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

Running title: Sharing decisions on reproductive goals

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Highlights (85 characters)

- Women with CF were highly motivated to participate in shared decision-making about reproductive health.
- Lack of information specific to people with CF about reproductive choices compromised capability to engage in shared decision-making.
- There was a perception of few opportunities for discussions with clinicians about reproductive goals.
- Multi-level interventions are needed to support person-centred care about reproductive goals, for example when choosing medication

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.

Key words

Cystic Fibrosis, pre-conception, shared decision-making, person-centred care, mixed-method, reproductive goals

Declarations of interest

No declarations of interest presented by authors.

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.

Method

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.

Participants and sampling

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

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.

Data collection

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.

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 (FEV₁%), 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.

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.

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 starting a family and managing 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).

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 starting 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).

SDM Opportunity: Social environment

Social support was assessed using a measure from the ENRICHED 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).

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.

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.

Analytical techniques

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, ENRICHED 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.

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.

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.

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
	<i>Missing data</i>	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

Preferences for having children	Termination	2	1.1
	Rather not say	1	0.5
	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

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

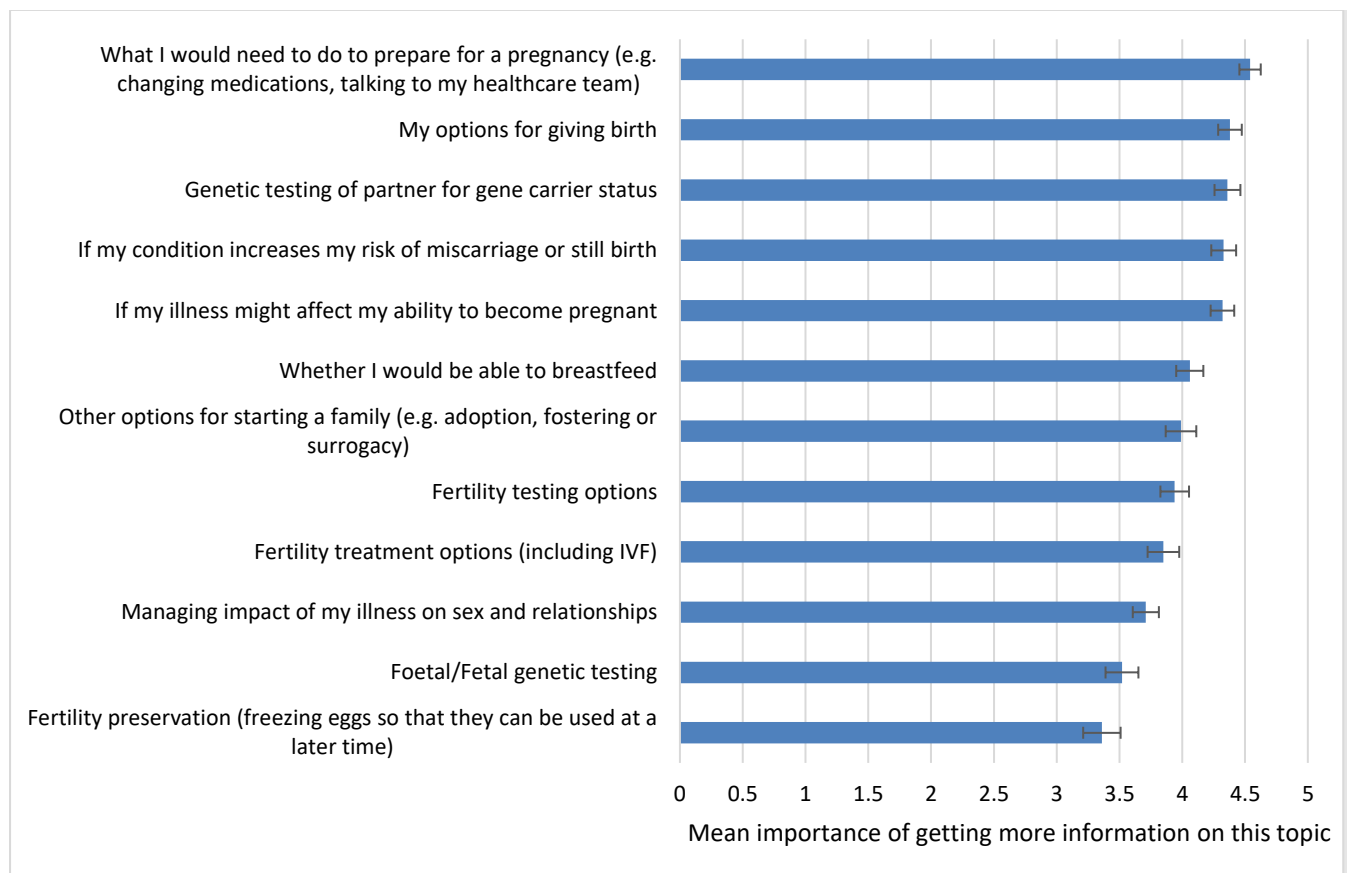
		All (n=182)	Have children already (n=45)	Do not have children (n=137)	95% CI of the difference
Variable	Scoring information Possible range and direction of scores	Mean (SD)	Mean (SD)	Mean (SD)	Lower, Upper
CFQ-R health perception score	<i>0 to 100, higher scores indicate better quality-of-life</i>	68.1 (23.1)	75.8 (23.1)	65.5 (22.6)	2.56, 17.99
CFQ-R treatment burden score	<i>0 to 100, higher scores indicate better quality-of-life</i>	58.5 (21.8)	58.3 (24.3)	58.6 (21.0)	-7.70, 7.13
ENRICHED social support total score	<i>0 to 20, higher scores indicate more social support</i>	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)	<i>0 to 48, higher scores indicate greater information needs</i>	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	<i>0 to 100, higher scores indicate greater self-efficacy</i>	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	<i>0 to 4, higher scores indicate greater consideration of preferences</i>	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	<i>0 to 27, higher scores indicate more SDM</i>	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)	

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 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 Figure 1.

Figure 1: Perceived importance of getting more information on topics relating to reproductive options for women considering having children/more children (n=123)



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.

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

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 20-53 minutes (average 38 minutes). Key themes identified are summarised in Table 4, mapped against the COM-B domains.

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

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.

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 outcomes 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.

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.

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 multidisciplinary approach was important in facilitating pre-conception decisions and supporting women through conception and pregnancy.

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.

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.

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 wide-spread 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 (39). 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 Australia. 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.

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.

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.

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Authorship

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