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ORIGINAL ARTICLE

# Assessment of a novel genetic counselling intervention to inform assisted reproductive technology treatments and other family-building options in adults with cystic fibrosis

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**KEYWORDS:** attitudes, beliefs, cystic fibrosis, infertility, knowledge

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**Abstract** Many patients with cystic fibrosis (CF) are living well into their adult years and contemplating parenthood. Previous studies have shown that there is an opportunity to improve understanding of inheritance and genetics among individuals with CF. This study explored whether a genetic counselling intervention would be associated with a change in knowledge and/or beliefs about genetics and family-building options. Adults (age  $\geq 18$  years) presenting to a CF clinic were approached for inclusion. Participants completed a pre-intervention survey to measure their knowledge of CF genetics, as well as perceptions and understanding of assisted reproductive technology treatments and other family-building options. Subjects then partook in a genetic counselling session. Subjects repeated the survey immediately after the session and 1–3 months later. Data analysis used one-way analysis of variance (ANOVA), repeated measures ANOVA and multiple linear regression. Thirty-five subjects [19 (54%) men and 16 (45%) women] with a mean ( $\pm$ standard deviation) age of  $28 \pm 5.64$  years were enrolled in the study. Before the intervention, 61.69%  $\pm$  4.50 of knowledge-based questions were answered correctly. Immediately after the intervention, the mean score increased to 77.71%  $\pm$  3.23, but this decreased to 69.48%  $\pm$  4.02 for the third test ( $P < 0.05$ , repeated measures ANOVA). Six individuals changed their family-building preference following the genetic counselling session. A short genetic consultation was associated with a significant improvement in CF-specific genetic knowledge. However, knowledge was not retained fully for a longer time period following the consultation. Multiple discussions regarding fertility options are needed to reinforce the key concepts related to CF genetics and fertility.

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## Introduction

Due to medical advances over the last few decades, most patients with cystic fibrosis (CF) are now living well into their adult years (Fair et al., 2000). With the increase in life expectancy, many patients contemplate family-building options as they enter early adulthood. Previous studies have identified fertility and family building as an important, yet underdiscussed, topic for patients with CF and their care team (Sawyer et al., 2005). CF is an autosomal-recessive genetic disorder that has systemic manifestations, including significant effects on the pulmonology, gastrointestinal and reproductive systems caused by mutations in the CF transmembrane conductance regulator (CFTR) gene (Fair et al., 2000). The majority of men with CF are infertile due to congenital bilateral absence of the vas deferens, which can result in obstructive azoospermia. Women with CF may also have reduced fertility, either due to intrinsic defects of mucus secretion affecting tubal function or other indirect complications of their chronic disease (Kazmerski et al., 2017). Large quantities of the CFTR protein are found in the cervix, endometrium and fallopian tubes, which could play a role in the decreased fertility seen in women with CF (Edenborough, 2001). The link between fertility issues and CF was first discovered in the late 1960s when average survival was 12 years. Now, survival into almost the fifth decade, if not longer, is expected (Foundation, 2020), which has altered the significance of fertility in this patient population (Sawyer et al., 2005). Recently, the introduction of CFTR modulator therapies has drastically changed the

quality of life of patients with CF. Patient-reported outcomes that aim to assess health-related quality of life have been shown to improve with CFTR modulators (Balfour-Lynn and King, 2020). The direct impact on fertility has not been assessed to date, given the novelty of the drug (Nichols et al., 2021). Although it is too soon to collect evidence about life expectancy directly, it is possible that CF may not be a life-shortening disease in the future, which will fundamentally change patients' perceptions about parenthood and family-building options (Balfour-Lynn and King, 2020).

With assisted reproductive technology (ART) progressing rapidly, there are many different reproductive treatments that may allow individuals with CF, both men and women, to conceive. Due to advances in ART, men with CF are able to father biological children after microsurgical sperm retrieval via intracytoplasmic sperm injection during in-vitro fertilization (IVF) (Hubert et al., 2006). CF is an autosomal-recessive condition. When both parents are carriers, defined as two individuals who each carry one deleterious allele of the same autosomal-recessive condition, they have a 25% chance of having a child affected with that condition (Myring et al., 2011). However, when a single individual with two deleterious alleles, diagnosed with an autosomal-recessive condition themselves, has a child with an individual who is a carrier of the same condition, the chance of having an affected child increases to 50% (Edenborough et al., 1995). This increased risk can play a large part in reproductive decision-making for adults with CF and their partners.

