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Psychosocial impact of genetic carrier status in the general population

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PSYCHOSOCIAL IMPACT OF GENETIC CARRIER STATUS IN THE GENERAL POPULATION

A Thesis

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of the Requirements for the Degree

Master of Arts

by

Margaret-Anne Mackintosh

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APPROVED FOR THE DEPARTMENT OF PSYCHOLOGY

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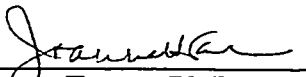

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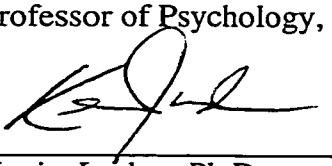

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ABSTRACT

PSYCHOSOCIAL IMPACT OF GENETIC CARRIER STATUS IN THE GENERAL POPULATION

by Margaret-Anne Mackintosh

Abstract

This study investigated four areas related to the psychosocial impact of carrying a gene for a recessive genetic disorder: affective responses to being a carrier, changes in health perceptions, the possible presence of stigma, and the influence of demographic and cognitive factors on attitudes about being heterozygotic. Reactions to the hypothetical possession of a genetic mutation were measured using a modified version of the Health Orientation Scale (HOS; Wooldridge & Murray, 1988). Measures of basic genetic knowledge and attitudes about genetic testing also were collected. Participants were 161 individuals at a state university. The research revealed that being a carrier of a cystic fibrosis mutation is viewed as a negative experience, producing reductions in how individuals view their health, and leading to slight feelings of stigmatization. None of the cognitive or demographic factors studied significantly influenced attitudes about being a carrier. These findings are compared with previous research using the HOS (Evers-Kiebooms et al., 1993; Wooldridge & Murray, 1988). The results point to the need for (1) greater education of genetic principles within the general population and (2) the absolute importance of genetic counseling within the genetic testing cycle.

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Footnote

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Abstract

This study investigated four areas related to the psychosocial impact of carrying a gene for a recessive genetic disorder: affective responses to being a carrier, changes in health perceptions, the possible presence of stigma, and the influence of demographic and cognitive factors on attitudes about being heterozygotic. Reactions to the hypothetical possession of a genetic mutation were measured using a modified version of the Health Orientation Scale (HOS; Wooldridge & Murray, 1988). Measures of basic genetic knowledge and attitudes about genetic testing also were collected. Participants were 161 individuals at a state university. The research revealed that being a carrier of a cystic fibrosis mutation is viewed as a negative experience, producing reductions in how individuals view their health, and leading to slight feelings of stigmatization. None of the cognitive or demographic factors studied significantly influenced attitudes about being a carrier. These findings are compared with previous research using the HOS (Evers-Kiebooms et al., 1993; Wooldridge & Murray, 1988). The results point to the need for (1) greater education of genetic principles within the general population and (2) the absolute importance of genetic counseling within the genetic testing cycle.

Psychosocial Impact of Carrier Status in the General Population

Recently, geneticists have isolated genetic mutations that influence many disorders, making it possible for medical practitioners to provide individuals with an unprecedented amount of information about themselves and their future health. The type of information now available includes an individual's potential for developing various genetically influenced disorders as well as any risk of having children with hereditary diseases. As more and more genes related to medical disorders are identified, people will become confronted increasingly with defects in their own genetic structure, those of their family members, and in those with whom they interact. What is the impact of discovering that one possesses a defective gene? What does the identification of such a gene mean when we, in fact, each possess five to ten disease related genes (Hietala et al., 1995)?

This paper will explore attitudes related to carrying a mutation for a recessive disorder, and how this relates to an individual's concept of health. First, the literature concerning the impact of identification as a carrier of a mutation for a recessive genetic disorder will be reviewed, focusing on the psychosocial impact of identification and factors that influence reactions to this genetic knowledge. Second, goals and specific hypotheses for this study are presented. The results then are discussed in relation to the previous research on genetic carrier status and within the larger context of the possible introduction of genetic screening programs in the United States.

Psychosocial changes related to carrier status

For many disorders, the possession of a mutated gene does not have any direct health effects for the individual. Autosomal recessive disorders, such as cystic fibrosis (CF), Tay Sachs, and sickle cell anemia, require that both parents of a child pass on a copy of a mutated gene in order for the child to have the disorder. Each parent may be unaffected by the disorder, but merely be heterozygotes, possessing one normal copy of the gene and one mutated gene. If both parents are carriers of the disease, each of their

children has a one in four chance of having the disorder. Carrier tests for autosomal recessive disorders focus on providing information to people who may be at higher risk for producing offspring with genetic disease (Axworthy, Brock, Bobrow, & Marteau, 1996; Berg, Pettersson, Riis, & Tranøy, 1995; Haan, 1993, Marteau, 1992).

Several studies have investigated the psychosocial impact of carrier testing for various autosomal recessive disorders, such as Tay Sachs (Clow & Scriver, 1977; Zeesman, Clow, Cartier, & Scriver, 1984) and CF (Bekker, Denniss, Modell, Bobrow, & Marteau, 1994; Childs, Gordis, Kaback, & Kazazian, 1976; Evers-Kiebooms, Denayer, Welkenhuysen, Cassiman, & Van den Berghe, 1994; Fanos & Johnson, 1995a & b; Mennie et al., 1992 & 1993). Recently, several CF screening trials have been conducted in the United States (Clayton et al., 1996; Witt et al., 1996), in Great Britain (Axworthy et al., 1996; Bekker et al., 1993; Harris et al., 1996; Mennie et al., 1993; Miedzybrodzka, Haites, & Dean, 1993; Watson et al., 1993) and in Germany (Jung, Urner, Grade, & Coutelle, 1994). Others have studied potential stigmatization of carriers by themselves and others, and discrimination in obtaining employment and health insurance (Billings et al., 1992; Evers-Kiebooms et al., 1994; Wooldridge & Murray, 1988)

Affective consequences. Many studies have found that individuals report being satisfied with discovering their genotype (Clow & Scriver, 1977; Harris et al., 1996; Jung et al., 1994; Mitchell, Scriver, Clow & Kaplan, 1993; Watson, Mayall, Lamb, Chapple, & Williamson, 1992; Witt et al., 1996). For example, Clow and Scriver (1977) investigated attitudes about genetic screening for Tay Sachs in high school students. Though 15% of the students tested considered the prospect of the screening test to be frightening, 95% of the non-carriers and 98% of the carriers were satisfied to know their genotype. In a follow-up study seven years later, the majority of these students still had positive attitudes about the testing experience and reported making reproductive decisions based on their

genetic test results (Zeesman et al., 1984). Similarly, several of the CF carrier screening trials recently conducted in Europe found high percentages of individuals (94-97%) who reported being glad to have been tested or willing to recommend testing to others (Harris et al., 1996; Jung et al., 1994; Witt et al., 1996).

In addition to positive attitudes about the testing experience, many studies have found no negative emotional impact of discovering one's carrier status or that any negative effects were ameliorated over time (Bekker et al., 1994; Childs et al., 1976; Mennie et al., 1993; Mitchell et al., 1993; Witt et al., 1996). Genetic counseling also has been found to be effective in relieving some of the anxiety generated from CF screening (Watson et al., 1992; Witt et al., 1996). For example, 71% of women who tested positive for a CF mutation during early pregnancy reported that genetic counseling was emotionally helpful (Witt et al., 1996). Although anxiety does not seem to be present in the months and years following testing for most individuals, several studies have shown that up to 20% of those tested had concerns several years following testing.

Though most individuals report satisfaction with gaining genetic information about themselves, negative affective consequences, such as anxiety, shock, guilt, and depression, have been reported for those identified as carriers (Axworthy et al., 1996; Childs et al., 1976; Clow & Scriver, 1977; Evers-Kiebooms et al., 1994; Harris et al., 1996; Jung et al., 1994; Loader et al., 1996; Mennie et al., 1993; Watson et al., 1992; Witt et al., 1996; Zeesman et al., 1984). Anxiety both during the genetic testing process and immediately after receiving positive carrier results has been reported (Bekker et al., 1994; Harris et al., 1996; Mennie et al., 1993; Mitchell et al., 1993). In a prenatal screening program of women without a family history of CF, 23% of those screened reported feeling anxious about their perceived carrier risk prior to testing, and had significantly higher scores on a measure of generalized psychological disturbance in comparison to baseline measures. While awaiting their own test results, partners of the women who had tested

positive for a CF mutation also had significantly higher anxiety and inadequacy subscores on the General Health Questionnaire compared to controls (Mennie et al., 1993). However, scores for the women and their partners on all measures returned to baseline after their partner had received negative test results for their own carrier status. This pattern of initial anxiety subsiding after the partners tested negative has been found in several other studies (Jung et al., 1994; Watson et al., 1993; Witt et al., 1996).

Longer term anxiety also has been found. In a study of 3000 individuals without a familial history of CF, carriers were found to have significantly higher scores on the Spielberger State Anxiety Inventory following testing as compared to pretest measures. Tested noncarriers, on the other hand, reported the opposite: significantly lower scores following screening compared to scores prior to screening (Watson et al., 1992). The same study found that three months after testing, 39% of the carriers reported still being slightly anxious and 38% reported being worried. A follow-up study of high school students tested for Tay Sachs also found that 19% of the carriers still reported being worried about their test results seven years later, which is significantly less than the 46% that expressed anxiety at the time of testing (Clow & Scriver, 1977; Zeesman et al., 1984).

It must be pointed out that although most individuals screened will receive negative results, some non-carriers are not relieved to discover they do not possess a disease-related gene. In a sample of adult siblings of individuals with CF, Fanos and Johnson (1995a & b) found that assumptions made by the siblings about their own carrier status were influenced by several factors, including level of resentment of the CF sibling while growing up, guilt, and size of the family of origin. For example, Fanos and Johnson (1995a) found that 11% of the siblings wished that they were carriers and 53% assumed that they were carriers of a CF mutation prior to genetic testing. These researchers conclude "perception of one's carrier status, at least in affected families, is a complex process by which positive carrier status may represent expected retribution for angry

wishes. It also represents a way of binding guilt, a desire to share the experience of the sick sibling" (p. 437). A study of individuals who had a parent with Huntington's disease found that eight of the nine individuals identified as non-carriers in the study initially reported feeling guilty and depressed (Tibben et al., 1990).

In summary, investigations of the affective consequences of carrier testing have provided mixed evidence for long term consequences of positive and negative results. Overall, there appears to be at least some short-term negative affective changes, such as anxiety, related to discovering that oneself is a carrier. Many studies have used unidimensional, self-report measures of the central concepts (e.g., anxiety and depression). Studies using standardized measures (e.g. Spielberger state/trait anxiety scale or General Health Questionnaire) or more in-depth qualitative probing may provide stronger evidence for emotional consequences, both negative and positive, resulting from carrier testing.

