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**PREDICTIVE GENETIC TESTING FOR CYSTIC
FIBROSIS CARRIER STATUS:
THE ROLE OF HEALTH BELIEFS AND
COPING STYLE**

by
Barbara Virley O'Connor, B.A.

A thesis submitted to
the Faculty of Graduate Studies and Research
in partial fulfilment of
the requirements for the degree of

Master of Arts

Department of Psychology

RUNNING HEAD: Genetic Testing for CF Carrier Status

Carleton University
Ottawa, Ontario
(September 12, 1996)
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Genetic Testing for CF Carrier Status

The undersigned recommend to the Faculty of
Graduate Studies and Research
acceptance of the thesis:

Predictive Genetic Testing for Cystic Fibrosis Carrier Status:

The Role of Health Beliefs and Coping Style

Submitted by Barbara Virley O'Connor
in partial fulfilment of the degree Master of Arts

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Chair, Department of Psychology

A handwritten signature in dark ink, appearing to read 'Maria Capelli', written over a horizontal line.

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September 12, 1996

Genetic Testing for CF Carrier Status

Abstract

In an attempt to explore health beliefs and coping style in relation to CF carrier screening decisions, the Health Belief Model and the Monitor/Blunter dimension were used as guiding frameworks. The objectives of the study were threefold: 1) to examine health beliefs as they relate to the intent to be tested for CF carrier status, 2) to examine the efficacy of the Monitor/Blunter theory as it relates to the intent to be tested for CF carrier status, and 3) to consider the relationship between characteristic coping style(i.e., monitoring, blunting) and health beliefs. Using an analogue survey design, information about health beliefs, personal coping style and demographic variables were collected from a sample of general population individuals. Results showed that decreased perceptions of carrier status severity, increased perceptions of susceptibility to carrier status and barriers to the testing procedure predicted intent to use genetic testing. Further, an inverse relationship between age and intent to be tested was observed. Coping style was not related to any of the HBM variables or to the intent to be tested for CF carrier status. It is proposed that further exploration of individual health beliefs surrounding CF carrier testing may be useful to facilitate effective decision making in counselling situations. However, identification of personal coping style may be more relevant later in the genetic testing process, during times of increased threat, and/or in situations where one's own health may be affected.

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Predictive Genetic Testing for Cystic Fibrosis Carrier Status:

The Role of Health Beliefs and Coping Style

With the advent of DNA predictive testing, unparalleled options in health care have exploded onto the medical scene. While the traditional medical model has relied on methods and interventions to diagnose and treat already ill individuals, predictive testing for some diseases affords the option for diagnosis prior to disease onset and symptom presentation in currently well individuals. In this way, lifestyle changes and preventive therapies may markedly decrease morbidity. Information from carrier testing may also assist prospective parents in making reproductive choices which may include avoidance of pregnancy, surrogacy, artificial insemination and/or prenatal testing. Cystic Fibrosis (CF) is unique in that the full range of options from gene identification to gene therapy is almost complete (Surh et al., 1995). As such, CF may provide a future model for predictive testing for other genetically-based diseases such as cancer and diabetes.

Understanding who benefits from genetic information is an important first step in maximizing the potential of predictive screening to assist individuals in making more informed reproductive decisions. In an applied setting, this information might be implemented in tailoring counselling protocols to match individual needs, thereby maximizing understanding and effective use of test results, while minimizing psychological impact. Yet, while there has been a wealth of research surrounding factors associated with uptake rates and the psychosocial impact of testing, there is a gap in existing

literature surrounding the specific personality variables and health beliefs which may affect genetic testing decisions. In essence, while past work has explored what individuals *take away* from genetic testing situations, what they *bring* to these situations has been largely neglected.

The Health Belief Model (HBM; Rosenstock, 1974) has previously been cited as a theoretical framework for understanding individual health beliefs and motivation in relation to genetic counselling decisions (Pearn, 1973; Rosenstock, 1979). In particular, the HBM may be useful in explaining individuals' intentions regarding genetic testing issues, given that these decisions involve a novel, singular (vs. habitual) action, and are not subject to a prescribed recommendation by professionals (Calnan, 1984; Sagi, Shiloh, & Cohen, 1992). Therefore, these conditions enhance the probability that decisions will be made with vigilance, based on relevant cognitions (Sagi et al., 1992).

A dimension of personal coping style which may also be relevant to genetic testing relates to individual preference for information under threat, as embodied by the Monitor/Blunter theory (Miller, 1987; Miller, Brody, & Summerton, 1988). Within a genetic testing framework, individual preference for information may mitigate the decision to be tested. For example, while some individuals may prefer not to know their carrier status, others may actually feel uncomfortable not knowing their carrier status, given the opportunity to be tested.

Thus, the primary objectives of the proposed study were threefold: 1) to examine health beliefs as they relate to the intent to be tested for CF carrier status, 2) to examine the efficacy of the Monitor/Blunter as a predictive framework, within a genetic testing context, and 3) to consider the relationship between characteristic coping styles and health beliefs. In addition, the impact of demographic variables on individual intent to be tested for CF carrier status was also considered. Following a brief overview of the literature to date surrounding CF carrier testing research, the HBM and the Monitor/Blunter theory, the rationale and methodology for the current study will be presented.

Background

Cystic Fibrosis - The Disease

CF is the most common and potentially severe autosomal recessive genetic disorder among Caucasian populations. Primarily a disease of the respiratory and digestive systems, CF shortens the lives of about 1 in 2927 Canadians (Sturgess, Czegledy-Nagy, & Corey, 1985), and it is estimated that about 1 in 25 Caucasians are asymptomatic carriers of the recessive gene mutation which causes CF (Boat & Welsh, 1989; Wilfond & Fost, 1990). Although the expression of CF is unique to each individual, patients typically suffer from a variety of symptoms which may include frequent lung infections and subsequent damage, intestinal malabsorption and vitamin and mineral deficiencies, intestinal obstruction, biliary cirrhosis, male infertility and reduced female fertility (Boat & Welsh, 1989). In addition, complications of the disease may include

pneumothorax, haemoptysis, diabetes mellitus and intussusception (Boat & Welsh, 1989). In conjunction with the observance of chronic pancreatic and/or pulmonary insufficiency, diagnosis of the disease is often made through the use of a "sweat test", which determines abnormally high levels of sweat chloride in CF patients (3-4 times higher than normal levels) (Belle White, Carvalho, Derse, O'Brien, & Dean, 1992; The Cystic Fibrosis Foundation Centre Committee and Guidelines Subcommittee, 1990).

The current average life expectancy for an individual with CF is approximately 28 years of age (Boat & Welsh, 1989; Fitzsimmons, 1993), and represents a significant improvement in prognosis over the last two decades (British Paediatric Working Party on Cystic Fibrosis, 1988). This improvement in outlook has been largely attributed to the development of comprehensive and multidisciplinary treatment protocols, including antibiotic regimens, nutritional treatment and chest physiotherapy (Canadian Task Force on the Periodic Health Examination, 1991; Dodge, 1991).

In addition to representing a significant physical and psychologic burden to CF patients and their families, the frequent hospitalizations and treatment costs associated with the disease pose a severe financial burden for afflicted individuals, their families and the health-care system at large. For example, a recent Canadian study estimates that the yearly cost of coping with CF (e.g., drugs, hospitalization, physician fees, social support fees) is an estimated \$75 000 in U.S. funds (Surh et al., 1992).

Cloning the CF Gene

In 1989, a three-base-pair deletion ($\Delta F508$) in the putative structural gene for CF was located on the long arm of chromosome 7 (Kerem et al., 1990; Riordan et al., 1989; Rommens et al., 1989), and was identified as the mutation responsible for an estimated 68% of CF chromosomes (Kerem et al., 1990; Riordan et al., 1989; Rommens et al., 1989; Ten Kate, 1989). Subsequent work has noted considerable ethnic difference in the incidence of CF mutations, with the frequency of the mutation estimated to be as large as 75.8% in North American Caucasian populations, and as low as 49%, 43% and 30% in Spanish, Italian and Ashkenazic Jewish populations, respectively (Estivill et al., 1989; Lemna, Feldman, & Kerem, 1990). Further, since the identification of the $\Delta F508$ mutation, at least 98 additional CF mutations have been identified (Beaudet, 1992). As of 1991, most DNA diagnostic laboratories tested for approximately 6 or 7 mutations, which together account for an estimated 85% of all CF chromosomes (Beaudet, 1992).

Prior to the localization of the CF chromosome mutations, the only methods of carrier detection were a) giving birth to a child with CF or, b) estimating carrier risk via degree of relation to an affected individual. With the ability to identify carrier status at hand, the door has been opened for prospective parents to make more informed reproductive choices that may include having no children, adopting, using in vitro fertilization, surrogacy, and/or prenatal testing.

The enthusiasm of this new technology, however, has been shadowed by debate

about to whom, when and where testing should be offered in order to maximize limited health care dollars, and minimize any potentially aversive psychosocial effects of screening (Beaudet, 1990; Beaudet, 1992; McGourty, 1989; Modell, 1990; Surh, Cappelli, MacDonald, Mettler, & Dales, 1994; Surh et al., 1995; Wilfond & Fost, 1990). More precisely, dissension continues surrounding specific questions such as: whether testing should be offered to the general public while the known number of mutations is only 85-90% (Beaudet, 1992; Biesecker, Bowles-Biesecker, Collins, Kaback, & Wilfond, 1992; Caskey, Kaback, & Beaudet, 1990; Cutting et al., 1992; Ober et al., 1991), when the optimum time for carrier testing is (e.g., before pregnancy, during pregnancy), and whether social discrimination as a result of carrier identification will result, as has been documented previously for other disease carrier states which are asymptomatic (c.f., Billings et al., 1992; Kenen & Schmidt, 1978; Wilfond & Fost, 1990). Indeed, the controversy among professionals surrounding the uses of predictive testing is confirmed by a recent survey of physicians and genetics professionals in the United States which reported little support for routinely offering CF carrier testing with a (then current) detection rate of 80% (Faden et al., 1994). By contrast, a survey of health care professionals in Britain cited that 75% supported the introduction of a widespread CF carrier screening program (Watson, Williamson, & Chapple, 1991).

At the present time, governing medical groups and agencies in North America recommend testing for only those individuals who have a known family history of CF until

a 95% detection rate and more affordable tests are realized (Canadian Task Force on the Periodic Health Examination, 1991; Caskey, Kaback, & Beaudet, 1990; National Institutes of Health Workshop on Population Screening for the Cystic Fibrosis Gene, 1990). In the meantime, pilot testing has been initiated around the world, to address key issues such as factors associated with screening and reproductive decisions, and the potential psychosocial costs of carrier screening.

Factors Related to Screening Decisions

Screening During Pregnancy

Several pilot studies considering the factors which influence uptake rates have focused on pregnant women and their partners. By drawing upon the fact that most women seek prenatal care at some time during their pregnancy, these studies have regarded antenatal clinics, hospitals and private offices as "turnstiles" where the offer of screening can be made (Brock, 1995).

One dimension of screening during pregnancy which has been scrutinized is the method of presentation. "Step-wise" or "sequential" testing (i.e., testing the woman first, then her partner if she is a carrier) has been one of the most widely observed models. In perhaps the largest trial of sequential screening, Mennie et al. (1992) studied over 3000 pregnant women participating in a prenatal screening program. The 111 women identified as CF carriers subsequently had their partners tested, and the 4 couples identified as heterozygous were offered prenatal screening. The overall uptake rate for Mennie et al.'s

model of two-step delivery was 73%.

In addition to their preliminary work, Mennie, Gilfillan, Compton, Liston & Brock (1993) explored the reasons why women attending an antenatal clinic might decline testing. Of those who declined testing, the main reason cited was unwillingness to terminate a pregnancy under any circumstance. Other reasons included partner's disapproval or non-participation, perceived risk of having a child with CF as low, and the error rate of the test.

Uptake rates for the sequential testing structure similar to those reported by Mennie et al. (1992) have been cited in Danish (Schwartz, Brandt, & Skovby, 1993) and German (Jung, Urner, Grade, & Coutelle, 1994) samples of pregnant women, where more than 80% of 3054 women and 99% of 638 women, respectively, accepted the offer to be tested. Women's reasons for not participating, however, were not reported in either of these two studies.

A second method for screening during pregnancy which has received considerable attention is "couple" screening (Wald, 1991). In this instance, both partners are asked to submit test samples for carrier status analysis. Initially, only one partner's sample is tested. If that partner's test result is negative, then the couple is issued a "negative" test result. Likewise, only if both partners are found to be positive, does the couple receive a "positive" result. In this way, couple testing purportedly avoids undue anxiety for those carriers whose partner is negative. The method has been criticized, however, in that it

fails to inform carriers of their risk with new partners, and to warn their families of potential risk (Miedzybrodzka & Hall et al., 1995).

A comparative analysis of both the step-wise and the couple methods of screening during pregnancy was undertaken by Miedzybrodzka, Hall and colleagues (1995). In a randomized trial, either women alone or women and their partners were offered genetic counselling and screening. In addition to uptake rates and anxiety, several other variables were measured, including knowledge of CF and carrier status, attitudes toward pregnancy, the baby and screening, and attitude toward use of carrier screening by other relatives. Results showed that uptake rates were not significantly different between the step-wise and couple approaches (90%). Likewise, there was no difference between the groups on measures of CF knowledge. Notably, carriers in both groups expressed a more negative attitude than non-carriers toward their pregnancy and the baby. However, this attitudinal difference changed after the partners of carriers were tested, where carriers actually expressed more positive attitudes toward their pregnancy and baby. Across groups, most carriers had told one or more relatives about their positive test result. Similar to the findings of Mennie et al. (1992), increased anxiety levels were present for women carriers in the step-wise condition before their partner's results were known, but anxiety dissipated after the partner was shown to be negative, and did not reappear during pregnancy. Results, however, also showed couple screening to be associated with more anxiety and more false reassurance among screenees with negative results. From an economic

perspective, the authors note that while stepwise screening required more staff time, couple screening required slightly more laboratory time to prepare (Miedzybrodzka & Hall et al., 1995).

The same authors also considered women's expressed preferences for both types of counselling (Miedzybrodzka, Semper, Shackley, Abdalla, & Donaldson, 1995). In this study, women attending an antenatal clinic heard descriptions of both types of counselling and were then asked which they would prefer. An estimate of the value of each type of counselling was also obtained from the women using "willingness to pay" (WTP), a method whereby individuals express how much they would be willing to sacrifice to obtain a particular "commodity", such as a screening test. Reportedly, more women preferred step-wise counselling to couple screening (62% vs. 26%), however the WTP for each method was not significantly different. In conjunction with their preference findings, Miedzybrodzka and Hall et al. (1995) concluded that stepwise screening is associated with less overall anxiety and false reassurance, and is the better method for implementation in an antenatal setting.

