WOMEN'S HEALTH: RESEARCH ON GENDER, BEHAVIOR, AND POLICY, 3(1), 31–51

Attitudes Toward Genetic Carrier Screening for Cystic Fibrosis Among Pregnant Women: The Role of Health Beliefs and Avoidant Coping Style

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In this study we examined the relations among psychosocial factors associated with pregnant women's attitudes toward genetic carrier testing for cystic fibrosis (CF). A sample of 511 pregnant women attending various health clinics for general prenatal

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care were educated about CF. Women's health beliefs, coping styles, and attitudes toward CF carrier screening were assessed. Results from structural equation modeling analyses indicated that women who perceived themselves as more likely to be carriers of the CF gene and who perceived greater benefits of screening were positively inclined toward genetic screening. Perceived barriers to screening were negatively associated with women's attitudes toward CF genetic testing. In addition, the findings suggest that some types of avoidant coping styles may indirectly influence one's decision to undergo genetic screening through perceptions of risk, benefits, and barriers. Given the advent of genetic screening options for many diseases, in this study we address some issues in women's attitudes toward prenatal screening that are relevant to a variety of genetic screening programs.

Key words: cystic fibrosis (CF), genetic carrier screening, health beliefs, avoidant coping

Cystic fibrosis (CF) afflicts approximately 30,000 people in the United States and is the most common recessive genetic disorder among Whites. Individuals with CF suffer from the production of abnormally viscous secretions in their lungs and digestive system, which may lead to frequent respiratory infections and pneumonia, persistent coughing and wheezing, and poor weight gain. These symptoms become progressively worse as the afflicted individual grows older and often lead to an early death. Although there are medical treatments to help control the symptoms of CF, there is no cure, and it is estimated that only 50% of individuals with CF live beyond the age of 25 (Elias, Annas, & Simpson, 1991; Office of Technology Assessment Report Brief, 1992; Rowley, Loader, Levenkron, & Phelps, 1993).

Because CF is an autosomal recessive disorder, individuals who carry one CF gene (i.e., a CF carrier) will never develop the disease and will not exhibit any symptoms of CF. However, if two CF carriers have children together, there is a 25% chance that each child will be born with CF (have both CF genes), a 50% chance that each child will be a CF carrier, and a 25% chance that each child will not be a CF carrier or have the disease. It is estimated that 1 in 25 White individuals of Northern European descent are carriers of the CF gene (Botkin & Alemagno, 1992; Elias et al., 1991; Office of Technology Assessment [OTA] Report Brief, 1992). Carrier rates for other ethnic groups vary from 1 in 40 Hispanics, 1 in 65 Blacks, to 1 in 150 Asians (OTA Report Brief, 1992). It is estimated that as many as 12 million Americans may be CF carriers. Each year in the United States, approximately 1 in 2,500 babies is born with CF (Rowley et al., 1993).

The identification of the CF gene on chromosome 7 led to the development of simple DNA tests that can determine whether or not an individual is a CF carrier (Elborn, 1991; Elias et al., 1991). In one version of this test, DNA from cells collected by the gentle brushing of the inside of one's mouth (buccal mucosa) can be analyzed for various mutations of the CF gene. Testing for the six most common

mutations will identify between 50% and 92% of CF carriers, depending on their ethnic background (see Shuber, Skoletsky, Stern, & Handelin, 1993). Given that the majority of the children born with CF have parents with no family history of CF, the information received from a genetic screening test may allow couples to make more informed decisions about family planning.

The development of these tests has introduced the possibility of widespread population screening for CF carrier status. In many cases, the point of entry for screening has been in prenatal clinics (Asch, Patton, Hershey, & Mennuti, 1993; Botkin & Alemagno, 1992; Elias et al., 1991; Loader et al., 1993; Witt et al., 1996), with the decision of whether to undergo screening falling primarily on women. However, reservations concerning the development of appropriate programs for CF carrier screening led to a moratorium on widespread carrier testing until studies addressed various educational, psychological, and logistical issues not previously addressed in other genetic screening programs (American Society of Human Genetics, 1992; National Institutes of Health, 1990; Wilfond & Fost, 1990). The National Institutes of Health have funded a series of studies designed to address these issues and to determine how best to offer voluntary CF carrier screening. As part of this effort, our primary goal in this study was to examine pregnant women's attitudes toward and interest in CF carrier screening in a prenatal setting.

We examined whether some known psychosocial determinants of preventive health behavior and disease detection behavior have an impact on pregnant women's attitudes toward genetic carrier screening for CF. The health belief model (HBM; Rosenstock, 1966) has been fairly successful in documenting some of the factors that influence an individual's decision to perform a variety of health behaviors, and it appears to be a useful theoretical framework for understanding attitudes toward CF carrier screening.

Several studies of carrier screening have found that various components of the HBM are effective in predicting attitudes toward genetic carrier screening and behavior (Becker, Kaback, Rosenstock, & Ruth, 1975; Ben-Sira & Padeh, 1978).

HEALTH BELIEF MODEL

The HBM was originally formulated in an attempt to understand why people fail to take action to prevent or detect disease (Rosenstock, 1966). According to the HBM, the likelihood that an individual will take action is determined by several factors: perceived vulnerability to the health condition, perceived severity of the disease, perceived benefits of performing the health behavior, perceived costs and barriers of performing this health behavior, and "cues" that trigger the individual to perform this action.

