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Pregnancy as Foreground in Cystic Fibrosis Carrier Testing Decisions in Primary Care

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Cystic fibrosis carrier testing (CFCT) is among the first of the DNA tests offered prenatally in primary care settings. This paper from a descriptive qualitative study describes the influence of pregnancy in CFCT decisions by women receiving community-based prenatal care. Twenty-seven women receiving prenatal care in Midwestern U.S. primary care clinics completed semistructured interviews. Audiotaped interviews were analyzed using content analysis. Participants described decision-making influences and strategies from the perspective of "being pregnant." Patterns of attitudes and beliefs include (1) dealing with emotions, (2) pregnancy is natural, and (3) thinking about the baby. Strategies in the decision-making process included (1) reducing stress, (2) choosing what is relevant, (3) doing everything right, (4) wanting to be prepared, (5) delaying information, and (6) trusting God. While other factors were mentioned by some women, major themes reflect the influence of currently being pregnant on the decision-making process. These findings suggest that pregnancy is a powerful influence on the decision-making process and may not be the optimal time to make fully informed decisions regarding genetic carrier testing. Further understanding of factors influencing the genetic testing decision-making process is needed. Offering CFCT prior to conception is advocated.

Introduction

Cystic fibrosis (CF) is a life-threatening autosomal recessive genetic disease resulting in chronic progressive respiratory involvement, malnutrition, electrolyte abnormalities, and infertility. It is the most common autosomal inherited condition in the population of White European origin, and has varying prevalence across ethnicities. Improved treatment has increased the life expectancy of CF patients from what was historically considered a fatal childhood condition to a present median survival of 36.8 years (Cystic Fibrosis Foundation, 2006). However, CF is still considered a serious, chronic condition without cure that results in considerable mortality and morbidity.

People with CF receive a genetic mutation from each parent (asymptomatic CF carrier), with the two mutations resulting in the CF clinical manifestations. The vast majority of CF mutation carriers are unaware of their mutation status. The most prevalent CF mutation in Caucasians, ΔF508 on the CF transmembrane conductance regulator (*CFTR*) gene, was identified in 1989 (Kerem *et al.*, 1989; Riordan *et al.*, 1989; Rommens *et al.*, 1989), and over 1500 CF mutations have been discovered since then. A screening test panel, the cystic fibrosis carrier testing (CFCT), was developed of the most common CF mutations in the panethnic U.S. population. CFCT clinical usefulness varies, however, as both the CF carrier frequency and the CFCT sensitivity differ according to the ethnicity of the person tested. While the CFCT is not diagnostic of CF in the pregnant woman or partner, and the identification of two CF mutations in a couple is not diagnostic of CF in the developing fetus, the screening test can provide an estimate of the risk that the fetus will receive a mutation on both copies on the *CFTR* gene. Interpretation of CFCT results by primary care providers is complicated by incomplete understanding of phenotypic clinical outcomes associated with genotype combinations. Couple carrier status information can be used by healthcare providers in helping to inform couples during the prenatal period regarding risk assessment for the fetus, prenatal diagnostic testing, and possible implications for pregnancy continuation. Couples can also use this information in preconception planning.

Following the 1997 National Institutes of Health consensus statement on CFCT (National Institutes of Health, 1999), screening implementation guidelines by the American College of Obstetricians and Gynecologists (ACOG) were released in October 2001. Guidelines stated that CFCT should be offered to the following groups: "couples with a family history of CF, partners of individuals with CF, and Caucasian couples of European or Ashkenazi Jewish descent planning a pregnancy or seeking prenatal care." In addition, "information about

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CF screening should be provided to patients in other ethnic and racial groups" with additional counseling and screening available upon patient request (American College of Obstetricians and Gynecologists, 2001b). With the release of the 2001 ACOG guidelines, CFCT began being offered in prenatal primary care settings.

Community prenatal providers have had screening testing experience with the maternal serum alpha fetoprotein (MS-AFP) testing for the detection of neural tube defects and other developmental abnormalities since 1985; CFCT is a newer addition to prenatal screening. Some implementation concerns between MS-AFP and CFCT are similar, but there are differences in the two prenatal screening tests. Differences include who is being tested, whether the test results provide information about the possible health of the fetus or an estimate of likelihood of the fetus' inheriting mutations that could cause disease, and implications for future reproductive planning. Research into CFCT screening practices in primary care has focused on provider knowledge (Kuller *et al.*, 1999; Metcalfe *et al.*, 2002; Vastag, 2003), screening standards and availability (Mennuti *et al.*, 1999; Schwind *et al.*, 1999), and controversies in community practice implementation (Vastag, 2003). However, patient decision making for CFCT screening has received limited focus in the literature.

