MATERNAL EFFORTS TO PREVENT TYPE 1 DIABETES IN GENETICALLY SCREENED INFANTS

By

AMY E. BAUGHCUM

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This dissertation is dedicated to my parents and grandparents who instilled in me the value of life-long learning.

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By

Amy E. Baughcum

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Chair: Suzanne Bennett Johnson
Cochair: Alexandra Quittner
Major Department: Clinical and Health Psychology

Currently, research programs exist to screen newborns in the general population for genetic risk of developing Type 1 diabetes, including the Florida Prospective Assessment of Newborn Diabetes Autoimmunity (PANDA) study. These screening programs are part of longitudinal studies addressing the etiology of type 1 diabetes, with the ultimate goal of developing preventative interventions. However, little is currently known about the impact of newborn genetic screening on maternal behaviors of newborns found to be at increased risk for the disease. Additionally, since we do not presently know how to effectively prevent type 1 diabetes, health care professionals are not able to offer definitive recommendations to mothers regarding specific behaviors to prevent diabetes in their at risk children. In the absence of this information, mothers may take their own actions in an effort to prevent the disease in their children. The purpose of this exploratory study was to examine maternal reported behavior changes associated with identifying at risk infants via genetic screening.

Structured telephone interviews were conducted with 192 mothers of children between the ages of 2 and 7 years who were previously identified as at increased risk through genetic screening. Interview questions elicited qualitative and quantitative information regarding maternal behavior changes affecting the child's diet, physical activity, stress level, environment, and health surveillance. Additional questions assessed mothers' anxiety, perceived control, perceived risk, information-seeking behaviors, and sources of information regarding their children's risk for diabetes.

Results indicated that most mothers reported engaging in behavior change (67%) and typically these behaviors involved increased health surveillance and healthy lifestyle changes. Significant predictors of behavior change included family history of diabetes, anxiety, coping, perceived risk, and information seeking. Overall, these findings suggest that genetic screening for type 1 diabetes has minimal negative impact on maternal behavior. Despite the positive nature of subsequent behavior modifications, such behavior changes that may occur in individuals' everyday lives in response to a health risk could threaten the internal validity of natural history studies and prevention trials if not carefully monitored.

CHAPTER 1
INTRODUCTION

Advances in the field of human genetics are rapidly changing the practice of medicine. The Human Genome Project (HGP), completed in 2000, has played a key role in this revolution by identifying and sequencing the genes that constitute the entire human genome. Genetic mutations account for an estimated 5,000 diseases and influence the development of thousands of others. Estimates suggest that 20 diseases account for 80% of the deaths in the Western world; and these diseases are due to the influence of 100 to 200 individual genes which will be identified in the next few years (Roberts, 2000; Patenaude, Guttmacher, & Collins, 2002). Tests are currently available to identify specific genetic markers that may lead to a disease in those who are at risk for developing the disease at some point during their lives. There are two types of tests. One type is known as "genetic testing," which involves using "specific assays to determine the genetic status of an individual already suspected to be at high risk for a particular inherited condition because of family history or clinical symptoms." The other, genetic screening, involves the use of "various genetic tests to evaluate populations or groups of individuals independent of a family history of a disorder" (Committee on Assessing Genetic Risks, Institutes of Medicine, 1994, p. 4). However, the terms "genetic testing" and "genetic screening" are often used interchangeably; and thus, these words are used interchangeably in this paper as well.

As we come to better understand human genetics, we have the opportunity to learn more about the role that specific genes play in the etiology of disease. It is estimated that

each person's genetic make-up contains 5 to 30 alterations in DNA that could predispose the development or transmission of a genetically based disease. It is apparent that advances in genetics will continue to expand, and screening for genetic susceptibility for diseases will become more commonplace in the coming decades (Juengst, 1995). Longitudinal research studies of genetically at risk individuals are necessary to learn about the natural progression of disease in order to develop effective prevention strategies. These studies will provide a better understanding of the interactions between multiple genes in disease development as well as interactions between genes and the environment. While there are diseases that are determined by a single gene mutation, such as Huntington's disease, many conditions are genetically more complex, involving multiple genes and environmental factors. Thus, as we learn more about genetic predispositions, it will be increasingly important to examine environmental factors (including individuals' health behaviors) to complete our scientific understanding of disease etiology. The exciting new opportunities in genetics are accompanied by many unanswered questions about how the public at large will accept and understand these new techniques and the risk information they provide.

Unfortunately, new advances in medical technology have outpaced the rate at which psychological research has proceeded. Genetic medicine can have a huge influence in life and death issues, raising ethical, social and legal concerns. It is ethically imperative to consider how the new genetic revolution impacts, both positively and negatively, the quality of life for individuals and their families. By identifying an individual's genetic risk for disease, there is potential for early treatment or disease prevention, or in the case of an incurable disease, the ability to initiate health surveillance

and/or plan for the future. Current literature suggests that certain health behaviors (i.e., diet and exercise) can moderate risk for several diseases, such as heart disease, cancer or type 2 diabetes. Therefore, awareness of one's genetic risk may directly affect behavior change, and consequently disease progression. Genetic counseling is now shifting to providing information regarding personal risk reduction; and allowing individuals to make better-informed medical decisions (Lerman et al., 2002). However, there are also many diseases for which there is no known method of prevention or cure. In these instances, it may not be possible to make behavioral recommendations regarding health behavior change, other than increased medical surveillance. Despite this, individuals on their own may engage in behaviors they perceive to be beneficial or preventative.

Recently, there has been a large push for clinical psychologists to become more involved in genetics; and to lend their expertise as clinicians, researchers, and educators to advance our understanding of the psychosocial costs and benefits of genetic screening (Fisher et al., 2002, Gallier, 2002; Lerman et al., 2002; Patenaude et al., 2002; Patenaude et al., 2003). A number of agencies have made the psychosocial implications of genetic advances a funding priority, including the Human Genome Project which designated 5% of the total budget to ethical, legal, and social issues (Jeffords & Daschle, 2001; cited in Patenaude et al., 2002).

Clinical psychologists can play an important role in answering how genetic risk information impacts individuals in cognitive, affective, and behavioral realms. Psychologists can assess the role that different personal, social, and cultural factors contribute to the development or prevention of disease. As clinicians, psychologists can help individuals and families understand risks, make informed behavioral and

reproductive choices, provide psychosocial support, and evaluate outcomes. A recent article by Patenaude et al. (2003) highlights the many important roles pediatric psychologists can play in the research and public policy arenas to inform ethical debates on the merits of genetic testing; and to provide competent clinical care to affected families (Patenaude et al., 2003).

While there has been some research addressing attitudes toward genetic testing, comprehension of genetic information, and the psychological impact of genetic testing, there is still much to be learned about the impact of genetic screening on individuals' behavior. This exploratory study examined the behavioral impact of newborn genetic screening for mothers whose children were found to be at risk for type 1 diabetes. Currently, little is understood about the specific behavior changes that may result from knowing one's child is genetically predisposed to a condition for which there is currently no known prevention method or cure. Additionally, our present understanding of the etiology of type 1 diabetes suggests that it develops from a combination of both genetic and environmental influences, which are not well-defined. In the absence of definitive recommendations from the health care community, mothers of newborns identified as "at risk" may take actions they believe are effective in preventing type 1 diabetes in their children. This study assessed the extent of mothers' self-reported behavior changes; and assessed associations between reported behavior change and maternal psychological (i.e., anxiety, perceived control, coping), and sociodemographic variables. The upcoming sections discuss existing literature on the psychological and behavioral impact of genetic testing, including newborn genetic screening; and our current knowledge of the etiology and prevention of type 1 diabetes.

CHAPTER 2
REVIEW OF THE LITERATURE

Psychological Impact of Genetic Testing

Literature assessing the psychological impact of genetic testing has largely focused on predictive testing for Huntington's disease and breast and ovarian cancer susceptibility (BRCA1/BRCA2 genes). Many studies have examined other uses of risk screening, including prenatal screening and carrier screening. This review focuses on predictive genetic testing. Generally, studies of predictive testing have focused on both the short- and longer-term effects of genetic testing on affective outcomes for screening participants and their family members. Overall, voiced criticisms that genetic testing leads to poor psychological adjustment appear unfounded based on published literature; and such claims may create unnecessary panic (Horowitz et al., 2001; Palmer et al., 2002).

Longitudinal prospective studies examined levels of anxiety and depression before genetic testing; and provided new information regarding the potential for poor psychological adjustment. Evidence suggests that contrary to earlier concerns, making the choice to be tested signals psychological preparedness for the outcome and ability to handle the news well. Most people who choose to participate in population-based screening programs do not have a family history and therefore, will most likely expect and receive a negative result. In screening members from high-risk families (those with a family history of the disease), patients tend to overestimate (not underestimate) their risk; and expect to receive positive results (Croyle & Lerman, 1999; Lynch et al., 1999; Lynch et al., 1993). While one might expect that receiving positive results would result in

clinically significant distress and increased mood symptomatology, studies have found that a positive test result is usually not associated with clinical levels of anxiety or depression (Broadstock et al., 2000; Lerman et al., 2002; Schwartz et al., 2002). This may be partly because reducing one's uncertainty regarding risk may actually decrease stress by providing relief from what was previously unknown (Marteau & Michie, 1995; Baum et al., 1997). While some studies suggest elevated scores on measures of distress, such as depression or anxiety (Shaw, Abrams & Marteau, 1999), scores generally return to baseline levels after 3 to 12 months (e.g., Croyle et al., 1997; Lerman et al., 1996; Wiggins et al., 1992). While there may be some immediate distress upon risk notification, it appears to be neither inevitable nor long-lasting.

A literature review conducted by Broadstock et al. (2000) examined existing prospective studies of the impact of genetic testing for Huntington's disease, ovarian and breast cancer, and familial adenomatous polypsosis (FAP). To be included in the review, studies had to contain both pre and post measures of psychological distress. Examples of measures of distress used in these types of studies include the Impact of Events Scale (IES; Horowitz, 1979), State-Trait Anxiety Inventory (STAI; Speilberger et al., 1970), and the Beck Depression Inventory (BDI; Beck, 1961). The authors' extensive search uncovered 11 studies, none of which found an increase in distress (defined as general or test-specific anxiety or depression) at any point in the 12 months after testing. After notification, distress decreased in individuals who received either a positive or negative test result. However, this decline was greater and more rapid in those who received negative test results. Furthermore, in regression models, the actual test result rarely predicted psychological outcomes beyond the first month post-risk notification.

Congruent with the Stress Disease Coping Model proposed by Baum et al. (1997), the individual's pretest emotional state, social support, and expectations were the most predictive of subsequent distress (Marteau & Croyle, 1998), suggesting that personal variables may play a role in how one handles genetic risk information.

Taken together, these studies generate little empirical support for the notion that genetic testing is associated with adverse psychological outcomes (Lerman et al., 2002). However, most of these studies involved participants in research registries and these results may not generalize to the broader population. It should be noted that those in clinical settings may be self-referred and more naïve, and thus, less equipped to cope with knowledge of their risk status (Broadstock et al., 2000).

Previous research suggests that psychological distress may be associated with specific personal (e.g., optimism) or demographic characteristics (e.g., race, education). Audrain et al. (1998) studied women with a first degree relative with breast or ovarian cancer before testing and found that pre-test distress was predicted by age, ethnicity, marital status, optimism, perceived control, and overestimated risk perception. Those who perceived less control, were younger, not Caucasian, married, and less optimistic were more likely to experience greater distress before risk screening (Audrain et al., 1998). Hughes et al. (1997) studied ethnic differences in knowledge and attitudes regarding testing for BRCA1 gene in at risk women; and found that African American women had lower levels of knowledge, but more positive attitudes toward genetic testing than Caucasian women. Risk perception appears to vary by ethnic status, with African American women who have a family history of breast cancer having greater concerns

about their own personal risk of breast cancer and appearing more likely to avoid breast cancer-related thoughts and feelings (Hughes et al., 1996).

Studies have examined coping strategies associated with receiving genetic risk information to determine if coping mediates distress in genetically at risk individuals. For these types of studies, coping has been conceptualized as the degree to which one either seeks more information (monitoring) or avoids or distracts oneself from the situation (blunting/avoidance) (Miller, 1987). Studies have used the Miller Behavioral Style Scale (MBSS; Miller, 1987), to determine the style in which individuals deal with risk information given. It has been hypothesized that individuals cope with health threats in one of two ways. In general, an interaction has been reported between the amount of information provided and whether monitoring or blunting characterizes the individual's coping style.

Studies have found that matching the amount of information received to the amount the individual desires, lowers distress (Ludwick-Rosenthal & Neufeld, 1993; Miller, 1980; Miller & Managan, 1983). There does not appear to be consensus regarding whether coping mediates distress in those notified of increased genetic risk status. For example, in a study of patients from high-risk families screened for BRCA1/BRCA2 genes, coping efforts (both active and avoidant) were associated with higher levels of distress prior to notification; whereas post-notification distress was associated with the test result, not coping (Tercyak et al., 2001a). Lerman et al. (1993) found that a high level of monitoring in women at risk for breast cancer predicted an increase in distress over a 3-month follow-up period; whereas Vernon and colleagues' (1997) study of FAP screening found the opposite to be true. Anxiety appears to be influenced by whether or

not the event is controllable and by the amount of information given to an individual (Miller et al., 1989). Sex differences may also play a role in risk appraisal and coping. Marteau et al. (1997) found that women have a greater fear of threat, worry more about negative outcomes, and perceive greater risks from technology than men; whereas men show higher threat minimization after positive carrier testing for cystic fibrosis (CF).

Behavioral Impact of Genetic Testing

A major question of interest to researchers is whether results of predictive genetic testing lead to increases or decreases in health behaviors and medical surveillance. Does informing people of genetic susceptibility to disease motivate them to take action to reduce their risk? Or does knowing that one is genetically predisposed suggest a sense of pre-determined destiny and perceived immutability (Senior, Marteau, & Peters, 1999; Senior, Marteau, & Weinman, 1999)? Marteau & Lerman (2001) reviewed literature related to cancer, smoking, and heart disease and espoused that providing genetic information may not increase motivation to change behaviors and may even result in reducing motivation. However, these authors also suggested that genetic information might better facilitate change if individuals are offered effective risk-reducing interventions tailored to their genetic risk. Most existing research studies in this area focused on cancer, particularly, breast cancer screening; and physician recommended behaviors, such as mammography and breast self-examinations.

Studies have examined the impact of distress caused by risk notification to determine if distress predicts health-protective or preventative behaviors. Croyle & Lerman (1999) reviewed studies on how coping and distress influenced the processing of genetic risk information and subsequent decision-making. Studies have found that risk information can be too anxiety provoking for some; and therefore, anxiety acts as a

barrier to following through with screening recommendations (Kash et al., 1992; Lerman et al., 1994; Lerman et al., 1993). Other studies have suggested that increased distress or worry actually increases health behaviors (Burnett et al., 1999; Diefenbach et al., 1999) and even leads to excessive health practices (i.e., breast self-examinations) (Epstein et al., 1997; Epstein & Lerman, 1997; Lerman et al., 1994; Lerman & Schwartz, 1993). Epstein et al. (1997) found that those who were excessive in their protective behaviors were more likely to be African American, older, and less educated. These findings may be explained by results from Audrain et al. (1998) suggesting that African American women undergoing genetic screening experience greater distress and have lowers levels of cancer-related knowledge. Others have found no significant relationship between distress and adherence to recommended medical surveillance (Lerman et al., 2000). Taken together, these studies suggest that an inverted U-shape curve may explain the relationship between distress and screening behavior, with highest rates of adherence predicted by a moderate level of anxiety (Hailey, 1991; Lerman et al., 1991; Lerman & Rimer, 1993).

There have been conflicting reports of how perceived versus actual risk impacts screening behaviors (Hailey, 1991). Overall, some cancer studies found an increase in screening rates in those informed they are genetically at higher risk (Meiser et al., 2000; Ritvo et al., 2002; Schwartz, Taylor, et al., 1999), while other studies have found adherence rates similar to those of the general population (Bratt et al., 2000; cited in Marteau & Lerman, 2001; Clavel-Chapelon et al., 1999). In a study of colon cancer screening and behavior intentions, half of respondents indicated that they would decrease their use of screening tests and make fewer attempts to reduce their dietary fat intake if

their test results indicated that they were at low risk (Lerman et al., 1996). Women with a family history of breast cancer were more likely to perceive higher genetic risk and engage in appropriate screening behaviors (Hailey et al., 2000; McCaul et al., 1996). Perceived risk has been found to predict screening compliance above and beyond the actual risk associated with family history of a disease (Aiken et al., 1994). Women who perceived themselves to be at greater risk were more likely to engage in initial as well as repeated screenings (Lerman et al., 1990; McCaul et al., 1996). However, the perception of risk does not appear to be necessarily related to the accuracy of risk. It has been suggested that accurate recall of risk information does not necessarily lead to risk-reducing behavior. Therefore, many have begun to examine the links between risk perception and risk-reducing behavior, particularly the potentially mediating variable of disease-specific worry or anxiety.

Many studies have been conducted examining the role of information-seeking and health behavior change, particularly as it relates to public health issues (i.e,. HIV/AIDS prevention). Rakowski (1990) conducted a randomized survey among adults in the general population and found a positive association between more frequent information seeking and personal health-related practices. However, hardly any studies have examined this issue in the context of genetic screening. One such study examined women at genetic risk of ovarian cancer; and found that monitors (information-seekers) demonstrated greater adherence to behavioral recommendations, such as attending cancer screenings (Wardle, 1995).

Demographics factors are also important in predicting health behaviors. Schwartz et al. (1999b) found that women with less education who were at risk for breast cancer

and screened, reduced their use of mammography after breast cancer risk counseling. Additionally, studies have found that women from ethnic minority groups and women with lower levels of education reported greater disease-specific worry (Aiken et al., 1994; Audrian et al., 1998) and retained less information about screening programs in general (Browner et al., 1996, Donovan & Tucker, 2000; Hughes et al., 1997)

Lerman et al. (1993) found that reproductive behaviors were also impacted by cancer screening. In a study of women under age 49, 22% reported that they would be less likely to have children if they tested positive; and 17% reported being uncertain whether they would continue a pregnancy. Other studies that assessed the reproductive impact of genetic testing, found that 46-83% of subjects within reproductive age in the general population would not have children or would limit further reproduction if they tested positive for a disease gene (Kessler et al., 1989; Schoenfeld et al., 1984; Zerres et al., 1986). Unfortunately, little is known about cancer screening and other lifestyle changes involving smoking, physical activity, or diet (Marteau & Lerman, 2001).

In the current study, both lifestyle and health surveillance behaviors were included as outcomes. Similar to other studies, relationships between reported behavioral outcomes and psychological variables (such as anxiety, perceived control, coping, and risk perception) were assessed.

Theoretical Models of Genetic Screening and Behavioral Change

Most studies in the area of genetic testing fail to use a theoretical model to conduct or interpret the findings. However, theoretical models are important as they can serve as a contextual framework for interpreting complex results. Generally, most models of health behaviors assume that the motivation for health-protective behavior comes from

both the anticipation of a negative health outcome and the hope of avoiding it (Weinstein, 1993).

Tercyak (2000) advocated for conceptualizing the impact of genetic testing as a family systems issue (Tercyak, 2000). The rationale for this was that genetic risk information impacts multiple family members and the family's dynamics as a whole. Additionally, Tercyak (2000) reasoned that families with no history or experience with a particular illness would fare differently than those with a family member who is already ill. Pre-existing illness in one member of the family provides personal experience and an increased knowledge base about that condition for other family members that families without a history of that particular disease would not have. Therefore, the meaning and implications of genetic test results would be different depending on family history. By translation, subsequent behavior change may also differ.

Rolland (1999) advocated the use of a specific model, the Family Systems-Illness Model, when examining the psychological impact of predictive testing. Rolland (1999) stated it to be a "useful guide," as it emphasizes the psychosocial demands of different disorders over time and emphasizes the key components of family functioning (i.e., multigenerational patterns), illness life cycles, and belief systems. Rolland recognized that psychosocial challenges varied according to biological variables, including the degree to which a disease is influenced by both the environmental and genetic factors and the degree to which prevention is possible (Rolland, 1999). Rolland advocated for longitudinal follow-up of families after genetic testing, as psychosocial strains related to knowledge of future risk do not just present themselves upon result notification, but will tend to surface at major life-cycle transitions (Rolland, 1999). These challenges

influence family decision making and health behaviors. In his descriptions, Rolland did not address specific mechanisms of his model or how they apply to screening results (Rolland, 1999). It is apparent that Rolland was using his model as a conceptual guide rather than as a testable model. To date, although genetic testing is recognized as impacting the daily lives of entire family units, there has been no formal testing of a family systems model pertaining to the psychosocial impact of genetic testing.

The Health Belief Model (HBM) (Rosenstock, 1974) has been used most frequently in previous genetic testing research. Investigators have used this model to explain preventative health care behavior in the context of one's perceived susceptibility to an illness, the perceived severity of that illness, and the potential benefits and costs of performing a specific behavior to reduce the risk (i.e., Aiken et al., 1994; Becker & Maiman, 1975; Frets et al., 1990; Rowley et al., 1991; Sagi et al., 1992; Shiloh & Saxe, 1989; Sorenson et al., 1987). These factors are hypothesized to be predictive of the decision to engage in health behavior change or to increase surveillance. Preventative action is most likely taken when individuals perceive themselves to be at risk for a serious disease and when the benefits to action outweigh the costs of not engaging in the specific health behavior. For the purposes of genetic screening studies, most have focused more on the perceived susceptibility component rather than the perceived severity.

The HBM has several significant weaknesses. The perceived severity component has not gained strong empirical support as a major predictor for preventative behavior (Leventhal et al., 1983). Additionally, the HBM has proven influential for health attitudes, but not consistently for health behaviors. HBM also assumes that health

behaviors arise from a single rational decision based on cost-benefit analysis, which may be an oversimplification (Horne & Weinman, 1998). Finally, this model does not specify underlying beliefs, how to change beliefs (Horne & Weinman, 1998) or what beliefs need to be changed in order to change behavior. Other social variables and personal factors, as reviewed in later sections, may have more importance in influencing health behavior than this model would suggest.

While many studies have used the HBM, Lerman et al. (1997) endorsed a different model, the Self-Regulation Model of Health Behavior (SRM; Leventhal, 1965), to more specifically address why women at risk for breast cancer experiencing too great or too little worry were less likely to practice risk-reduction behavior. According to Leventhal (1970), a health threat results in both cognitive and affective responses, which occur in parallel. The SRM suggests that moderate levels of perceived health threat (e.g., diagnosis of cancer in a relative) engender a moderate level of concern/worry, which in turn, leads individuals to take actions that will reduce the anxiety caused by a health threat. Fear arousal coupled with an action plan leads to a "cognitive representation" of the threat (Horne & Weinman, 1998). Excessive cancer-related anxiety might produce avoidance of screening, and at least a minimal level of anxiety is necessary to motivate these behaviors. Similar to this model is the Fear Arousing Communications Theory (Janis and Feshback, 1953), which states some degree of fear arousal is needed to predict adoption of health care behaviors. If individuals are not concerned, they may deny the threat; and if they are overconcerned, they may come to avoid preventative health practices (Kash et al., 1992).

However, contrary to the aforementioned models, communication of a threat alone may be insufficient to change one's behavior (Horne & Weinman, 1998). Relatively few studies have used the SRM, perhaps because of its complexity, which makes it difficult to operationalize. However, there does appear to be some empirical support for this model in studies of medication adherence in hypertension (Meyer et al., 1985) and regimen adherence in diabetes (Gonder-Frederick & Cox, 1991; cited in Horne & Weinman, 1998).

The model that best informs the current study is a transactional model of stress and coping, known as the Stress-Disease Risk-Coping Model, which is a comprehensive model specifically designed for studies of genetic testing (Baum et al., 1997). Baum and colleagues' (1997) model is based on the concept of risk appraisal espoused by Lazarus & Folkman (1984), in which primary appraisal involves the judgment of the threat of a stressor; and secondary appraisal consists of a judgment regarding available resources to deal with the threat. This model is particularly concerned with the relationship between uncertainty and risk perception, which influence one's stress response, and consequently affect one's behavior. Proponents believe this model is useful in predicting psychological and behavioral responses to genetic testing results (Lerman, 1997). This model hypothesizes that distress and behavior changes will be affected by the interaction between personal factors (perceived risk influenced by family history, optimism), actual test results, characteristics of disease, and degree of uncertainty remaining after testing (Figure 1). The central component of the model involves the appraisal process regarding the test results. In this step, appraisal of increased certainty regarding future outcomes is coupled with perceived available options for action. This appraisal process is influenced

by the degree to which one perceives him/herself to be at risk; and is influenced by surveillance and prevention options, along with other variables (such as social support, optimism, perceived control, etc.). This appraisal process is associated with the stress response to the information. The more resources available, the better one may be able to cope with the stressor (Wallston, 2000). This stress response and these coping mechanisms in turn, relate to behavioral consequences. This model suggests that the adoption of health behaviors is influenced by personal factors, perceived risk, perceived control, distress, and coping resources. In describing the model, Baum et al. (1997) review the studies that influenced the design of the model, indicating that this model was originally informed by both theoretical and empirical evidence.

Baum and colleagues' (1997) model is fairly new; and presently, no known published studies have tested this model. Despite a lack of available empirical support, this appears to be the most comprehensive and relevant model to use when examining the behavioral impact of genetic screening. The model incorporates many variables that have been examined in the context of genetic screening studies (i.e., perceived risk distress). It should be noted that this model applies to individuals and unfortunately, does not directly incorporate the family unit, which is undoubtedly affected by results of genetic testing. Despite this limitation, for the purposes of the current study, this model was applied to maternal behavior change in response to risk identification in their children.

Newborn Genetic Screening

The current study examined the impact of genetic screening of infants, which is ethically more complicated than testing within adult populations. The Institutes of Medicine (IOM) reports that 3% of children have an illness or disorder of probable genetic origin (IOM, 1994). Understandably, while one would want to extend the

benefits of biomedical advances to children, additional considerations are involved. Consequently, organizations have generated ethical guidelines for performing genetic tests in pediatric populations, including the American Academy of Pediatrics (AAP), American Society of Human Genetics and American College of Medical Genetics, Clinical Genetics Society, and Institutes of Medicine (American Society of Human Genetics and American College of Medical Genetics, Boards of Directors, 1995; Clarke et al., 1994; Wertz et al., 1994). These guidelines are especially important for testing for diseases for which there are no known cures or modes of prevention. In the absence of clearly beneficial treatments or effective methods of prevention, it is difficult to justify the genetic testing of children and adolescents, including newborn screening. Because young children are unable to understand the value of genetic information for their own lives, particular care must be exercised by parents and pediatricians when making decisions about genetic testing for children (AAP Committee on Bioethics, 2001). Other important factors to consider include the psychological and economic impact on the family, time of disease onset, degree of risk, and possible medical benefits.

For these reasons, newborn genetic screening is controversial, especially for those diseases with no known cure. The Institute of Medicine (IOM) report recommended three principles (IOM 1994) to govern the maintenance of existing screening tests and the introduction of new newborn tests:

- identification of the genetic condition must provide a clear benefit to the child
- a system must be in place to confirm the diagnosis
- treatment and follow-up must be available for affected newborns

In other words, newborn genetic screening is supported only if the infants would benefit from early identification and prevention/treatment. Other guidelines exist that allow

regulated research protocols to test children when no immediate medical benefit exists but the contribution to scientific knowledge is great (American Society of Human Genetics/American College of Medical Genetics Board of Directors, 1995; Clark, 1994).