Previous studies have shown that more discussions about fertility and family-building options are needed (Kazmerski et al., 2017; Sawyer et al., 2005). One study found that 43% of men and 26% of women with CF had not had discussions about fertility with their healthcare providers (Fair et al., 2000). Another study that aimed to develop and validate a questionnaire regarding all aspects of CF disease knowledge found that comprehension was highest for lung- and gastrointestinal-related categories, and was lowest for categories involving reproduction and genetics (Siklosi et al., 2010). Furthermore, studies have found that patients are dissatisfied with the lack of communication around fertility (Kazmerski et al., 2017; Sawyer et al., 2005). Although these studies have demonstrated a lack of knowledge and communication in reproductive and genetic topics in patients with CF, there is limited information about how a patient educational intervention influences knowledge and perceptions about these topics. No studies to date have assessed patient knowledge about ART treatments as they apply to family-building options for patients with CF. This study aimed to measure if and how a genetic counselling intervention may influence knowledge and/or perceptions about genetics, fertility, ART treatments and family-building options among adult patients with CF.

## Materials and methods

This prospective, single-arm, intervention study explored the effects of a genetic counselling intervention on CF-specific genetic knowledge and family-building preference. The Emory Institutional Review Board (IRB) reviewed and approved the study protocol, and each participant provided informed consent prior to study participation (IRB 00102445).

## Recruitment

This study was conducted at the Emory + Childrens' Adult Cystic Fibrosis Center located in Atlanta, GA, USA. Investigators approached both men and women with CF aged  $\geq 18$  years for study participation. Study investigators excluded adults from study participation if they were unable to take the questionnaire independently and/or could not speak or understand English.

## Survey measure

Pre- and postintervention surveys measured the influence of a genetic counselling intervention (see Supplementary Material 1). SK and WH created the survey with feedback from the other authors. Participants received the survey a total of three times. All three surveys were identical except for demographic questions. Participants only had to answer the demographic questions during the first survey distribution. Men and women received identical surveys; however, there were questions specific to pregnancy for women who already had children. Study participants completed the first survey before the intervention, completed the second survey immediately after the genetic counselling intervention, and completed the final survey 1–3 months after

the intervention in order to measure retention of knowledge. This survey had 21 questions regarding the inheritance of CF, impacts of CF on fertility and basic knowledge about ART. Following the questions, the survey asked participants to pick one of four options regarding their family-building preference: wanted children in the future; no intention of having children; already had children and wanted more children in the future; or already had children and did not want more in the future. Based on this question, participants received a specific subset of questions regarding perceptions that related to their family-building preference. Those who already had children received and completed the same subset regardless of whether or not they wanted more children in the future. For those who wanted children, perception questions aimed to measure an individual's likelihood of considering the use of different family-building options, in addition to their feelings and motivations regarding having a family. These family-building options included IVF, pre-implantation genetic testing for monogenic disorders [PGT-M, previously known as pre-implantation genetic diagnosis], donor egg with IVF, donor sperm with intrauterine insemination (IUI), adoption and surrogacy (IVF with gestational carrier). For those who did not want children, perception questions aimed to understand the participant's feelings and/or motivations for why they did not want children. Finally, participants who had children received questions regarding carrier testing, pregnancy complications, and their perceptions about having a child. SK administered the genetic counselling intervention and focused on all topics covered in the survey. Study participants participated in the intervention counselling session after they had completed an appointment with their clinical CF care team. The study intervention took place in either a clinic room or in a separate office space connected to the CF clinic. On average, sessions lasted for approximately 30 min and covered topics such as inheritance of CF, fertility issues seen in patients with CF, and different ART methods in addition to other family-building options such as adoption. Study investigators hosting the counselling session used a basic outline/script to reduce variability of the information given to each patient.