Informed decision making, such as in genetic testing decisions, is assumed to occur during the process of individuals integrating new knowledge with their personal beliefs (Anderson, 1998). While factors related to prenatal decisions have been identified, little is known regarding how individuals use new genetic knowledge along with personal beliefs to make informed healthcare decisions (Feetham *et al.*, 2002; Etchegary *et al.*, 2008). In a study by van den Berg *et al.* (2006), even when most participants were knowledgeable about prenatal screening (82%), made deliberate decisions (75%), and the decisions were consistent with patient values (82%), only 51% of prenatal screening decisions were described as "informed." Decision-making research into other prenatal testing decisions cites the importance of provider support, prevalence of the general lack of test understanding, balancing the test worry and fetal health, and differences in risk perception as affecting prenatal testing decisions (Kenen *et al.*, 2000). Prenatal amniocentesis testing decisions have been described as "embodied" knowledge, where women transform, interpret, and integrate received information with their experiences, understandings, feelings, and beliefs as the basis for decision making (Lippman, 1999). The influence of experiential knowledge, both as embodied knowledge and empathetic knowledge (subjective knowledge developed from the interactions with others), is described as a contextual influence in the prenatal testing decision-making process (Etchegary *et al.*, 2008). A systematic review of decisions to accept or decline CFCT has identified that most of the common factors to either accept or decline testing are consistent with aspects of the Health Belief Model, including perceived benefits and barriers to testing (Chen and Goodson, 2007). How the decision-making process for CFCT transpires in the primary care setting when the test does not directly indicate fetal health, has no predictive disease implication for the pregnant woman, and for which the pregnant woman may not have any prior knowledge of risk is unclear.

Among persons under going genetic testing, gender differences in risk perception, emotional response, psychological affect, self-perception, and testing convenience have been noted (Fang *et al.*, 1997; Sorenson *et al.*, 1997; Cheuvront *et al.*, 1998; Callanan *et al.*, 1999; Newman *et al.*, 2002). While the length of time needed for carrier decision making is not established, the limited time available for decision making during pregnancy has been identified as problematic for some couples (Honnor *et al.*, 2000). Differences in partner perspective on prenatal decision making may affect the ultimate genetic testing decisions made in prenatal care (Kenen, 2000; Kenen *et al.*, 2000). CFCT requires collection of DNA samples from both the woman and her partner; how a pregnant woman makes the decision and the ways in which the partner participates in this process are not known (Kenen, 2000; Henneman *et al.*, 2002).

The purpose of this study was to describe patterns of decision making for CFCT in pregnant females in the primary care setting. This report focuses on describing how being pregnant influenced consideration of CFCT during genetic testing decision making by women receiving community-based prenatal care.

Materials and Methods

A descriptive qualitative design was used to elicit descriptions of prenatal decision making regarding CFCT. Qualitative descriptive methodology is appropriate when describing a process or phenomenon that has not been widely studied or understood, or to elicit previously undetermined insights (Sandelowski, 2000). The synthesis of qualitative data and derived insights obtained further inform the development of hypotheses, research questions, or theory for further study. Inductive techniques such as analysis of direct observation and interviews with participants are important to identify social and contextual factors in decision behavior and identify modulating variables (Klein *et al.*, 1993; Patel *et al.*, 1996). Qualitative semistructured interviews, an important tool for descriptive and exploratory research, are appropriate when the researcher knows enough about the phenomena to formulate questions, but cannot predict all possible responses (Morse and Richards, 2002). In this descriptive qualitative study, semistructured interviews regarding how the CFCT decision was made, structured questions of demographic information, and researcher field notes of participant interviews were used to identify patterns of how women progress through the process of CFCT decision making during pregnancy. The interview schedule was rehearsed with a doctorally prepared nurse researcher and a pregnant colleague to ascertain the clarity and acceptability of the questions (Fontana and Frey, 2000; Patton, 2002). The study was conducted from May 2005 through early 2006.

Sample

Participants were recruited in collaboration with three Midwestern community-based primary care obstetrics and gynecology (OB/GYN) practices. Pregnant women were eligible for inclusion if they (1) were 14–45 years old and receiving prenatal services at one of the three sample clinics, (2) lived in the bistate metropolitan region or in an adjacent county, (3) were English speaking, (4) had been offered CFCT in the prenatal setting and had made a decision whether or not to accept testing, (5) had not received results of CFCT, and (6) did not have physical, cognitive, or emotional deficits that precluded participation. Thirty-eight women were referred by

the OB/GYN practices for more information about the study, and 27 women ultimately participated. Despite multiple attempts, the researcher was either unable to contact or to arrange interviews with the other 11 potential participants. The study sample consisted of 27 pregnant women; 23 participants had declined CFCT and 4 expressed intent to accept CFCT. None of the women had yet had DNA collection for CFCT at the time of the interview. The women ranged from 19 to 38 years of age and self-reported 11–19 years of education. To provide insights on decision making across economic strata, there was purposive sampling of women from lower socioeconomic status to be in the study. The Federal Poverty Guidelines or a percentage multiple are used as eligibility criteria for several federal programs. For this study, a threshold of 200% of the Federal Poverty level according to family size was used to differentiate participants according to socioeconomic status (SES). For example, for a family size of two, the U.S. Department of Health and Human Services 200% Poverty threshold was 25,660 dollars. Fifteen of the 27 women reported family income for family size above 200% of the Federal Poverty Guidelines for 2005, while 12 of the 27 participants lived in families earning below the 200% Federal Poverty Guidelines. Fifteen of the pregnant women had other children, while this was a first pregnancy for 12 of the participants. Parity was distributed between the SES levels. Table 1 shows characteristics of the sample.

