Original Article

Sharing decisions on reproductive goals: A mixed-methods study of the views of women who have cystic fibrosis

Denitza Williams [a,*], Oluwaseun B Esan [b], Daniela K Schlüter [c], David Taylor-Robinson [b], Shantini Paranjothy [c], Jamie Duckers [d], Natalie Goodchild [e], Rhiannon Phillips [f]

[a] Division of Population Medicine, School of Medicine, Cardiff University, Cardiff, UK
[b] Department of Public Health, Policy and Systems, University of Liverpool, Waterhouse Building (2nd Floor, Block F), 1-5 Brownlow Street, Liverpool L69 3GL, UK
[c] Aberdeen Centre for Health Data Science, Institute of Applied Health Sciences, University of Aberdeen, Aberdeen, UK
[d] All Wales Adult CF Centre, Cardiff and Vale University Health Board, Cardiff, UK
[e] Public Involvement Partners, UK
[f] Cardiff School of Sport and Health Sciences, Cardiff Metropolitan University, Llandaff Campus, Cardiff CF5 2YB, UK

ARTICLE INFO

ABSTRACT

Background: There are complex medical, psychological, social and economic aspects to becoming a parent with Cystic Fibrosis (CF). A shared decision-making (SDM) approach could help women with CF make informed decisions about their reproductive goals that are sensitive to their individual values and preferences. This study investigated capability, opportunity, and motivation to participate in SDM from the perspective of women with CF.

Methods: Mixed-methods design. An international online survey was completed by 182 women with CF, to investigate participation in SDM in relation to reproductive goals, and measures of capability (information needs), opportunity (social environment) and motivation (SDM attitudes and self-efficacy) to engage in SDM. Twenty-one women were interviewed using a visual timelines method to explore their SDM experiences and preferences. Qualitative data were analysed thematically.

Results: Women with higher decision self-efficacy reported better experiences of SDM relating to their reproductive goals. Decision self-efficacy was positively associated with social support, age, and level of education, highlighting inequalities. Interviews indicated that women were highly motivated to engage in SDM, but their capability was compromised by lack of information, perception of insufficient opportunities for focused discussions about SDM.

Conclusions: Women with CF are keen to engage in SDM about reproductive health, but currently lack sufficient information and support to do so. Interventions at patient, clinician and system levels are needed to support capability, opportunity and motivation to engage equitably in SDM in relation to their reproductive goals.

© 2023 The Authors. Published by Elsevier B.V. on behalf of European Cystic Fibrosis Society. This is an open access article under the CC BY license (http://creativecommons.org/licenses/by/4.0/)

1. Background

Since the introduction of Cystic Fibrosis Transmembrane Conductance Regulator (CFTR) modulator therapies, people with Cystic Fibrosis (CF) are living longer healthier lives than in previous decades [1,2]. More people with CF are now considering having families of their own [3–7]. There are complex medical, psychological, social and economic aspects to becoming a parent with CF [6–8]. Women with CF would like to discuss sexual and reproductive health with their CF teams, but report difficulty accessing the information and support they need [9,10]. Pro-active discussions about reproductive choices should be included as part of the routine healthcare for CF to optimise pre-conception health and reduce the risk of unintended pregnancies [6,11,12].

Shared decision-making (SDM) is a person-centred approach that enables patients to make more informed decisions that are aligned with their personal preferences, become more active and empowered in their own healthcare, have better relationships with their health care professionals, and feel more satisfied with their choices [13]. Implementation of SDM in clinical practice involves preparation for SDM, conversations about options, development of

'informed preferences' by patients and clinicians, distributed and multi-stage decision-making (as opposed to a single discrete decision), and open-ended discussions about planning [14]. Patient readiness for SDM can be influenced by a number of factors, including health literacy, skills (consideration, self-awareness, communication), attitudes towards SDM, and socio-demographic factors [15].

Providing effective and timely support to people with CF with their decisions about their reproductive goals is vital in enabling them to make informed decisions that are sensitive to their individual values and preferences. SDM is a complex process [14,15] and a multi-level approach is required in understanding patient engagement in SDM. The Capability, Opportunity, and Motivation (COM-B) model helps to explain how interactions between people's physical and psychological capability (**C**), social and physical opportunity (**O**), and automatic and reflective motivation (**M**) can influence health-related behaviour [16]. In this study, we investigated how capability, opportunity, and motivation can influence women's participation in SDM relating to their reproductive goals. In particular, we focused on experiences of SDM, preference for SDM, unmet information needs, self-efficacy and social support. Please note the term 'women' is used to represent all individuals with CF who are able to become pregnant.

2. Method

2.1. Design

Mixed-methods study, including:

1. A cross-sectional online survey, aiming to establish to what extent women felt their information needs had been met and to identify psychological determinants of perceptions of participation in SDM
2. One-to-one semi-structured interviews with women with CF, aiming to understand in detail the experiences of women with CF of SDM in relation to reproductive goals
3. Quantitative and qualitative data was triangulated to provide an understanding of factors that could act as barriers and facilitators to participation in SDM from the perspectives of women with CF and to map this against behaviour change intervention types.

The COM-B model [16] was used to identify the target behaviour (participation in SDM) and relevant measures of capability (knowledge), opportunity (social environment) and motivation (attitudes towards SDM and perceived capability to engage in SDM) for inclusion in the survey. The model informed the development of the survey and coding framework for the qualitative analysis, providing a structured framework for data triangulation.

2.2. Participants and sampling

2.2.1. Online survey

Using a convenience sampling method, we aimed to recruit at least 120 women to the online survey between May 2020 and April 2021. Inclusion criteria were being a woman diagnosed with CF, between 18 and 49 years, and currently resident in the United Kingdom, Ireland, New Zealand, Australia, Canada or United States of America (OECD countries that have CF registries where English is recognised as an official language). Compulsory eligibility questions were set at the start of the survey and participants were asked to confirm that they had been diagnosed with CF, that they were able to become pregnant and were resident within the countries of interest . Eligibility was not further verified e.g. by access to patient records). The study was advertised through project social media feeds (Twitter, Facebook) and patient-facing organisa-

tions (CF Trust, CF Foundation) who shared the study advert on social media and relevant newsletters.