Regardless of the type of screening offered to pregnant women and/or their spouses, there is general agreement among professionals that pregnancy is not the optimum time for offering screening because at that time, couples find themselves with a limited range of reproductive options (Brock, 1990). However, work in this area is still important, because clinical experience suggests that most individuals are not motivated to

seek carrier testing until a pregnancy occurs, irrespective of their objective risk status (Brock, 1990; Wright Clayton et al., 1996). Further, the more frequent use of CF carrier screening during pregnancy than before conception is consistent with findings about carrier-screening tests for other diseases, which are also primarily used after conception (Kaback et al., 1993; Tambor et al., 1994).

Screening Before Pregnancy

Considerable work has also been done in the area of factors affecting CF carrier screening decisions with non-pregnant individuals and couples of reproductive age. In contrast to screening programs during pregnancy, the rationale behind these screening programs is to allow individuals to maximize their choices in reproductive options, before a pregnancy occurs (Brock, 1995).

As in pilot work with pregnant women and their partners, the manner in which screening is offered has been found to play a pivotal role in the screening decisions of general population individuals. For example, Watson et al. (1991) found participation rates of 87% in a family planning clinic and 66% in a general practice setting, when adults of reproductive age were offered screening in person by a researcher. In contrast, only 10% of individuals who received a written invitation to participate sought testing. Similarly, Bekker et al. (1993) found that uptake rates were 70% for those offered testing in a general practice setting by a physician, while other methods of presentation (e.g., written invitation) yielded minimal participation. In conclusion, the authors assert that the

most important factor determining participation rates for screening is a personal approach by a professional, and the offer of immediate testing (Bekker et al., 1993).

A third study confirms that the method of presentation may affect utilization of testing. Tambor et al. (1994) offered carrier screening to individuals of reproductive age who were members of a Health Maintenance Organization (HMO). Uptake rates were low (3.7%) when individuals were invited to participate in a free testing trial that required attendance at a pre-trial educational session. By comparison, the participation rate was considerably higher (23.5%) when individuals were approached and offered immediate testing while waiting for an unrelated appointment. Thus, evidence suggests that in order to generate participation in screening programs, some sort of personal contact is necessary, while letters and leaflets are not effective (Tambor et al., 1994).

Unfortunately the economic implications of this degree of professional involvement in the testing process are considerable. As Brock (1995) points out, the high cost of professional involvement has rendered pilot screening programs in primary care settings virtually impossible. Still others suggest that while extending direct offers of testing from health care professionals to individuals might achieve high experimental uptake rates, these rates are artificial indicators of actual uptake rates (Surh et al., 1994). For example, despite their reports of high uptake rates, most past work (e.g., Watson et al., 1991, Bekker et al., 1993) has neglected to assess participants' understanding of the implications of screening, aside from accepting or declining an easy, painless test (buccal swab).

Further, individuals recruited in this manner may have opted for the test not because of their own personal conviction, but rather because the health care professional appeared to advocate it (Surh et al., 1994). In support of this argument, Bekker and colleagues (1993) suggest that high uptake rates achieved by active recruitment signal more of a "supply push" than actual "demand pull" for screening services. To date, Wilfond and Fost (1990) have provided the most comprehensive review of expressed concerns over the implications of widespread, general population screening.

An alternative to recruitment of general population individuals by health care professionals has been examined, where carrier testing for known relatives of a patient with CF is advertised via other family members. In this model, testing information moves or "cascades" down from the index case to immediate, extended and distant family members. For example, in Super and colleagues' (1994) pilot screening project of "cascade" testing, nuclear family members of an index patient were encouraged to invite their relatives to participate in screening. Those relatives who were found to be carriers were subsequently asked to encourage their relatives to seek counselling and screening. Results showed a substantial increase in the number of relatives and partners tested, following the introduction of the cascade method. In total, 1563 relatives and partners in 129 families with an affected individual sought testing. Of this sample, 15 heterozygous couples were identified, and 8 of these couples subsequently opted for prenatal diagnosis. Although Super et al. (1994) failed to provide an estimate of the total population from

which they sampled (and therefore overall uptake rates), they did assert some striking conclusions. By extrapolating to a total sample of 10 000 relatives and partners, the authors suggested that 100 heterozygous couples would be detected. Further, the authors concluded that in their hypothetical sample, 25 affected fetuses would be diagnosed: a figure equal to the number of affected children born each year in their Manchester, England region. Given their results, the authors heralded pro-active cascade screening as a viable and effective alternative to widespread population screening (Super et al., 1994).

Subsequent critics of Super et al.'s findings (1994) have pointed out, however, that it is impossible to measure the theoretical effectiveness of cascade testing by simple extrapolation (Brock, 1995). Moreover, it has since been shown that cascade testing must actually reach the fourth cousin level in order to be comparative to the effectiveness of population screening (Holloway & Brock, 1994). Parallel to criticisms of general population screening models, Super et al.'s (1994) endorsement of cascade screening has been criticized on the basis that the authors did not consider how effectively information about reproductive options was disseminated, nor how well it was understood before testing decisions were made (Marteau, 1994).

In fact, a recent Canadian study of the cascade method supports the idea implicit in the criticism of past work: that CF carrier screening research should move beyond a simple analysis of uptake rates, and ask how information is disseminated, what motivates individuals to be tested, and what perceived relevance the results of carrier testing hold for

those being tested. In their study of relatives of CF patients, Surh et al. (1994) relied solely on the family members of an affected individual to disseminate information about the availability of free CF carrier testing. In contrast to Super et al.'s (1994) findings, these authors found an uptake rate of only 10%. The factors which predicted participation in testing were 1) greater degree of relationship to a pediatric patient with CF, and 2) the presence of an unsolicited family member who took on self-appointed recruiting role. Surh and colleagues hypothesized that the observed lack of participation shown by relatives may have been because they 1) did not know about the test due to limited dissemination of test results, 2) knew about the test, but lived outside the regional catchment area, or 3) knew about the test, but did not perceive the results as very useful. In support of the third explanation, the authors cite past work which also suggests that the desire to know one's own carrier status may not be an important motivator in an individual's decision to be tested (Denayer, Evers-Kiebooms, De Boeck, & Van den Berghe, 1992; Page, Eng, Chang, & Waye, 1992).

In a subsequent analysis of factors differentiating between those who are motivated to seek testing and those who are actively recruited, Surh et al. (1995) compared three groups of individuals: "Tested" individuals who had previously sought carrier testing and had a family history of CF, "Recruited" individuals who had a family history of CF but had not sought carrier testing, and "General Population" individuals who had not previously been carrier tested. All three groups demonstrated similar preferences for health states

associated with CF. Further, no differences were observed between the previously Tested group and the Recruited group on measures of CF and carrier testing knowledge, degree of information dissemination, or attitude toward carrier testing. However, despite not seeking testing on their own, 97% of the Recruited group agreed to be tested when offered, as compared to only 63% of the General Population group. Indeed, the only discernable difference between the previously Tested and Recruited groups was that the latter was offered the *convenience* of home testing using a non-invasive procedure (buccal swab). The findings of Surh and colleagues (1995) suggest findings that clearly diverge from past work. Rather than attitudinal factors and a desire to know one's carrier status, the decision to be tested may be entirely contingent on the convenience of testing and the perceived "value" of (or lack thereof) carrier test results. Perhaps most importantly, these authors' results underscore the idea that high uptake rates do not necessarily reflect "need" nor "usefulness" on behalf of those tested.

Factors Related to Reproductive Planning

Relatively few studies have examined if and how the results of carrier testing are actually used to assist in making reproductive decisions. This paucity of literature is somewhat paradoxical, given that the primary objective in the genetic counselling process is usually regarded as the facilitation of more informed reproductive decisions (Conway, Allenby, & Pond, 1994; Hsia, Hirschhorn, Silverberg, & Godmilow, 1979; Wilfond & Fost, 1990). Two studies have considered the effect of CF carrier status knowledge in an

indirect manner, by studying the impact of the birth of an afflicted child on subsequent reproductive plans (Evers-Kiebooms, Denayer, & Van den Berghe, 1990; Wertz, Janes, Rosenfield, & Erbe, 1992). Evers-Kiebooms et al. (1992) interviewed parents of children with CF to assess the effect of their child's birth on family planning decisions. Three years later, parents completed a follow-up questionnaire to update their initial responses, and to assess their knowledge and intentions concerning new possibilities in DNA testing. Results showed that the birth of a child with CF had a major impact on subsequent family planning, as evidenced by parents' reported plans and the occurrence of pregnancies in the follow-up period. The most important factor associated with the decision to forego having more children was the objective risk factor. A second relevant factor was the birth-order of the child with CF. If the child was the first born, the occurrence of a subsequent pregnancy was more likely than in families who already had a healthy child. Regardless, only 47% of the parents whose first child had CF opted for another pregnancy. In addition, it was shown that all parents used more reliable contraception after the birth of an affected child, including sterilization at a younger age than in the general population.

Wertz et al. (1992) also studied factors associated with reproductive decisions, in a sample of parents of children with CF. Specifically, this study explored the psychosocial factors associated with the intent to use prenatal diagnosis for CF. Of the 227 couples surveyed, fewer than one fifth were considering more children, and a majority (69%) were

surgically sterile. Of those who did intend to have more children, 77% had considered or had already used prenatal diagnosis. The three factors most commonly associated with an increased likelihood of intent to use prenatal diagnosis were a) a greater willingness to abort for CF, b) a siblings' approval of abortion for CF, and c) a subjective list of no accomplishments (i.e., social, academic) for the child with CF. Notably, the majority of families rejected selective abortion for CF, opting instead to curtail childbearing, rather than use prenatal diagnosis. The authors asserted that the high level of CF knowledge among their respondents indicated "successful efforts" on behalf of the health care community to disseminate accurate information. Moreover, while noting that it is difficult to predict population attitudes from those of families already afflicted by CF, Wertz et al. contend that the decisions made by those in the general population are also likely to be contingent on the accuracy of CF information relayed to them via health professionals.

Denayer et al. (1992) is the only comprehensive study to date to consider the reproductive decision making of relatives of a CF patient. Respondents' subjective evaluation of their risk did indeed play a role in the reproductive decision-making of at least 39% of the sample. Nonetheless, only 9% actually curtailed having any or more children because of the perceived risk factor. The authors note, however, that the family sizes in those sampled were smaller than expected in relation to a representative sample of cohorts in the general population. The majority of the sample indicated that they would likely use carrier testing, and would seek prenatal diagnosis should they become pregnant.

Further, the intention to use prenatal diagnosis was significantly correlated with age and educational level. The acceptance of abortion of a fetus with CF was positively related to both perceived burden of raising a child with CF and respondents' age, while negatively related to the health state of the afflicted relative. In support of Wertz et al.'s (1992) assertion about the importance of accurate dissemination of information in relation to subsequent family planning decisions, Denayer and colleagues found a positive relation between knowledge of how CF is transmitted, and the presence of "worries" about the risk of having an affected child. In general, however, respondents tended to underestimate their carrier risk, and have inaccurate information about carrier testing issues.

The work of Frets et al. (1990) is unique in that these authors actually considered the intentions of those who knew their carrier status, but did not have an affected child. Follow-up interviews with individuals were completed 2-3 years after their initial genetic counselling session to assess factors associated with reproductive decisions. Results showed that even when the disorder was perceived as severe, and individual risk was interpreted as high ($>15\%$), 72% of couples indicated that they still planned to have children. Thus, neither the magnitude of genetic risk nor the perceived severity of the disease was foremost to reproductive planning. Rather, intent to have children was associated with the desire to have children, and an absence of personal experience with the disorder. Unfortunately, Frets et al.'s (1990) results are of limited application to specific reproductive decisions surrounding CF; the results reported are pooled for couples

screened for a variety of genetic disorders, and the report does not include a disease-specific analysis.

Clearly, knowledge surrounding factors which predict the actual use of carrier screening results in subsequent reproductive decisions is limited. For example, while it would appear that the birth of a child with CF is a significant factor in shaping some parents' subsequent family planning decisions, this is not the case for all. Similarly, it appears that although objective risk information influences the decisions of some, the effect of perceived risk is more profound for others. In addition, the specific interplay of factors such as personal experience with CF, current number of children, age, information about CF and willingness to abort an affected fetus remain largely unexplored, and in need of an integrative model.

Psychological Impact of Screening

One of the most frequently studied aspects of carrier screening has been the psychosocial impact on those tested. The majority of work in this area has been undertaken with general population samples. In perhaps the largest study of psychosocial effects of carrier screening programs, Watson, Mayall, Lamb, Chapple, and Williamson (1992) assessed over 3000 individuals in a primary care setting. Although varying degrees of anxiety were initially correlated with a positive test result, most carriers' fears were reportedly alleviated through counselling. Further, Watson et al. (1992) found that six months after testing, the majority of carriers reported feeling positive about their decision

to be tested and were unconcerned about their test result. Carriers also indicated that they intended to use their knowledge in future family planning decisions. In support of their intentions, follow-up measures revealed that carriers retained greater knowledge about CF and its inheritance than did non-carriers. In light of their findings, Watson and colleagues concluded that the possible social costs of screening were unfounded, and that the information gained from community screening programs would be used in a proactive manner to cope with the risk of having a child with CF.

Mennie et al. (1992) also observed significant anxiety levels among 64 pregnant women who were identified as carriers, particularly during the period of waiting for their partner's results (about 4 days). However, in support of Watson et al.'s (1992) results, these elevated anxiety levels disappeared after the partner's test results (all of which were negative) were known, and did not reappear during the course of the pregnancy.

Kaplan, Clow and Scriver (1991) also found that the majority of those identified as non-carriers in a high school sample understood the residual risk of being identified as a non-carrier (95%), were satisfied that they were tested (84%), and stated that receiving a "carrier" test result would not have affected their self-image (70%). The overwhelming majority of non-carriers also advocated genetic testing in high schools (96%). The views of the carriers in the sample, however, are not reported, except for the general statement that they and their families "were found to have positive views about their new awareness" (Kaplan et al., 1991; p. 241).

Although not a direct analysis of the psychological impact of CF carrier screening, the findings of Marteau and her colleagues (Marteau, Van Duijn, & Ellis, 1992) regarding Tay-Sachs screening are nonetheless related to the literature surrounding psychosocial effects of screening for CF. Perceptions of general health were compared in three groups: carriers of the genetic mutation for Tay-Sachs disease (TSD), non-carriers of the TSD gene mutation, and a community based sample who had not undergone screening for TSD. The three groups did not differ in their perceptions of their current or past health. Carriers of the TSD mutation, however, did view their future health with less optimism than the other two groups. The authors contend that their results underscore the importance of preliminary examinations of individuals' experiences of screening programs, before any such programs are undertaken.

Similarly, in an analysis of CF carriers and non-carriers, Evers-Kiebooms and colleagues (Evers-Kiebooms, Denayer, Welkenhuysen, Cassiman, & Van den Berghe, 1994) also report finding that carriers reported significantly fewer positive feelings about themselves, even a year after they had received test results. Both groups also attributed more negative feelings to carriers than to non-carriers. Thus, the authors suggest that their findings may point not only to a self-stigmatizing effect, but also to a social stigmatizing effect of CF carrier status.