Setting

Three primary care OB/GYN clinics in a metropolitan bistate Midwestern area of about 500,000 people were used to recruit participants for this study. All of the clinic sites were private practices; however, Clinic 2 had a specific mission to offer healthcare services to women regardless of ability to pay. For Clinics 1 and 2, both nurse midwives and obstetricians were primary care providers for the women, while in Clinic 3 all of the women recruited were cared for by the nurse midwife. All the women recruited from Clinic 1 were from the higher SES grouping, while participants recruited from Clinics 2 and 3 were divided between lower and higher SES groupings.

All the clinical sites had been offering CFCT for 1–3 years when data collection was initiated in spring 2005. They each used a couple's model with securing DNA samples by blood sample from both parents, but only tested the father's blood if the mother's DNA indicated a CF mutation. They offered the CFCT during the first prenatal visit; patients could make the CFCT decision at the time the test was offered or inform staff of their decision at the second prenatal visit. All the clinics used the ACOG pamphlet (American College of Obstetricians and Gynecologists, 2001a) or a state-designed pamphlet as the teaching material distributed during the CFCT educational session. For participants who accepted the CFCT, peripheral blood samples were obtained for DNA analysis and transported to the state hygienic laboratory for processing. CFCT results were available to the clinic within 2 weeks.

Procedure

The researcher oriented clinic personnel from each clinic site to assist in study recruitment. Clinics used their usual standards and procedures for providing prenatal care. During

Table 1. Demographic Characteristics of Study Participants

Participant	*SES*	*CFCT decision*	*Age*	*Years of education*	*Religious affiliation*	*Parity*	*Race/ethnicity*	*Marital status*	*Household size*
1	Above	No	29	17	Lutheran	1	Cauc	M	3
2	Above	No	27	19	Catholic	0	Cauc	M	2
3	Above	No	26	13.5	Catholic	0	Cauc	M	2
4	Above	No	28	16	Christian	1	Cauc	M	3
5	Above	No	23	17	Catholic	0	Cauc	M	2
6	Below	Yes	24	11	Catholic	0	Cauc	S	2
7	Above	No	28	17.5	Catholic	1	Cauc	M	3
8	Below	Yes	25	12	Baptist	2	Cauc/Native Am	S	3
9	Below	No	38	12.5	None	3	African-American	S	4
10	Above	Yes	23	14	Christian	0	Cauc	M	2
11	Below	No	28	12	None	2	Cauc/Native Am	S	3
12	Above	Yes	23	12	Lutheran	2	Cauc	M	4
13	Above	No	23	16	Lutheran	0	Cauc	M	2
14	Below	No	22	14	None	0	Cauc	S	3
15	Above	No	28	16	Catholic	0	Cauc	S	2
16	Below	No	36	12	None	4	Cauc	M	6
17	Above	No	22	14	Christian	2	Cauc	S	4
18	Above	No	31	16	Catholic	3	Cauc	M	5
19	Below	No	27	12	Catholic	1	Cauc	M	3
20	Below	No	19	13	None	1	Cauc	S	4
21	Below	No	27	14	Pentecostal	0	Cauc	S	3
22	Below	No	19	12	Christian	0	Cauc	M	2
23	Above	No	22	13	None	1	Cauc	M	3
24	Above	No	26	16	Catholic	1	Cauc	M	3
25	Below	No	20	12.5	None	0	Cauc	S	1
26	Above	No	27	16	Methodist	0	Cauc	M	2
27	Below	No	24	12	Christian	2	Cauc	M	4

the course of their regular prenatal care, the women are offered the opportunity to have CFCT and make the decision whether or not to accept testing. After the CFCT decision was made, clinic personnel introduced the opportunity to be a participant in this study. For interested potential participants, the researcher contacted the woman, further explained the study, and made arrangements to obtain consent and collect data. Data collection was through single interviews of the pregnant women regarding how the woman progressed through the CFCT decision process. Patient-identified influences, how the pregnant woman considered the option of CFCT to make her decision, and recommendations for healthcare providers were obtained through a semistructured interviews conducted at the woman's home, or a location of her choice. Participant verification of interview content was assured by having the interviewer summarize the interview with the participant and verify or clarify content at the end of the interview. This helped ensure data accuracy, enabled clarifications, and provided an opportunity for participant-added observations (Seng *et al.*, 2003). Sampling continued until there was redundancy in the interview data.

