Then she explained the 'big aims' of the Euro HD project, that these were two fold, to facilitate and increase research, and to have a big group of patients ready for drug trials. She also explained that the projects tend not to hit the press headlines because the disease is less common than Parkinson's disease for example. However, she pointed out that this is quite a good thing, although people do regard Huntington's as a 'model condition' more than other neurological conditions. The clinician then gave a warning about being hopeful regarding headlines in the press. She advised them to try to read what the actual studies say rather than the press interpretations. (Extract from fieldnotes 23, November 2007)

Professor James' description gave the research a much larger, more dramatic scale and contrasted with the husband's view of 'not seeing much in the press', because she provided a context for the research in the form of 'post-genetic test progress' and 'explosion'. The clinician referred to the huge number of laboratory based tests, as a way of describing the unseen activities taking place on an industrial scale, out of view of the public and occasionally hinted at via controlled press release. The emphasis of the explanation was 'big science' getting ready to intervene, on a scale previously

unheard of and the general tone was highly optimistic without any mention of a timescale or a cure. Professor James spoke with enthusiasm for the scale of the research and balanced this with the rarity of Huntington's disease, and a reference to Parkinson's disease being more common. It remained ambiguous whether 'a good thing' referred to the lower incidence of Huntington's disease or the lack of press coverage because it avoided people being too hopeful too soon. In another way, this could have been saying that the lack of press coverage gave more time to do the science without the pressure of public expectations. In referring to Huntington's as a model for other neurological conditions, Professor James subtly gives the research high status and increased importance, indicating that other neurological conditions will benefit from this key research, which aims to find solutions. The Professor's explanation was built on the contrast between the large industrial scale research for a small (rare) affected population, and the huge importance of the research in terms of neurological disease generally, plus the warning against too much early optimism and press hyperbole. The clinician's task is to balance the enthusiasm and optimism for research with the reality of the translation into clinical practice, hence the ambiguity of the research message. The aim is to keep patients and carers engaged with the research aims, yet not raise expectations to unmanageable levels which could be harmful to the research, and possibly the patient's frame of mind.

High rates of optimism and the patient's unending quest for treatment lead to more direct requests for experimental brain surgery. This raises the problem of surgical tourism for the health service in the UK, who may have to deal with the after effects of patients travelling abroad and for the patients who may pay large amounts of money for unlicensed and ineffective treatments (Campbell, 2010) The prospect of foetal or stem cell transplantation has caught the public imagination so much in the case of neurological diseases that there is a ready supply of willing participants, even though the technology is not yet proven and is not yet ready (Cameron, 2010).

Julia and Adrian, a young couple, in their forties, had travelled several hundred miles from another part of the country to attend the clinic. Julia was affected with HD and used a wheelchair, she was accompanied by her husband and full time carer, Adrian. In the waiting room, the couple and I had an interesting and lively conversation

about their recent holiday, they proudly showed Julia's electric wheelchair and listed where it had been in the world. This discussion continued into the consulting room, and the couple told their 'holiday story' to Professor James. They presented themselves as fully participating in society, willing and able to benefit from travel, technology and conversation. Once in the consulting room Professor James tried to steer the conversation to the purpose of their visit and initially there was resistance from Adrian, but compliance from Julia.

Prof James: Now, you usually see another consultant, but you're interested in the transplant programme so you've come along here. Well, it may not be that simple, but I'll explain.

Adrian: This is what we do (shows travel brochure to consultant)

Julia: We listened to Dave Smith [*pseudonym, of person who had spoken about transplant*]/s at the conference.

Adrian: Julia was very impressed and keen to

Prof James: I will explain everything, but I must tell you it is *very* experimental. Can you tell me, how long have you had HD?

In the initial stage of the consultation, Professor James sets out the reason for the couple's visit, but Adrian was still keen to discuss the holiday. Julia offered her conference experience as an opening, and the name of the speaker as a reference. Professor James immediately returned to Julia's medical condition which placed the couple firmly back into the position of patient and carer in front of the doctor.

Julia: Ten years

Prof James: So if we go back twelve years, you were perfectly ok?

Julia: Well, no, my hand was shaking. I'd gone to the doctor's to get tablets for it.

Adrian: We were aware of HD in the family history, Julia's mother had it. But her father took the children away, so she didn't see her. It was 1996, she had a nasty fall, but after that she seemed to become

accident prone. Then it was her fingers, in 1997, when she was driving the car, I could see them moving, but there were no other symptoms. Anyway, because of the family history we had to see a specialist. I mean she'd seen one in 1989, she saw a Professor, and there were no symptoms, but he thought she had it. So the test was later, and the HD was diagnosed. She was given tetrabenzine, but she deteriorated, it was terrible. The doctor would not take her off it, but we complained and we saw someone else, thank god. They took her off the drugs and she was better immediately. So after that we transferred to Dr. Lawrence.

Prof James: There is a very good set up there, you're in good hands.

Professor James validated Adrian's account without commenting on the disease history. The couples' previous encounter with 'a Professor' was mentioned and dismissed by Adrian, but from this information we can see that the previously consulted Professor had given his medical opinion based on clinical consultation. The diagnostic or confirmatory genetic test had come later. Adrian continued:

(Adrian gave a list of medications and Professor James wrote notes about these and family history in the file, while asking one or two questions about a fish oil trial and swallowing capability.)

Adrian: Previous medical history, pneumonia, there was a swallowing problem and food got into the trachea.

Prof James: Do you get around at home? In your chair or around the furniture?

Julia: Yes

Prof James: Any other illnesses?

The Professor began to direct her questions to Julia who made two responses before Adrian took up the disease progression story again. He was dedicated to being the most accurate witness to his wife's condition, and played down her increasing symptoms.

Adrian: She's healthier than me! Sorry, I'm taking over Julia, you want to explain?

Julia: I get dressed, just the fiddly bits are difficult, I wash myself.

Adrian: She does the housework, not the ironing though

Prof James: How about handling money?

Julia: That's fine I can do that.

Prof James: Family finances?

Adrian: I'm in charge but she is involved.

Prof James: Do you use the phone?

Julia: Yes

Prof James: Cooking meals?

Adrian: I do that.

Professor James watched as Julia reversed the wheelchair and got out, she took a few steps. She sat back in it. Then the neurological examination took place, in the main room this time, following the finger with eye movements, finger tapping, stretching out the arms, sticking out the tongue.

Prof James: You're right handed, has this stayed or do you use the left more?

At the end of this examination, the Professor continued with the conversation.

Prof James: Your intellectual function is so good, that's what's keeping you going so well.

Adrian: Speaking as the main carer, there are some problems with compulsion, sometimes she gets into scrapes because she's so determined and likes to keep to her routine. Deviation from it is very difficult, like you know, she's fanatical about keeping the house clean. That's to do with the family history, because from the age of six she kept the place clean for her baby brothers, would you agree Julia?

Julia: Yes.

(Adrian is now very relaxed, dangling his arms behind the chair, he appears very comfortable in this environment).

Julia's disease progression was relayed by her husband Adrian, who was claiming that her signs of disease were always minimal and isolated. Yet, he realised at one point that he was speaking for his wife a great deal, and apologised, saying he was 'taking over'. Julia was able to answer again briefly, but when intellectual function was mentioned as 'keeping her going well', he switched roles and as the 'main carer' explained his concerns about compulsion and strict routines. This was a definite switch from the attempt to define Julia as being minimally affected and potentially the 'right' sort of patient for a transplant treatment. His reasoning about her early life came across as back tracking; Adrian was caught between wanting to have his real concerns as main carer taken into account, and the picture he had tried to create of Julia as a perfect transplant candidate. The hope for a cure did not only rest on being in this clinic, it was also dependant on being in the early stages of the disease. In his attempt to achieve 'right type of patient status' for Julia, Adrian had dominated the conversation and also revealed his true concerns about her stage of disease.

Julia and Adrian's visit to the clinic opens a window on the hopes and expectations associated with this particular clinic, the stage of neurological research, and a professional account of the experimental transplant procedure. It is also revealing about the negotiations in the clinic research encounters and how the clinician has to try to manage the hopes and expectations of patients and carers.

Prof James: Let me tell you about the transplant trial. What I most need to tell you is, it is *very* experimental. Basically, we put foetal cells into the part of the brain where the HD is. There are two places in the world where this is done, one in the UK, and one by our collaborators in France, and there are a couple of subtle differences. In the UK, five patients with HD were transplanted, but this is now on hold due to the Human Tissue Act. We had to completely change our environment to do that, because it has to be GMP, good enough for

drug manufacture. I think it will be another year before it restarts. In France, they don't process tissue, it's transplanted direct. The supply of tissue in the UK is from terminations, and we've had ethical approval. The next five patients have to be quite early in the disease stage, and who they will be in UK is decided at a high scientific level, and this will be probably a year before it all starts again. Then over five years, there will be another five, so we can't promise you anything at this stage. It wouldn't be fair. It will all be reviewed after the next patients.

Adrian: Are you trying to ask a question young lady? (To Julia)

Julia: Am I having it now or not?

Prof James: No, not at the moment.

Julia: I want a trial.

Prof James: It's not possible yet. You used to be able to get it on demand in the USA and pay, but it was not strictly controlled. In my opinion, it was money in the wind.

With a clear explanation, the clinician let Adrian and Julia know that there is no possibility of a trial operation at the present time. In addition, when trials recommence in a couple of years time, it will be for people who are in the very early stages of disease. Professor James tried to carefully reduce their expectations without alienating them from the research; she does this by using ambiguity in her response. She emphasised the seriousness of the procedure ('only two places in the world'), the experimental nature, the source of the foetal cells (terminations of pregnancy) and the credentials of the clinic (ethical approval). There is an important disclaimer about the choice of transplant patients 'who they will be, is decided at a high scientific level'; this indicates those decisions will be taken elsewhere, by even more eminent scientists. So, although this clinic has excellent credentials, and ethical approval for very important world class research, there is no influence to be obtained by being seen here. Again, the Professor employed ambiguity to deal with a difficult situation; it was necessary to manage the expectations of patients and to keep the support for research aims high.

The chance to take advantage of current research aims, and the willingness to use experimental technology to achieve it, is a strong motivator for this couple who have travelled to find the best treatment available. They have assumed (like others who attend this clinic) that there is a direct benefit associated with coming here.

The Professor is candid that nothing can be promised and that it would not be fair to make any suggestions, in other words, 'I am being honest with you even though it's not what you want'. Julia's direct request for a 'trial' is then directly turned down. Professor James switched from the soft language of fairness to the direct refusal of a request and qualifies this with a warning regarding unscrupulous money-making by other medical institutions without the necessary credentials. Here, she may have been trying to pre-empt their determination to pursue a cell transplant at any cost.

In the next section of the conversation, there is a struggle for 'control' in the dialogue, with Adrian attempting to show that he knows other people elsewhere, who may be influential.

Adrian: I think we're talking about Julia, about a couple of years. We know a couple of people at Trentsides....

Prof James: Oh who do you know? I started out in Trentsides.

Adrian: Dave Smith and Ann Farrell.

Prof James: Ah, well you are in the same position as her, so we'll have to see how things are progressing. There are more things coming through, I mean ten years ago not much was known about the disease, but its very positive now, there is a lot going on. What we can say is - there is a lot of activity happening through the labs all the time. You are in a position where more trials will become available, but it is all experimental.