Newborn screening is the most widely used type of genetic screening, with nearly all states in the U.S. mandating newborn screening for phenylketonuria (PKU) and congenital hypothyroidism, in which early diagnosis leads to treatment and better medical outcomes (IOM, 1994). Recently, in some states, newborn screening has expanded to include testing for congenital adrenal hyperplasia and cystic fibrosis (CF) (in WI, CO, and WY). As a point of comparison for CF screening, only 6% of newborn U.S. children are screened versus 92% of newborns in Australia (Wilcken & Travert, 1999). In the past decade, newborn screening has been implemented in research settings to test for risk of type 1 diabetes (discussed in a later section).

Currently, there has been relatively little research on the psychological implications of screening newborns. Much of the research has occurred in other countries. Studies from Wales on newborn screening for Duchenne muscular-dystrophy, an incurable X-linked condition eventually leading to death during early adulthood (Fenton-May et al., 1994), suggest that the screening has been well-received, with few adverse psychological outcomes reported and a participation rate of 90% for eligible families (Bradley et al., 1993; Parsons et al., 2002). Such a favorable outcome is not always the case. A screening program for alpha-1-antitrypsin (lung disease) in newborns in Sweden had to be terminated prematurely because of adverse effects. These included negative changes in family dynamics and parental nonadherence to medical recommendations, including increased smoking behavior (McNeil et al., 1989).

Most of the newborn screening literature has been dedicated to screening for cystic fibrosis (Kerem et al., 1989). CF screening remains controversial (especially for those without a family history) as some view the psychological costs as outweighing the medical benefits of early diagnosis (Wald & Morris, 1998). Adverse psychological outcomes have included greater parenting stress (Baroni et al., 1997) and a small percentage of mothers experiencing short-lived feelings of rejection toward their child (Al-Jader et al., 1990). It should be noted that these effects might also be present when diagnosis is made through conventional means when children are a little older (Al-Jader et al., 1990; Boland & Thompson, 1990; Wilcken et al., 1983). Boland & Thompson (1990) found newborn screening versus traditional screening did not produce greater overprotectiveness in mothers. The delay in diagnosis that occurred when screening was not conducted resulted in greater maternal distress and anger. Therefore, these psychological risks do not appear significant when the potential benefits of newborn screening include better health outcomes due to earlier initiation of treatment (Waters et al., 1999).

Further exploration of the psychological effects of newborn screening is an important area of research as new genetic tests become available; and decisions will need to be made regarding the appropriateness of their use. Whether testing is conducted in the general population or in research settings only; and whether it is conducted with all families or just those with a family history of the disease, are important questions to be answered. How risk information is understood and used by families; and whether it then translates into emotional and/or behavioral changes are key areas for future research.

Type 1 Diabetes: Etiology and Prevention

In the U.S., the prevalence of insulin dependent diabetes mellitus (IDDM; type 1 diabetes) is approximately 2-3/1,000 children, which makes it one of the most prevalent childhood chronic illnesses (Arslanian et al., 1997; LaPorte et al., 1995). Annual incidence is estimated to be over 12,000 children each year, with peak incidence of diagnosis occurring between five and six years of age and again between the ages of eleven and thirteen. The prevalence of type 1 diabetes is higher among Caucasians (National Diabetes Data Group, 1995). In type 1 diabetes, the body produces little or no insulin due to the autoimmune destruction of islet cells in the pancreas. This leads to high blood glucose levels. Type 1 diabetes is thought to be the endpoint of an immunologically mediated attack on pancreatic beta cells. It is an autoimmune disorder where islet cells are destroyed by an immune response, or more simply, destroyed by cells within one's own body that normally protect a person from germs. Complications of type 1 diabetes can include retinopathy, blindness, renal disease, neuropathy, lower extremity ulcers, digestive disorders, heart disease, and vascular disease (National Diabetes Data Group, 1995). The average life span for those with diabetes is generally shortened due to vascular complications. With no cure available, type 1 diabetes is currently medically managed by administering insulin on a daily basis and adhering to a specialized diet and exercise program. These daily treatment demands can greatly affect an individual and their family's lifestyle.

In addition to the impact on the family, type 1 diabetes is a substantial societal and economic burden. Therefore, an obvious need exists for diabetes prevention. Currently, diabetes (including treatment, prevention, and research) consumes one in every seven dollars spent on health care in the U.S. (Schatz et al., 2002). Often diabetes is not

diagnosed until a patient is having a crisis episode (ketoacidosis), which can lead to increased medical complications and longer hospitalizations (Beisswinger, 2000; cited in Schatz, 2002).

Unfortunately, we do not fully understand the etiology of type 1 diabetes. Nearly 90% of type 1 diabetes occurs in families with no history of the disease (Dalquist et al., 1985) and there is only a 30-50% concordance rate among monozygotic twins (National Diabetes Data Group, 1995; Kyvik et al., 1995; LaPorte et al., 1995). However, approximately 3-6% of first-degree relatives with type 1 will develop the disease as well (Tillil & Kobberling, 1987). The chance of developing diabetes for the general population is about 1 in 300 while, for those with first-degree relatives with diabetes, the chances increase to 1 in 20 (National Diabetes Data Group, 1995). These data suggest IDDM is caused by a combination of genetic and environmental factors.

It is generally thought that environmental triggers initiate an autoimmune process that leads to the destruction of pancreatic beta-cells and consequently, type 1 diabetes. It is still unclear the degree to which these environmental factors play a role. In order to determine the interactions between genetics and the environment, longitudinal studies are needed to follow at risk individuals over time. To date, research studies have suggested viral illness (enterovirus and rotovirus) may be one class of environmental triggers (Akerblom & Knip, 1998, Couper, 2001, Dorman et al., 1995). Additionally, Classen & Classen (2001) argue that timing of vaccines increases the risk of type 1 diabetes. The risk of type 1 diabetes decreases when children receive vaccinations after at least two months of age, arguing for the benefits of delayed immunization schedules. A recent study found increased social mixing in young children (i.e., attendance of daycare) in

early infancy was protective against the development of type 1 diabetes because it increased exposure to infections and strengthened immunity (McKinney et al., 2000). However, there has been no other direct evidence in favor of such an association (Akerblom & Knip, 1998). Finally, early emotional stress may also be a contributing factor (Thernlund et al., 1995).

Dietary factors have been implicated as important environmental contributors to the development of type 1 diabetes. Such dietary factors included not breastfeeding (Akerblom & Knip, 1998), early introduction of cow's milk (Akerblom, et al, 1993; Gerstein, 1994; Virtanen et al., 2000), high intake of nitrites/nitrates (Virtanen & Aro, 1994), accelerated prenatal growth (Dahlquist et al., 1996), high intake of proteins (Akerblom & Knip, 1998), high intake of carbohydrates (Akerblom & Knip, 1998) and increased weight gain in infancy (Hypponen et al., 1999). Although, based on both animal and human studies, the most likely putative dietary factors are hypothesized to be cow's milk, proteins, and nitrates/nitrites (Akerblom & Knip, 1998

The greatest amount of research regarding environmental factors related to type 1 diabetes has examined whether breastfeeding is protective and how this interacts with exposure to cow's milk in infancy. Cow's milk is implicated because it has a higher protein content, specifically the protein casein, than that found in human breastmilk. Many studies have been conducted to address this issue with no firm consensus reached (Akerblom & Knip, 1998; Couper, 2001). To examine the role of cow's milk, the multi-national Trial to Reduce IDDM in the Genetically at Risk (TRIGR) is ongoing to determine if delayed exposure to cow's milk until after 6 months of age will have an effect on the subsequent development of diabetes (Karges et al., 1997; Schatz, 2002;

Virtanen et al., 1997). Schatz & Maclaren (1996) warn it is premature to recommend eliminating cow's milk from an at risk child's diet as there is no convincing evidence to suggest the nutritional benefits of milk for young children outweigh the potential dangers.

To answer questions regarding the prevention of type 1 diabetes, The Diabetes Prevention Trial (DPT-1) was initiated in 1994 to determine whether subcutaneous or oral insulin could prevent or delay the onset of diabetes in at risk relatives (DPT-1 Study Group, 1995, 2002). Within this large-scale randomized, nonblind study, there were two separate trials for the two types of insulin administration. Three hundred and thirty nine participants, who were between 3 and 45 years of age and had a first degree relative with type 1 diabetes were randomized in the subcutaneous insulin trial (out of 84,228 screened first degree relatives). To be eligible, participants had to be determined as "high risk," defined as a 50% chance of developing type 1 diabetes over the next five years. This was determined by the absence of protective genetic markers, positive antibody testing, and a low first-phase insulin response in glucose tolerance testing. Participants were randomized to either the intervention group, which received low dose subcutaneous insulin, or the close observation group, and all of whom were followed for an average of 3.7 years. Results from the subcutaneous insulin trial were recently published. Results suggested that injected insulin does not delay or prevent type 1 diabetes (DPT-1 Study Group, 2002). The oral insulin trial is ongoing and results are not currently available.

In contrast to type 1 diabetes, type 2 diabetes (non-insulin dependent diabetes) is a different form of diabetes that is considered a metabolic disorder, rather than an autoimmune disease. It is usually diagnosed in adulthood, although it can develop in childhood. In type 2 diabetes, the body is unable to make enough or properly use insulin;

however, beta cells are preserved. This is in contrast to type 1 diabetes, in which beta cells are destroyed, leading to insulin deficiency. Type 2 diabetes accounts for 90-95% of diabetes and researchers have found obesity and a sedentary lifestyle to be contributing factors, as well as genetic predispotion (Fletcher et al., 2002). Because the prevalence of type 2 diabetes is rapidly increasing to epidemic proportions, the health care community and the media have recently focused significant attention on type 2 diabetes, advocating for healthy lifestyle changes. Recent research has indicated moderate diet and exercise reduces risk for type 2 diabetes more effectively than even oral insulin (Tuomilehto, et al., 2001). A healthy diet is effective because it reduces the insulin load and exercise is effective because physical inactivity reduces tissue glucose tolerance and is associated with insulin resistance. Scientific evidence is not clear as to whether these same behaviors have an impact on the development of type 1 diabetes; however, at the present time, it seems unlikely (Schatz, personal communication). People who do not understand the distinction between type 1 and 2 diabetes may apply recommendations for type 2 to their children at risk for type 1. The current study explored whether this hypothesis was true for our sample population of mothers of at risk young children.

Prediction and Pre-Symptomatic Screening for Type 1 Diabetes

While we do not fully comprehend the natural history of the development of diabetes, we do know that the destruction of pancreatic cells is a precursor to type 1 diabetes and begins long before overt symptoms. It is currently possible to detect pancreatic cell destruction and identify those at risk for developing Type 1 diabetes. Riley et al. (1990) found the determination of islet-cell antibodies in relatives of probands with Type 1 diabetes increased an individuals' risk for developing the disease in the future.

Currently there are two types of screening for diabetes, autoantibody screening and genetic screening. The most recently developed test, genetic screening, is typically done in newborns to determine the present of high-risk genetic markers (DR 3/4, DR 4/4, DR 3/3) in the Human Leukocyte Antigen (HLA) region (the Major Histocompatability Complex (MHC)) on chromosome 6. This is an area that helps control immune response, and such markers are known to confer 50% of the genetic risk for Type 1 diabetes (Yu et al., 1999) (Table 1). The second type of testing, antibody screening, is a process that has been in existence for longer and detects islet-related autoantibodies, including autoantibodies to insulin (Christie et al., 1994; Landin-Olsson et al., 1992), GAD or islet antigen-2 (IA-2), as well as islet cell antibodies (ICA) (Riley et al., 1990; Schatz et al., 1994) present in blood serum. It has been shown the presence and number of these antibodies is directly related to risk for type 1 diabetes (Knip, 1998). An ICA positive result signifies that the process of beta cell destruction has begun and therefore, those who are ICA positive are farther along in the process of developing type 1 diabetes. For example, individuals who test positive for ICA have approximately a 45% chance of developing diabetes in the next ten years. Antibody screening has been conducted with children and adults and used as a primary screening method and as follow-up to newborn genetic screening.

While critics oppose screening for risk of developing type 1 diabetes before symptoms appear, Schatz, et al. (2002) argue it is very important to the future of diabetes prevention research. The authors assert screening helps us in a number of ways: it allows us to better understand the prediabetic period and diabetes pathogenesis, assists in identification of individuals for prevention trials, facilitates earlier diagnoses which

reduces the mortality and morbidity associated with type 1 diabetes (Schatz et al., 2002). Currently, genetic screening is only conducted within research settings since widespread screening of the general population, when there is no available effective intervention, is considered unethical. Many longitudinal studies are now ongoing to follow newborns found to be genetically at risk for Type 1 to better study the development of diabetes. These trials, taking place in Germany (BABYDIAB), Finland (DIPP), Denver, CO (DAISY), and Gainesville, FL (PANDA), include studies of the participants from the general public and at risk families (e.g., Nejentsev et al., 1999; Rewers et al., 1996; Schatz et al., 2000; Schenker et al., 1999; Ziegler et al., 1999).

Opponents of screening argue that without a prevention strategy, studies should avoid disclosing results to participating families and that if disclosure is necessary then research should only be conducted with infants who have a first degree relative with type 1 diabetes (Friedman Ross, 2003). Critics argue that screening under any other circumstances may result in harm to children and their parents. Friedman Ross (2003) stated that genetic screening can only convey at most a susceptibility that is a 20% probability. She claims that as a result of these tests, parents may prepare unnecessarily and treat their child as ill (Friedman Ross, 2003). As the debate continues regarding the merits of genetic screening of the general population and as interest in diabetes prevention continues to rise, research on the psychological and behavioral impact of genetic screening becomes timely and highly relevant.

Psychological Impact of Diabetes Screening

Relatively little research has been conducted examining the parents' psychological reactions to participation in a newborn screening program for type 1 diabetes. However, parents have indicated favorable attitudes towards risk screening and prevention trials for

type I diabetes (Lucidarme et al., 1998; Ludvigsson et al., 2002). To explore the psychological impact of risk screening, Dr. Johnson and her research group have conducted several studies of adults and children identified as at risk via autoantibody and newborn genetic screening (Carmichael et al., 2003; Johnson, 2001; Johnson & Carmichael, 2000; Johnson & Tercyack, 1995; Johnson et al., 1990). As explained above, a determination that an individual is at risk as identified through presymptomatic screening does not mean an individual will definitely develop diabetes. How this information and level of uncertainty impacts individuals, particularly newborns and their families, is an important factor to consider when evaluating the ethical nature of genetic risk screening.

In one of the first studies in this area, Johnson et al. (1990) reported individuals found to be at high risk (as identified through ICA screening) and their family members exhibited clinically significant levels of anxiety subsequent to at risk notification. Those testing ICA+ were told their chances of developing diabetes were 50%. Johnson and Tercyak (1995) subsequently found notification of islet cell antibody positive (ICA+) status had an emotional impact on the at risk individual (adults and children) and their family members (i.e., spouses, parents). Initial notification was associated with considerable situationally-specific anxiety (as measured by the state portion of the State-Trait Anxiety Inventory (STAI; Speilberger, 1970) and the State-Trait Anxiety Inventory for Children (STAI-C; Speilberger, 1973) in both individuals with the risk and their family members. This was especially true in parents of ICA+ children. In addition, parent and child anxiety was highly correlated. However, initial anxiety seemed to decrease to normal levels over time, as measured in a 4-month follow-up interview.

In a similar study with fewer participants, Galatzer et al. (2001) examined antibody positive children (n=10) and their parents using the Impact of Events Scale (IES; Horowitz, 1979) and found that high levels of distress reported by parents upon results notification decreased by the 3-month interview. Galatzer et al. compared their results with a study of parents of children newly diagnosed with type 1 diabetes (Kovacs, 1985) and found similarly strong emotional reactions, but more so in the group of parents of children with diabetes. Another small-scale study conducted by Yu et al. (1999) (n= 88) found notification of high-risk genetic status in newborns was not associated with increased parenting stress as measured by total stress score (TSS) of the Parenting Stress Index (PSI; Abidin, 1990) more than three months after notification.

A follow-up study to Johnson & Tercyak (1995) examined how individuals found to be at risk (ICA+) coped with their own or a loved one's at risk status, by administering the Ways of Coping Checklist-Revised (WCC-R; Folkman & Lazarus, 1980) (Johnson & Carmichael, 2000). Using this multi-dimensional measure allows for closer examination of coping styles (i.e., problem-focused, seeking social support, wishful thinking, avoidance, self blame) beyond the concept of monitoring vs. blunting found in previous cancer genetic screening studies. Johnson & Carmichael (2000) found at risk children used more avoidance coping (e.g., tried to forget the whole thing, kept your feelings to yourself; slept more than usual) than at risk adults, mothers of at risk children, or spouses of at risk adults. At risk children also used more wishful thinking (e.g., hoped a miracle would happen; wished the situation would go away) than at risk adults. Initial state anxiety in response to risk notification was related with subsequent coping as mothers who were more anxious tended to use more wishful thinking, avoidance, and they tended

to blame themselves for their child's at risk status. Coping strategies appeared to influence the maintenance of anxiety over time as mothers who blamed themselves tended to remain anxious.

In the late 1990s, testing moved from biological to genetic markers, and from family cohorts to the general population. Carmichael et al. (2003), Johnson & Carmichael (2000) and Johnson et al. (submitted) interviewed mothers of infants at risk for developing Type 1 diabetes as identified through participation in the longitudinal Prospective Assessment of Newborns for Diabetes Autoimmunity (PANDA) study (Schatz, 2000). As described earlier, PANDA involves HLA genotyping and serial antibody screenings over time. Interviews assessing the psychological impact of participation in PANDA were conducted approximately 4 weeks post-notification, and again 4 and 12 months after notification. Similar to the ICA+ studies, they found maternal anxiety levels were clinically elevated after initial notification of risk status, but appeared to dissipate over time to normal levels (Johnson et al., submitted).

Risk understanding was examined in mothers who participated in the initial and 4-month follow-up PANDA interviews (Carmichael et al., 2003). Almost 75% of mothers gave a correct estimate of their child's genetic risk at the initial interview; however, over time, mothers were less likely to be accurate and more mothers underestimated their child's risk. Overall, very few mothers overestimated their child's risk. Mothers who were Caucasian and who had higher levels of education were more likely to be accurate. Mothers whose children were in the highest risk group were least accurate. Mothers of children with a family history of a first degree relative with diabetes were more likely to underestimate their child's risk at the initial interview. Maternal anxiety was a predictor

of risk underestimation at the 4-month interview, but was not significant in predicting to earlier underestimation or accuracy at either time point. As one might expect, mothers who were more anxious were less likely to underestimate their child's risk.

In studies of maternal anxiety in this population, initial anxiety levels were found to be higher in mothers who were Hispanic, with less education, in those whose infants were at greater risk, and in mothers who overestimated their child's actual risk (Johnson et al., submitted). Coping strategies also appeared to be related to anxiety as wishful thinking and blaming one's self predicted anxiety at the 4 and 12-month follow-up interviews (unpublished data). As explained in later chapters, participants for this study were recruited from this larger sample.

These studies, taken together, suggest newborn screening does not have long-term detrimental effects on parental adjustment, as measured by either anxiety or stress. Additionally, it appears that a majority of mothers correctly recall their infant's risk with few mothers overestimating their child's risk and consequently becoming more anxious. These findings are congruent with other studies of genetic testing previously discussed. It is likely parents' reactions to the news and subsequent coping style may influence an individual's or family's decision to participate in longitudinal trials or natural history studies, such as PANDA, that will provide the scientific bases of a prevention or cure for type 1 diabetes. These studies can play an important role in informing debates about the ethics of newborn screening.

Behavioral Impact of Diabetes Screening

Johnson & Tercyak's (1995) study of ICA+ children and adults, assessed after notification of screening results, found 52% of ICA+ children and 24% of ICA+ adults reported making a change in their behaviors and/or lifestyle in an attempt to delay or

prevent the onset of Type 1 diabetes. While details were not reported, the authors made a general statement that these reported changes most often reportedly occurred in the areas of diet and increased exercise. The authors also found that a higher level of anxiety was associated with greater lifestyle/behavior modifications. Similarly, in a later study of genetically at risk infants, mothers who continued their child's participation in the longitudinal PANDA study tended to be more anxious with infants at higher risk. Mothers who believed their at risk children would never get diabetes were less likely to continue study participation (Carmichael et al., 1999b).

In a recent study of intentions for behavior change, Hendrieckx et al. (2002) surveyed a sample of 403 adults with first-degree relatives with type 1 diabetes who were undergoing antibody screening for type 1 diabetes and were assessed prior to results notification. This novel study sought to better understand the relationships between perceived control, distress, and behavioral intentions. Results indicated 73% of participants stated they intended to make a lifestyle change if found to be at high risk, with diet (87%) and exercise (30%) most frequently endorsed (Hendrieckx et al., 2002). These results suggested individuals' beliefs regarding the prevention of type 1 diabetes did not correspond well with current scientific knowledge; however, beliefs appeared more congruent with an understanding of type 2 diabetes (Hendrieckx et al., 2002). Hendrieckx et al. (2002) found general anxiety did not appear to be a significant predictor of behavior change, nor were behavioral intentions predicted by education level. However, similar to Johnson & Tercyak (1995), diabetes-specific worry was related to intentions towards behavioral change, along with perceived internal control. Hendrieckx et al. (2002) also found those who were female, married, and older were more likely to

report anticipating making lifestyle changes. Additionally, perceived internal control was related to beliefs regarding the causes of diabetes. More specifically, those who believed their relative developed diabetes largely due to heredity or chance, were more likely to believe they were unable to do something to reduce their risk of developing diabetes. While this study provided important exploratory data, the results were limited because the data were collected prior to the screening results, and intentions --rather than actual behaviors-- were assessed. Additionally, it used several new measures, which have not been psychometrically validated.

Additional data on diabetes screening and behavioral change come from the Participant Experience Survey, designed by Johnson, for participants who completed the DPT-1 study (Johnson, 2002). The survey was administered anonymously across study sites to examine subjective experiences of participants (who were at least 10 years old) in the trial, as well their parents (of participants under the age of 18). Questions assessed a broad range of issues, including study adherence, satisfaction, reasons for participation, perceived need for psychological support, and efforts to prevent or delay type 1 diabetes from developing. Items that assessed efforts to prevent/delay type 1 diabetes were designed to reflect intentional changes in weight diet, exercise, lifestyle, stress level, monitoring, and alternative medication use. Items were scored as either "yes" or "no" to reflect whether or not an individual reported engaging in a specific behavior. Only data from those who were unaware of the study's results were analyzed. Sixty-five percent of DPT-1 participants, who were all over the age of 10, responded to the survey, with 82 from the intervention (IN) group (which received preventative insulin) and 81 from the close observation (CO) group. Over half (54%) of all participants reported modifying at

least one behavior in an effort to delay or prevent type 1 diabetes onset, with no significant differences between groups, with the exception of alternative medication use (significantly greater use in IN group). Results indicated dietary changes were the most common behaviors reported, with approximately one-third of participants stating they reduced candy or sweets intake, reduced intake of regular soda, or increased intake of diet soda. Twenty-eight percent of participants indicated they would increase their physical activity. Seventeen percent stated they took alternative medications. Ten percent of participants reported attempting weight loss. Seven participants (4 in the experimental group and 3 in control) stated they took extra insulin in an effort to delay or prevent diabetes onset. No significant predictors of behavior change were found in this study. These results were congruent with Tercyak & Carmichael (1995) and Hendrieckx et al. (2002) and indicated a substantial proportion of individuals' who are found at risk for type 1 diabetes engage in behaviors that correspond to those found to be effective in the treatment and prevention recommendations for type 2 diabetes (ADA, 2002b; Tuomilehto, et al., 2001). Data are currently available from parents whose children participated in the DPT-1 study. These data were used as a comparison group in the current study.

These studies described herein suggest there are unanswered questions to be explored regarding the behavioral outcomes associated with risk screening for type 1 diabetes. Existing data suggest individuals report engaging in behavior changes in response to risk information, although it is unclear what predicts these behavioral efforts. Based on existing literature, risk perception, perceived control, and psychological distress appear important factors to consider. Behavior changes that result from risk notification

may or may not be related to scientifically validated methods of risk reduction. However, in the case of type 1 diabetes, whether a behavior is scientifically valid is not necessarily important, since we do not currently know what delays or prevents the onset of the disease.

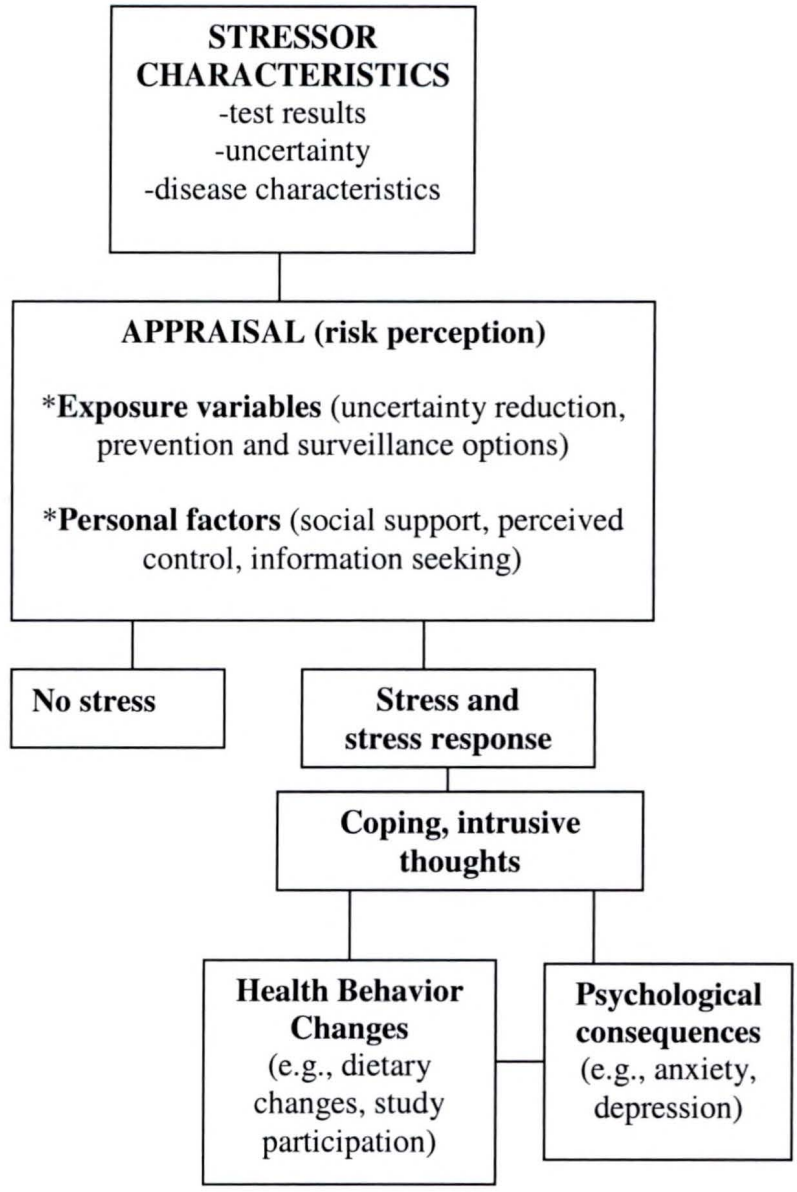

Figure 2-1. Partial representation of the Stress-Disease Risk-Coping Model adapted from Baum et al. (1997)

CHAPTER 3
RATIONALE AND PURPOSES

The purposes of this study were to better understand predictors of self-reported behavior change in mothers of newborns who were identified as at–risk for type 1 diabetes through genetic screening. Currently, little is understood about the specific behavior changes that result from knowing one's child is genetically predisposed to a condition for which there is currently no known prevention method or cure. Additionally, our present understanding of the etiology of type 1 diabetes suggests it develops from a combination of both genetic and environmental influences, which are not well-defined. In the absence of definitive recommendations from the health care community, mothers of newborns identified as "at-risk" may take actions they believe are effective in preventing type 1 diabetes in their children.