Data analysis

Demographic questions describing the participants were analyzed through descriptive statistics. Interviews were audiotaped and transcribed verbatim, and field notes were recorded. Interview transcripts and field notes data were analyzed through content analysis methods (Knafl and Webster, 1988; Sandelowski, 2000). Both within and across case analysis techniques of the interviews to identify patterns in the data were employed (Ayres *et al.*, 2003). Data analysis started concurrent with data collection and continued throughout data collection. Interview transcripts were read numerous times by the primary investigator and a doctorally prepared nurse researcher. Data were sorted into emergent categories and patterns or associations among specific personal, demographic, or contextual variables and experiences with CFCT were sought throughout the data analysis process. A codebook of definitions was maintained and updated regularly to enable consistency in identification of themes. These definitions were refined, and redundant or irrelevant codes eliminated with ongoing analysis and data interpretation. A sampling of interviews was independently coded by a doctorally prepared nurse researcher, and regular meetings to discuss coding, emerging themes, and data interpretation were held. To increase trustworthiness of the analysis, categories or themes identified from the data were reviewed by the researchers for clarity and consistency with the interview transcript data, the participant check summary at the end of the interviews, and the field notes (Morse and Field, 1995; Holloway and Wheeler, 2002). An audit trail of all decisions regarding data gathering and analysis were also maintained as a quality assurance mechanism. NVivo 7 (QSR, 2006) qualitative computer software was used for data management.

Results

The central theme emerging from the data was "The influence of pregnancy as being at the foreground of the decision" whether or not to accept genetic carrier testing for CF. This was present throughout their descriptions of their decisions. For women offered CFCT during the early stages of their pregnancy, it is the pregnancy that functioned as the "perceptional lens" through which the woman's decision making was viewed. Using this lens, three patterns of attitudes and beliefs about CFCT in pregnancy and six management strategies were identified. The three patterns of attitudes and beliefs were (1) dealing with emotions, (2) pregnancy is natural, and (3) thinking about the baby. The six management strategies used by the women included (1) reducing stress, (2) choosing what is relevant, (3) doing everything right, (4) wanting to be prepared, (5) delaying information, and (6) trusting God. Exemplars are shared and subcategories delineated for each identified pattern. These themes are listed in Table 2.

Attitudes and beliefs about CFCT in pregnancy

Dealing with emotions. Overwhelmed by decision making: Eight participants noted feeling overwhelmed by the amount of information and number of decisions they were expected to make. "It's just a crazy time because it was my first pregnancy and everything and a lot of stuff to think about." One participant who researched pregnancy complications explained, "Oh my goodness this is overwhelming. I didn't realize that they could go in and do all of this stuff."

Anxious about testing: The prospect of CFCT and considering the possibility of CF made some women anxious or fearful. Anxiety associated with considering prenatal testing and possible subsequent testing or decisions made some not want to think about testing, "[additional testing and possible decisions about the pregnancy] made me very uncomfortable and I didn't even want to continue thinking about it." Another described the stress associated with testing and not wanting to worry about these issues during pregnancy, "You're stressed out, you're thinking about enough stuff while you're pregnant. . . . I don't think I should be worried about [CFCT]."

Table 2. Themes of CFCT Decision Making by Pregnant Women

Central theme: pregnancy as foreground

I. Attitudes and beliefs about CFCT in pregnancy-
- Dealing with emotions
  - Overwhelmed by decision making
  - Anxious about testing
  - Excited to be pregnant
- Pregnancy is natural
  - Acceptable routine care in pregnancy
  - Prospect of follow-up testing post-CFCT
  - Differentiating between prenatal tests
- Thinking about the baby
  - Protecting the baby
  - Treatment *in utero*
  - Valuing life
  - Forming parental attachments
  - Making sure the baby's healthy

II. Self-management pregnancy strategies used by pregnant women in decision making
- Reducing stress
- Choosing what is relevant
- Doing everything right
- Wanting to be prepared
- Delaying information
- Trusting God

For two women, the fear associated with pregnancy and possible adverse outcomes of pregnancy prompted accepting CFCT, "I think I took every test that was available. . . . I just really scared of anything—if there was any test to find out anything I did it." One woman was "scared" and wanted as much information as she could about possible problems with the baby's health, "I don't know how to deal with a sick kid."

Excited to be pregnant: Excitement about the pregnancy was an emotion that affected how three participants considered CFCT. For these women, a history of past pregnancy loss, infertility, or a current pregnancy with twins heightened anticipation of childbirth. They described not being ready to consider health issues with the developing child or risk testing that could result in a spontaneous or therapeutic abortion. Participants described being "excited about being pregnant and ecstatic . . . nothing was going to keep me from having this baby."

Pregnancy is natural. Routine care in pregnancy: Ten women discussed their childbearing philosophy as defining acceptable medical interventions during pregnancy. These women envisioned most prenatal testing, both to identify birth defects, as in MS-AFP screening, and genetic testing, such as CFCT, as "extra tests" that were not needed. They were only interested in basic medical management testing, such as routine blood work, urine testing, and ultrasounds to identify the baby's sex. Some expressed a desire to keep the pregnancy interventions simple and natural, "I believe that pregnancy and birth are a natural process, and the least amount of medical intervention possible is what I choose to go with." Some felt that less testing was less stressful both physically and emotionally during the pregnancy, "I just guess that I feel that the least amount of testing to do to the baby before it is born can actually result in a safer pregnancy." In contrast, women who accepted CFCT had more positive attitudes about the beneficial role of prenatal testing. "Yeah, there's only benefits to it, you know, there are really no consequences to taking a test. . . . It can't make it worse for cystic fibrosis, it can only make it better."