Adrian's, attempt to re-establish some control and influence by mentioning people they know at another research centre was reduced somewhat because Professor James also has good connections in the same research centre. Adrian was visibly subdued. The Professor tried to offer something, but much less than brain surgery. Ambiguity

was still being used, especially when she said ‘What we *can* say is’. This strongly indicated what could not be said, i.e., ‘what we cannot say is, there will be a transplant opportunity’. In addition, ‘You are in a position where more trials will become available’, moved the ambiguity in time from the present ‘you are’ to an imagined future ‘will become’. This extract shows clearly how doctor-patient interaction/rhetoric about futures is used to keep people engaged with the clinic.

The consultation was not quite over, and Adrian asked directly for clarification on Julia’s position in the research clinic and possible future research. The question was rather obviously parried by Professor James with a change of subject.

Adrian: Where does Julia stand with what you are doing now?

Prof James: Do you see Dr. Lawrence regularly? Yes? We are part of the network, its probably best if I keep her updated. I don’t know if a transplant will ever be available. I don’t want to drag you over here in one year to say the same thing. But, you are welcome to come anytime to chat.

Adrian: What we need to do Julia, is wait.

Prof James: I can always see you if you’re here.

Adrian: (explaining to Julia) it’s going to be one year before there are any, then another year before any results, then possibly consider...

Julia: (begins to cry and shout) I want it to be now! I thought I could have it! I want to be better!

Prof James: I do understand

Julia: I thought I could be made better (crying, as Adrian comforts her with his arm around her)

At this point I am asked to go and make some tea and coffee for the couple.

Professor James avoided the direct question and moved to include Julia’s usual consultant. This was bringing the couple back to reality and their usual routines and location rather than the quest for treatment. The invitation to return

to this clinic was also ambiguous because it will not change anything, even though the Professor agreed to see the couple. In this way, the Professor avoids closing the door of the research clinic to the couple but also avoids making empty promises. The Professor enrolled me as another person in the clinic to change the focus of the talk and to provide a distraction with refreshments. It was also a signal that the consultation 'business' was concluded.

When I returned with the tea, the consultation was continuing, despite the earlier signals that it should end. Adrian was trying to discuss complex US research on mice and neuronal interface disruption with the Professor, and Julia was still crying and wanting to have a cell transplantation. The Professor looked calm but also keen to bring the consultation to a close.

Julia: You are doing good things, frontline things here. This is the leading research centre, and in London too. Your name is at the front of everyone's lists.

Adrian: (To Julia) So, we'll probably see the Professor in a year's time and if there are any changes, Dr. Lawrence will let us know.

Julia: (crying again) I thought I was having my stems done.

Prof: (spoke quietly and gently) It is major surgery, its drilling into your head...

Jul: I want to be better, I've had enough of this damned illness, just had enough, (sobbing).

The Professor stood up and asked me if I would take the couple into another room so they could finish their drinks, which I did.

Julia's appeal to the reputation of the clinic and to the Professor herself was a final attempt to enlist support for the transplant trial. Her evident disappointment was the conclusion to the visit which had begun so happily with the talk of holidays and travel. Once again, I was enrolled to create a distance between the emotional reaction and the consulting process. I did not mind too much, although it caused me to reflect on my role as observer and how the researchers perceived me in the clinic. I thought I was

becoming more involved, and I thought it might be about time to leave the clinic observations.

Several points are evident from this observation, firstly that the reputations of the research centre and the lead researcher go far beyond the local area, and secondly people are so desperate for treatment that they will consider experimental brain surgery with no guarantees. The Professor was put in a difficult position of having to turn down a request for experimental treatment, with the risk of alienating these people from all the studies.

The management of information and emotions was challenging for both the clinician and the patient and carer, because the latter were so determined to seek a particular type of experimental research. It is a consequence of the move to a neurological framing and management of the disease that more patients will be aware of the neurological research agendas and seek to participate when there is no other treatment available. Whilst the disease framework remained in genetics, even though the cause of the disease was known to be neurological, experimental surgical treatments were not contemplated, although drug therapy for symptoms was sometimes available. It is a question of the timing of the discovery of potential foetal/stem cell implantation, with the social acceptance of neuro-surgery and its increased successes in other conditions, the change in attitudes to the human body and enhancements, the reduction of stigma, the non-acceptance of long term fatal illness, and increasing longevity which all contribute to a situation where people actively seek intervention. In addition, there is an element of health consumerism, which at this time in western medicine expects people to choose technology and healthy options in food and activities as part of their citizenship. We should not be surprised that the most seriously ill people also want to take advantage of the latest technology to improve their quality of life and are willing to push at the scientific boundaries to obtain it. This rather extreme example from the clinic aligns with Clarke et al's concept of customising bodies in the biomedicalisation era (Clarke et al, 2003).

Opting for experimental surgery is also a way to delay facing disease progression and in the case of Huntington's disease, the premature decline towards incapacity and death; it underlines the importance we place on health as part of our

individual social value and identity, and reflects the more general wish to remain healthy and active well into old age.

Patients and carers can be just as determined to refuse treatment as another way of keeping disease progression at a distance. Denying the future can be attractive if it seems to delay disease progression and keep the situation as it is, even though the real effect may be to cause more suffering. This is another situation where the research clinicians are dealing with difficult clinic situations concerning appropriate disease management.

Hope for the patient's recovery

Carers repeatedly expressed their hopes in the clinic for their loved one to return to a former state in some way. They typically told the patients' biographies as people who had enjoyed active outdoor lives, sports and social activities before the illness had reduced their capabilities. This was referred to as part of the patient's identity, such as someone being a 'good eater' or a 'keen walker'. This represented the pre-HD identity of the individual patient and this was often established by the carers early in the consultation, especially when the patient could no longer have this type of conversation. Some patients and carers expressed their former physical and mental activities in the clinic talk in lively terms, it was as if they wanted the clinician and researchers to know that the person in front of them was once fit, active, healthy and vital, and was still that same person despite their increasing disability. One couple talked about appearing in a TV show some years ago, another about feats of strength, and another of multiple outdoor hobbies such as fell walking, camping and canoeing.

The following extract is an example of one of the problems of Huntington's disease suffered by some people, that of extreme hunger. The person loses the capacity to know when they are eating, but can feel hungry. The nutritional requirements of a Huntington's patient are thought to be between four and five thousand calories per day, more than double that of an unaffected person. Mrs Jones' has seen a small improvement in her husband being able to climb the stairs due to a change in drug

dosage. The extract indicates that she would also like to see an improvement in Mr Jones' regarding eating.

The patient, Mr Jones, had begun to lose weight and felt very hungry most of the time. Mrs Jones appeared to be exhausted.

Prof James asks the patient: Do you feel hungry? Yes?

Mr Jones: Yes. (He is very mobile in the wheelchair, he makes grimaces and noises)

Mrs Jones: He has trouble with getting up in the night at least four or five times. He's better if he's been out in the car though. It's a prostate thing, the Dr told me. He's changed his tablets, now its 5 + 5, not the 10, but he's more alert. He can climb the stairs now.

Prof James: The movements, they're not too bad. We could add a sleeping tablet at night? Does he see the dietician?

Mrs Jones: Yes, we have tried things, Suppligen. The thing is, he thinks he's drinking but he's not. Then he puts his head back like he's swallowing.

Prof James: Do you have any help?

The mentioning of the dietician and 'Suppligen' indicated to Professor James that the food problem was an ongoing matter. Thickening agents are often added to drinks when patients have problems with throat control when swallowing liquids. Professor James was appreciating the increasing difficulties of Mr and Mrs Jones' daily lives.

The clinician and the patient came out of the examination room. Professor James started to discuss the option of PEG¹ feeding. She said she advised this option because the patient was unable to keep himself from being hungry. The use of the PEG device was explained, and she also said it might help to stop the chest infections. Professor James went on to say there is no right or wrong about the device, and

¹ PEG is the acronym for percutaneous endoscopic gastrostomy, a surgical opening in the body directly into the stomach, to allow constant feeding with high calorific food supplements.

no need for a quick decision. There was no rush, and they should take some time to decide but, it would be advisable if they considered having the PEG, to have that within the next six months to a year's time.

Mrs Jones: He thinks he is eating- but he's not.

Mr Jones: (Makes speech sounds- but unintelligible)

Professor James: The gastro-intestinal team will come over and talk about it if you like?

Mrs Jones: We won't have to have it will we?

The PEG system works by directly feeding a balanced nutritional food into the stomach via a pump. Mrs Jones wanted to make sure it was not compulsory, and there was a choice about the procedure.

Professor James: No, not at all, but they can tell you exactly....

Wife: (quietly finishes sentence)...what is involved, mmm. Well I don't want to consider it at the moment. I want him eating again. (Pause- wife laughs nervously, fiddles with envelopes on desk). He's always been a good eater.

Patient: No. (very clear)

Mrs Jones' refusal to consider the option highlights her resistance to disease progression in her husband, she desires a return to his former state of eating hearty meals that she provides. The acceptance of PEG feeding would mean disappointment and not hope. Even the offer of a discussion with the GI surgical team is considered risky by Mrs Jones and she decides it is not relevant at the moment, and reinforces this with her explanation of her husband's 'agitation'.

Mrs Jones: (nervously) He gets agitated you see.

Professor James: Well, you let me know if you want an appointment with the GI team. Now, what drugs is he on?

Mrs Jones: He has a hernia, trimethoprin to help stop the bladder infections.

Professor James: I'll get the occupational therapist to come and look at your house. And we'll see you again in a year.

(Mr and Mrs Jones leave the consulting room)

Professor James dictates a letter to the GP about the occupational therapist visit, and completes the blue forms in the file. She turns to me and says she thinks the wife is not keen to pursue the PEG feeding.

(Extract from observation 18, September 2007)

Mr and Mrs Jones were experiencing the multiple problems of a chronic long term illness, including sleep problems, food problems and movement problems. Professor James had recognized the very difficult situation and that there was little assistance, and she suggested an intervention to relieve the pressure of Mr Jones' constant hunger and Mrs Jones' tiredness. The suggestion was not recognized by the couple because in Mrs Jones' case she was hoping for a return to normal, she said 'I want him eating again, he's always been a good eater'. Their resistance was evident even though Mrs Jones appeared quite a timid, quiet person and Mr Jones could barely speak. It is impossible to say if Mr Jones held the same opinion as his wife, but he was able to say no. The PEG would be an admission that the disease was progressing in a fundamental way which would affect them both. Mrs Jones was not ready to relinquish her hope of her husband eating again. It was not simply a case of getting nutrition into the man's body, there were other feelings and emotions connected with food and eating, which were having an impact on the couple's lives. Professor James was required to identify a technical problem and provide a solution for the patient's well being, and the neurological research clinic environment was not ideal for a detailed discussion about the food issue, hence the proposal to involve the surgical staff. The procedure to create a PEG was not without risks, Professor James and I had talked about this on another occasion, and she had explained that some people die 'on the table' during the operation because the surgery has come

too late. It was of the utmost importance to consider the ability of the patient to recover from surgery and not be in such a weak state that there was no benefit.

The next appointment was in a year's time, which would allow the couple to consider the PEG feeding option. Professor James was in a difficult position; there was a fine balance in giving the optimum treatment and care to a patient, pursuing research objectives, not giving false hope and not criticizing the decisions people made. It means dealing with disappointment and the gradual reduction of hope, plus acceptance of the disease progression as well as optimism about research. This provokes several questions about the role of the clinic and the researchers. There are some conflicting priorities which are not easily resolved; on one hand there is the need to produce excellent research with fully informed participants, and on the other hand it is necessary to deal with the complex social issues of terminal disease.