Based on previous studies, possible behavioral changes may include altering their children's environment, feeding schedules, activity patterns, and/or medical surveillance behavior (Hendrieckx, 2002; Johnson, 2001; Johnson, 2002). These efforts may represent mothers' attempts to reduce their anxiety and better cope with the situation. However, we have yet to document the nature and extent of such behavior changes, including the incidence of excessive prevention efforts that may become burdensome and impact daily functioning. Further, since the onset of diabetes is thought to be an interaction between genetics and the environment, it is unclear to what extent certain types of behaviors could advance or delay disease onset. Although current science does not permit us to

recommend certain behaviors as preventative, it is important for us to monitor the role of relevant behaviors if we are to understand the natural history of this disease.

Monitoring possible behavior change associated with high-risk notification is equally important to current and future diabetes prevention trials. In the DPT-1, for example, behavior change efforts taken by the control group (e.g., taking insulin or nicotinimide) could undermine the trial's internal validity. Unless these behavior changes are monitored, interpretation of study results can become exceedingly difficult. This is not unique to diabetes-or genetic screening-specific trials as certain behavior changes could potentially impact other types of clinical trials as well.

This study involves both qualitative and quantitative data to examine reported behavioral outcomes associated with participation in the Perspective Assessment of Newborn Diabetes Autoimmunity (PANDA) study. Findings will be examined in the context of Baum and colleagues' (1997) model of genetic testing, in which behavior change in response to genetic test results is influenced by one's risk appraisal, affective response to the information, and available coping resources. Objectives of the study are listed below.

Objective 1: To Investigate the Extent of Reported Maternal Behavior Change as a Result of Genetic Screening for Type 1 Diabetes

Hypothesis 1.1: Reported behavior changes will most likely correspond to recommendations for the treatment of diabetes (American Diabetes Association (ADA), 2002a, 2002b) and the prevention of type 2 diabetes (Pierce et al., 1995), including changes in diet and physical activity patterns (Forsyth et al., 1997; Pierce et al., 1995).

Rationale: There is scientific uncertainty regarding the environmental factors associated with the development of type 1 diabetes. The health care community and

media have recently focused significant attention on type 2 diabetes, advocating for healthy lifestyle changes. In a study of parents with type 2 diabetes, nearly half thought they could reduce their children's risk of developing diabetes by altering their children's diet and exercise patterns (Pierce et al., 1999). Mothers, who may not understand the distinctions between type 1 and 2 diabetes may apply such recommendations to their children at-risk for type 1. This hypothesis is congruent with findings from Johnson & Tercyak (1995), Hendrieckx (2002), and unpublished data from the DPT-1 survey (Johnson, 2002).

Objective 2: To Assess Predictors of Maternal Behavior Change as a Result of Genetic Screening for Type 1 Diabetes

Hypothesis 2.1: Mothers who perceive they have control over their child developing diabetes will be more likely to report engaging in behavior changes.

Rationale: Mothers may be more likely to report taking action to help prevent such an outcome if they believe they have some control over the situation. Behavioral change to reduce a health threat is more likely if there is a belief that change can be affected (Diefenbach et al., 1999; Hendrieckx et al., 2002). Perceived control is related to both uncertainty reduction and available prevention/surveillance options, which are integral to risk perception, a key component of health behavior change (Baum et al., 1997). Therefore, perceived control will be examined in the context of other predictors of behavior to determine possible interaction effects.

Hypothesis 2.2: Mothers who perceive their children to be at greater risk will be more likely to report engaging in behavior change.

Rationale: Perceived risk, more so than infant's actual risk, will be a better predictor of behavior. Mothers of children who perceive their children to be at higher

risk than has been identified through testing (overestimate their risk) will be more likely to engage in behavior change. Mother of children who underestimate their child's actual risk will be less likely to engage in behavior change. Children who are perceived to be at high or extremely high risk may have mothers who will be more likely to try to intervene. Studies of genetic screening for breast cancer have found that increased perceived risk predicts likelihood of engaging in health behavior change and health surveillance behaviors (e.g., Aiken et al., 1994; Meiser et al., 2000; Ritvo et al., 2002; Schwartz et al., 1999).

Perceived control may also interact with risk perception as mothers who perceive their child to be at greater risk may be more likely to engage in behavior change if they also believe they are able to control whether their child develops diabetes.

Hypothesis 2.3: Mothers who are more anxious will be more likely to report engaging in behavior change.

Rationale: Mothers who are more concerned and worried about their child developing diabetes will be more likely to report taking preventative actions. Studies indicate disease-specific worry (Hendrieckx, 2002; Johnson and Tercyak, 1995) and beliefs regarding the effectiveness of preventative actions (Diefenbach et al., 1999) predicted either intentions for behavior change or increased adherence to health-protective behaviors. Maternal anxiety may also be related to the degree of maternal perceived control, which may in turn influence behavior. Mothers who are more anxious/worried may report more behavior change if they also believe they have some control over the situation.

Hypothesis 2.4: Mothers who use more coping strategies, particularly active coping (i.e., problem-focused, seeking social support), will be more likely to report behavioral changes.

Rationale: Engaging in risk reducing behavior can be seen as a means of coping with a health threat. Behavior change is an active coping approach and likely to be associated with other ways of coping, particularly those that are also more active, namely problem-focused coping and seeking social support. Mothers who use avoidant strategies, and try not to think about the problem, may be less likely to engage in risk-reducing behaviors.

Additionally, those who perceive greater control over the situation may be more likely to engage in more proactive coping methods, whereas, mothers who perceive less control may engage in more avoidant coping and be less likely to report behavior change.

Hypothesis 2.5: Mothers who report information seeking and/or report receiving recommendations from medical professionals or other family members related to behavior change will be more likely to report engaging in behavior change.

Rationale: Mothers who are given advice to change their behavior by those they feel are authoritative will be more likely to follow through with recommendations. Research findings suggest "monitors" (or information-seekers) are thought to cope more effectively with stressful situations using more problem-focused, information-obtaining coping strategies rather than avoiding the situation and not seeking out information (Scheier et al., 1986; Carver et al., 1989). Participation in research studies may be viewed as an additional form of information seeking about health status. Information seeking may also influence one's sense of perceived control, and consequently, may

influence behavior both directly and indirectly. Those who seek and utilize information from various sources may perceive greater control over the situation, and consequently, be more likely to report behavior changes. Therefore, information seeking will be measured in this study and used as an independent predictor, as well as in conjunction with perceived control.

Hypothesis 2.6: Mothers who continue participation in the PANDA Part II study (repeated blood testing for antibodies), will be more likely to report other behavior changes.

Rationale: Health surveillance behaviors, such as participation in additional blood draws, may be a likely outcome following risk notification. Increased health surveillance may also signify increased contact with health care professionals. For those who continue in the PANDA study, the risk of developing diabetes may be more salient and seen as something they should address. Mothers who continue in the study have contact with investigators and study staff over time and therefore, this contact may influence their behavior. Mothers who are sufficiently concerned about their child's risk enough to monitor their child's risk more closely, may be more likely to report other behavior changes. Data on participation in PANDA Part II blood draws are available and these data can be compared with maternal report.

Objective 3: To Assess Psychological Effects (i.e., Anxiety) of Maternal Behavior Change Over Time

Hypothesis 3.1: Mothers who report modifying behaviors will show a greater reduction in anxiety over time than mothers who do not report behavior change.

Rationale: Behavior changes, including health surveillance behaviors, may represent means of coping with a health threat. Engaging in behaviors perceived as risk

reducing may help lower maternal anxiety regarding the situation. However, the influence of reported behavior change on maternal anxiety may be influenced by maternal perceived control (i.e., mothers who perceive control over diabetes onset and engage in behavior change may show greater reduction in anxiety).

Objective 4: To Compare Reported Behavior Change between Mothers of Children Genetically at Risk for Developing Type 1 Diabetes with Mothers of Children in the Diabetes Prevention Trial Who Were ICA+, and Therefore, at Even Greater Risk for Diabetes Onset

Hypothesis 4.1: Mothers of genetically at-risk children will be less likely to report behavior change than mothers of ICA+ children enrolled in Diabetes Prevention Trial-1 (DPT-1).

Rationale: Participants enrolled in the DPT-I trial were at increased risk for diabetes as identified through positive family histories and positive ICA screening. Their risk level was collectively higher than 98% of our original total sample population for the PANDA Part III study (of whom 7 of 435 were ICA positive). For this reason, we believe that mothers in the current study will report fewer behavior changes than mothers of children in the DPT-I since children of mothers in the proposed study are at relatively less risk than the DPT-1study children.

Examining this hypothesis will allow us the unique opportunity to explore differences between maternal reports of behavior change in two at-risk groups: children identified at birth as genetically at-risk and higher-risk children who have entered a prevention trial.

Pierce et al's (1999) study of parents with type 2 diabetes found that those who believed they could prevent their children from developing diabetes and who perceived

their child's risk to be higher, were more likely to experience greater anxiety (Pierce et al., 1999)

For the purposes of examining this hypothesis, maternal data from the DPT-1 survey (n = 134) will be compared with mothers' reported from the PANDA study. Based on DPT-1 participant data, over half of the sample reported at least one behavior change, with dietary changes most often reported, followed by increased exercise, weight loss attempts, and alternative medicine use (i.e., vitamins) use (Johnson, 2002).

CHAPTER 4
METHODS AND MATERIALS

Prospective Assessment of Newborn Diabetes Autoimmunity (PANDA) Study Procedures

Part I

Participants were mothers whose infants were screened at birth to determine their genetic risk for the development of Type 1 diabetes (1997-1999) through Part I of the Prospective Assessment of Newborn Diabetes Autoimmunity (PANDA) study. This study is a National Institutes of Health and Juvenile Diabetes Research Foundation Internal-supported registry that uses genetic testing to identify newborns at risk for type 1 diabetes (Schatz et al., 2000). In this study, mothers were contacted at the time of their child's birth and asked permission to screen the newborn for the presence of the high-risk HLA-DQB1 alleles using blood spots on filter paper (obtained by heel stick at the time of state-mandated phenylketonuria testing). Informed consent was obtained and consenting participants were told they would only be re-contacted if their child was at increased risk for type 1 diabetes. The majority of these women gave birth at participating locations in Gainesville, Florida, or Pensacola, Florida, were all English speaking, and were over the age of 18.

PANDA genetic testing results placed infants into one of six risk categories: very low risk (1/6000), low risk (1/300), slightly increased risk (1/125), moderate risk (2/100) high risk (5-10/100) and extremely high risk (20-25/100) (Table 4-1). Only children who

were at moderate, high or extremely risk children are followed longitudinally in the PANDA study.

If a child was determined to be "at risk," in other words, classified as either at "moderate," "high" or "extremely high risk," mothers were sent letters notifying them that their children's genetic test results were available. Results were usually available after approximately 12-20 weeks following birth. In the letters, participants were requested to call the PANDA study coordinator to discuss the results and possible continued study participation in PANDA Part II according to PANDA protocol. If no response was received approximately 30 days after sending the notification letter, the PANDA study staff attempted to contact the mother by phone to notify her of her infant's risk.

For mothers who called for results or were contacted by phone, the study coordinator followed a scripted presentation of the risk information, including both categorical and numerical risk figures. Additionally, she presented available options, including participation in the PANDA Part II study, and an opportunity to ask questions about the study or the meaning of the results. Parents had the option at that time to decline further participation, continue with the study, or delay their decision. Regardless of participation status in the PANDA Part II study, all mothers were asked for their permission to be contacted by a second individual from the Pediatric Psychology Research lab who would ask them questions about their understanding of the study and its psychological impact. See Figure 4.1 for procedural outline of entire PANDA study.

Part II

Part II of the PANDA study involves longitudinal follow-up of children screened at birth. These children are periodically screened (via blood draws) starting when the child

is at least six months of age for the presence of autoantibodies, which are additional markers of diabetes disease progression. A positive screening for autoantibodies would suggest that the child is at even greater risk of developing type 1 diabetes. Blood draws could be conducted either by (1) mailing out supplies to parents and to have their pediatricians draw the blood and mail back to the PANDA staff or (2) scheduling directly at study sites in Gainesville, Orlando, or Pensacola, Florida. Blood draws were expected to occur every at 3, 6 ,or 12 months, depending on risk level.

Part III

Part III of the PANDA study examined the psychological impact of participation in PANDA, including maternal affective (i.e., anxiety) and cognitive responses (i.e., risk understanding) as well as coping response. Mothers who agreed to be contacted at the time of notification were interviewed by telephone approximately 4 weeks following notification ($\underline{M}$ = 3.50, $\underline{SD}$ = 1.96) and again at 4 ($\underline{M}$ = 3.93, $\underline{SD}$ = 1.96) and 12 months ($\underline{M}$ = 12.83, $\underline{SD}$ = 2.45) post-notification (see Figure 4-1).

For the initial interview, the Part III participation rate was high. Approximately 90% ($\underline{n}$ = 435) of the mothers we were able to contact (of over 700 eligible) agreed to complete the initial interview, 79% participated in a second interview ($\underline{n}$ = 344), and 62% participated in the third interview ($\underline{n}$ = 269). Sixty percent ($\underline{n}$ = 262) completed all three interviews. Of those who did not complete all three interviews, 67 declined to be contacted beyond the first interview (no attempts were made to contact these mothers to participate in the current interview), and 106 were unable to be contacted by phone due to either disconnected numbers or the time that had elapsed between or study personnel could not reach them.

Participants

To be eligible for the current study, mothers must have completed at least the initial interview of the PANDA Part III study and at no point declined participation in either of the subsequent two interviews (n = 368). Out of 368 eligible mothers for the current study (i.e., those with > 1 previous interview and who did not previously decline participation), 204 were successfully contacted (55%). Of these mothers, 192 (94%) completed the interview, ten declined participation (5%), and two mothers were no longer eligible because their "at risk" children had recently developed type 1 diabetes (1%). Of the 163 mothers who could not be contacted (44%), 145 had disconnected numbers and/or had no forwarding contact information, and 18 mothers with presumably correct up-to-date contact information were "unable to be contacted" after multiple attempts. Families were deemed "unable to be contacted" when there was no response after at least fifteen attempts were made over a two-month period, with at least three messages left if a family member or answering machine was available.

Maternal Characteristics

Mothers who completed the current interview ranged in age from 20 to 46 (M = 33.67, SD = 5.38). Eighty five percent of mothers were married and 44% had a 4-year college degree at the time of interview (Table 4-2). Eighty-five percent of mothers where Caucasian and therefore, minority members were under-represented compared to the population in Alachua County, Florida, and Florida in its entirety, where among women of childbirth age (18-44 years) approximately 25% and 23% are minorities, respectively (Florida Office of Economic and Demographic Research, 2001). On average, mothers in this sample had two children at the time of interview (M = 2.09, SD = 1.11). Eighty three

percent of participating mothers completed all three interviews and 62% attended at least one blood draw.

Child Characteristics

Target at risk children of participating mothers were between the ages of 2 and 7 years ($\underline{M}$ = 4.25, $\underline{SD}$ = 0.89) and evenly split between males and females (Table 4-3). Within this at risk sample, the majority of infants were at "moderate" risk (56%), 37% were at "high" risk, and 7% were at "extremely high" risk. Five out of the six eligible mothers of children who were antibody positive participated in the current interview. Most children of participating mothers were reported as having a family history of diabetes (72%) (type 1 or 2). Sixty-five percent of children have at least one distant ($\geq$ second degree) relative with diabetes. Thirty-seven children (19%) have at least one first-degree family member with diabetes. Of these, 30 children have immediate family members with type 1 diabetes (81%), including 15 participating mothers themselves, along with seven fathers and 14 siblings. In five of these families, two immediate family members have type 1 diabetes.

Procedures

The current interview was conducted at least one-year post PANDA Part III study completion and therefore, two to four years post-notification ($\underline{M}$ = 3.60, $\underline{SD}$ = 0.78). Attempts were made by the Principal Investigator or research staff to contact all eligible mothers (n = 368) for an additional follow-up interview to measure reported maternal behavior changes resulting from knowledge of their children's risk for type 1 diabetes. Contact information for these mothers was kept within a computerized database with restricted access, so telephone numbers were available only to study staff.

When participants were contacted, they were reminded of their earlier participation in PANDA Part III interviews and asked if they would agree to participate in an additional interview. Participants were reminded of the voluntary and confidential nature of the study and those who agreed to participate, were given a $5 gift certificate to Publix or Target (their choice) as a token of appreciation.

Asking mothers about their behavior might have had the potential to raise mothers' anxiety and curiosity levels regarding what they should or should not be doing to help their children. Therefore, at the beginning and end of the interview, there was a disclaimer read to remind mothers that we do not currently know what causes type 1 diabetes, and that we did not have specific recommendations to offer other than encouraging a health lifestyle, including a healthy diet, physical activity, and rest. For mothers who asked more specific questions beyond this, we had prepared documents from the American Medical Association (AMA) on developmentally appropriate guidelines regarding eating, exercise, and sleep (found at www.ama.org; last accessed 6/1/02), which could be mailed to mothers upon request. Eleven mothers requested additional information. The most frequently requested materials pertained to information regarding signs and symptoms of type 1 diabetes.

For quality assurance purposes, data were entered twice into a computerized database, systematically compared and cleaned, before analyses were conducted. Data from this interview were linked to previously collected data (PANDA Part III) on these study participants through their unique identification numbers assigned by the PANDA study staff. This allowed for longitudinal analyses of the data. This study was approved by the UF Health Science Center Institutional Review Board (9/1/02) and documentation

of written consent was waived. Funding for this study was obtained from the North Central Florida's Children's Miracle Network.

Measures

Descriptive Variables

Descriptive data were collected to examine the maternal and child demographic characteristics, overall participation rate as well as demographic differences between mothers who agreed to participate versus those who declined or were unable to be contacted (for further detail see section on "Predictor Variables"). These two groups of mothers were compared across outcome and predictor variables based on data from the initial interview.

Outcome Variable: Reported Behavior Change

A component of the structured interview was developed to assess behavioral changes across six domains: (1) diet/eating patterns, (2) physical activity, (3) emotional stress, (4) medical interventions, (5) medical surveillance, and (6) illness prevention behaviors. These questions and constructs were adapted from the Participant Experiences Survey used in the Diabetes Prevention Trial-1 (DPT-1) study (Johnson, 2002) and constructs were classified based on the DPT-1 survey and Hendrieckx et al. (2002). Additional questions were added to address other potential environmental triggers or influences hypothesized in the research literature to be related to diabetes development (Akerblom, et al, 1998) (see Appendix A).

Diet and eating patterns (α = .58). Sixteen questions addressed changes in the frequency, amount, and types of food/drink (i.e., sweets, soda, juice, cow's milk) given to the child as well as attempts to modify the child's weight. Also, included were questions assessing changes in early feeding history, including timing of the introduction of solid

foods and breastfeeding. The 16 questions represented ten different types of behavior changes, as some questions are paired to assess in which the direction changes occurred (i.e., decrease vs. increase). Additionally, two questions referred to behaviors for which the concepts of frequency and duration do not apply, and therefore, these detailed follow-up questions were not asked.

Physical activity/Physical stress ($\underline{\alpha} = 0.54$). Four questions assessed whether mothers increased or decreased their children's physical activity or physical exertion in response to their child's risk for type 1 diabetes.

Emotional stress ($\underline{\alpha} = 0.47$). Four items were designed to assess lifestyle changes that foster the reduction of the child's level of emotional stress.

Medications ($\underline{\alpha} = 0.54$). Five items addressed whether mothers provided their children with medications, such as dietary supplements, vitamins, or insulin.

Illness Prevention ($\underline{\alpha} = 0.72$). Eight questions representing seven unique concepts, assessed the degree to which mothers altered their children's environment to minimize risk of illness or infection.

Medical surveillance ($\underline{\alpha} = 0.37$). Five questions were designed to assess whether participants engaged in health- monitoring behaviors for their children, such as more frequent doctor's visits, glucose monitoring, and reported participation in PANDA Part II study (autoantibody screening). However, reported PANDA Part II participation was not included in calculating the domain score, as it was used for reliability purposes and also used as a predictor variable.

This portion of the interview began with a simple "yes/no" question assessing if, in general, participants felt they engaged in any behavior change to prevent diabetes in

their child. This question was followed by more detailed questions regarding different types of behaviors relevant to the six domains described above. For each section, participants were first given an open-ended question to solicit spontaneous answers (e.g., Have you done anything different with your son's physical activity patterns to prevent him from developing diabetes?) followed by more detailed forced-choice questions. When a response was given to an open-ended question that would be later addressed by a forced choice item, the corresponding forced choice item was also endorsed.

Within each domain, forced choice items were designed to assess a wide variety of behavior changes. For each question, participants were reminded that these questions apply only to behaviors initiated specifically to prevent diabetes in their children. For forced choice questions in which the response was "yes," follow-up questions were asked to assess duration/consistency and frequency of given behavior. Forced choice items were scored as either "yes" or "no." Duration or consistency of the behavior was scored as "never" (0), if the behavior never occurred, "inconsistent" (1), if the behavior was initiated early on but stopped, or began only recently; or "consistent" (2), if the behavior has been ongoing since results notification. Frequency of a behavior was scored as "never" (0), "occasionally" (1) or "always/nearly everyday" (2).

Each question was scored as dichotomous "yes/no" (0 or 1) as well as given a continuous composite score value for duration, frequency, and duration x frequency. However, duration was relatively static as 86% of those endorsing a certain behavior reported consistent engagement since time of notification. As for frequency of behaviors, 60% reported engaging in the behavior "always/nearly everyday" (Table 5-8).

Due to relatively low frequencies for most items and low variability in duration scores, only the dichotomized "yes/no" scores were used for analyses. Domain scores were calculated in two ways: (1) calculating sum of the number of behaviors endorsed and (2) whether at least one behavior change occurred with each domain. A total score for behavior change was similarly obtained by collapsing domains. A factor analysis of this measure was not conducted due to low variability on the items and inadequate sample size for the number of items in the measure. To determine the statistical strength of the scores for the six domains and total score, coefficient alphas were calculated to determine the reliability of each construct (Table 4-4).

Reliability was relatively strong for the total behavior score ($\alpha = 0.77$) and illness prevention domain ($\alpha = 0.72$), but weaker for the other five domains, with alphas ranging from 0.37 to 0.58. Correlations between domain scores ranged from 0.10 to 0.44 (Table 4-4). As expected given the data, the total behavior score was best correlated with diet and health surveillance behavior scores. Due to the low frequency of behavior changes within several domains, as well as the non-normal distribution and relatively poor reliability of domain scores ($\alpha < 0.60$), no further analyses of domain-specific behaviors were conducted (Table 4-5). Additionally, due to the relative low frequency of endorsement of items overall and the non-normal distribution of the total behavior score, the total behavior score used in subsequent analyses was the dichotomous variable of whether at least one behavior change was reported (1 = 'yes') versus no behavior change reported (0 = 'no') (see Figure 4-2).

Reliability of self-reported behavior change. Self-reported participation in Part II of the PANDA study was collected in the structured interview and compared with data on

actual Part II participation. These data were available through the PANDA computerized database. Continuation in the PANDA Part II study was defined as those mothers who brought their child in for at least one blood draw for autoantibody screening, coded as "participated" (1) and "did not participate" (0). Actual participation in PANDA II blood draws was the only observed behavioral data available to us. It permitted us to examine the validity of maternal self-report data concerning this particular component of medical surveillance. PANDA Part II participation data indicated that 61% of mothers participated in at least one subsequent blood draw and 26% participated in two or more. When asked in the interview, 174 mothers reported accurately whether they participated in Part II of the study (91%), with 72 accurately reporting they had not continued participation and 102 correctly reported they had. Three mothers reported participating when they actually had not (1%), and 15 reported they had not participated when they actually had (8%). These findings suggest mothers may have been open and honest when completing the interview and that social desirability effects were not strong. If anything it is possible mothers may have underreported efforts to prevent diabetes.

Predictor Variables

Sociodemographics

The following variables were assessed during the first PANDA Part III telephone interview: date of interview (to calculate length of time since notification), maternal date of birth, child date of birth, maternal and paternal education level, family income bracket, maternal and child ethnicity, marital status, number of children and whether or not this is her first child. The number of first-degree relatives, second-degree relatives, or greater relatives of the child with type 1 or type 2 diabetes was also assessed, if known. In the current interview, several non-static demographic variables were updated in this current

interview to ensure that information was current, including marital status, number of children, family income bracket, maternal and paternal education level, and family diabetes history. Mothers were also asked for a current address in order for gift certificates to be sent.

Perceived control

This construct was assessed by a series of questions adapted from a questionnaire developed by Bradley et al. (1999) and used in Hendrieckx et al. (2002). These questions assessed whether participants believed there was anything a parent or a medical professional could do to prevent diabetes in the children, as well as a question about diabetes onset being determined by chance or fate. Responses were scored on a 5-point Likert scale, anchored by "strongly disagree" (scored as 1) and "strongly agree (scored as 5)." Internal consistency of this 3-item scale was $\underline{\alpha} = 0.55$. This was unsatisfactory based on the study's criteria of using an alpha score of 0.60 as the cut-off for acceptable reliability. However, when chance was not retained as a part of this composite, internal consistency increased ($\underline{\alpha} = 0.66$), therefore, only the two-item measure of perceived control was retained as a composite measure (Table 4-7). The composite score of perceived control was calculated by averaging the scores of the two items (Table 4-6).

Risk perception

(1) *Perceived absolute risk.* An absolute measure of perceived risk and its accuracy was assessed in the previous interviews and was assessed in a similar way in the current interview. Mothers were presented with a list of the possible risk categories (with numerical estimates) and asked whether or not any of these were the risk group they were told their child was in. "I don't remember" was recorded if they were unable to recall or recognize their child's risk category or number. (2) *Perceived estimated risk.* Perceived

risk was considered accurate if the participant was able to recognize the infant's correct risk status from the list. Responses were classified as "accurate" (scored 2), "overestimates" (scored 3), "underestimates" (scored 0), or "unknown" (scored 1) based on the relationship of the response to the child's actual risk status This component reflected perceived absolute risk while controlling for actual risk and was included in the composite score of risk perception, whereas, absolute risk was not. (3) *Perceived comparative risk.* A question adapted from Hendrieckx et al. (2002) assessing perceived comparative risk was included. The question was stated as follows: "How do you think your child's risk for developing diabetes compares to other children?" The response was rated on a 5-point Likert scale ranging from 1 to 5, anchored by "much lower" and "much higher." (4) *Expectations.* A question used in all previous interviews assessed whether participants believed their child will develop diabetes. This question was coded as "yes, my child will develop diabetes in the near future" (scored 3), "my child will eventually develop diabetes but not for a long from now," (scored 2) "my child will not ever develop diabetes," (scored 0) or "I am unsure." (scored 1) This variable was previously used in Carmichael et al. (1999).

Intercorrelations between the three risk perception variables (estimate risk, relative risk, and expectations) were examined and a composite score was calculated. To accomplish this, scores were transformed into z-scores and mean of the three variables was derived as the composite. Additionally, reliability of the composite score was assessed ($\underline{\alpha}$.= 0.61; Table 4-7). For those whose response to an item was "unknown," when calculating reliability for the composite score, their score was replaced by the item

mean. When computing the individual's composite score in cases with an "unknown" response, the individual's score was the average of the other two risk items.