## Data analysis

SPSS (IBM Corp., Armonk, NY, USA) was used for statistical analysis. The study statistician determined whether categorical demographic variables influenced baseline knowledge score utilizing descriptive statistics and one-way analysis of variance (ANOVA). Following one-way ANOVA, analysis with a Tukey post-hoc test determined which specific demographic variables influenced baseline knowledge score. The statistician used repeated measures ANOVA to determine significant differences in knowledge scores and differences in perceptions across all three surveys. Following ANOVA, post-hoc testing using Bonferroni's correction was undertaken to correct for multiple samples at multiple time points. In addition to ANOVA, multiple linear regression accounted for the combination of categorical and continuous demographic variables, and determined if any demographics impacted baseline knowledge. Chi-squared

tests assessed associations between different demographic variables.  $P < 0.05$  was considered to indicate significance.

## Results

This study enrolled 35 participants with CF out of 48 potential subjects approached (72.9%). The most commonly stated reason for deferring study participation was other competing obligations limiting the subject's time in clinic. Table 1 describes the baseline characteristics of the study participants. The 35 subjects (19 men and 16 women) had a mean ( $\pm$ standard deviation) age of  $28 \pm 5.64$  years. Most participants self-identified as being in a committed relationship or engaged/married ( $n = 25$ , 71.5%). The majority reported that they would like children in the future ( $n = 24$ , 68.6%) (Table 1). Furthermore, most participants indicated that they had never discussed fertility options with a provider ( $n = 20$ , 57.1%) and had not seen a genetic counselor previously ( $n = 22$ , 62.9%). Of those who had discussed fertility options with a provider, most had spoken with the pulmonologist who managed their CF.

Before the intervention, the mean score for the survey questions was  $61.7\% \pm 4.5$ . Immediately after the intervention, the mean knowledge score increased to  $77.7\% \pm 3.2$  (Fig. 1). Of the 35 participants enrolled in the study, 22 completed the third survey, which was administered 1–3 months after the intervention (response rate 63%). For the third survey, the mean score was  $69.5\% \pm 4.0$ . In summary, there was a large increase in the percentage of correct answers immediately after the genetic counselling

session, and this decreased 1–3 months after the intervention.

When analysing individual knowledge questions at baseline, most participants gave a correct answer to the question regarding autosomal-recessive inheritance and the chance of two carriers having an affected child ( $n = 30$ , 86%). Most participants answered the question asking about the risk of their future child being a carrier for CF incorrectly (correct answer 100%), with only 31% ( $n = 11$ ) answering correctly. There were two specific questions on the survey that asked about the process of IVF and PGT-M. Just under half of the participants ( $n = 16$ , 47%) answered at least one of these questions incorrectly, and eight individuals (23%) answered both questions incorrectly. Finally, most patients did not know the category of their CF mutation ( $n = 29$ , 83%) prior to the genetic counselling intervention.

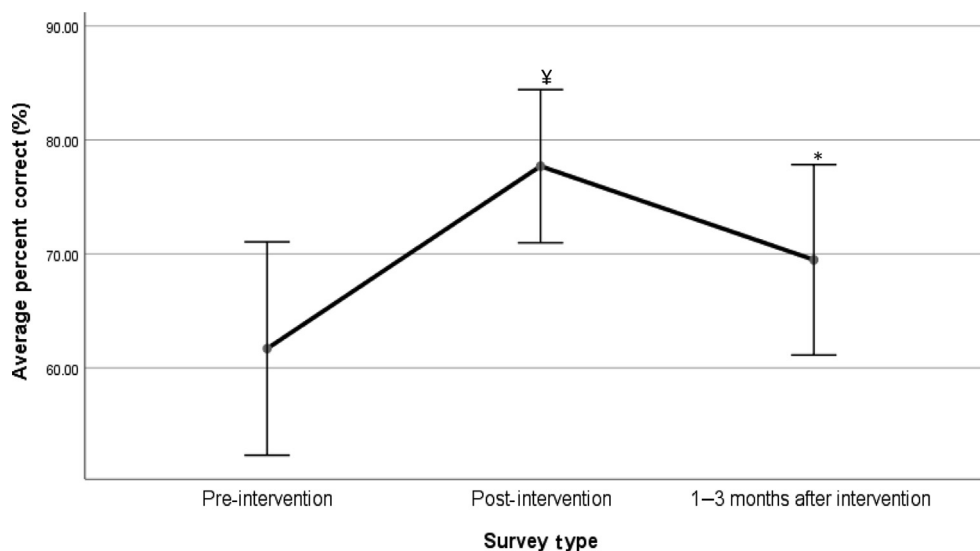
One-way ANOVA determined that there was a difference in baseline knowledge score and demographic groups. There was a significant difference in baseline knowledge score between different marital status groups [ $F(2,32) = 3.43$ ,  $P < 0.05$ ]. A Tukey post-hoc test revealed that participants who were engaged/married had a significantly higher baseline knowledge score than participants who were in a committed relationship ( $14.40 \pm 4.84$  versus  $9.50 \pm 5.19$ ,  $P < 0.05$ ). Further analysis found no other significant differences between marital status groups. There was a significant difference in baseline knowledge score between participants who had different levels of education [ $F(4,29) = 3.92$ ,  $P < 0.05$ ]. Post-hoc analysis revealed that participants who had less than a high school education had a significantly lower baseline knowledge score compared