Prospect of follow-up testing post-CFCT: More invasive than the peripheral venipuncture blood test used for CFCT is a follow-up amniocentesis if the couple CFCT results are positive. The topic of amniocentesis was raised by women in nine of the interviews. For women who expressed the philosophy of keeping pregnancy management "natural" and who had difficulty conceiving or carrying a pregnancy, the option of an amniocentesis was not acceptable due to the perception of possible risk of miscarriage. Taking that chance was described as "totally off limits." Others described negative stories they had heard about the amniocentesis test itself or poor outcomes from the procedure, "I have heard more bad stories about amniocentesis than good stories." Some women confused the accuracy of an amniocentesis that would examine the fetus' DNA with the possibility of false-positive results in MS-AFP testing, "they said sometimes they have them come back false as positive, and that's one of the things that was kind of a concern. I wouldn't have wanted to go through the test and have it falsely come back positive." This confusion regarding test accuracy made it less likely women would decide to have CFCT.

Two women thought that an amniocentesis was the first test done for CFCT; one had agreed to the test, while the other chose not to have CFCT because she thought that it initially involved an amniocentesis rather than a peripheral blood draw. Only a few participants cited concerns about the amniocentesis as a major consideration in the CFCT decision. Most participants had not considered the possibility of an amniocentesis, were ambivalent, or thought that if they were in a situation where they needed to make that decision, would probably choose not to have the test.

Differentiating between prenatal tests: Participants who wanted to minimize prenatal testing did not differentiate between the MS-AFP testing and the CFCT in their decision making. They either indicated that they were not interested in "extra" testing during the pregnancy or added that "none of the testing was needed unless there was something that could be done [treatment of the condition *in utero*]." The four women who accepted CFCT also accepted MS-AFP testing, seeing the extra tests as not an additional burden, "I just signed the paper for it and stuff like that. I had to sign a paper for all the other tests too. I guess this was just one more kinda added in there."

Some women did see a difference in these two types of prenatal testing, however. Of the eight women who accepted MS-AFP testing, only half also opted for CFCT. For a few, it was a difference in how much the test was a part of routine medical care during pregnancy, "MS-AFP, it's kind of a standard test I think anymore . . . cystic fibrosis I felt was like, more optional."

Thinking about the baby. Attitudes regarding the developing fetus influenced how the participant thought about the CFCT decision. The pregnant women referred to the developing pregnancy as a "baby" rather than a "fetus." Protection of the baby, the ability to enhance the baby's health through treatment *in utero*, philosophy on the value of life, and attachment to the baby regardless of the child's health were the main considerations.

Protecting the baby: Participants who had considered amniocentesis tended to be less favorable toward this procedure if they interpreted this intervention as a risk to the child. "That's [amniocentesis procedure] more risky than blood." Another shared, "We both realized that there was a risk to the baby for doing any type of prenatal testing. Um, it was kind of an instant decision that we would not do anything to harm our child."

Treatment *in utero*: Whether the condition identified during pregnancy could be treated *in utero* was a common consideration. "I would like them to [test for CF] because sometimes you can fix stuff before the baby's even born." Most participants who verbalized considering whether the test could lead to treatment during pregnancy understood the CF was not treatable during pregnancy, and differentiated their choices on testing with that in mind. "A lot of times you can catch things in ultrasounds, like heart problems, so that was important that I had an ultrasound, but I know it's not good to have more than the needed ultrasound." Another reasoned, "There's not anything therapeutic that can be done [*in utero*] . . . to where you were going to take special precautions, or anything." Some indicated knowing the information, without recourse for action, would also increase the worry during pregnancy, "If it's not something that can be fixed, then there is no reason to have a test done and to worry about it until the baby's born."

Valuing life: Seven participants spoke about their views of the value of life, while four indicated that the quality of life was a consideration in deciding about testing. They shared

that basic feelings of how they viewed life was a "mindset" that was maintained while considering prenatal testing options. These women commonly viewed that they would "accept the child with, with or without any disabilities." They talked about the child not being at fault, and instances where working with people who had a disability or chronic illness convinced them these persons' lives had value and should not be dismissed. Three women deliberated about quality of life, whether the condition was more of a mental impairment, or a pulmonary ailment, "With AFP I felt the child could be severely disabled and you would be finding something out about that, but with cystic fibrosis, the disease that affects the lungs . . . I felt like it could be something that could be managed." There was also the question of parental responsibility toward having children who could have a "normal life." "You would like to say I would love that child regardless, and accept them . . . , but at the same time you would be thinking 'I want to give my child the most normal life they could have.' " These considerations were not commonly discussed, and emerged with a highly educated subset of participants.

Forming parental attachments: Women shared attitudes of attachment to the fetus, and anticipation of loving and caring for the baby regardless of the baby's health. They identified that attachment to the developing child was a consideration, "We're still gonna love it, we're still gonna take care of it. . . . We both believe that no matter what the baby has, we're still gonna have the baby." This feeling of attachment influenced perceptions of viable decision options, "Mostly with me it wasn't really an option. I figured either way I'm gonna to love my child, so there was no reason to worry about whether or not she was going to have cystic fibrosis." Women also minimized other prenatal testing if the results were not going to make a difference in how the parents felt about the child or their decision whether to continue the pregnancy. "No, just the basic [tests during pregnancy] . . . no matter what the baby has we are going to love it and keep it. So we really didn't test anything because it didn't matter to us really."