Anxiety

Anxiety was measured by a 10-item short form of the state-component of the State Trait Anxiety Inventory STAI (STAI; Spielberger, 1970). Respondents were asked to rate the questions according to how anxious they presently felt about their child's risk for developing type 1 diabetes on a four-point scale (i.e., Not at all, Somewhat, Moderately, or Very much). The 10-item STAI was also administered at all previous interviews and results were reported on in published studies (Johnson et al., submitted; Carmichael et al., 2003).

The 10-item short form was derived from a sample of 231 mothers who completed the full 20-item scale at the initial interview. Ten items were selected by examining the items that most highly correlated with the full 20-item scale scores for these participants. This form was found to be highly reliable at the initial ($\underline{\alpha} = 0.93$), four month follow-up ($\underline{\alpha} = 0.92$) and 12 month ($\underline{\alpha} = 0.90$) follow-up interviews. The 10-item short and 20-item full forms of the STAI were highly correlated ($\underline{r} = 0.97$). The practice of creating a short form of this measure is not unusual. The STAI-SF, a six-item Short Form, was developed and used in a prior study related to genetic screening (Marteau & Bekker, 1992).

A regression equation was developed which converts the short form scores into scores compatible with STAI norms to allow for comparisons with normative data provided in the STAI Manual. Data compiled by Carmichael et al. (2000) provides additional comparisons to similar samples including mothers learning of their child's

increased risk status as a result of ICA testing and pregnant women undergoing amniocentesis.

An additional question, adapted from Hendrieckx (2001) was asked to assess how often mothers worry about their children's risk. This question was stated as "how often do you worry about your child's risk for developing diabetes?" and rated on a 5-point Likert scale ranging from 0 to 4 anchored by "never" and "very often".

A composite score was derived by converting both scores into z-scores and calculating the mean z-score of the two items ($\underline{\alpha} = 0.80$) (Table 4-8).

Coping

The Ways of Coping Checklist-Revised (WCC-R) (Folkman & Lazarus, 1980) is a 69-item dichotic (yes/no) questionnaire used to assess the use of coping strategies and preferred coping style. In the PANDA Part III study, the WCC-R was administered at the 4 follow-up interviews to assess maternal coping regarding their infant's genetic risk of developing Type 1 diabetes (n = 178). This measure has also been used in similar risk screening studies (Johnson & Tercyak, 1995; Johnson & Carmichael, 2000). Factor scores were calculated using Vitaliano et al. (1985) factor structure, which uses 42 items. The five factors included the following coping styles/strategies: Problem-focused Coping, Seeking Social Support, Wishful Thinking, Self-Blame, and Avoidance. The WCC-R was not administrated in the current interview. However, subscale scores obtained at the second (4 month) interview were used as predictor variables. To be able to compare across factors having a varying number of items, mean scores were calculated for each subscale as well as for the total measure.

For this sample reliabilities for the subscales Wishful Thinking ($\alpha = 0.70$), Seeks Social Support ($\alpha = 0.73$) and Problem-Focused coping ($\alpha = 0.81$) were satisfactory. However, reliabilities for the Avoidance ($\alpha = 0.36$) and Self-Blame ($\alpha = 0.53$) subscales were poorer and did not meet criteria for further analyses ($\alpha < 0.60$) (Table 4-9). The reliability scores of the factors were consistent with previously published studies of similar populations (Johnson & Carmichael, 2000). Correlations between variables were significant, particularly between the total coping score and Problem Focused coping, Seeks Social Support, and Wishful Thinking (Table 4-9). Mean factor scores were similar to previous studies. Seeks social support was the most favored used coping style, followed by Problem-Focused coping. Self-blame was the least used coping style (Table 4-10).

Information seeking

A self-report measure of information-seeking was given to assess participants' sources of information regarding diabetes risk and/or behavior change. Questions were designed to assess if participants consulted with their physicians, family members of friends, including those who may have diabetes themselves. Follow-up questions were asked to determine if participants were given specific advice from these sources and if they followed the advice. Additional questions assessed behaviors such as searching the internet, consulting written materials about diabetes, or watching diabetes-related television news stories. This measure was scored as a continuous variable by calculating the number of information sources reported and as a dichotomous variable denoting whether any information seeking occurred (1 = 'Yes' and 0 = 'No') (Table 4-11).

Questions regarding the nature of the relationship and the content of the advice were used as descriptive data.

Participation in PANDA Part II

Data on Part II participation was available through the PANDA computerized database. Continuation in the PANDA Part II study was coded as two different variables, (1) number of blood draws and (2) at least one blood draw (1 = 'participated') versus no blood draws (0 = 'did not participate'). For more details, please refer to earlier section entitled "Reliability of self-reported behavioral change".

Statistical Analyses

Data analyses were conducted using SPSS 11.0. Internal reliability of predictor and outcome scores were calculated using Cronbach alpha and only constructs with alphas greater than 0.60 were retained for regression analyses. Additionally, components of risk perception and anxiety composite scores were transformed into z-scores because they were measured on different numerical scales. Consequently these composite scores reflect a z-transformation as well. Descriptive statistics were conducted, including ANOVA, t-tests and chi-square analyses, to compare demographic variables between participants and non-participants. When expected cell size was < 5, Fisher's Exact test statistic was used instead of chi-square. Hierarchical logistic regressions were used to predict to behavior change, as well as linear regression to predict to the continuous outcome measure of anxiety. For further details, refer to Chapter 5 (Results).

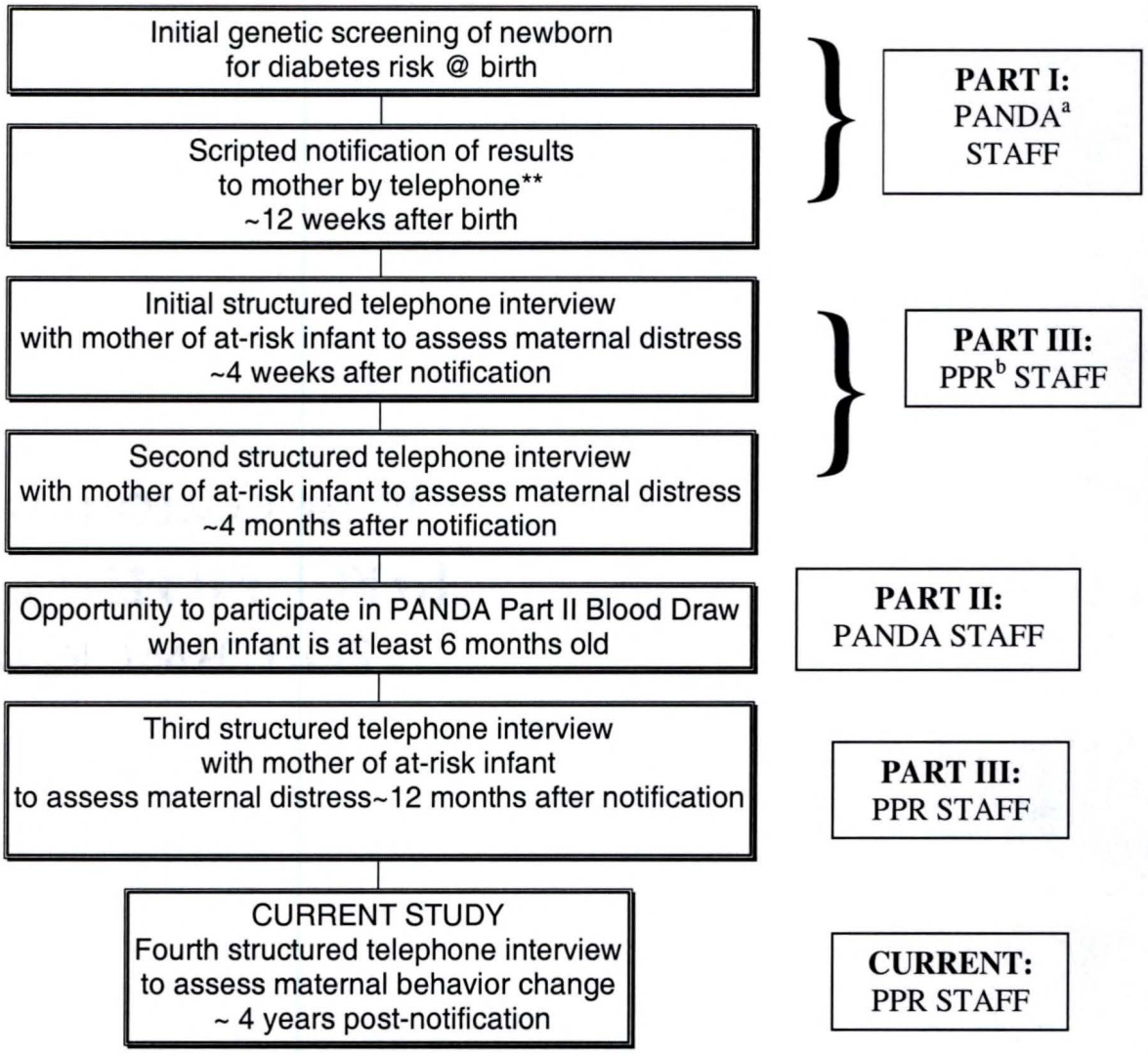

Figure 4-1. Procedural outline of PANDA study. PANDA = Prospective Assessment of Newborn Diabetes Autoimmunity. PPR = Pediatric Psychology Research Lab.

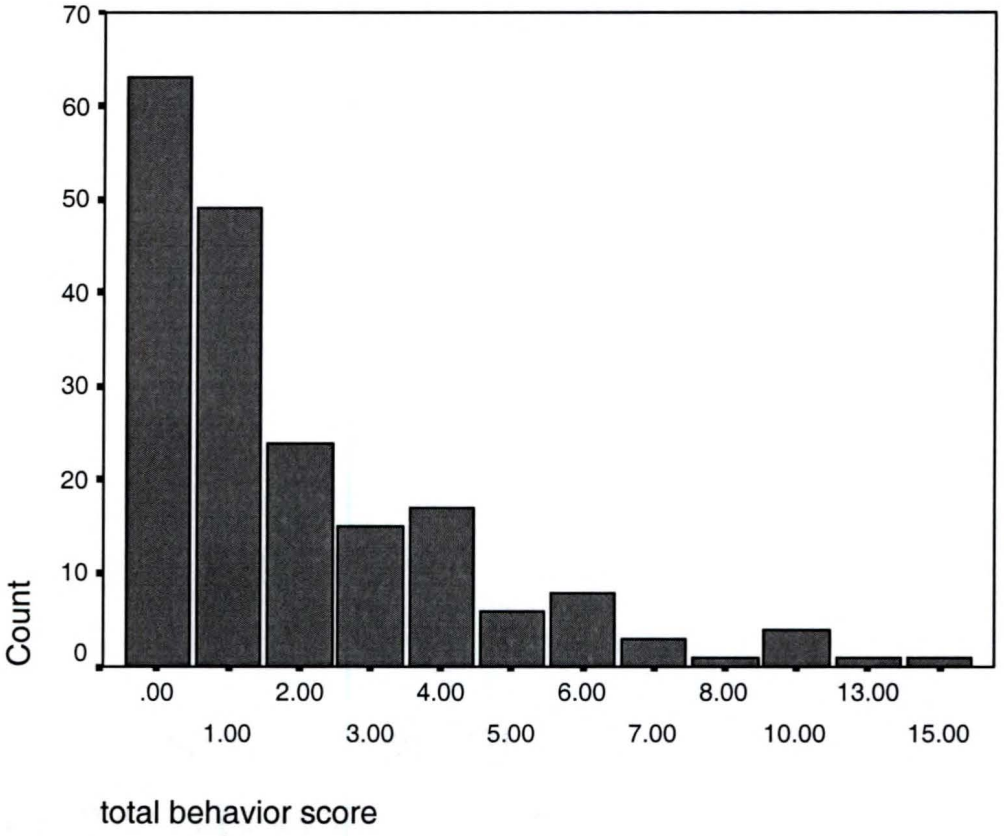

Figure 4-2. Frequency distribution of total behavior score (n = 192)

Table 4-1. Diabetes genetic risk factors

DR/DQ Alleles/genotypes	With first degree relative	Without first degree relative	% General population	% Type 1 patients
DR 3/4, DR 4/4 DQ 0201/0300 DQ 0300/0300	20-25/100 **Extremely high risk**	5/100 **High risk**	5%	>50%
DR 3/4, DR 4/X[a] DQ 0201/0201 DQ 0300/X[b]	10/100 **High risk**	2/100 **Moderate risk**	10%	30-40%
DR 3/4 or X/X DQ X/X	1/125 **Intermediate risk**	1/600 **Very low risk**	85%	10%
DR 3/4 or X/X DQ 0602	1/15,000 **Protective**	1/15,000 **Protective**		

X is a non-defined allele, 75% of the time X= DR 4 or DQ 0301. X allele is not 0602.

Table 4-2. Maternal characteristics of current sample (n = 192)

Variable	
Maternal age at notification	30.49 ± 5.36
Current maternal age	33.67 ± 5.38
Race	
Caucasian	162 (85%)
African American	6 (3%)
Hispanic	13 (6%)
Asian/other	10 (5%)
Mothers level of education	
High school or less	45 (23%)
Some college/trade school	62 (32%)
College degree or beyond	85 (44%)
Marital status (married)	164 (85%)
Annual income (in $10,000 intervals)	4.97 ± 2.50
Number of children	2.09 ± 1.11
Number of previous interviews	2.79 ± 0.51
1	9 (5%)
2	23 (12%)
3	160 (83%)
Number of blood draws (Part II)	1.19 ± 1.17
0	74 (39%)
1	47 (25%)
2	29 (15%)
3	41 (21%)

Note: Data are n (%) and means ± SD.

Table 4-3. Child characteristics of current sample (n = 192)

Variable	
Infant risk classification	
Moderate (2/100)	108 (56%)
High (1/10)	71 (37%)
Very high (1/5)	13 (7%)
Child age at notification (mo.)	7.85 ± 6.24
Current child age (years)	4.25 ± 0.89
Child sex (Male)	97 (51%)
Only child (Yes)	62 (33%)
Family history	
No family history	50 (26%)
Third degree relative	98 (51%)
Second degree relative	82 (43%)
First degree relative	37 (19%)

Note: Data are n (%) and means ± SD.

Table 4-4. Intercorrelations and coefficient alphas for domain scores of reported behaviors

Domain	1	2	3	4	5	6	7
1. Health surveillance	**0.37**						
2. Diet	0.44**	**0.58**					
3. Physical activity	0.27**	0.32**	**0.54**				
4. Illness prevention	0.35**	0.24**	0.36**	**0.72**			
5. Medications	0.28**	0.28**	0.12	0.36**	**0.54**		
5. Stress	0.16*	0.18*	0.10	0.28**	0.22**	**0.47**	
6. Total	0.74**	0.79**	0.58**	0.65**	0.47**	0.35**	**0.77**

Note: Coefficient alphas are presented in boldface along the diagonal. * $p < 0.05$ ** $p < 0.01$.

Table 4-5. Mean domain scores of reported behavior changes for total sample

Domain	# Items	Range	M	SD
Health surveillance	4	0-3	0.85	0.86
Diet	16	0-6	0.69	1.15
Physical activity	4	0-3	0.21	0.57
Illness prevention	8	0-5	0.18	0.69
Medications	5	0-3	0.04	0.27
Stress	4	0-2	0.04	0.24
Total	41	0-15	2.00	2.53
# of domains	6	1-6	1.22	1.20

Table 4-6. Mean values or frequencies for perceived control scales

Item	
I can do something[a]	2.95 ± 1.18
Strongly disagree	28 (15%)
Somewhat disagree	39 (20%)
Neutral	53 (28%)
Somewhat agree	58 (30%)
Strongly agree	14 (7%)
Doctors can do something[a]	2.63 ± 1.11
Strongly disagree	28 (15%)
Somewhat disagree	74 (39%)
Neutral	38 (20%)
Somewhat agree	47 (23%)
Strongly agree	8 (4%)
It is up to chance[a, b]	3.26 ± 1.11
Strongly disagree	15 (8%)
Somewhat disagree	37 (19%)
Neutral	39 (20%)
Somewhat agree	83 (24%)
Strongly agree	17 (9%)
Composite score (z-score)	2.79 ± 0.99

Note: Data are n (%) and means ± SD. [a] Scored as follows: Strongly Disagree = '1', Disagree = '2', Neutral = '3', Agree = '4', Strongly Agree = '5'. [b] Variable not used in composite score.

Table 4-7. Mean values or frequencies for perceived risk scales

Item	
Relative risk[a]	3.46 + 1.05
Much less	13 (7%)
Somewhat less	15 (8%)
About the same	61 (32%)
Somewhat higher	74 (39%)
Much higher	27 (14%)
Belief about when child may develop diabetes[b]	1.88 + 0.68
Never	53 (28%)
Unsure	113 (59%)
Yes, but not for a long time from now	22 (12%)
Yes, in the near future	4 (2%)
Risk estimation[c]	
Overestimate	12 (6%)
Accurate	76 (40%)
Underestimate	80 (42%)
Don't know/don't remember	24 (13%)
Risk composite (z-score)	-.004 ± 0.74

Note: Data are n (%) and means ± SD. [a] Scored as follows: Much less = '1', Somewhat less = '2', About the same= '3', Somewhat higher = '4', Much higher = '5'. [b] Scored as follows: Never = '0', Unsure = '2', Yes, but not for a long time from now = '3', Yes, in the near future = '4'. [c] Overestimate = '3', Accurate = '2', Underestimate = '1', Don't know/don't remember= '0' but value not included in analyses.

Table 4-8. Mean values or frequencies for anxiety/worry scale

Item	
Worry[a]	1.00 ± 1.02
Never	73 (38%)
Rarely	66 (34%)
Sometimes	39 (20%)
Often	8 (4%)
Always	6 (3%)
Anxiety (10-item STAI)[b]	30.79 ± 9.66
Anxiety composite (z-score)	0 ± 0.91

Note: Data are n (%) and means + SD. [a] Scored as follows: Never = '0', Rarely = '1', Sometimes = '2', Often = '3', Always = '4'. [b] Predicted full scale score based on 10 item measure.

Table 4-9. Intercorrelations and coefficient alphas for coping variables

Domain	1	2	3	4	5	6	7
1. Problem focused	**0.81**						
2. Seeks social support	0.64***	**0.73**					
3. Avoidance	0.15	-0.02	**0.36**				
4. Wishful thinking	0.53***	0.31***	0.37***	**0.70**			
5. Self-blame	0.22**	0.12	0.33***	0.28***	**0.53**		
6. Total score	0.84**	0.78***	0.37	0.74***	0.44***	**0.86**	

Note: Coefficient alphas are presented in boldface along the diagonal. ** p< .01. *** p < 0.001.

Table 4-10. Mean scores for Ways of Coping-Revised (WCC-R) scales

Construct[a]	# Items	
Problem focused	15	0.48 ± 0.24
Seeks social support	6	0.54 ± 0.31
Avoidance	6	0.13 ± 0.11
Wishful thinking	10	0.25 ± 0.23
Self-blame	3	0.03 ± 0.13
Total score	42	0.29 ± 0.14

Note: Data are means ± SD. [a] Scored as the mean of the items in each subscale. Each item in subscale is scored as '0' = No, '1' = Yes.

Table 4-11. Mean values or frequencies for information seeking scale

Item	
Any information source[a]	115 (60%)
Literature	63 (33)
Doctor	46 (24)
Family/friend	33 (17)
Television	27 (14)
Internet	22 (12)
# of information sources	0.99 ± 1.04

Note: Data are n (%) and means ± SD. [a]Scored as Yes= '1', No = '0'.

CHAPTER 5
RESULTS

Sample Characteristics

Compared to mothers who were eligible but did not complete the current interview ($\underline{n}$ = 176), participants in this current study ($\underline{n}$ = 192) were significantly more likely to be married ($\underline{p} < 0.001$) and older at time of notification ($\underline{p} < 0.01$) and the current interview ($\underline{p} < 0.001$) (for those who were not contacted, age was estimated based on end date of data collection 4/1/03) (Table 5-1). Additionally, they had higher levels of education ($\underline{p} < 0.01$) and annual family income ($\underline{p} < 0.001$). There were no differences between the two groups in terms of ethnicity. Overall, these results suggest the current sample was a highly select sample of mothers who were more economically stable and possessed more personal resources than mothers in the original larger sample. It is important to consider the sample bias in interpreting results of this study, as the behaviors of these mothers may not be reflective of the general population.

For mothers who participated in the current interview, 85% completed all three previous PANDA Part III interviews versus 57% of eligible non-participating mothers (χ^2 (1, $\underline{N}$ = 368) = 35.12 , $\underline{p} < 0.001$). Participating mothers had a higher number of completed interviews ($\underline{p} < 0.001$). Participation rates differed significantly for the longitudinal component of the PANDA study (Part II), which involves periodic autoantibody screening. Sixty one percent of current study participants and 49% of non-participants completed at least one autoantibody screening (χ^2 (1, $\underline{N}$ = 368) = 5.02 , $\underline{p} < 0.05$). Participating mothers had a higher number of blood draws ($\underline{p} < 0.001$). These

participation rates are less than rates reported in Finland, where approximately 80% of infants who were genetically screened joined their antibody surveillance study (Kupila et al., 2001).

As assessed in the initial interview, there was no significant difference in anxiety scores, as measured by the state STAI, between participating and non-participating mothers (Table 5-1). There was no significant difference between the two groups of mothers in their perceived likelihood that their child would develop diabetes in the future. However, at the time of the initial interview, mothers who participated in the current interview reported greater accuracy in estimating their child's risk status than mothers who did not participate ($p < 0.01$) and fewer mothers underestimated their child's risk ($p < 0.05$) (Table 5-1).

There were no differences between children of participants versus non-participants in regards to age, genetic risk status, sex, only child status, or family history (Table 5-2).

Objective 1

Hypothesis 1.1

At the outset of the study, it was hypothesized that reported behavior changes endorsed would most likely correspond to recommendations for the treatment of diabetes (ADA, 2001) and the prevention of type 2 diabetes (ADA, 2002a, 2002b; Pierce et al., 1995), including changes in diet and physical activity patterns.

The questionnaire's design permitted the use of both open and closed ended questions within each behavioral domain. Descriptive analyses of each behavioral construct, both as dichotomous and continuous variables, were conducted, including frequencies, means, standard deviations, and correlations. Qualitative data from other

open-ended questions addressing advice received and perceived control were also coded as descriptive data.

Open-Ended Questions

The initial open-ended item assessing behavioral change simply asked whether mothers did anything special to reduce their child's risk of developing type 1 diabetes (yes or no). Fifty-five mothers (29%) responded that they had done something preventative. Open-ended questions were also asked at the beginning of each of the six domains and again, at the end of the interview to assess maternal recall of behavior changes. At least one spontaneous behavior was reported in response to domain specific open ended questions by sixty nine mothers (39%), somewhat more than were identified through the initial broad question, yielding a total of 118 spontaneous responses (Table 5-3). Of these, 51 mothers indicated making a change in their child's diet and/or exercise (74%), corresponding with recommendations to prevent and/or treat type 2 diabetes (Table 5-4). One domain, medications, yielded no spontaneous responses and stress only yielded one response. Sixty three percent of the responses to the open-ended questions were later addressed in forced choice items asked subsequently in each domain.

To further examine the hypothesis that mothers were following recommendations for the prevention of type 2 diabetes, responses to open-ended questions regarding advice received and maternal beliefs were analyzed to determine if mothers' reported actions were based on a premise that a healthy lifestyle is an effective prevention method for type 1 diabetes. An open-ended question assessing maternal beliefs about what they could do to prevent their child from developing type 1 diabetes was asked of mothers who agreed or strongly agreed that they could do something to prevent their child from developing type 1 diabetes (Table 5-5). Seventy two mothers (38%) reported believing they could do

something preventative, with 108 responses generated. Of these, 61 mothers reported dietary and/or exercise changes (92%).

Additionally, 46 (24%) mothers reported receiving advice from a medical professional, generating 63 pieces of advice, and 33 (20%) mothers reported receiving advice from family or friends, generating 39 pieces of advice (Table 5-6). Ninety percent of mothers reported following advice from a medical professional and 95% reported following advice from family members/friends. Of the advice received from medical professionals, 43% suggested making healthier dietary and physical activity changes. Of the advice received from family members or friends, 28% of advice from family suggested healthy lifestyle changes in diet and exercise.

Forced Choice Questions

Forced choice items were asked with yes or no responses to assess maternal recognition of reported behavior changes. These items were used to assess specific behaviors and were expected to yield more positive responses than the use of open-ended questions.

Results based on the forced choice items within each domain, indicated that out of 192 mothers, 129 (67%) reported changing at least one behavior in an attempt to prevent diabetes from developing in their at risk child ($\underline{M}$ =2.00, $\underline{SD}$ = 2.53). Domain scores were calculated for each of the six possible categories of behavior determined a priori. Of those who reported at least one behavior change, 30% reported two to three changes, 24% reported four to six changes, and 8% reported changing more than six behaviors ($\underline{M}$ = 2.98, $\underline{SD}$ = 2.57) (Table 5-7). Changes in health surveillance behaviors were most frequently endorsed (59%), including blood glucose monitoring and watching for signs of diabetes development. Changes in child's diet (34%) were the next most commonly

reported, followed by changes in physical activity (14%), illness prevention (9%), medications (3%), and stress (3%) (Table 5-8). The item most frequently endorsed (≥ 10%) was checking for specific signs of type 1 diabetes (50.5%). An open-ended follow-up question asked mothers to specify the nature of the symptoms they look for in their children. Ninety seven mothers reported they look for signs of diabetes in their at risk child, each responding with approximately two signs each (M = 2.16). Seventy nine percent of mothers reported at least one correct diagnostic criterion type 1 diabetes (i.e., polyuria, polydipsia, weight loss, and increased appetite), 32% identified behaviors that were not indicative of diagnosis, but were related to diabetes symptomatology (i.e., signs of hyperglycemia or diabetic ketoacidosis), and 45% identified signs that were not related to diabetes (Table 5-9). Only 5% of mothers did not identify one correct or related symptom of type 1 diabetes. Of those who reported an accurate symptom, 38% also listed inaccurate symptoms. Additionally, testing the child's blood glucose level either at home or at a physician's office, feeding the child less soda, juice and other sweet foods, and encouraging the child to exercise more often were the next most commonly endorsed behavior changes. Items that might indicate maternal overprotectiveness or items suggesting unwarranted use of medications were rarely endorsed. Reported behavior changes ranged across domains for those endorsing more than one behavior change with only 19% reporting changes within only one domain ($\underline{M}$ = 2.33, $\underline{SD}$ = 1.02). This suggests that mothers engaged in a wide variety of behavior changes.

In comparing forced choice item responses with responses to open-ended questions, results indicated that significantly more mothers endorsed forced choice items rather than made spontaneous responses. This suggests that mothers may either have had difficulty

recalling behaviors that were not as salient, with forced choice items serving as a recognition task to help refresh their memory. Or, perhaps there may have been a demand characteristics associated with presenting individual specific behaviors in yes/no format. According to forced choice items, 67% of mothers reported making at least one behavior change versus 36% of mothers responding to open ended questions (Table 5-10). All mothers who spontaneously reported behavior changes also responded similarly to forced choice items, so there were no mothers who spontaneously reported behavior change who did not also report changes according to forced choice items. In comparing forced choice versus open-ended questions, the primary difference was that mothers were less likely to spontaneously report changes in health surveillance that were later identified through forced choice items. Mothers may not consider increased health surveillance as a way of actively preventing diabetes.