Changes in perception of health. In addition to affective changes, individuals' assumptions about their health status also have been found to be influenced by carrier status for several different recessive disorders such as CF (Axworthy et al., 1996; Fanos & Johnson, 1995b), Tay Sachs (Marteau, van Duijn, & Ellis, 1992), and sickle cell anemia (Hill, 1994). Others studying individuals screened for CF, however, have not reproduced this finding (Bekker et al., 1994; Mitchell et al., 1993). Researchers found that parents often do not understand the results of carrier tests performed on their children. For example, 43% of parents of identified carriers in a sickle cell carrier screening program believed that their child had a disease, and 66% of those parents incorrectly believed that the child needed dietary supplements because of the condition (Hampton, Anderson, Lavizzo, & Bergman, 1974). Fanos and Johnson (1995a) also found that 30% of the CF siblings as well as 13% of the siblings' spouses believed that there were health problems associated with CF carrier status. In addition, 55% of the siblings and 41% of the spouses

exhibited moderate to severe anxiety concerning their children's health. Interestingly, carriers and non-carriers were equally as likely to exhibit this type of anxiety.

People's perception of their future health also can be affected when they discover they are heterozygotic for a recessive genetic disorder. In a retrospective study of individuals screened for Tay Sachs carrier status, identified carriers were found to hold less optimistic views of their future health when compared to noncarriers, even though carrier status poses no threat to physical health. Axworthy and colleagues (1996) found that carriers of a CF mutation perceived their current health as significantly poorer compared to those who tested negative, but found no differences in past or future health perceptions.

In summary, changes in an individual's perception of their present and future health have been identified following genetic testing. Further research needs to be conducted on how people would interpret the effects of carrier status on health apart from the testing process.

Presence of stigmatization. Other psychosocial changes, such as downward shifts in self-image and stigmatization related to being identified as a carrier, have been important considerations in genetic testing. Self-image changes, as measured by self-report measures and willingness to disclose one's carrier status, have been used as measures of stigmatization produced by genetic testing. Goffman (1974) describes stigma symbols as "signs which are especially effective in drawing attention to a debasing identity discrepancy, breaking up what would otherwise be a coherent overall picture, with a consequent reduction in our valuation of the individual" (p. 44). He identifies two kinds of stigmas: ones that are readily apparent during social interactions and ones that are not directly visible, but that still separate the possessor from those that do not possess them. For the first group, whom Goffman labels the "discredited," social interactions are governed by the need to manage the social tension caused by the stigma. The second group, the "discreditable," focus on managing information during social interactions,

deciding whether or not to disclose information that proves that they are different from what others assume them to be. Individuals possessing a gene for a recessive genetic disorder could fall into this second group. Therefore, stigmatization can be measured both as a set of characteristics that lead us to degrade how we see ourselves as well as information we try to conceal during most social interactions.

Several studies have shown changes in self-image related to discovering heterozygotic status. In one study, nearly one out of ten identified Tay Sachs carriers reported diminished self-image upon discovering their status, while the same proportion of noncarriers reported improved self-image (Clow & Scriver, 1977). In another Tay Sachs screening program, 20-25% of non-carriers reported that their self image would have been lowered if they had discovered they were carriers (Mitchell et al., 1993).

Several studies have not found any changes in self-image associated with carrier status (Childs et al., 1976; Watson et al., 1992). In a high school screening program for CF, students who received positive results for CF carrier status reported no changes in self-image (Mitchell et al., 1993). Ninety-four percent of Tay Sachs carriers reported no need to keep the information a secret, and carrier couples were more likely than noncarriers to discuss genetic testing and their results with their cousins, aunts, and uncles (Childs et al., 1976). Similarly, Watson and colleagues (1992) found that individuals identified as carriers of a CF mutation also showed little reluctance in disclosing their carrier status. For example, six months after testing 89% of carriers had discussed the information with their partner, 83% with their parents, 82% with siblings, and 63% with friends.

However, stigmatization based on results of genetic carrier testing has been identified as a potentially significant problem with population based screening for many genetic disorders (Kenen & Schmidt, 1978; Markel, 1992; Rowley, 1984; Wilfond & Fost, 1990). Clow and Scriver (1977) found that only one third of the Tay Sachs carriers discussed their status with their friends, while 85% of noncarriers did discuss their results.

Witt et al. (1996) found 11% of those consenting to CF carrier testing reported that they felt people would look at them differently if they were found to be a carrier. The disclosure of one's carrier status is important, because Goffman's (1974) theory of stigma described "discrediting" stigma symbols as characteristics that people would be uncomfortable revealing because it would make them appear to be different from what they are assumed to be.

Two studies have been conducted specifically to investigate stigmatization associated with heterozygotic carrier status (Evers-Kiebooms et al., 1994; Wooldridge & Murray, 1988). These studies used a multidimensional quantitative measure, the Health Orientation Scale (HOS), to explore changes in self-image and stigmatization following carrier screening, and both found stigmatization to be associated with carrier status.

Wooldridge and Murray (1988) were the first to use the HOS to investigate the impact of carrier testing for sickle cell anemia as well as to assess the effects of genetic counseling. The HOS asks respondents to imagine themselves in a number of situations, and then rate how they would feel using twelve pairs of bipolar adjectives. The researchers found that those who tested negative for the sickle cell trait reported significantly more negative feelings than did carriers in the following situations: when discovering they had the sickle cell trait, disclosing one's carrier status to friends, considering the birth of a child with sickle cell anemia, and when describing how they believe carriers feel about themselves. When respondents were asked to imagine that they were the boss of several individuals with sickle cell trait, people found not to carry the sickle cell trait reported that the employees who were carriers would feel significantly less able, worse, sadder, weaker, angrier, sicker and more marked, afraid, ashamed, and inactive in comparison to ratings made by actual carriers of the trait.

Carriers and non-carriers did not differ significantly on direct measures of self-image. However, there was some evidence of self-stigmatization. Carriers reported they

would feel bad, sad, angry and shocked when faced with discovering they had the sickle cell trait. Overall, Wooldridge and Murray (1988) conclude that "the greater potential for stigmatization clearly appears to lie with non-carriers who consistently reveal more negative attitudes than carriers toward sickle cell trait and sickle cell disease and the capabilities of carriers" (p. 134).

Evers-Kiebooms et al. (1994) used a modified version of the HOS to assess respondents' feelings upon discovering whether or not they carried a CF mutation. These researchers replaced the nine original HOS situations with three that measured (1) individuals' self-descriptions, (2) feelings they attributed to most carriers, and (3) feelings they attributed to most non-carriers. They found that when both carriers and non-carriers were grouped together, all respondents rated carriers as reporting significantly more negative feelings on all twelve scales. That is, they reported that carriers of a CF mutation feel more shocked, worse, less happy, more afraid, more angry, weaker, less healthy, more marked, less active, more ashamed, less able, and more guilty in comparison to tested non-carriers.

Actual CF carriers also were found to have significantly more negative self-description than tested non-carriers, with carriers reporting being less happy, weaker, and more shocked, angry, and afraid than tested noncarriers. Interestingly, carriers also reported that other carriers would describe themselves in more negative terms. They rated other carriers as feeling more shocked, less healthy, more ashamed, more guilty, more marked, more afraid, worse, weaker, and more angry than they do themselves as carriers.

Even though evidence of stigmatization following carrier identification has produced mixed results, studies using a multi-dimensional measure or in-depth interviews have found some evidence of stigmatization. This effect includes not only a downward change in self-image by those identified as carriers, but also the fact others believe that carriers feel worse about themselves than the carriers actually do. Further research needs to

be conducted to investigate whether or not individuals in the general population would also view the possession of a recessive mutation as a stigma symbol.

Demographic and cognitive factors related to carrier status

In addition to studying the consequences of genetic testing, researchers have investigated factors that influence individuals' reactions to discovering they are heterozygotic for a recessive genetic disorder. Many researchers have studied the influence of demographic variables, such as age, income level, and education level, on attitudes and reactions to carrier testing (Childs et al., 1976; Evers-Kiebooms et al., 1994; Fanos & Johnson, 1995a; Surh, Cappeli, MacDonald, Mettler, & Dales, 1994; Witt et al., 1996). As a whole, the research investigating the influence of individual demographic factors on the uptake and reactions to genetic testing have been inconclusive. Several studies have found that age and level of general education do not influence attitudes about carrier testing (Denayer, De Boeck, Evers-Kiebooms, & Van den Berghe, 1992; Childs et al., 1976; Evers-Kiebooms, 1987; Fanos & Johnson, 1995a). Witt and colleagues (1996), however, found that individuals who declined CF carrier testing were younger and less educated. In a study of Tay Sachs carriers, attitudes were not influenced by the number of children an individual had or was planning to have (Childs et al., 1976). However, planning to have children was found to influence desire for carrier testing in aunts and uncles of CF patients (Denayer et al., 1992). Witt et al. (1996) found that women who already had had successful pregnancies were less likely to participate in a prenatal CF carrier screening program than women who did not have children.

Research investigating the impact of religious views on attitudes about and acceptance of carrier testing has produced mixed findings. Several studies have found religious views influence people's decision to be tested (Axworthy et al., 1996; Botkin & Alegmagno, 1992; Clayton et al., 1996; Fadden, Tambor, Chase, Geller, Hofman, & Holtzman, 1994; Miller & Schwartz, 1992). Botkin and Alegmagno (1992) found that

pregnant women who attended religious services frequently were less likely to desire carrier testing during pregnancy. However, not all groups within a particular religion view the use of genetic services in a similar fashion. For example, Miller and Schwartz (1992) found that even within several highly conservative, Christian religious sects (Mennonites, Amish, Hutterites and Rochesterians), these groups felt differently about the acceptability of using carrier testing, prenatal diagnosis, and abortion. Others have found no such influence (Denayer et al., 1992; Witt et al., 1996). For example, religious attendance had no impact on aunts' and uncles' of CF patients desire to undergo carrier testing (Denayer et al., 1992).

The impact of gender also has been found to be inconclusive. Clayton et al. (1996) found that women were more likely to consent to carrier testing. Another study reported that while gender did not influence feelings attributed to most carriers, there was a significant interaction between gender and carrier status. Female carriers of CF reported significantly more negative self-descriptions compared to male carriers, while female non-carriers had significantly more positive self-descriptions than male non-carriers (Evers-Kiebooms et al., 1994). Tambor et al. (1994), however, found that prior to conception, there were no differences in uptake rate for CF testing between women and men.

Interestingly, one study found that when the husband was found to be a carrier and the wife was not, both members of the couple were likely to report being "upset," but when the wife was a carrier, she was significantly more likely to suffer alone (Childs et al., 1976). One researcher hypothesizes that perhaps men's self-image is more strongly influenced by their perceived health and their ability to reproduce (Marteau et al., 1992).

The ethnicity of the those tested and an individual's cultural setting also may affect the use of genetic services and the subsequent reaction to the information provided. Studies have found that individuals consenting to CF testing are more likely to be Caucasian (Clayton et al., 1996; Witt et al., 1996), which is consistent with the prevalence

of CF mutations within that population. Hill (1994) interviewed lower income, African-American women and found that constraints based on ethnicity and social class kept women who knew they were carriers from being able to use this information to prevent births of children with sickle cell anemia. The women did not have the power to convince their partners to undergo genetic testing once the women had discovered that they were carriers of the sickle cell trait. Hill concludes that motherhood was "one of the few status-attaining and satisfying options available to these low-income women, and they were unwilling to sacrifice their right to have children" (p. 43).