Research on the HBM has examined the extent to which certain variables from the model successfully explain why people engage in health-promoting behaviors

(see Janz & Becker, 1984). For example, the HBM has been examined in the context of mammography utilization (Aiken, West, Woodward, & Reno, 1994; Stein, Fox, & Murata, 1991), vaccination behavior (Aho, 1979; Fosu, 1991), and even carrier screening for Tay-Sachs Disease (Becker et al., 1975). In the study of carrier screening for Tay-Sachs Disease, 500 screening program participants were compared with the 368 nonparticipants on their health beliefs. In general, participants perceived themselves to be more vulnerable to being carriers of the Tay-Sachs gene than nonparticipants. In addition, those who believed that knowing their carrier status would be highly disruptive to future family planning (high perceived severity) were less likely to participate in the screening program than those who did not perceive such an impact. Perceived benefits and barriers of testing were only asked of those couples who expressed an intention to have children (or additional children). Of these respondents, nonparticipants were more likely than participants to indicate that they would change their reproductive plans if one or both members of a couple were found to be carriers. These findings suggested that individuals who did not perceive themselves to be vulnerable to this health threat and who believed that learning of a negative health state would be personally disruptive were more likely to avoid participation in a screening program.

RESEARCH ON CF CARRIER SCREENING

Although the HBM has not been examined in the context of CF carrier screening, some studies in this area have used constructs similar to those found in the HBM, such as perceived risk or vulnerability and perceived barriers (Tambor et al., 1994; Witt et al., 1996). Tambor et al. (1994) found that among nonpregnant women who were planning to have children, those who perceived a higher risk of being a carrier (i.e., high perceived vulnerability) were significantly more likely to be tested. In addition, testing rates were significantly higher when the testing could be obtained with minimal effort (i.e., low perceived barriers). Similarly, Witt et al. (1996) found that a major factor involved in pregnant women's decisions to be screened was the ease of testing (i.e., low perceived barriers). In addition, pregnant women who declined screening reported that they believed their risk for having an affected baby was very low (i.e., low perceived vulnerability). The findings from these studies suggest that several of the HBM constructs are associated with genetic carrier screening behavior.

However, genetic carrier screening introduces several issues that are different from those found in preventive health behaviors and disease detection behaviors. First, CF screening is unlike disease detection behaviors because the "detection" of the CF gene is of no particular health threat to the tested individual (i.e., a CF carrier), but rather, poses the possibility of a serious threat to the health of any future children. Furthermore, knowing that one is a CF carrier still does not provide complete information regarding risk for any future children. Instead, the additional

step of having a partner tested is required for a more accurate assessment of risk. Therefore, unlike diagnostic tests for illness, information regarding the potential for this health threat necessitates the involvement of not only oneself, but also of one's partner.

Second, if both partners are found to be carriers of the CF gene and they decide to have children together, they have the option of testing each unborn child for CF. However, because there is no known cure for CF, early detection of an affected fetus leaves most couples with difficult decisions. Consequently, more general attitudes regarding abortion and future family planning issues may have a considerable impact on the behavior of carrier screening.

Third, current CF screening tests can detect only a small fraction of the more than 400 possible mutations of the CF gene, and therefore have only a 50% to 92% sensitivity for carrier detection, depending on the individual's ethnic background. Thus, there is the possibility, albeit small, that one might receive a false-negative test result and unknowingly have a child with CF, even after one has performed this health behavior.

Therefore, learning about CF and the carrier screening program may be very anxiety-provoking for pregnant women. That is, the women learn that participation in a screening program may lead to negative outcomes, such as finding out that they are a CF carrier, which may lead to subsequent stigmatization and discrimination (Kass, 1992; OTA Report Brief, 1992), and in some cases, discovering that their unborn child has CF. On the other hand, nonparticipation in a screening program (i.e., not getting screened) may also lead to negative outcomes, such as unknowingly having a child with an incurable genetic disease. The use of psychological defense mechanisms has been implicated in preventive health behaviors in which both engagement and nonengagement of the prescribed behavior have a probability of leading to negative outcomes (Ben-Sira & Padeh, 1978). In other words, knowing that both the performance as well as the nonperformance of the health behavior may potentially lead to negative outcomes may cause such distress that individuals attempt to relieve this distress through the use of certain defense mechanisms, such as denial or avoidance of the situation. Specifically, by denying that this threat is present in oneself, perceptions of susceptibility to this health threat may diminish, thereby alleviating psychological and emotional distress. In a study of carrier screening for Tay-Sachs disease, Ben-Sira and Padeh (1978) found that pregnant women, who were more emotionally distressed by the potential outcome of the Tay-Sachs screening than nonpregnant women, tended to use denial-like mechanisms more than nonpregnant women, and the use of these defense mechanisms was negatively associated with women's beliefs in their chances of being a Tay-Sachs carrier and giving birth to a child with Tay-Sachs disease. Their results suggest that these psychological defense mechanisms may affect carrier screening behavior by altering individuals' perceptions of vulnerability (Ben-Sira & Padeh, 1978).