Reported behaviors specific to healthy lifestyle changes, within diet and exercise domains, consistent with recommendations for prevention and treatment of type 2 were coded and compared to address the hypothesis that behavior change would likely correspond with recommendations for prevention of type 2 diabetes. Overall, based on responses to open-ended questions, 51 mothers (27%) reported making at least one such behavior change (Table 5-4) and 59 mothers (31%) indicated a similar behavior change via responses to forced choice items (Table 5-8). Behaviors related to recommendations for prevention of type 2 diabetes were more prevalent among open-ended responses than among forced choice responses, in which health surveillance changes and overall dietary changes in general were more frequently reported.

Objective 2

Exploratory model testing was conducted through the use of logistic regression analyses predicting whether mothers reported behavior change. As stated previously, due to the non-normal distribution of reported behavior scores, the outcome measure of behavior was examined dichotomously, comparing mothers who reported at least one behavior change ($1 = \geq 1$ behavior change) versus mothers who reported none ($0 =$ no behavior change). Regressions were also conducted to predict whether a behavior change was spontaneously reported in response to open ended questions; however, results were nearly identical to using the forced choice items and therefore, quantitative analyses based on responses to open ended questions were reported.

In each regression model, predictor variables were entered in blocks according to hypothesized relationships from prior literature. Each block of variables was added successively. When each block was added to the model, only variables that were significant at $p< 0.10$ were retained. For these analyses, several variables were recoded for ease of interpretation. Due to the sample's unbalanced distribution by maternal race, minority ethnic groups were collapsed into one group and maternal ethnicity was categorized as "Caucasian" (1) and "not Caucasian" (0). Maternal marital status was coded as "1" for married and "0" for single, separated, widowed or divorced. Child's sex was coded as 1 "male" and 2 "female". Only child status coded as 1 "yes" and 0 "no".

The first block of variables entered into the regression model contained one variable, time elapsed between notification and current interview, to control for effects of time. The second block of variables contained maternal demographic variables, including maternal education level, ethnicity, marital status, number of children, and age at the time of the interview. The third block entered contained child demographic

variables, including child's sex, whether an only child, and age at the time of interview. Family history of diabetes was also included in this block, using two dichotomous variables: (1) the presence of a first-degree relative with diabetes (yes/no) or (2) the presence of a second or higher degreed relative (yes/no).

The fourth block of variables contained the hypothesized predictor variable. Predictor variables consisted of standardized composite scores on measures of perceived control, risk perception, and anxiety, as well as total scores on measures of coping and information seeking. Participation in PANDA Part II study was also used as dichotomous predictor variable (yes/no). Reliability analyses were conducted on composite scores suggesting that internal consistency was fair for these variables (Table 4-6). Each of the following hypotheses was examined separately. Within each model, main effects were examined as well as interactions between perceived control and other predictors, where noted. Only significant predictors were retained. Ultimately, a final model was produced from these separate models to account for the highest classification rate in the behavioral outcome variable. To account for type 1 error, a more conservative level of significance was chosen at $p < 0.01$ and this is noted where appropriate.

Hypothesis 2.1

It was hypothesized that mothers who perceived they have control over their child developing diabetes would be more likely to report engaging in behavior changes. Based on statements regarding perceived control, which required an agree/disagree response, 38% percent of mothers reported believing they could do something and 27% believed doctors could do something to prevent their child from developing type 1 diabetes. Meanwhile, 52% reported believing it was up to chance or fate whether their child develops type 1 diabetes (Table 4-6).

Hierarchical logistic regression analyses were conducted, using the composite score for perceived control (belief that mother could do something or medical professional could do something) to predict whether any behavior change was reported (yes/no) when controlling for demographic factors (Table 5-11). Results indicated that mothers whose children had a first degree relative with diabetes were significantly more likely to engage in behavior change (odds ratio = 24.22, $p < 0.001$) and maternal perceived control was not a significant predictor of behavior change, resulting in an overall model that accounted for 67.5% overall correct classification.

Hypothesis 2.2

It was hypothesized that mothers who perceived their children to be increased risk for type 1 diabetes would be more likely to report engaging in behavior change. Hierarchical logistic regression was conducted as described previously; however, both actual risk and the risk composite score were entered as the final block in the logistic regression model. Results indicated that again, the presence of a first degree relative was a significant predictor of behavior change (odds ratio = 18.98, $p < 0.01$). The child's actual risk was found not to be significant. When controlling for actual risk, perceived risk was a significant predictor (2.32, $p< 0.01$) (Table 5-12). Mothers who perceived their children to be at greater risk were more likely to engage in behavior change. This model resulted in an overall classification rate of 68.4%. An interaction between perceived control and perceived risk was also tested and was not significant.

Hypothesis 2.3

It was hypothesized that mothers who were more anxious would be more likely to report engaging in behavior change. This hypothesis was tested by entering the anxiety composite score as the final block in the logistic regression model. Results indicated that

anxiety, as measured at the initial interview following notification, was not a significant predictor of subsequent behavior change and was not retained in the final model. However, mothers who were more anxious at the time of the current interview were more likely to report behavior change (odds ratio = 2.98, $p < 0.001$). This model resulted in a correct classification rate of 72.9% (Table 5-13). Follow-up analyses were conducted to determine if there was an interaction between anxiety and perceived control; however, none was found. Results demonstrated that while anxiety remained a significant predictor, there was no main effect of perceived control, nor was the interaction term significant, suggesting that mothers who were more anxious were more likely to report behavior change to prevent diabetes in their child regardless of their level of perceived control over the situation.

Hypothesis 2.4

It was hypothesized that mothers who used more coping strategies, particularly active coping (i.e., problem-focused, seeking social support), would be more likely to report engaging in behavioral change. Data were available for 176 mothers who completed the Ways of Coping Checklist- Revised (WCC-R) at the 4-month interview. In separate logistic regression models, each coping scale score was entered as the final block of variables. Results indicated that after controlling for the significant effect of the presence of a first degree relative, problem-focused coping (odds ratio = 10.72, $p < 0.01$), seeking social support (odds ratio = 4.99, $p < 0.01$), and wishful thinking (odds ratio = 14.48, $p < 0.01$) were significant predictors of behavior change (Tables 5-14, 5-15, and 5-16). While the two active coping factors were significantly related to behavior change, a more passive coping style, wishful thinking, was also significant and to a relatively

higher degree. Item analysis of the wishful thinking scale indicated that this scale included items related to optimistic thinking but also a desire for the problem to "go away" or "be over with." It may be that wishful thinking reflects a sense of optimism and urgency that might be associated with engaging in preventative actions believed by mothers to be efficacious. Additionally, total coping as measured by the mean of all reported coping behaviors was a significant predictor of reported behavior change (odds ratio = 160.06, $p < 0.001$) (Table 5-17).

Hypothesis 2.5

It was hypothesized that mothers who engaged in information seeking and/or were given recommendations by medical professionals or other family members related to behavior change, would be more likely to report engaging in behavior change. Overall, 60% reported receiving information from at least one source, and the mean number of sources was 0.99 (SD = 1.04). Overall, 33% reported receiving diabetes-specific information from a book or other literature, 14% reported watching diabetes-related television programming, and 12% reported seeking information using the internet (Table 4-11). As stated previously,

Fifty nine percent of mothers reported talking to their physician about their child's genetic risk screening results. Of those, 41% reported receiving advice from their physician, with over 89% reportedly taking their physician's advice. When specifically asked in an open-ended question about the nature of the guidance given, mothers specified a wide range of advice (Table 5-6). The most frequent advice given was to monitor their child and promote a healthy lifestyle. Additionally, six mothers were told to continue with PANDA study and five mothers were told by their physicians not to worry about their child's risk.

Advice from family friends was similarly assessed. Eighty-six percent reported talking to a family member or friend. Seventy percent reported talking with their spouse about their child's genetic screening results, 63% reported talking with the child's grandparent, 13% reported taking to a family member of friend who has diabetes and 32% reported talking with a family member or friend who does not have diabetes. Seventeen percent of mothers reported receiving advice from at least one family member/friend. Typically, advice was given by a child's grandparent (62%), followed by spouse (15%) and friend or family member who does not have diabetes (15%), then friend or family member who does have diabetes (5%). When specifically asked in open-ended questions about the advice that was given, most frequent advice was to help child maintain a healthy diet and five mothers were told not to worry. (Table 5-7)

Logistic regression was used to determine if the number of information sources predicted the likelihood of engaging in behavior change. The number of information sources was entered as the last block of predictor variables in a logistic regression model (Table 5-18). When controlling for the presence of a first degree relative (odds ratio = 26.31, $p < 0.01$), those with more sources of diabetes-specific information were significantly more likely to report engaging in behavior change (odds ratio = 2.27, $p < 0.001$). The presence of a first degree relative combined with the degree of information sources together resulted in an overall classification rate of 74.3%.

Follow-up logistic regression analyses were conducted to determine if an interaction was present between perceived control and the number of information sources; none was found.

Hypothesis 2.6

It was hypothesized that mothers who continued their participation in the Prospective Assessment of Newborn Diabetes Autoimmunity (PANDA) study by participating in periodic blood testing for antibodies, would be more likely to report behavior changes. Overall, 61% of mothers participated in at least one subsequent blood draw. Twenty-six percent participated in two or more. However, when asked in the interview, 174 mothers reported accurately whether they participated in part II of the study (91%), and 3 reported participating when they actually have not (1%), and 15 reporting they had not participated when they actually had (8%).

Surprisingly, in the logistic regression model, registry participation using either number of blood draws or continued participation (yes/no) did not predict to reported behavior change (Table 5-19a, b). Mothers who continued with the PANDA study were no more likely to report engaging in preventative efforts, despite their already active participation in health surveillance.

Summary Model

Logistic regression was conducted to determine which of the previously listed variables were most predictive of behavior change. As in previous analyses, family history, characterized by presence of a first-degree relative, was entered as the first block of variables as it had been found to be consistently significant in all previous models. Actual risk was entered next in the model followed by all six variables also found to be significant at the 0.01 level in previous models (i.e., perceived risk, anxiety, number of information sources, problem focused coping, seeking social support, and wishful thinking) were subsequently entered simultaneously. Problem focused coping and

seeking social support were dropped from the resulting model, as they were not significant.

The final logistic regression model showed the presence of a first degree relative was once again a significant predictor of behavior change (odds ratio = 19.34, $p = 0.01$). Number of information sources, anxiety, perceived risk, and wishful thinking were also significant predictors (Table 5-20). Overall, the model's classification rate was 77.7%.

Objective 3

It was hypothesized that mothers who reported modifying behaviors would show a greater reduction in anxiety over time than mothers who did not report engaging in behavior change. To examine this hypothesis, hierarchical linear regression was conducted similarly to logistic regression procedures described for Objective 2, except the dependent variable was the anxiety composite score, a continuous variable. Anxiety at the initial interview was entered as the first block of variables followed by same ordering of blocks of variables of demographic variables described previously. Reported behavior change and the composite score for perceived control were entered as the final (fourth) block of predictor variables to determine if behavior contributed significantly to anxiety at the final follow-up interview, above and beyond the effect of initial anxiety and demographic predictors. In a follow-up model, the interaction term between behavior change and perceived control was added as the fifth block of predictors.

Results indicated that initial anxiety was a significant predictor of anxiety at the current interview ($\beta = 0.42$, $p < 0.001$) accounting for 22% of the variance (Table 5-21). Current age of the child ($\beta = -0.18$, $p < 0.01$) along with the presence of a first degree relative ($\beta = 0.29$, $p < 0.001$) and the presence of a second or higher degree relative ($\beta = 0.18$, $p < 0.01$) together accounted for an additional 13% of the variance ($p < 0.001$.

Reported behavior change ($\beta = 0.24$, $p < 0.001$) and perceived control ($\beta = 0.20$, $p < 0.01$) were entered as the final block and both were found to be significant predictors, accounting for an additional 9% of variance ($p < 0.001$). Overall, the model accounted for 43% of the total variance. In the follow-up model, the interaction term was added and not found to be significant. Results indicated that initial anxiety was the primary predictor of anxiety at the time of the current interview. However, above and beyond initial anxiety, mothers whose children were younger and had a relative with diabetes were more anxious at the time of the current interview. Mothers who reported at least one behavior change were significantly more anxious at both post-notification and current interviews (as measured by the STAI only) than mothers who reported no behavior changes (initial interview: M = 42.75, SD = 14.54 versus M = 36.80, SD = 12.54, $t(1, 189) = 2.76$, $p < 0.01$) (current interview M = 32.57, SD = 10.28 versus M = 27.14, SD = 7.02, $t(1, 190) = 4.29$, $p < 0.001$). Contrary to the original hypothesis, when controlling for demographics and initial anxiety, mothers who reported at least one behavior change and who perceived greater control over the onset of diabetes in their children were more anxious at the time of current interview than mothers who did not. This suggests behavior change maintains, rather than reduces anxiety over time for these mothers.

Objective 4

Questions regarding behavior change used in the current interview were developed from the DPT-1 survey (Johnson, 2002) and therefore, dichotomous scoring for some of the questions in the current interview were comparable. The database from the current study was merged with the maternal report data from the DPT-I study. Only data collected from mothers who were not aware of the study results were included ($n =$

116). Of these mothers, 63 (53%) had children who participated in the control group of the study and 53 (47%) had children enrolled in the experimental arm. Children whose mothers completed the DPT-1 survey were significantly older than children in the PANDA sample, ranging in age from 5 to 19 years old ($\underline{M} = 12.18$, $\underline{SD} = 3.24$) ($\underline{t(2, 306)} = 32.02$, $\underline{p} < 0.001$). Reported maternal behavioral data from these two populations were compared on 17 overlapping variables. Analyses were conducted across the corresponding individual questions and similar domain scores.

We hypothesized mothers of genetically at risk children would be less likely to report behavior change than mothers of ICA+ children enrolled in Diabetes Prevention Trial-1 (DPT-1) ($\underline{n} = 116$). On questionnaire items that were shared by both studies, 43.2% of mothers whose children were in the DPT-1 study and 33.3% of mothers in the current sample reported at least one behavior change (Table 5-22). However, this difference was not significant ($\underline{p} = 0.08$). Mothers in the two samples reported similar proportions of behavior change in the domains of diet and exercise; however, medications differed by groups with mother from the DPT-1 sample reporting greater use of medications/supplements.

There were few significant differences between the two samples of mothers on specific items. Mothers in the DPT-1 sample were nearly four times more likely to report feeding their children more diet and sugar free drinks ($\underline{p} < 0.001$), and more often reported feeding their children less regular soda ($\underline{p} < 0.05$), whereas, mothers in the current sample more often reported feeding their children less juice ($\underline{p} < 0.05$). Administering vitamins ($\underline{p} < 0.05$) and administering insulin at home ($\underline{p} < 0.05$) were practices that were significantly more common in the DPT-1 sample. This is not surprising given that 53 of

the mothers had children who were in the experimental arm of the study involving home insulin injections (46%) and the question involved giving "extra" insulin above and beyond study protocol. Out of the 5 mothers who reported giving their child insulin, 4 (80%) were mothers of children enrolled in the experimental group.

Table 5-1. Comparisons of maternal demographic variables between participants in current sample versus those eligible who were unable to be contacted or declined participation (N= 368)

	Completers (n = 192)	Unable to contact/declined (n = 176)	Total (n = 368)	F (1, 434) or χ^2
Maternal age at notification	33.49 + 5.36	27.71+5.35	29.28+5.63	14.42***
Current maternal age[a]	33.67 + 5.38	31.21 + 5.33	32.41+5.44	12.18**
Race				7.32
Caucasian	162 (84.9)	135 (76.7)	297 (80.9)	
African American	6 (3.1)	17 (9.7)	23 (6.3)	
Hispanic	13 (6.8)	17 (9.7)	30 (8.2)	
Asian/Other	10 (5.2)	7 (4.0)	17 (4.6)	
Mothers level of education				11.72**
High school or less	45 (23.7)	50 (28.2)	95 (25.9)	
Some college/trade school	62 (32.1)	74 (41.8)	137 (36.8)	
College degree or beyond	85 (44.2)	53 (29.9)	138 (37.3)	
Marital Status (married)	164 (85.4)	112 (64.4)	276 (75.5)	18.71***
Annual income (in $10,000 intervals)	4.97+2.50	3.82 + 5.30	4.43 + 2.40	20.22***
Number of Children	2.09 + 1.11	2.03 + 1.26	2.06 + 1.18	
Number of previous interviews				39.83***
1	9 (4.7)	36 (20.3)	45 (12.0)	
2	23 (12.0)	41 (23.2)	64 (17.2)	
3	160 (83.3)	100 (56.5)	260 (70.8)	
Number of blood draws (II)				44.03***
0	74 (39.1)	90 (50.8)	164 (44.7)	
1	47 (24.5)	59 (33.3)	106 (28.6)	
2	29 (15.1)	20 (11.3)	49 (13.4)	
3	41 (21.4)	8 (4.5)	49 (13.4)	
Anxiety[b]	40.74+14.13	39.83 + 14.29	40.30 + 14.19	0.70
Belief about when child may develop diabetes[b]				1.90
Never	43 (22.4)	34 (19.3)	77 (20.8)	

Table 5-1 Continued

	Completers (n = 192)	Unable to contact/declined (n = 176)	Total (n = 368)	F (1, 434) or χ^2
Unsure	135 (70.3)	126 (71.6)	261 (71.0)	
In distant future	13 (6.8)	12 (6.8)	25 (6.8)	
Soon	1 (0.5)	4 (2.3)	5 (1.4)	
Risk estimation[b]				
Overestimate	7 (3.6)	7 (4.0)	14 (3.8)	
Accurate	155 (81.3)	117 (65.5)	272 (73.6)	11.12**
Underestimate	20 (10.4)	28 (15.8)	48 (13.1)	3.64*
Don't know/don't remember	9 (4.7)	26 (14.7)	35 (9.5)	

Note: Data are n (%) and means + SD. Comparisons tested using chi-square or t-tests between completers and non-completers. Non-significant p values not reported.
[a]Estimated from 4/1/03 (end date of data collection). [b] Scores from initial interview (3-5 weeks post-risk notification). * $p < 0.05$. ** $p < 0.01$. *** $p < 0.001$.

Table 5-2. Comparisons of child demographic variables between participants in current sample versus those eligible who were unable to be contacted or declined participation (N= 368)

	Completers (n = 192)	Unable to contact/ declined (n = 176)	Total (n = 368)	F (1, 434) or χ^2
Infant risk classification				2.75
Moderate (2/100)	108 (56.3)	113 (63.8)	221 (60.2)	
High (1/10)	71 (37.0)	56 (31.6)	125 (34.1)	
Very high (1/5)	13 (6.8)	8 (4.5)	21 (5.7)	
Child age at notification (mo.)	7.85 + 6.24	7.29 + 4.34	7.59 + 5.43	3.35
Current child age (years)[a]	4.25 + 0.89	4.45 + 0.72[a]	4.34 + 0.82	0.17
Child sex (Male)	97 (50.5)	88 (50.9)	185 (50.4)	0.25
Only child (Yes)	62 (32.6)	72 (41.4)	134 (36.5)	3.05
Family history				5.71
No family history	50 (26.0)	37 (20.9)	87 (23.7)	
Third degree relative	98 (51.0)	104 (58.8)	202 (55.0)	
Second-degree relative	82 (42.7)	74 (41.8)	156 (42.5)	
First-degree relative	37 (19.3)	23 (13.0)	60 (16.3)	

Note: Data are n (%) and means + SD. Comparisons tested using chi-square or t-tests between completers and non-completers. Non-significant p values not reported.
[a]Estimated from 4/1/03 (end date of data collection). $^{*}p < 0.05$. $^{**}p < .01$. $^{***}p < 0.001$.

Table 5-3. Mothers' responses to open-ended questions in each behavioral domain regarding behavioral change

Responses	# of responses	Addressed in questionnaire
Diet/eating patterns		
Healthier diet total	53	
Less sugar	20	•
Ate healthy foods	12	
Decreased carbohydrates	5	
Monitored eating	3	
Ate more vegetables	3	
Limited juice	3	•
Limited fast food/junk food	2	
Ate more protein	1	
Drank more water	1	
Ate smaller portions	1	•
Taught good nutrition	1	
Ate a balanced diet	1	
Ate a low-fat diet	1	
Delayed milk	1	•
Varied diet	1	
Physical activity		
Increased exercise	14	
Encouraged activity	3	•
Kept active	3	•
Played outside in yard	2	
Daily exercise	1	•
Dance class	1	•
Exercised as family	1	•
Joined gymnastics team	1	•
More walking	1	•
More exercise	1	•
Health surveillance		
Checked blood glucose	15	•
Blood draw with PANDA	8	•
Looked for symptoms	7	•
Watched weight	2	•
Blood draws	2	
Tested for ketones	1	
Took to specialist	1	

Table 5-3 Continued

Responses	# of responses	Addressed in questionnaire
Checked at doctor's office	1	•
Annual exams	1	•
Illness prevention		
Protect child from the cold	2	
Avoiding things allergic to	1	
Keep away if someone is sick	1	•
Monitor allergies	1	
Prevent ear infection	1	
No daycare	1	•
Stress		
Decreased child's stress	1	•
Extra (not included in domains)		
Prayer	2	

Note: No responses for the medications domain of behavior change.

Table 5-4. Mothers' responses to open-ended questions in each behavioral domain

Responses	N	% of open ended responses (n= 69)	% of total (n = 192)
Type 2 diabetes recommendations	51	74%	27%
Diet/eating patterns	49	71%	26%
Health surveillance	30	43%	16%
Physical activity	13	19%	7%
Illness Prevention	8	12%	4%
Prayer (extra)	2	3%	1%
Stress	1	1%	0.5%

Note: No responses for the medications domain of behavior change.

Table 5-5. Mothers' statements on what they believe they or health care professionals can do to prevent type 1 diabetes in their children

Question	n
What I believe I can do (n = 72)	
Balanced diet/Healthy eating	42
Exercise/activity	25
Limit sweets/sugar/carbohydrates	10
Look for symptoms	6
Don't know	6
Teach healthy lifestyle	4
Keep healthy	2
Prayer	2
Positive attitude/mental empowerment	3
Breastfed	1
PANDA study/Research	2
Set example/model behavior	1
Learn more	1
Insulin early	1
Give child omega 3 fatty acid	1
Lower stress	1
What I believe doctors can do (n = 51)	
Research	13
Don't know	11
Educate parents	10
Monitor child's health	4
Give medications	5
PANDA study	5
New technology	2
Early testing	2
Prevention	2
Develop new medications	1
Provide support	1
Help in treatment	1
Public awareness	1
"Hoping they can"	1
"Nothing now, maybe later on"	1

Table 5-6. Advice received by mothers regarding their child's risk screening results

	n
Medical professional (n = 46)	
Monitor	17
Healthy diet/eating habits	11
Exercise/activity	7
Continue with PANDA study	6
Don't worry	5
Limit sugar	5
Check blood glucose	4
Avoid milk	1
Changed vaccine schedule	1
Lower risk of illness	1
Flu short	1
Be careful for insurance purposes	1
Recheck it later	1
Nothing you can do	1
Watch weight	1
Family member or friend (n = 33)	
Healthy diet/eating habits	10
Don't worry	8
Monitor symptoms	5
Pray	4
Continue with PANDA study	3
Continue to check	2
Talk to doctor	2
Get more information	2
Watch child's weight	2
Exercise	1

Table 5-7. Mean domain scores of reported behavior changes for those who made at least one behavior change

Domain	# Items	Range	M	SD
Health surveillance	4	0-3	1.26	0.77
Diet	16	0-6	1.02	1.28
Physical activity	4	0-3	0.31	067
Illness prevention	8	0-5	0.27	0.83
Medications	5	0-3	0.06	0.32
Stress	4	0-2	0.05	0.29
Total	41	1-15	2.98	2.57
# of domains	6	1-6	2.33	1.02

Table 5-8. Prevalence of reported behavior changes according to forced-choice items

Item	Yes n (%)	Consistent[a]	Always/nearly everyday[b]
Health surveillance	**114 (59)**		
Watched for signs	97 (51)	87 (90)	46 (48)
Tested child's blood glucose level at doctor's office	36 (19)	19 (53)	n/a[c]
Tested child's blood glucose level at home	27 (14)	21 (78)	2 (7)
Attended more frequent pediatrician visits	3 (2)	2 (67)	n/a
Diet/eating behaviors	**65 (34)**		
Fed child less sweet foods	38 (20)	34 (90)	28 (73)
Fed child less soda	30 (16)	29 (95)	23 (77)
Fed child less juice	23 (12)	22 (96)	20 (87)
Fed more diet and sugar free drinks	12 (6)	9 (75)	8 (67)
Increased duration of breast feeding	8 (4)	n/a	n/a
Fed child more often	5 (3)	5 (100)	5 (100)
Delayed introduction of cow's milk	5 (3)	n/a	n/a
Tried to get child to lose weight	4 (2)	3 (75)	2 (50)
Fed child less to eat	3 (2)	2 (67)	3 (100)
Fed child more juice	2 (1)	2 (100)	2 (100)
Fed more to eat	1 (1)	1 (100)	1 (100)
Fed child less often	1 (1)	1 (100)	1 (100)
Tried to get child to gain weight	0 (0)	--[c]	--
Changed timing of introduction to solid foods	0 (0)	--	--
Avoided cow's milk altogether	0 (0)	--	--
Physical activity	**26 (14)**		
Child exercise more often	19 (10)	19 (100)	15 (79)
Encouraged child to be active everyday	17 (9)	15 (88)	13 (81)
Encouraged child to rest more during exercise	3 (2)	2 (67)	2 (67)
Encouraged child to exercise less often	1 (1)	0 (0)	1 (100)
Illness prevention	**18 (9)**		
Worked harder to protect child from germs	15 (8)	14 (93)	12 (80)
Limited child's exposure to other kids	8 (4)	7 (88)	4 (50)
Kept child out of daycare	5 (3)	4 (80)	5 (100)
Had child wash hands more often	2 (1)	2 (100)	2 (100)
Avoided child exposure to chemicals (i.e., pollution, food additives)	2 (1)	2 (100)	2 (100)
Delayed immunizations for child	2 (1)	n/a	n/a
Avoided child's exposure to smoke	1 (1)	1 (100)	1 (100)
Increased child's exposure to other children to boost immunity	0 (0)	--	--
Medications	**6 (3)**		
Administered vitamins to child	6 (3)	5 (83)	5 (83)
Administered diabetes medications to child	1 (1)	0 (0)	0 (0)

Table 5-8 Continued

Item	Yes n (%)	Consistent	Always/nearly everyday
Administered insulin to child at home	1 (1)	0 (0)	0(0)
Administered nicotinamide	0 (0)	--	--
Used herbal supplements	0 (0)	--	--
Stress	**5 (3)**		
Had child rest more often	3 (2)	2 (67)	2 (67)
Actively lowered child's stress level	2 (1)	2 (100)	2 (100)
Actively distracted child's focus during stressful situations	1 (1)	1 (100)	0 (0)
Avoided distressing situations for child	1 (1)	1 (100)	0(0)

Note: Data reported in n (%). [a] Frequency of those who reported engaging in specific behavior since risk notification. [b] Freequency of those who reported engaging in specific behavior for "always/nearly everyday." c Not applicable· [d] Not calculated (n = 0).