2.2.2. Qualitative interviews

Women were purposively sampled from those who had completed the survey and had expressed an interest in an interview. A maximum variation strategy was used when sampling to ensure a broad representation of individuals. Sampling considerations included people with differing disease status, family status, socio-economic background, and geographical location. We aimed to interview up to 30 women, with recruitment continuing until no significant new themes were identified [17]. Interview participants were given a £10 gift voucher as reimbursement for their time.

2.3. Data collection

2.3.1. Online survey measures

The survey was developed in collaboration with stakeholders in the UK and the US, using a combination of validated and new measures. The survey was adapted from previous research on reproductive choices with women with rheumatological conditions [18,23,24]. Cognitive interviewing was completed with three women with CF using the 'think aloud' method, and the survey was subsequently modified to improve clarity and face validity.

2.3.2. Demographics, self-reported health, and quality-of life

A range of socio-cultural, demographic, and clinical factors can influence patient readiness for SDM [25]. Therefore, demographic information was gathered on age, country of residence, employment status, family status, sexual orientation, gender identity, ethnicity, relationship status, and highest level of education. Participants were asked about their treatments, transplant status, antibiotic use, hospitalisation, Body Mass Index (BMI), lung function (FEV_1%), whether they had been diagnosed with a P. aeruginosa infection, and co-morbidities, method of contraception (if applicable).

Disease-related quality of life was assessed using six items from the treatment burden and health perceptions components of the Cystic Fibrosis Questionnaire Revised (CFQ-R) questionnaire [19,26]. The mean score from each domain is calculated and standardized to provide a score from 0 to 100, with higher scores indicating better quality-of-life.

2.3.3. Reproductive goals

Women were asked whether they intended on having children (or more children), were currently pregnant, were trying to conceive or undergoing fertility treatment, or had decided not to have children.

2.3.4. SDM behaviour: participation in SDM when accessing routine CF healthcare

A single item was included to assess general experiences of incorporation of preferences into medical decision making; "Have your Cystic Fibrosis healthcare team considered whether or not you would like to have children when talking about your treatment options (e.g. types of medication, surgery, organ transplantation)?". This was rated from 0 – not considered at all to 4 – fully considered.

Women were asked to rate a memorable conversation they had with a health professional about their options for stating a family and manging their condition using the collaboRATE measure [20]. This included three items relating to how much effort was made to: 1. help them understand your options about having children and managing your conditions; 2. to listen to the things that matter most to you; and 3. to include what matters most to them in

choosing what to do next? The items were rated from 0 (no effort was made) to 9 (every effort was made). Any item scoring <9 was coded as 0, responses of 9 are coded as 1, with the percentage of encounters coded as 1 forming the collaboRATE 'top score', indicating the extent to which 'gold standard' SDM took place [27].

2.3.5. SDM capability: unmet information needs

Women who were considering having children or were undecided were asked how important it was for them to have more information on 12 items that were relevant to reproductive goals. These were scored from 0 (not important at all) to 4 (extremely important). Topics included sex and relationships, fertility, genetic testing, other options for stating a family (e.g. adoption), preparing for pregnancy, risk of miscarriage or still birth, options for giving birth and breastfeeding. The information needs items were summed to produce a total reproductive options information needs score (Cronbach's alpha = 0.91).

2.3.6. SDM opportunity: social environment

Social support was assessed using a measure from the EN-RICHED study [28–30]. Ability to obtain social support from various sources when needed was rated from 0 (none of the time) to 4 (all of the time). A total social support score was derived by summing items 1, 2, 3, 5, and 6 of the scale [29]. Low perceived social support was defined as having a score of ≤2 on at least 2 of the 5 items, and a total score of ≤18 [29].

2.3.7. SDM motivation: SDM attitudes and decision self-efficacy

General preference for involvement in decision making was assessed using a single item from the Control Preferences Scale [21]. Women's perceived ability to make informed decisions about having children was assessed using seven items from the Decision Self-efficacy Scale (DSE)(22). The items related to two components of decision making: ability to obtain information and ability to ask questions. Items were rated on a five-point scale from 0 (not at all confident) to 4 (very confident). A mean score for the items was calculated and multiplied by 25 to provide a total score ranging from 0 to 100, with higher scores indicating higher self-efficacy [22].

Details of these measures and how they relate to the COM-B framework are provided in Appendix 1.

2.3.8. Qualitative data collection

Interviews were carried out via the telephone or video-call (e.g. Skype, Zoom, MS Teams) to gather in-depth information about factors influencing women's decision-making process in relation to their reproductive goals. Consent was obtained before interview. Visual timelines were used as an optional elicitation tool during the interviews to facilitate women in telling their own story when discussing this complex and sensitive topic [18,31]. A resource pack was emailed to participants ahead of the interview containing the participant information sheets, consent form and 'what to expect' sheet outlining the focus of the interview, and a timeline template (Appendix 2). Preparing a timeline was voluntary and women were encouraged to use their own formats [e.g. notes, diagrams they had drawn] as an alternative if they wished to do so .The researcher used a set of prompts to further explore women's decision-making preferences and experiences. Interviews were audio-recorded, transcribed verbatim and anonymised prior to analysis. DW who does not have CF and who has two children conducted all the interviews.

2.4. Analytical techniques

2.4.1. Survey data

IBM SPSS v27 was used for statistical analysis. Descriptive analysis was carried out to characterise the study population in terms of their demographic characteristics, health and experiences of SDM. We carried out exploratory analysis to establish whether variables relating to capability, opportunity and motivation were independently associated with perceived experience of participation in SDM in relation to reproductive goals and impact of having children already. We fitted a multivariable linear regression model for the CollaboRATE mean score in relation to discussion with health professionals about reproductive choices as the outcome variable and the following variables as predictors: CFQ-R treatment burden, CFR-Q health perceptions scores, Decision Self-Efficacy, EN-RICHED Social Support score, age, and highest level of education (college educated/not college educated). The 'enter' method of regression was used with missing cases excluded listwise. To better understand the determinants of women's confidence in their ability to make choices about their reproductive goals, a multivariable linear regression model was also fitted for Decision Self-Efficacy using the same set of predictor variables, with the exception of the Decision Self-efficacy score. Based on Green's [32] rule of thumb for testing individual predictors, $N = 104 + m$ (where m is the number of predictors), we estimated that a minimum sample size of 109 people would be required for our planned regression analysis.