**Table 1** Participant characteristics ( $n = 35$ ).

Characteristic	Value <sup>a</sup>
Age in years (mean $\pm$ SD)	$28 \pm 5.64$
Gender	
Male	19 (54.3)
Female	16 (45.7)
Ethnic group	
Caucasian	29 (82.9)
African American	2 (5.7)
Other	4 (11.5)
Education	
Graduate degree	3 (8.6)
Bachelor's degree	10 (28.6)
Some college but no degree	11 (31.4)
High school degree	6 (17.1)
Less than high school degree	4 (11.4)
Relationship status	
Engaged/married	15 (42.9)
Committed relationship	10 (28.6)
Single	10 (28.6)
Family-building preference	
Wants children	24 (68.6)
Does not want children	8 (22.9)
Has children	3 (8.6)

SD, standard deviation.

<sup>a</sup>Values are  $n$  (%), unless otherwise stated.



**Fig. 1** Effect of genetic counselling intervention and the passage of time on level of knowledge, as measured by the change in the average percentage of correct answers to survey questions. ¥ $P < 0.05$  compared with pre-intervention score; \* $P < 0.05$  compared with postintervention score. Error bars = 95% confidence interval.

with participants who had a Bachelor's degree ( $6.50 \pm 1.91$  versus  $15.70 \pm 3.34$ ,  $P < 0.05$ ). There were no significant differences in baseline knowledge scores when compared by age, gender, quality of life, family-building preference or previous genetic counselling encounter. Multiple linear regression analysis failed to find any significant predictive value of demographic variables and baseline knowledge score.

Repeated measures ANOVA determined that the mean knowledge score differed significantly between the three survey time points [ $F(2,42) = 12.97$ ,  $P < 0.05$ ]. Post-hoc tests using Bonferroni's correction revealed that there was a significant increase in the percentage of correct answers between baseline score and the score immediately after the genetic counselling intervention ( $61.69 \pm 4.50$  versus  $77.71 \pm 3.23$ ,  $P < 0.01$ ). Furthermore, the percentage of correct answers immediately after the genetic counselling intervention was significantly higher than the percentage of correct answers 1–3 months after the intervention ( $77.71 \pm 3.23$  versus  $69.48 \pm 4.02$ ,  $P < 0.05$ ). There was an increase in the percentage of correct answers between baseline and 1–3 months after the intervention; however, this was not significant.

The survey also assessed participants' confidence in explaining various fertility options to a friend, including the concepts of IVF, IUI, pre-implantation genetic testing (PGT), and donor egg and sperm. The final analysis excluded three individuals due to incomplete answers ( $n = 32$ ; Table 2). Changes in confidence immediately after the genetic counselling session were assessed. The majority indicated that they felt more comfortable explaining IUI and PGT-M following the genetic counselling intervention. Seven individuals reported a decrease in confidence in explaining these fertility options to a friend.