Reputation, expertise and the creation of hope

The research clinic fulfilled far more than its main research purpose for those people who chose to attend. The clinic held symbolic importance for families because here they were in contact with the best experts, and they also felt they were accepted and understood which did not happen in every medical setting. The clinic was also a way to access specialist advice about palliative drugs, referrals to assistance with day to day living, and held the possibility of experimental treatment. Attending this clinic gave them hope for future treatment. People who live long distances away also specifically asked to attend this clinic and their reasons for attending were similar to the more local patients, but in addition, they said they wanted to come to this clinic in particular because of the expertise and reputation. They knew the research profiles of the clinicians and wanted to be seen by them. Patients and carers typically said they were glad to be able to see the 'top people', and 'to get into this clinic'. They described their attendance at the clinic as lucky, fortunate, as though there was some privilege associated with coming here, rather than voluntary research participation. Therefore it is perhaps unsurprising that the majority of clinic attenders appeared enthusiastic and

keen to be there. It may naturally follow that enthusiastic and keen clinic attenders would be more hopeful than others about what their clinic attendance would achieve.

Angela cared full time for her husband, Dave. She referred to the clinic research 'talk' making her feel hopeful about a future cure for the disease.

Angela: I just feel it's progressed and they've got so much knowledge and, I don't know, I come from there sometimes and I think 'there's hope' that they will find a cure.

Jacki: What sort of things give you that hope?

Angela: The way that they're saying about the research, the amount of research that's being done – although it doesn't sometimes mean a lot to me and I can't see it happening but there's always talk about research, and when you look online, there's always different things they're saying about research isn't there.

(Interview 5, Angela, wife main carer &, patient)

The amount of knowledge gained about Huntington's disease and the way current research is talked about in the clinic was important in maintaining hope for Angela. She trusted in the words of the clinic researchers, whilst acknowledging that some of the research talk was beyond her understanding. It was symbolic of hope to her that expert people were engaged in research, even though she could not physically see it. She was motivated to seek out online research information, and drew her conclusions from these two reliable sources that there was a large amount of research activity, and therefore hope for the future.

Patients and carers mentioned being referred to this clinic by their local neurologists who knew very little about Huntington's disease. Carers often described their local hospitals as having no idea about the disease, or how it affected the patients and the families. Several told of apparently insensitive medical staff that were afraid of the patients and assumed all symptoms were associated with Huntington's disease, which made it difficult to get treatment for anything. This also applied to some GP's who had never seen the disease even if they had many years of experience. This comparing of other medical encounters with the HD research clinic experience was also

a way of expressing the greater expertise of this clinic, and its staff and the knowledge they held about the disease. The experiences people had elsewhere in the medical world meant that they were highly appreciative of the clinic and how they were received. The horror stories people told also underline the effects of a lack of rare disease knowledge outside specialist clinics.

The Huntington's disease support worker, Kate, explained how she thought patients viewed the clinic.

Jacki: And how do you think the patients feel? I mean I know, I do take on board and I've been there when people have said that they've started out quite early and have travelled a long way ...

Kate: Mmm, it's a long day. Yes.

Jacki: What do you think their overall impression is of coming to this clinic and the purpose of it as well?

Kate: Yes. I think a lot of people feel it's the only place they can come where people know anything about Huntington's Disease.

Jacki: Right.

Kate: And that is a comfort that at least ... at least there is somewhere..

Jacki: Yes.

Kate:... so that is a – just important in itself that people know what they're talking about. And so a lot of people like to think the clinic is there in the background and that, you know, it's a resource that can be used.

(Interview Support worker, March 2008)

Using the clinic 'as a resource' highlights the multiple purposes placed on the clinic by families, and goes beyond a simple research centre or a yearly appointment. The need for families to have contact with experts who understand the problems of Huntington's disease is clearly expressed, as is the rarity of finding such a place.

Rebecca is a co-coordinator for the research programme and she explained to me the importance of the clinic for patients and carers.

Jacki: And it's like 'we're here now let the show begin', sort of thing.

Rebecca: Yes, I would agree. It's a big event and it might be an annual visit whether things have changed or has anything new happened as well.

Jacki: It's got a lot of impact on them, how they come in, how they approach the clinic, what happens to them in there and what they're leaving with.

Rebecca: More impact than one might realise I think.

Jacki: I think there is a lot of impact. And I'm just trying to work out – part of my sort of questioning of myself really and what I'm seeing, is how much of that is the research, how much of that is the event and having a visit. Would it be like that if ...

Rebecca: My gut reaction would be, it's the actual visit more than the research.

Rebecca understood that the patients and carers needed the contact with the clinic, and that their visits took on an important role, which was not all based on being in the research. She also appreciated the impact that the visit may have on families, it was an event to prepare for, it was stimulating in some ways but it could also be very tiring. This finding was confirmed by several carers who referred to the extra preparations made for getting to the clinic appointment with the patient. Once there, patients and carers had longer conversations than they usually had, saw more people than they usually did, and had to think about the research tasks which could be stretching. They may also have to confront the disease in ways they perhaps could avoid in their normal day to day lives, which could be emotionally draining. There was however, a benefit to them in the supportive clinic atmosphere and knowing there were other families in similar circumstances.

The meanings of research participation for HD families

Patients and carers considered attendance at the clinic as a positive act towards an eventual cure for Huntington's disease. No one said they came to the clinic specifically to take part in research, most people said the clinic purpose was to 'keep an eye on the patient', or to have 'a chat' with Professor James. However, when asked directly about research, most people did remember that this was one of the clinic purposes.

Clinic participation effectively changed meanings surrounding the disease for patients and carers. The emphasis of this neurologically based clinic is on the individual patient and their current neurological problems. This is a biomedical, quantitative approach, based on the measurement of various neurological functions in a standardised European framework. The previous clinical genetics model used the similar measuring techniques in a more flexible way, but was also concerned with family dynamics and genetic risk assessments. In contrast, the more standardised and extended research programme in the present clinic is solely a biomedicalised approach concerned with the functions of the brain, with the effects evident in the rest of the body. The emphasis of the research as a whole is to find out how to re-enable the brain to function normally in the patient, it is an active process, which is encouraging to people. Isolating the biology to a specific part of the body, separates the disease from the individual person, and allows the disease its own identity rather than focusing on a malfunctioning personality. This removes the moral aspects of potential blame and shame from the disease, in a shift away from genetics and inheritance and allows a further disembodiment of the disease (Mol, 2002). This appears to be something that people respond positively to in the clinic. However, there are also disadvantages to this approach because families and carers do not have a dedicated space in which to seek support and feel cared for; the difficult family and social issues remain outside the clinic boundary for the majority of the time. This was mentioned by several carers and will be further examined in the discussion.

In the following extract, Edward explained what research participation meant for his late wife Anna, and himself. Anna had Huntington's disease and had died

unexpectedly the year before our interview. He told me how vital the clinic was to both of them.

Edward: I obviously realised we were in research if I recall, because of the things that were happening to us when we were out there, but we just participated in it because we could see that (a) there's a monitoring process, it's a work in progress in that respect, and (b) you've got, you know, it's doing some good. I mean that's paramount to us really, in terms of the inevitable is going to happen but can we get that reduced for somebody else if you know what I mean? Or make life easier for somebody else.

(Interview 10, Edward, Carer)

Edward and Anna found a benefit in being part of something larger and believed there would be an eventual practical benefit for someone else. He refers to the 'inevitable' meaning the end of life, in this case for Anna, his wife. He wanted to do something meaningful in spite of this, to make a contribution of some kind, in the face of his own loss. It is difficult for patients and families to participate in many activities that others can take for granted, so research participation can provide a means of being involved in society. The altruistic stance of helping others in the future may also fulfill the families' need to feel that they have a purpose in life, and that they can make a positive contribution. This may assist them to cope with the illness and the effect on their family.

Edward went on to explain his and Anna's frame of mind, and how social attitudes to serious disease and disability were changing.

Edward: Yeah, I think [it's] more aware (sic) now for sure.

Jacki: Yeah, there's more awareness but there's also the social pressure of having a disease and I think we're in a different position now, we see people with long term diseases and socially we treat them

...

Edward: With much more acceptance.

Jacki: ... yeah, in a different way. And I just wondered if the clinic is kind of part of that thought for you? You had a really positive outlook – you and Anna together, you were together on it and you were moving forward.

Edward: Well I think we were, a hundred per cent, but I also think the clinic gave us that positivity as well.

Jacki: Really?

Edward: Mm. Because you feel you're contributing to something as part ... you're participating in it as well as having the progression and monitoring, you are ... the more I think about it, the more I think back, we ... you know, I feel quite proud really of the approach we had and I felt very much part of a team.

Jacki: That's very important isn't it? That's quite an interesting idea, that you were as much a part of that team because without the patients they couldn't do their research anyway so you were quite vital to that

...

Edward: Yeah. Whether it's driven by the personalities involved I don't know but obviously the Professor was there. I mean it was only a short time we knew her really, when you think about it. Every six months or whatever it is but she's got the sort of demeanour that she's there to help you as well as learn from you, if you know what I mean.

It's a two way process then.

Jacki: So there's a lot of trust?

Edward: Oh huge. Huge. Yeah.

(Interview 10, Edward, carer)

Edward was clearly proud of the contribution he and his late wife had made to the research effort, it was important to be able to take part in something that mattered. He felt appreciated by the research staff too, so Edward and Anna's contribution was validated by the experts in the clinic. This indicates the role played by the clinic and its

actors in reducing carer isolation and raising self esteem. The validation by the researchers is also indicative of how important the patients and carers are to the research teams in the effort to find treatments. This continues a relationship pattern with families which began in the Genetics department almost forty years ago.

The patients and carers who attend the clinic today are often well informed about the disease, unlike thirty years ago when the Genetics service began. A retired research scientist explained the motivation to produce a genetics service

Prof Dean: You know starting up, well it wasn't just for genetic counseling, but they couldn't get any decent information about anything. I think partly that was why we met a very good reception because anything was better than what had been there before, because there hadn't been anything there before, and it was just exceptional having Miriam as such an outstanding person.....Well, you know her contribution was an absolutely unique one, and apart from anything else, there was the continuity of it you see.

(Interview, retired research Professor, Jan 2008)

Professor Dean explained the lack of available information for HD families prior to the start of the Genetics service, and refers to a medical social worker who had made contact with many families who had never had any support before. The ideals of continuity and trust were important factors, because there were often several branches of the same family affected, and they came to know the Genetics service, the social worker Miriam, and Professor Dean as sources of information and support. This is continued in the present research clinic, but with the focus more on neurological research. The families who attend the research clinic however, had noticed that they lacked a central supporting medical service for the disease even though the research clinic was helpful. This will be explored further in the discussion chapter.

There were definite social benefits attached to research clinic participation, and these will be explained next. Clinical management now appears as research based, and this changes the status of the participation for the patients and families. Even though,

on one level they describe clinic visits as 'just a chat', or 'keeping an eye on things', there is acknowledgement of the research as a chance to look forward again rather than back.

Social benefits of the clinic

Many people were keen to extend their clinic day into something social, whilst others felt the pressure to return home as soon as possible. Angela and David had been attending the clinic for several years, previously as part of the Genetic service and more recently as part of the neurological research. They gave this account of previous clinic visits and more recent ones.