2.4.2. Qualitative data

Qualitative data were analysed thematically through a process of familiarisation, generation of initial codes, searching for themes, reviewing and refining themes, and defining and naming themes [33]. NViVO software (Version 12) was used. DW coded the data. The qualitative team met regularly over the course of the study to debrief and reflect on their experiences. The timelines were not used as part of the analysis process, as they were used primarily as elicitation and engagement method. Themes identified in the qualitative data were organised under the capability, opportunity, and motivation elements of the COM-B [16]. The data was then triangulated with the quantitative survey findings to provide an understanding of factors that could act as barriers and facilitators to participation in SDM from the perspectives of women with CF.

3. Findings

Overall, 184 women gave consent and started the survey. Two indicated that they did not live in the countries listed and were excluded. The survey was therefore completed by 182 women from the USA ($n = 102$, 56%), UK ($n = 58$, 31.8%), and Canada ($n = 20$, 11.1%). Location data was missing for $n = 2$ (1.1%) people. Participants were aged between 20 and 49 years (mean age 31.9, SD 6.53). The majority of participants were college educated ($n = 155$, 85.2%), in a long-term relationship ($n = 152$, 83.5%), white ($n = 173$, 95.1%), and heterosexual ($n = 163$, 89.6%). Forty-five women (24.7%) had children already. In terms of reproductive goals, 59 (32.4%) women had decided that they didn't want to have children/more children, 66 (36.3%) wanted to become pregnant, were pregnant, or receiving fertility treatment, 38 (20.9%) were undecided, and 19 (10.4%) would like to have a child but did not plan on getting pregnant (e.g. through adoption or surrogacy). Details of self-reported health and reproductive status of participants is provided in Table 1. Descriptive statistics for disease-related quality of life and SDM measures for women who did and did not have children already are provided in Table 2.

Most women ($n = 173$, 95%) felt that they and their doctor should be involved in making decisions about their care and the average level of decision self-efficacy in this sample was high (mean of 80.51 on a scale from 0 to 100). Women who had children already had better perceived health 95% CI [2.56,17.99], higher decision self-efficacy 95% CI [5.90,17.13], rated the extent to which their preferences for having children had been considered by their

Table 1
Self-reported health and reproductive status of survey participants ($n = 182$).

Variable	Category	N	%
CF Treatments	CFTR modulators (e.g. Ivacaftor, Kalydeco, Lumacaftor, Ivaca)	128	70.3
	Medicines to help with mucus (e.g. DNase, dornase alpha, hypertonic saline, Mannitol dry powder)	148	81.3
	Inhaled/nebulized antibiotics	132	72.5
	Insulin injections	72	39.6
	Medicines to replace pancreas enzymes (e.g. Creon or Pancrease)	161	88.5
	Other treatments for CF	65	35.7
Antibiotics in the last year	Oral antibiotics received in the last year (in addition to long-term antibiotics if applicable)	159	87.4
	IV antibiotics received in the last year	93	51.1
Hospital in-patient stays in the last year	Yes	75	41.2
FEV1%	40% or under	18	9.9
	41–70%	68	37.4
	70% and above	94	51.6
	Missing data	*2*	*1.1*
Ever diagnosed with a P. aeruginosa infection	Yes	131	72
Lung transplant	Have had a lung transplant	9	4.9
	Told by doctor that a lung transplant is likely to be needed in the next 5 years/on waiting list for transplant	9	4.9
Body Mass Index	Less than 18.5	13	7.1
	18.5 to 24.9	128	79.2
	25 to 29.9	29	15.9
	30 and above	7	3.8
How many children do you have?	None	137	75.3
	1 child	28	15.4
	2 children	14	7.7
	3 or more children	3	1.6
Ever been pregnant	Yes	48	26.4
	No	134	73.6
Pregnancy loss (ever experienced)	Miscarriage	16	8.8
	Ectopic pregnancy	1	0.5
	Still birth	2	1.1
	Termination	2	1.1
	Rather not say	1	0.5
Preferences for having children	Don't want to have children/more children	59	32.4
	Not sure	38	20.9
	Would like to become pregnant	55	30.2
	Pregnant	6	3.3
	Receiving fertility treatment	5	2.7
	Would like to have a child, but don't plan on getting pregnant (e.g. adoption/surrogacy)	19	10.4
	Not sure	7	15.6

healthcare team more highly 95% CI [0.83,1.70], and had higher mean CollaboRATE scores 95% CI [0.44,2.19] than those who did not have children. There were no differences between these groups in decision control preferences (Phi = 0.51, approximate $p = 0.976$).

Unmet information needs reported by women who were considering pregnancy or who were unsure about their reproductive goals are summarised in Fig. 1.

3.1. Multivariable regression models

The multivariable linear regression model for CollaboRATE mean score was statistically significant ($F_{6,141} = 18.04$, $p < 0.001$, adjusted R square = 0.41). The multivariable linear regression model fitted for decision self-efficacy was statistically significant ($F_{5,143} = 5.180$, $p < 0.001$, adjusted R square = 0.124). Beta values and 95% CIs for predictors included in the multivariate linear regression models for CollaboRATE mean score and decision self-efficacy are provided in Table 3. Decision self-efficacy was the only variable in the model that was independently associated with the CollaboRATE mean score. In turn, women reporting more social support, who had been college educated, and were older had higher levels of decision self-efficacy.

3.2. Qualitative interview findings

Twenty-one women were interviewed. Participants were based in the UK ($n = 7$) or the US ($n = 14$), between the age of 26–45yrs. Most participants were: heterosexual ($n = 18$), college educated ($n = 18$), employed ($n = 14$), did not have children already ($n = 11$), were on CFTR modulators ($n = 16$) and had a lung function over 70% ($n = 15$). Eight participants used a timeline to structure the account of their decision-making experiences. Interviews lasted between 20 and 53 min (average 38 min). Key themes identified are summarised in Table 4, mapped against the COM-B domains.