### Participants who wanted children in the future

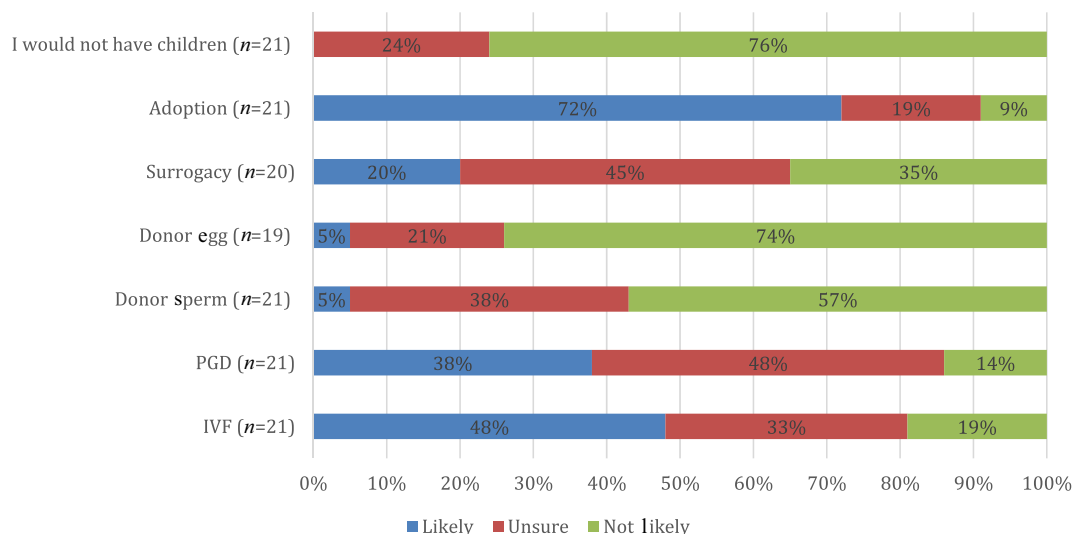
Twenty-four (68.6%) participants reported that they wanted children in the future. When asked about perceptions of their own health and having a child in the future, the majority indicated that they were worried that their health would not allow them to care for a child properly ( $n = 14$ , 58.3%) and that their health would decline when caring for a child ( $n = 14$ , 58.3%). However, 75% of participants believed that they were healthy enough to care for a child at that time. All participants indicated that they were interested in learning about various family-building options. Before the genetic counselling intervention, participants answered Likert-scale-type questions regarding which family-building preference they were likely or unlikely to utilize in the future if they were unable or chose not to have children naturally (Fig. 2). Most participants reported that they were likely to pursue adoption ( $n = 15$ , 72%) as a family-building option and were unlikely to utilize donor egg as an option ( $n = 15$ , 74%). Furthermore, the majority reported that they would not forgo having children altogether, and would pursue at least one of the family-building options described to them in order to have children ( $n = 16$ , 76%). When asked about the factors that were most important to the participant with regard to pursuing different reproductive options, many reported that risk of treatment was the most important factor ( $n = 6$ , 35%) followed by success rate ( $n = 5$ , 26%). The majority of participants ( $n = 12$ , 63%) ranked religious beliefs as the least important factor to them when considering the pursuit of different reproductive options.

Following the genetic counselling session, 15 of 24 (62.5%) participants did not change their preference about which family-building options they would utilize; there

**Table 2** Self-reported change in confidence in explaining fertility treatment options immediately after a genetic counselling intervention ( $n = 32$ ).

Confidence level	Fertility treatment option			
	IVF	IUI	PGT-M	Donor egg/sperm
Increased	14 (43.8)	20 (62.5)	18 (56.3)	14 (43.8)
No change	17 (53.1)	12 (37.5)	19 (31.3)	16 (50.0)
Decreased	1 (3.1)	0 (0)	4 (12.5)	2 (6.3)

IVF, in-vitro fertilization; IUI, intra-uterine implantation; PGT-M, pre-implantation genetic testing for monogenic disorders. Values are the number of individuals reporting each confidence level (%).



**Fig. 2** Family-building preference prior to genetic counselling. PGD, pre-implantation genetic diagnosis; IVF, in-vitro fertilization.

was no significant difference in the frequencies of utilizing these options immediately after the intervention. Overall, nine of 24 (37%) participants did change their preference about which family-building option he/she would utilize after the genetic counselling session. Four participants (19%) who previously indicated that they were unsure about or unlikely to utilize IVF or IVF with PGT-M changed their preference, and reported that they would consider utilizing IVF or IVF with PGT-M. With regard to gamete donation, one individual who was previously unsure about donor sperm changed his/her preference to being unlikely to utilize donor sperm following the genetic counselling session. One other individual who previously indicated that he/she was unsure about utilizing donor egg changed their preference to being likely to utilize donor egg following the genetic counselling session. Fourteen percent of individuals ( $n = 3$ ) were less likely to utilize surrogacy following the genetic counselling session. Finally, there were no changes in preference regarding adoption.