Further evidence for the use of distress-reducing coping strategies can be found in the literature on monitoring versus blunting (Miller, 1980, 1981). When an individual is faced with a stressful, aversive event (such as one in which both performing and not performing a health behavior may lead to negative outcomes), the individual can reduce emotional arousal and distress by using a variety of strategies to remove himself or herself from psychological awareness of this event. Miller (1980, 1981) proposed that individuals may use "blunting" strategies, such as distraction, to help remove themselves psychologically from these aversive events. For example, individuals can distract themselves by thinking about other activities or events. This strategy should not only reduce emotional arousal, but also reduce processing of threat-relevant external information (see Miller, 1980, p. 149).

Although the educational materials used in this study were simplified as much as possible, information regarding carrier screening for genetic diseases is a complex issue because it involves mathematical probabilities of carrier risk, gene detection and transmission, and the potential for false-negative test results. Previous studies have found educational level to be positively correlated with comprehension of genetic concepts (Yuen, Hsia, & Hall, 1988). Therefore, the educational level of participants was hypothesized to be a factor in determining whether one is able to fully understand and grasp the concepts and risk ratios presented in the educational materials. A more complete understanding of the educational materials will no doubt directly influence one's perceptions of vulnerability or risk as well as one's perceptions of the barriers and benefits associated with genetic carrier screening.

Consequently, in this study, it was hypothesized that avoidant coping strategies, such as denial and distraction, would influence women's perceptions of their risk for being a carrier of CF and of giving birth to a baby with CF. Specifically, it was proposed that these avoidant coping mechanisms would act to reduce women's perceptions of vulnerability to being a carrier of CF and having a baby who has CF. In addition, it was predicted that individuals who use avoidant coping mechanisms would process less information about CF and genetic screening issues, resulting in reduced comprehension of the topics presented in the educational materials. The impact of avoidant coping on perceptions of severity, barriers, and benefits associated with genetic carrier screening was also examined.

The constructs of perceived vulnerability, perceived severity, perceived barriers to screening, and perceived benefits of screening were hypothesized to covary among themselves and to be predictive of pregnant women's attitudes toward CF carrier screening. Based on previous research (for a review, see Janz & Becker, 1984), it was hypothesized that greater perceptions of vulnerability or risk would be associated with more positive attitudes toward, or greater interest in, genetic carrier screening. In addition, perceived severity was expected to be positively associated with greater interest in or more positive attitudes toward genetic carrier screening. Perceiving more benefits to screening was also expected to be posi-

tively related to favorable attitudes toward genetic carrier screening, whereas perceiving many barriers to screening was hypothesized to be negatively related to interest in genetic carrier screening.

METHOD

Participants

A sample of 1,625 women attending the University of California, Los Angeles (UCLA) Prenatal Diagnosis Center or Kaiser Permanente clinics in the greater Los Angeles area for general prenatal care or amniocentesis counseling were recruited for participation in this study. There were 577 women (35.5%) who declined to participate in the study. Of the 1,048 participants, the sample consisted of 601 non-Hispanic Whites, 93 Asians, 65 Blacks, 230 Hispanics, and 59 participants of mixed or other ethnic background.

Only non-Hispanic Whites were selected for this study's analyses for several reasons. First, as mentioned before, non-Hispanic Whites (and especially Whites of northern European descent) are at greatest risk for being a carrier of CF, compared with other ethnic groups. Consequently, most interventions in this area will be targeted at this high-risk group. Second, previous research has indicated the existence of ethnic differences in health perceptions (for examples see Prohaska, Albrecht, Levy, Sugrue, & Kim, 1990; Stein et al., 1991; Takeuchi, Leaf, & Kuo, 1988). Our own preliminary analyses also demonstrated that there are ethnic differences in health perceptions in the context of CF carrier screening (Fox et al., 1993); however, the numbers of ethnic minorities were too small to perform meaningful structural equation modeling analyses designed for this study.

Of the 601 non-Hispanic White participants, 70 participants failed to respond to nine or more items, and an additional 20 participants did not indicate educational background and were treated as missing and dropped from the sample, leaving a total of 511 participants. An examination of the data revealed no systematic differences in health beliefs (all $ps > .05$) between the 90 omitted cases and the 511 that were retained.

All participants were either in their first or early second trimester of pregnancy and had no family history of CF. Women who were more than 18 weeks pregnant were not asked to participate in this study because if it were discovered that the fetus was affected with CF, termination of the pregnancy might not be possible. The participants ranged in age from 17 to 44, with a mean age of 34. The mean number of years of education in this sample was 15.03 years, and approximately 45% of the participants had received some advanced education beyond a college degree. Finally, 87% described themselves as being currently married, and 53% had one or more children.

Materials

Two questionnaire packets, an educational brochure, and a video were developed for use in this study. The brochure and video, designed by a research team including physicians, psychologists, and genetic counselors at UCLA's Prenatal Diagnosis Center, provided information on the clinical features and genetics of CF and the variable sensitivity of the screening test in different ethnic populations (see Fox et al., 1994; Tatsugawa et al., 1994). The brochure presented information in a question-and-answer format by addressing questions such as "How is CF inherited?" "Am I at risk for being a carrier of CF?" and "How is the test performed?" The 5-min video covered the same information as the brochure.