Theme 1SDM capability: knowledge gaps in making informed reproductive decisions

3.2.1. Knowledge about impact of CF on fertility

Although some individuals had discussed reproductive health with their healthcare professionals, women often stated that they did not have sufficient knowledge about this. Women reported a lack of information specific to making decisions about their reproductive goals, particularly in relation to the impact of CF and CF medications on their contraception options, fertility, impact on their CF, impact of their CF on the infant and breastfeeding.

3.2.2. Knowledge about impact of pregnancy on CF

Women reported a need for more information about the potential impact of pregnancy on their CF. Generally, women who reported lower lung functions perceived that having children would detrimentally impact their condition or that they were too unwell for pregnancy and were less likely to plan on having children naturally. Women also wanted scientific information focusing on out-

Table 2
Disease-related quality-of-life and SDM overall and for women who did and did not have children already.

| Variable	Scoring information Possible range and direction of scores	All (n = 182) Mean (SD)	Have children already (n = 45) Mean (SD)	Do not have children (n = 137) Mean (SD)	95% CI of the difference Lower, Upper
CFQ-R health perception score	*0 to 100, higher scores indicate better quality-of-life*	68.1 (23.1)	75.8 (23.1)	65.5 (22.6)	2.56, 17.99
CFQ-R treatment burden score	*0 to 100, higher scores indicate better quality-of-life*	58.5 (21.8)	58.3 (24.3)	58.6 (21.0)	−7.70, 7.13
ENRICHED social support total score	*0 to 20, higher scores indicate more social support*	18.0 (3.0)	17.4 (3.7)	18.2 (2.7)	−2.05, 0.55
Reproductive options information needs total score for women considering having children (n = 130)	*0 to 48, higher scores indicate greater information needs*	46.5 (11.9)	44.7 (14.9)	47.1 (10.7)	−8.14, 3.20
Decision-self efficacy Scale: Making choices about having children	*0 to 100, higher scores indicate greater self-efficacy*	80.5 (19.0)	89.1 (15.4)	77.6 (19.2)	5.90, 17.13
Preferences for having children considered by healthcare team in decisions about treatment	*0 to 4, higher scores indicate greater consideration of preferences*	2.4 (1.7)	3.3 (1.1)	2.0 (1.7)	0.83, 1.70
CollaboRATE mean score: discussion with health professional about preferences for having children	*0 to 27, higher scores indicate more SDM*	6.3 (2.6)	7.3 (2.2)	6.0 (2.7)	0.44, 2.19
Control Preferences Scale		N (%)	N (%)	N (%)	
I prefer to leave all decisions regarding treatment to my doctor		3 (1.6)	1 (2.2)	2 (1.5)	
I prefer that my doctor makes the final decision about which treatment will be used, but seriously considers my opinion		30 (16.5)	8 (17.8)	22 (16.1)	
I prefer that my doctor and I share the responsibility for deciding which treatment is best for me		87 (47.8)	22 (48.9)	65 (47.4)	
I prefer to make the final decision about my treatment after seriously considering my doctor's opinion		56 (30.8)	12 (28.9)	43 (31.4)	
I prefer to make the decision about which treatment I receive		6 (3.3)	1 (2.2)	5 (3.6)	
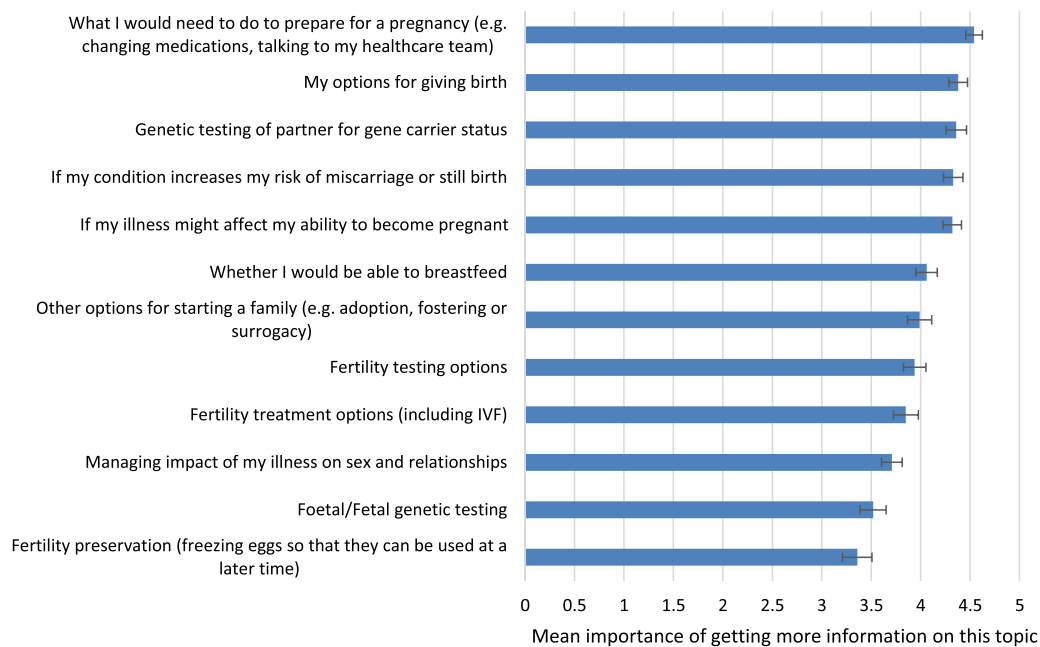
Fig. 1. Perceived importance of getting more information on topics relating to reproductive options for women considering having children/more children (n = 123).

Table 3
The association of decision self-efficacy and shared decision making as assessed by CollaboRATE mean score.

Variable	Beta	95% CI lower bound	95% CI upper bound	P value
Model 1: Collaborate mean score				
CFQ-R health perceptions	−0.006	−0.02	0.011	0.499
CFQ-R treatment burden	0.009	−0.007	0.026	0.268
Decision self-efficacy	0.088	0.069	0.107	<0.001
Social support	0.054	−0.064	0.172	0.363
Not college educated vs college educated	0.853	−0.166	1.872	0.100
Age	0.014	−0.041	0.069	0.613
Model 2: Decision self-efficacy				
CFQ-R health perceptions	0.149	−0.002	0.300	0.052
CFQ-R treatment burden	−0.042	−0.190	0.106	0.574
Social support	1.097	0.060	2.133	0.038
Not college educated vs college educated	−13.86	−22.684	−5.041	0.002
Age	0.829	0.359	1.299	<0.001

Table 4
Summary of themes identified in the participatory qualitative interviews with women with CF (*n* = 21).