Making sure the baby's healthy: Some women equated CFCT with making sure the baby was healthy, or helping to ensure the baby's health. They did not focus on whether the CF could be treated *in utero* or future decision making regarding the pregnancy. "I thought it was like something that they take a test to make sure your baby's healthy. That's the first thing that pops in my mind. Anything that's gonna check on the baby, I'm in for." A second participant said, "They just asked me, and I want my baby to be healthy, so I did, I said 'yes,' " while another noted, "It's my child, and if there are steps I can take to prevent it, or if I know about it ahead of time, or any test that's given to me as an opportunity, I might as well take it."

This attitude of being likely to agree to more testing during pregnancy as a positive benefit of medical management was in sharp contrast to the attitude of minimizing medical care during pregnancy and viewing pregnancy as a natural process.

Women's strategies for decision making during pregnancy

Consistent with the beliefs and attitudes professed related to pregnancy, the women described strategies for decision making. Women used these pregnancy management strategies to guide their day-to-day pregnancy decisions and the CFCT decision, and for coping with the potential of having a child with CF.

Reducing stress. Stress reduction was a strategy employed by almost half of the participants. Most women interested in reducing stress chose not to accept CFCT, as the worry associated with prenatal testing and a possible false result was something to be avoided. "I'm such a worrier, that I would probably have worried the rest of my pregnancy about it, and probably created more health problems." Concerns raised during pregnancy were stressful for some pregnant women; not knowing CFCT results served as a strategy to prevent additional anxiety, "[If] they have concerns about anything, it's just gonna raise that level of anxiety and stress . . . but I don't know, I just knew that I didn't, I didn't want to know." The cumulative load of life stresses in addition to prenatal testing was evident by this participant's comment, "Well I think by having these tests . . . I would be very stressed out about it, and thinking about it all the time. I have enough stress at work and everything else, that I don't need that, so . . .."

Choosing what is relevant. Whether the woman saw CFCT as relevant to her pregnancy management was an important consideration. Eighteen of the 27 participants discussed how they asked themselves whether it would make a difference if they had CFCT. For these women, they universally decided that it was not relevant to the pregnancy management. These women chose a strategy to have testing that would enhance their own health or that of their unborn child, but not to have testing they deemed irrelevant in their situation. "What mainly influenced my decision would be we didn't think it would change our mind on the pregnancy, we didn't think it would be of any benefit in knowing whether our child had cystic fibrosis or not."

Doing everything right. A few women talked about using a strategy for prenatal care that included accepting provider-offered testing, with the motivation of doing everything right. Even for some participants who declined CFCT, they spoke of having more difficulty with the decision with a first pregnancy, feeling scared, and not wanting to make any mistakes. One woman who accepted CFCT with a prior pregnancy indicated, "The only reason I did it was because she was my first child and I was scared of everything . . . I wanted to know. I think I took every test that was available." The least educated participant had a more global view of accepting testing, "They asked me about it . . . if I was interested in it, and I said 'Sure, yeah, I'm interested in it'—I do all their tests, I do everything there."

Wanting to be prepared. Some women identified strategies for coping with the potential risk of having a child affected by CF, based on the principle of wanting to be prepared. Two women who accepted testing indicated that "being prepared" was an important part of their decision making. "So I wanna know, I just want to know what kind of thing I'm dealing with," and "it's not going to affect anything, you know? Except to mentally prepare myself." Others who declined CFCT considered whether that knowledge would have made them more prepared in coping with a child with

CF. They could understand some wanting that information, but did not feel this consideration was important enough to decide the issue for themselves. One woman with a family history of CF stated, "It's more of a curiosity type thing. Or to know if we were both carriers—to know that risk, I would probably have wanted him to get some sort of education on cystic fibrosis." Another noted, "Maybe I could be a little bit more prepared . . . read up about it before the baby was born."

Delaying information. Commonly, women coped with the risk for CF by delaying information until after the baby is born. "I think, just for me, I'd rather find out after the baby's here." One participant indicated delaying this information till after birth served to avoid a predelivery mindset about the child and potential problems with bonding. She explained,

"I know if I had done the testing and it had said it was cystic fibrosis, that's what would be the first thing on my mind. . . . How do I deal with the cystic fibrosis and like the personality of the baby separately? Because I think when you see your baby you are not going to think, 'What is wrong with this baby?' You're just going to go, 'Oh my gosh, my baby's here and I love him . . .' "

Trusting God. Twenty of the participants identified a religious preference when asked, while five described their faith-based beliefs as providing an intrinsic backdrop for their CFCT decision. They expressed a willingness to accept whatever God gave them, and to trust that the outcome would not be more than they could handle. "Your children are the way they are for a reason . . . that's God's plan for you, and we were willing to accept our child no matter what."