Jacki: Do you tend to make a day of it then?

Angela: Yes we usually do. And before David was PEG fed we always used to stop on the way back, didn't we? That was our day out. We used to stop at a little pub on the outskirts and go in for a bar meal on the way back. But just, we just stop off now and have a coffee somewhere when we travel back home. And then ... we like that don't we? making a day of it. Sometimes in the summer, ...— if we've got an early appointment, we go into town shopping.

(Interview 5, Angela, wife, main carer)

The couple explained how they liked to make something social of their day away from the home and care routine. Changes had been made to their day out since David had to be fed by PEG, but they compromised and went for coffee and shopping, which were also rare social outings for them.

Alan and his wife Jane, who was very disabled and in a wheelchair, described what they thought about the clinic.

Alan: Well, we just hope the same as everybody else that possibly in a year or two or, nobody knows you know, that there might be a cure for it or possibly a way of holding it back. Other than that we just come

down for a day out more than anything! Change from the supermarket! (laughs)

(Interview 1, Alan, husband, main carer)

These couples had found a way to escape from the mundane routine of home and the burden of caring. In addition, there was an element of hope expressed by Alan, that there may be some treatment available, and this was also part of their reason for coming to the clinic. The issue of hope for research progress, 'same as everybody else...possibly in a year or two', uncovers the motivation to be in the right place when a breakthrough happens, to have a chance of some treatment and not be left out.

Carers also welcomed the chance of interaction with other families at the clinic and this made them feel less isolated in caring for the patient. The social benefits of attending were immensely important and gave them increased self esteem in the job of caring. Angela described her feelings about seeing other people at clinic.

Angela: But we was on research once for that, erm, we had a placebo for the er..

David: Fish. Fish.

Angela: Fish oil, that's right, Dave. And that was lovely because you saw different people and there was a few of us at the same time.

David: Six.

Angela: Six, was there? Yeah, that's right it was yeah. And that was lovely because people had come from way down – Devon, somebody down that way – on this trial and it was... that was lovely for me to talk to other people.

Jacki: Even in that short time that you were there? Sort of an hour or a couple of hours perhaps?

Angela: Yes, yes. That was lovely. I don't know about [?] with David. David was OK.. Because they were more or less all at the same stage of the illness as David. But David felt that he was looking at himself – er, looking at somebody else with HD. David could see

somebody else with the movements and see himself doing it. So it did... he didn't feel so good when there was lots of people there.

(Interview, Angela, wife, carer and David, patient. Feb 2008)

Angela benefitted from seeing other families, but David felt differently. This shows how the needs of carers and patients can be quite different. The isolation of caring for a seriously disabled person with a rare disease means that social contact with others in a similar situation is unusual, yet it can be a very supportive experience. However for the patient, like David, seeing other people with HD can be an unpleasant way to face the disease, seeing your own problems mirrored in someone else. This was something that the clinic researchers were aware of and they tried to accommodate when arranging appointments, because they 'shuffled' the order of patients sometimes, so that patients did not have to face this situation. (Referred to this elsewhere in thesis).

The research clinic reputation has a strong presence and influence beyond the immediate family as Angela explained. The idea of hoping for some future treatment due to the advances in research was not limited to the people who attended the clinic. David has two children, who know they have a fifty percent risk of developing HD.

Angela: At the moment in time because Dave's got two girls, it's nice, because they don't ... they can't handle it and accept it at the moment.

Jacki: Right. Are they teenagers then or ...?

Angela: No, 29, but they don't come and see their Dad or anything.

Jacki: Ah. Right.

Angela: Because they just can't handle the illness. They do like to know when we go down the clinic, 'is there any outcome of the stem cell' – it all seems to boil down to the stem cell all the time.

Jacki: Does it?

Angela: Mmm. You know ...

Jacki: What do they say about it? Is it just that they refer to stem cells or do they ...

Angela: Yes. Yes. Because they think that's the only cure for them.

Jacki: Right.

Angela: If ... you know.

Jacki: So they're not in testing or research or anything at the moment?

Angela: No, no. They can't ... (to David, her husband) I know it's awful to talk about your children like this love, but it is the truth isn't it? They don't come here or ... you know ...

Jacki: (to David) if you don't want to go into that area that's fine – we don't have to talk about that.

Angela: Do you want to or not?

David: What? What?

Angela: Say about the girls and you?

David: Why not?

Angela: Why not. Yeah, they don't see their Dad at all. That's hurtful for David.

Jacki : Right, yeah. But they sort of are still interested because they want to know about the clinic.

Angela: That's me, I try to keep them in touch with their Dad as much as possible because they're not my children – it's a second marriage. Erm, that's my doing – I hope to let them know what's going on.

Jacki: Yeah. Yeah. OK.

Angela: And, er, it's always ... 'What about the stem cell? What about the stem cell?'

Jacki: Right, OK. What do you think about that? About the stem cells ...

Angela: I'd like to know more because all we know is that the government stopped it and then they were building a new lab in Handforth weren't they?

Jacki: Yeah, that's right.

Angela: And I don't know whether it's going or ...

(Interview 5, Angela, wife, main carer, and David patient)

Angela explained clearly that the influence of the clinic went beyond the immediate contact with patients and carers, to the wider 'at risk' community of children, siblings and extended family. The 'stem cell' possibility is evident in the hopes of people who have never attended a research clinic, and shows that there is knowledge to be gained even by an indirect link with the clinic. People at risk of developing Huntington's disease at least know where they may obtain information, even if they cannot engage with it yet.

The changes in family understanding of HD

People who knew of their own family history of the disease, and had seen older relatives with HD, were very aware of the positive social changes towards degenerative disease in the last two decades. Theresa had experienced a difficult and troubling childhood because her mother was affected with HD. She discussed the very different disease experience of her sister, who was found to have Huntington's disease several years after their mother's death.

Jacki: And you'd had that childhood where you didn't really understand ...

... what on earth ... Why things were a bit disjointed, you know, and a bit..

Theresa: Not a clue. Not a clue what was the matter.

Jacki: ... a bit difficult, did you?

Theresa: No idea. No idea what was wrong.

Jacki: Yeah. Yeah. What ... what ... when you saw it on the certificate then, did that stimulate you to find out more or was it ... was more information given then , or...?

Theresa: Well, I asked my GP, I said what do you know about Huntington's disease? Chorea. Huntington's chorea, that what was on there. And he said – 'nothing'. He said 'there's about that much in my

medical books' [makes gesture of small amount between finger and thumb].

So, no. And beyond that I didn't pursue it because I just wanted ... I wanted to close the book on it. That was all, I didn't want to know anything about it at all. Just lock it up and throw away the key.

Jacki: Yeah. Did you perhaps think that that part of your life was over then, that it had finished with your Mum's death and ...?

Theresa: Yeah. I did. I really thought, that's it, that's the end of it.

Hooray. Hooray. I didn't want to find out about it ... I didn't want ... I didn't want to know. Just bury it all away.

Jacki: Completely understandable.

Theresa felt great relief when her mother died because she and her siblings had never been able to understand their mother's behaviour. Information about the disease was not easily available, and there was no clue that the cause was genetic.

Theresa: It was only when I saw the symptoms coming on my sister, which was a long, long, long time after that, er ... and even then, if she hadn't been so keen, she was the one who wanted to join the support group which was a sort of ... in its embryonic form then. And, erm ... and find out – she wanted to know ... know everything. Because she ... she was wonderful, she just sort of met it head on, there was no denial, nothing like that, just met it head on – let's find out about it, get on with it, you know.

(Interview 8, Theresa, Sister, main carer,)

The disease experience for Theresa's affected sister was very different to that of their mother who was ill throughout their childhood. Life for the two young girls was very worrying, and sometimes the social disapproval of their mother extended to them too. People in their community thought that the unusual movements and unpredictable behaviour were caused by drunkenness. Their mother had hidden away, taking walks only at night. The sister's attitude of 'meeting it head on' was in direct contrast to their

previous experience. Social change was evident from the existence of the newly formed support group which indicated a more open and accepting attitude to the disease.

Increased openness and awareness does allow people access to care and potential future treatment, and in the last few years this has included research as part of the package. The research is focused on the individual patient and can be intense, it is therefore not surprising that some people assume they are getting better care from being involved in research.

Research means better treatment

People chose to attend this research clinic because of its reputation and the reputation of the researchers. When asked about their reasons for attending that particular clinic, people expressed their preference for a clinic where research was taking place. They justified this by saying that they wanted to be in the right place to receive the best treatment. This shows that people equate high profile research reputation with the hope or expectation of new and better treatments.

The mother Enid, and brother, of an affected woman explained their reasons for choosing the clinic, even though they had to travel quite a distance to reach it.

Jacki: Yeah. I mean is it a choice that you go to that clinic...?

Enid, Mother: Er, I think Graham (another brother) wanted to go

[he] was going to take us wasn't he, he wanted.....

Alan, Brother: It's probably because it's more renowned for research isn't it?

Enid: Yeah, that's right.

Alan: You want to be in the place where the research is don't you?

Jacki: Right, OK. Do you want to explain a bit more about that to me – that's quite an interesting idea for me to ...

Alan: Well if there's research going on in a hospital then you're in the right place.

Enid: Or you think you are!

Alan: To be helped or assessed, or you'd get the best treatment, the best medication.

Enid: Or you'd be perhaps more so in front of the queue, if it's a research hospital, if they're researching there. That's my way of looking at it.

Alan: I mean if they're researching a certain tablet then you'd be the first ... their patients would be the first to know about it wouldn't they, surely ...? hopefully.

(Interview 3 , Enid, Mother and Alan, brother)

The explanation of clinic choice began with the preference of the driver, Graham, another brother, but quickly turned to the idea of a renowned research centre, so it implies that reputation is involved. There is also the issue of positioning in order to benefit from potential treatment and when this is probed, the assertion is made about being in 'the right place', to get the 'best treatment', the 'best medication', and to receive this best treatment first. There is an assumption, a hope, that being in this clinic will bring preferential treatment even if experimental. The expression 'first in the queue' raises ideas of scarcity, and suggests treatment is not available to everyone at the same time. The assumption is that some advantage will be gained by having new treatment before other people. In most random controlled trial research projects it is completely erroneous to assume there will be an advantage and this is known as the 'therapeutic misconception' (Applebaum et al, 1987). However, the misconception is only partly erroneous in this case, because when patients present the clinician with a disease related problem, she may offer a trial of something experimental to relieve symptoms. The patient (and often the clinician, in a double blind trial) would not know if they received a drug or a placebo, but may assume they are receiving treatment. This is a consequential feature of a combined clinic for research and disease management, and neither activity can be 'pure' in operation.

Patient and carer awareness of research hopes and limitations

Clinic attendance and test participation became a signifier that the patient was still 'in the running' to be able to benefit from a research drug trial or potentially, foetal cell transplantation to the brain. This was not discounted by the researchers who acknowledged the enthusiasm but felt they had to play it down.

Prof James: ...The expectation issue is a really important one so.... We had to face that early on... a lot of contact with the media....with patients... actually lowering that expectation,..obviously we wouldn't be doing it if we didn't think it had some prospect but you know, very, very, early days.

(Interview 6a: Research Clinician)

Prof James:.....whether or not patients are individually involved in the research, I think they find...and even though it hasn't yet produced..a cure or treatment, I think the fact that it's going on is a bit of a light in the tunnel to them....