The pre-education questionnaire consisted of a demographics inventory and a 13-item Avoidant Coping scale. The Avoidant Coping scale items and instructions were taken from the Ways of Coping scale (Folkman & Lazarus, 1985; Folkman & Lazarus, 1988) and the COPE Inventory (Carver, Scheier, & Weintraub, 1989). For each item, participants were asked to indicate how often they coped with stress in a particular manner. Specifically, items were drawn from the Escape–Avoidance and Distancing subscales of the Ways of Coping scale (e.g., "I try not to think about it," "I try not to talk about it," "Even when I have a lot of feelings about problems, I don't deal with them"), and supplemented with Denial items from the COPE measure (e.g., "I refuse to believe that it has happened," "I pretend that it hasn't really happened," "I act as though it hasn't even happened"). Responses were made on a 4-point scale, ranging from 1 (*Usually don't do this at all*) to 4 (*Usually do this a lot*). This hybrid of features from two commonly used coping measures was developed to examine the avoidant coping styles that were believed to be most theoretically relevant to this study.

The posteducation questionnaire was composed of 18 items designed to measure the HBM constructs. Four items were designed to assess Perceived Vulnerability ("There is the chance that I have a cystic fibrosis gene," "There is a chance that my child will have a cystic fibrosis gene," "There is a chance that a child of mine will be born with cystic fibrosis," and "My partner could have a cystic fibrosis gene"). Three items were used to measure Perceived Severity ("Cystic fibrosis is one of the worst diseases there is," "Seeing someone with cystic fibrosis would really upset me," and "It would upset me to know that I have a cystic fibrosis gene"). Five items were designed to assess Perceived Barriers ("It is inconvenient for me to have a test for the CF gene today," "Even if I wanted to, I am not able to have a test for the cystic fibrosis gene today," "There is nothing preventing me from having a test for the cystic fibrosis gene today" (reverse coded), "I am uncomfortable about being tested to see if I have a cystic fibrosis gene," and "It would be too upsetting to be tested for the cystic fibrosis gene"). Six items were used to measure Perceived Benefits ("It is important for me to know if I have a cystic fibrosis gene," "It would be helpful for me to know if the child I am carrying has cystic fibrosis," "Knowing

if my partner and I each have a cystic fibrosis gene is valuable information," "Knowing my CF genetic test result will help me make better decisions about having children in the future," "I think my partner would want me to have the test for the cystic fibrosis gene," and "Everyone in the United States should be offered testing for the cystic fibrosis gene"). All items were measured on a 5-point Likert-type scale with endpoints ranging from 1 (*strongly agree*) to 5 (*strongly disagree*). In addition, two items were used to assess participants' attitudes toward, or interest in, CF carrier screening. The first item was worded, "I feel fine about being tested to see if I have a cystic fibrosis gene," and was measured using the same 5-point Likert-type scale just mentioned. The second item instructed participants to select one of four statements that best described their feelings about CF genetic testing: (a) "I do not want to be tested for the cystic fibrosis gene now or in the future"; (b) "I am not sure whether to be tested"; (c) "I would like to be tested for the cystic fibrosis gene sometime, but not today"; and (d) "I want to be tested for the cystic fibrosis gene today."

Procedure

Participants were recruited from the UCLA Prenatal Diagnosis Center and several prenatal clinics at a large health maintenance organization in the greater Los Angeles area. Pregnant women who were undergoing routine prenatal care and had a negative family history for CF were considered eligible for the study. All women were approached individually while they were in the waiting room prior to their regular appointment. Women who wished to participate in the study completed an informed consent form. Then, participants were given the pre-education questionnaire. After completing the pre-education questionnaire, the participants were randomly assigned to receive either the written educational materials or the video educational materials as part of an intervention study, one goal of which was to compare the two modes of delivering information. An examination of participants' knowledge and understanding of the materials revealed no differences between the written and video materials (Fox et al., 1994). Consequently, analyses are reported for data collapsed across the two conditions. Following the presentation of the educational materials, the participants completed the posteducation questionnaire.

After the participants completed the posteducation questionnaire, they were fully debriefed by a genetic counselor, who explained the purpose of the study and answered any remaining questions. During this debriefing period, genetic counselors discussed the option of undergoing genetic carrier screening for mutations of the CF gene, and they provided additional information about the screening procedure. All participants were assured that their test results would remain confidential. After this consultation, participants were offered the CF screening test free of charge. Previous CF screening programs have reported screening rates of only 1% to 4% in the general population (Clayton et al., 1996; Tambor et al., 1994). To

our surprise, 501 of the 511 participants in this study chose to be screened. Due to the restrictions of the research setting, however, we were unable to obtain any further measures of health beliefs or attitudes following this consultation. Moreover, because the final consultation varied from participant to participant (e.g., for some participants, the consultation was a straightforward question and answer session, whereas for other participants it involved a deeper discussion of the participant's individual concerns), it is impossible to know exactly how our psychosocial and attitudinal measures were related to the participants' screening behavior. Therefore, the analyses in this article are restricted to the women's attitudes that were formed after learning about CF, but before this final individualized consultation.