COM-B Domain	Main theme	Sub-theme	Example quotes
Capability	Psychological capability: Knowledge	Lack of information on contraceptive options for women with CF	I've been told I could take the pill, there were ones that I could take, but they couldn't guarantee that I could just take it and not have to worry, that I would still have to be on top of what antibiotics am I taking. They couldn't guarantee that my decline in health wouldn't affect it, so I thought, what's the point? (P2)
		Need for more information about the potential impact of a pregnancy on women's CF	I think mostly as far as health wise, 'If you get pregnant, here's what the possibilities of what could happen CF wise. Here's the kind of medications that you could take and that you probably wouldn't want to take, and here's what the effect of that might be on you.' (P12)
		Need for information about the potential impact of CF on the child	I think just what to watch for, what to, you know, and maybe signs of more information about what about babies that have come, you know, that were born from CF parents that do, and don't have CF, you know, what's their mortality rate? You know, like, what's their, you know, more of that information as well, because you don't really, nobody really knows. (P21)
		Need for information about fertility and fertility treatments	What I understand is that sometimes a woman with CF do have a little bit of trouble conceiving. But no one ever spoke to me about it, so I don't know if that was because I was already approaching my 30 s, or if it was a result of CF. I still don't know but I think that would be really helpful now. (P11)
			The fertility treatments I did were pretty non-invasive. If I was doing IVF or something like that, then I think there would be need to be communication with the CF doctor. Physically, can my body handle this? Where I am on my CF road, can my body handle this? Is that gonna be a good choice? (P12)
	Psychological capability: Planning	Need for genetic screening of partner	There's no fun-ness, is there, you know. I, I've sort of spoken to [name] who's the psychologist and you know, okay, so, first thing we have to get him (partner) tested. Well, firstly we have to decide that we'd want to pursue this. Then he needs to go and get tested. We wait for that result. Okay, and then from the result, it's the two parts. Okay, if he's a carrier, we're going to have to go down a scientific route, and if he's, you know, if he's not a carrier, great, we can go down a more natural route. (P10)
			Like I was fine if I had a child with CF, that I would love them really well and take great care of them. I thought, like, if I needed vitro fine, but I wanted to have biological children. (P19)
		Planning to have children, but not via pregnancy (e.g. adoption, surrogacy)	But now it's planning for what now? Do we stay and do nothing? Do we try to adopt? Like that, that was our, our focus was adoption. You know I don't know and what I had hoped for when I hit adults [adult CF services], would there be someone to talk with you, like about your options, about your family planning, if you want it. You know not everyone's gonna want it, but at least have that option be there. (P7)
		Planning for periods of ill health/reduced life expectancy	Early on it's really difficult to get a decent night's sleep and if you get to a place where you're sufficiently tired, or your lungs are sufficiently, you know, poorly working or infected, that, that's really starting to take a toll on your health, I think you need a, you need a, somebody to step in when you wave the white flag. You know like you need a grandparent to come help, you need your spouse to like really step up and give you a couple of nights of like actual sleep. ...But if people aren't sort of thinking about that ahead of time, you know, like that would be something to, to think about as like what are you gonna do when you're not feeling well and you need help? What are your choices for like finding those extra, extra helpers? (P13)
			So that was also really a fear of mine, of, you know, would it [pregnancy] make me sicker, would it make handling my illness more difficult, or what would happen if I were to pass away, what kind of an impact would that have on a young child or, you know, even a teenager. (P17)

Table 4 (*continued*)

COM-B Domain	Main theme	Sub-theme	Example quotes
Opportunity	Physical environment: the healthcare system	Clinical reviews	It was more that they, it was quite, yeah it did, it, mainly annual reviews with the CF nurse/specialist and it would be more a, kind of, tick box exercise of did, like 'Are you planning to have children? And obviously my answer would always be no ...And it would be left at that or they'd be like, "Okay well that's fine but if you change your mind... (P 1)
		Competing priorities during healthcare consultations	Googled a lot of stuff, instead of... you know. Not that I want, well didn't want to bother the CF clinic, I just, you know, there's more important things that the CF clinic is doing, so I tend not to bother them unless I have to. (P15)
		Holistic and multi-disciplinary healthcare services	And my diabetes doctor, who's not part of my CF clinic here, she got me onto an insulin pump and I like learned how to manage that [interference on recording 00:12:42] in the months leading up to the first pregnancy, so that I would be sort of better able to stay really on top of the, the diabetes aspect of it. (P13) I had my OB appointments scheduled and my nurse actually got it to where she kind of pencilled me in with my doctor, my CF doctor, so that I could see her on the same day, so I didn't have to make both trips. (P16)
	Social environment: learning from experiences of peers	Influence of personal stories of women with CF who had children on reproductive goals	So, this was when I lived in [place] and it was quite a small CF community and everyone, kind of, knew everyone, and one of the girls that had CF fell pregnant when she was 18, and had the baby. And I remember talking to my physiotherapist at the time about [the woman who became pregnant] and saying, like, "Has she recovered well?" And she said, "Oh no she's lost quite a bit of her lung function," which probably she shouldn't have disclosed but anyway she did. And at the time I thought, "Oh my gosh, like, I can't risk doing that. (P23) And they [health professionals] did tell me, you know, that they had, they had patients who had given birth and were doing great, and it seemed like many women with CF don't have issues with fertility or anything like that. So, it gave me a lotta hope regarding that. (P14)
Motivation	Reflective motivation	Impact of emergence of new treatments on reproductive goals and expectations	Had my 26th birthday, I was talking to my doctor and she told me about so many CF women who were pregnant now because of the Trikafta, and it, kind of, got me a little excited... it's actually really exciting for me cos I can look forward to actually having a family now, instead of a few years back where I was just crushed and just... One of my biggest dreams is, is what I refer to it too as, as being a mother and having a family, and when I was 22 it just was taken away from me. (P18) So when I was a kid I was told I'd live till two, so I was therefore given no suggestion at all of, of marriage or children or anything like that as I grew up. It was 'if you make it to 18 you will have been a real survivor'. Because I was born a long time ago, obviously, 1974 CF care was pretty minimal." (P14) And especially like now I think I, I'm on a, you know, one of the Facebook groups and I've been reading about women who, like, for years have not been able to get pregnant but now that they're on Trikafta they're getting pregnant. (P11)
		Emotional impact of previous experience of communication with health professionals	She [the doctor] said, "If you do decide to have children and bear your own child you might not make it to term, and you, your lung function will decrease and you'll never get it back and you'll need a lung transplant immediately. And that's if your lungs can go through all of the things that go with pregnancy and still manage to have the baby. Right after that you would have to have a lung transplant because you, your lung function would be 15 at best." I took, I took that really, really hard, and my doctor was, like, "There are other options though, there's always adoption and, you know, some people already have children that you can get to know and relationship wise – (P18) I'm thinking back to what the hospital were saying, they weren't saying, having children was a good idea. In fact I remember one doctor... a long, long time ago. He's a lovely gentleman, but was very traditional in, in sort of the way that things were thought, and he said, you know, getting pregnant if you're a CF woman is, is a suicide event really, you're, you're not going to do well after it, we've got research that says that (P14). It's all very well and good doctors saying to you, oh well you could have [had children], you could have, if you'd thought of this before we could have done this. And it's like, but we didn't know what was gonna be invented. You can't, you can't live your life in that level of hope. I've only stayed about a year ahead of things as it is, you know. So when I was given Kaftrio I was still being prescribed oxygen. You, at that point you are preparing for end stage, you are not preparing for having a child. (P14)