The work of Bekker, Denniss, Modell, Bobrow and Marteau (1994) likewise points to some negative psychological consequences as a result of carrier screening. In

their evaluation of effects of population based carrier screening, measures of health perceptions, anxiety, and test result understanding were compared before testing, upon receipt of results, and 3 months later. While there was no difference between carriers and non-carriers on perceptions of health, anxiety levels were higher among those who received a positive result. By the three month interval, however, this anxiety had dissipated. Most disconcerting to the authors were the findings regarding knowledge retention. Overall knowledge of CF and its transmission reportedly improved from pre-test to post-test. Yet, at the three month follow-up, 17% of those who received a negative result had disregarded and/or forgotten about the residual risk explained to them and erroneously reported that they were at no risk for having a child with CF. Similarly, 36% of the carriers incorrectly believed that they probably, but not definitely, carried the gene for CF. Bekker et al. (1994) maintain that in the long term, the greatest consequence of population based screening may actually be false reassurance, rather than false anxiety.

Thus, while there is evidence to suggest that the psychosocial impact of carrier screening programs may be minimal, the work is far from unanimous in concluding that population based screening for CF is beneficial (Harper, 1995). Likewise, more work remains to be done surrounding if and how carrier screening can pose a threat to those with a positive result. Despite these differing results, there is consensus about one area: the need for accurate information dissemination and effective counselling as an integral part of any testing process (Brock, 1990; Colten, 1990; Harper, 1995; Stewart, 1989;

Watson et al., 1992). Genetic counselling is recognized as crucial in minimizing the misunderstanding of test results, the impairment of self-image and the perceived unsuitability as a partner and a parent (Rowley, Loader, Levenkron, & Phelps, 1993; Wilfond & Fost, 1990).

The Role of Health Beliefs and Coping Style

A substantial body of research surrounding factors associated with screening and reproductive decisions, and the psychosocial impact of testing has surfaced since the cloning of the CF gene. Nonetheless, a gap is apparent in the existing literature. The personality variables and health beliefs which may affect genetic testing and counselling situations remain largely unexplored. In this sense, past work has explored what individuals *take away* from genetic testing situations, but what they *bring* to these situations has been largely neglected.

An understanding of the contribution of relevant personality and belief information to genetic screening decisions may be paramount to developing counselling protocols which could maximize personal understanding, retention, and effective use of test results, while minimizing their psychological impact. In fact, surprisingly little work has been done surrounding factors, both intrapersonal and interpersonal, which combine to create an effective counselling infrastructure. Moreover, the poor research record in this area continues even in spite of numerous studies which have shown that counselees retain a limited amount of knowledge from genetic counselling sessions (Bekker et al., 1994;

Besson & Golbus, 1985; d'Ydewalle & Evers-Kiebooms, 1987; Rona et al., 1994) and/or misinterpret the information as it applies to themselves (Bekker et al., 1994). In a related way, past literature surrounding the genetic counselling/testing process has yet to offer a comprehensive theoretical framework to explain why some individuals make decisions that do not seem rational in relation to their recurrence risk (Welshimer & Earp, 1989).

The HBM (Rosenstock, 1974) has previously been cited as a theoretical framework for understanding individual beliefs in relation to genetic counselling decisions (Pearn, 1974; Rosenstock, 1979). In theory, the HBM may be relevant in explaining individuals' motivation regarding the decision to be tested for CF carrier status, since the decision involves a novel and singular (vs. habitual) action, and there are no guidelines for decision-making according to professional advice (Calnan, 1984; Sagi et al., 1992). Under these conditions, there is increased likelihood that decisions will be made with vigilance, based on relevant cognitions (Sagi et al., 1992).

A second personality dimension which may be relevant to genetic testing is the preference for information under threat, as embodied by Monitor/Blunter theory (Miller, 1987; Miller et al., 1988). An individual's preference for information may be key to her/his decision to be tested. Those with a higher preference for information under threatening conditions may be more motivated to learn their carrier status, thereby decreasing their uncertainty about the future. On the other hand, those with a decreased preference for information may prefer not to know carrier status information, and may in fact be

adversely affected by knowing their results.

The Health Belief Model

The HBM (Rosenstock, 1974) predicts health behaviour according to an individual's perceptions and beliefs. The basis of the model is derived from a credible body of behavioural and psychological literature, whose various models hypothesize that behaviour depends primarily on: a) the value placed by an individual on a particular goal, and b) the individual's estimate of the likelihood of attaining that goal (Becker & Maiman, 1975). Within the health setting, these variables have been conceptualized in terms of health related behaviour: a) the desire to avoid illness (or if ill, to become well), and b) the belief that a certain health behaviour will prevent (or alleviate) illness (Janz & Becker, 1984).

In particular, the model hypothesizes that before an individual takes preventive health action against a disease, he/she must: a) perceive himself or herself to be susceptible to the disease (*perceived susceptibility*), b) believe the disease to be severe (*perceived severity*),

c) perceive the result of the health behaviour to be effective (*perceived benefits*), and d) believe that the obstacles to taking the health action, either psychological (e.g., fear of procedure) or physical (e.g., time taken from workday, cost), can be overcome (*perceived barriers*). Notably, the model emphasizes individual perception (rather than objective status) of the four dimensions in forming a decision to take health action. In addition to

the original four dimensions (Rosenstock, 1974), later work with the HBM has included other modifying factors in the model, such as demographics (i.e., age, sex, education, socio-economic status), and knowledge of the disease (Becker & Maiman, 1975).

Early work considering preventive health and sick role behaviours including immunizations, ambulatory clinic use, antihypertensive regimens, diabetic regimens, confirmed the predicted HBM effects for all but perceived severity (c.f., Janz & Becker, 1984). Subsequent work has examined various components of the HBM, particularly in attempt to explain and increase utilization of preventive health behaviours (i.e., mammography) for breast cancer (Aiken, West, Woodward, & Reno, 1994; Bastani, Marcus, & Hollatz-Brown, 1991; Calnan, Moss, & Chamberlain, 1985; Champion, 1992; Lerman, Rimer, Trock, Balshem, & Engstrom, 1990; Rimer, Keintz, Kessler, Engstrom, & Rosan, 1989; Rutledge, Hartman, Kinman, & Winfield, 1988; Slenker & Grant, 1988; Stein, Fox, & Murata, 1991; Stein, Fox, Murata, & Morisky, 1992; Zapka, Stottard, Costanza, & Greene, 1989). As with earlier work, these studies have yielded little support for the severity component of the HBM, while confirming positive relationships between mammography behaviour and perceived susceptibility and perceived benefits, and negative relationships between mammography behaviour and perceived barriers (Aiken et al., 1994). In addition, support has been found for the predictive value of the added HBM components of demographics (e.g., age, socio-economic status, education) (Bastani et al., 1991; Hyman, Baker, Ephraim, Moadel, & Philip, 1994; Rimer et al., 1989; Rutledge et

al., 1988) and disease knowledge (Aiken et al., 1995; Champion, 1992) and experience (Lerman et al., 1990). Interventions designed to alter health beliefs in relation to mammography use have been successful in modifying all but perceived susceptibility in the expected direction (e.g., Champion, 1994).

The Health Belief Model and Genetic Testing

Only a few empirical studies have considered the HBM variables in relation to genetic counselling issues. Becker, Kaback & Rosenstock (1975) examined health beliefs as predictors of participants in a screening program for Tay-Sachs disease (TSD). Using a questionnaire format, participants and non-participants were asked about their perceptions of the likelihood of being a carrier of the TSD gene, the severity and impact of learning that one was a carrier (especially within the context of family planning decisions), and the potential barriers and benefits associated with screening. Results showed that significantly more participants than non-participants felt they were susceptible to being carriers of the Tay-Sachs mutation. Perceived severity was also related to participating. Low or moderate perceptions of the severity of positive carrier status appeared to motivate individuals to be tested, while high perceptions of severity had an inhibiting effect on participation. The combined "susceptibility" and "severity" perceptions of an individual's spouse also increased the accuracy of predicting screening participation. Findings for the perceived benefits and barriers were also in the hypothesized direction, although no statistical analyses were reported. The authors concluded that these HBM variables were

mitigated by knowledge that amniocentesis could identify a fetus with TSD, and by willingness to undergo an abortion for an affected fetus (Becker et al., 1975).

Past work has also found a link between the "susceptibility" component of the HBM and families' decisions surrounding genetic risks. For example, reproductive intentions of genetic counselees have been positively related to their subjective perceptions of recurrent risk (Sagi et al., 1992; Sorenson, Scotch, Swazey, Wertz, & Heeren, 1987), and with counselees' estimates of the burden of the genetic disorder (Meryash, Bernier, & Ahmad, 1989). A relation between individuals' beliefs about the risks of performing different prenatal diagnostic procedures and their preferences for these procedures has also been observed (Lippman-Hand, Perry, Mandel, & Cartier, 1985).

Recently, Rowley and colleagues (Rowley, Loader, Sutura, & Kozyra, 1991) explored the feasibility of the HBM in explaining the differences between a) pregnant women identified as carriers for a hemoglobinopathy who opted to have their partner tested and those who did not, and b) couples at risk for a child with a hemoglobinopathy who wanted prenatal diagnosis and couples who did not. Support was found only for the HBM variables of "perceived severity" and "perceived barriers", and only in regard to the partners' testing decision. Women who perceived the potential illness as severe were more likely to have their partners tested. The authors also found an important predictive effect of proximity to the partner: women who lived with their partners were more likely to have their partners tested. Living away from one's partner was thus identified as a "perceived

barrier" to testing. Accordingly, these findings lend tentative support to the applicability of the HBM in predicting partner testing for hemoglobinopathy screening.

Sagi et al. (1992) also considered the relation of HBM variables to parents' intentions to use prenatal diagnosis of cleft lip and/or palate. Parents' beliefs regarding perceived susceptibility and severity to abnormalities, and perceived benefits and barriers of prenatal diagnosis were evaluated using a questionnaire method. Significant correlations were found between parents' intentions and their beliefs. When the malformation was perceived as more severe and more likely, and when the diagnosis was perceived as beneficial and less distressing, intentions to use prenatal diagnosis increased. Perceived benefits were noted as the strongest predictor of both intentions to use prenatal diagnosis and termination of an affected pregnancy. This finding corresponds with a trend in HBM literature which has shown perceived benefits to be among the best predictors of health behaviour (Sagi et al., 1992).

The Monitor/Blunter Theory

A key environmental factor which has been found to mediate stressful situations, particularly within the medical setting, is the extent to which patients have information about the situation which they are about to experience (Auerbach, 1989; Miller, 1990; Miller, 1992; Suls & Fletcher, 1985). The study of personal coping strategies in relation to the availability of information has largely been conceptualized across two independent dimensions called "monitoring" and "blunting" (Miller, 1987; Miller et al., 1988).

According to this theory, when faced with threatening situations, individuals react in one of two ways. They may *monitor* threat-relevant cues, actively seeking out information about the procedure and/or their condition. Alternatively, they may avoid or *blunt* threatening information in order to minimize anxiety.

By using coping interventions such as distraction and relaxation to blunt threatening stimuli, the theory reasons that blunting individuals are able to selectively process events in a less negative way, thereby reducing anxiety and controlling their emotions in the face of threat (Miller, 1989). For these individuals, the effect of threat-related information is negative. Information forces them to attend to the aversive nature of the situation, thereby disabling their habitual "blunting" responses and increasing their anxiety.

In contrast, threat-related information has the opposite effect for those who will not or cannot use these cognitive interventions. For these "monitors", threat-relevant information is considered positive because it brings increased certainty and the reassurance that predictive cues allow (Leventhal, 1988; cited in Miller, 1989), thus minimizing their stress. Based on this rationale, the theory predicts that how well an individual copes should be a function of the match between information provided and the individual's dispositional style (Miller, 1992).

The first conclusion which has emerged from the work on coping styles is that when faced with threatening conditions, those identified as monitors do in fact prefer more

detailed and extensive information than blunters (Miller, 1988). Laboratory studies have demonstrated this finding within settings involving the cold pressor task (Efran, Chorney, Ascher, & Lukens, 1989), threat of electric shock, and cognitively challenging paper and pen tasks (Miller, 1987). These results have also been extended to applied medical and surgical settings involving gynaecological surgery (Miller & Mangan, 1983; Steptoe & O'Sullivan, 1986), cardiac catheterization (Watkins, Weaver, & Odegaard, 1986) and metastatic cancer (Steptoe, Sutcliffe, Allen, & Coombes, 1991).

Secondly, past work has also shown that there may be a psychological and physical "cost" associated with a monitoring style when the stressful situation is largely uncontrollable, in both laboratory settings (Efran et al., 1989; Miller, 1987) and applied medical settings involving gynaecological surgery (Miller & Mangan, 1983), preparation for cardiac catheterization (Watkins et al., 1986), amniocentesis (Phipps & Zinn, 1986), general outpatient surgery (Caldwell, 1991) and chemotherapy (Gard, Edwards, Harris, & McCormack, 1988). Notably, Miller, Leinbach and Brody (1989) have also cited a link between a monitoring coping style and chronic disorders, noting that hypertensive patients were significantly more likely to display a monitoring coping style.

Overall, past findings also tend to support a third conclusion: the idea that how well an individual copes is a function of the fit between her/his characteristic coping style and the individual situation. This finding has been demonstrated independently of major trait factors such as depression and anxiety (Caldwell, 1991; Ludwick-Rosenthal &

Neufeld, 1993; Miller, 1987; Miller, 1992; Miller et al., 1988; Steptoe & O'Sullivan, 1986; Watkins et al., 1986). In particular, these findings have been demonstrated using behavioral, subjective and physiological measures of stress in situations involving gynaecological stress (Stewart, 1989), and preparation for cardiac catheterization (Watkins et al., 1986; Ludwick-Rosenthal & Neufeld, 1993; Davis, MacGuire, Haraphongse, & Schaumberger, 1994).

Given monitors' propensity to seek out threat-related cues, several studies have also considered the relationship of coping style to health seeking behaviour. For example, Steptoe and O'Sullivan (1986) considered two questions in a sample of women undergoing gynaecological surgery: 1) whether differences existed between monitors and blunders on the amount of factual information they had about medical conditions and their desire for further information, and 2) whether monitoring was linked to a "vigilant" health behaviour, or actions that may prevent disease. Indeed, it was found that monitors were more likely to undergo annual cervical smears than blunders. Likewise, 21% of monitors reported that they completed breast self-exam on a daily or weekly basis, as compared to 5% of blunders ($p < 0.07$). In addition, monitors who thought they understood the details of their procedure actually had more factual knowledge. By contrast, factual knowledge of blunders was unrelated to their reported understanding, suggesting that blunders reported understanding is more a function of preferred coping style than actual understanding (Steptoe & O'Sullivan, 1986). Based on their sample of primary care

patients, Miller et al. (1988) also concluded: 1) that a monitoring coping style may be predictive of greater health care utilization, independent of depression level, and 2) that although monitors appear to suffer from less severe medical problems, they complain about them equally and take longer to report recovery.