Data Analyses

Factor analytic methods were used to examine the HBM factors and the avoidant coping factor. Before testing the measurement model, preliminary exploratory factor analyses were conducted to determine the optimum configuration for each construct. Confirmatory factor analyses (CFAs) were then performed to evaluate the goodness of fit of the factor structure. Once a well-fitting measurement model was obtained, the hypothesized path model was tested using the EQS structural equation modeling program (Version 5; Bentler & Wu, 1995). Modifications of this model were then implemented and tested. All model parameters were estimated using a maximum likelihood method. For both the CFA and the path model, goodness of fit was assessed with the chi-square statistic and the comparative fit index (CFI; Bentler, 1990). A chi-square to *df* ratio of less than 2:1 and fit index values greater than .90 are reflective of good model fit.

RESULTS

Factor Analyses

HBM. An exploratory factor analysis was performed on the 18 HBM items. Four factors were extracted using the maximum likelihood method and were rotated using a direct oblimin solution. All but one item loaded on its hypothesized construct. That item, written to measure perceived severity, was found to load weakly on both perceived severity and perceived barriers. In addition, the low internal consistency of the perceived severity scale, $\alpha = .47$, was considered problematic for subsequent analyses, and therefore, this measure was dropped from further examination.

To examine the factor structure of the remaining three factors, CFA methods were then used. Each of the 15 remaining items was constrained to load onto one

factor, and the three factors were allowed to covary. The initial CFA resulted in fit indices of $\chi^2(87, N = 511) = 201.57$, $p < .05$, CFI = .94. The Lagrange-Multiplier Test (L–M Test) indicated that the addition of four paths from manifest variables to latent constructs would improve model fit. These complex factor loadings were theoretically defensible, allowing two barrier items to load negatively onto the benefits factor and allowing two benefits items to load negatively onto the barriers factor. These modifications improved model fit, $\chi^2(83, N = 511) = 169.79$, CFI = .96.

Avoidant coping styles: Denial and distancing. An exploratory factor analysis was conducted on the avoidant coping items. Four factors were extracted using the maximum likelihood method and rotated using a direct oblimin oblique solution. After examination of the factors, the first two factors were labeled as *denial* and *distancing*. The third and fourth factors were not interpretable, and were therefore not examined further.

CFA was used to examine the goodness of fit of a two-factor model of avoidant coping. Each item was constrained to load onto one factor, and the two factors were allowed to covary. The initial CFA resulted in fit indices of $\chi^2(8, N = 511) = 16.65$, $p = .034$, CFI = .98. As suggested by the L–M Test, we allowed one distancing item to also load onto the denial factor. This modification improved model fit, $\chi^2(7, N = 511) = 6.49$, $p = .48$, CFI = .996.

Correlations Among Latent Factors and Attitudes Toward CF Genetic Screening

A composite variable of attitudes toward CF carrier screening was formed by summing the standardized scores from the two attitude items. The means of the two items were 4.45 and 3.91 (*SD*s = 0.62 and 0.42, respectively). Mean scores of Perceived Vulnerability, Barriers, Benefits, Denial, and Distancing were created for each participant by averaging the individual items comprising each factor. Educational level was computed by standardizing and summing across the number of years of school and educational degree. Mean scale scores, standard deviations, and alpha coefficients are reported in Table 1. The correlation matrix among the HBM factors, avoidant coping factors, educational level, and attitudes toward genetic carrier screening are presented in Table 2.

Path Analysis

To test a formal model of the relation between the HBM constructs and avoidant coping and attitudes toward CF genetic screening, a structural modeling analysis was performed. The model included both latent constructs, measured by several

TABLE 1
Means and Standard Deviations of Scales

Scale	*M*	*SD*	*Range*
Perceived vulnerability (α = .92)	3.27	.85	1–5
Perceived barriers (α = .79)	1.72	.57	1–5
Perceived benefits (α = .82)	4.26	.53	1–5
Denial (α = .84)	1.19	.39	1–4
Distancing (α = .76)	1.74	.65	1–4

Note. Educational degree: No degree, 9.6%; high school, 0.9%; A.A., 28.4%; B.A. or B.S., 14.7%; M.A. or M.S., 33.9%; M.D., Ph.D., or J.D., 11.4%.

TABLE 2
Correlations of Health Belief Model Scales, Avoidant Coping Factors, Education, and Genetic Screening Attitudes

	Perceived Barriers	*Perceived Benefits*	*Denial*	*Distancing*	*Educational Level*	*Attitudes*
Perceived vulnerability	−.245***	.190***	−.069	−.125**	.118**	.249***
Perceived barriers		−.577***	.127**	.121**	−.057	−.628***
Perceived benefits			−.099*	−.076	.029	.545***
Denial				.302***	−.037	−.079
Distancing					−.078	−.028
Years of schooling						.090*

*$p < .05$. **$p < .01$. ***$p < .001$.

indicators (e.g., perceived vulnerability, perceived barriers, perceived benefits, educational level, distancing and denial), and measured variables assessed with one indicator (e.g., knowledge of CF and genetic carrier screening issues was measured as the number of knowledge items answered correctly after education). The dependent variable of attitude toward CF genetic screening was computed so that higher scores represent more positive attitudes toward CF genetic screening. Because the dependent variable was positively skewed, we used the Satorra–Bentler chi-square, which is a scaled test statistic that is designed to improve the distribution of the standard test statistic under violations of the normality assumption (Chou, Bentler, & Satorra, 1991).