comes for children of women with CF as evidence relating to this is currently lacking. Women felt that information about the reality of having a child whilst managing CF would be useful and reflected on the need for preparation for support with parenting, particularly when they are unwell.

3.2.3. Planning

Planning was an integral part of pre-conception decision-making for all women. Women felt that they had to make a conscious and deliberate decision about whether to have children. Planning involved reflection on changes in prognosis and morbidity, health status, healthcare professional recommendations about feasibility of having children, potential impact on CF, genetic

screening of partner, and personal stories of women with CF who have been pregnant and/or have children.

3.2.4. Theme 2 Sdm opportunity for preference based reproductive discussions

Some participants reported a lack of initiation of discussions focusing on pre-conception decision making by healthcare professionals, whilst others were more satisfied with the healthcare communication. Participants reported a desire to be seen holistically as an individual, rather than being defined by their disease. A multi-disciplinary approach was important in facilitating pre-conception decisions and supporting women through conception and pregnancy.

3.2.5. Annual review

Often discussions about reproductive goals would take place during routine annual reviews. However, this was often perceived to be part of a brief "tick box" exercise within a wider process, without providing an opportunity for focused discussion. Some individuals felt that they did not wish to 'bother' the CF clinicians, who they felt had more pressing priorities, with questions about their reproductive goals.

3.2.6. Support

Some women reported high satisfaction with support received from healthcare professionals with making decisions about starting a family. However, lower satisfaction was reported when women were advised not to have children if this was not aligned with their personal goals. Some women reported these discussions were not handled sensitively and that there was with a lack of follow-on support.

Personal stories of women with CF who had children were important when deciding on their reproductive goals. Direct comparisons with the level of disease management and health status were important in influencing decisions. The source of the patient stories did not seem to be significant. However, the alignment between the values and health status of the individual in the patient story and the participants was important.

Theme 3 SDM motivation for preference based reproductive discussions.

Women reflected on the enormity of the improved treatments for CF throughout their lifetime in terms of the impact of life expectancy, identity, and aspirations for having a family. For some, the advances in CF treatments had not arrived soon enough for them to consider having children and it was important that healthcare professionals were sensitive to the resulting sense of loss they experienced. The availability of new treatments was often seen as a key motivator for discussions focusing on reproductive goals.

4. Discussion

In this study, we investigated women with CF's perspectives on how capability, opportunity, and motivation influenced participation in SDM in relation to reproductive goals. Women with CF were highly motivated to engage in SDM but there were significant gaps in the provision of information which affected their capability to do so. Opportunities to have focused discussions with CF healthcare teams about reproductive goals was limited. Social support was important for confidence in engaging in SDM, in particular the opportunity to learn from the experiences of other women with CF. Motivation to engage in SDM was influenced by changing attitudes towards reproductive goals as a result of new treatments becoming available and the new possibilities this bought. The emotional impact of past experiences of discussing reproductive goals with health professionals was an important aspect of motivation to engage in SDM.

As has been previously reported [9,10], we also observed an unmet need for information on reproductive health, however we identified this at an international level. This shows that widespread effort is needed to promote person-centred decision making for pre-conception care. Similar to a recent review our study identified fragmented care [34] experienced by patients, with need for focused pre-conception conversations. Our study identified the effect of self-efficacy on experience of shared-decision making in consultations and the potential impact this could have on pre-conception decision making. Decision support tools can provide tailored information, help women understand their options and clarify their preferences. Decision support tools that focus on facilitating a SDM discussion with health professionals focusing on disease specific, reproductive goals, such as 'My Voice CF', provide

a promising approach to facilitating preference-based decision-making for women with CF [12,35]. Further development and evaluation of such decision aids is required.

Women with CF in our study reported lack of opportunities for focussed discussion of their reproductive goals in their routine CF healthcare, similar to what has been found in other long-term conditions [18]. For long-term conditions, decision-making is often a distributed and multi-stage process and open-ended planning has been identified as an important aspect of implementation of SDM in routine practice [14].

The relationship between patient confidence, knowledge and skills and engagement in SDM appears to be bi-directional [36]. Ensuring that clinicians have the information, time, skills, and confidence to engage in these complex and emotive conversations about reproductive goals is important in motivating women to take part in SDM and ensuring that their values and preferences are considered. For women who are not able to achieve their reproductive goals, or who feel a sense of loss relating to what could have been had they been given different advice or had access to effective treatments sooner, emotional support is important following on from these conversations.