Punales-Morejon and Penchaszadeh (1992) reported an overall increase of 100% (75 to 150 patients per year in a community of 100,000 individuals of Chinese ancestry) in utilization of genetic services during the first two years when these services were designed and implemented in an ethnocultural-sensitive manner. They argued that ethnocultural differences are one of the major barriers to the use of genetic services, because the larger culture in which individuals are immersed often has its own interpretations of the causes of birth defects and genetic conditions as well as culture-specific definitions of health, illness, and disability.

Though many have investigated the impact of demographic variables on willingness to undergo genetic testing and reactions subsequent reactions to the information, few have looked at the impact of cognitive variables, such as basic genetic knowledge, attitudes about genetic testing, and previous exposure to information about the disorder, on reactions to being heterozygotic for a recessive disorder. Research has shown that a higher understanding of basic genetic material influences individuals' reactions to carrier status (Denayer et al., 1992; Massarik & Kaback, 1981). In a study of individuals at higher risk for Tay Sachs, Massarik and Kaback (1981) found that the highest level of basic genetic knowledge was related to the highest readiness to seek testing. In contrast, Quaid and Morris (1993) found that a greater understanding of basic genetic principles was related to

low uptake of genetic testing services for Huntington's Disease (HD), while there was a high level of interest in HD testing among those who know little about testing (Meissen & Berchek, 1987). This difference possibly could be due to the differences between testing for an autosomal recessive disorder (Tay Sachs) and an autosomal dominant disorder (Huntington's Disease); however, further research needs to be conducted. For the present study it was predicted that a higher level of genetic knowledge would be related to less negative reactions to being identified as a carrier.

In summary, many of the studies on the affective responses to carrier testing and results concerning moderating factors have been mixed. Overall, it appears that there are long-term consequences to being identified as a carrier of a gene for an autosomal recessive disorder. Anxiety, depression, and even guilt for individuals coming from families with a history of a disorder have been found in several studies using either multi-dimensional measures or in-depth interviews to measure these responses. Ethnic and cultural differences seem to have an effect, but it is not clear what carrier status means in each population. For other demographic and cognitive variables, their effects are not as clear.

Given the findings associated with heterozygotic status, research needs to be conducted investigating factors underlying the meaning of carrier status in the general population. With the proliferation of genetic tests and the continued identification of genes that are responsible for or influence the development of numerous disorders, people will be increasingly faced with genetic information about themselves, potential marital partners, and others in society. Various strategies have been suggested to control for misuse of information derived from these tests. For example, genetic counselors are charged with the tasks of helping clients understanding genetic information and to help ameliorate any negative impact of genetic testing. Broad-based educational efforts also have been suggested and modeled to reach the general population through high school and college biology courses (Clow & Scriver, 1977; Decruyenare, Evers-Kiebooms, Denayer, & Van

den Berghe, 1992; Massarik & Kaback, 1981; Mitchell, Capua, Clow, & Scriver, 1996). Researchers have focused on the development of information leaflets and videos to aid in the education and decision making prior to CF screening (Livingstone, Axton, Mennie, Gilfillan, & Brock, 1993). By identifying the factors or characteristics that are likely to influence individuals' attitudes about carrier status, both genetic counselors and educators can tailor their efforts to meet the "needs" of the individuals with whom they are working.

Purpose and hypotheses

The present study focused on two areas related to the impact of being heterozygotic for a CF mutation. The first explored the psychosocial impact of being a carrier by specifically looking for (1) changes in affective consequences to being a carrier, (2) changes in health perceptions, and (3) the possible presence of stigmatization. The second goal was to assess the impact of demographic and cognitive factors on attitudes about being a carrier.

Goal 1: Psychosocial changes related to carrier status. Hypotheses investigating the psychosocial impact of possessing a genetic mutation on individual's self-concept and the way in which this may influence social interactions was explored by comparing individuals' reactions to several situations related to discovering that one has a genetic mutation. Responses to each situation included measures of affective response, health perception, and possible stigmatization (See Table 1 for a description of each situation). Specific hypotheses were:

A. Individuals' present self-descriptions of themselves would be more positive than if they believed themselves to be carriers (present self vs. carrier self).

B. Disclosure of one's carrier status would be a more negative experience than one's present description of self (present self vs. self disclosure).

C. Carriers would describe themselves in more positive ways than they would other carriers (carrier self vs. other carrier).

Table 1: Situations included in Modified Health Orientation Scale - Modified (in order of presentation).

	Modified Health Orientation Scale Situations
Carrier-other	I am in charge of 50 people who do different kinds of jobs. I learn that several of my employees are carriers of the cystic fibrosis trait. I imagine that the person carrying the gene for CF might feel:
Carrier-self	My doctor has just told me that I am a cystic fibrosis carrier. I feel:
Self-disclosure	I have the cystic fibrosis trait. Over a game of cards, there is a conversation about cystic fibrosis. As I consider whether or not to mention that I have the cystic fibrosis gene, I feel:
CF-diagnosis	My doctor has just told me that I have cystic fibrosis. I feel:
Schizophrenia-diagnosis	My doctor has just told me that I have schizophrenia. I feel:
AIDS-diagnosis	My doctor has just told me that I have AIDS. I feel:
Present-self	The following terms best describe me in general:

This study investigated the impact of one being a carrier compared to learning that one has various medical disorders. To investigate further the impact of a genetic mutation on people's perception of health, an individual's ability to differentiate between being a carrier for CF (no health consequences) and having the disease (severe health consequences) was assessed. Second, being a carrier was contrasted with known stigmatizing conditions: schizophrenia (Clausen, 1980) and AIDS (Herek & Capitano, 1993). Specific hypotheses were:

D. Being told that one is a carrier would be a less negative experience than discovering one has CF.

E. Being told that one is a carrier would be significantly less negative experience than being told that you have schizophrenia or AIDS (less stigmatization and no direct health effects).

Goal 2: Demographic and cognitive factors related to carrier status. Finally, this study sought to identify factors that influenced individual attitudes about being carriers. Most research to date has concentrated on demographic factors influencing an individual's uptake of genetic services. The presence of a family history for a particular disorder and ethnic or cultural background seem to be the demographic factors that most likely influence the uptake and reactions to genetic testing. Other factors with mixed support include religion and its frequency of practice, gender, and the number of children one would like to have. In addition, the effect of cognitive variables, such as attitudes about genetic testing in general, the level of basic genetic knowledge, and previous exposure to information about CF, were used to predict response to being heterozygotic.

Method

Participants

One hundred and sixty-one students from introductory psychology courses at San Jose State University participated in the survey. Data from six participants were dropped

due to too much missing data. The remaining sample of 155 individuals consisted of 91 females and 64 males. The sample was ethnically diverse, consisting of 32% Asian, 22% Caucasian, 16% Filipino, 12% Hispanic, 6% African-American, and 12% other ethnicities. Ages ranged from 17-51 with an average age of 20 years. Most (97%) were single and had not yet started families (median number of present children was zero). The majority of individuals did want to have children in the future (median number of children desired = 2). Only 3.2% (5 individuals) reported having known a family in which CF had occurred, but 93% had heard of CF prior to this study.

Materials

Health Orientation Scale-Modified. The original version of the HOS was designed to measure the affective response to the possession of a defective gene, changes in self-image related to carrier status, and to investigate attitudes related to selected health concerns. The HOS used the semantic differential scaling technique (Osgood, Suci, & Tannenbaum, 1957) to measure individuals' responses to various situations. For example, participants were presented with the following situation, "*You are in charge of fifty people, and you just found out several are carrying the sickle cell trait. You imagine they feel: ...*"

Each situation is followed by 13 bipolar word pairs measuring affective response to the situation. Individuals rated their perception of how a carrier would feel along each set of bipolar adjectives, such as *healthy:sick* or *happy:sad.* One additional bipolar word pair was included in this study, *anxious:relaxed*, to directly assess anxiety. The adjective pairs represented the anchors of a seven point scale (See Appendix B for complete scale). Adjectives representing more positive attributes (e.g., healthy, happy, capable) were scored as the low end of the scale. Therefore, situations with higher total scores were being perceived as more negative life occurrences. The order in which the adjective pairs appeared within the situations was randomly determined, though their position remained constant across all situations.

Several modifications were made to the HOS for this study. The HOS-M included seven situations, five original situations and two new ones (See Appendix B for the complete scale). These seven situations can be divided into two subgroups, which measure the emotional reaction to genetic carrier status from two different perspectives. The first section of the HOS-M contains four situations, which were designed to investigate the psychosocial impact of possessing "discreditable" information about oneself and others according to Goffman's (1974) theory of stigma. The first three situations were constructed to assess an individual's initial self-esteem (present-self), feelings upon discovering that one is a carrier (carrier-self), and feelings in sharing one's heterozygotic status (self-disclosure). The final situation asked individuals to describe how they believe *others* feel about themselves (carrier-other). Goffman's theory of stigma predicts that people would report that being identified as a carrier and disclosing this information would be more negative experiences than initial ratings of self-esteem.

The last three situations measured reactions to diagnoses of various medical conditions. One of the original HOS situations was used; diagnosis of CF. Two new situations also were added: "You have just been told you have (1) schizophrenia and (2) AIDS." These three medical conditions were chosen to provide a backdrop against which to judge the meaning of genetic carrier status to provide two comparisons: respondent's ability to differentiate between CF the disease and being heterozygotic for CF, and whether stigmatization may be associated with being a carrier. Several studies have found stigmatization following the diagnosis of mental illness (schizophrenia) for both the individual identified with the condition and for family members (Clausen, 1980; Wahl & Harman, 1989). Feelings of stigmatization also have been associated with testing positive for HIV (Herek & Capitano, 1993).

The second major modification to the HOS-M was to create three sub-scales from the 13 bipolar adjective pairs used in response to the situations. The affective subscale was

created to measure overall emotional response. The affective subscale was the average of six of the adjective pairs (*good:bad, unafraid:afraid; relieved:shocked, happy:sad, angry:pleased;* and *anxious:relaxed*). The health subscale sought to measure changes in an individual's perception of their health and ability when considering the various situations. Four adjective word pairs were averaged for this scale (*strong:weak, able:unable, active:inactive,* and *healthy:sick*). Finally, the stigma scale was created by averaging the three remaining word pairs (*unmarked:marked, unashamed:ashamed*, and *not guilty:guilty*). These three scales were developed because previous research supported changes in these concepts following carrier testing.

The third modification made to the HOS-M was to alter the gene mutation being studied. The original HOS used sickle cell trait as the identified mutation, while this study used CF. There are at least two advantages to changing recessive disorders. First, even though the CF carrier tests have not satisfied all the points necessary before population based screening can occur (Wilfond & Fost, 1990; Williamson, 1993), pilot programs have been funded in the United Kingdom and United States (National Institutes of Health Workshop, 1990). Second, using a different recessive disorder will strengthen the validity of the HOS as a measure of reactions to the possession of a recessive genetic mutation, rather than just the possession of the sickle cell trait. This will help broaden the possible uses of the HOS. Also, CF mutations are the most common recessive genes in the United States, appearing in 4-5% of the Caucasian population, and leading to 1 in 2500 Caucasian children having the disease (Cystic Fibrosis Research Inc., 1995). Though the sickle trait is carried by a larger percentage of individuals in the African-American population (approximately 8%), and has a higher incidence rate of 1 in 835 births (Agency for Health Care Policy and Research, 1993), the CF gene is more common because of the larger Caucasian population in the United States.