The hypothesized model proposed that greater levels of perceived vulnerability and perceived benefits would be positively associated with attitudes toward CF genetic screening, whereas perceiving more barriers would be negatively associated with attitudes toward CF genetic screening. Due to the interrelations among perceived vulnerability, barriers, and benefits, the error terms (or disturbances) for

those three factors were allowed to covary (Bentler & Wu, 1995, p. 244).[1] In addition, it was hypothesized that the avoidant coping factors of distancing and denial would be negatively associated with perceived vulnerability as well as with less knowledge. Educational level was expected to be positively associated with knowledge. In addition, all paths among the predictor latent factors (denial, distancing, and educational level) and the outcome variable (attitudes toward CF genetic screening) were included, and the factors of distancing, denial, and educational level were allowed to covary. Exploratory paths from distancing and denial to perceived barriers and perceived benefits were also included.

The test of the hypothesized model resulted in an adequate model fit, $\chi^2(256, N = 511) = 441.15$, CFI = .94, although there were a number of nonsignificant paths indicating that the model could be trimmed and improved. Modifications suggested by the Wald test (Chou & Bentler, 1990) included dropping several nonsignificant paths from the latent constructs to the outcome variable (e.g., the direct paths from educational level, distancing, and denial to attitudes toward CF genetic screening were dropped). The final model, $\chi^2(258, N = 511) = 426.49$, CFI = .95, is illustrated in Figure 1, with all significant paths and covariances included.

As expected, perceived vulnerability and perceived benefits were positively related to attitudes toward CF genetic screening, whereas perceived barriers were negatively related to attitudes toward CF genetic screening. As predicted, educational level was positively related to knowledge of CF and genetic screening issues. Higher educational attainment was also significantly correlated with less use of distancing. As predicted, distancing was negatively related to knowledge of CF and genetic screening issues. Although distancing did not have any direct effects on perceived vulnerability, perceived barriers, or perceived benefits, it did indirectly affect perceived vulnerability and perceived barriers through a negative association with knowledge. Knowledge was positively related to perceived vulnerability and negatively associated with perceived barriers. Distancing was also positively correlated with denial. Contrary to predictions, denial was not directly related to knowledge. However, denial was significantly related to greater perceived barriers and fewer perceived benefits. As mentioned previously, there was no direct effect of distancing, denial, or educational level on attitudes toward CF genetic screening. However, each of these factors did have a significant indirect effect (via the HBM factors and, in some cases, knowledge) on participants' attitudes toward CF genetic screening. The total indirect effects of educational level, distancing, and denial on the outcome variable are presented in Table 3.

[1] In this model, the latent constructs of perceived vulnerability, barriers, and benefits are considered dependent variables that are explained by other variables. The statistical program, EQS, does not allow dependent variables to covary with other variables. Consequently, covariances among the disturbances, or error terms of the dependent variables, were used to reflect these relations (Bentler & Wu, 1995).