Women's social environment was important to their decision-making process. Social support encompasses informational, emotional, instrumental and appraisal support, which can come from a variety of sources [37]. Provision of emotional support (empathy, care, and concern) and informational support (assistance with knowledge, information and skills) are particularly important aspects of the patient-clinician relationship [37], and clinicians' interpersonal skills are likely to influence women's confidence and motivation to engage in SDM. Planning for parenting involved women's informal social support networks, particularly when considering contingency plans for caring for children in the event of deteriorated health. Women also expressed a desire to learn about the experiences of peers. This highlights the need to involve women's support network when designing and implementing SDM interventions.

This mixed-methods study applied an established behaviour change theory, the COM-B model, in a novel way to identify determinants of women's engagement in SDM. In-depth information was gathered from 182 women in three OECD countries where different healthcare systems are in place and the socio-cultural context differs. Women had varied health status and reproductive goals yet there was a striking consistency in their experiences, particularly with regard to unmet information needs and lack of opportunities within routine CF care to engage in SDM in relation to reproductive goals.

To reduce inequalities in health and to facilitate a person-centred approach to pre-conception decision making within CF, changes at individual (micro) and organisational (macro) level are needed [38]. Recommendations from this study include: investment in shared decision-making training for clinical staff, initiation of pre-conception conversation by healthcare professionals, incorporation of a broader focus on reproductive and pre-conception health options in consultations, pre-consultation preparation for women for person centred conversations, co-development of decision support tools for women, and specific support for those who are disadvantaged.

Limitations of this study were the use of a cross-sectional self-report method in a non-random sample. Due to the recruitment method, a survey response rate is not available and it is likely that women who had a particular interest in pre-conception decision-making or those who have had particularly positive or negative experiences self-selected to participate. Our participants were predominantly highly educated and ethnic minority communities were under-represented. There was no representation of experiences from individuals living in Ireland, New Zealand and Aus-

tralia. The women who had children were generally older than others in the CF population who had children [8]. The aim of this study was not to compare the experiences of women who had children and those who did not, however longitudinal research with larger groups would be useful to facilitate further understanding of how the decision-making process unfolds over time and at different life stages. While this study provided in-depth information on the experiences of women who took part, the methodology did not allow for meaningful comparisons between healthcare systems in different countries and results cannot be generalised to the general Cystic Fibrosis population. Further research focusing on other populations such as under-served groups would be of benefit in understanding inequalities in engagement with SDM in relation to reproductive goals.

5. Conclusions

Treatment advances for CF have led to a rapidly changing landscape for patients, where having a family is now a real possibility for many women. There is an increased need to provide person-centred support with these complex and emotive decisions. The implementation of SDM in relation to reproductive goals for women with CF is likely to require a multi-level approach that supports women with their decisions, providing information, social support, and regular opportunities to have focused discussions about with healthcare professionals about their reproductive goals. Planning should be part of a person-centred package including appropriate follow-up and emotional support when needed. Considerations need to be made for those who are less likely to engage with healthcare services to support person-centred and equitable healthcare provision as well as preferences for virtual or face-to face discussions.

5.1. Healthcare professional considerations

- Half of women would like their healthcare professionals to initiate reproductive conversations.
- Reproductive goals should be explored periodically, possibly during annual reviews with follow-up focused discussions if required.
- Healthcare professionals need reliable up-to-date information about the likely impact of pregnancy on CF and vice versa, which can be shared with patients.
- Healthcare professionals need to be aware of the impact of rapidly changing treatment scene and new possibilities within CF care and the impact that can pose of women's identity.

Declaration of Competing Interest

No declarations of interest presented by authors.

CRediT authorship contribution statement

Denitza Williams: Conceptualization, Methodology, Formal analysis, Writing – original draft. **Oluwaseun B Esan:** Writing – review & editing. **Daniela K Schlüter:** Writing – review & editing. **David Taylor-Robinson:** Writing – review & editing. **Shantini Paranjothy:** Writing – review & editing. **Jamie Duckers:** Supervision, Conceptualization, Funding acquisition. **Natalie Goodchild:** Conceptualization, Writing – review & editing. **Rhiannon Phillips:** Conceptualization, Methodology, Writing – original draft, Supervision.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jcf.2023.02.007.

References

[1] The Cystic Fibrosis Trust. Cystic fibrosis strength in numbers: uk cystic fibrosis registry annual data report 2016. London; 2017.

[2] McBennett KA, Davis PB, Konstan MW. Increasing life expectancy in cystic fibrosis: advances and challenges. Pediatr Pulmonol 2022;57(Suppl 1):S5–s12 Suppl 1.

[3] Heltshe SL, Godfrey EM, Josephy T, Aitken ML, Taylor-Cousar JL. Pregnancy among cystic fibrosis women in the era of CFTR modulators. J Cyst Fibros 2017;16(6):687–94.

[4] Edenborough FP, Borgo G, Knoop C, Lannefors L, Mackenzie WE, Madge S, et al. Guidelines for the management of pregnancy in women with cystic fibrosis. J Cyst Fibros Off J Eur Cyst Fibros Soc 2008;7(Suppl 1):S2–32.

[5] Sawyer SM, Phelan PD, Bowes G. Reproductive health in young-women with cystic-fibrosis - knowledge, behavior and attitudes. J Adolesc Health 1995;17(1):46–50.

[6] Jain R., Kazmerski T.M., Zuckerwise L.C., West N.E., Montemayor K., Aitken M.L., et al. Pregnancy in cystic fibrosis: review of the literature and expert recommendations. J Cyst Fibros.

[7] Hailey CE, Tan JW, Dellon EP, Park EM. Pursuing parenthood with cystic fibrosis: reproductive health and parenting concerns in individuals with cystic fibrosis. Pediatr Pulmonol 2019;54(8):1225–33.

[8] Esan O, Schlüter D, Phillips R, Cosgriff R, Paranjothy S, Williams D, et al. Pregnancy rates and outcomes in women with cystic fibrosis in the UK: comparisons with the general population before and after the introduction of disease–modifying treatment. BJOG Int J Obstet Gynaecol 2021;n/a(n/a):2003–17.