Several women indicated that trusting the outcome to a higher spiritual being helped them be more relaxed and feel less stressed about the pregnancy.

"Sometimes I feel like it [prenatal testing] just stresses you out more to have to go through all of this, because, you know, it's never 100%. . . . So, I think before we even got pregnant, we knew that this was something that we wouldn't do, we just kind of thought 'we'll get pregnant, and just let God do it.' Whatever He wants."

Discussion

Pregnancy is an assumed condition for decisions made in the prenatal period, including medication use and testing decisions. For most prenatal testing decisions, the opportunity to choose the test only presents itself because the woman is pregnant. For CFCT, this is not the case. A CFCT can occur at any time during the lifespan. Recommendations for offering CFCT to pregnant couples were designed to facilitate informed and autonomous decision making regarding reproductive planning and the risk of the fetus being affected by CF. However, the state of pregnancy itself may increase the challenges for making CFCT decisions.

Pregnancy is a time of vulnerability for the pregnant woman, in terms of both emotional variability and as a developmental task. It has been described as a transition into the unknown (Lundgren and Wahlberg, 1999), a crisis (Raphael-Leff, 1991), and a part of a woman's transformation to motherhood (Bergum, 1997). Pregnant women are also viewed as vulnerable according to the Belmont Report and protection of human subjects for research (U.S. National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research, 1979). In addition, biological changes (hormonal factors) during the prenatal period increase the pregnant woman's vulnerability to anxiety-related symptoms and mood changes (Ross *et al.*, 2004; Ross and McLean, 2006). Research evidence suggests a relationship between emotional disturbances during pregnancy and the emotional development of the fetus (Salisbury *et al.*, 2005). While it is recognized that the prevalence of anxiety in the prenatal women is higher than the general population, implications of maternal anxiety on the woman's quality of life and decision making are not well described.

Genetic carrier testing literature notes that studies generally do not address data on emotional factors that may influence the decision-making process (Lerman *et al.*, 2002). However, affective states are recognized as influential in decision making, particularly as they relate to fear arousal, uncertainty, and predecision processing (O'Hair *et al.*, 1996; Brownstein, 2003; Crano and Prislin, 2006; Peters *et al.*, 2006). In this study, participants described the affective states such as excitement and anxiety that may have affected the processing of prenatal decisions. Women in this study did not deliberate this decision at length, making the CFCT decision quickly. This is consistent with a van den Berg *et al.* (2006) study that many prenatal screening decisions were not deliberated, and often were not informed decisions. Many women noted approaching the CFCT decision in a way that would minimize their stress level and their feelings of anxiety during pregnancy. Some indicated that increasing their stress through considering and choosing CFCT would be both difficult for them to cope with during this time and potentially harmful for the baby. However, for one participant who identified multiple life stresses, the CFCT represented a way to decrease stress of not knowing if she was carrying a CF-affected child. For women at particular risk due to social circumstances, substance abuse, or mental health issues, the possible impact of CFCT options during pregnancy is largely unknown. Both the belief systems related to pregnancy and wanting to manage their pregnancy in a way that decreased their physical and psychological vulnerability were important considerations for the women in this study. The underlying belief systems, heightened vulnerability, and personal stress management strategies identified were all related to the participants making the CFCT decision during pregnancy.

CFCT (and other prenatal testing) is offered early in pregnancy, when the woman is establishing her own maternal identity role and attachment to the fetus. The process of becoming a mother is described as a dynamic psychosocial development process throughout the pregnancy and postpartum period (Mercer, 2004). Commitment to the pregnancy and attachment to the fetus are key components of the initial stages in becoming competent in the maternal role. Anxiety is noted as having a negative effect on the attachment. The women in this study spoke of feeling attached to their baby and committed to the pregnancy. Likewise, they made an effort to avoid situations that they felt would increase feelings of anxiety. While the possible influence of genetic testing in pregnancy and maternal attachment is not clear, one study of the pregnancy experience where the unborn child had been diagnosed with a nonlethal congenital abnormality illustrated that prospective mothers were able to develop a maternal attachment to the child *in utero* (Hedrick, 2004).

CF carrier mutation status does not need to be determined during pregnancy, but can be assessed prior to conception. While the relative value and logistics of carrier screening prior to conception versus prenatally have been debated internationally (Wildhagen *et al.*, 1998; Wille *et al.*, 2004; Mennuti, 2008), traditionally, there has been less emphasis on the healthcare needs of the "potentially expectant mother" prior to conception. However, the option of preconceptional CFCT when a couple requests the test was endorsed by the majority of respondents in a survey of Dutch general practitioners, gynecologists, and pediatricians (Baars *et al.*, 2004). The preference of nonpregnant women for preconception fragile X carrier testing was documented by researchers who are developing a model for offering carrier screening for fragile X syndrome in primary care practices in Australia (Metcalfe *et al.*, 2008).