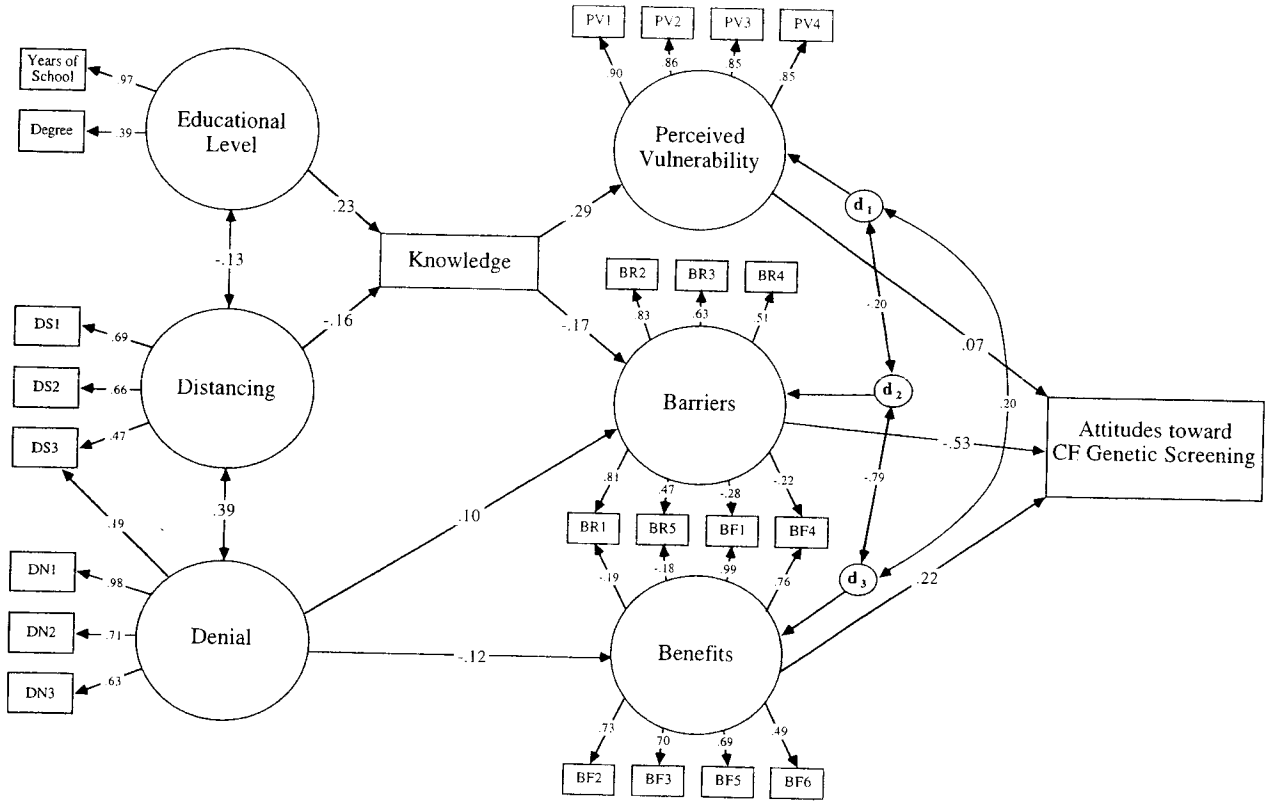

FIGURE 1 An examination of the effects of educational level, distancing, and denial on health belief model constructs and attitudes toward CF genetic screening. Following the conventional practice in the causal modeling literature, measured variables are represented by rectangles, and latent factors are represented by circles. All paths are significant at $p < .05$.

TABLE 3
Effects of Educational Level and Avoidant Coping Styles on Attitudes Toward Cystic Fibrosis Genetic Screening

Variable	Total Effect	Direct Effect	Indirect Effects Via		
			Perceived Vulnerability	Perceived Barriers	Perceived Benefits
Educational level	.024**	0	.004	.020	
Distancing	−.017*	0	−.003	−.014	
Denial	−.079*	0		−.053	−.026

Note. Total effects are defined as the sum of direct and indirect effects. The total indirect effect (in this case, it is equivalent to the total effect) is associated with a standard error that is used to compute the z-test statistic.

*$p < .01$. **$p < .001$

DISCUSSION

This is the first study to examine the utility of the HBM constructs and avoidant coping in the context of pregnant women's attitudes toward genetic carrier screening for CF. The results indicate that perceived vulnerability, barriers, and benefits were all significant predictors of women's attitudes toward CF genetic carrier screening. The strongest predictor of carrier screening attitudes was perceived barriers. As expected, greater perceived barriers predicted less favorable attitudes and less interest in undergoing genetic screening. Perceived vulnerability and perceived benefits were also associated with carrier screening attitudes, with higher levels of perceived vulnerability and benefits predicting more positive attitudes toward genetic testing. These results are comparable to findings from other studies that have used the HBM as a predictive tool (see Janz & Becker, 1984, for a review), but are the first to emerge in the context of CF carrier screening.

Furthermore, our investigation of the relations among avoidant coping and health beliefs has extended the utility of the HBM in this new domain of genetic screening. For example, denial was found to indirectly influence CF screening attitudes through the perception of greater barriers and fewer benefits. The relations found among denial and health perceptions are interesting in light of research that has suggested that avoidant coping styles may be associated with poorer health outcomes (e.g., Frenzel, McCaul, Glasgow, & Schafer, 1988) or may impede adaptive coping processes (Suls & Fletcher, 1985). Although previous research has shown that denial may be useful in enabling people to gradually adjust to threatening events (e.g., Janoff-Bulman & Timko, 1987; Lazarus, 1983; Taylor & Aspinwall, 1993), the regular use of denial as a coping strategy in the area of disease detection or preventive health may be maladaptive because it prevents the perform-

ance of the health behaviors necessary to reduce one's risk. In other words, individuals who tend to use avoidant coping strategies may not undertake preventive health measures or initiate difficult lifestyle changes because they selectively perceive more barriers and fewer benefits of that health behavior.

The other coping factor, distancing, was found to be negatively correlated with education, replicating findings from a previous study of cancer patients (Dunkel-Schetter, Feinstein, Taylor, & Falke, 1992). As predicted, distancing was associated with less knowledge of CF and thus may reflect reduced processing of threat-relevant information (Miller, 1980). Distancing was also negatively related to lower perceptions of CF carrier risk indirectly. This finding partially parallels the finding of Ben-Sira and Padeh (1978) of a negative relation between avoidant coping and perceived risk of being a Tay-Sachs carrier. Our results also indicate that distancing and denial had separate and distinct roles. This study is not the first to identify these different dimensions of coping (see Carver et al., 1989), but it reinforces the growing recognition that distancing and denial are distinct aspects of avoidant coping with distinct correlates.