(Interview 6a: Research clinician)

Professor James explained that lowering public expectation was a key part of the early publicity surrounding the project. This shows that scientists and researchers are aware of the potential media 'hype' which accompanies new research and take action to keep the news realistic. However, the news-making is part of science and an important motivator of public support for new and contentious procedures. Several years ago, foetal cell transplantation from aborted fetuses would have made the headlines for rather different reasons, and success in science is dependant on taking the public with you.

There was acknowledgement by researchers and families that nothing had changed in terms of disease treatment for many years, but also the suggestion that there would be a successful outcome. This signalled more use of ambiguity in the prediction

of what science will achieve; the public are just as likely to be alienated by a lack of belief in success, as contentious procedures. Patients and carers remain involved with the clinical professionals 'just in case' something became available.

Ken cared for his wife Sue, his view was typical of clinic participants and he even used the same metaphor as Professor James:

Ken: ...if there was a light at the end of the tunnel and we were coming here and there was a progression then perhaps it would be different you know. But there is no cure and we're just coming down just for them more or less to monitor us and...no, there's no other reason..... there's no sign of Sue getting better you see, at the moment anyway. Nobody knows you know, that there might be a cure for it or possibly a way of holding it back.

(Interview 1, Ken, husband, carer)

Ken's view that there was only monitoring of the disease taking place was realistic, but he also hopes for more, at some time in the future. He acknowledges the limits of current knowledge with the last sentence "Nobody knows...there might be a cure for it", and yet still frames this as a possible outcome.

Hope was also mitigated by reality; the day to day burden of living with a degenerative disease for the patient and the carer often sharply focused against the increasing research agenda. It was as though families felt so responsible to be involved and there was a social expectation that they should hold onto hope, it was almost treachery to accept that research may not produce anything, or to state anything other than a positive outlook. This 'just in case' outlook has been written of as 'promissory capital', and features in the development of umbilical cord blood banking by parents as a measure to avoid future 'moral recrimination' (Brown and Kraft, 2006; Rubin 2008)

People spoke often of 'reality', 'the facts', but with a growing realization that the research would be unlikely to produce anything for their own situation. This sometimes remained in the background and was unsaid.

Alan: I still think it's one of the most awful diseases I've ever seen. I mean even if there's a cure or a help with tablets – you've just said it could be ten years down the line.

Enid: It's such a ... in my estimation it's such a long disease, isn't it? I mean Alice's twelve, thirteen years now – I mean no-one knows ...

(Interview 3 , Mother and brother, Feb 2008)

What was particularly striking in this exchange was the length of time expressed by the two family carers. 'One of the most awful diseases I've ever seen' was a heartfelt statement and not spoken in exaggeration or self pity, just factually. The patient in this family has already been affected for thirteen years. This is not only the patient's life time illness, it is perhaps twenty five years in each family member's life time, whether or not they have the disease themselves. Enid, the mother, had also cared for her husband who had the disease for many years, so practically all her adult life was taken up as a carer. The cumulative total of years lived with the disease can easily be over a hundred in each family.

In looking at the length of time involved between diagnosis, continuing decline and eventual death, it is more understandable that the ideal clinic performs an important function in maintaining contact, continuity and hope in families and patients, and that the meaning of going to the clinic, extends way beyond simple research participation.

Transplant hopes

The suggestion that research with embryonic stem cells could lead to potential regeneration of the human body is known as 'therapeutic promise', a concept which first appeared in 1995 and it was elaborated and restated in 1998 (see Rubin 2008, pp18-20 for a full discussion). Since then, the public phase of stem cell discourse has continued with high level support and patronage as the following statement from the scientific journal *Science* shows:

With dramatic results like these coupled with growing public acceptance, the stem cell field is poised for progress. If it lives up to its early promise, it may one day restore vigour to aged and diseased muscles, hearts and brains – perhaps even allowing humans to combine the wisdom of old age with the potential of youth. (Vogel, 1999, p.2239, cited in Rubin, 2008, p 20)

This high profile vision was part of the ‘scientific breakthrough’ award made by Science to the authors of publication of the work concerning the isolation of human embryonic stem cells (Rubin, 2008, p20)

Patients and carers in this clinic study explained that they signed up for research in the hope that brain cell transplantation would become available for themselves or their relatives. Enid, the mother of a patient explained clearly that this is what she hoped for.

Jacki: Can you remember the first time you were asked about research in the clinic? Or before you went to the clinic?

Enid: Erm, well in the clinic yes. We signed up for any research because you grab at any straw don’t you? you know, and at that time, going back quite a few years now, there was the stem cell wasn’t it to the brain, they’d done two operations, how many years ago- about six years ago....Yeah. So, we thought it would have been... I mean I’m silly, no, because Alan was lucky with his heart transplant, I thought oh, Alice’s going to be lucky with hers you know but...keep our fingers crossed you never know do you? (Interview 3, Enid, mother)

Enid’s other adult child Alan, had received a successful heart transplant some years ago. Enid was critical of her initial optimism for assuming brain cell transplantation would also be easily available to stop the Huntington’s disease, then diagnosed in Alice, the other adult child. The optimism was based in her previous heart transplant experience so perhaps it was understandable in her case. However, it does

point to a normalising of transplantation to treat the failing body, and a move to the most invasive experimental measures when the disease is incurable and fatal.

Patients and families believed in the 'natural suitability' of stem cells rather than manufactured drugs, as a means of halting decline or curing Huntington's disease. Carol the mother of an adult man with the disease, also recognised that this could be a contentious matter for some people who did not agree with stem cell technology for reasons of principle or religion.

Carol: ...I have no problems with stem cell research. But, saying that, I'm not about to say to someone who has got problems with it, that they're wrong, because if you said to me stem cell research was the answer to HD for Stephen, I'd go for it....So, I personally thought there was great optimism with stem cell research. I thought it might have actually a lot more answers....

Jacki: Yes,...I was going to ask you- we're talking about it as if it was a past tense and I just wondered if you...

Carol: I thought it...it took a lot of criticism didn't it?

Jacki:had changed your mind or?.

Carol:and also it took a lot of ethical- what they could do and could not do. ...I mean for a start, America... they stopped stem cell research didn't they in the States with George Bush but now.... I'm not being funny, I do think something like the States is an asset in that- the size and resources, it's a bit like saying you've got a Europe HD network rather than just the UK..... the more you share the more chance I think you've got of picking something useful up.I sometimes think of drug research as having a limited line... I don't think I can foresee someone inventing a pill that you will take that will cure you of HD. Stem cell research seems to me much more viable....

Jacki: What is it particularly about stem cells that make you optimistic?

Carol: I don't know, I suppose it's the basis of life, isn't it? I think it comes back to origin if you know what I mean. But we could be totally wrong- there might not be an answer there. But it always seems to me as if it was a more logical way to go than drugs....I always thought stem cell research was probably where they might get a lot of answers.

(Interview 6, Carol, mother, carer)

Carol was astute in her appraisal of the possibility of a stem cell treatment, and when expressing her thoughts about the changing political and research situation in the United States. She felt there was more potential success in the global research project, with more people working towards a solution and sharing their information. This position assumes the total beneficence of science and that scientists are not in competition but are working with the same altruistic objectives, i.e. focussing on reducing suffering. Previous studies of science have shown this is not exactly how scientific collaboration works in every case (Atkinson et al, 1997, Collins, 1992), but it is a common public perception which is rather useful in gaining public support for scientific endeavours.

Carol was distinctive in being the only non-scientist in the whole study who voiced the opinion that stem cells may not provide a solution; only one researcher also expressed a vague doubt about this.

Conclusion

This chapter has discussed the hopes and benefits of clinic participation from the perspective of the patients, carers and families affected by Huntington's disease. It was evident that people attended the clinic to have disease management, moral support and assistance. The future and research objectives of the clinic were of secondary importance for participants. However these played a major and important role in keeping people engaged with the clinic. Patients and carers found research involvement a meaningful part of their clinic attendance but there may have been bias in these responses, due to willing clinic attenders being more supportive of research in any case.

The researchers negotiated the difficult position of trying to maintain their research objectives and deal with the various problems brought to the clinic by patients. Some of the problems were research related, such as requests for neurological surgery for example, but these cannot be fulfilled as the patients would often wish. Clinical disease related issues require much more time, expertise and resources than are available to the clinic researchers, and so they are often passed outside the clinic, to other agencies or left for the families to try to deal with themselves through primary care services.

The potential of future treatment such as neuro-surgery was the most important motivator in keeping people engaged with the clinic. The research team were aware of the challenges this created, and sought to achieve a balance of cautious optimism and realistic approaches in order to maintain research interest and to be fair to patients and families.

With respect to the research questions of how do the two areas of research and care work effectively in the combined clinic, and what does the research clinic mean for the actors, the chapter highlights the important role of hope and reveals how that is created and maintained. Researchers encourage the production of hope by means of ambiguity and expertise, families and patients see hope as an imperative to their existence.

Chapter Seven

Local clinics, global research

Introduction

This thesis considered research questions concerning the change in the research basis of Huntington's disease from genetics to neurology. The first question was - what are the effects of this shift in disease categorisation, (from a previous framework of genetics to the current position as prime neuroscientific research), and how does the move affect the relationships between patients, carers, researchers/clinicians and the disease? Together with the overarching idea of a disease trajectory which has placed Huntington's firmly into neurology for the time being, the neurological research surrounding the disease has taken on global proportions in a manner previously unseen. It is against this background of increased participation levels and high profile neuroscience, that local research clinics take place all over the world, gathering vast amounts of epidemiological data from the diagnosed Huntington's patients.

The second research question was – what exactly does research participation mean for the patients, families and researchers? This study of the Huntington's research clinic has found that there are differences in the meaning of the clinic activity for patients and researchers as a result of the 'movement' of the disease from genetics to neurology, and the emphasis on clinical neurological research. The differences in meaning are spread right across the different clinic actors, including patients, carers, clinic researchers, other NHS management staff, support workers funded by charitable organisations, and even the extended families of patients. To answer the broad question of the effects of the disease move to neurology, I have analysed ethnographic data of the research clinic routines, the experiences of patients, carers and researchers, the rhetorical and performative strategies of clinicians, patients and carers, and the methods of utilising hope which help to maintain the research activity.

I have put forward three main themes or findings in the analysis chapters; the 'blurring' of the traditional boundaries between research and clinical management

activity; the performance of roles and strategies in the clinic by both researchers and patients and carers; and the utilisation of hope as a means of recruitment, justification for participation, and retention of participants.

Blurring between clinical management and research

First, I will concentrate the discussion on a ‘blurring’ of traditional boundaries between research and clinical management. Patients and researchers constantly negotiate their clinic activities of research and disease management in a manner which effectively creates ‘blurring’ between these two areas, which are usually carried out separately, and have quite separate and distinct protocols attached to them. In late modernity, the two activities are traditionally considered to be distinct, at least in western medicine (Gieryn, 1983), and one of the important reasons according to Gieryn, is that a clear distinction protects researchers from outside or political interference and is helpful in supporting professional research goals. The boundaries between research and clinical management of Huntington’s disease are fast being breached to the effect that patients cannot distinguish between them, as highlighted in the data analysis of this project. I have shown that the two activities take place at the same time in the same clinic, and are carried out by a team of researchers, and some medical staff who are also researchers. The change observed in this study of neurological HD research clinics, is that the separation of care and research is no longer distinct for either the researcher or the patient - this is what is meant by the blurring of the boundaries. The mechanisms by which each activity was formerly discrete, are pushed together in a more obvious manner. For example, recruitment to different studies takes place within the clinic and may involve several research team members in a discussion before, during and after the clinic day. Care of patients is intertwined with research activity, the latter may be used to potentially achieve the former.