Table 5-9. Signs used by mothers to monitor diabetes symptoms in their children (n = 97)

Response	# of responses	Accurate symptom of type 1 diabetes	Related symptom of type 1 diabetes
Thirst/Drinking more	59	•	
Urination	47	•	
Tired/fatigue	21		•
Weight loss	12	•	
Weight gain	6		
Changes in child's behavior (general)	6		
Irritability	6		
Eating more	6	•	
Hyperactivity	6		
Craving sweets	5		
Headaches	4		•
Bedwetting	4		•
Sweating	3		•
Shakiness	3		•
Dizziness	2		•
Blurred vision	2		•
Seizures	1		•
"List from doctor/don't remember"	1		
Blood pressure	1		
Dry mouth	1		
Not eating	1		
Appetite change	1		•
Vomiting	1		•
Wounds not healing	1		
Skin rash	1		
Fever	1		
Cold symptoms	1		
Shortness of breath	1		•
General sickness	1		
Poor circulation	1		

Table 5-10. Prevalence of reported behavior changes as reported in responses to forced choice versus open-ended questions for those who reported at least one behavior change

Responses	Forced Choice (n = 129)	Open ended (n = 69)
Health surveillance	114 (88)	30 (43)
Diet/eating patterns	65 (50)	49 (71)
Type 2 recommendations	59 (46)	51 (74)
Physical activity	26 (20)	13 (19)
Illness Prevention	18 (14)	8 (12)
Prayer[a]	n/a	2 (3)
Medications	6 (5)	0 (0)
Stress	5 (4)	1 (1)

Note: Data reported as n (%). [a]Spontaneous response - item is not included in domains of forced choice items.

Table 5-11. Logistic regression demonstrating relationship between perceived control and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative	3.19	1.03	24.22	9.60**
Control composite	0.08	0.17	1.09	0.24

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as 0 = No and 1 = Yes. ** $p < 0.01$.

Table 5-12. Logistic regression demonstrating relationship between perceived risk and reported behavior change[a]

Predictor variable	B	SE	Odds ratio	Wald statistic
First degree relative[b]	2.94	1.07	18.98	7.53**
Actual risk[c]	-0.35	0.35	0.70	1.06
Risk composite	0.84	0.27	2.32	9.59**

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as 0 = No and 1 = Yes. [c]Coded as 4= moderate, 5 = high, 6 = extremely high. ** $p < 0.01$.

Table 5-13. Logistic regression demonstrating relationship between anxiety and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	2.70	1.04	14.92	6.72**
Current anxiety composite	1.09	0.28	2.98	15.54**

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. ** $p < 0.01$. *** $p < 0.001$.

Table 5-14. Logistic regression demonstrating relationship between problem focused coping and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	3.19	1.03	21.91	8.92**
Problem focused	2.34	0.76	10.41	9.55**

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. ** $p < 0.01$.

Table 5-15. Logistic regression demonstrating relationship between seeking social support and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	3.01	1.03	20.26	8.50**
Seeks social support	1.61	0.56	4.99	8.28**

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. ** $p < 0.01$.

Table 5-16. Logistic regression demonstrating relationship between wishful thinking and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	2.96	1.03	19.37	8.23**
Wishful thinking	2.67	0.86	14.48	9.76**

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. ** $p < 0.01$.

Table 5-17. Logistic regression demonstrating relationship between total coping score and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	3.00	1.03	20.04	8.40**
Total coping score	5.08	1.38	160.06	13.56***

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. ** $p < 0.01$. *** $p < 0.001$.

Table 5-18. Logistic regression demonstrating relationship between number of information sources and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	3.27	1.04	26.21	9.96**
# of information sources	0.82	0.21	2.27	16.00***

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. ** $p < 0.01$. *** $p < 0.001$.

Table 5-19a. Logistic regression demonstrating relationship between continued participation in PANDA study (yes/no) and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	3.22	1.03	24.95	9.71*
Attended at least one blood draw	-0.12	0.33	1.89	0.14

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. * $p < 0.05$.

Table 5-19b. Logistic regression demonstrating relationship between continued participation in PANDA study (# of blood draws) and reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
First degree relative[b]	3.09	1.04	22.01	9.79*
# of blood draws	0.07	0.16	1.07	0.21

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. * $p < 0.05$.

Table 5-20. Summary logistic regression model predicting to reported behavior change[a]

Predictor variable	β	SE	Odds ratio	Wald statistic
Step1				
First degree relative[b]	2.96	1.15	19.34	6.66**
Step 2				
Actual risk	-0.65	0.44	0.52	2.16
Step 3				
Risk composite	0.73	0.34	2.08	4.67*
Anxiety composite	0.73	0.32	2.07	5.14*
# of information sources	0.69	0.24	1.99	8.02**
Wishful thinking	2.47	1.01	11.79	5.91*

Note: [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. [b]Coded as Yes = '1' and No = '0'. * $p < 0.05$. ** $p < 0.01$.

Table 5-21. Hierarchical linear regression model predicting to anxiety composite score

Predictor variable	B	SE	β	R^2	ΔR^2
Step1				0.22^{***}	
Anxiety at initial interview	2.70e-02	4.00e-03	0.42^{***}		
Step2				0.34^{***}	0.13^{***}
Child age (current)	-0.19	0.06	-0.18^{**}		
First degree relative[a]	0.68	0.14	0.29^{***}		
Second or higher degree relative[a]	0.34	0.11	0.18^{**}		
Step 3				0.43^{***}	0.09^{***}
Behavior change[b]	0.46	0.12	0.24^{***}		
Control composite	0.18	0.05	0.20^{***}		

Note: [a] Coded as 0= no and 1 = yes. [b] Coded as At least one reported behavior change = '1', No reported behavior change = '0'.. * $p< 0.05$. ** $p< 0.01$. *** $p< 0.001$.

Table 5-22. Prevalence of reported behavior changes among current study and Diabetes Prevention Trial –1 (DPT-1) study participants

Item	Current (n = 192)	DPT-1 (n = 116)	χ^2 (346)
Any behavior change[a]	64(33%)	50 (43%)	2.96
Diet/eating patterns	59 (31%)	42 (36%)	0.99
Fed more diet and sugar free drinks	12 (6%)	28 (24%)	20.48***
Fed child less soda	30 (16%)	30 (26%)	4.83*
Fed child less juice	23 (12%)	5 (4%)	5.15*
Fed child less sweet foods	38 (20%)	25 (22%)	0.14
Tried to get child to lose weight	4 (2%)	1 (1%)	0.68
Fed child less to eat	3 (2%)	0 (0%)	1.83
Fed child more juice	2 (1%)	1 (1%)	0.05
Fed more to eat	1 (1%)	0	0.61
Tried to get child to gain weight	0	0	n/a
Physical activity	20 (10%)	11 (10%)	0.07
Child exercised more often	19 (10%)	11 (10%)	0.01
Encouraged child to exercise less often	1 (0.5)	0	0.61
Medications	6 (3%)	15 (13%)	10.94**
Administered vitamins to child	6 (3%)	15 (10%)	6.48*
Administered insulin to child at home	1 (0.5%)	5 (4%)	5.44*
Used herbal supplements	0	2 (1%)	3.33
Administered nicotinamide	0	1 (1%)	1.66
Stress			
Had child rest more often	3 (1.6%)	3 (3%)	0.40

Note: Data reported as n (%). [a]Reported behavior change coded as At least one reported behavior change = '1', No reported behavior change = '0'.. *$p < 0.05$. **$p < 0.01$. ***$p < 0.001$.

CHAPTER 6
DISCUSSION

Understanding the behavioral impact of genetic risk knowledge helps to inform us about the ethical and social implications of genetic screening and has the potential to influence future legislative policies as screening becomes more widespread. Studies are needed in this area, as there is a lack of empirical data examining the effects of genetic screening on families' everyday lives. This study was exploratory in nature and represents a first look into the behavioral impact of newborn screening for type 1 diabetes. As such, results should be interpreted with some caution, especially due to psychometric weakness in some of the measures used and an unavoidably select sample of mothers.

Results of this study demonstrated risk identification of type 1 diabetes through newborn genetic screening did not lead to widespread maladaptive behaviors that might endanger the child or family or disrupt daily functioning. Contrary to this, results indicated healthy lifestyle changes and increased health surveillance. At a bare minimum, these contribute positively to a child's health and well-being overall with the added benefit of the increased potential for earlier detection and diagnosis should the child develop symptoms of type 1 diabetes. In addition, it is possible that in the long run, researchers may find that some of these specific behaviors may have protective effects for type 1 diabetes.

Hypotheses

Objective 1: To Investigate the Extent of Reported Maternal Behavior Change as a Result of Genetic Screening for Type 1 Diabetes

Over 60% of mothers reported engaging in at least one behavior change. These results are consistent with results from the Hendrieckx et al. (2002) study on intentions for behavioral change in adults with a first degree relative with type 1 diabetes who were undergoing antibody screening and assessed prior to results notification. Hendrieckx et al. (2002) found that 73% reported an intention to modify at least one behavior if testing results placed them at a high risk for diabetes. Our results are slightly higher than the 52% of children and 24% of adults who reported making a behavioral change as in the Johnson & Tercyak (1995) study on participants who were identified as antibody positive. Taken together, these results indicated that participants in screening studies may not only intend to make behavior changes prior to notification, but will actually take action if identified as "at risk" for developing type 1 diabetes.

In making comparisons between studies, it should be noted these are not direct comparisons as the studies used similar, but not identical self-report measures of behavior change. Hendrieckx et al. (2002) used a prospective design with an open-ended question to assess intentions for behavioral change. Respondents were asked to report what they would do differently in their daily lives if they were found to be at risk and responses were coded and categorized. Johnson & Tercyak (1995) used a retrospective design, but asked a very similar open-ended question in assessing actual behavior change following antibody screening. However, they did not present results on specific types of behavior changes. For this current study, a more detailed assessment was conducted with both forced choice and open-ended questions across different domains of behaviors.

Reported changes were most frequently reported in response to forced choice items (yes or no) rather than open-ended questions, suggesting that it was more difficult for mothers to spontaneously recall specific behaviors than it was for them to recognize behaviors that were presented to them. This may be particularly true of health surveillance behaviors, which mothers may not immediately recall, as they may not consider monitoring to be preventative in nature. According to forced choice items, increased health surveillance behaviors were the most commonly reported behavior changes. It is also possible that forced choice items may have indirectly created a demand characteristic for mothers to endorse behaviors.

In open-ended questions, spontaneous responses most frequently corresponded with healthier diet and exercise changes. In forced choice items, alterations in diet and exercise patterns were less common than health surveillance behaviors, but more prevalent than illness prevention, medications, or stress reduction. This is consistent with Henrieckx et al. (2002) who found that modifications of diet and exercise patterns were often cited as behaviors participants reportedly intended to change if found to be at risk for type 1 diabetes. While prevalence rates were not specified for different types of behavior changes, according to Johnson & Tercyak (1995), the majority of behavior changes reported in their study were in the areas of diet and increased physical activity.

Nearly all changes in diet and exercise reported in this study would be considered healthy lifestyle changes, consistent with current recommendations for the prevention of type 2 diabetes. Almost one half of participants who reported engaging in at least one behavioral prevention effort, reported engaging in specific behaviors congruent with current recommendations for type 2 diabetes prevention. While not as prevalent as

increased health surveillance, these results suggest that mothers may have applied recommendations for type 2 to their children at risk for type 1 diabetes. Our descriptive data indicated that 92% of mothers who believed they could do something reported changing their child's diet and exercise changes as possible methods of prevention. These findings are consistent with Pierce et al (1999) who found that nearly half of parents with type 2 diabetes reported believing they could reduce their child's risk of developing diabetes. Nearly all efforts noted by these parents to be perceived as effective were diet changes and increased exercise (Pierce et al., 1999). The key difference between these studies as that diet and exercise are effective in preventing type 2 diabetes (Ryan et al., 2003; Tuomilehto, et al., 2001).

This reflects a lack of understanding about the etiology of type 1 diabetes and highlights the fact that mothers may believe that changes in diet and exercise may be as important to the prevention of type 1 diabetes as they are to the prevention of type 2 diabetes. In fact, there are no currently available data that lifestyle behaviors are linked to the etiology of type 1 diabetes. Therefore, mothers appear to rely on available information to fill in where current scientific evidence is absent. Additional evidence of this lack of understanding comes from data indicating that while nearly 80% of mothers who reported checking for signs of diabetes in their children correctly identified at least one symptom, 45% of mothers also reported at least one incorrect symptom. Furthermore, approximately one fourth of mothers received advice from health professionals and family and friends; nearly all mothers reported following the advice they were given. Results indicated that of these, almost half received recommendations

for healthy lifestyle changes from health care professionals and one fourth received similar advice from family members or friends.

Current research suggests that there are environmental triggers of the immunological process leading to the development of type 1 diabetes. At the present time, these triggers are unknown. Thus far, lifestyle behaviors, specifically diet and exercise, have not been implicated as such triggers. There is no scientific evidence that diet and exercise changes can prevent type 1diabetes. Results of this study suggest that the public believes that healthy lifestyle changes advocated to prevent type 2 diabetes might also be effective at preventing type 1 diabetes. Therefore, as results indicated, many mothers of children at risk for developing type 1 diabetes reported engaging in behaviors that foster lifestyle changes in their children, such as healthier diets and increased exercise. Since researchers do not know the exact triggers of type 1 diabetes, when conducting natural history studies or prevention trials, these behavioral changes need to be monitored. In the future, researchers may find that certain environmental influences or lifestyle choices may indeed trigger an immunological process leading to type 1 diabetes. Similarly, researchers may find that healthy diet and physical activity may be protective factors for developing type 1 diabetes. In the meantime, these results suggest that participation in genetic screening and risk identification may have promoted healthier lifestyles for these children, through increased health surveillance, healthier eating, and increased physical activity.

These behavioral outcomes are important to consider in the context of ethical debates about newborn screening for type 1 diabetes. Critics argue that screening creates undue burden and causes distress in at risk families. Staunch opponents to screening

advocate that without a definitive prevention strategy, population screening should not be conducted at all. Further, if screening is conducted in only within a research context, families should not be notified of results (Friedman Ross, 2003).

Results suggest that mothers report healthy changes, and very few mothers are reportedly engaging in maladaptive or inappropriate behaviors suggestive of overprotectiveness or undue parental burden. Although, this cannot be confirmed as this study did not explicitly examine the negative or positive impact of these changes on families' quality of life. However, data collected in this study on the duration of behaviors suggests that most mothers who reported initiating a behavior continued their efforts consistently over time. Perhaps if certain behaviors were burdensome, mothers would have discontinued them. Furthermore, the benefits of increased health surveillance and healthy lifestyles are obvious in this population. Increased health surveillance may aid families in earlier detection and diagnosis should their child eventually develop diabetes.

Objective 2: To Assess the Predictors of Reported Maternal Behavior Change as a Result of Genetic Screening for Type 1 Diabetes

Results of regression analyses were generally consistent with Baum et al's Stress Disease Risk Coping Model (1997); however, this model was not intended to address issues pertaining to other family members and family dynamics (other than medical history) -- variables that other researchers believe are key in understanding the psychological impact of genetic testing results (Rolland, 1999; Tercyak, 2000). While this model was not intended to pertain to behavior changes in parents in response to a child's risk, it appears that this model fit well with current data and was useful in interpreting results of the current study. To date, the Stress Disease Risk Coping Model

has not been formally tested and therefore, future studies are needed to further develop and generate empirical and statistical support of this model.

In every model, the presence of a first degree relative with diabetes also predicted greater likelihood of behavior change. Most of these relatives had type 1 diabetes and in nearly half of these families, the relative was the mother herself. Mothers, who live with the disease and understand its severity, may be more inclined to take actions to prevent it from developing in their offspring. Additionally, having a close relative with diabetes is concrete evidence and a visible sign of genetic predisposition. This may increase one's perceived risk above and beyond results of screening for genetic (HLA) markers, and increase the likelihood of preventative actions.

These findings are consistent with Baum et al's (1997) model in which family history is considered a personal variable associated with risk appraisal. It is difficult to compare findings of the current study, which was conducted in the general population, with other published genetic screening studies as most involved selected samples of participants with family histories of a particular disease. However, in other health screening studies, Marteau & Lerman (2001) suggest that family history has an inconsistent relationship to behavior change. For example, in studies on heart disease, those who perceived a family history were no more likely to engage in risk reducing behavior (Becker & Levine, 1987) and in a separate survey, 15% of those with a family history of heart disease perceived a sense of fatalism (Hunt et al., 2001), which may in part explain why more people do not change their behavior if they perceive themselves as greater risk (Marteau & Lerman, 2001). However, it should be noted that these studies

were on adults about their own health risks and behavior, and it is possible that parents may be more active to prevent disease in a child.

As opposed to family history, actual risk, as determined by genetic screening, was not a significant predictor of behavioral change. While this study did not contain a control group to directly test this, it is possible that mothers of children with first-degree relatives may have engaged in these behavior changes regardless of whether screening had occurred. To explore this issue, future studies should include a sample of families with a first-degree relative with diabetes whose children did not undergo genetic screening.

As hypothesized, perceived risk, anxiety, coping, and information seeking were all predictors of behavior change. These findings support the Baum et al.'s (1997) Stress Disease Risk Coping Model, in which health behavior changes are impacted by both affective and cognitive variables, including psychological distress, coping resources, personal factors (i.e., information seeking).

Mothers who perceived their child to be at greater risk were more likely to report engaging in behavior change, consistent with previous studies on breast cancer risk and mammography screening (Meiser et al., 2000, Ritvo et al., 2002). It seems logical that mothers who perceive their child to be more susceptible to a disease would be more likely to take action to reduce their risk, as hypothesized in several models of health behavior (Leventhal, 1970; Rosenstock, 1974). Pierce et al. (1999) found that those who hypothesized their risk to be high were also more likely to worry. It is possible that mothers who perceived their child's risk to be higher would be more likely to feel anxious and thus, more likely to initiate behavior change.

In this study, actual risk was not a significant predictor, which is similar to findings of Aiken et al. (1994), in which perceived risk above and beyond actual risk predicted behavior change. These results suggest that in predicting behavior, it was more important how mothers viewed their child's risk than the risk itself. However, it is important to keep in mind that all children in this sample were identified as at increased risk for type 1 diabetes (as compared to the general population) to begin with. If this study had included a wider range of risk groups (i.e., protected to extremely high risk), actual risk may have been a predictor. Additionally, it should be noted that the highest level of actual risk ("extremely high risk") identified through screening signifies the child has a 20% probability of developing diabetes.

Anxiety at the time of the current interview was also a significant predictor of behavior change, even when controlling for initial anxiety. Congruent with Johnson & Tercyak (1995) and Hendrieckx et al. (2002), mothers who reported greater diabetes-specific worry and anxiety were more likely to report engaging in preventative behaviors. This lends further support to studies of other populations suggesting that a person's affective response (i.e., distress) to a health threat is related to whether they adopt health-protective behaviors (e.g., Diefenbach et al., 1999). Furthermore, it supports the Stress Disease Risk Coping Model, which theorizes that a person's stress response will in turn influence modifications in health behaviors (Baum et al., 1997).

Again, consistent with Baum et al. (1997), mothers who reported using more coping strategies at the 4-month follow-up interview were more likely to report taking preventative action. Engaging in behaviors viewed as potentially risk-reducing may represent another method of coping with a health threat. As hypothesized, the total score

from the Ways of Coping Checklist-Revised (WCC-R) and three of its coping scales were significant predictors of behavior change. Active coping, as characterized by seeking social support and problem-focused coping constructs, were both significant predictors. Somewhat surprising, passive coping, as exemplified by the construct of wishful thinking, was also a significant predictor of behavior change. When all three coping scales were entered into the summary model, only wishful thinking remained significant. Based on analyses of items in this scale, it appears that the construct includes items that reflect optimism but also a desire for the problem to "go away." Mothers who wished the problem would resolve and felt optimistic about the possibilities were more likely to engage in preventative behaviors despite the lack of available recommendations.

As hypothesized, information seeking was related to behavior change, more so than other predictor variables, such as anxiety, risk perception, and coping style. Mothers who reported having more sources of information regarding type 1 diabetes were more likely to report engaging in behavior change. Mothers in this study reported following advice given to them by others and it is not surprising that more information was related to greater likelihood of taking actions. Information seeking can be viewed as a form of active coping when dealing with a health threat. Seeking out more information is a proactive step, much like changing one's behavior. Additionally, most of the information in the media as well as data collected in this study on information from family and friends appear to be directed at type 2 diabetes and healthy lifestyle changes. The more information that mothers receive about diabetes, they more likely they may be to apply available recommendations to their at risk child.

Contrary to the original hypothesis, perceived control was not a significant predictor nor did it demonstrate any significant interaction effects with other predictor variables. This was incongruent with Baum et al.(1997) because perceived control was originally considered part of the appraisal process described as "risk perception." Our findings are in contrast to Hendrieckx et al (2002) who found that adult participants who perceived greater internal control were more likely to report intentions for behavior change. It should be noted that Hendrieckx (2002) explored adults' intentions for their own behavior, rather than the current study of mothers' reported behavior changes related to their children.

Another unexpected finding from this study was that participation in the PANDA study (Part II) did not predict to whether behavior change occurred. Study participation could be viewed as health surveillance behavior in and of itself, but it was surprising to find that this was not be related to other types of health surveillance or changes in other behavior domains. However, in interacting with participants, PANDA study staff remind mothers there are no current recommendations for prevention of type 1 diabetes, so continued contact with staff would not necessarily increase the likelihood of behavioral change.

Objective 3: To Assess Psychological Effects (i.e., Anxiety) of Maternal Behavior Change Over Time

Contrary to the original hypothesis, behavior change did not appear to decrease anxiety over time. Mothers who reported behavior change and perceived greater control were more anxious at time of the current interview. In essence, behavior change did not decrease anxiety over time, it actually was associated with higher levels of anxiety as compared to those who did not reported behavior change and who perceived less control.

These findings are consistent with Pierce et al. (1999) who found that parents who thought prevention was possible were more likely to worry about their children developing type 2 diabetes. It is somewhat surprising that mothers who perceived greater control were more anxious. Perhaps, when mothers feel they have control, it engenders a sense of increased responsibility for their child's health and therefore, increased anxiety. Similarly, it may also be that there is stress that accompanies believing that there is something one ought to be doing to prevent diabetes in one's children.

Sociodemographic predictors of anxiety at the current interview, above and beyond initial anxiety, included child's age and family history of diabetes. Mothers of children who were younger and who had relatives with diabetes were more anxious at the current interview. Previous studies within the larger PANDA population are consistent with these findings that family history is a significant predictor (Johnson et al., submitted). This may reflect the relationship between anxiety and elevated perceived risk, as may be present when there is a positive family history. Current data indicated a significant correlation between composite scores of perceived risk and anxiety as have previous studies using data from the PANDA study (Johnson et al., submitted). The lack of other significant sociodemographic predictors as found in Johnson et al. (submitted) when predicting to earlier assessments of anxiety (i.e, PANDA Part III interviews), may be due to the homogeneity of this current sample, length of time since notification, and controlling for the effect of initial anxiety.

Objective 4: To Compare Reported Behavior Change between Mothers of Children Genetically at Risk for Developing Type 1 Diabetes with Mothers of Children in the Diabetes Prevention Trial Who Were ICA+, and Therefore, at Even Greater Risk for Diabetes Onset

In terms of reported behavior, there were relatively few differences between these two samples of mothers. As hypothesized, mothers of children enrolled in the DPT-1 trial were more likely to report at least one behavior change, but the difference only approached significance, with 43% of mothers in the DPT-1 sample versus 33% in the PANDA sample. Data collected from DPT-1 participants themselves indicated that 54% reported at least one behavior modification, which was slightly higher than maternal report (Johnson, 2002). The prevalence of reported behavior change in the DPT-1 sample is comparable to rates reported in other studies of people who are ICA+. Johnson and Tercyak (1995) found 52% of children and 25% of adults reported engaging in prevention efforts. This may be explained by the difference in actual risk between ICA+ samples, including the DPT-1 (Johnson, 2002) and Johnson & Tercyak (1995), and genetic screening samples, such as this PANDA sample. Those who are ICA+ are at significantly greater risk of developing type 1 diabetes (i.e., approximately 45% chance of developing diabetes), versus those who are identified through genetic screening. The highest risk classification, "extremely high risk" signifies having a 20% chance of developing diabetes. Additionally, while the rates of behavior change overall did not differ between the current sample and the DPT-1 sample, the difference may also reflect differences in anxiety, as previous studies have found that mothers of ICA+ children report higher levels of anxiety than mothers of genetically at risk children (Johnson et al., submitted). Mothers who were more anxious may indeed be more likely to report

engaging in prevention efforts, as found in this study (see Objective 2) and this may also be true of the DPT-1 study population.

In analyses of individual items, mothers in both samples more frequently endorsed changes in diet and physical activity, consistent with previous studies (Johnson, 2002; Johnson & Tercyak, 1995). Differences between the two samples included mothers in the DPT-1 trial more often reporting feeding their children less regular soda, less sweet food, and more diet or sugar-free drinks. However, mothers in the current sample more often reported feeding their children less juice. Additionally, mothers in the DPT-1 sample reported more often administering vitamins and insulin. It is not surprising that administering insulin was more prevalent in the DPT-1 study as presumably all mothers understood the purpose of the study was to determine if early administration of insulin was a effective prevention method. Additionally, 45% of children were enrolled in the experimental arm of the study in which they received to begin with, and therefore, it may have been more readily available. Increased use of vitamins may be explained by the use of a paper questionnaire. In our survey, many mothers frequently reported administering vitamins to their children but when asked again by the interviewer if this was because of their child's diabetes risk, they no longer endorsed this item. With a paper survey as used in the DPT-1, no one is there to remind the mothers these practices are intended to only reflect specific efforts to prevent diabetes.

Strengths and Limitations

The proposed study design has several strengths. Hendrieckx et al. (2002) was the first study to systematically explore behavioral efforts to prevent type I diabetes in at risk individuals. However, outcomes were measured as intentions, not behavior changes, and assessment occurred prior to screening. In contrast, this proposed study was

conducted several years post-notification and examined actual reported behavior change. The study design also permitted linkage to data collected during previous interviews. This allowed for prospective statistical analyses to interpret temporal relationships between variables and predictors of reported behavior change, including the relative contributions of personal, affective and cognitive appraisal variables (e.g., psychological distress, risk perception, perceived control, sociodemographics). Behavioral outcomes in this proposed study sample were also compared with those of mothers of children at even greater risk for developing type 1 diabetes who entered a prevention trial (DPT-1).

Additionally, our study's design permitted analyses of both recognition and recall data, through the use of open-ended and forced choice questions. This was possible due to the retrospective nature of our study. While results indicated that recognition (i.e., forced choice items) yielded a higher rate of behavior change and more detailed information, the recognition method, if used prospectively in prevention trials might prompt behavior change in participants. For example, providing mothers with a list of possible behaviors may inadvertently encourage mothers to initiate behaviors they otherwise might not otherwise engaged in. Consequently, any subsequent data collected on behavior may not be valid as it was not an action mothers engaged in on their own. Researchers should be cautious about using a recognition approach since a long list of forced choice items may increase social desirability.

The study provided important guidance as to which domains of behavior change are most commonly reported and should be targeted for more expensive observational data collection. Indeed, this study has shown a large proportion of mothers reported at least some behavior change as a result of genetic screening. Health surveillance and

behaviors characterized as healthy lifestyle changes were most commonly reported. These are behaviors that could be operationally defined and tracked in future research studies.

This study has several limitations. First, nearly all of the behavior change data collected were based on self-report and may be subject to social desirability bias. Whether mothers had their child participate in PANDA blood-draws was the only observational behavioral data available. These data indicated mothers were largely accurate in their report of participation, suggesting their responses in the rest of the interview may also be valid. Mothers were more likely to report not participating in blood draws when they actually had than inaccurately stating they had participated when they had not. Additionally, the rates of behavior change were consistent with other studies suggesting that mothers were not overly endorsing practices (Hendieckx et al., 2002; Johnson, 2002, Johnson & Carmichael, 2000). The apparent lack of social desirability may have been a direct result of our efforts to use language in the interview to minimize such bias, reiterating at several points in the interview that there were no right or wrong answers, nor are their known methods to prevent the development of type 1 diabetes. However, as with all self-report data, social desirability is a factor to consider.