[9] Kazmerski TM, Gmelin T, Slocum B, Borrero S, Miller E. Attitudes and decision making related to pregnancy among young women with cystic fibrosis. Matern Child Health J 2017;21(4):818–24.

[10] Gage LA. What deficits in sexual and reproductive health knowledge exist among women with cystic fibrosis? A systematic review. Health Soc Work 2012;37(1):29–36.

[11] Havermans T, Abbott J, Colpaert K, De Boeck K. Communication of information about reproductive and sexual health in cystic fibrosis. Patients, parents and caregivers' experience. J Cyst Fibros 2011;10(4):221–7.

[12] Stransky OM, Pam M, Ladores SL, Birru Talabi M, Borrero S, Godfrey EM, et al. Engaging stakeholders in the development of a reproductive goals decision AID for women with cystic fibrosis. J Patient Exp 2021;9. doi:10.1177/23743735221077527.

[13] DaSilva D. Helping people chare decision making. London: The Health Foundation; 2012.

[14] Joseph-Williams N, Williams D, Wood F, Lloyd A, Brain K, Thomas N, et al. A descriptive model of shared decision making derived from routine implementation in clinical practice ('Implement-SDM'): qualitative study. Patient Educ Couns 2019;102(10):1774–85.

[15] Keij SM, van Duijn-Bakker N, Stiggelbout AM, Pieterse AH. What makes a patient ready for Shared Decision Making? A qualitative study. Patient Educ Couns 2021;104(3):571–7.

[16] Michie S, van Stralen MM, West R. The behaviour change wheel: a new method for characterising and designing behaviour change interventions. Implement Sci 2011;6:42.

[17] Fusch PI, Ness LR. Are we there yet? Data saturation in qualitative research. Qual Report 2015;20(9):1408.

[18] Phillips R, Pell B, Grant A, Bowen D, Sanders J, Taylor A, et al. Identifying the unmet information and support needs of women with autoimmune rheumatic diseases during pregnancy planning, pregnancy and early parenting: mixed-methods study. BMC Rheumatol 2018;2(1):21.

[19] Henry B, Aussage P, Grosskopf C, Goehrs JM. Development of the cystic fibrosis questionnaire (CFQ) for assessing quality of life in pediatric and adult patients. Qual Life Res 2003;12(1):63–76.

[20] Elwyn G, Barr PJ, Grande SW, Thompson R, Walsh T, Ozanne EM. Developing CollaboRATE: a fast and frugal patient-reported measure of shared decision making in clinical encounters. Patient Educ Couns 2013;93(1):102–7.

[21] Degner LF, Sloan JA, Venkatesh P. The control preferences scale. Can J Nurs Res 1997;29(3):21–43.

[22] O'Connor AM. User manual - decision self-efficacy scale. Ottawa: Ottawa Hospital Research Institute; 1995.

[23] Phillips R, Williams D, Bowen D, Morris D, Grant A, Pell B, et al. Reaching a consensus on research priorities for supporting women with autoimmune rheumatic diseases during pre-conception, pregnancy and early parenting: a Nominal Group Technique exercise with lay and professional stakeholders [version 1; peer review: 2 approved]. Wellcome Open Res 2018;3(75).

[24] Ackerman IN, Jordan JE, Van Doornum S, Ricardo M, Briggs AM. Understanding the information needs of women with rheumatoid arthritis concerning pregnancy, post-natal care and early parenting: a mixed-methods study. BMC Musculoskelet Disord 2015;16(1):194.

[25] Stransky OM, Pam M, Ladores SL, Birru Talabi M, Borrero S, Godfrey EM, et al. Engaging stakeholders in the development of a reproductive goals decision AID for women with cystic fibrosis. J Patient Exp 2022;9. doi:10.1177/23743735221077527.

[26] Quittner AL, Buu A, Messer MA, Modi AC, Watrous M. Development and validation of the cystic fibrosis questionnaire in the united states: a health-related quality-of-life measure for cystic fibrosis. Chest 2005;128(4):2347–54.

[27] Barr PJ, Thompson R, Walsh T, Grande SW, Ozanne EM, Elwyn G. The psychometric properties of CollaboRATE: a fast and frugal patient-reported measure of the shared decision-making process. J Med Internet Res 2014;16(1):e2.

[28] Enhancing recovery in coronary heart disease patients (ENRICHD): study design and methods. The ENRICHD investigators. Am Heart J 2000;139(1 Pt 1):1–9.

[29] Enhancing recovery in coronary heart disease (ENRICHD): baseline characteristics. Am J Cardiol 2001;88(3):316–22.

[30] Vaglio J, Conard M, Poston WS, O'Keefe J, Haddock CK, House J, et al. Testing the performance of the ENRICHD Social Support Instrument in cardiac patients. Health Qual Life Outcomes 2004;2(1):24.

[31] Pell B, Williams D, Phillips R, Sanders J, Edwards A, Choy E, et al. Using visual timelines in telephone interviews: reflections and lessons learned from the star family study. Int J Qual Methods 2020;19:1609406920913675.

[32] Green SB. How many subjects does it take to do a regression analysis. Multivariate Behav Res 1991;26(3):499–510.

[33] Clarke V, Braun V. Thematic analysis. Encyclopedia of critical psychology. Springer; 2014. p. 1947–52.

[34] Kazmerski TM, Borrero S, Tuchman LK, et al. Provider and patient attitudes regarding sexual health in young women with cystic fibrosis. Paediatrics 2016;137(6).

[35] Leech MM, Stransky OM, Talabi MB, Borrero S, Roe AH, Kazmerski TM. Exploring the reproductive decision support needs and preferences of women with cystic fibrosis·. Contraception 2021;103(1):32–7.

[36] Poon BY, Shortell SM, Rodriguez HP. Patient activation as a pathway to shared decision-making for adults with diabetes or cardiovascular disease. J Gen Intern Med 2020;35(3):732–42.

[37] Ommen O, Thuem S, Pfaff H, Janssen C. The relationship between social support, shared decision-making and patient's trust in doctors: a cross-sectional survey of 2,197 inpatients using the Cologne Patient Questionnaire. Int J Public Health 2011;56(3):319–27.

[38] Whitehead M. A typology of actions to tackle social inequalities in health. J Epidemiol Community Health 2007;61(6):473–8.