There is a trend in clinical medicine towards the dual activities of research and clinical management happening at the same time in the same clinic (Hallowell et al 2010), when previously they have been considered very separate activities, with separate ways of operating, and clear demarcation between research and ‘care’.

Hallowell (2009) points out that ‘blurring’ as I describe it, “appears to be particularly prevalent within clinical genetics”, and several other authors also refer to this (Easter et al, 2006; Henderson et al, 2007; Miller et al 2008; Hallowell et al, 2010). However, the situation has arisen partly due to 1) the fact that many fatal diseases with a genetic basis are relatively rare in the overall population, and 2) the usual basis for causal determination relies on large numbers of samples to show the isolated ‘difference’ in affected persons vs. non-affected, then 3) the isolated ‘similarity’ in affected populations. Obtaining large groups of samples of rare disorders takes up many resources including time and money, so in order to make the recruitment efficient, one would require specialist groupings of affected people such as those who attend a specialist clinic for example. In the location where my research took place, the NHS formerly had a specialist clinic for people affected by Huntington’s disease which was based in the Genetics department of a large teaching hospital. A major effect and sub theme is, that in this study Huntington’s disease management has moved away from the NHS framework for cost reasons, and also because there is no standard treatment available yet.

The Huntington’s disease management is therefore effectively funded by research money from an outside organisation. This is a fundamental difference in disease management from the previous arrangements within the NHS framework via the Genetics department. Cost cutting meant the specialist clinic existence was in jeopardy; because there is no approved treatment for affected people, the main purpose of the specialist clinic was to monitor Huntington’s disease progression (as in many long term illnesses), and also provide patient and family support and palliative care. The neurological research group had already begun to monitor the disease using well known clinical measures, however this research group had no care function, or particular expertise in that direction. When the decision was made to cut back the NHS Huntington’s management clinic, this meant that patients had the choice of attending the research clinic for disease monitoring instead. In effect, the research funding was supporting the clinical management function but without the NHS staff or expertise previously provided.

The majority of HD clinic patients participate in several forms of epidemiological research known as European Huntington's disease Registry, or Euro-HD. It is funded by a non profit organisation formerly known as the High Q Foundation, and currently known as CHDI, Inc., and it is based in the USA. The role of this organisation is to facilitate more and better co-ordinated research into Huntington's disease. The CHDI website sets out the objectives of the organisation.

“CHDI is a private, not-for-profit research organization. We work with an international network of scientists to discover drugs that slow the progression or delay the onset of Huntington's disease (HD).

We seek to accelerate scientific progress by serving as a collaborative enabler. We encourage and support cooperation and collaboration among HD researchers. Our strategy is to encourage researchers to develop practical ideas, useful research materials, and powerful technologies, often by providing financial support.

Our activities extend from exploratory biology to the identification and validation of therapeutic targets, and from drug discovery and development to clinical studies and trials.”(www.highqfoundation.org)
Accessed 10/9/2010.

The financing of research is often outside UK state funding, from research councils, charitable trusts, and other organizations in partnership with pharmaceutical companies, health authorities, and private companies and the majority is US based (Lock & Nguyen, 2010). There is a global market in research subjects, and anthropological work has established that the world's poor and 'naïve' patients are much in demand to furnish clinical trials (Petryna, 2009). The 'naïve' patients are those who do not practice self medication or who have not received previous treatment for a condition, so the clinical drug trials are far less likely to be subject to drug interaction, and the disease is thought to exist in an untainted, more natural state, for example, hypertension (Lock & Nguyen, 2010). One such area of Venezuela assisted

the earlier genetic research which eventually discovered the site of the gene for Huntington's disease on chromosome 4; the population of Barranquitas, Venezuela, has a much higher than usual incidence of HD, possibly as high as one in ten. The US based Hereditary Disease Foundation¹ has sponsored some help for the affected families there, but would like to research further with the co-operation of the Venezuelan government (HDF, 2008; BBC, 2010). The Venezuelan government however, is not in agreement with the idea of blood samples and genetic material leaving the country. It is beyond the scope of this study to discuss this area further, but the potential for a social science study of the Venezuelan community and the effects of further research there, may reveal important structures of global HD research in a similar way to those observed in Eastern Europe (Petryna, 2009). Closer to home, there is a research survey included in the Euro-HD project to assess self-medication in HD patients, with the aim of reducing it. This too, may offer a worthwhile area for future sociological research.

Standardisation, measurement and reproducibility in research

The basic research programme in the UK is part of a global scale epidemiological study of Huntington's disease, incorporating disease progression measures which are coded and stored in several different forms. Importantly, it is a non interventionist study, but within its framework is the option to re-contact patients to invite them to participate in intervention studies (Euro HD, 2010). The basic study research instruments are based closely on existing internationally approved scales of disease management, and are subject to further standardisation in this study, in order to provide suitable data for large data sets to be created and retained indefinitely. The standardisation of the epidemiological research includes, 1) standards for the implementation, interpretation and result recording of neurological, cognitive and physical motor function tests which are applied globally, 2) the collection of global and national bio-banks of blood, DNA, human tissue and urine, 3) the collection of visually recorded data of standardised neurological testing, and in addition to the basic epidemiology, 4) data from multi-

¹ This organisation also has strong links with CHDI, the funding body of Euro-HD.

national clinical trials of drugs where recruitment is from the epidemiological group. The move to neurological research has brought about some further standardisation in the clinic testing procedures and recording of data, and if we consider that the medical record is an organisational memory (Berg & Bowker, 1996/7), the current function of it is to provide a memory of detailed neurological data. However, there are advantages for the neurologist, because it is much easier to see at a glance the condition of the patient at the last visit and this saves time in the next consultation. The neurologist 'sees' a standardised picture of the patient's condition according to the parameters of the medical record. The standardisation of recording and what is measured, produces a disembodied form of Huntington's disease known only to the researchers and the scientists, and it is a partial view. Creating the disease as an entity is an ongoing project. The demands of epidemiology and globalisation require certain types of tests producing certain types of disease; this is the standardisation of the disease itself via measurement and reproducibility. The collection of many different national datasets in the Euro-HD project may produce ideas about subtle differences in the manifestation of disease which are so far unexplained by bioscience alone. This is increasingly the way of research, it has become transnational in many respects, as described by Lock and Nguyen (2010, p.360), although the point they make is that biomedicine has tended to assume the homogeneity of the human body even in transnational terms.

The research clinic observed for this project is part of a European research framework in the first instance, and a global research framework in the second, which will eventually require large groups of patients to be available for the future testing of new interventions. There are at least 19 countries collecting the epidemiological data, including, Russia, UK, Germany, Italy and Spain. There are close links to the USA based Huntington Study Group, and projects in Australia and South America. Further recruitment of the Registry patients is possible from the basic epidemiological research as already mentioned; patients may be invited for other research projects to test drugs, interventions or surgical procedures and patient suitability is often based on their particular disease manifestation or disease stage. This appeared to be presenting the patients with an opportunity to benefit from a trial, in effect, experimental medicine (Lewis, Hughes & Atkinson, 2009). In contrast, the NHS would only consider

treatment of Huntington's patients if there was evidence of effective intervention; this is the policy position of evidence based medical treatment in the current UK health service arrangements. New potential therapies for Huntington's disease would be expensive initially, because they would be classed as 'orphan' drugs and may not be approved for use in every geographical area of the UK. Therefore, this targeted recruitment of Huntington's patients represents a speeding up of the process in terms of potential availability of treatment, and disease management.

The role of therapeutic misconception in blurring care and research

Patients and carers are aware of the possibility of being asked to join a trial, and are keen to take part because they assume they will receive some experimental treatment which may help them. A great deal has been written on the 'therapeutic misconception' or 'TM' (Applebaum, Roth et al, 1987; Applebaum, Roth & Lidz, 1982 ; Applebaum, 2002; King, 2000; Miller & Brody, 2003), which means that participants in research expect to receive some benefit from taking part, in the way of treatment or better care. There are clearly therapeutic expectations of this clinic within the patients' and families' expectations of the HD clinic; the majority of people said openly that they want the chance of a stem cell transplant or a drug trial. Although the existence of 'therapeutic misconception' is not usually desirable in research or clinical trials according to the governance of ethical research, King (2000), nevertheless considers that there are different types of therapeutic benefit and some of these beneficial effects may be helpful to patients. The benefits in the HD clinic are intrinsic and are concerned with collateral or inclusion benefit, plus aspirational benefit for the future of society (King, 2000). These effects are clear in this study because patients and families were very proud to have made a contribution to the research, despite their hopes of treatment not being realised. The blurring of research and clinical care is assisted by the 'therapeutic misconception' which according to Glannon (2006a) is 'an unavoidable part of the imperfect process of recruitment and consent in medical research'. Glannon is making an important point, because the reality of medical research is often somewhat different to the ideal situations enshrined in biomedical ethics frameworks, and this

reality is rarely acknowledged in research protocols, or post project reports. It is as if the biomedical ethics surrounding research and care are part of an idealistic view of a past golden age of pure research, untainted by care issues, and pure care, unsullied by research objectives.

Effects of blurring on clinic practice

Patients, carers and researchers acknowledge there is a 'service gap'. The guidelines on treatment for Long Term Neurological Conditions (LTNC) recommend close links between neurology, rehabilitation and palliative care services, and included in this is a requirement for a specialist neurological service (Turner-Stokes et al, 2008). The HD research clinic was not designed for this multi-function role, although the researchers and clinicians make every attempt to fill this service gap to the best of their ability and with available resources. However, this has an impact on both disease management and the operation of research, where neither can function in an optimal manner. For example, a research clinic would normally concentrate on gathering data and observing the 'gold standards of research'. Recruitment would be planned, so that trials and experimentation would pursue a definitive path. For disease management, the patients and carers would have a regular point of contact and all the specialist help they require from allied health professionals, in rehabilitation services and palliative care. The guidelines as mentioned above, make an important point that rehabilitation care is required in long term neurological conditions up to the end of life, not only for those conditions where an improvement or recovery is expected (Turner-Stokes et al, 2008).

This is a particular problem for the clinic researchers in this location, who have to expend research time and effort 'passing on' these issues, rather than concentrating exclusively on the research aims. Therefore, a secondary effect of Huntington's disease move to neurology research shows that a management decision within the NHS, has effectively turned patient care over to a research group. The blurring of the traditional boundaries between research activity and clinical care have been brought about in this case, because the neurological research activity is viewed on some level (not by

everyone), as almost identical to clinical care and there is a need to conserve health service resources.

However, research funding cannot provide the full level of support required, certainly nothing near to that recommended by the guidelines for Long Term Neurological Conditions (Turner-Stokes et al, 2008) and this is what has transpired in the move from genetics to neurology. Carers and Huntington's disease support workers are very aware that the research clinic is better than no clinic, but they also recognise that there is a fundamental flaw in this approach, which does not serve either the patients' care or the researchers' tasks to the best effect. Other research (Miller et al, 2008) has found that in some cases, research clinics became quasi-clinical care and were ineffective.

What is lost when blurring occurs?