The Monitor/Blunter Theory and Genetic Testing

The interactive approach suggested by Miller's theory of monitoring/blunting may provide an illustrative model for assessing person-by-situation interactions in the genetic counselling setting. If how well an individual copes is in part a function of the "fit" between the situation and her/his coping style, then important clinical and educational implications must be considered (Miller, 1990). It may be possible to improve health-care outcomes by tailoring the level of information presented to patient coping style. Indeed, past work has demonstrated the feasibility of assessing patient coping style via the MBSS and using the outcomes to tailor instructions to patient coping style (Watkins et al., 1986). The use of a brief test to assess patient coping style also has the added benefit of being more efficient, widely applicable, and cost-effective, as compared to the use interventions employing health-care professionals to instruct patients in the maximal use of their preferred coping style (Kendall et al., 1979; Tan & Poser, 1982).

A logical prediction following from monitors' propensity to engage in more health-seeking/vigilant behaviour is that they would be more likely to seek carrier status information, via genetic testing. Yet, despite the apparent applicability of the

monitor/blunter theory to genetic testing issues, only a few studies to date have considered the role of monitoring/blunting within the context of genetic testing issues.

Phipps and Zinn (1986) assessed differences between monitors and blunters in a group of women undergoing amniocentesis, and a control group not having the procedure. In the amniocentesis group, monitors experienced significant increases in subjective arousal before and during the procedure. However, the interaction between coping style and anxiety disappeared after communication of results. These findings suggest that women's responses to amniocentesis are more than a function of the objective components of the procedure - they are a function of individual differences in coping style that influence subjective appraisal of stressors involved (Phipps & Zinn, 1986).

Although they did not consider individual differences in coping style, Decruyenaere, Evers-Kiebooms & Van den Berghe, (1993) considered young women's perceptions of genetic testing for Huntington's disease. They found that a positive attitude toward predictive testing was systematically related to perceiving a more certain future as a benefit. Likewise, those who had more negative attitudes toward testing cited "knowing everything in advance" as a disadvantage. Indeed, these authors call for more work exploring the personality profile of those who prefer certainty and those who prefer uncertainty, within the context of genetic testing decisions.

Rationale for the Current Study

Based on the foregoing literature review and theoretical framework, potential intrapersonal predictors of the intent to be tested for CF carrier status were considered in the present study. The primary objectives of the study were as follows: 1) to examine health beliefs as they relate to the intent to be tested for CF carrier status, and 2) to examine personal coping style as it relates to the intent to be tested for CF carrier status. Specifically, the following hypothesis were developed and tested:

1) Individuals who intend to be tested will perceive themselves as more susceptible to carrier status, carrier status as more severe, and more benefits and fewer barriers to the testing procedure. Further, individuals who intend to be tested will have an increased knowledge of CF.

2) Individuals who have a characteristic monitoring coping style will be more likely to report an intention to be tested for CF carrier status, when compared with individuals who have a characteristic blunting coping style.

A third objective of the study was to consider the relationship between the HBM and the Monitor/Blunter theories, within a genetic testing context. Although an analysis of the two theories has not previously been undertaken, it is expected that a relationship will exist between the two theories. In particular, the following hypothesis was tested:

3) Individuals who have a characteristic monitoring style will perceive carrier status as more severe, themselves as more susceptible to carrier status, and more

benefits of screening for CF carrier status. In addition, it was expected that monitoring individuals would perceive fewer barriers to the screening process, as compared to blunting individuals.

Finally, the following demographic variables were considered in relation to intent to use genetic testing for CF carrier status: gender, age, income, religion, marital status and number of children.

Methods

Participants

From April until June, 1996, general-population individuals were sampled from a regional catchment population of over one million in Eastern Ontario and Quebec. This sample population is congruent to the majority of pilot studies which have been conducted with individuals from the general population. English-speaking adults who accompanied children to an orthopaedic out-patient clinic at the Children's Hospital of Eastern Ontario were invited to participate in the study by completing the questionnaire package at their convenience, and returning it to the researcher in the stamped, addressed envelope provided. In particular, those within the reproductive age range of 18–45 years were targeted for inclusion in the study, given that past work and clinical experience have shown that this age group most actively seeks genetic counselling and testing (Surh et al., 1995). Further, individuals who had previously undergone CF carrier testing, or had a child with any chronic illness (including CF) were excluded from the study sample. Prior to commencing data collection, the proposal for this study was reviewed and deemed acceptable by ethics committees at the Children's Hospital of Eastern Ontario, and Carleton University.

Measures

CF Carrier Testing Survey.

In order to examine knowledge, affect, attitudes and intended behaviours, the CF Carrier Testing Survey was used (Appendix A), as developed by Surh and colleagues (1994; 1995). The survey contains four subscales which measure a) knowledge about the disease and its inheritance/experience with the disease (8 items), b) possible affective changes arising from carrier testing (6 items), c) attitudes toward genetic testing (9 items) and d) intended behaviours after being carrier tested (8 items). In addition, the survey addresses demographic information such as number of children, age, sex, religion and education level (11 items). Finally, the survey contains an item which asks individuals if they would consider having CF genetic testing for carrier status (p. 6, Appendix A). For purposes of the present study, answers to this dependent measure were dichotomized into yes/no responses.

In Surh and colleagues' preliminary work with the CF questionnaire, reliability analysis yielded Cronbach alphas for each subscale of 0.75 (knowledge), 0.82 (affect), 0.83 (attitudes) and 0.74 (behaviours following testing). Acceptable concurrent validity was also demonstrated through the testing and confirming of three hypotheses: 1) anticipated family planning behaviour was correlated with a positive attitude toward genetic screening ($r=0.56$, $p<0.001$), 2) individuals reported significantly less pleasant feelings with respect to a hypothetical positive carrier test result scenario than a negative

carrier test scenario ($t = 16.08$, $p < 0.001$), and 3) individuals considering screening had significantly less negative attitudes and more positive behaviours (e.g., encouraging other family members to be tested) when compared with individuals who did not consider testing ($t = 4.52$, $p < 0.001$, and $t = 2.50$, $p < 0.01$, respectively).

Health Belief Model Measures.

Currently, no established measure of perceptions pertaining to the HBM exists. although efforts have been undertaken to develop HBM scales tailored to disease-specific hypotheses, such as mammography compliance (e.g., Champion, 1995; Hyman & Baker, 1992). In order to assess HBM concepts of perceived benefits and barriers, this study drew on two of the established attitude items in the CF questionnaire, which assess both the perceived benefits and perceived barriers surrounding carrier screening. In particular, two items (p. 10, Appendix A) ask individuals to indicate which of several statements they perceive as advantages to testing, and which of several statements they perceive as potential disadvantages to carrier testing. Although the items in the original CF questionnaire are measured as yes/no responses, the current study evaluated the same items with a Likert-scale, which ranged from "Strongly Agree" (1) to "Strongly Disagree" (5). This adjustment was made in order to make responses to this question consistent with the other HBM measures (see below), and to provide participants with a wider range of options for assessing the personal relevance of each item.

Perceived susceptibility to CF carrier status was assessed with two existing measures contained in the CF survey and two new items. The two existing items (pp.4-5, Appendix A) ask respondents to indicate 1) the probability that they are a CF carrier, and 2) how concerned they are about the indicated probability. A third measure of perceived susceptibility (p.5, Appendix A) was taken by asking respondents to mark their perceived risk of being a carrier on a scale from 0 (no risk) to 100 (certain risk). In addition, a fourth measure of individuals' perceived susceptibility was taken (p.5, Appendix A) by asking respondents to indicate, based on their answer to the preceding rating scale question, whether they thought that their risk was a) very high, b) high, c) moderate, d) low, or e) very low.

Perceptions of severity were measured via the addition of four new items to the existing CF survey (p.12, Appendix A). Following the format of the majority of the CF questionnaire, respondents were asked to indicate their beliefs surrounding the following questions on a Likert-scale ranging from 1 (Very Likely) to 5 (Very Unlikely):

- 1) I would have great difficulty adjusting emotionally if I learned that I was a CF carrier.
- 2) Learning that I was a CF carrier would make me feel hopeless.
- 3) Finding out that I was a CF carrier would hurt my relationship with my (future) partner.
- 4) Finding out that I was a CF carrier would cause me a lot of stress.

The Miller Behavioural Style Scale (MBSS).

Prior to the early 1980's, most of the available measures of individual coping styles were psychometrically lacking or invalidated (Miller, 1987; Miller, 1992), or were focused exclusively on the role of psychodynamic variables (e.g., Byrne, 1961). The Miller Behavioral Style Scale (MBSS; Miller, 1987; Miller, 1992) is directly related to hypothetical behavioural descriptions of how an individual prefers to cope with physically and psychologically aversive situations (Appendix B).

The MBSS has predicted responses in several different medical settings including aversive gynaecological procedures (Miller & Mangan, 1983; Steptoe & O'Sullivan, 1986), chemotherapy (Gard et al., 1988), cardiac catheterization (Davis et al., 1994; Ludwick-Rosenthal & Neufeld, 1993; Watkins et al., 1986), amniocentesis (Phipps & Zinn, 1986), and laboratory settings involving threat of electric shock (Miller, 1987), stressful cognitive tasks (Miller, 1987) and cold pressor tasks (Efran et al., 1989). Moreover, the MBSS has been shown to predict coping style independently of other characteristics such as trait anxiety (Miller, 1990). In addition to good predictive validity, convergent and discriminative validity have been demonstrated for the scale (Miller, 1987; Miller & Mangan, 1983). A test-retest reliability¹ showed the MBSS to be highly stable and in the 0.80 range, over an acceptable time-frame of three months (Miller, 1992).

¹In an electronic mail reply (dated June 28, 1996) to my request for more information about the cited reliability analyses, Miller confirmed that her data for the MBSS have never been published, and that the manuscript containing the reliability data is "currently unavailable".

Scores on the MBSS are calculated in one of three ways. The monitoring measure is the sum of all items endorsed on the monitoring subscale. Individuals who score above the sample median on the monitoring scale are considered to be "high" monitors, while those scoring below are low monitors. Likewise, blunting style is measured by summing all blunting scores, and dividing individuals into high and low blunners according to a sample median split. Finally, an overall monitor/blunter score may be obtained by subtracting the total number of blunting items endorsed (16 total) from the total number of monitoring items endorsed (16 total). Thus, a higher score indicates more monitoring behaviour and the calculated difference score is used to separate individuals into monitoring and blunting groups, respectively.

Although the majority of early research with the MBSS used a difference score as a measure of monitoring/blunting behaviour, Miller suggests that given the complex, multidimensional process of coping, it is probably most useful to consider the dimensions of monitoring and blunting separately (Miller, 1987; Miller, 1990). As such, more current work with the MBSS has heeded Miller's advice, and employed separate measures. For most analyses, the current study maintained a separate monitoring and blunting score for each individual in order to explore the monitoring and blunting concepts separately. However, to test the specific hypothesis that characteristic monitors would be more likely to report an intention to use CF carrier testing, difference scores were used.

Spielberger State-Trait Anxiety Inventory.

In order to assess baseline levels of anxiety, the State-Trait Anxiety Inventory was used (Appendix C). The scale is one of the most widely employed measures of anxiety, and comprises 20 items which measure state anxiety (i.e., "how I feel right now"), and 20 items which measure trait anxiety (i.e., "how I feel in general"). Two forms of the inventory have been published, form X and the more recent form Y. In general, form Y is recommended because of its superior psychometric properties (Spielberger, Gorsuch, & Lushene, 1970). Based on the responses of 1387 federal aviation workers in the United States, alpha coefficients of 0.93 (state) and 0.91 (trait) are reported for the measure. Test-retest reliability coefficients for the trait score, based on the responses of 357 high school students at 60 and 90 days, are cited as 0.71, 0.68 (males) and 0.75, 0.65 (females). Using the responses of the same sample, reliability coefficients for the state score are reportedly 0.62, 0.51 (males) and 0.34, 0.36 (females). Inclusion of an anxiety index as a control measure is consistent with both past work in the personality domain (Pervin, 1990), and previous work with the CF questionnaire (Surh et al., 1994; Surh et al., 1995).

Procedure

The package given to participants who were approached at the orthopaedic outpatient clinic at the Children's Hospital of Eastern Ontario included the following pen and paper items: 1) CF Carrier Testing Survey, 2) MBSS, 3) STAI, and 4) a CF disease

description (Appendix D). The package also contained an introductory letter explaining the study's purpose, time requirements, confidential nature, and instructions (Appendix E). A consent form to participate in the study (Appendix F) was also completed by participants, in the presence of the researcher, prior to completion of a questionnaire package.

The steps required to complete the questionnaire package were outlined in detail in the introductory letter to participants. To begin, participants were asked to read through a CF disease description which gives a brief overview of a "typical" disease process over the lifetime of a person with CF (Appendix D). Notably, this CF disease description does not give factual information about genetic inheritance or relative risk, which might bias responses to the knowledge portion of the CF Survey. The disease description has been reviewed in the past for comprehensiveness, unbiasedness and clarity by a CF focus group comprised of 8 parents of children with CF and 4 adults with CF (Surh et al., 1995). Next, the introductory letter asked each participant to complete the aforementioned pen and pencil measures of affect, knowledge, health beliefs, intended behaviours and personal coping style in the following order: 1) CF Carrier Testing Survey, 2) MBSS, 3) STAI. Finally, participants were asked in the letter to return the completed package to the investigator in the self-addressed, stamped envelope which was provided to them. Participants who did not return survey packages within three weeks from date of contact were followed-up with a telephone call to ask if they still intended to complete the survey.

Results

A total of 258 individuals were approached to participate in the survey, of which 133 returned the completed package, yielding an overall participation rate for the study of 50%. The participation rate differed according to gender, with more women (63%) than men (54%) returning the questionnaire. Only 33 individuals of the 258 (12%) approached declined participation at the outset, and the most common reasons given for not participating included: 1) no stated reason (30%), 2) self-expressed inability to write/read English at a level sufficient to comprehend and complete the questionnaire (21%), and 3) too busy (18%).

Demographics & Awareness of CF Carrier Screening

Demographic statistics for the sample are presented in Table 1. In relation to awareness about CF carrier screening, most individuals *had not* heard of the test (60%, n=80) prior to filling out the questionnaire. Of the remaining individuals who *had heard* of carrier screening, the most frequently cited sources of information were the media (26%, n=34) and scientific literature (11%, n=14). A slight majority of individuals (56%, n=75) indicated that they *would not consider* testing; the two most commonly endorsed reasons for this decision included having no family history of the disease (41%, n=55), and having finished their family (38%, n=51). Of those who indicated that they *would consider* testing (44%, n=57), the two most frequently endorsed reasons were to find out their chance of being a carrier (29%, n=38), and to find out their children's chances of

being carriers (28%, n=37). The situations cited as most influential of one's decision to undergo carrier screening were having a family member diagnosed with CF (65%, n=86), finding out that a family member is a carrier (64%, n=85), and pregnancy (26%, n=34). Despite the fact that a majority of individuals indicated that they would not consider testing for themselves, 87% (n=115) of the sample said that they would tell others about the availability of testing, including "people in general" (53%, n=70), siblings (50%, n=66), and their spouse (48%, n=64). The two people cited as most influential of one's decision to undergo screening were family doctor (37%, n=49) and spouse (34%, n=45).