Over the past 25 years, guidelines and standards of preconception care in the United States have evolved from an initial acknowledgement of the role of prevention prior to pregnancy in 1979, to incorporating preconception care and counseling in Healthy People 2000, and including genetic counseling and genetic testing in the current ACOG guidelines (Freda *et al.*, 2006). The Centers for Disease Control and Prevention (CDC) and 35 partner organizations convened a national summit on preconception care in 2005, and released policy recommendations in 2006 (Posner *et al.*, 2006). Identification of carrier status for genetic conditions (including CF) that could be assessed prior to pregnancy is advocated in the recently released policies (Atrash *et al.*, 2006; Posner *et al.*, 2006; Shapira and Dolan, 2006). In addition, updated 2005 ACOG recommendations for CFCT placed more emphasis on offering CFCT to all couples, rather than focusing primarily on couples planning pregnancy or during pregnancy (Committee on Genetics ACOG, 2005). Offering genetic screening and associated patient education for carrier status of autosomal recessive disorders prior to pregnancy may promote a less complex and more deliberative decision making of genetic testing decisions. This may also have implications for healthcare provider education and clinical care within the limitations of the primary care environment. Recommendations emphasize the need for comprehensive planning (Payne *et al.*, 1997), ongoing monitoring of long-term psychological and social effects of screening (Henneman *et al.*, 2002), and the importance of pre- and posttest genetic counseling (Wildhagen *et al.*, 1998; Ensenauer *et al.*, 2005). How the needed logistical, educational, and counseling resources will be coordinated to support the primary care health system both in the United States and internationally is yet to be defined.

Also identified in the recommendations for preconception care in the United States are the financial limitations of limited insurance and third-party payer coverage to fund preconception care, as well as the diminished awareness of many women and some practitioners on the importance of these interventional strategies. Funding initiatives to facilitate preconception care and increase public and provider awareness of the importance of preconception care may promote positive health outcomes and consideration of genetic testing outside of pregnancy. To support informed and deliberative decision making, CFCT implementation may best focus on the preconception period as an initial screening strategy, with prenatal testing for CF carrier status becoming a secondary screening strategy. Decision making after a positive prenatal diagnosis following amniocentesis for Down syndrome has been described as a paradoxical lack of choice, or a travesty of choosing, where couples are forced to contemplate choosing against a wanted pregnancy (Sandelowski and Barroso, 2005). In the case of CF, the possible dilemmas encountered after positive prenatal diagnosis are avoided by focusing screening efforts outside of pregnancy. Appropriate policies, fiscal support, and alterations in provider practice will be needed for successful implementation.

Research findings must be viewed within several limitations. Because participants were recruited from community care settings and clinic personnel introduced the study, it is undetermined whether the study was offered to eligible participants systematically. It is unknown if the 11 women who indicated initial interest in the study but were either unable to be contacted or declined study participation differed from the women who agreed to consent to the study. There were no teens recruited for this study, and there was little variation in race/ethnicity among the participants. The decision-making insights provided were predominantly from women who did not choose to have CFCT, so may not adequately illustrate decision making for women who accept CFCT. Also, standardized instruments on concepts of interest such as personality and coping styles were not included. While interview data suggest that a few of the women seemed to agree with all testing suggested by their healthcare provider, most demonstrated using information management styles that decreased anxiety, were consistent with their beliefs about pregnancy and pregnancy management, and eliminated the potential for further prenatal testing decisions.

Other limitations are reflective of the design and sample in that the study takes place in one geographic area, with a small sample. The nature of qualitative research limits generalizability of the results to a larger population. It does, however, identify nuances of the dynamics of genetic carrier decision making within the context of primary care that may not have been anticipated through a quantitative design.

This sample of women from both rural and metropolitan settings, a range of patient circumstances regarding marital status, socioeconomic status, number of children, and experience with making prenatal decisions served to illustrate the complexity of the decision-making phenomenon under study. This further underscores that despite the demographic and experiential differences, there was commonality in the general approach to the CFCT decision-making process.

In this study, the importance of pregnancy emerged as the perceptual lens through which women described their decision-making process. Emotions, beliefs, attitudes, and strategies for these women also did not exist outside of the overlying reality, or perceptional lens, of being in a state of pregnancy. Findings from this study support examination of mechanisms by which CFCT can be incorporated into preconception care for couples of reproductive age. When CFCT is offered during pregnancy, it may be one of many decisions the pregnant woman is asked to make. Because some women may have difficulty differentiating the purpose, process, and meaning of different screening tests, beginning this discussion prior to pregnancy may help those women make an informed decision. If offered during pregnancy, not offering CFCT at the same clinical visit as other prenatal testing (such as MS-AFP) may also decrease the confusion between testing options and implications for testing. Regardless of the timing of offering CFCT, discussions of this option may be most useful

when they include the opportunity to weigh the decision within the context of the woman's or couple's attitudes and beliefs. Understanding how patients make genetic testing decisions is important for appropriate implementation of genetic screening initiatives and to enable enhanced provider–patient collaboration in complex clinical decision making (Pletcher *et al.*, 2008). This understanding can avoid misdirected program development, unwanted genetic testing, or missed opportunities for patients to find out information they may consider valuable for reproductive decision making. As CFCT is one of the first genetic tests offered in primary care, insights from patient perspectives in this situation have implications for future genetic testing not only in prenatal settings, but also throughout clinical practice.

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