The second study limitation was the lack of representativeness of the study's sample. To some extent this was unavoidable as a significant proportion of mothers were unable to be contacted, despite multiple attempts by staff. In this sample, minorities were under-represented, even more so than even in the original PANDA sample. Mothers who completed the current interview, as compared to mothers who were unable to be contacted, were significantly older, more educated, had higher family incomes, and were

more likely to be married. Additionally, they were more accurate in their reported understanding of their child's risk. Most of these mothers had completed previous interviews, suggested a higher level of concern and motivation. The lack of socioeconomic variability in this sample is a weakness of this study as results may not be generalizable to PANDA participants as a whole or to the general population. Additionally, it is possible that some demographic variables may predict behavior change, although, this was not apparent due to the restricted range in the current sample.

In terms of genetic risk, this sample contained few mothers of children who were at extremely high risk, although the prevalence rate in the current study is representative of the entire PANDA study population. The current study did not include participants who were at low risk for developing diabetes to use as a comparison group. This was not feasible at the present time as PANDA only longitudinally follows children at moderate, high, or extremely high risk. However, gathering data on those in lower risk groups could be useful information in future studies.

Third, measures used in this study were largely new and were not psychometrically sound. Predictor variables often had too few items (i.e. perceived control, perceive risk) with moderate levels of internal consistency. It is important in future studies to develop better measures for these constructs by adding more items, conducting in depth pilot testing and/or create new measures which specifically relate to genetic screening. In terms of the study's outcome measure, most of the behavior interview was new, with many items adapted from a similar survey used in only one previous study. Thus, it was difficult to determine an accurate data analysis plan a priori. Results indicated there was satisfactory internal consistency for only two of the behavior

domains. Additionally, behavioral data was not normally distributed and therefore, fine-grained analyses including continuous behavior scores that reflected duration and frequency could not be successfully analyzed.

Despite these limitations, this study represents an important exploratory step in understanding maternal reactions to the news a child is at risk for a disease with no known method to prevent disease onset. This information is critical in efforts to develop the best risk communication methods, provide families with appropriate support in response to risk communication, design natural history studies of disease onset and progression, and design clinical trials aimed at disease prevention.

Implications and Directions for Future Research

This study provides unique contributions to the literature in several ways. First, very few studies have examined the behavioral impact of genetic screening, especially non-health surveillance behaviors (e.g., diet, exercise, stress reduction). Second, this study improves our understanding of the public's awareness of diabetes, including the lack of discrimination between types 1 and 2, as well as beliefs regarding diabetes prevention efforts. Many of behaviors reported by mothers in this sample were consistent with recommendations for type 2 diabetes, suggesting the medical community's efforts to increase awareness of the importance of a healthy lifestyle have been effective. However, many mothers believed they could prevent their child from developing type 1 diabetes, when there is currently no evidence to support this belief. These studies highlight the continued need to keep the public informed of current medical research and provide education regarding the distinctions between types of diabetes. Furthermore, it speaks to providing participants in genetic screening and natural history studies with more detailed information regarding the nature of type 1 diabetes.

Data collected in this study allowed us to gain a better understanding mothers' advice-seeking behavior. A significant proportion of mothers received advice from a medical professional; however, a sizable portion also received advice from family and friends. While its is unclear exactly how this advice may have influenced behavior in this study, data indicated that nearly all mothers followed advice given to them by family or friends while slightly less followed the advice given to them by medical professionals. Future studies are needed in this area to explore how people seek out medical advice and which sources are trusted the most for accurate information. While it was not a focus of this study, results from open-ended questions indicated several mothers relied on prayer to help prevent their child from developing diabetes. The role of religion in coping with a health threat, particularly as it may pertain to predictive genetic testing and counseling, may be an area worthy of further exploration.

Finally, it is crucial when designing clinical trials, particularly natural history and prevention studies, to understand the behavior changes that may occur in individuals' everyday lives in response to a health threat. Unless carefully monitored, such behavior modifications could threaten the internal validity of natural history studies and prevention trials. The scientific community is still unclear regarding effective prevention methods for type 1 diabetes and it is possible some of the measures taken by these mothers to prevent diabetes from developing in their children may ultimately be determined to be beneficial or harmful. It is clear that genetics is only part of the story. Environmental triggers, which could include behavioral factors such as diet or exercise habits, do play a role. However, scientists do not yet know what those environmental triggers are,

highlighting the importance of the continued study of attempted behavior change in response to risk notification.

APPENDIX
STRUCTURED TELEPHONE INTERVIEW

Sample Script for Male Child

Date:_____

Hello, is *(say mom's name)* there please? This is _____ *(principal investigator or research assistant)* and I'm calling from the Diabetes Research Office at the University of Florida. You might remember that your son *(say child's name)* participated in the PANDA study, in which his blood was tested to see if he had any genes that might put him at risk for developing type I diabetes. You were told about his results and then we talked with you over the phone several times to ask you some questions about your feelings regarding your child's involvement in this study. Does this sound familiar to you?

It has been awhile since we last talked with you, but if you don't mind I'd like to ask you a few more questions. Like the other times we have talked with you, all the information is kept private and confidential – we won't tell anyone else- and you can refuse to answer any question at any time. For this interview, it does not matter if you brought you child in for more blood draws or not. If you feel uncomfortable answering a question, feel free to skip that item. You are free to discontinue your participation in this interview at any point in time.

In doing this interview, there are no direct benefits to you. But, to thank you for helping us we would like to send you in the mail a $5 gift certificate to either Target or Publix—your choice. The interview should take about 15-20 minutes. Would you be interested in participating? Is now a good time for you? *(If not, find a good time. If mom refuses interview, thank her for her earlier participation in PANDA and previous interviews).*

To get started, I would like to make sure that we have current basic information about you and your family. You may remember these same questions from before and I would like to know if anything has changed.

Family's mailing address (to be used solely for mailing gift certificates)

Street Address: _____

City: _____ **State:** _____ **Zip Code:** _____

Parents Marital Status:_____
1=single parent, involved father
2=single parent, non-involved father
3=married
4=separated
5=divorced

How far did you go in school? (mom):_____ **father:** _____
1=some high school
2=graduated HS/GED
3=some college or trade school
4=graduated from college/trade school
5=some graduate school or professional program
6=graduated from graduate or professional school

# of children in the family:_____(first child? Y/N)

Annual Total Family Income: _____

1=0-10,000	4=30,001-40,000	7=60,001-70,000	10=90,001-100,000
2=10,001-20,000	5=40,001-50,000	8=70,001-80,000	11=100,000+
3=20,001-30,000	6=50,001-60,000	9=80,001-90,000	

Does anyone in your son's family have diabetes? (TO INTERVIEWER: If there is family history reported in earlier interviews, say: "Does anyone else in your son's family besides _____ *[list relations as previously reported]* have diabetes now?"
(include information regarding the mother's estimating of diabetic control and how problematic the disease has been for all diabetic family members, including those from before)

Relationship to child (circle)	Type	Age at onset	Live with you?	Duration	Control*	Problems**
Mother						
Father						
Full Sibling						
Full Sibling						
Half Sibling						
m-grandmother						
m-grandfather						
m-grt.grandmother						
m-grt.grandfather						
p-grandmother						
p-grandfather						
p-grt.grandmother						
p-grt.grandfather						
(other)						

Note. Indicate if they DON'T KNOW.*1=excellent 2=good 3=fair 4=poor 5=DK **1=none 2=a few3=several 4=a lot 5=DK

The PANDA study coordinator might have told you that your son was more or less at-risk for developing diabetes than other people in the general population. I'm going to read some categories and give you some numbers, and I want you to tell me which risk category you were told your son was in. Okay? Were you told your baby was at: (circle one)

1. Very low risk (~1/5000)
2. Low risk (~1/600)
3. Intermediate risk (~1/125)
4. Moderate risk (~2/100)
5. High risk (~5 or 10/100)
6. Extremely high risk (~1/5)
7. Don't know/don't remember

What are your thoughts on having had your son tested at birth for diabetes risk? (check one)

- ❒ Glad you participated in the study
- ❒ Not sure about your participation in the study
- ❒ Wish you had not participated in the study

What do you think will happen to your child in regards to developing diabetes? (check one)

- ❒ My child will develop diabetes in the near future
- ❒ My child will eventually develop diabetes but not for a long time from now
- ❒ My child will not ever develop diabetes
- ❒ I am unsure what will happen

How do you think your son's risk compares to other children for developing diabetes? *(circle one)*

1 Much lower
2 Somewhat lower
3 About the same
4 Somewhat higher
5 Much higher

How often do you worry that your son will get diabetes? *(circle one)*

0 Never
1 Rarely
2 Sometimes
3 Often
4 Very often

I want you to think about your son's risk for developing diabetes. I'm going to describe some feelings to you, and I want you tell me how you feel right now at this moment about the situation.

GIVE STAI-SF10
Would you say you currently feel: (circle one for each)

Not at all calm	somewhat calm	moderately calm	very calm
Not at all secure	somewhat secure	moderately secure	very secure
Not at all tense	somewhat tense	moderately tense	very tense
Not at all at-ease	somewhat at-ease	moderately at-ease	very at-ease

You are presently worrying over possible misfortunes:

Not at all	somewhat	moderately	very much
Not at all frightened	somewhat frightened	moderately frightened	very frightened
Not at all comfortable	somewhat comfortable	moderately comfortable	very comfortable
Not at all nervous	somewhat nervous	moderately nervous	very nervous
Not at all relaxed	somewhat relaxed	moderately relaxed	very relaxed
Not at all worried	somewhat worried	moderately worried	very worried

INFORMATION SEEKING

Did you talk to your family doctor or pediatrician about your son's diabetes risk screening results?
❒ **Yes** ❒ **No**

IF YES: Did you get any advice from your physician? ❒ **Yes** ❒ **No**
What was the advice?

_____

_____

Did you take the advice? ❒ **Yes** ❒ **No**

Did you talk about your child's risk with family members or friends? ❒ **Yes** ❒ **No**

Who?

- ❒ **Child's grandparent**
- ❒ **Spouse**
- ❒ **Other family member or friend who has diabetes**
- ❒ **Other family member or friend who does not have diabetes**
- ❒ **Other (please specify)**_____

Did you get any advice from them? ❒ **Yes** ❒ **No**

From whom did you get advice?

- ❒ **Child's grandparent**
- ❒ **Spouse**
- ❒ **Other family member or friend who has diabetes**
- ❒ **Other family member or friend who does not have diabetes**
- ❒ **Other (please specify)**_____

(if any of above checked)

What was the advice from _____(#1 listed)?

_____

_____

Did you take the advice? ❒ **Yes** ❒ **No**

What was the advice from _____(#2 listed)?

_____

_____

Did you take the advice? ❒ **Yes** ❒ **No**

What was the advice from _____(#3 listed)?

_____

Did you take the advice? ❒ **Yes** ❒ **No**

Did you search the Internet for information on your child's risk?

❒ **Yes** ❒ **No**

Did you read a book or other literature regarding diabetes and diabetes risk?

❒ **Yes** ❒ **No**

Did you watch diabetes-related programming on television (i.e., news stories)?

❒ **Yes** ❒ **No**

Are there any other things I did not mention that you did in order to get more information about diabetes and/or your child's risk?

_____

Right now, doctors and scientists don't know how to stop type 1 diabetes from developing. But even so, people may have different ideas about what might work or not. Some people might think there is something special you can do to stop your son from getting type 1 diabetes. Other people might think there is nothing you can do. We are interested in knowing how you have responded to the news that you son is at risk for diabetes. This information may help in future research. Whether you did something or did nothing is equally important information. There are no right or wrong answers. We want you to be as honest as possible.

Did you do anything special to try to prevent diabetes in your son?

❒ **Yes** ❒ **No**

How much do you agree or disagree with the following statement: "I can do something to reduce my son's risk for developing type I diabetes"?

❒ **Strong Disagree**
❒ **Disagree**
❒ **Neutral**
❒ **Agree**
❒ **Strongly Agree**

IF AGREE/STRONGLY AGREE: What types of things do you think you can do to try to prevent your child from developing type I diabetes?

_____

How much do you agree or disagree with the following statement: "Medical professionals can do something to reduce my son's risk for developing type I diabetes"?

❒ **Strong Disagree**
❒ **Disagree**
❒ **Neutral**
❒ **Agree**
❒ **Strongly Agree**

IF AGREE/STRONGLY AGREE: What types of things do you think medical professionals can do to try to prevent your child from developing type I diabetes?

_____

How much do you agree or disagree with the following statement: "It is up to chance or fate whether my son develops type I diabetes"?

❒ **Strong Disagree**
❒ **Disagree**
❒ **Neutral**
❒ **Agree**
❒ **Strongly Agree**

****

Now, I am going to get a little more detailed in the questions I will be asking you. Again, I want you to keep in mind that there are no right or wrong answers. We just want to know about anything you have tried or are doing specifically to try to prevent type I diabetes from developing in your son.

DIET: Have you done anything different with your son's diet or eating patterns to prevent him from developing diabetes? *(list verbatim below and check off corresponding items below followed by questions in right-hand column)*

_____

I am going to list off other things you may not have mentioned or thought of right now and I want you to tell me if you have done them. The responses to these questions are just yes or no.

DIET/EATING PATTERNS

#	QUESTION			IF YES, ASK THE FOLLOWING...		
1.	Have you fed your son less candy, cookies, cake and other sweet foods?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
2.	Have you fed your son less regular soda and sweet drinks?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
3.	Have you fed your son more diet soda and sugar free drinks?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		

4.	Have you given your son less juice to drink?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
5.	Have you given your son more juice to drink?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
6.	Have you tried to get your son to lose weight?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
7.	Have you tried to get your son to gain weight?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		

8.	Have you fed your son more often than you otherwise would have?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
9.	Have you fed your son less often than you otherwise would have?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
10.	Did you introduce solid foods, such as baby food, table food, or cereal, earlier than you had planned?	**NO**	**YES**	By how long? _____ (in months) How old was your child when he was first given solid foods? _____ (in months)		
11.	Did you introduce solid foods, such as baby food, table food, or cereal, later than you had planned?	**NO**	**YES**	By how long? _____ (in months) How old was your child when he was first given solid foods? _____ (in months)		
12.	Did you delay giving your son cow's milk?	**NO**	**YES**	By how long? _____ (in months) How old was your child when he was first given cow's milk? _____ (in months)		

13.	Have you avoided giving your son cow's milk altogether?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
14.	Have you encouraged your son to eat more than you otherwise would have?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
15.	Did you encourage your son to eat less than you otherwise would have?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
16.	Did you breastfeed your son?	**NO**	*YES*	How long did you breastfeed your son? _____months/years (circle one) Was your decision to breastfeed him influenced by his risk status for developing type I diabetes? Did you breastfeed your son for a longer or shorter time than you expected because of his risk? (circle one) Longer Shorter Neither About how much longer/shorter? _____ months/years (circle one)	**NO**	**YES**

PHYSICAL ACTIVITY/PHYSICAL STRESS: Have you done anything different with your son's physical activity patterns to prevent him from developing diabetes? *(list verbatim below and check off corresponding items below followed by questions in right-hand column)*

_____

I am going to list off other things you may not have mentioned or thought of right now and I want you to tell me if you have done them. The responses to these questions are just yes or no.

PHYSICAL ACTIVITY/PHYSICAL STRESS

#	QUESTION			IF YES, ASK THE FOLLOWING…		
1.	Have you encouraged your son to be active, doing something physical, every day?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
2.	Have you encouraged your son to exercise more often?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
3.	Have you encouraged your son to be less active so that he would not get tired?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		

4.	When playing hard, have you encouraged your son rest more so that he would not overdo it?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

HEALTHY LIFESTYLE/EMOTIONAL STRESS: Have your done anything differently to lower your son's stress in order to prevent him from developing diabetes? *(list verbatim below and check off corresponding items below followed by questions in right-hand column)*

_____

I am going to list off other things you may not have mentioned or thought of right now and I want you to tell me if you have done them. The responses to these questions are just yes or no.

HEALTHY LIFESTYLE/EMOTIONAL STRESS

#	QUESTION			IF YES, ASK THE FOLLOWING…		
1.	Have you encouraged your son to get more rest?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
2.	Have you actively tried to lower your son's stress level?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

3.	When your son gets upset, have you tried harder to take his attention away from the situation or get him focused on something different?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
4.	Have you tried to keep your son away from situations that you felt might upset him?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

MEDICAL SURVEILLANCE: Have you done anything to monitor or keep an eye on your son's risk of developing diabetes? *(list verbatim below and check off corresponding items below followed by questions in right-hand column)*

_____

I am going to list off other things you may not have mentioned or thought of right now and I want you to tell me if you have done them. The responses to these questions are just yes or no.

MEDICAL SURVEILLANCE

#	QUESTION			IF YES, ASK THE FOLLOWING…		
1.	Have you taken your son to doctor's visits more frequently?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

2.	Have you checked your son's blood glucose level at home?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
3.	Have you had your pediatrician check your son's blood glucose level at their office?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
4.	Have you had your son's blood drawn to test for autoantibodies as part of PANDA study?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
5.	Have you watched for signs in your son that you think may be related to symptoms of type I diabetes?	**NO**	**YES**	**What kinds of things have you looked for?** _____ _____ **A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

MEDICATIONS: Have you given your son any pills or medications to prevent him from developing diabetes? If yes, what kinds? *(list verbatim below and check off corresponding items below followed by questions in right-hand column)*

_____

I am going to list off other things you may not have mentioned or thought of right now and I want you to tell me if you have done them. The responses to these questions are just yes or no.

MEDICATIONS

#	QUESTION			IF YES, ASK THE FOLLOWING…		
1.	Have you given your son vitamins?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
2.	Have you given your son any medications for diabetes such as glucophage or insulin?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
3.	Have you given your son insulin shots at home?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		

4.	Have you given your son herbal supplements?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**
5.	Have you given your son nicotinamide?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

PROTECTIVE BEHAVIORS: Have you actively done anything special to lower your son's chances of getting sick in order to prevent him from developing diabetes? *(list verbatim below and check off corresponding items below followed by questions in right-hand column)*

_____

I am going to list off other things you may not have mentioned or thought of right now and I want you to tell me if you have done them. The responses to these questions are just yes or no.

PROTECTIVE BEHAVIORS

#	QUESTION			IF YES, ASK THE FOLLOWING…		
1.	Have you worked harder to protect your son from germs?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk? **B.** Is this something you did early on and then stopped **OR** Did you start doing this just recently? **C.** During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**	**NO** (ask B & C) **NO** **NO**	**YES** (ask C) **YES** **YES**

2.	Have you kept your son out of daycare with other children to protect him from germs?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
3.	Have you increased your son's exposure to other children (i.e., put him in daycare with other children) to boost his immunity?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
4.	Have you had your son wash his hands more often?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
5.	Have you limited your son's exposure to other children because you were worried he might get sick and increase his risk?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		

6.	Have you limited your son's exposure to tobacco smoke (i.e., cigarettes, cigars)?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
7.	Have you actively try to reduce your son's contact with harmful chemicals (i.e., pollution, food additives)?	**NO**	**YES**	**A.** Is this something you have been doing ever since you found out about your son's risk?	**NO** (ask B & C)	**YES** (ask C)
				B. Is this something you did early on and then stopped **OR** Did you start doing this just recently?	**NO** **NO**	**YES** **YES**
				C. During the time you were doing this, was it ❒ **Occasionally** ❒ **Always/Nearly Everyday**		
8.	Have you delayed immunizations (i.e., DPT, MMR) for your son?	**NO**	**YES**			

Is there anything else that we have not covered that you have done or are still doing to try to prevent type I diabetes from developing in your child?

We have come to the end of the interview. I know I have asked you about a lot of things today. I want to say again that unfortunately, scientists and doctors still do not know exactly what causes type I diabetes in children. Because of this, we don't have any specific recommendations to give you and the questions I just asked you should not be considered to be things you should or should not be doing. As in all matters of health, the best we can suggest is maintaining a healthy lifestyle for your child, including a balanced diet, activity, and rest. Thank you so much for your time and helping us with our study. We will be mailing you a gift certificate in the next few weeks. If you would like, we would also be willing to send you a summary of our results once the study is completed.

LIST OF REFERENCES

Abidin, R.R. (1997) Parenting Stress: A measure of the parent child system. In Zalaquett, C.P. & Wood, R.J. (Eds.), *Evaluating stress: A book of resources* (pp. 277-291). Lanham: Scarecrow.

Aiken, L. S., West, S. G., Woodward, C. K., & Reno, R. R. (1994). Health beliefs and compliance with mammography-screening recommendations in asymptomatic women. *Health Psychology, 13*, 122-129.

Akerblom, H., & Knip, M. (1998). Putative environmental factors in type 1 diabetes. *Diabetes Metabolism Review, 14*, 31-67.

Akerblom, H. K., Savilahti, E., Saukkonen, T. T., Paganus, A., Virtanen, S. M., & Teramo, K. (1993). The case for elimination of cow's milk in early infancy in the prevention of type 1 diabetes: The Finnish experience. *Diabetes Metabolic Review, 9*, 269-278.

Al-Jader, L. N., Goodchild, M. C., Ryley, H. C., & Harper, P. S. (1990). Attitudes of parents of cystic fibrosis children towards neonatal screening and antenatal diagnosis. *Clinical Genetics, 38*, 460-465.

American Academy of Pediatrics Committee on Bioethics (2001). Ethical issues with genetic testing in pediatrics. *Pediatrics, 107*, 1451-1455.

American Diabetes Association (2001). Nutrition recommendations and principles for people with diabetes mellitus. *Diabetes Care, 23*, S44-50.

American Diabetes Association (2002a). Evidence-based nutrition principles and recommendations for the treatment and prevention of diabetes and related complications. *Diabetes Care, 25*, S50-S60.

American Diabetes Association. (2002b). The prevention or delay of type 2 diabetes. *Diabetes Care, 25*, 742-749.

American Society of Human Genetics (1996). ASHG Report: Statement on informed consent for genetic research. *American Journal of Human Genetics, 59*, 471-474.

American Society of Human Genetics, American College of Medical Genetics (1995). Points to consider: Ethical, legal, and psychosocial implications of genetic testing in children and adolescents. *American Journal of Human Genetics, 57*, 1233-1241.

Arslanian, S., Becker, D., & Drash, A. L. (1997). Diabetes mellitus in the child and adolescent. In M. S. Kappy, R. M. Blizzard, & C. J. Migeon (Eds.), *The Diagnosis and Treatment of Endocrine Disorders in Childhood and Adolescents* (pp. 961-1026). Springfield: Thomas.

Audrain, J., Boyd, N. R., Roth, J., Main, D., Caporaso, N. S., & Lerman, C. (1997). Genetic susceptibility testing in smoking-cessation treatment: One-year outcomes of a randomized trial. *Addictive Behaviors, 22*, 741-751.

Audrain, J., Schwartz, M. D., Lerman, C., Hughes, C., Peshkin, B. N., & Biesecker, B. (1998). Psychological distress in women seeking genetic counseling for breast-ovarian cancer risk: The contributions of personality and appraisal. *Annals of Behavioral Medicine, 19*, 370-377.

Baroni, M. A., Anderson, Y. E., & Mishcler, E. M. (1997). Cystic fibrosis newborn screening: Impact of early screening results on parenting stress. *Pediatric Nursing, 23*, 143-151.

Baum, A., Friedman, A. L., & Zakowski, S. G. (1997). Stress and genetic testing for disease risk. *Health Psychology, 16*, 8-19.

Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbaugh, J. (1961). An inventory for measuring depression. *Archives of General Psychiatry, 4*, 561-571.

Becker, D. M., & Levine, D.M. (1987). Risk perception, knowledge, and lifestyles in siblings of people with premature coronary artery disease. *American Journal of Preventative Medicine, 3*, 45-50.

Becker, M. H., & Maiman, B. A. (1975). Sociobehavioral determinants of compliance with health and medical care recommendations. *Medical Care, 13*, 10-22.

Beisswinger, P. J. (2000). Type 1 diabetes. In J. L. Leahy, N. G. Clark, & W. T. Cefalu (Eds.), *Medical Management of Diabetes Mellitus* (pp. 95-114). New York: Marcel Dekker.

Boland, C., & Thompson, N. L. (1990). Effects of newborn screening of cystic fibrosis on reported maternal behaviour. *Archives of Diseases in Childhood, 65*, 1240-1244.

Bradley, C. (1994). Diabetes Onset Locus of Control (DOLoC). In C. Bradley (Ed.), *Handbook of Psychology and Diabetes* (pp. 397). Chur, Switzerland: Harwood Academic.

Bradley, D. M., Parsons, E. P., & Clarke, A. J. (1993). Experience with screening newborns for Duchenne muscular dystrophy in Wales. *BMJ, 306*, 357-360.

Bratt, O., Damber, J. E., Emanuelsson, M., Kristofferson, U., Lundgren, R., & Olsson, H. (2000). Risk perception, screening practice and interest in genetic testing among unaffected men in families with hereditary prostate cancer. *European Journal of Cancer, 36*, 235-241.

Broadstock, M., Michie, S., & Marteau, T. (2000). Psychological consequences of predictive genetic testing: A systematic review. *European Journal of Human Genetics, 8*, 731-738.

Browner, C. H., Preloran, M., & Press, N. A. (1996). The effects of ethnicity, education and an informational video on pregnant women's knowledge and decisions about a prenatal diagnostic screening test. *Patient Education and Counseling, 27*, 135-146.

Burnett, C. B., Steakley, C. S., Slack, R., Roth, J., & Lerman, C. (1999). Patterns of breast cancer screening among lesbians at increased risk for breast cancer. *Women and Health, 29*, 35-55.

Carmichael, S.K., Johnson, S.B., Baughcum, A.E., North, K., Hopkins, D., Dukes, M.G., She, J., & Schatz, D.A. (2003) Prospective assessment in newborns of diabetes autoimmunity (PANDA): Maternal understanding of infant diabetes risk. *Genetics in Medicine, 5(2), 77-83.*

Carmichael, S. K., Johnson, S. B., Fuller, K. G., Taylor, K. R., She, J. X., & Schatz, D. A. (1999). Newborn screening for type 1 diabetes: Maternal understanding of infant risk. *Diabetes, 48*, A322.

Carver, C. S., Scheier, M. F., & Weintraub, J. K. (1989). Assessing coping strategies: A theoretically based approach. *Journal of Personality and Social Psychology, 56*, 267-283.

Christie, M. R., Roll, U., Payton, M. A., Hatfield, E. C., & Ziegler, A. G. (1997). Validity of screening for individuals at risk for type I diabetes by combined analysis of antibodies to recombinant proteins. *Diabetes Care, 20*, 965-970.

Clarke, A. (1994). The genetic testing of children. Working Party of the Clinical Genetics Society (UK). *Journal of Medical Genetics, 31*, 785-797.

Classen, J.B. & Classen, D.C. (2001). Vaccines and the risk of insulin-dependent diabetes (IDDM): potential mechanisms of action. *Medical Hypotheses, 57*, 532-538.

Clavel-Chapelon, F., Joseph, R., & Goulard, H. (1999). Surveillance behavior of women with reported family history of colorectal cancer. *Preventative Medicine, 28*, 174-178.

Committee on Assessing Genetic Risk (Ed.). (1994). *Assessing Genetic Risks: Implications for Health and Social Policy*. Washington, D.C.: National Academy Press.