Insert Table 1 about here

Relationship of the HBM Variables to Intention to Be Tested

Scale Properties

Given that the development of the HBM items (except for knowledge items) required modifications and/or additions to the original CF questionnaire, the subscales of severity, susceptibility, barriers and benefits were considered for internal reliability (Table 2). Acceptable standardized Cronbach alpha coefficients were demonstrated for the items comprising the severity subscale and the benefits subscale. Coefficients for the subscales of barriers and susceptibility were modest, at best. For the purpose of analyses, the item values within each of the subscales were standardized and summated, and then a standardized composite score was calculated for each of severity, susceptibility, barriers

and benefits. In the case of knowledge items, a percentage correct score was calculated for each individual, based on the number of correct responses she/he indicated out of eight questions.

Insert Table 2 about here

Relationship of the HBM Variables to Intention to Be Tested

Hypothesis 1

The hypothesis concerning the relationship of the HBM variables to intention to be tested for carrier status was tested using logistic regression procedures (Hosmer & Lemeshow, 1986; SPSS for Windows, Standard Version 6.1). Univariate analyses of the HBM variables, six unique cross-product terms and the knowledge score suggested that only the composite scores for susceptibility, severity and benefits should be considered in the logistic regression model. Using forward-stepping likelihood ratio procedures, the three HBM components were considered. Results demonstrated that the model was highly significant ($\chi^2_{(3)} = 24$, $p < 0.001$), with 70% of cases classified correctly (Table 3).

Insert Table 3 about here

The relationship between the HBM variables and continuous demographics (i.e., age, number of children, state and trait anxiety, income) was also considered using linear regression. For purposes of analyses, gender was coded 0 and 1, and treated as a continuous variable in the regression equation (Tabachnick & Fidell, 1989). A small correlation was observed between susceptibility and age, however, no other significant results were found (Table 4). MANOVA procedures using religion and marital status as independent variables showed no significant main effects or interactions with the HBM variables.

Insert Table 4 about here

Relationship of the Monitor and Blunter Subscales to Intent to Be Tested

Hypothesis 2

Descriptive statistics for the monitoring and blunting subscales are shown in Table 5. Both subscales and their interaction were considered in relation to intent to be tested for carrier status using logistic regression procedures. Based on a liberal univariate inclusion criteria of $p < 0.25$ for individual predictors (Hosmer & Lemeshow, 1986), preliminary results demonstrated that only the blunting subscale was appropriate for inclusion in a logistic regression model. However, the overall model using the blunting subscale as a

predictor was not significant ($\chi^2_{(1)} = 1.45$, ns).

Insert Table 5 about here

In order to test the specific hypothesis that individuals with a characteristic monitoring coping style (i.e., high monitors) would be more likely to consider testing than individuals with a characteristic blunting coping style (i.e., high blunters), each of the two subscales was dichotomized into high and low scores using a median sample split (Miller, 1987). Preliminary univariate results suggested that both the dichotomized monitoring and blunting subscales should be further explored as predictors. However, analysis of the model did not yield significant prediction ($\chi^2_{(2)} = 5.2$, ns).

Multiple regression analyses of the monitoring and blunting subscales on demographic variables indicated no relationship with any of age, gender, income, state or trait anxiety. However, a minimal relationship between blunting score and number of children was observed (Table 6). Results of a MANOVA analysis indicated no main effects or interactions between monitoring or blunting and religion or marital status.

Insert Table 6 about here

Relationship of HBM Variables, Monitoring and Blunting Subscales

Hypothesis 3

The relationship between HBM variables and the monitoring and blunting

subscales was explored using linear regression procedures. As shown in Table 7, neither the monitoring subscale nor the blunting subscale was significantly related to perceptions of severity, susceptibility, barriers, benefits or knowledge ($F_{(5, 116)}=0.611$, ns; $F_{(5, 116)} = 1.18$, ns, respectively).

Insert Table 7 about here

Relationship of Demographic Variables to Intent to Be Tested

Results of a univariate logistic regression with demographic variables showed that only age and marital status should be considered in a logistic regression model. Using forward-stepping likelihood ratio procedures, only age was shown to be a significant predictor of intent to be tested ($\chi^2_{(2)} = 11.28$, $p < 0.01$), with the model classifying 65% of cases correctly. As noted in Table 8, the relationship of age to intent to be tested was inverse.

Insert Table 8 about here

Overall Prediction Model of Intent to Be Tested

As expected, forward-stepping logistic regression procedures using age, susceptibility, severity and benefits as predictors revealed a significant overall model ($\chi^2_{(4)} = 31.4$, $p < 0.001$). In total, 72% of cases were classified correctly (Table 9). Notably, the

correlation between age and susceptibility does not preclude inclusion of both variables in the model. Rather, stepwise logistic regression eliminates the covariate component of the two variables and indicates the extent to which each variable is independently related to the intention to use CF carrier screening (c.f., Hosmer & Lemeshow, 1986).

Insert Table 9 about here

Synopsis of Results

In summary, the following results were observed in the present study:

- 1) Increased perceptions of benefits and susceptibility were positively related to individual intent to use CF carrier screening.
- 2) Perceptions of severity were also related to the intent to use testing, although the relationship was inverse.
- 3) No support was found for the predictive value of the monitor/blunter dimension in relation to genetic testing decisions or to the HBM.
- 4) Age was the only significant demographic predictor of intent to be tested for carrier status, and the relationship was inverse.
- 5) An overall prediction model including age, perceived susceptibility, severity and benefits was highly significant and classified 72% of cases correctly.

Discussion

We cannot choose an option if we are not aware that it exists. Moreover, before we can make a decision which is in line with our personal goals, values and morals, we must be able to compare and contrast among all options available to us. In this regard, knowledge and understanding are the cornerstones of rational decision-making, informed consent and, ultimately, of autonomy. Where predictive genetic testing is concerned, the advent of carrier testing for CF has provided prospective parents with a host of new options to make more informed reproductive decisions.

Yet, while the factual information about carrier screening results can readily be presented to individuals, past work has shown that the information is not always interpreted by counselees in an objective manner (Becker et al., 1994, Watson et al., 1991), or even retained in the future (Becker et al., 1994; Besson & Golbus, 1985; d'Ydewalle & Evers-Kiebooms, 1987; Rona et al., 1994). In fact, it has been suggested that the subjective version of the information gleaned from genetic counselling sessions is what people actually use in making reproductive decisions (Lippman-Hand & Fraser, 1979a; Lippman-Hand & Fraser, 1979b).

Understanding the health perceptions and personal coping styles which colour individual interpretation of genetic testing information may be a preliminary step in identifying how to aid individuals in the decision process leading up to testing, and facilitate understanding, interpretation and retention once results are known. In the case

of carrier testing for CF, however, the specific health beliefs and personal coping styles which may impact on the genetic testing decision process have not been explored. In response to this gap in the CF carrier testing literature, the main focus of the present study was to test the relationship between individual intent to be tested for CF carrier status, the HBM variables of susceptibility, severity, benefits and barriers, and individual propensity to either seek out or avoid threat-related information. In addition, the relationship between the HBM and the Monitor/Blunter theory was considered.

Awareness of and Attitudes Toward CF Carrier Screening

The finding that over one half of the present general population sample had not heard of CF carrier testing was not surprising, given that testing is currently only available to those with a family history of the disease. Notably, the most common source of information for those who had heard of the test was the media. This finding suggests that media campaigns may provide one effective way to reach individuals with accurate information, should population testing ever become available.

Perhaps the most interesting finding in regard to attitudes explored was in relation to support for CF carrier testing and dissemination of information about its availability. Despite the fact that only a minority (40%) of individuals stated that they would be interested in finding out their carrier test results, the vast majority (87%) of participants stated that they would tell other people about the availability of testing. These results lend further support to the findings of other researchers. For example, Decruyenaere et al.

(1992) noted in their sample that while the majority of individuals had no objections to carrier screening for CF, the proportion of individuals who stated an interest in knowing their own carrier status was lower. Likewise, Ten Kate & Tijnstra (1990) found that 80-96% of individuals were in favour of CF carrier testing, however, only 54-77% of them were interested in knowing their own carrier status. Therefore, while it appears that the majority of general population individuals see value in telling others about the availability of carrier testing, considerably fewer individuals see value in finding out their own carrier status.

The Importance of the Health Belief Model Variables

Perceived Susceptibility and Benefits

In general, the present study provides support for several HBM variables in predicting who may use CF carrier testing. As predicted, those who perceived themselves as more susceptible to carrier status and who saw more benefits to carrier testing were more likely to report that they would consider testing. Both of these findings are in line with the few studies that have examined the HBM in a genetic testing context. For example, in their study of the factors distinguishing between those who accepted an invitation for Tay-Sachs carrier screening and those who did not, Becker and colleagues (1975) found that participants in the screening program saw themselves as more susceptible to being carriers of the Tay-Sachs mutation. Likewise, a positive relationship between parents' intentions to use prenatal screening for cleft lip/palate and their

perceptions of susceptibility has previously been demonstrated (Sagi et al., 1992). These results lend support to work with the HBM in other domains (e.g., mammography screening, diabetes regimens), where perceived susceptibility and perceived barriers have also been significant predictors of health behaviour (Janz & Becker, 1984).

Perceived Severity

Results for the severity scale, although significant, were not in the hypothesized direction. Those who saw carrier testing as more severe were actually *less* likely to report an intention to be tested. Although unexpected, these results are similar to the findings of Bekker et al. (1975), who found that increased perceptions of severity had an inhibitory effect on participation in a Tay-Sachs screening program. As suggested by these researchers, it may be that while a low or moderate estimation of severity is necessary to prompt individuals to take action, the perception that being a carrier would be highly disruptive of future family planning may have an inhibiting effect on participation. Hence, there is perhaps an "optimal level" of fear or motivation associated with readiness to act. Rosenstock (1979) has also suggested this concept in relation to genetic counselling situations, where he notes that "too little or too much seriousness" (p.140) can produce a response that is inappropriate to the situational context.

Becker and colleagues also found that spousal perceptions of severity had an impact on individual behaviour in relation to screening. Although these findings were not directly tested in the current study, one third of the present sample did indicate their

spouse as someone who might influence their decision to be tested. Given that the implications of positive carrier status are only relevant to reproductive planning, it is logical that the opinions of one's partner would be highly pertinent where the decision to be tested is concerned. Further, these results are consistent with the results of past studies which have shown that approval by important others is strongly associated with the use of genetic testing (Wertz et al., 1992). Therefore, notwithstanding the importance of individual perceptions, the current findings underscore that the decision to undergo carrier testing is likely influenced by the norms and pressures of one's social group, and particularly by one's spouse.

To summarize, the results of the present study, in tandem with the work of Becker and colleagues tend to support some type of relationship between perceived severity and intention to use carrier testing. However, both sets of findings diverge from the conclusions of the majority of past research, which has found severity to be the poorest predictor of health behaviour (c.f., Janz & Becker, 1984). This apparent contradiction in findings may actually be a reflection of the nature of the health threat. Consider that while other diseases may unanimously be perceived as severe (e.g., cancer, heart disease), perceptions of the severity of CF/Tay-Sachs carrier status are likely quite variable - probably as a result of poor general knowledge. This assertion is supported by the fact that most individuals in the present study had never heard of carrier testing prior to the study, and had poor knowledge about CF and its inheritance. This general lack of

knowledge among the sample is not surprising, given that carrier screening (and therefore information about it) has not been made widely available to the general public. In turn, it is probable that individual perceptions about the severity of being a CF carrier are largely based on speculation, and therefore subject to more variability than perceptions of more common and widely publicized diseases, such as cancer.

Perceived Barriers and Knowledge

The finding that perceived barriers was not related to the intent to be tested is not consistent with past work, particularly in the area of mammography screening use, which has found considerable support for a negative relationship between barriers and health action. From a methodological standpoint, the present findings concerning barriers may be related to the low internal reliability for the barriers subscale ($\alpha=0.57$). However, aside from the obvious methodology issues, there may be an alternative explanation. It is plausible that individuals are simply not consistent about their perceptions of barriers to CF testing. Again, this assertion is supported by the fact that in the general population individuals typically have poor knowledge about testing and what it entails, and perhaps about what disadvantages it might pose. Further, it may be that the most important barrier to carrier testing for CF is, as Surh and colleagues (1995) suggest, perceived inconvenience - a variable which was not directly measured in the current study.

As with the barriers hypothesis, the prediction that an increased knowledge of CF would predict intent to use carrier status was not supported. In the past, it has been

suggested that lack of knowledge is generally a barrier to medical testing (Deber, 1994). Likewise, it has been asserted that the general population's lack of knowledge about CF carrier testing may pose a barrier to being tested (Youn, 1996). Clearly, it is difficult to support or refute either of these claims without a direct comparison to a control group of high risk participants. However, it appears that knowledge about CF and carrier status among this general population sample was independent of the intention to undergo CF carrier screening.

The Monitor/Blunter Theory in Relation to Carrier Testing Decisions

Despite its impressive track record in a variety of health care contexts, the MBSS did not discriminate in the current study between those who would and would not consider testing for CF carrier status. The most obvious explanation for why there was no observed relationship between coping style and the intent to use carrier testing may be that personality variables simply do not mitigate the decision to use CF carrier testing services. If taken in this context, the current results lend support to past findings (e.g., Bekker et al., 1993; Surh et al., 1995; Youn, 1996) which have demonstrated the ultimate importance of factors such as convenience, and immediate offer of testing by a health professional.

On the other hand, it may be that some personality variables truly do predict intent to use carrier testing, but that those variables were not measured in this study. Perhaps the importance of personality in relation to genetic testing lies not in the monitor/blunter

dimension, but in some other personality dimension, or a complex interaction of many personality variables. Preliminary work by Chase et al. (1995), for example, found that individuals with a low tolerance for test uncertainty (as in the case of CF carrier testing which is neither 100% sensitive nor 100% specific) were twice as likely to decline the offer of CF carrier testing made to them, as compared to those with a high tolerance for test uncertainty. Notwithstanding their significant findings about this personality dimension, Chase and colleagues still concluded that those who had a high tolerance for test uncertainty probably did not understand and accept the uncertainty associated with CF carrier testing. Rather, the authors speculate that these individuals may have accepted the uncertainty of the test because it fulfilled the expectations of the medical practitioner involved (Chase et al., 1995). Thus, even when personality variables do enter into the decision to be tested for carrier status, they may be overshadowed by perceptions about what the health practitioner expects, in addition to how convenient the testing is.

Aside from these two explanations, there may be third reason why the said hypothesis about the monitor/blunter theory was not confirmed. As previously described. Miller does not purport that monitoring/blunting behaviour surfaces in everyday events. Rather, she emphasizes that differences between monitors and blunters emerge in response to a perceived threat (Miller et al., 1988; Miller, 1990; Miller, 1992). Thus, where there is no perceived threat, there is likely no evidence of differences between monitors and blunters on intention to complete a health behaviour.