Patients and carers whilst appreciative of the time and effort taken by the researchers in the HD clinic study, notice that the 'clinic work' is so focussed on the individual patient, that there is no 'space' for any real support for the carer or family members, and no direct means of obtaining required assistance easily. Indeed, not all patients with HD attend the research clinic for various reasons. One view of this came from a support worker who explained that some families and patients feel aggrieved that their data is 'mined' in clinics, yet there is no specialist health service care available from all the 'disease experts'. Those non clinic attenders are reliant on whatever services are available from their primary care providers and probably social services. In reality, they may not be any worse off than people who attend the clinic, who have their problems 'turfed' back to primary care. Families and patients arrive at the clinic, where they have access to what they consider to be the best expert knowledge of this disease, they take part in research and monitoring for disease progression, and then they return to their day to day struggles with patchy care provision and primary care non-expertise. This brings me to consideration of what aspects of Huntington's disease have legitimacy to be aired in the research clinic, and who can raise the issues around the disease which are not directly concerned with the research. There is no equity of

availability or provision outside the clinic, with each geographical area or governing health board setting their own levels of care. The small amount of contact and resources available via the research are very precious and no one wants to sound ungrateful. It is an important point that research can be organised on almost an all encompassing scale and be standardised, yet patient care does not receive the same attention in practice. The move to research is also linked perversely with a reversal of the argument for care or cure. Bury (1997) claimed that the shift from cure to care in many chronic illnesses is representative of the changing demographics in society, including ageing, longevity, chronic illness and disability. In the case of the HD clinic, this appears not to be the case; the resources are firmly directed at the research effort in the search for treatment and cure. These are private resources, not supplied by the state, and potentially subject to shifts in private priorities. It is possible that the change to more research and away from clinical management and support is fundamentally more important for people than the move to neurology instead of genetics.

Normalisation of research ‘work’ into clinical care ‘work’

Quite apart from the thorny question of the lack of expert support from the main health service provider in the UK (the NHS) for families, carers and patients, is the matter of the normalisation of research into clinical care. May (2006) claims that in the latter half of the twentieth century, medicine began to include the patients’ individual meaning and subjectivity into practice, and at the same time, there was a move towards the global processes and organisation of biomedicalisation (Clarke et al, 2003). May explains that there is nothing special about the clinical encounter as it has been variously portrayed in the work of Parsons (1951), and more recently in Science and Technology studies by Langstrup (2008) and at a further extreme by Timmermans and Haas (2008). He goes further to say that the clinical encounter, in essence, is now the same as many other professional social or business encounters, because it is constrained by the complexity of business and management objectives which are often asymmetrical in terms of power and knowledge. In this respect, May is talking about the management of medicine, which is now subject to corporate regulations, and the

asymmetry he refers to is between the privatised relations of clinician and patient, rather than the power of clinicians over patients, or medical dominance. The Huntington's research clinic shows some signs of this - the arrangements for this clinic to operate are made, not primarily in the interests of the patients, but in the interests of research and the reducing of health service costs. The patients recognise there could be a 'research bargain' being struck and seek their experimental therapy. Aside from any political point this may raise, it is an important effect of management of health technology on patient's bodies. May considers this to be an assault on the 'special moral character' of the ideal type of clinical encounter (2010), because it is a set of corporate impulses (2010, p5).

May (2010) goes even further with 'Normalisation Process Theory' in respect of clinical encounters, which he constitutes as 'work' to be progressed through. The research clinic data highlights that researchers and carers are often highly aware that there is a research schedule to 'get through', and this can easily become delayed if other types of 'work' (care matters) are brought into the clinical encounter by carers or patients. Carers acknowledged some discomfort in failing to find a 'space' to approach these matters, but only when they were away from the clinic and paused to reflect. Graham (2006) notes that many sociological analyses are unsympathetic to the pressures of medical 'work', and structure their criticisms around institutional medicine that is 'done to people'. This position does not take account of the patient 'doing' illness, or the difficulties faced by clinicians in the medical encounter (Graham, 2006). However, in the study of the HD clinic I have shown that in almost every encounter, the clinician must engage with difficult negotiations, hard 'work', the result of the blurring of the boundary between research and care of the patient. The study also shows clearly the patients and carers performing the work of their 'ideal candidate' roles, in effect, examples of 'doing' the sick role, in the hope of obtaining a treatment benefit. It is the performance of roles in the clinic to which I will turn next.

Performances become linked to the research objectives

Performances enacted in the clinic by patients, carers and researchers are directly linked with the movement of Huntington's disease into neurological research. Researchers perform many roles, they are required to be both dispassionate researchers and sympathetic clinicians, undertake the roles of friendly and understanding research experts, disease knowledge experts, and at least facilitators to sources of care, rather than direct care providers. Patients and family carers in the HD clinic study usually portray themselves as 'ideal patients' and 'ideal carers' in the hope that some form of treatment can be secured. The patients and carers explain their routines and day to day living in ways which show their expertise with the effects of Huntington's disease, and with the enabling of the disabled patients lives. Carers like to portray the quality of life they are sustaining for the patient in order to be seen as expert carers, and because they want to be engaged with and included in the research.

I begin the discussion of the theme of 'performance' with reference to the siting and setting up of the clinic because it is not a permanent physical entity. The website for the clinic refers to the 'virtual' centre for the research of Huntington's disease, and the twice monthly or weekly clinics are created each time by the meeting of several researchers from different parts of the university, in the research centre. However, to an outsider arriving for an appointment, the creation of the clinic is invisible; when they arrive it already exists, there is a waiting room, consulting rooms, researchers, and a programme of work or research tasks. Patients and carers asked me during the course of my observations and interviews if all patients with genetic diseases use the clinic, and they presume this is a high status NHS funded centre. The reality of the clinic being a 'research set' each fortnight or so, was not something which occurred to the participants, and it did not occur to me initially. It was during my second or third clinic visit that I appreciated the gathering together of separate researchers and workers for the creation of an HD research clinic day.

The clinic has a 'rehearsal' and 'directors' instructions' issued at a pre-clinic meeting, where the researchers discuss the expected patients and how the research work will be distributed and in what order, there is effectively a rotation of multiple research

tasks. The operation of the clinic day is lean compared to other parts of the NHS which patients may have experienced, and this contributes to the feeling of receiving a personal service. There is direct contact between the patients and the researchers who carry out the testing which may take several hours to complete. Some patients have attended the clinic for several years with their carers, which is time for them to become familiar with the surroundings, clinic routine and the researchers, in ways they do not in other appointments or research. Most families enjoy being recognised and remembered in a very collegiate atmosphere, patients and researchers know each others first names. These factors contribute to the ideas held by families that this clinic is specialised, and personalised with the patients at the centre of the whole operation.

Performance in preserving the clinic ceremony

The researchers remarked in their interviews, that if they were in the patients' position and doing the tests themselves, they would want to know 'how they had done', but patients only ask about physical testing, and never about their cognitive abilities. This reticence to enquire, appeared to demonstrate Strong's bureaucratic format of a 'ceremonial order' (1979, p.18 & 192) in that it preserved the surface ceremony, which is an unspoken compromise between the patient and the researcher. Direct questions concerning the patient only took place during the neurological consultation, where in contrast to the other clinic research exercises, the surface ceremony was breached by patients and carers where people objected to suggestions, or tried to gain treatment. Patients and carers who did challenge the bureaucratic format were subject to reminders of medical authority, and 'moral work' which occasionally took a covert form (Featherstone et al, 2006). The research work performed in the clinic was complex and nuanced; it was not simply a data gathering exercise from disinterested participants. This was portrayed in the pre and post clinic meetings and via informal researcher interactions during the clinic day. For example, a family who came to clinic without the patient's drugs, or who did not accept advice on certain matters, were described as 'difficult'. Their non-cooperation took place outside the clinic and was contained in aspects of 'living with the disease'. Those carers who did not take up suggestions, or

who behaved in a light-hearted way in the consultation may have been described as ‘being in denial’ about the disease, and for patients, this was pathologised into ‘losing cognition’. These performances held the moral work in the unofficial ‘clinic talk’, and were not recorded in patient files, so once the consultation was over, there was no trace of the moral work or the medical authority. In contrast to this, most of the moral work done in previous decades was actually written down in the files.

The ‘surface ceremony’ was not breached by the consultants or researchers in any obvious dramatic way, but there were subtle indications that this clinic was different in important ways. For example, the consultant would regularly appear in the waiting room to greet patients and accompany them to the consulting room, and this represented a lowering of barriers and removal of medical hierarchy to the patients. Time given to patients and carers was never rushed; there was a relaxed atmosphere which contributed to patients and carers feeling valued in the clinic. ‘Surface ceremony’ boundaries between research work and care were only marked by researchers who did not have the authority to medically treat patients in the clinic especially, if there was a high expectation of care from patient or carer. However, researchers never made this explicit to the patients in the clinic, by refusing a request for example. Researchers would either retreat into neutrality or may refer the patient and the enquiry to the neurologist. This represents an adherence to the surface ceremony (Strong 1979); it served to preserve the medical authority and patients’ confidence in the clinic.

Biomedicalisation of care and research functions

Clarke et al (2003) discuss biomedicalisation in a way that can be mapped onto the HD research clinic, with one or two areas remaining incomplete. Using Clarke et al’s theoretical position, we see the HD clinic patients transformed into data providers, bar-coded entries on standardised paperwork, bar-coded identifiers on blood, tissue, and urine samples, all producing new biomedical knowledge. The distribution of the new knowledge is to a much wider audience than in previous decades, different national and

international groups of scientists may be permitted access to the data for use in their research work, producing further new biomedical knowledge. However, according to the criteria given by Clarke et al, the shift to biomedicalisation also includes transformations of bodies and identities in terms of bodily customisation (2003). This is only happening in terms of the data transformation in the HD research clinic at the moment, and some drug trials for groups of HD patients. There are no individually tailored drugs or therapies generally available to patients, however, the targeted recruitment for clinical trials appears to be a step in this more experimental direction.

I have shown that patients and carers create performances, present themselves in such a way as to be considered suitable for trials and interventions, in effect as 'ideal candidates', and that they maintain links with this clinic in particular because they consider it to be the 'best place' for a chance of treatment. The focus for researchers is firmly on the collection of research data; the day to day problems of living with the disease which are brought to the clinic by patients are not ignored, but are delegated elsewhere, outside the boundaries of the clinic, and often outside the hospital, back to the primary care level. Further evidence of patients and carers performing roles in the clinic was highlighted in interview data, where people openly disclosed that they maintained links with the clinic because of the hope of some treatment, and the high regard they have for the performance and reputation of the clinic and researchers. This is all part of hope for treatment now and in the future, which I will discuss in the next section.

The effects of raised research hopes and optimism

Increased hope in a treatment or cure is a feature of large, well-publicised research programmes (Geesink et al, 2008; Martin et al, 2008). It is because neural regenerative research has reached a stage of potential intervention with foetal cells or stem cells, (Dunnett et al, 1997) that the raised expectations of future treatments for neurological diseases exist practically everywhere. There is great optimism surrounding the invasive neurological procedures to the point that the question is 'when' it will become available as a standard treatment, and not 'if'. The existence of raised hope, expectation, and

research optimism, serves to further enhance the research clinic reputation, and helps to maintain the clinic research recruitment. This recruitment is of a long term nature, there is no end date and no limit on the number of participants, although there is a general aim to obtain data from ten thousand patients in the European sector of the study, by the end of 2010. The current participants will not benefit, but their data (written, coded and biomedical samples) will potentially continue to be used long after they are deceased. Patients and carers acknowledge this and express a desire to assist future generations (who will often be related of course). This hope for a future where HD can be treated if not cured, and the continued co-operation of the research participants is due in part to decades of work previously carried out by the Genetics department in providing support, counselling and a service to the families in the local population who have Huntington's disease in their lives. This has created a research(ed) community who are now hoping to benefit from the recent advances in stem cell technology.