Attitudes toward genetic testing. Respondents' attitudes concerning genetic testing were assessed (see Appendix C for complete scale). Respondents were asked to rate the degree to which they agree/disagree with 14 statements used by Hietala et al. (1995) to identify attitudes about genetic testing as well as reasons for testing in the general population of Belgium. Answers were scored such that higher scores represented a more positive attitude toward genetic testing. This information was used as one of the cognitive predictor variables of respondent's reactions to the possession of a mutated gene.

Test of basic genetic knowledge. A 14-question, multiple-choice test of genetic knowledge developed by Massarik and Kaback (1981) was administered to assess the influence of basic genetic knowledge on reactions to the possession of a genetic mutation (see Appendix D for complete test). The number of items correctly identified was summed and used as the measure of genetic knowledge. The test scores were used as one of the cognitive predictor variables. This test was administered prior to providing participants with information about CF and the meaning of carrier status.

Demographics questionnaire. Demographic variables, such as the respondent's age, gender, religious practices, and ethnicity, were collected (see Appendix E). These variables were used as demographic factors that possibly influence attitudes towards genetic carrier status.

Introductory information about Cystic Fibrosis. Participants were provided with a two-page description of CF, including brief sections on symptoms, how CF is treated, and the genetic transmission of CF (see Appendix F for information sheets). The information was adapted from a teacher's curriculum on the prevention of genetic and birth disorders (Burhansstipanov, Giarratanu, & Keger, 1987). The original document was prepared before the CF gene had been discovered, so the section on genetic transmission was updated to reflect recent progress in gene identification and the availability of carrier testing. Also included was a diagram and a written description of the transmission of a recessive

trait. This information was provided to students immediately prior to filling out the HOS-M to expose them to basic information about the nature of the disease and the principles of recessive transmission.

Procedure

Participants were tested in groups as they were recruited from the Psychology Participant Pool at San Jose State University. Students were informed about the general nature of the research, questions concerning the study were answered, and informed consent was obtained. Research packets containing materials in Appendices B-F were then distributed. Participants were instructed to work through the packets sequentially. It took 30 min to read and respond to all the questions. Upon completion of the questionnaire, participants received a written debriefing statement explaining the goals and hypotheses of the research, and any further questions about the study were answered.

Results

Goal 1: Psychosocial changes related to carrier status

Hypothesis A: Present-self vs. carrier self. Goffman's (1974) theory of stigma predicts that if CF carrier status is a stigmatizing condition, individuals would report a lowering of self-esteem when comparing self-esteem prior to testing to self-esteem when they believed themselves to be a carrier. To test the hypothesis that CF carrier status may be a stigmatizing condition a 2 (HOS-M situation) x 3 (HOS-M subscales) repeated measures Analysis of Variance (ANOVA) was conducted on the average subscale scores for the present-self and carrier-self situations. A Situation x Subscales interaction was found, $F(2, 296) = 92.2$, $p < .001$ (see Figure 1 for graph of means and standard errors). For the present-self situation, the affective and stigma subscales were significantly higher than the activity subscale (Ms = 3.2, 3.2, and 2.8, respectively). For the carrier-self situation, all subscales were significantly different from one another with the affective scale

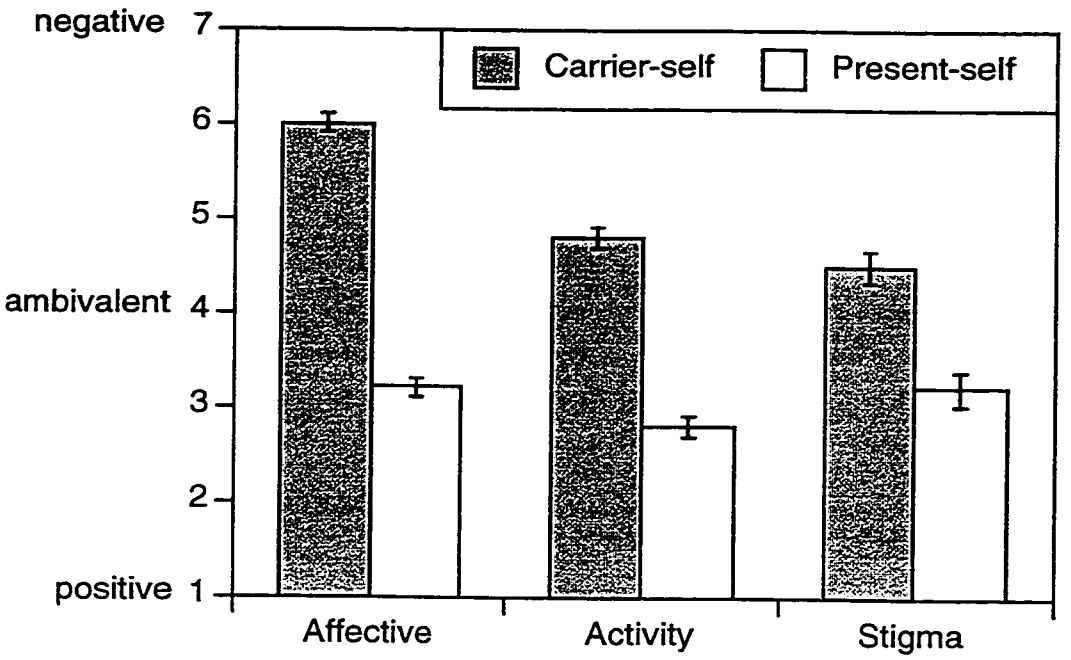

Figure 1. Means with Standard Error Bars for the Health Orientation Scale - Modified Subscales for the Present-self and Carrier-self Situations

being the most negative (M = 6.0), followed by the activity subscale (M = 4.8), and then the stigma subscale (M = 4.5).

More theoretically interesting is the main effect for situations, F(1, 148) = 285.8, p < .001. An individuals description of themselves were significantly more negative when they thought about themselves as being a carriers. The largest difference between situations was on the affective subscale with a mean difference of 2.8 points on a 7-point scale. The mean difference on the activity subscale was 2.0 points and 1.3 points on the stigma subscale. Thus, the hypothesis was supported: people had a negative affective response to thinking about themselves as a carrier, carrier status would lower their view of their health, and they would feel somewhat stigmatized by carrier status.

Hypothesis B: Present-self vs. Carrier-disclosure. Goffman's theory also predicted that if a condition was stigmatizing, disclosing one's carrier status would be a negative experience, since people would want to hide the discrediting characteristic. To test this theory, a 2 (HOS-M situation) x 3 (HOS-M subscales) repeated measures ANOVA was conducted on the mean ratings for the present-self and self-disclosure HOS-M situations. The self-disclosure situation asked the respondents to describe how they would react to disclosing their hypothetical CF carrier status. An interaction between situations and subscales was found, F(2, 296) = 11.7, p < .001 (see Figure 2 for graph of means and standard errors). As with the first hypothesis, the affective and stigma subscales for the present-self situation were significantly higher than the activity subscale (M = 3.2, 3.2, and 2.8, respectively). For the carrier-disclosure situation, however, the affective subscale (M = 5.3) was significantly higher than the activity and stigma subscales (Ms = 4.7 for both).

Again, the more theoretically interesting finding was the main effect for HOS-M situations, F(1, 148) = 216.8, p < .001. The ratings for the carrier-disclosure subscales were significantly more negative than those for present-self. Similar to the first hypothesis, the largest mean difference was on the affective subscale (mean difference = 2.1 points).

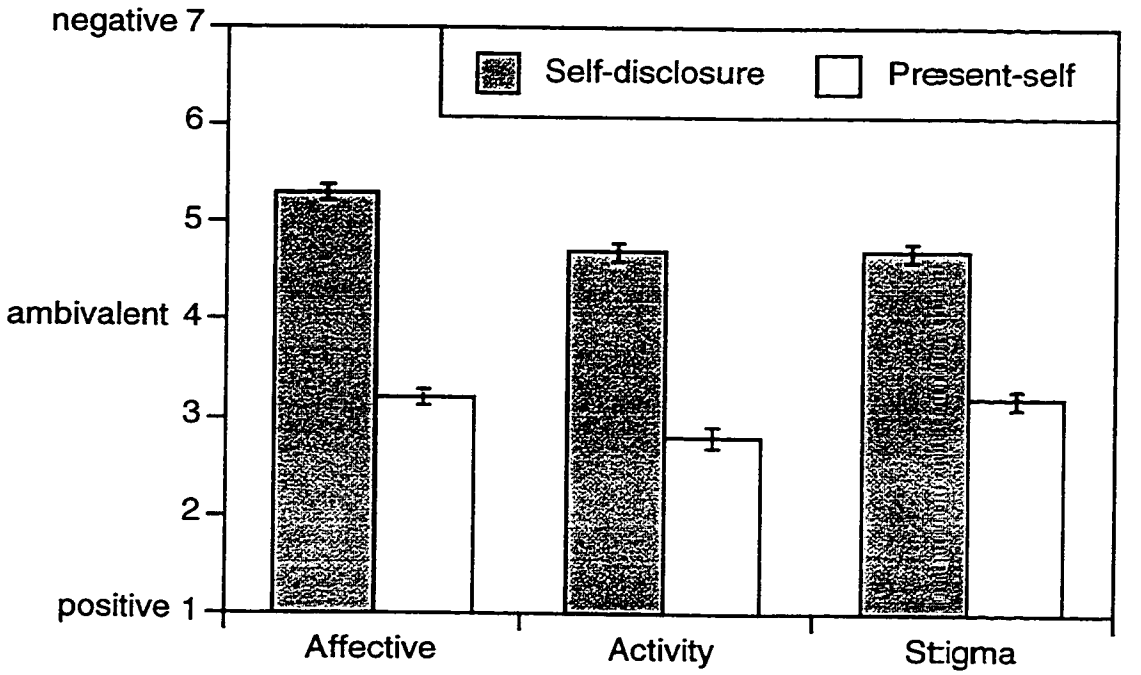

Figure 2. Means with Standard Error Bars for the Health Orientation Scale - Modified Subscales for the Present-self and Self-disclosure Situations

The difference on the activity subscale was 1.9 points, and 1.5 points on the stigma subscale. The hypothesis based on Goffman's theory was supported in that disclosing one's carrier status was viewed as a negative affective experience. As predicted, disclosure of one's carrier status also was related to downward shifts in health ratings and increased level of stigmatization.