A salient example of the importance of perceived threat in relation to the monitor/blunter theory is found in the work of Phipps and Zinn (1986), who studied a sample of women undergoing amniocentesis. As noted earlier, these authors found that monitors undergoing amniocentesis experienced significant increases in subjective arousal before and during the procedure, as compared to characteristic blunters. However, when the results of the amniocentesis were known, the monitors' anxiety levels returned to normal. These findings underscore the idea that propensity to either seek out or avoid threat-related information is a function of *perceived* threat, and not simply a response to the presence or absence of an objective stimuli. Thus, the non-significant relationship demonstrated between coping style and intent to be tested may suggest that being a CF carrier (a state which has no direct effect on personal health) is not perceived as threatening by general populations individuals.

Even if the carrier testing decision is generally perceived as severe, the nature of the present study may not have afforded a true reflection of the perceived threat, because participants were only asked to indicate their *intent* to use carrier testing (i.e., "Would you consider testing for CF carrier status?"). In this regard, the testing decision was purely hypothetical, and perhaps too removed from any potential threat that an *actual* testing situation might pose.

The HBM in Relation to the Monitor/Blunter Theory

If one accepts the argument that the proposition of CF carrier testing did not pose

a significant threat to participants in the current study, then the same argument provides a rational explanation for why no relationship was demonstrated between coping style and the HBM - especially in regard to perceived severity. Again, if individuals do not perceive being a carrier for CF as severe, then there is no reason to suspect that differential coping styles will be observed, and no opportunity to observe a relationship between health perceptions and coping style.

It is important to note, however, that these results run contrary to the recent work of Schwartz and colleagues (Schwartz, Lerman, Miller, Daly, & Masny, 1995), particularly in relation to perceptions of susceptibility. These authors cite a strong relationship between dispositional attentional style (i.e., monitoring), and the susceptibility perceptions of women who had a first degree relative with ovarian cancer. Results showed that high monitors (as opposed to low monitors) were most likely to overestimate their risk of ovarian cancer (regardless of their objective risk status), and suffer from increased psychological distress. The authors suggest that as a result of their tendency to overestimate their susceptibility to ovarian cancer, high monitors likely experience unnecessary psychological distress. Further, they assert that high monitors may be more motivated to seek out prophylactic surgery or genetic testing for susceptibility, despite the fact that their objective risk level may not warrant such drastic measures.

Clearly, the results of the current study do not support those of Schwartz and colleagues, since in the current case there was 1) no observed relationship between health

beliefs and coping disposition, 2) no observed relationship between monitoring and intent to be tested for carrier status, and 3) no differences between high and low monitors on anxiety levels. Yet, comparison of the present study to the work of Schwartz and colleagues (and indeed to the work of most researchers surrounding the monitor/blunter theory) is limited by a fundamental distinction. In contrast to ovarian cancer (or cardiac catheterization, colposcopy, outpatient surgery, etc.), carrier testing for CF does not involve a direct threat to one's own health. The only threat associated with being a CF carrier is to one's reproductive health, and then only if one's partner is also a carrier. In this way, the study of carrier testing for CF is a unique situation: it is a "once removed" perspective on health action. Again, the non-significant relationship between health perceptions and coping style in the present study lends further credence to the assertion that carrier status may not be seen as a severe health stressor by general population individuals.

Taken together, the current results and those of Schwartz et al. (1995) suggest that the monitor/blunter dimension may be a useful framework for considering how people cope with genetic testing for illnesses which pertain to their own well-being. On the other hand, it appears that situations which do not affect one's own health status may not be perceived as particularly threatening - and therefore these situations do not elicit characteristic monitoring and blunting behaviours. Indeed, the monitor/blunter framework may be more relevant to counselling protocols *after* the initial decision process and/or later in the process, during times of increased perceived threat (e.g.,

pregnancy, prenatal testing).

The Relationship of Demographics to Intent to Be Tested for CF Carrier Status

In brief, it was not surprising that only age added predictive value to the hypothesized model of intent to use carrier testing, since past work has not shown demographics to be an important factor in the decision to use carrier testing. Further, the inverse relationship between age and intent to use carrier testing is logical, since younger individuals are also less likely to have finished their families. It is also noteworthy that age was modestly and inversely correlated to the susceptibility subscale. In relative terms, older individuals appear less likely to consider testing.

Implications for Counselling

The results of the present study show that younger age, decreased perceptions of severity, and increased perceptions of susceptibility and benefits may be important factors in the decision to use genetic testing for CF carrier status. However, at this point, it is important to emphasize a key point. Although it is hoped that this study has contributed to a better understanding of what motivates people to use CF carrier testing, the purpose of this work was not to examine how to manipulate uptake rates. A considerable body of work in this field has already demonstrated that near perfect uptake rates can be achieved through the offer of immediate testing by a professional. Moreover, these results have been demonstrated in both high risk (e.g., Surh, 1994, Youn, 1996) and general population samples (e.g., Bekker, 1994, Wright Clayton et al., 1996). Even in samples of pregnant women, where consistently high uptake rates have been

demonstrated (e.g., Mennie et al., 1992), convenience of testing appears paramount, since interest in carrier screening declines markedly if a minimal fee (\$50), or a short travel distance is introduced (Asch, Patton, Hershey, & Mennuti, 1993).

Rather than adding to the existing knowledge base surrounding uptake rates, the purpose of the present study was to explore individual factors which may mitigate the decision to be tested in order to develop more effective genetic counselling protocols. Implicit in the interpretation of the current results is the idea that a "successful" genetic testing program should not be contingent on high uptake rates, since these are easily affected by method of presentation (Surh et al., 1995). Likewise, "success" cannot be defined in terms of by positive attitudes toward testing and/or a willingness to tell others about its availability, since these variables do not necessarily predict that individuals will use the test for themselves (see page 58).

The present study proceeded on the premise that the success of carrier testing for CF lies in the extent to which those who present for testing (of their own accord) are able to make decisions which are informed and consistent with their beliefs. In this regard, the value of understanding the personality factors and perceptions that affect genetic testing decisions rests in helping individuals to form a better understanding of what they might gain from test results, and assist them in making decisions which are in line with their belief systems - all while minimizing unnecessary anxiety. For if clients come for counselling with preconceived notions (as presumably they do) about the severity of carrier status, their own susceptibility to it, and what the benefits of screening are, then

the information they hear from a genetic counselling is certainly coloured by their pre-existing frame of reference (Rosenstock, 1979; Welshimer & Earp, 1989). Therefore, a move toward studying the personality variables and health beliefs which predict CF carrier testing is not redundant; rather, it is a first step in the development of a new way to measure success - one that is independent of uptake rates.

It is also important to note that this call for more personalized genetic counselling is somewhat at odds with the currently accepted practice of "non-directive" genetic counselling. In brief, the principle of "non-directive" genetic counselling involves imparting knowledge to individuals in the most consistent and objective way possible, to avoid suggestion by the counsellor that one option is better than another (Welshimer & Earp, 1989). From the current perspective, the practice of non-directive counselling may be detrimental because it lacks the specificity needed to ensure that information is personalized in an accurate and helpful way. While objectivity may still be emphasized, counselling and decision-making may be enhanced by using a strategy of "focused" counselling, as suggested by Welshimer and Earp (1989):

Targeted at the specific reproductive attitudes, beliefs, and expectations individuals bring to counselling, focused counselling may improve peoples' understanding of the counselling information and its subsequent use for making sound and personally satisfying reproductive decisions (p. 241).

Thus, within the context of genetic counselling for CF carrier screening, the HBM may provide a useful departure point from which to begin exploring individual perceptions and testing the efficacy of counselling protocols which are tailored to meet individual need.

Methodological Limitations of the Study

The Design and Sample

The most important shortcoming of this study is the fact that the study presented a hypothetical testing situation to participants, and only asked about their intentions to be tested. In this way, it was impossible to measure intention as it related to some behavioural criterion. As mentioned earlier, the hypothetical situation that was created in the current study may have produced results that were too far removed from any threat that an actual testing situation might pose. The wording of the question posed to participants may also have been problematic in that answering yes to "Would you consider testing..." may not have indicated an actual intent to use carrier screening. This is illustrated in the response of one participant who tempered his "yes" response with this note: "I would only consider testing. I'm not sure that I would have it". Thus, further examination should necessarily include examination of actual behaviour in carrier testing situations rather than intent, such that the contribution of health perceptions and coping style can be assessed in actual clinical context.

Ideally, an analysis of the impact of coping style and health beliefs on carrier screening decisions should include a longitudinal perspective which spans the entire

decision-making process, including subsequent reproductive decisions. In theory, this type of analysis would represent a research shift in this area, from asking the question, "*who comes for testing*" to asking the question, "*how can we enhance the full range of experience for individuals who come for testing?*". From a clinical perspective, understanding the entire spectrum of the decision-making process is key to providing the necessary medical and emotional support to aid individuals in reaching their desired reproductive outcome.

A second critique of this current work pertains to the homogeneity of the sample. Specifically, the results are of limited applicability, since the participants were predominantly English-speaking, well-educated and in the mid-upper income bracket. Certainly, this limits generalizability of the current results to individuals with different cultural and socio-economic and educational backgrounds. Perhaps more importantly, the vast majority of participants were parents, therefore it is impossible to generalize these results to individuals without children. The opportunity to observe differences between individuals with and without children would surely provide more insight into how these two groups compare and contrast, in terms of their health perceptions and coping styles. For example, carrier testing should have more relevance for people who are in the planning stages of having a family (Becker et al., 1975; Goldstein, Greenwald, Nathan, Massarik, & Kaback, 1977). One might also expect that the experience of being a parent would be highly influential of reproductive attitudes, and therefore significant where

subsequent reproductive decisions are concerned. In addition, when compared to people without children, parents may perceive different benefits to carrier testing (i.e., for their children's reproductive plans), particularly if they have finished their family.

It must also be noted that demand characteristics may have influenced the responses of participants in this sample, since all participants were attending the hospital in order to receive a service for the children in their care. Further, the Children's Hospital of Eastern Ontario is a well-known institution in the community, which is noted for its successful research endeavours and fundraising campaigns for children. In this regard, participants may have felt compelled to: 1) participate in the study to "help research for children" and 2) respond in a positive way to the study questions, particularly on the CF Questionnaire. Respondents may have felt it socially desirable, for example, to say that they would tell others about the availability of genetic testing for CF carrier status - not because they view testing as a particularly useful service, but rather because they wanted to show support for the services offered at the Children's Hospital. Hence, a more representative reflection of the health beliefs and coping styles of general population individuals in relation to genetic testing might be achieved if participants were approached in more "neutral" settings (e.g., malls, shopping centres, mail-out surveys).

A better design might also have included a comparison of both general population and high risk individuals. It is reasonable to hypothesize that the health beliefs (especially those surrounding susceptibility) of high risk populations would be markedly different than

the general population's, given that the former may have experience with CF, and increased knowledge about CF, resulting from their known family history. One might also speculate that the carrier testing situation is more threatening for high risk individuals, again because of their family history and personal experience with the disease. Nonetheless, future study which compares high risk individuals and general population individuals is imperative if a better understanding of the role of health beliefs and coping style is to be fostered.

Lack of a Validated Measure for The HBM

One of the most important methodological considerations concerns the HBM, and the lack of a validated measurement device. In particular, the low internal reliability coefficients observed for the susceptibility and benefits subscales may suggest either inconsistencies on behalf of the participants, or inconsistencies with the measurement devices. In either case, the interpretation of low reliability coefficients must be qualified with the knowledge that potential relationships (and therefore predictive value) may have been overestimated. Hence, it is important that any future work which seeks to explore the HBM, and its value in CF carrier screening protocols, be undertaken with a well-validated instrument which captures its theoretical underpinnings.

Conclusion

*The prudent text-books give it
In tables at the end-
The stress that shears a rivet
Or makes a tie bar bend
What traffic wrecks macadam
What concrete should endure
But we, poor sons of Adam,
Have no such literature,
To warn or make us sure!*

Rudyard Kipling, *Hymn of Breaking Strain*

Perhaps more than any other medical technology, predictive genetic testing raises a myriad of complicated social, ethical, legal and economic questions which, for the most part, have no apparent answers. While the technical aspects of genetic testing progress at speeds nearly too fast to document, the emotional, psychological facets of the testing process have been slow to catch up. In the case of carrier screening for CF, state of the art technology is available to provide individuals with reproductive options. Further, we now understand that individuals can be recruited for carrier screening simply by manipulating how, when and where the testing is offered to them. Yet, regardless of how one defines "effective" use (i.e., more informed reproductive choices and/or avoidance of the birth of an affected child), we must realize that it is neither logistically nor economically viable to spend precious resources on enticing individuals to use testing services if the information they take from their experience is not remembered, or

remembered incorrectly - because if this is the case, then information is wasted - no matter what the advocated use is.

Clearly, the CF carrier screening experience has not provided the anticipated "ideal model" for other genetically-based diseases. However, to a certain extent, a model has been built for the types of questions that must be asked before carrier screening can be offered in a socially and economically responsible way. Perhaps the next step in building the CF carrier screening model is to focus on research that will address the personal factors which individuals bring to testing situations, and explore how clinicians can use this information to enhance and maximize the genetic testing experience.

The current study argues that the "measure of success" for a carrier screening protocol lies in the ability of its genetic counselling component to support clients in making decisions that are analogous to their personal beliefs and values. Further, it is suggested that an effective genetic screening program is one that can maximize understanding and retention of results and options, and minimize undue anxiety. The HBM provides a promising framework for developing personalized counselling protocols which would assist clients, particularly during the decision to undergo carrier testing. On the other hand, it is proposed that the monitor/blunter theory may be more relevant later in the carrier screening process, particularly during times of increased anxiety (e.g., pregnancy and/or prenatal diagnosis), or in situations where carrier screening is of personal relevance to one's own health (e.g., breast/ovarian cancer, Huntington disease).

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Table 1
Demographic Characteristics of the Sample

	<u>Mean</u>	<u>SD</u>	<u>Median</u>
<u>No. of Children</u>	2	1	2
<u>Age in years</u>	36	7	37
<u>State Anxiety</u>	47	8	46
<u>Trait Anxiety</u>	50	10	50
	<u>% of sample</u>	<u>n</u>	
<u>Gender</u>			
Male	38	(51)	
Female	62	(82)	
<u>Income</u>			
<\$20 000	10	(18)	
\$20 000-\$39 000	20	(27)	
\$40 000-\$59 000	18	(24)	
\$60 000-\$79 000	14	(18)	
\$80 000-\$99 000	7	(10)	
>\$100 000	12	(16)	
<u>Religion</u>			
Roman Catholic	44	(59)	
Protestant	40	(30)	
No preference	16	(21)	
Other	7	(9)	
<u>Education</u>			
Less than High School	5	(6)	
High School	14	(14)	
College Diploma /Trade School	39	(52)	
University Degree	26	(35)	
Graduate Degree	15	(20)	
<u>Language spoken in home</u>			
English	82	(107)	
French	15	(20)	
Other	3	(4)	

Table 2

Health Belief Model Subscale Statistics

Subscale (# of Items)	Max. Possible	Mean	SD	Alpha
1) Severity (4)	40	10.7	0.37	0.76
2) Susceptibility (4)	117	9.6	2.1	0.48
3) Barriers (6)	30	19.3	0.71	0.57
4) Benefits (8)	40	30.7	0.60	0.81
5) Knowledge (8)	8	4.1	1.60	0.49

Table 3

Prediction of Intent to be Tested for CF Carrier Status from Health Belief Model Variables

Predictor	Raw Coefficient	Odds Ratio
Severity	-0.59*	0.55 ^a
Susceptibility	0.72*	2.10 ^b
Benefits	0.51*	1.70 ^c
Overall χ^2 (3, n=131) = 24.5, p<0.0001		
Per cent cases classified correctly: 70		

*p<0.01

^a For every one unit increase in perceived severity, the likelihood that an individual will consider testing *decreases* by a factor of 0.59: therefore higher the severity score, the less likely an individual is to consider carrier testing.