### Participants who did not want children in the future

Eight participants reported that they did not currently have children and did not want any children in the future (23.5%). Likert-scale-type questions assessed this subset of partici-

pants regarding their perceptions of personal health, and how that influenced their choice not to have children in the future. The majority reported that they did not want children because they did not feel healthy enough to raise a child ( $n = 5$ , 62.5%). In addition, half of this subset of participants ( $n = 4$ ) reported that they did not want children because they did not want their child to have CF. None of the participants did not want to have children because of past experiences of being told that they could not have children. Finally, all participants who reported that they did not want children in the future indicated that they did not want to have children due to other personal reasons as well. These perceptions did not change for any of the participants after the genetic counselling session.

### Participants who already had children

Three female participants reported that they already had children. None of these women were actively trying to conceive when they got pregnant. In addition, no fertility treatments or ART treatments were used to conceive. Only one participant had been referred to a genetic counsellor during her pregnancy. Two of three participants had their partner undergo carrier testing for CF. None of the three participants reported any complications during pregnancy, and all of them declined invasive prenatal testing such as amnio-

centesis or chorionic villi sampling. When asked about their perceptions regarding their health and pregnancy, all three women indicated that they felt healthy enough currently to care for a child; however, two of the three participants were worried that their health might decline in the future while caring for a child. These perceptions did not change for any of this subset of participants after the genetic counselling intervention.

## Discussion

To the authors' knowledge, this is the first study to assess how a genetic counselling intervention influences both knowledge and perceptions of ART treatments and other family-building options within the adult CF population. Furthermore, this study was able to capture participants at multiple points in time, and measure retention of knowledge following a genetic counselling session. This study not only reiterates that knowledge levels regarding ART are low, but also shows that genetic counselling could help increase the level of knowledge. Furthermore, the results show that providing more information through a single genetic counselling session can influence patient choice of the family-building options that he/she would utilize. Low knowledge levels could be due to many different factors, such as low awareness of ART or not being able to talk to a provider about fertility options.

With regard to personal perceptions, four of the eight participants who reported that they did not want children stated that they were afraid their child would have CF. Even after the genetic counselling intervention, where it was stated that their future child would only be at risk if his/her partner was a carrier, this perception of fear did not change in three of the four participants who expressed this fear. Further qualitative studies to assess this perception of fear may be needed to better provide for patients.

Consistent with previous data, few participants reported that they had previously had a structured discussion with their healthcare provider regarding the fertility implications of CF or available fertility options (Hubert et al., 2006; Kazmerski et al., 2017). Similar to other studies, the majority of participants in the present study reported that they would like children in the future, and were interested in learning about family-building options (Fair et al., 2000; Kazmerski et al., 2017; Sawyer et al., 2005). There has been a drastic increase in life expectancy for patients with CF over the last three to four decades, leading to a large increase in the number of patients wanting to become parents. However, patients have expressed the desire to start a family since 1990 (Cromer et al., 1990). The introduction of highly effective modular therapy may have a drastic effect on physical health and life expectancy in this population, which will undoubtedly change patients' perceptions about parenthood and family building. However, given the novelty of these therapies, more studies are needed to assess the fertility impacts of modulator therapy, along with mental health and quality-of-life impacts (Balfour-Lynn and King, 2020; Nichols et al., 2021). As more is learned about these novel therapies and how they physically impact fertility, qualitative studies regarding changing perceptions about family building and parenthood are necessary. This will

improve our understanding of patients in the context of this disease, which seems to be changing rapidly.

The study results indicate that a genetic counselling intervention resulted in a significant increase in knowledge immediately after the session. Although there was a trend showing that knowledge retention scores were higher compared with baseline scores, this increase was not found to be significant. These findings suggest that multiple discussions regarding fertility options may be needed to reinforce key concepts and keep patients fully informed as they enter different phases of their adult life and want to explore different family-building options. Furthermore, genetic counselling for this patient population should be tailored not only to the individual's level of knowledge, but also to concepts that are specifically important in CF. Based on the frequency of incorrect answers in this study, this may include counselling about a patient's individual mutations and what their disease status means for their children's carrier status.