Hypothesis C: Carrier-self vs. Carrier-other. This hypothesis sought to explore the psychosocial impact of discovering that one was a carrier compared to discovering that someone else was a carrier. To this end a 2 (HOS-M situation) x 3 (HOS-M subscales) repeated measures ANOVA was conducted on the mean subscales ratings for the carrier-self and carrier-other situations. A significant interaction was found, $F(2, 288) = 10.9$, $p < .001$ (see Figure 3 for a graph of the means and standard errors). Although all means are significantly different, Figure 3 shows that there are larger mean differences between the situations for the affective and stigma subscales (mean differences = .9 and .6, respectively) than for the activity subscale (mean difference = .4). This may account for the interaction.

The hypothesis that individuals would describe themselves in more positive ways than they would other carriers was not supported by the data situation (main effect for situations; $F (1, 144) = 75.36$, $p < .001$). In fact, the opposite was found; means on the subscales for carrier-self situations were significantly higher than for carrier-other. This suggests that individuals thought that they would feel worse, less healthy, and more stigmatized than they thought others would feel about being a carrier.

A second set of hypotheses investigated the impact of carrier status compared to other medical conditions. The first hypothesis was meant to test how well individuals differentiated between having CF and being heterozygotic for a CF mutation. The second hypothesis compared CF carrier status to known stigmatizing conditions.

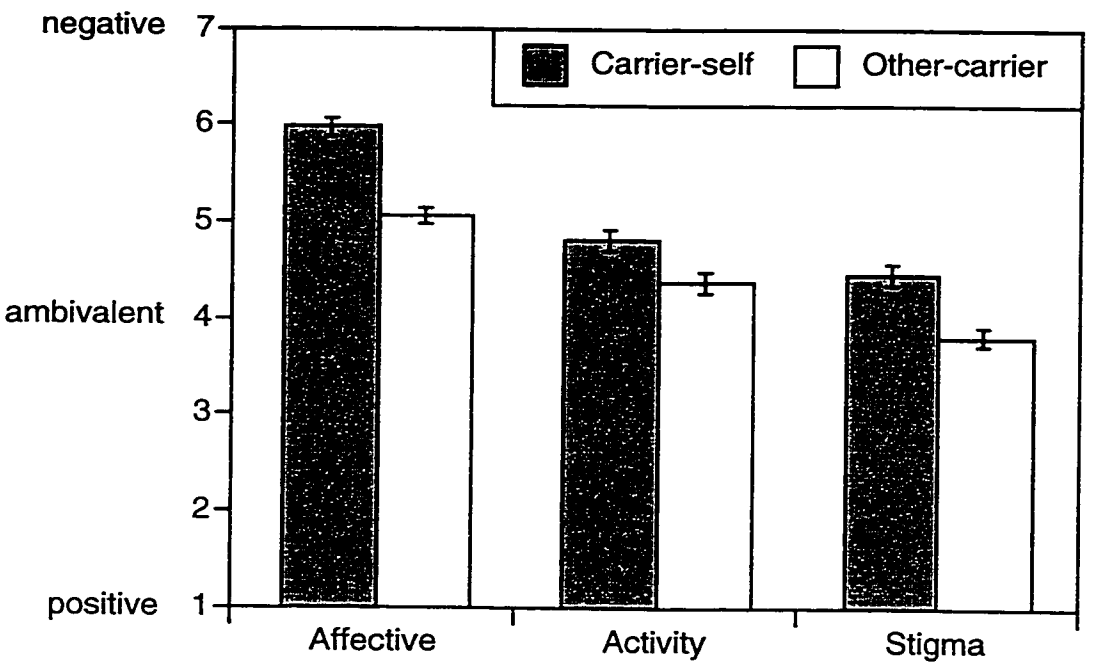

Figure 3. Means with Standard Error Bars for the Health Orientation Scale - Modified Subscales for the Carrier-self and Other-carrier Situations

Hypothesis D: CF vs. Carrier-self. To investigate to what degree individuals differentiated having CF and being a carrier, a 2 (HOS-M situation) x 3 (HOS-M subscales) repeated measures ANOVA was conducted on the mean subscale ratings for the CF and carrier-self situations. A Situation x Subscales interaction was found, F (2, 294) = 8.1, $p < .001$ (see Figure 4 for a graph of means and standard errors). The affective and activity subscales (Ms = 6.1 and 5.3, respectively) were significantly more negative than in the carrier-self situation (Ms = 6.0 and 4.8, respectively) for the CF situation. For the stigma subscale, however, the means were not significantly different (Ms = 4.6 for both situations).

The means for the affective subscale were found to be significantly different F (1, 152) = 10.47, $p = .001$, despite there being only a .1 mean difference between situations. There are two points that must be considered. First, the ratings for the affective subscale were created by averaging six of the bipolar word pairs, compared to four for the activity subscale and three for the stigma subscale. An increased number of measurement points should provide a better estimate of the population parameter and reduced variance. This, coupled with the large sample size (N = 153), and a within-subjects design reduced the standard errors of the means to .07 for each subscale and a pooled standard error of .055. This allowed for negligible differences between subscales to be significantly different.

In summary, it appears that participants rated both being a carrier for CF and having CF as equally negative experiences and similar in producing slight feelings of stigmatization. In addition, participants reported that having CF would have significantly more negative effect on health perception than being a carrier for CF.

Hypothesis E: Carrier-self vs. Stigmatizing conditions (AIDS and schizophrenia). This hypothesis compared an individual's reaction to being a carrier of a CF mutation to a stigmatizing medical condition (AIDS) and a stigmatizing mental disorder (schizophrenia). A 3 (HOS-M situation) x 3 (HOS-M subscales) repeated measures ANOVA was conducted

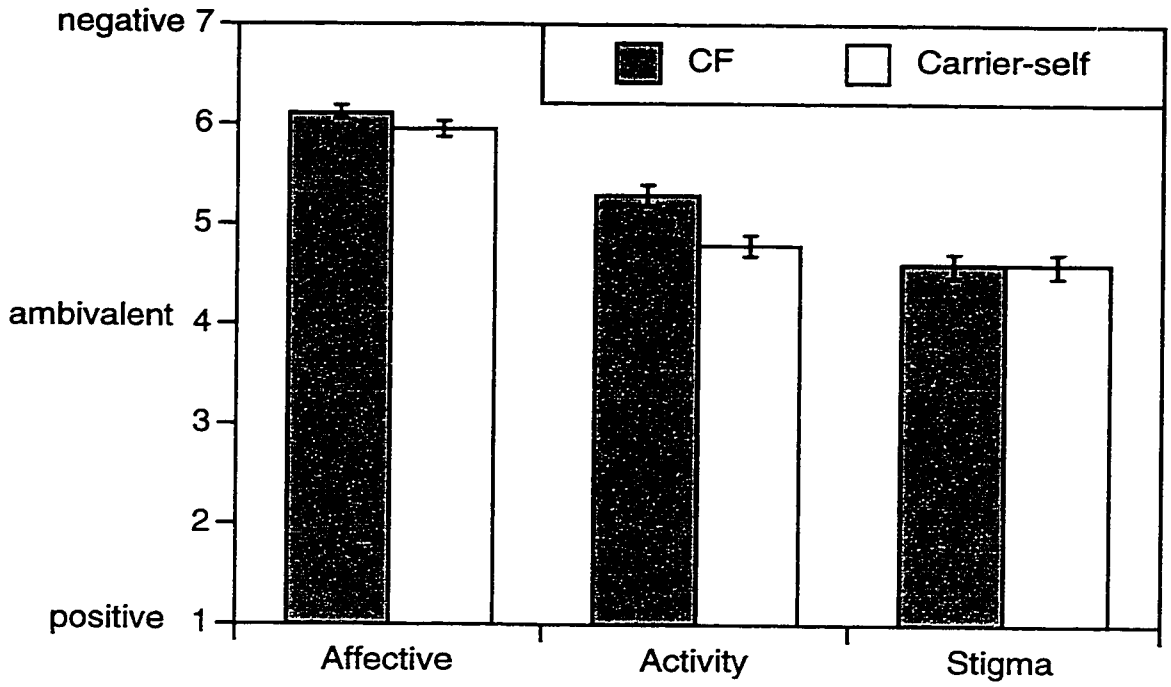

Figure 4. Means with Standard Error Bars for the Health Orientation Scale - Modified Subscales for the CF-diagnosis and Carrier-self Situations

on the mean subscale ratings. A significant Situation x Subscale interaction was found, $F(4, 476) = 11.12$, $p < .001$ (see Figure 5 for a graph of the means and standard errors). For the AIDS situation, all three subscales were significantly different from the carrier-self situation. For the schizophrenia situation, the affective and activity subscales (Ms = 6.0 and 5.2, respectively) were not significantly different from the carrier-self situation subscales (Ms = 6.0 for the affective subscale and 4.9 for the activity subscale). For the stigma subscale, the schizophrenia situation was significantly higher than in the carrier-self situation (Ms = 5.0 and 4.6, respectively).

This hypothesis attempted to compare one's reactions to stigmatizing conditions with being identified as a carrier. When carrier status is compared to a stigmatizing medical condition (AIDS), participants clearly rated AIDS as a more negative condition on all three subscales. AIDS is viewed as more emotionally detrimental, reduces perceptions of health, and produces more feels of stigmatization. Responses about being heterozygotic and having a stigmatizing mental disorder (schizophrenia) are not distinct. Both conditions produced high levels of negative affective feedback and reductions in views about health. However, schizophrenia was viewed as a more stigmatizing condition than CF carrier status.

Goal 2: Demographic and cognitive factors related to carrier status

A third set of analyses investigated the impact of a set of cognitive variables and a set of demographic factors on one's reaction to being a carrier for a CF mutation. The response variable was the sum of all 13 adjective pairs for the carrier-self situation. The demographic variables (and range of responses) included in the analyses were: (1) gender (female, male), (2) ethnicity (Asian, Caucasian, other), (3) religion (Christian, Buddhist, none, other), (4) frequency of attendance at religious services (frequent attendance, infrequent attendance, not practicing), and (5) number of children desired (0 - 5). Due to the limited range of ages in the present sample (90% of the sample was 18-21 years old),