^b For every one unit increase in perceived susceptibility, the likelihood that an individual will consider testing *increases* by a factor of 0.72: therefore, the higher the susceptibility score, the more likely individuals are to consider carrier testing.

^c For every one unit increase in perceived benefits, the likelihood that an individual will consider testing *increases* by a factor of 0.51: therefore, those who perceive more benefits are more likely to consider carrier testing.

Table 4

Correlation Between Health Belief Model Variables and Continuous Demographic Variables

	Severity	Susceptibility	Barriers	Benefits	Knowledge
Age	- 0.13	- 0.26**	0.08	- 0.02	-0.02
Gender	- 0.04	0.12	- 0.00	- 0.01	0.10
Income	- 0.05	- 0.12	0.04	- 0.06	0.06
No. of Children	- 0.05	- 0.11	- 0.03	0.09	- 0.16
State Anxiety	0.21	0.13	0.01	0.07	- 0.03
Trait Anxiety	0.17	0.10	0.07	0.14	- 0.03

Note: Correlation coefficients are point biserial for categorical demographic variables coded 0 and 1, and Pearson for continuous demographic variables.

**p<0.01

Table 5

Descriptive Statistics for Subscales of the Miller Behavioural Style Scale

	<u>Monitoring</u> (n=123)	<u>Blunting</u> (n=123)
Mean	9.5	3.5
Median	10	3.0
St. Deviation	3.2	2.1
^a No. above median	65	57
^b No. below median	58	66

^a number of "high" monitors/blunters

^b number of "low" monitors/blunters

Table 6

Correlations Between Monitor and Blunter Scores and Continuous Demographic Variables

	Monitoring Score	Blunting Score
Age	-0.12	0.17
*Gender	-0.03	0.04
Income	-0.11	0.02
No. of Children	-0.05	0.28**
State Anxiety	-0.02	0.00
Trait Anxiety	0.17	0.02

***Note:** Correlation coefficients are point biserial for dichotomous demographic variables coded 0 and 1, and Pearson for continuous demographic variables.

** $p < 0.01$

Table 7

Correlation Between Health Belief Model Variables and The Monitoring and Blunting Subscales

	Severity	Susceptibility	Barriers	Benefits	Knowledge
Monitoring Score	-0.06	-0.01	-0.08	0.06	0.13
Blunting Score	-0.04	-0.06	0.15	0.02	0.14

Note: All coefficients are Pearson

Table 8

Prediction of Intent to Be Tested for CF Carrier Status From Demographic Variables

Predictor	Raw Coefficient	Odds Ratio
Age	-0.08*	0.92 ^a
Marital Status	-0.13	0.87
Overall χ^2 (2, n=131) = 11.3, p<0.01		
Per cent cases classified correctly: 65		

*p<0.01

^a For every one year increase in age, the likelihood of an individual considering testing decreases by a factor of 0.08: thus, for a five year increase in age, the likelihood that an individual will consider testing drops from 0.92 to 0.67, where $0.67 = [(-0.08 \times 5)^{-1}]$.

Table 9

Prediction of Intent to be Tested for CF Carrier Status from Health Belief Model
Variables and Demographics

Predictor	Raw Coefficient	Odds Ratio
Severity	-0.86*	0.42 ^a
Susceptibility	0.53*	1.70 ^b
Benefits	0.54*	1.70 ^c
Age	-0.10*	0.91 ^d
Overall χ^2 (4, n=131) = 31, p<0.0001		
Per cent cases classified correctly: 72		

*p<0.01

^a For every one unit increase in perceived severity, the likelihood that an individual will consider carrier testing *decreases* by a factor of 0.86; therefore, people who see carrier status as more severe are more likely to consider testing.

^b For every unit increase in perceived susceptibility, the likelihood that an individual will consider testing *increases* by a factor of 0.52; therefore, people who see themselves as more susceptible to being carriers are more likely to consider testing.

^c For every unit increase in perceived benefits, the likelihood that an individual will consider testing *increases* by a factor of 0.54; therefore, people who see more benefits to carrier testing are more likely to consider testing.

^d For every one year increase in age, the likelihood that an individual will consider testing *decreases* by a factor of 0.10; therefore, as people age, they are less likely to consider carrier testing.

Appendix A
CF Carrier Testing Survey

GENETIC TESTING
FOR
CYSTIC FIBROSIS (CF)

For information:

Dr. Mario Cappelli or Barbara Virley O'Connor
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Children's Hospital of Eastern Ontario
401 Smyth Road
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GENETIC TESTING FOR CYSTIC FIBROSIS (CF)

We would like you to participate in a study by the Children's Hospital of Eastern Ontario. The study is about genetic screening for the disease, cystic fibrosis (CF).

A new genetic test is available which lets people know if they carry a CF disease gene. We are interested in your opinions concerning genetic testing for CF carrier status and how useful you might find the test results. The information you provide to us by completing the attached questionnaire will help us to better understand people's views surrounding this issue and to improve our service in the future.

It should take about 40 minutes of your time. Individual responses will be kept strictly confidential; your name will not appear with the responses you give.

Your assistance with this survey is greatly appreciated. Thank you for your time!

Before you begin, remember that you do not have to answer any questions you would rather not answer.

The following questions refer to the disease known as cystic fibrosis or CF. Choose the single best answer from the list provided. Circle one letter only for each statement.

1. Cystic fibrosis (CF) is:
 - a. a disease causing breast cysts
 - b. a disease affecting muscle control
 - c. a disease affecting lungs and digestive system
 - d. a rare type of bone disease.
 - e. don't know

2. The cause of CF is:
 - a. a virus illness
 - b. an inherited illness
 - c. a toxic medicine taken during pregnancy
 - d. a genetic accident during pregnancy
 - e. don't know

3. A CF "carrier" is:
 - a. someone who has only mild symptoms of CF
 - b. someone who does not have CF but who could have a child with CF
 - c. someone who does not have CF disease now but will develop it later
 - d. all of the above
 - e. don't know

4. CF "carriers" are diagnosed by:
 - a. their chest problems (i.e. asthma, colds)
 - b. a "sweat" test
 - c. a blood test
 - d. All of the above
 - e. Don't know

5. On average, individuals with CF live:
- a. until infancy
 - b. until childhood
 - c. until adulthood
 - d. people with CF live a normal lifespan
 - e. don't know
6. If there is no family history of CF, the chance of being a CF carrier is:
- a. 4 out of 4 (100%)
 - b. 1 out of 4 (25%)
 - c. 1 out of 25 (4%)
 - d. very low risk but not zero
 - e. no risk
 - f. don't know
7. Imagine that your neighbour and his pregnant wife find out that they are both CF carriers. What is the chance of their unborn child having CF ?
- a. 4 out of 4 (100%)
 - b. 1 out of 4 (25%)
 - c. 1 out of 25 (4%)
 - d. very low risk but not zero
 - e. no risk
 - f. don't know
8. What do you think is your chance of being a CF carrier?:
- a. 4 out of 4 (100%)
 - b. 2 out of 3 (67%)
 - c. 1 out of 4 (25%)
 - d. 1 out of 8 (12%)
 - e. 1 out of 25 (4%)
 - f. very low risk but not zero
 - g. no risk
 - h. don't know

9. How concerned are you about your own chance of being a CF carrier (as indicated in question 8?)

- a. not at all concerned
- b. slightly concerned
- c. moderately concerned
- d. very concerned
- e. extremely concerned

10. I think that my risk of being a CF carrier is (*please indicate by placing a mark on the scale, where 0=no risk and 100=certain risk*)

11. I think that the risk I indicated in question 10 is

- a. very low
- b. low
- c. moderate
- d. high
- e. very high

12. The following statements are about the possible effects of CF disease:

- i) CF patients usually need treatment every day
- ii) There is a cure for some CF patients
- iii) CF patients are less able to have children because of their disease

Please indicate the true statement or statements, by choosing one of the following answers:

- a. (i) only
- b. (ii) only
- c. (iii) only
- d. (i) and (iii)
- e. (ii) and (iii)
- f. don't know

In the following sections, there is information given to assist you in answering the specific questions. Please DO NOT GO BACK to previous questions or change your answers

II To help with the following questions, here is some information about CF:

- (1) CF disease is caused by having two CF genes.
- (2) A CF carrier is a person who does not have the disease, but does have one CF gene and one normal gene.
- (3) If two CF carriers have a child, then that child has a 1 out of 4 chance (25 %) of having the disease (i.e. the chance of that child getting a CF gene from each parent).

The following are some general questions about testing for cystic fibrosis carriers, also referred to as "CF genetic testing".

13. (a) Before today, have you heard about CF carrier (genetic) testing?

YES ☐ NO ☐

- (b) If YES, from whom or where did you hear? (You may check more than one).

- ☐ Parents
- ☐ Brothers/Sisters
- ☐ Aunts/Uncles
- ☐ Spouse/Partner
- ☐ Cousins
- ☐ Friends
- ☐ Family Doctor
- ☐ Genetic Counsellor
- ☐ Media (e.g., T.V., radio, newspapers)
- ☐ CF Foundation
- ☐ Scientific Literature
- ☐ Other (please specify) _____

14. (a) Would you consider having CF genetic testing?

YES ☐ NO ☐

(b) If YES, why? (*you may check more than one box*)

- ☐ To find out my chance of being a carrier
- ☐ To find out my children's chance of being carriers
- ☐ To find out my siblings' chance of being carriers
- ☐ To find out other family members' chance of being carriers
- ☐ To learn more about CF
- ☐ To help research
- ☐ To help find a cure
- ☐ Other (*please specify*) _____

(c) If NO, why not? (*you may check more than one box*)

- ☐ I have no known family history of CF
- ☐ I have finished my family
- ☐ I do not want to have children
- ☐ Other (*please specify*) _____

15. What person (e.g. relative, friend, etc.) might influence your decision to have CF genetic testing? (*you may check more than one box*)

- ☐ Parents
- ☐ Brothers/Sisters
- ☐ Aunts/Uncles
- ☐ Spouse/Partner
- ☐ Cousins
- ☐ Friends
- ☐ Family Doctor
- ☐ Genetic Counsellor
- ☐ No one, it is my choice
- ☐ Other (*please specify*) _____

16. What situation (e.g. marriage, pregnancy, etc.) might influence your decision to have CF genetic testing? (*you may check more than one box*)

- ☐ Family member found to be a CF carrier
- ☐ Family member diagnosed with CF
- ☐ Marriage
- ☐ Pregnancy
- ☐ No situation would influence my situation
- ☐ Other (*please specify*) _____

17. How do you think you would use the information from the CF genetic test? (*you may check more than one box*)

- ☐ For my family planning
- ☐ For my children's family planning
- ☐ For my sibling's family planning
- ☐ For other family members' family planning
- ☐ To prepare for the birth and care of a child with CF
- ☐ Spouse/Partner selection
- ☐ Have my spouse/partner tested
- ☐ I would not use the information from the CF genetic test
- ☐ I don't know how I would use the information
- ☐ Other (*please specify*) _____

18. (a) Would you tell anyone else about the availability of CF genetic testing?

YES ☐ NO ☐

(b) If YES, who would you tell? and why? (*you may check more than one box*)

- ☐ Parents
- ☐ Brothers/Sisters
- ☐ Aunts/Uncles
- ☐ Spouse/Partner
- ☐ Cousins
- ☐ Friends
- ☐ Acquaintances (e.g., coworkers, neighbours)
- ☐ Couples who are planning to have children
- ☐ People who ask
- ☐ No one
- ☐ Other (*please specify*) _____

(c) If NO, why not? (*you may check more than one box*)

- ☐ No chance to do so
- ☐ I don't know enough about CF genetic testing
- ☐ I don't want others to ask about my test results
- ☐ It is not my place to bring it up
- ☐ Other (*please specify*) _____

III Here are some possible advantages to CF genetic testing. For *EACH* of the following statements, please circle your own level of agreement.

		Strongly Agree 1	Agree 2	Neutral 3	Disagree 4	Strongly Disagree 5
19.	Information about my carrier status	1	2	3	4	5
20.	End worry and uncertainty	1	2	3	4	5
21.	Family planning	1	2	3	4	5
22.	Prenatal testing	1	2	3	4	5
25.	Spouse/Partner selection	1	2	3	4	5
26.	Lead to a cure	1	2	3	4	5
27.	Early detection of CF	1	2	3	4	5
28.	Help research	1	2	3	4	5

IV Here are some possible disadvantages to CF genetic testing. For EACH of the following statements, please circle your own level of agreement.

	Strongly Agree 1	Agree 2	Neutral 3	Disagree 4	Strongly Disagree 5
29. People who are carriers may be "labelled" or treated differently	1	2	3	4	5
30. Emotional upset	1	2	3	4	5
31. People may restrict themselves from having children	1	2	3	4	5
32. It may lead to abortion	1	2	3	4	5
33. Testing is expensive	1	2	3	4	5
34. Testing is expensive	1	2	3	4	5
35. It's better not to know	1	2	3	4	5

V Here are some opinions that people have about CF genetic testing. For EACH of the following statements, please circle your own level of agreement.