Patients with CF view their pulmonologist and the rest of their CF team as their de-facto primary care physicians, and go to them with all of their concerns (Kazmerski et al., 2016). This further highlights the need for these discussions to occur in the multidisciplinary CF clinic. Previous studies have indicated that conversations regarding fertility and reproduction should begin in adolescence (Fair et al., 2000; Sawyer et al., 2005). The timing of these multiple conversations is likely dependent on the individual patient, but individuals should be asked screening questions regarding fertility concerns or family building at intake in order to identify those individuals who may benefit from having a discussion regarding fertility options. Studies have reported that patients are not comfortable bringing up these topics on their own, highlighting the need for providers to not only initiate the conversation, but also to be trained to give the desired information regarding fertility (Fair et al., 2000; Sawyer et al., 2005). Although there was no significant association between increased knowledge and change in the utilization of different family-building options, a few individuals in the study cohort did change their preferences following the genetic counselling session, showing that it did influence their perceptions of family building. It is important to create a system that could potentially identify those individuals who would gain the greatest benefit from genetic counselling.

When comparing the present results regarding utilization of different family-building options with those with other genetic conditions, both similarities and differences were found. Similar to the present results, one study looking at individuals with BRCA1/2 mutations found that women are very interested in learning about pre-implantation technologies such as PGT-M, and that 48% would consider PGT-M and 55% would consider adoption if they were found to test positive for mutations (Chan et al., 2017). Another study looking at reproductive decision-making among people with epilepsy found that concerns about the ability to care for a child and passing epilepsy on to a child were associated with having fewer children (Helbig et al., 2010). Similar feelings regarding health and passing down disease were found in the present study cohort. Similarly, a study looking at reproductive decision-making in those with sickle cell disease or sickle cell trait showed that participants did not want their children or grandchildren to inherit the condition

or trait. Participants in this study also had a low level of knowledge about IVF and PGT-M; however, this study found that most participants would not consider adoption as a family-building option (Gallo et al., 2010). This is the opposite of the findings of the present study, which found that adoption was the family-building option that most participants would utilize if they were unable to have children naturally. There are likely many reasons for this difference, but it is important to note the potentially substantial costs associated with both ART and adoption. These options may not be accessible to each family, greatly impacting their choice of family building. Overall, it is important to consider the different effects that these diseases have on reproduction itself. Of all the conditions talked about, CF is the only one that has direct implications on fertility itself, while the others simply have genetic risk of passing down the condition.

Given that the present study did not find a significant difference in family-building preferences after genetic counselling, an increase in knowledge does not necessarily change an individual's perceptions and utilization of different fertility options. There are likely other factors that play a greater role in these preferences. Cultural factors have been shown to play a major role in a couple's decision to pursue different fertility options given the relative importance of having a child in certain cultural groups (Bos and van Rooij, 2007). In addition, it has been shown that there are substantial differences in the frequency of IVF pregnancies among groups of different socio-economic status (Raisanen et al., 2013). In terms of religious factors, previous data show that religion can play a major role in a patient's decision regarding which family-building option to pursue (Sallam and Sallam, 2016). The present cohort indicated that religious factors would not be a factor in decision making. However, this was a small sample and the participants were not asked in detail about their religious preferences. In addition, other health factors, such as lung transplantation and mental health, were not explored to see how these factors could influence a patient's family-building preference. In all, it is likely a combination of many different factors that influences a patient's preference regarding family building.

This study did have limitations to consider. The participants represent a small convenience sample recruited from one centre. In addition, the knowledge level of patients at Emory + Childrens' Adult Cystic Fibrosis Center may have been higher than average as previous genetic studies had been undertaken using this patient population. Thus, the findings cannot be generalized to all adult patients with CF. In order to direct participants to a specific subset of perception questions, they were asked about their parenting intention in a dichotomous way (i.e. whether they 'did want children' or 'did not want children'). This questioning did not allow respondents to indicate an ambivalent response, which may be a common attitude in this population but is unreported here. Validation of findings among other CF centres and with a larger sample is needed. Further studies should aim to explore ways to retain knowledge among this patient population over time.

In conclusion, genetic counselling is beneficial to this patient population, as evidenced by the immediate increase in knowledge and confidence following the genetic coun-

selling intervention. However, other methods of education or multiple discussions are needed to retain this knowledge. Future studies should aim to explore different methods for knowledge retention, and to explore other factors that may influence a patient's family-building preference through qualitative studies.

## Acknowledgements

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## Appendix A. Supplementary material

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.rbms.2021.05.001>.

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