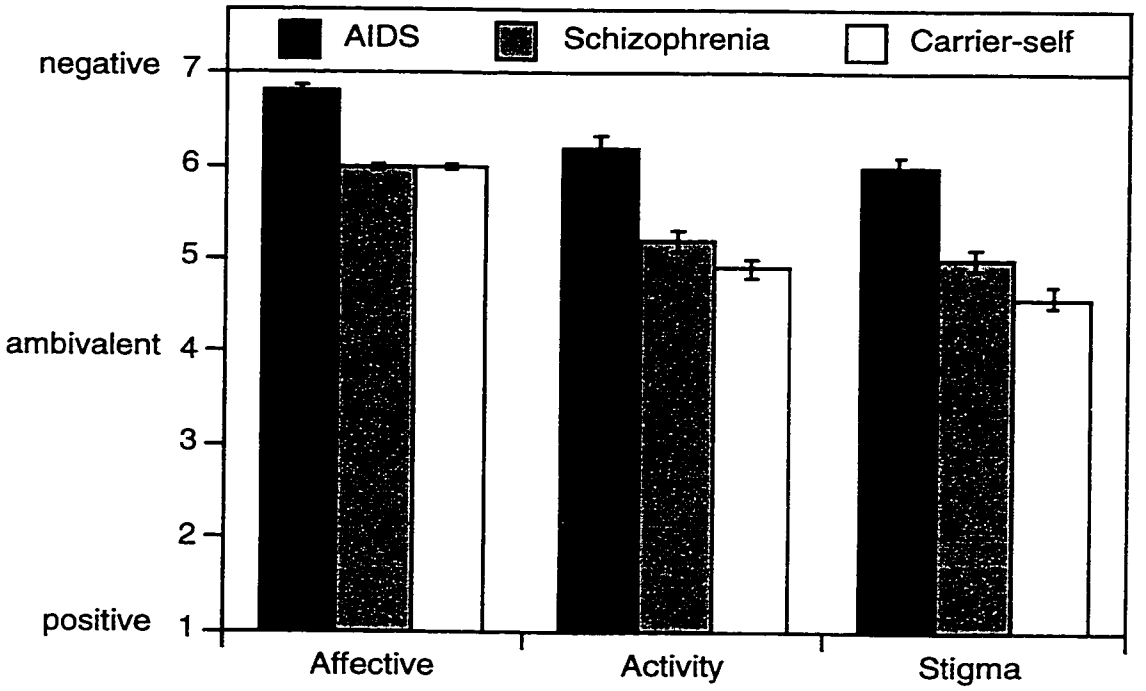

Figure 5. Means with Standard Error Bars for the Health Orientation Scale - Modified Subscales for the AIDS-diagnosis, Schizophrenia-diagnosis and Carrier-self Situations

age was not included as a variable in these analyses. The cognitive set of variables (and range of responses) included the following: (1) the number of sources of information about CF selected (ten specific sources plus an "other" option), (2) score from the attitude scale (20-50), and (3) score from test of genetic knowledge (0-18).

To assess the impact of each set of variables on the total scale score for the carrier-self situation, two separate hierarchical multiple regressions were conducted. The first entered the set of demographic variables into the analysis followed by the set of cognitive variables to assess the impact of cognitive variables after taking into account the demographic variables. The second multiple regression assessed the independent impact of cognitive variables and then assessed what additional information could be derived from the demographic variables. See Table 2 for descriptive statistics for each predictor.

In the first multiple regression, the demographic variables were entered first as a block. Neither the overall set of variables, $F(9, 138) = .84$, $p = .58$, nor any of the individual variables were able to predict significantly the scores on the carrier-self. Overall, only 5% of the variance of the carrier-self situation could be predicted by the demographic variables ($R = .23$). Next, the cognitive variables were entered as a block. An additional 2% of the variance in the scale scores could be accounted for when the set of cognitive variable were added, which was not a significant increase in R^2, ($F_{change} = .92$, $p = .43$). Overall, the combination of both demographic and cognitive variables did not significantly predict respondent's reactions to being identified as a carrier with a multiple R of .27 ($F(12, 135) = .86$, $p = .59$). No individual variable was a significant predictor of the total scale score for the carrier-self situation.

A second hierarchical multiple regression was conducted to assess the independent impact of the cognitive variables, followed then by the demographic variables. The cognitive were entered as a block, and did not significantly predict carrier-self scores, $F(3, 144) = 1.25$, $p = .29$. Overall, the cognitive variables only accounted for 3% of the

Table 2. Descriptive Statistics for Demographic and Cognitive Variables

Demographic Factors	Frequencies
Gender	
females	88 (59%)
males	60 (41%)
Ethnicity	
Asian	71 (48%)
Caucasian	34 (23%)
Other	43 (29%)
Religion	
Christian	91 (62%)
Buddhist	12 (8%)
None	30 (20%)
Other	15 (10%)
Frequency of Religious Practice	
Frequent	44 (30%)
Infrequent	72 (49%)
Not Practicing	32 (21%)
Number of Children Desired	median = 2 range = 0 - 5

Cognitive Variables	Descriptives
Attitude about genetic testing scale	mean = 34.7 SD = 6.1 range = 20 - 50
Genetic Knowledge test	mean = 8.4 SD = 2.4 range = 1-13
Number of sources of information about CF	mean = 1.9 SD = 1.7 range = 0 - 5

variance ($R = .16$). The demographic variables were entered next, and as a block accounted for an additional 4% of the variance in carrier-self scores. This was not a significant increase in R^2 ($F_{change} = .73$, $p = .68$).

Discussion

This study investigated the possible psychosocial changes related to being a hypothetical carrier for a CF mutation as well as demographic and cognitive factors that may influence how people interpret carrier status. Using three subscales developed from a modified version of the Health Orientation Scale, several negative psychosocial changes related to being a hypothetical carrier for a CF mutation were identified. Specifically, hypotheses regarding negative affective changes, reductions in health perception, and possible stigmatization were supported by the data. Unlike much of the previous work concerning attitudes about being heterozygotic for a recessive disorder, these data were sampled from a population that had not been exposed to the testing process. This population is important because they represent how individuals in the general population may react to the possession of a mutation in themselves and others.

Psychosocial changes related to carrier status

Affective consequences. The affective scale of the HOS-M consistently revealed large differences between situations representing negative emotional responses. When comparing one's present self-esteem with ratings of the self as a carrier and with feelings about disclosing such information, individual mean ratings increased two to three scale-points on a seven point measure, and were clearly in the negative range of the scale. This negative affective response is similar to previous studies which found the individuals tested for CF mutations often reported negative feelings immediately following testing (Axworthy et al., 1996; Childs et al., 1976; Evers-Kiebooms et al., 1994; Harris et al., 1996; Witt et al., 1996) and others have found changes in reported self-esteem following testing (Clow & Scriver, 1977; Mitchell et al., 1993).

It is important to note that many studies have found that negative affective reactions to being identified as a carrier are often ameliorated over time (Mennie et al., 1993; Zeesman et al., 1984) or following genetic counseling (Watson et al., 1992; Witt et al., 1996). Since this study took ratings at only one point in time, there are no data to investigate changes in affective response across time or following exposure to more information relating to the meaning of genetic carrier status. The data are important in that some trial screening programs have not provided post-test counseling, leaving the individual to deal with the strong negative response without knowledgeable, trained professionals to explain the meaning of the complex, genetic information.

Changes in perception of health. Being a carrier of a genetic mutation also seems to influence individuals' perception of their health. Ratings for the activity scale of the HOS-M showed large differences when comparing present health perception and projected health ratings as a carrier. Similar reductions in health status were found when participants thought about disclosing carrier status. Other studies have found similar reductions in health perceptions related to carrier status (Axworthy et al., 1996; Marteau et al., 1992; Hill, 1994). For example, Marteau and her colleagues (1992) found that people's interpretation of their present health status was significantly lower after being identified as a carrier.

The negative impact of carrier status on health perceptions also is supported by the poor differentiation found between actually being affected with CF as opposed to being a carrier of a CF mutation. The data showed that people, in general, do not draw very clear distinctions between the disease, which is presently fatal, and carrier status, which has no known direct health effects for the individual. Though significant differences were found for two of the three HOS-M scales (affective and activity scales), the largest difference between the two situations was .5 for the activity scale. It could be argued that, because of the extremely small standard errors, these small scales differences were statistically

significant. Conceptually, there seems to be little difference between individuals' ratings of having CF and being a carrier for it. This is similar to Hampton et al. (1974) who found that parents of children identified as being carriers of a sickle cell gene believed that the children had the disease or required a special diet because of their carrier status.

Presence of stigma. The present study also supports the presence of stigmatization associated with carrier status. The data showed a significant increase on the stigma scale when comparing present self-esteem and projected self-esteem as a carrier, indicating that self-esteem would be lowered if an individual discovered he/she was a carrier. In addition, an increase in stigmatization was found when comparing present self-esteem to feelings about disclosure of one's carrier status to friends. Goffman's (1974) theory on stigma predicted that people would be less willing to volunteer information that would point out differences between themselves and other that could be interpreted as negative. It seems that not only is carrier status interpreted as a negative state, but people interpret discussing such information as a negative experience.

The possibility of stigmatization associated with CF carrier status also was assessed in this study by comparing ratings of carrier status to known stigmatizing conditions (AIDS and schizophrenia). As seen in Figure 5, even though CF carrier status was seen as a negative experience for many people, it is not rated nearly so negative as being diagnosed with AIDS. AIDS clearly was viewed an extremely negative and stigmatizing condition with mean scale ratings of six or higher on the seven-point scale.

The differences between carrier status and schizophrenia, on the other hand, were not as clear. It appears that people would view being identified as a CF carrier similar to a diagnosis of schizophrenia on measures of emotional response and implication for health and activity. Both conditions were viewed as quite negative experiences. However, schizophrenia was viewed as a more stigmatizing condition than CF carrier status.

Significant difference also were found between people's rating of themselves as carriers and feelings they ascribe to others as carriers. For all three HOS-M scales, people rated their own experience of possibly being a carrier as more negative compared to how they thought others would feel as carriers. This is the opposite to what was found by Evers-Kiebooms et al. (1994) and Wooldridge and Murray (1988). Evers-Kiebooms and her colleagues found that carriers' self-descriptions were significantly more positive than feelings they projected for most carriers. Similarly, Wooldridge and Murray (1988) found that non-carriers ascribed more negative feelings to how carriers might feel compared to how identified carriers of a sickle cell mutation believe other carriers feel.

Though the ratings for the self-as-a-carrier were significantly higher than for other-as-carriers for all scales, it needs to be pointed out that these differences were less than one scale-point. An important point to consider from Figure 3 is that all means are at or above the neutral scale position (represented as a dashed line of the scale).

Comparison with previous research using the HOS. Overall, the data in this study suggest that being identified as a carrier is interpreted as largely a negative experience for many individuals in the general population. The three HOS-M situations in this study used to assess psychosocial reactions to carrier testing (carrier-self, self-disclosure, and carrier-other) all were rated as negative experiences. Using the HOS scaling categories described by Wooldridge and Murray (1988), the results of this study can be compared with the two other studies that have used the HOS methodology. When comparing total scale scores on comparable situations, the results from this study are similar to what was found by Evers-Kiebooms et al. (1994) and Wooldridge and Murray (1988), who used the HOS in their studies of individual's reactions to the genetic testing process. (See Table 3 for a comparison of the data from the three studies using the HOS.) It appears that tested non-carriers and untested individuals consistently rated that others would feel that being heterozygotic for a genetic mutation would be an unfavorable state. This consistency is

Table 3. Comparison of Health Orientation Scale Results from the Present Study, Wooldridge and Murray (1988), and Evers-Kiebooms et al. (1994)

HOS Situation	Present Study	Wooldridge & Murray (1988)	Evers-Kiebooms et al. (1994)
Present Self-Esteem	Untested*: Favorable**	Carriers: Favorable Non-carriers: Favorable	Carriers: Favorable Non-carriers: Very Favorable
Self-as-a-Carrier	Untested: Unfavorable	Carriers: Ambivalent Non-carriers: Ambivalent	
Self-disclosure	Untested: Unfavorable	Carriers: Ambivalent Non-carriers: Ambivalent	
Others-as-a-Carrier	Untested: Unfavorable	Carriers: Ambivalent Non-carriers: Unfavorable	Carriers: Ambivalent Non-carriers: Unfavorable
CF diagnosis	Untested: Unfavorable	Carriers: Unfavorable Non-carriers: Unfavorable	

* Present study studied individuals not exposed to the genetic testing procedure. Wooldridge and Murray (1998) and Evers-Kiebooms et al. (1994) studied individuals who had been tested for either sickle cell anemia or cystic fibrosis, respectively.