As expected, a positive relation was found between educational level and participant's knowledge of CF and carrier screening issues. This relation replicates previous research findings that education is correlated with comprehension of genetic information (Yuen et al., 1988), and more specifically, with the comprehension of CF carrier screening issues (Clayton et al., 1995; Hannig et al., 1994). This finding may be because genetics involves complex concepts not familiar to the average person. Despite our efforts to simplify the educational materials, the information contained in them (e.g., probability of carrier status, gene detection and transmission, reliability of testing) remained quite technical. Consequently, the relation between knowledge and risk perception may be a function of more educated participants having a more complete understanding of the educational materials, and thus, of their vulnerability to their CF carrier status.

An understanding of genetic concepts and CF genetic transmission was also related to perceiving fewer barriers to screening. This relation suggests that knowledge of the relevant issues may eliminate any misconceptions that might act as potential barriers. The absence of a relation between knowledge and perceived benefits may be due to the fact that the educational materials were geared toward providing information about CF and the screening test, whereas perceived benefits of genetic screening was operationalized as the personal advantages or benefits, such as making informed family planning decisions, that one would gain through genetic screening. These "benefits" are not determined by whether an individual has learned the educational materials, but rather by subjective personal judgments of priorities. Indeed, not everyone would agree that informed family planning decisions, or knowing that one's unborn child has an incurable genetic disease, is a "benefit."

Although the model fit was quite good, it may have been constrained by the limited variability of the screening attitudes variable. As previously mentioned, participants' attitudes toward CF carrier screening were positively skewed, indicating a generally positive attitude toward screening. This generally positive predisposition of women in our prenatal CF screening program limited the predictive power of the model. Genetic screening projects in other settings can help determine whether these results are replicable and if a better model fit might be obtained.

As previously mentioned, 98% of the participants chose to be tested for mutations of the CF gene following the individual consultation with the genetic counselor (Grody et al., 1996). Compared with other studies of CF carrier screening (Clayton et al., 1996; Tambor et al., 1994), this project yielded an unusually high rate of screening. One factor that may have contributed to this high screening rate was the absence of barriers to screening (i.e., the test was free, did not involve drawing blood, and could be quickly and simply performed). Findings from previous studies have indicated that fewer barriers to testing are associated with higher testing rates (Tambor et al., 1994; Witt et al., 1996). In addition to minimizing the number of barriers, our findings also indicate that increasing participants' awareness of the potential benefits of screening is likely to elicit more positive attitudes toward screening, and possibly, higher screening rates. The high rate of screening in this study may also have been due, at least in part, to the individualized consultation, during which time participants were able to discuss the option of testing with a genetic counselor. Because we were unable to collect psychosocial and attitudinal measures after the final consultation, we cannot speculate further as to how the final consultation may have affected screening behavior.

One limitation of this research is that the sample consisted of generally well-educated White women. Consequently, these findings may not be generalizable to other populations. However, because non-Hispanic Whites are the ethnic group at greatest risk for being carriers of the CF gene, most screening programs will be targeted at this high-risk group. Future studies of carrier screening should also investigate the interrelations among health beliefs and avoidant coping style in other ethnic groups. This need is highlighted by our preliminary analyses on data from Black, Asian, and Hispanic women that show some differences in the relations among the HBM constructs and genetic screening attitudes (Fang et al., 1996; Fox et al., 1994).

These findings suggest several avenues for future research. One avenue for future research is to examine how actual coping behaviors (both avoidant and nonavoidant) are related to health perceptions and attitudes toward prenatal testing. A second focus for future studies is to determine what factors may have influenced the "decliners," or those women who did not participate in this study. Finally, other directions for future research involve the identification of additional factors not included in our model that may also influence attitudes toward genetic screening,

such as concerns about confidentiality of test results, attitudes toward abortion, or negative affectivity. These variables may contribute to our further understanding of women's attitudes toward prenatal genetic screening.

CONCLUSION

In summary, we would like to highlight two key findings. First, perceived barriers were the strongest predictor of pregnant women's interest in and attitudes toward genetic carrier screening for CF. These findings indicate that both physical and psychological barriers are important factors to consider in understanding women's carrier screening attitudes and behavior. Second, this study contributes to the understanding of the precursors of women's health perceptions. Knowing what factors may influence perceptions of vulnerability to disease and assessments of potential benefits and barriers associated with particular health attitudes in pregnancy is valuable information for researchers and for the providers of screening programs. The findings from this study of pregnant women's attitudes toward prenatal genetic screening can provide valuable insight into how women engage in decision-making processes regarding genetic risk and their behavioral options.

As the mapping of the human genome continues to advance at a rapid pace, the availability of carrier screening tests for other genetic diseases will undoubtedly follow. These tests will present similar issues to CF genetic screening and additional problems as well. Given that carrier screening behavior differs from preventive health behavior in some significant ways, the development of models to predict attitudes toward and interest in genetic screening will be of paramount importance.

ACKNOWLEDGMENTS

This research was sponsored by the Ethical, Legal, and Social Implications Program of the National Center for Human Genome Research and supported by the National Institute of Child Health and Human Development Grant HD29337–03. Preparation of this article was supported in part by the National Institute of Mental Health predoctoral training Grant MH15750.

We acknowledge Fabiana Wallis and Juliana Fuqua for their assistance in recruiting participants for this study. We thank Leona S. Aiken, Peter M. Bentler, and Annette L. Stanton for their helpful comments on earlier versions of this article.

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