	Strongly Agree 1	Agree 2	Neutral 3	Disagree 4	Strongly Disagree 5
36. I would have great difficulty adjusting emotionally if I learned that I was a CF carrier	1	2	3	4	5
37. Learning that I was a CF carrier would make me feel hopeless.	1	2	3	4	5
38. Finding out that I was a CF carrier would hurt my relationship with my (future) partner	1	2	3	4	5
39. It would be very stressful for me if I knew that I was a CF carrier	1	2	3	4	5
40. Testing would make me worry unnecessarily	1	2	3	4	5
41. Testing could lead to a cure for CF	1	2	3	4	5
42. Testing is too much trouble	1	2	3	4	5
43. Testing would give me peace of mind	1	2	3	4	5

VI Imagine that your test results show you are a CF carrier. For EACH of the following statements, please indicate how likely you are to:

	Very Likely 1	Likely 2	Neutral 3	Unlikely 4	Very Unlikely 5
44. Keep the test results to yourself	1	2	3	4	5
45. Try not to think much about the test results	1	2	3	4	5
46. Tell your relatives about your test results	1	2	3	4	5
47. Tell your partner about your test results	1	2	3	4	5
48. Encourage your partner to be tested	1	2	3	4	5
49. Tell your family doctor about your test results	1	2	3	4	5
50. Tell your children about your results	1	2	3	4	5

VII Again imagine that you have just been informed that you are a CF carrier. Here are some feelings people have expressed after they received similar test results. *For EACH of the following feelings, please circle the ONE NUMBER that best indicates how you might feel:*

	Very Likely 1	Likely 2	Neutral 3	Unlikely 4	Very Unlikely 5
51. Anxious	1	2	3	4	5
52. Depressed	1	2	3	4	5
53. Happy	1	2	3	4	5
54. Relieved	1	2	3	4	5
55. Angry	1	2	3	4	5
56. Hopeless	1	2	3	4	5
57. "Labelled" or "Abnormal"	1	2	3	4	5

VIII **Imagine that your test results show you are not a CF carrier. For EACH of the following statements, please indicate how likely you are to:**

	Very Likely 1	2	Likely 3	Neutral 4	Unlikely 5 Very Unlikely
58. Keep the test results to yourself	1	2	3	4	5
59. Try not to think much about the test results	1	2	3	4	5
60. Tell your relatives about your test results	1	2	3	4	5
61. Tell your partner about your test results	1	2	3	4	5
62. Encourage your partner to be tested	1	2	3	4	5
63. Tell your family doctor about your test results	1	2	3	4	5
64. Tell your children about your test results	1	2	3	4	5

IX Again, imagine that you have just been informed that you are not a CF carrier.
For EACH of the following feelings, please circle the ONE NUMBER that indicates how you might feel :

	Very Likely 1	Likely 2	Neutral 3	Unlikely 4	Very Unlikely 5
65. Anxious	1	2	3	4	5
66. Depressed	1	2	3	4	5
67. Happy	1	2	3	4	5
68. Relieved	1	2	3	4	5
69. Angry	1	2	3	4	5
70. Hopeless	1	2	3	4	5
71. "Labelled" or "Abnormal"	1	2	3	4	5

X **Imagine** that your risk (or chance) to be a CF carrier has been lowered from 1/20 to 1/200 (i.e. 5% to 0.5%). Therefore, you have a very low risk, **but not zero**, for being a CF carrier. *For EACH of the following statements, please indicate how likely you are to:*

	Very Likely 1	Likely 2	Neutral 3	Unlikely 4	Very Unlikely 5
72. Keep the test results to yourself	1	2	3	4	5
73. Try not to think much about the test results	1	2	3	4	5
74. Tell your relatives about your test results	1	2	3	4	5
75. Tell your partner about your test results	1	2	3	4	5
76. Encourage your partner to be tested	1	2	3	4	5
77. Tell your family doctor about your test results	1	2	3	4	5
78. Tell your children about your test results	1	2	3	4	5

XI Again, imagine that you have a very low risk, but not zero, for being a CF carrier. For EACH of the following feelings, please indicate the ONE NUMBER that best indicates how you might feel.

	Very Likely 1	Likely 2	Neutral 3	Unlikely 4	Very Unlikely 5
79. Anxious	1	2	3	4	5
80. Depressed	1	2	3	4	5
81. Happy	1	2	3	4	5
82. Relieved	1	2	3	4	5
83. Angry	1	2	3	4	5
84. Hopeless	1	2	3	4	5
85. "Labelled" or "Abnormal"	1	2	3	4	5

XII The following questions deal with how you THINK you would use the test results for planning your own family.

86. (a) Would the test results affect your family planning decisions? (*you may check more than one box*)

YES ☐ NO ☐

(b) If YES, in what way? (*you may check more than one box*)

- ☐ I would get more information about CF
- ☐ I would have my partner tested
- ☐ I would not marry a carrier
- ☐ I would use a more effective birth control method
- ☐ I would consider adoption
- ☐ I would consider artificial insemination or egg donor
- ☐ I (or my partner) would go for prenatal testing
- ☐ I would not have any more children if both myself and my partner are carriers
- ☐ I would consider abortion
- ☐ Other (*please specify*) _____

(c) If NO, why not? (*you may check more than one box*)

- ☐ I am probably not a carrier
- ☐ I would prefer to let nature take its course
- ☐ I have finished my family
- ☐ I do not want any children
- ☐ Religious reasons
- ☐ I (or my partner) am already pregnant
- ☐ Any child is worth the risk

XIII Remember that you do not have to answer any questions that you would rather not answer. All responses will be kept confidential.

For the following questions, mark the appropriate box or fill in the blank.

87. What is your postal code?

88. What is your gender? Male ☐ Female ☐

89. What is the highest grade or level of education that you have completed?

- ☐ Less than high school
- ☐ Completed high school
- ☐ Trade certificate/diploma
- ☐ Some college
- ☐ Community college diploma/degree
- ☐ University Bachelors degree
- ☐ University Masters degree
- ☐ Doctorate

90. What year were you born?

19 (Year)

91. What language do you speak most often in the home?

- ☐ English
 - ☐ French
 - ☐ Other
- Specify _____

92. What is your religion?

- ☐ Roman Catholic
☐ Protestant
☐ Jewish
☐ No religious preference
☐ Other
Specify _____

93. What is your current marital status?

- ☐ Single
☐ Married or Common-law
☐ Separated
☐ Divorced
☐ Widowed

94. (a) Do you have any children?

YES ☐ NO ☐

(b) If YES, please indicate their gender, age, and whether or not they have CF.

	Gender		Year of birth	Affected With CF?	
1	M ☐	F ☐	19 ☐ ☐	Yes ☐	No ☐
2	M ☐	F ☐	19 ☐ ☐	Yes ☐	No ☐
3	M ☐	F ☐	19 ☐ ☐	Yes ☐	No ☐
4	M ☐	F ☐	19 ☐ ☐	Yes ☐	No ☐
5	M ☐	F ☐	19 ☐ ☐	Yes ☐	No ☐
6	M ☐	F ☐	19 ☐ ☐	Yes ☐	No ☐

95. Is it possible that you will have children in the next two years?

YES ☐ NO ☐ UNCERTAIN ☐

96. (a) What is your current occupation?

- ☐ Looking for work
- ☐ Student
- ☐ Retired
- ☐ Homemaker
- ☐ Currently employed as: _____
- ☐ Other:
Specify _____

(b) Number of hours worked per week? ☐☐

97. What is your family income per year? (If married or common-law, state the combined income of you and your partner).

- ☐ Under \$10,000
- ☐ \$10,000 to 19,999
- ☐ \$20,000 to 29,999
- ☐ \$30,000 to 39,999
- ☐ \$40,000 to 49,999
- ☐ \$50,000 to 59,999
- ☐ \$60,000 to 69,999
- ☐ \$70,000 to 79,999
- ☐ \$80,000 to 89,999
- ☐ \$90,000 to 99,999
- ☐ \$100,000 or above

THANK YOU FOR TAKING THE TIME TO ANSWER OUR QUESTIONS.

If you have any questions or comments, please feel free to contact us at
(613) 738 - 3904 or (e-mail) ci110@freenet.carleton.ca

Appendix B

The Miller Behavioural Style Scale

1. Vividly imagine that you are afraid of the dentist and have to get some dental work done. Which of the following would you do? Check all of the statements that might apply to you.

- ☐ I would ask the dentist exactly what he/she was going to do.
- ☐ I would take a tranquilizer or have a drink before going.
- ☐ I would try to think about pleasant memories.
- ☐ I would want the dentist to tell me when I would feel pain.
- ☐ I would try to sleep.
- ☐ I would watch all the dentist's movements and listen for the sound of the drill.
- ☐ I would watch the flow of water from my mouth to see if it contained blood.
- ☐ I would do mental puzzles in my mind.

2. Vividly imagine that you are being held hostage by a group of armed terrorists in a public building. Which of the following would you do? Check all of the statements that might apply to you.

- ☐ I would sit by myself and have as many daydreams and fantasies as I could.
- ☐ I would stay alert and try to keep myself from falling asleep.
- ☐ I would exchange life stories with the other hostages.
- ☐ If there was a radio present, I would stay near it and listen to the bulletins about what the police were doing.
- ☐ I would watch every movement of my captors and keep an eye on their weapons.
- ☐ I would try to sleep as much as possible.
- ☐ I would think about how nice it's going to be when I get home.
- ☐ I would make sure I knew where every possible exit was.

Appendix B, continued

3. Vividly imagine that, due to a large drop in sales, it is rumoured that several people in your department at work will be laid off. Your supervisor has turned in an evaluation of your work for the past year. The decision about lay-offs have been made and will be announced in several days. Check all of the statements that might apply to you.

_____ I would talk to my fellow workers to see if they knew anything about what the supervisor's evaluation of me said.

_____ I would review the list of duties for my present job and try to figure out if I had fulfilled them all.

_____ I would go to the movies to take my mind off things.

_____ I would try to remember any arguments of disagreements I might have had with the supervisor that would have lowered her/his opinion of me.

_____ I would push all thoughts of being laid off out of my mind.

_____ I would tell my spouse that I'd rather not discuss my chances of being laid off.

_____ I would try to think which employees in my department the supervisor might have thought had done the worst job.

_____ I would continue doing my work as if nothing special was happening.

Appendix B, continued

4. Vividly imagine that you are on an airplane, thirty minutes from your destination, when the plane unexpectedly goes into a deep dive and then suddenly levels off. After a short time, the pilot announces that nothing is wrong, although the rest of the ride may be rough. You, however, are not convinced that all is well. Check all of the statements that might apply to you.

_____ I would carefully read the information provided about safety features in the plane and make sure I knew where the emergency exits were.

_____ I would make small talk with the passenger beside me.

_____ I would watch the end of the movie, even if I had seen it before.

_____ I would call for the flight attendant and ask him/her exactly what the problem was.

_____ I would order a drink or tranquilizer from the flight attendant.

_____ I would listen carefully to the engines for unusual noises and would watch the crew to see if their behaviour was out of the ordinary.

_____ I would talk to the passenger beside me about what might be wrong.

_____ I would settle down and read a book or magazine or write a letter.

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APPENDIX C - PAGES 120 & 121

Appendix D

Description of Cystic Fibrosis

In the beginning your baby will seem healthy, however, you will begin to notice certain persistent signs of illness such as less playfulness, some coughing, and foul smelling bowel movements. You may need to give pills to your child many times a day. Your child will occasionally need to stay in the hospital (i.e. once a year). You will, however, be able to enjoy many of the joys of parenthood, with allowances made for the illness.

Imagine that the symptoms steadily progress. As a teenager your child is often tired, has a poor appetite, and has a constant cough. In addition to needing more medication, there is now a need for more frequent hospital stays (i.e. 2-3 times a year) and physical therapy to clear your child's lungs. You find that you are spending an increasing amount of time caring for your child. There is, however, still some time for normal family activities.

In their 20's, your child's activity level becomes severely limited so that he/she cannot work. Hospital stays are more frequent. Your child is taking as many as 40 pills a day and friends notice obvious signs of weight loss and breathing problems. In their 30's your child may die from CF after a prolonged hospital stay.

Throughout your child's lifetime there will be no intellectual impairment but there may be some social and emotional difficulties related to the disease.

At times, many parents may feel sadness and distress dealing with the additional responsibilities of caring for their CF child. In spite of this, some parents find hidden strengths in themselves and appreciate the special joys in caring for such children.

Appendix E

Introductory Letter to Participants

Dear Participant:

My name is Barb Virley O'Connor and together with Drs. Mario Cappelli and Linda Surh at the Children's Hospital of Eastern Ontario, I am conducting a research project concerning the factors which influence individuals' decisions to use genetic testing for CF carrier screening,

As you may know, a genetic test is available which lets people know if they carry a CF disease gene. The purpose of this study is to better understand peoples' opinions concerning genetic testing for CF carrier status, and how useful you might find the test results. The information you provide to us in the questionnaires enclosed will help us to better understand peoples' views toward, and reasons for, seeking genetic testing.

Participating in this study will take about 40 minutes of your time. You will be required to fill out several brief questionnaires. All of your responses will be kept strictly confidential, meaning that no one except the researcher will ever know who gave which response to which question. All data is coded such that your name is not associated with answers you gave. You have the right to withdraw your information from the study at any time.

IF YOU AGREE TO PARTICIPATE, please follow these 7 steps:

- 1) Read and sign the enclosed "Consent-CF Carrier Testing Study" form
- 2) Read the form entitled "Description of Cystic Fibrosis"
- 3) Complete both sides of the survey entitled "Genetic Testing for Cystic Fibrosis"
- 4) Read the instructions and complete both sides of the form entitled "MBSS"
- 5) Read the instructions and complete both sides of the "Self-Evaluation Questionnaire" form
- 6) Read the instructions and complete the form entitled, "Rating Scale Exercise"
- 7) Put all forms from # 1-6 in the enclosed, addressed and stamped envelope and return package as soon as possible to the researcher.

continued...

Appendix E, continued

Your time in answering these questions is greatly appreciated. Your responses are very important in helping us to understand how people feel about genetic testing. If you have any ethical concerns about this study, please contact Dr. L. Paquet (Chair, Department of Psychology Ethics Committee, Carleton University, Ottawa, 613-520-2600), or Dr. A. Hunter (Chair, Research Ethics Committee, CHEO, Ottawa, 613-737-2275).

If you have any questions or comments about this study, please contact us by telephone at **(613) 738-3904**, or contacting Dr. Cappelli directly at **(613) 737-2492**. You may also reach us by e-mail at **ci110@freenet.carleton.ca**. Once again, thank you for your time and consideration.

Sincerely,

Barbara Virley O'Connor
Research Assistant, CHEO
M.A. Candidate, Carleton University

Dr. Mario Cappelli,
Psychologist
CHEO

Dr. Linda Surh
Director, Molecular Diagnostics Laboratory
CHEO

Appendix F

Consent - CF Carrier Testing Study

As you may know, a genetic test is available which lets people know if they carry a CF disease gene. We are interested in your opinions concerning genetic testing for CF carrier status, and how useful you might find the test results. The information you provide us in the questionnaires enclosed will help us to better understand peoples' views toward, and reasons for, seeking genetic testing.

Participating in this study will take about 40 minutes of your time. All of your responses will be kept strictly confidential, meaning that no one except the researcher will ever know who gave which response to which question.

This study is designed to gather information only. If you decide to answer the enclosed questionnaires and return them to the researcher, you have the option to withdraw your answers from the study at any time by contacting Dr. Mario Cappelli, Dr. Linda Surh or Barbara Virley O'Connor at (613) 738-3904, or by e-mail at ci110@freenet.carleton.ca.

♦ I, (*please print name and telephone number*) _____

_____ consent to participating in this study of
people's attitudes toward CF carrier screening.

♦ Signed, _____

♦ This date, _____, 1996.

♦ As witnessed by (*please print name and sign*) _____

♦ This date, _____, 1996.