** Five categories were developed by Wooldridge and Murray (1988); very favorable, favorable, ambivalent, unfavorable, and very unfavorable. Categories for each HOS situation were determined by summing the scores for each bipolar adjective pair. Total scale scores in Wooldridge and Murray (1988) and Evers-Kiebooms et al. (1994) ranged from 12-60 consisting of 12 adjective pairs using a 5-point likert type scale. Total scale scores for the present ranged from 13-91 consisting of 13 adjective pairs using a 7-point likert type scale.

important because of the differences that were present in the nature of the studies. For example, the three studies covered both tested (Wooldridge & Murray, 1988, Evers-Kiebooms et al., 1994) and untested populations (this study); different recessive disorders, CF (Evers-Kiebooms et al., 1994; this study) and sickle cell anemia (Wooldridge & Murray, 1988); and studied groups in different countries, United States (this study; Wooldridge & Murray, 1988) and Denmark (Evers-Kiebooms et al., 1994).

Demographic and cognitive factors related to carrier status

This study found no impact of either demographic or cognitive factors on an individual's reaction to being heterozygotic. This is not too surprising given the mixed evidence found in previous research investigating demographic factors.

Genetic knowledge and carrier status. Surprisingly, one's level of basic genetic knowledge and attitude about genetic testing also were not related to the HOS-M situation. It was predicted that increased understanding of basic genetics would be related to more positive views about being heterozygotic for CF. However, no relationship between level of genetic knowledge and the meaning of being a carrier was found.

Further investigation of responses to the genetic knowledge scale provided some important insights into this population. Overall, participants had a somewhat low level of understanding. The participants correctly answered an average of 66% of the questions. Questions related to autosomal recessive disorders like CF were correctly answered understood by many. For example, 44% correctly answered a question on recessive transmission (some genetic diseases require that both parents pass on a copy of the gene) and 67% correctly a question regarding the effects of recessive genes on the carriers health (you can have a disease related a gene and still be healthy) of this sample. Several other facts important to understand the genetic testing process were not well understood. For example, (1) most cases of genetic disease are **not** due to a new mutation in the affected individual (31% correct response); (2) the definition of a gene as a chemical unit of

hereditary information (46% correct response); and (3) that a genetic disorder can occur almost exclusively in a selected population (60% correct response). The low level of knowledge found in this sample is similar to other studies of individuals undergoing genetic testing (Jung et al., 1994; Nance et al., 1993; Watson et al., 1991) as well as in studies of groups apart from genetic services (Decruyenare et al., 1992; Denayer, De Boeck et al., 1992; Magnay, Wilson, El Hait, Balhamar, & Burn, 1992; Massarik & Kaback, 1981; Nance, Sevenich, & Schut, 1994).

The appropriate use of genetic services in the general population requires that individuals understand the procedure they are undergoing and why it is important. The level of incorrect information described should raise concerns since many genetic testing programs are focused on finding members of certain populations that carry the common mutations for these genetic diseases. For example, screening programs have been conducted to identify Tay Sachs carriers in predominantly Jewish population, sickle cell carriers in African-American population, and CF carriers in the Caucasian population.

Another disturbing trend that has been identified in the literature is that the medical professionals generally identified as handling the genetic testing services (mainly primary care physicians and obstetricians/gynecologists) have been found to have limited knowledge about genetic principles important to providing genetic testing services (Hoffman et al., 1993; Kershner, Hammond, & Donnenfeld, 1993; Rowley, Levenkron, Loader, & Phelps, 1993; Thies, Bockel, & Bochdalofsky, 1993). In a study of primary care physicians knowledge about CF and CF testing, Rowley et al. (1993) conclude "the aggressive marketing of carrier testing by commercial firms could result in many individuals being tested, yet receiving inadequate pretest education and posttest counseling. In this setting, population screening may burden the individual with misunderstanding, impairment of self-image, and perceived unsuitability as a partner and as a parent" (pp. 265). The lack of appropriate levels of genetic information on the part of both the

providers and consumers of genetic testing services is a concern as population based screening programs begin to be implemented.

Attitudes about genetic testing and carrier status. It also was expected that individuals' attitudes about genetic testing in general would be related to their affective response to being a carrier. As with basic genetic knowledge, this hypothesis was not supported. Further investigation of attitudes about genetic testing in general showed that individuals felt that genetic testing should be available and provided to those who wish it, especially in relation to family planning decisions and as information during early pregnancy.

One important finding is that approximately three-fourths of all individuals preferred that genetic testing to determine a couple's risk of producing a child with a serious genetic defect should be offered to couples prior to pregnancy for both genetic defects that were not preventable/treatable and for defects that could be identified through fetal diagnosis. This is important information, since many of the trial screening programs conducted in the United States and in Europe were done through maternity clinics when the couples were already pregnant. Also, National Institutes of Health Consensus Development Conference (1997) has recently recommended that the best time for screening is during the prenatal period. Testing during pregnancy could lead to an exaggeration of the negative consequences identified in this and other studies (e.g., anxiety, depression and reduction in health perception).

Limitations of study

There are several limitations that need to be recognized. Of primary importance is the fact that the sample represented a very narrow group -- young, college students who have not yet begun to raise families. Feelings associated with genetic carrier status may vary as people get older and start raising families and after they have completed having children. Also to be noted is the fact that this study primarily measured individuals'

attitudes and responses to hypothetical situations. Many studies have shown low correlations between a person's attitude about a subject and their behavior (LaPiere, 1934; see Wicker, 1969 for a further review). Within the field of genetic testing, many studies have found poor correspondence between a person's stated willingness to undergo genetic testing and their actual willingness to undergo genetic testing(National Institutes of Health Consensus Development Conference, 1997). The data from the present study, however, do provide insight into the thoughts of how untested individuals may interpret heterozygotic carrier status in their family, friends, and society at large.

Future Research

Further research on the possible stigmatization related to CF carrier status needs to be done. More in-depth projects need to be conducted to try to understand the subtle nature of people's feelings about being heterozygotic and the effects on perceived health. Most of the research conducted in actual CF testing programs used measures of anxiety and depression or single questions about changes in self image to assess stigmatization following screening. The HOS methodology has shown to be easily adapted to different research settings as well as easily used by research participants.

Studies on the effects of people's negative interpretation of CF carrier status in social interactions also needs to be assessed. One concern with the full-scale implementation of carrier screening programs has been the possibility of discrimination based on positive carrier status. How does an individual's negative reaction to someone else being a carrier affect their interaction? How does the stigmatized individual manage the social tension created by having to hide discrediting information?

Also important to assess is the impact that one's culture has on connotative meaning of heterozygotic status. The views that one's predominant culture has about health and reproduction have been shown to influence an individual's uptake of genetic services (Punales-Morejon & Penchaszadeh 1992), and his/her ability to use that information in

child-bearing decisions (Hill, 1994). The present study used broad ethnic categories to assess the impact of ethnicity on feelings about carrier status. Future research should focus on a particular culture that individuals identify as their own and that culture's understanding of health and illness rather than ethnic category alone.

Implications and Conclusions

This study suggests that being a carrier of a CF mutation is a negative experience, perhaps producing reductions in an individual's view of their health, and possibly leading to some feelings of stigmatization. These data help to shed light on possible reactions to the introduction to population-based screening programs for CF and other recessive disorders. Many pilot programs testing individuals without a family history of CF have been conducted, and recently the National Institutes of Health Consensus Development Conference (1997) issued a document describing requirements for such CF screening programs. The results of this and other studies finding psychosocial consequences associated with being heterozygotic for a recessive gene point to the need for both greater education for genetic principles within the general population and the absolute importance of genetic counseling within the genetic testing cycle.

The results of the study suggest that there is a need for increased genetic education within the general population. A moderate to poor understanding of genetic principles related to understanding of recessive disorders and poor differentiation between CF and being a heterozygotic carrier for the disorder were found in this sample of college students. Given the major advances that continue to be made in science and technology, it seems unlikely that we can assume that high school biology courses will prepare individuals to live and interact in a world of proliferating genetic tests. Community/culture-based education may need to be developed to aid groups in understanding test results within the group's unique understanding of health and illness.

Finally, given the inevitable introduction of widespread genetic testing programs coupled with poor understanding of genetic principles, genetic counseling must remain an integral part of the genetic testing process. Genetic counseling may be required both before the testing process to ensure that those who choose to undergo testing understand the process and it's limitations, and to help individuals understand test results and alleviate any concerns raised during the testing procedure. Genetic counseling has been shown to reduce anxiety following positive test results. It is expected that genetic counselors also could be effective in lessening the negative health interpretation and stigmatization suggested by this study and previous research.

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Appendix A. Signed Approval Forms

SJSU SAN JOSE STATE UNIVERSITY

A campus of The California State University

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One Washington Square • San José, California 95192-0025 • 408/924-2480

TO: Margaret-Anne Mackintosh
408 Casa View Dr.
San Jose, CA 95129

FROM: Serena W. Stanford Serena W. Stanford
AAVP, Graduate Studies & Research

DATE: May 2, 1996

The Human Subjects-Institutional Review Board has approved your request to use human subjects in the study entitled:

"Attitudes Concerning Carrier Status in the General Population"

This approval is contingent upon the subjects participating in your research project being appropriately protected from risk. This includes the protection of the anonymity of the subjects' identity when they participate in your research project, and with regard to any and all data that may be collected from the subjects. The Board's approval includes continued monitoring of your research by the Board to assure that the subjects are being adequately and properly protected from such risks. If at any time a subject becomes injured or complains of injury, you must notify Serena Stanford, Ph.D., immediately. Injury includes but is not limited to bodily harm, psychological trauma and release of potentially damaging personal information.

Please also be advised that all subjects need to be fully informed and aware that their participation in your research project is voluntary, and that he or she may withdraw from the project at any time. Further, a subject's participation, refusal to participate, or withdrawal will not affect any services the subject is receiving or will receive at the institution in which the research is being conducted.

If you have any questions, please contact me at (408) 924-2480.

Appendix B. Health Orientation Scale - Modified

Health Orientation Scale

Below is a list of items which get at your feelings about cystic fibrosis and some other health concerns. *There are no wrong or right answers,* only information about how you feel. Please complete all the items. Give your first response and do not go back over items already answered. Indicate your feelings by placing a mark on the line closest to the word which best describes your feelings.