Research hopes ongoing and adjusted

Published scientific papers on the outcomes of the previous foetal cell neural transplants are variable in terms of outcomes and expectations for further work (Bachoud-Levy et al, 2000; Bachoud-Levy et al, 2006; Cichetti et al, 2009). The most promising results gave a small improvement in some areas of the brain for one or two patients for the first two years, but after this, the patients continued to decline. Other patients did not show any improvement at all but the neural graft sites remained intact, and some patients had degeneration in the areas of the grafts. Yet this information does not seem to have reached the clinic participants, and it is not discussed in these terms in the clinic when issues of transplantation are raised by patients. The researcher 'talk' is of hoping to re-start the project, in the next few years, with emphasis on the experimental nature of the invasive procedure. This is an example of an 'imagined future', undertaken by a 'conglomerate of diverse groups' according to Martin et al (2008), 'nevertheless, they are often spatially and temporally situated with often limited opportunity for actual engagement', (2008, p.32). This may account for the enthusiasm

expressed by patients and carers, for attendance at the clinic where they engage with 'some of the best experts', as one carer put it.

The purpose of embryonic or foetal stem cell transplantation has also undergone at least two shifts from the original hope of regenerating neural tissue from the embryonic cells and improving brain function (Bachoud-Levi, 2000). It was thought that there may be a protective effect of neural grafts which had potential to delay progression of Huntington's disease because the grafts survived, even though the patients eventually died a few years later (Keene et al, 2007). This was a less dramatic effect than the regeneration of the brain but was still a potential intervention. More recent results showed that neural grafts can be subject to HD like degeneration, with the authors stating that this raises uncertainty about this potential therapeutic approach to HD (Cichetti et al, 2009). At the time the clinic observations were made for this study, this latter information was not available, and the type of 'hope' expressed was undifferentiated, and was utilised as a legitimisation strategy (Kitzinger & Williams, 2005). However, an earlier study pointed to the benefits of simple environmental enrichment in mice models, and suggested that strategies of occupational therapy and environmental enrichment could help to delay the onset of Huntington's disease in humans (Hockly et al, 2002).

Hope, commitment, and ambiguity

The hopes of the patients and carers play an important part in maintaining their support for the research clinic, and an aspect of the clinicians' and researchers' clinic roles was to foster this support and manage the expectations of the clinic participants who often wanted to know more about the research. Explanations of the research progress were given in terms of the future, of potential, of large numbers of research projects with large numbers of participants. The rationale given was the aim of finding suitable interventions to delay the disease progression; there was no talk of a cure. Patients' participation in the research was supported by researchers and clinicians with encouragement, optimism and gratitude for attendance.

One of the difficulties which arose in the clinic for researchers was to find the right balance between optimism and reality for patients. There are an increasing number of cases of stem cell tourism (Campbell, 2010), where desperate patients travel the world and use their life savings to try to obtain a cure for their illness at unlicensed 'stem cell clinics'. None of these so called miracle clinics have any peer reviewed results for examination, there is no evidence of the treatments being helpful, and they may even be potentially harmful (Campbell, 2010). Clinicians and researchers found that in order to balance hopes and expectations with reality in the clinic, and to keep patients engaged with the research process as a whole, it was necessary to use ambiguous language. The language pushed the promise of treatment further into the future, but also kept the patients engaged with the current research activity, which was portrayed as a massive enterprise and vital to the future promise of a cure. Researchers also included their own carefully optimistic expectations in these portrayals, and gave reassurance that efforts now would produce results in the future. There was use of inclusive language, for example 'we hope that in the future..', and 'we are hoping the trial will show...', 'the research shows us that..', which extended the impression of partnership in the research enterprise to clinic participants. The alternative, the end of hope in a UK research clinic, may be enough to send patients to seek treatment abroad. Researchers have the task of maintaining support for well regulated UK research; a few reports of bad experiences abroad may have a negative effect on support for UK stem cell research, prompting public questioning and mistrust similar to that of the MMR vaccination controversy.

Patients and carers found the research clinic gave them time, and participation in something of importance which, greatly increased their self esteem. They were proud to be associated with the clinic and to make a contribution to research, however small that may be. This was also a justification for their hope in the future where treatment may become possible. They expressed their hopes mostly in terms of the future but also potentially for their own benefit, or for the benefit of their descendants. Petryna (2009) points out the paradox of this position in terms of the basis of ethical clinical trials. Those who take the position that clinical trials are a 'social good' because they enable patients to have treatment they would not otherwise receive, are effectively agreeing

that the voluntarism and altruism, deemed to be a necessary element of ethical clinical trials, are sometimes suspended.

It is important to say at this point, that issues of informed consent were not included in the research questions I sought to answer, but they are worthy of comment. Lock (2010) considers that Petryna forces a reconsideration of the rules of ethical research, when people have little choice but to opt for research in the absence of medical care. Petryna was talking about globalisation and the appropriation of the bodies of the poor in developing countries for the outsourcing of clinical trials (2009). This is a growing trend due to the difficulty of finding sufficient numbers of willing populations in the west to undertake experimental trials. It is therefore important to consider the position of patients with rare conditions such as Huntington's disease, and how their bodies may be appropriated in a similar way, even with the ethical and clinical safeguards present in the practice of western medicine.

The usual meaning of signing up for research is consciously displayed as altruistic desire to help unknown future patients, along the lines of 'wanting to do anything to help'. This is also the western bioethical, social expectation of research from many angles, the clinician, the research funders, the drug companies if they are involved, and research ethics committees. However, together with the idea that the 'therapeutic misconception' is an error made by patients, and the patients' make a rational choice of research participation or no prospect of treatment, there is incongruence with the accepted bioethical framework of informed consent. In this study, research is considered differently by the people for whom no other treatment is available. Mascalzoni et al (2009) take the position that informed consent in the genomics era should be reconsidered due to the implications of gene wide association studies, and there is evidence from Petryna (2009) that this also applies to globalised, outsourced, clinical trials. It is fair to say this is a matter for the neuroscience era too, or any era where there is a shortage of patients available for clinical trials.

Concluding remarks

The thesis has attempted to answer the broad question of the effects of the movement of Huntington's disease, from genetics to neurology. All the localised clinic activities of care, neurological research data collection and disease management, took place against a background of the global bioscientific research project intended to create biomedical interventions for Huntington's disease. The local ethnography of the clinic has provided insight into three main themes which have impact on the patients, families and researchers. These are firstly, the blurring of clinical disease management and research, with new areas of negotiation present in each consultation. Secondly, the research clinic produced different types of performance from the actors which are directly associated with the prospect of experimental treatment, for example, the 'ideal patient', the 'ideal carer', and the optimistic but cautious researcher, who must negotiate the present and the future in each patient. The performance of the bioscientific research also has an effect on the way Huntington's disease is constructed within neurology, and is creating a standardised 'research friendly' form of the disease. The current placing of Huntington's disease in neurology, and the related research of foetal cell transplantation, provides the third theme, the significantly increased hopes and expectations of researchers, and of the families at risk. The three themes have important effects on the research clinic operation and serve to maintain the position of Huntington's disease as a model for other neurological disorders. Additionally there is an important requirement for the research clinician to manage the hopes of patients and families in ways which will be helpful to the research effort, maintain project recruitment and still convey the futuristic nature of any translation of research outcomes into clinical treatments.

According to Petryna (2009) the field of global experimental activity has taken shape over the last few years virtually uncharted, and although it may bring health benefits, it also leaves behind 'partial scientific evidence, unforeseen harms and new medical and legal realities'. It is to be hoped that the current Huntington's bioscientific project will produce some accessible treatment for the disease, with few if any, of the associated disadvantages that Petryna observed in her international study. In spite of the

hopes and cooperation of the patients and families, it remains to be seen if the global standardisation of the disease will yet produce a global standard treatment for Huntington's disease.

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Appendices



Mrs Jacqueline Needs
PhD student
Cardiff University
ESRC Centre for the study of Economic
& Social Aspects of Genomics
6 Museum Place, Cardiff
CF10 3BG

02 August 2007

Dear Mrs Needs

Full title of study: Changing Perspectives of Huntington's disease:
scientific, clinical and personal.
REC reference number: 07/WSE04/51

Thank you for your letter of the 1 August 2007, 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].

Ethical review of research sites



The Committee has designated this study as exempt from site-specific assessment (SSA).

There is no requirement for [other] Local Research Ethics Committees to be informed or for site-specific assessment to be carried out at each site.

Conditions of approval

The favourable opinion is given provided that you comply with the conditions set out in the attached document.

You are advised to study the conditions carefully.



Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Application	5.3	16 May 2007
Investigator CV	Paul Atkinson	11 July 2007
Investigator CV	K Featherstone	11 July 2007
Investigator CV	2 - Angus Clarke	01 January 2007
Investigator CV	Jacqueline Needs	10 July 2007
Protocol	1	12 February 2007
Covering Letter		14 May 2007
Letter from Sponsor	Cardiff University	26 March 2007
Peer Review		30 March 2007
Interview Schedules/Topic Guides	2	01 July 2007
Letter of invitation to participant	Contact 2.0	01 July 2007
Letter of invitation to participant	Access Notes 2.0	01 July 2007
Letter of invitation to participant	Observation 2.0 AC	01 July 2007
Letter of invitation to participant	Observation 2.0 AR	01 July 2007
Letter of invitation to participant	Interview 2.0	01 July 2007
Letter of invitation to participant	1 scientists & clinicians	01 July 2007
Participant Information Sheet: scientists & clinicians	2	01 July 2007
Participant Information Sheet: Observation	2	01 July 2007
Participant Information Sheet: Interview	2	01 July 2007
Participant Information Sheet: General	1	01 July 2007
Participant Information Sheet: Access to Notes	2	01 July 2007
Participant Consent Form: Access to Notes	2.0	01 July 2007
Participant Consent Form: Contact	1.0	01 July 2007
Participant Consent Form: scientists & clinicians	2.0	01 July 2007
Participant Consent Form: Interview	2.0	01 July 2007
Participant Consent Form: Observation	2.0	01 July 2007
Response to Request for Further Information		13 July 2007
Response to Request for Further Information		01 August 2007
Letter of support from Prof A Clarke		14 May 2007

R&D approval

All researchers and research collaborators who will be participating in the research at NHS sites should apply for R&D approval from the relevant care organisation, if they have not yet done so. R&D approval is required, whether or not the study is exempt from SSA. You should advise researchers and local collaborators accordingly.

Guidance on applying for R&D approval is available from
<http://www.rdforum.nhs.uk/rdform.htm>.

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.

Feedback on the application process

Now that you have completed the application process you are invited to give your view of the service you received from the National Research Ethics Service. If you wish to make your views known please use the feedback form available on the NRES website at:

<https://www.nresform.org.uk/AppForm/Modules/Feedback/EthicalReview.aspx>

We value your views and comments and will use them to inform the operational process and further improve our service.

07/WSE04/51	Please quote this number on all correspondence
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With the Committee's best wishes for the success of this project

Yours sincerely

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