I am in charge of 50 people who do different kinds of jobs. I learn that several of my employees are carriers of the cystic fibrosis trait. I imagine that the person carrying the gene for CF might feel:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

My doctor has just told me that I am a cystic fibrosis carrier, I feel:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

----------*----------

I have the cystic fibrosis trait. Over a game of cards, there is a conversation about cystic fibrosis. As I consider whether or not to mention that I have the cystic fibrosis gene. I feel:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

My doctor has just told me that I have cystic fibrosis. I feel:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

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My doctor has just told me that I have schizophrenia. I feel:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

My doctor has just told me that I have AIDS. I feel:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

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The following terms best describe me in general:

Bad	____ : ____ : ____ : ____ : ____ : ____ :____	Good
Afraid	____ : ____ : ____ : ____ : ____ : ____ :____	Unafraid
Not guilty	____ : ____ : ____ : ____ : ____ : ____ :____	Guilty
Ashamed	____ : ____ : ____ : ____ : ____ : ____ :____	Unashamed
Strong	____ : ____ : ____ : ____ : ____ : ____ :____	Weak
Shocked	____ : ____ : ____ : ____ : ____ : ____ :____	Relieved
Sad	____ : ____ : ____ : ____ : ____ : ____ :____	Happy
Unmarked	____ : ____ : ____ : ____ : ____ : ____ :____	Marked
Unable	____ : ____ : ____ : ____ : ____ : ____ :____	Able
Pleased	____ : ____ : ____ : ____ : ____ : ____ :____	Angry
Inactive	____ : ____ : ____ : ____ : ____ : ____ :____	Active
Sick	____ : ____ : ____ : ____ : ____ : ____ :____	Healthy
Relaxed	____ : ____ : ____ : ____ : ____ : ____ :____	Anxious

APPENDIX C. Attitudes toward Genetic Testing Scale

For each question select the response that best describes you and your behaviors. Select from among the following response alternatives:

Strongly **A**gree **A**gree **N**eutral **D**isagree **S**trongly **D**isagree

Circle the letter that corresponds to your choice.

1. Gene testing should be obligatory for everybody before fertility age:
   SA A N D SD
2. Gene testing should be performed on newborn babies:
   SA A N D SD
3. Gene testing should be available to anybody who wishes to have information about disease genes he/she carries:
   SA A N D SD
4. Gene testing should not be performed at all:
   SA A N D SD
5. Individuals should be offered a gene test when choosing a spouse:
   SA A N D SD
6. Individuals should be offered a gene test in family planning:
   SA A N D SD
7. All pregnant women should be offered a gene test:
   SA A N D SD
8. Society would save on the costs of treatment of diseases if gene testing was available
   SA A N D SD
9. People have the right to know about their genes to be able to affect their own health and lives:
   SA A N D SD
10. People have the right to know about their genes to be able to affect the health and life of their offspring:
    SA A N D SD
11. There are bigger public health problems that should be taken care of first:
    SA A N D SD

Strongly Agree **Agree** **Neutral** **Disagree** **Strongly Disagree**

12. The natural order should be respected without interference:
    SA A N D SD
13. Knowledge of test results may lead to discrimination against disease gene carriers (e.g., in employment and insurance policies)
    SA A N D SD
14. Testing would make abortions more common.
    SA A N D SD

APPENDIX D. Test of Basic Genetic Knowledge

Please answer the following questions by placing a mark on the line next to your responses.

1. While illness can have many causes (infection, trauma, malignancy), certain diseases are due almost entirely to inherited factors. What percentage of all illnesses do you think are predominantly due to inherited factors (genetic causes)? (Select only one answer)

_____ Less than 1%	_____ 25%
_____ 5%	_____ 50%
_____ 10%	_____ 70% or more

2. Is an "inherited disorder" the same as a "congenital disorder"?

_____ Yes _____ No _____ Don't know

3. A "gene" is best defined as (check one):

The chromosome	_____
A chemical unit of hereditary information	_____
A group of cells with hereditary information	_____
The egg and the sperm	_____
Don't know	_____

4. Respond to each of the following statements concerning genetic diseases:

A. Certain genetic diseases can skip generations:

_____ Yes _____ No _____ Don't know

B. Certain genetic diseases can be inherited from *one* parent only

_____ Yes _____ No _____ Don't know

C. Certain genetic diseases require inheritance of the disease gene from *both* parents:

_____ Yes _____ No _____ Don't know

D. Most cases of genetic disease are due to a new mutation in the affected individual:

_____ Yes _____ No _____ Don't know

5. Do you believe that genetic disease can be cured?

_____ Always _____ Sometimes _____ Never _____ Don't know

6. Do you believe that genetic disease can be treated?

_____ Always _____ Sometimes _____ Never _____ Don't know

7. Do you believe that genetic diseases can be prevented?

_____ Always _____ Sometimes _____ Never _____ Don't know

8. Do you believe that genetic diseases can be predicted?

_____ Always _____ Sometimes _____ Never _____ Don't know

9. Can a person carry a disease-related gene and still be perfectly healthy?

_____ Always _____ Sometimes _____ Never _____ Don't know

10. Can a genetic disorder occur almost exclusively in males?

_____ Yes _____ No _____ Don't know

11. Can a genetic disorder occur almost exclusively in females?

_____ Yes _____ No _____ Don't know

12. Can a genetic disorder occur almost exclusively in a selected population (racial, ethnic, or national group)?

_____ Yes _____ No _____ Don't know

13. How often is a genetic disease likely to occur in a selected population?

_____ Frequently _____ Occasionally _____ Very rarely

14. If a couple is told that they have a 25% risk for having children with a genetic disease this would mean:

A. that none of their children would be affected

_____ Yes _____ No _____ Don't know

B. that each child would be 25% affected

_____ Yes _____ No _____ Don't know

C. that if the first child was affected, the next three would be entirely unaffected

_____ Yes _____ No _____ Don't know

D. that each pregnancy would have a one in four chance of resulting in an affected child

_____ Yes _____ No _____ Don't know

APPENDIX E. Demographics Questionnaire

1. Age: _____

2. Gender: _____ male _____ female

3. Ethnicity: _____

4. Marital status: _____Single _____Married _____Divorced _____Separated _____Widowed

5. Education level: _____ freshman _____ sophomore _____ junior _____ senior _____ graduate student

6. Which religion do you presently practice, if any?

_____

If you attend a religious service, how frequently do you attend?

___ once a week or more _____ 1 or 2 times a month ___ a few times a year ___ once a year

7. How many children do you presently have? _____

8. How many children would you like to have? _____

9. Prior to this study, had you ever heard of cystic fibrosis?

_____ Never _____ Something _____ A lot

10. Have you ever known a family where cystic fibrosis has occurred?

_____ Yes _____ No _____ Don't know

11. Has cystic fibrosis ever occurred in your family?

_____ Yes _____ No _____ Don't know

12. If you have heard of cystic fibrosis, where did you obtain your information (check as many as apply)?

Friends	_____	Relatives	_____
Magazine	_____	School	_____
Newspaper	_____	Scientific Literature	_____
Physician	_____	Television	_____
Clergy	_____	Other	_____
Radio	_____		

13. Have you ever heard about a testing program for cystic fibrosis prevention?

_____ Yes _____ No

14. Has any genetic disorder occurred in your family?

_____ Yes _____ No _____ Don't know

APPENDIX F. Introductory Information about Cystic Fibrosis

CYSTIC FIBROSIS

Cystic fibrosis (CF) is the number one fatal inherited disease among young people in the United States. At present, only half of those with CF survive to age 30. It primarily affects the lungs and digestive system. Approximately one baby in every two thousand is born with CF. It occurs in males and females alike, and mainly among Caucasians.

In most people, mucus is thin and slippery, and it helps to clean the dust and germs from the lungs and breathing passages. A person with CF produces abnormal amounts of thick, sticky mucus that tends to clog the air passages in the lungs and interfere with breathing. Mucus may also block the ducts of the pancreas, preventing digestive juices from reaching the intestines. As a result, food passes through the body without being broken down and absorbed, and the patient doesn't get proper nourishment.

Since CF is a genetic disease, it is always present at birth, but it may not develop for months or even years later. Some of the symptoms are common to nearly all patients. Others with CF have fewer symptoms of the disease. This puzzles researchers who wonder why people are affected differently.

WHAT ARE THE SYMPTOMS OF CF?

Generally there are three major symptoms of this disease: respiratory problems; digestive problems, which occur in 85% of the patients; and high amounts of salt in the sweat. Most CF patients develop lung disease at some time in their lives. The thick mucus produced in CF clogs the airways of the lung and respiratory system, interfering with normal breathing, and harboring infections, which eventually damage lung tissue. Respiratory complications are the main cause of death of patients with CF. These patients are also susceptible to lung infections that are resistant to certain kinds of antibiotics.

The pancreas and other organs of the digestive system secrete enzymes into the intestine where they help to break down food into elements that the body needs for energy, growth, and maintenance. In CF, thick, sticky mucus blocks the passageways that carry these enzymes to the intestine. This causes problems with digestion, and the food that is not broken down cannot be used by the body, so it is excreted.

CF affects the sweat glands so that there is a large amount of salt in the perspiration of CF patients, especially when sweating is increased as during exercise, in hot weather, or with a fever. The excessive salt loss can cause heat exhaustion or dehydration.

WHAT IS THE TREATMENT FOR CYSTIC FIBROSIS?

Because there is no cure, the aim of treatment is to help the person with CF live as normal a life as possible. Because the disease affects each patient in a different way, individual treatment is necessary. Treatment generally includes bronchial drainage, a form of chest physical therapy that helps loosen mucus from the lungs and keep lung passages open, antibiotic therapy to help fight respiratory infections, and dietary management.

To combat pancreatic enzyme deficiencies, the treatment includes oral pancreatic enzymes with meals and snacks. This replaces enzymes blocked in the pancreatic ducts. Children with CF generally need to eat more because their bodies require more energy. Other dietary measures include generous salting of foods, and salt supplements in hot weather to combat sweat loss. A balanced, highly nutritious diet is very important, and generally vitamin supplements are prescribed.

Some CF patients take as many as forty to sixty pills a day. The individual cost of CF treatment and care can range from six thousand dollars to twelve thousand dollars per year, and sometimes higher if there are complications that require frequent hospitalizations.

HOW IS CYSTIC FIBROSIS TRANSMITTED?

CF is a hereditary disease caused by a defective or abnormal gene on chromosome 7. It is estimated that one in every twenty Americans is a carrier of this defective gene. Inheriting a single gene for CF doesn't cause a problem, though, and often people don't even know they are carriers until they become parents of a child with CF.

The child with cystic fibrosis has inherited a pair of recessive CF genes. One gene from the pair is from the father and one from the mother. Just because both parents are carriers, it doesn't mean that all their children will have CF. There is a 25% chance with each pregnancy that their child will be unaffected, a 50% chance that their child will be a carrier, and a 25% chance that their child will have the disease. So this means that there is a 75% chance that their child will not have CF. The chances are the same for each pregnancy, even if there is a child in the family who already has CF.

Researchers have identified the gene responsible for CF, and have developed safe, effective tests for detecting the CF gene. This allows doctors to identify carriers of CF who have no outward symptoms of the disease. It can also be used to tell a pregnant woman, who has already had a child with CF, whether her developing baby is affected as well.