Couper, J.J. (2001). Environmental triggers of type 1 diabetes. Journal of Paediatrics and Child Health, 37, 218-222.

Croyle, R. T., & Lerman, C. (1999). Risk communication in genetic testing for cancer susceptibility. *Journal of the National Cancer Institute Monographs, 25*, 59-66.

Croyle, R. T., Smith, K. R., Botkin, J. R., Baty, B., & J., N. (1997). Psychological responses to BRCA1 mutation testing: Preliminary findings. *Health Psychology, 16*, 63-72.

Dahlquist, G., Bennich, S. S., & Kallen, B. (1996). Intrauterine growth pattern and risk of childhood onset insulin dependent (type 1) diabetes: Population based case-control study. *BMJ, 313*, 1174-1177.

Dahlquist, G., Blom, L., Holmgren, G., Hogglof, B., Larsson, Y., & Sterky, G. (1985). The epidemiology of diabetes in Swedish children 0 to 14 years - a six year prospective study. *Diabetologia, 28*, 802-808.

Diabetes Prevention Trial Study Group (1995). The Diabetes Prevention Trial (DPT-1): Implementation of screening and staging of relatives. *Transplant Processes, 27*, 3.

Diabetes Prevention Trial Study Group (2002). Effects of insulin in relatives of patients with type 1 diabetes mellitus. *New England Journal of Medicine, 346*, 1685-1691.

Diefenbach, M. A., Willer, D. M., & Daly, M. B. (1999). Specific worry about breast cancer predicts mammography use in women at risk for breast and ovarian cancer. *Health Psychology, 18*, 532-536.

Donovan, K. A., & Tucker, D. C. (2000). Knowledge about genetic risk for breast cancer and perceptions of genetic testing in a sociodemographically diverse sample. *Journal of Behavioral Medicine, 23*, 15-36.

Dorman, J. S., McCarthy, B. J., O'Leary, L. A., & Koehler, A. N. (1995). Risk factors for insulin-dependent diabetes. In M. I. Harris, P. H. Bennett, E. J. Boyko, & e. al. (Eds.), *Diabetes in America* (pp. 165-173). Bethesda, MD: National Institutes of Health.

Epstein, S. A., Hsiang Lin, T., Audrain, J., Stefanek, M., Rimer, B., Lerman, C., & Consortium, T. H. R. B. C. (1997). Excessive breast self-examination among first-degree relatives of newly diagnosed breast cancer patients. *Psychosomatics, 38*, 253-261.

Epstein, S. A., & Lerman, C. (1997). Excessive health behaviors in those at risk for physical disorder. *Journal of Psychosomatic Research, 43*, 223-225.

Fenton-May, J. (1994). Screening for Duchenne muscular dystrophy. *Archives of Disease in Childhood, 70*, 551-552.

Fisher, E. B., Walker, E. A., Bostrom, A., Fischoff, B., Haire-Joshu, D., & Johnson, S. B. (2002). Behavioral science research in the prevention of diabetes: Status and opportunities. *Diabetes Care, 25*, 599-606.

Fletcher, B., Gulanick, M., & Lamendola, C. (2002). Risk factors for type 2 diabetes mellitus. *Journal of Cardiovascular Nursing, 16*, 17-23.

Florida Office of Economic and Demographic Research (2001). Available at: http://www.state.fl.us/edr/population. Last accessed 8/02.

Folkman, S., & Lazarus, R. (1980). An analysis of coping in a middle-ages community sample. *Journal of Health and Social Behavior, 21*, 219-239.

Forsyth, L.H. & Goetsch, V.L. (1997). Perceived threat of illness and health protective behaviors in offspring of adults with non-insulin-dependent diabetes mellitus. Behavioral Medicine, 23, 112-119.

Friedman Ross, L. (2003). Minimizing risks: the ethics of predictive diabetes mellitus screening research in newborns. *Archives of Pediatric and Adolescent Medicine, 157*, 89-95.

Frets, P. G., Duivenvoorden, H. J., Verhage, F., Niermeijer, M. F., van de Berge, S. M., & Galjaard, H. (1990). Factors influencing the reproductive decision after genetic counseling. *American Journal of Medical Genetics, 35*, 496-502.

Galatzar, A., Green, E., Ofan, R., Benzaquen, H., Yosefsberg, A., Wentrob, N., Karp, M., & Vardi, P. (2001). Psychological impact of islet cell antibody screening. *Journal of Endocrinology and Metabolism, 14*, 675-679.

Gerstein, H. C. (1994). Cow's milk exposure and type 1 diabetes mellitus: A critical review of the clinical literature. *Diabetes Care, 17*, 13-19.

Hailey, B. J. (1991). Family history of breast cancer and screening behavior: An inverted U-shaped curve? *Medical Hypotheses, 36*, 397-403.

Hailey, B. J., Carter, C. L., & Burnett, D. R. (2000). Breast cancer attitudes, knowledge, and screening behavior in women with and without a family history of breast cancer. *Health Care for Women International, 21*, 701-715.

Hendrieckx, C., DeSmet, F., Kristoffersen, I., & Bradley, C. (2002). Risk assessment for developing type 1 diabetes: Intentions of behavioural changes prior to risk notification. *Diabetes/Metabolism Research and Reviews, 18*, 36-42.

Horne, R., & Weinman, J. (1998). Predicting treatment adherence: An overview of theoretical models. In L. B. Meyers & K. Midence (Eds.), *Adherence to treatment in medical conditions* (pp. 25-50). Amsterdam, Netherlands: Harwood Academic.

Horowitz, M., Sundlin, E., Zanko, A., & Lauer, R. (2001). Coping with the grim news from genetic testing. *Psychosomatics, 42*, 100-105.

Horowitz, M., Wilner, N., & Alvarez, W. (1979). Impact of Events Scale: A measure of subjective stress. *Psychosomatic Medicine, 41*, 209-218.

Hughes, C., Gomez-Caminaero, A., Benkendorf, J., Kerner, J., Issacs, C., Barter, J., & Lerman, C. (1997). Ethnic differences in knowledge and attitudes about BRCA1 testing in women at increased risk. *Patient Education and Counseling, 32*, 51-62.

Hughes, C., Lerman, C., & Lustbader, E. (1996). Ethnic differences in risk perception among women at increased risk for breast cancer. *Breast Cancer Research and Treatment, 40*, 25-35.

Hunt, K., Emslie, C., & Wattm G. (2001). Barriers rooted in biography: how interpretations of family patterns of heart disease and early life experiences may undermine behavioural change in mid life. In: Graham, H., ed. *Understanding health inequalities.* Oxford: Oxford University Press.

Hypponen, E., Kenward, M. G., Virtanen, S. M., Pitulainen, A., Virta-Autio, P., Tuomilehto, J., Knip, M., & Akerblom, H. K. (1999). Infant feeding, early weight gain, and risk of type 1 diabetes. Childhood Diabetes in Finland (DiME) Study Group. *Diabetes Care, 22*, 1961-1965.

Institutes of Medicine (1994). *Assessing Genetic Risks: Implications for Health and Social Policy.* Washington, DC: National Academy Press.

Janis, I. L., & Feshback, S. (1953). Effect of fear-arousing communications. *Journal of Abnormal and Social Psychology, 48*, 78-92.

Jeffords, J. M., & Daschle, T. (2001). Policy issues: Political issues in the genome era. *Science, 291*, 1249-1251.

Johnson, S. B. (2001). Screening programs to identify children at risk for diabetes mellitus: Psychological impact on children and parents. *Journal of Pediatric Endocrinology and Metabolism, 14*, S653-659.

Johnson, S. B. (2002, May). Participant Experiences in the DPT-1: Preliminary Results. Invited Presentation to NIH Type 1 TrialNet Study Group. Bethesda, MD.

Johnson, S.B., Baughcum, A.E., Carmichael, S.K., She, J., & Schatz, D.A. (submitted) Maternal anxiety associated with newborn genetic screening for type 1 diabetes. *Diabetes Care.*

Johnson, S. B., & Carmichael, S. K. (2000). At-risk for diabetes: Coping with the news. *Journal of Clinical Psychology in Medical Settings, 7*, 69-78.

Johnson, S. B., Riley, W. J., Hansen, C. A., & Nurick, M. A. (1990). Psychological impact of islet-cell antibody screening. Preliminary results. *Diabetes Care, 13*, 93-97.

Johnson, S. B., & Tercyak, K. P. (1995). Psychological impact of islet cell antibody screening for IDDM on children, adults and their family members. *Diabetes Care, 18*, 1370-1372.

Juengst, E. T. (1995). The ethics of prediction: genetic risk and the physician-patient relationship. *Genome Science and Technology, 1*, 21-36.

Karges W., Hammond-McKibben D., Cheung R.K., Visconti M., Shibuya N., Kemp D., Dosch H.M. (1997). Immunological aspects of nutritional diabetes prevention in NOD mice: a pilot study for the cow's milk-based IDDM prevention trial. *Diabetes, 46*, 557-564.

Kash, K. M., Holland, J. C., Halper, M. S., & Miller, D. G. (1992). Psychological distress and surveillance behaviors in women with a family history of breast cancer. *Journal of the National Cancer Institute, 84*, 24-30.

Kessler, S. (1989). Psychological aspects of genetic counseling: VI. A critical review of the literature dealing with education and reproduction. *American Journal of Medical Genetics, 34*, 340-353.

Knip, M. (1998). Prediction and prevention of type 1 diabetes. *Acta Pediatrica, 425*, S54-62.

Kovacs, M., Finkelstein, R., Feinberg, T. L., Crouse, -. N., M., Paulaskas, S., & Pollock, M. (1985). Initial psychologic responses of parents to the diagnosis of insulin-dependent diabetes mellitus in their children. *Diabetes Care, 8*, 568-575.

Kupila, A., Muona, P., Simell, T., Arvilommi, P., Savolainen, H., Hamalainen, A.M., Korhonen, S., Kimpimaki, T., Sjoroos, M., Ilonen, J., Knip, M., & Simell, O. (2001). Feasibility of genetic and immunological prediction of type 1 diabetes in a population-based birth cohort. *Diabetologia, 44*, 290-297.

Kyvik, K. O., Green, A., & Beck-Nielson, H. (1995). Concordance rates of insulin dependent mellitus: A population based study of young Danish twins. *BMJ, 311*, 913-917.

Landin-Olsson, M., Palmer, J. P., Lernmark, A., Blom, L., Sundkvist, G., Nystrom, L., & Dahlquist, G. (1992). Predictive value of islet cell and insulin autoantibodies for type 1 (insulin-dependent) diabetes mellitus in a population-based study of newly-diagnosed diabetic and matched control children. *Diabetologia, 35*, 1068-1073.

Laporte, R. E. (1995). Patterns of disease: Diabetes mellitus and the rest. *BMJ, 310*, 545-546.

Lazarus, R. S., & Folkman, S. (1984). *Stress, appraisal, and coping*. New York: Springer.

Lerman, C. (1997). Psychological aspects of genetic testing: Introduction to the special issue. *Health Psychology, 16*, 3-7.

Lerman, C., Croyle, R. T., Tercyak, K. P., & Hamann, H. (2002). Genetic testing: Psychological aspects and implications. *Journal of Clinical and Consulting Psychology, 70*, 784-797.

Lerman, C., Daly, M., Masny, A., & Balshem, A. (1994a). Attitudes about genetic testing for breast-ovarian cancer susceptibility. *Journal of Clinical Oncology, 12*, 843-850.

Lerman, C., Daly, M., Sands, C., Balshem, A., Lustbader, E., Heggan, T., Goldstein, L., James, J., & Engstrom, P. (1993). Mammography adherence and psychological distress among woman at risk for breast cancer. *Journal of the National Cancer Institute, 85*, 1074-1080.

Lerman, C., Hughes, C., Croyle, R. T., Main, D., Durham, C., Snyder, C., Bonney, A., Lynch, J. F., Narod, S. A., & Lynch, H. T. (2000). Prophylactic surgery decisions and surveillance practices one year following BRCA 1/2 testing. *Preventive Medicine, 31*, 75-80.

Lerman, C., Kash, K., & Stefanek, M. (1994b). Younger women at increased risk for breast cancer: Perceived risk, psychological well-being, and surveillance behavior. *Monographs of the National Cancer Institute, 16*, 171-176.

Lerman, C., Marshall, J., Audrain, J., & Gomez-Caminero, A. (1996a). Genetic testing for colon cancer susceptibility: Anticipated reactions of patients and challenges to providers. *International Journal of Cancer, 69*, 58-61.

Lerman, C., Narod, S., Schulman, K., Hughes, C., Gomez-Caminero, A., Bonney, G., Gold, K., Trock, B., Main, D., Lynch, J., Fulmore, C., Snyder, C., Lemon, S. J., Conway, T., Tonin, P., Lenoir, G., & Lynch, H. (1996b). BRCA1 testing in families with hereditary breast-ovarian cancer: A prospective study of patient decision making and outcomes. *JAMA, 26*, 1885-1892.

Lerman, C., Rimer, B., Trock, B., Balshem, A., & Engstrom, P. F. (1990). Factors associated with repeat adherence to breast cancer screening. *Preventive Medicine, 19*, 279-290.

Lerman, C., & Schwartz, M. (1993). Adherence and psychological adjustment among women at high risk for breast cancer. *Breast Cancer Research and Treatment, 28*, 145-155.

Lerman, C., Schwartz, M. D., Lin, T. H., & Hughes, C. (1997). The influence of psychological distress on use of genetic testing for cancer risk. *Journal of Consulting and Clinical Psychology, 65*, 414-420.

Lerman, C., Schwartz, M. D., Miller, S. M., Daly, M., Sands, C., & Rimer, B. K. (1996c). A randomized trial of breast cancer risk counseling: interacting effects of counseling, educational level, and coping style. *15*(75-83).

Lerman, C., Trock, B., Rimer, B. K., Jepson, C., Brody, D., & Boyce, A. (1991). Psychological side effects of breast cancer screening. *Health Psychology, 10*, 259-267.

Lerman, C. E., & Rimer, B. K. (1993). Psychosocial impact of cancer screening. *Oncology, 7*, 67-72.

Leventhal, H. (1970). Findings and theory in the study of fear communications. *Advances in Experimental and Social Psychology, 5*, 119-186.

Leventhal, H., Safer, M. A., & Panagis, D. M. (1983). The impact of communications on the self-regulation of health beliefs, decisions, and behavior. *Health Education Quarterly, 10*, 3-29.

Leventhal, H., Singer, R., & Jones, S. (1965). Effects of fear and specificity on recommendation upon attitudes and behavior. *Journal of Personality and Social Psychology, 2*, 20-29.

Lucidarme, N., Dominues-Muriel, W., castro, D., Czernichow, P., & Levy-Marchal. (1998). Appraisal and implications of predictive testing for insulin-dependent diabetes mellitus. *Diabetes and Metabolism, 24*, 550-553.

Ludvigsson, J., Gustafsson-Stolt, U., Liss, P., & Svensson, Y. (2002). Mothers of children in ABIS, a population-based screening for prediabetes, experience few ethical conflicts and have a positive attitude. *Annals of New York Academy of Science, 958*, 376-381.

Ludwick-Rosenthal, R., & Neufeld, R. W. (1993). Preparation for undergoing an invasive medical procedure: Interacting effects of information and coping style. *Journal of Consulting and Clinical Psychology, 61*, 156-164.

Lynch, H. T., Watson, P., Conway, T. A., Lynch, J. F., Slominski-Caster, S. M., Narod, S. A., Feunteun, J., & Lenoir, G. (1993). DNA screening for breast/ovarian cancer susceptibility based on linked markers. *Archives of Internal Medicine, 153*, 1979-1987.

Lynch, H. T., Watson, P., Tinley, S., Snyder, C., Durham, C., Lynch, J., Y., K., Serova, O., Lenoir, G., Lerman, C., & Naord, S. A. (1999). An update on DNA-based BRCA1/BRCA2 genetic counseling in hereditary breast cancer. *Cancer Genetics and Cytogenetics, 109*, 91-98.

Marteau, T. M., & Bekker, H. (1992). The development of a six-item short-form of the state scale of the Speilberger State-Trait Anxiety Inventory (STAI). *British Journal of Clinical Psychology, 31*, 301-306.

Marteau, T. M., & Croyle, R. T. (1998). Psychological responses to genetic testing. *BMJ, 316*, 693-696.

Marteau, T. M., Dundas, R., & Axworthy, D. (1997). Long-term cognitive and emotional impact of genetic testing for carriers of cystic fibrosis. *Health Psychology, 16*, 51-62.

Marteau, T. M., & Lerman, C. (2001). Genetic risk and behavioural change. *BMJ, 322*, 1056-1059.

Marteau, T. M., & Michie, S. (1995). A positive screening test for an untreatable condition provides psychological relief from uncertainty. *BMJ, 11*, 58-59.

McCaul, K. D., Schroeder, D. M., & Reid, P. A. (1996). Breast cancer worry and screening: Some prospective data. *Health Psychology, 16*, 430-433.

McKinney, P. A., Okasha, M., Parslow, R. C., Law, G. R., gurney, K. A., Williams, R., & Bodansky, H., J. (2000). Early social mixing and childhood type 1 diabetes: A case control study in Yorkshire, UK. *Diabetic Medicine, 17*, 236-242.

McNeil, T. F., Thelin, T., Aspergren-Jansson, E., & Sveger, T. (1986). Identifying children at high somatic risk possible long-term effects on parents' attitudes and feelings associated with the child. *Acta Psychiatrica Scandinavica, 74*, 341-346.

Meiser, B., Butow, P., Friedlander, M., Schnieden, V., Gattas, M., Kirk, J., Suthers, G., Haan, E., & Tucker, K. (2000). Intention to undergo prophylactic mastectomy in women at increased risk of developing hereditary breast cancer. *Journal of Clinical Oncology, 18*, 2250-2257.

Meyer, D., Leventhal, D., & Gutmann, M. (1985). Common models of illness: The example of hypertension. *Health Psychology, 4*, 115-135.

Miller, S. M. (1987). Monitoring and blunting: Validation of a questionnaire to assess styles of information seeking under threat. *Journal of Personality and Social Psychology, 52*, 345-353.

Miller, S. M., Leinbach, A., & Brody, D. S. (1989). Coping style in hypertensive patients: Nature and consequences. *Journal of Consulting and Clinical Psychology, 57*, 333-337.

Miller, S. M., & Managan, C. (1983). Interacting effect of information and coping style in adapting to gynecologic stress: Should the doctor tell all? *Journal of Personality and Social Psychology, 45*, 223-236.

National Diabetes Data Group (1995). *Diabetes in America* (2nd ed.). Bethesda, MD: National Institutes of Health.

Nejentsev, S., Sjoroos, M., Soukka, T., Knip, M., Simell, O., Lovgren, T., & Ilonen, J. (1999). Population-based genetic screening for the estimation of Type 1 diabetes mellitus risk in Finland: selective genotyping of markers in the HLA-DQB1, HLA-DQA1 and HLA-DRB1 loci. *Diabetic Medicine, 16*, 985-992.

Palmer, S. C., Kagee, A., Kruus, L., & Coyne, J. C. (2002). Overemphasis of psychological risks of genetic testing may have "dire" consequences. *Psychosomatics, 43*, 86-88.

Parsons, E. P., Clarke, A. J., Hood, K., Lycett, E., & Bradley, D. M. (2002). Newborn screening for Duchenne muscular dystrophy. *Archives of Disease in Childhood, 86*, F91-95.

Patenaude, A. F. (2003). Pediatric psychology training and genetics: What will twenty-first-century pediatric psychologists need to know. *Journal of Pediatric Psychology, 28*, 135-145.

Patenaude, A. F., Guttmacher, A. E., & Collins, F. S. (2002). Genetic testing and psychology: New roles, new responsibilities. *American Psychologist, 57*, 271-282.

Pierce, M., Hayworth, J., Warburton, F., Keen, H., & Bradley, C. (1999). Diabetes mellitus in the family: perceptions of offspring's risk. *Diabetic Medicine, 16*, 431-436.

Pierce, M., Keen, H., & Bradley, C. (1995). Risk of diabetes in offspring of parents with non-insulin dependent diabetes. *Diabetes Medicine, 12*, 6-13.

Rakowski, W., Assaf, A.R., Lefebvre, R.C., Lasater, T.M., Niknian, M., Carleton, R.A (1990). Information-seeking about health in a community sample of adults: correlates and associations with other health-related practices. *Health Education Quarterly, 17*, 379-93.

Rewers, M., Bugawan, T. L., Norris, J. M., Blair, A., Beaty, B., Hoffman, M., McDuffie, R. S., Hamman, R. F., Klingensmith, G., Eisenbarth, G. S., & Erlich, H. A. (1996). Newborn screening for HLA markers associated with IDDM: Diabetes Autoimmunity Study in the Young (DAISY). *Diabetologia, 39*, 807-812.

Riley, W. J., Atkinson, M. A., Schatz, D. A., & Maclaren, N. K. (1990). Comparison of islet autoantibodies in "pre-diabetes" and recommendations for screening. *Journal of Autoimmunity, 3*, 47-51.

Ritvo, P., Irvine, J., Robinson, G., Brown, L., Murphy, K. J., Matthew, A., & Rosen, B. (2002). Psychological adjustment to familial-genetic risk assessment for ovarian cancer: Predictors of nonadherence to surveillance recommendations. *Gynecologic Oncology, 84*, 72-80.

Rolland, J. S. (1994). *Families, illness, and disability: An integrative treatment model*. New York: Basic Books.

Rolland, J. S. (1999). Families and genetic fate: A millennial challenge. *Families, Systems, and Health, 17*, 123-132.

Rosenstock, I. M. (1974). The Health Belief Model and preventative health behaviors. *Health Education Monographs, 2*, 254-386.

Rowley, P. T., Loader, S. L., Sutera, C. J., Walder, M., & Kozyra, A. (1991). Prenatal screening of hemogloibnopathies III. Applicability of the Health Belief Model. *American Journal of Human Genetics, 48*, 452-459.

Ryan, D.H. Diabetes Prevention Program Research Group. (2003). Diet and exercise in the prevention of diabetes. *International Journal of Clinical Practice, 134*, 28-35.

Sagi, M., Shiloh, S., & Cohen, T. (1992). Applications of the Health Belief Model in a study of parents' intentions to utilize prenatal diagnosis of cleft lip and palate. *American Journal of Medical Genetics, 44*, 326-333.

Schatz, D., Krishner, J., & She, J. (2002). To screen or not to screen for pre-type 1 diabetes? *Hormone Research, 57*, 15-17.

Schatz, D. A., Krischer, J., Horne, G., Riley, W. J., Atkinson, M. S., Winter, W. E., Silverstein, J. H., Allen, L., Shah, S., Malone, J., & Maclaren, N. K. (1994). Islet cell antibodies predict insulin dependent diabetes in U.S. School age children as powerfully as in unaffected relatives. *Journal of Clinical Investigations, 93*, 2403-2407.

Schatz, D. A., & Maclaren, N. K. (1996). Cow's milk and insulin-dependent diabetes mellitus: Innocent until proven guilty. *JAMA, 276*, 647-648.

Schatz, D. A., Muir, A., fuler, K., Atkinson, M., Crockett, S., Hsiang, H., Winter, B., Ellis, T., Taylor, K., Saites, C., Dukes, M., Fang, Q., Clare-Salzer, M., & She, J. (2000). Prospective assessment in newborns for diabetes autoimmunity (PANDA): A newborn screening program in the general population of Florida. *Diabetes, 49*, 67A.

Scheier, M. F., Weintraub, J. K., & Carver, C. S. (1986). Coping with stress: Divergent strategies of optimists and pessimists. *Journal of Personality and Social Psychology, 51*, 1257-1264.

Schenker, M., Hummel, M., Ferber, K., Walter, M., Keller, E., Albert, E. D., & Janka, H. U. (1999). Early expression and high prevalence of islet autoantibodies for DR3/4 heterozygous and DR4/4 homozygous offspring of parents with type 1 diabetes: the German BABYDIAB study. *Diabetologia, 42*, 671-677.

Schoenfeld, M., Myers, R. H., Berkman, B., & Clark, E. (1984). Potential impact of a predictive test on the gene frequency of Huntington disease. *American Journal of Medical Genetics, 18*, 423-429.

Schwartz, M. D., Peshkin, B. N., Hughes, C., Main, D., Isaacs, C., & Lerman, C. (2002). Impact of BRCA1/BRCA2 mutation testing on psychological distress in a clinic-based sample. *Journal of Clinical Oncology, 20*, 514-520.

Schwartz, M. D., Taylor, K. L., Willard, K. S., Siegel, J. E., Lamdan, R. M., & Moran, K. (1999). Distress, personality, and mammography utilization among women with a family history of breast cancer. *Health Psychology, 18*, 315-326.

Senior, V., Marteau, T. M., & Peters, T. J. (1999a). Will genetic testing for predisposition for disease result in fatalism? A qualitative study of parents' responses to neonatal screening for familial hypercholestrolaemia. *Social Sciences Medicine, 48*, 1857-1860.

Senior, V., Marteau, T. M., & Weinman, J. (1999b). Impact of genetic testing on causal models of heart disease and arthritis: An analogue study. *Psychology and Health, 14*, 1077-1088.

Shaw, C., Abrams, K., & Marteau, T. M. (1999). Psychological impact of predicting individual's risk for illness: A systematic review. *Social Science Medicine, 49*, 1571-1598.

Shiloh, S., & Sagi, M. (1989). Effect of framing on the perception of genetic recurrence risks. *Psychological Health, 3*, 45-61.

Sorenson, J. R., Scotch, N. A., Swazey, J. P., Wertz, D. C., & Heeren, T. C. (1987). Reproductive plans of genetic counseling clients not eligible for prenatal diagnosis. *American Journal of Medical Genetics, 28*, 345-352.

Spielberger, C. D. (1973). *Test Manual for the State-Trait Anxiety Inventory for Children*. Palo Alto, CA: Consulting Psychologists.

Spielberger, C. D., Gorsuch, R. L., & Lushene, R. E. (1970). *STAI Manual*. Palo Alto, CA: Consulting Psychologist Press.

Tercyak, K. P., Lerman, C., Peshkin, B. N., Hughes, C., Main, D., Isaacs, C., & Schwartz, M. D. (2001). Effects of coping style and BRCA1 and BRCA2 test results on anxiety among women participating in genetic counseling and testing for breast and ovarian cancer risk. *Health Psychology, 20*, 217-222.

Tercyak, K. P., Streisand, R., Peshkin, B., & Lerman, C. (2000). Psychological impact of predictive testing for illness on children and families: Challenges for the new millennium. *Journal of Clinical Psychology in Medical Settings, 7*, 55-68.

Thernlund, G. M., Dahlquist, G., Hansson, K., Ivarsson, S. A., Ludvigsson, J., Sjobald, S., & Hagglof, B. (1995). Psychological stress and the onset of IDDM in children. *Diabetes Care, 18*, 1323-1329.

Tillil, H., & Kobberling, J. (1987). Age-corrected empirical genetic risk estimates for first degree relatives of IDDM patients. *Diabetes, 36*, 93-99.

Tuomilehto, J., Lindstrom, J., Eriksson, J. G., Valle, T. T., Hamalainen, H., Ilanne-Parikka, P., Leinanen-Kiukaanniemi, S., & Laakso, M. (2001). Prevention of type 2 diabetes mellitus by changes in lifestyle among subjects with impaired glucose tolerance. *New England Journal of Medicine, 344*, 1343-1350.

Vernon, S. W., Gritz, E. R., Peterson, S. K., Amis, C. I., Perz, C. A., Perz, C. A., Baile, W. F., & Lynch, P. M. (1997). Correlates of psychologic distress in colorectal cancer patients undergoing genetic testing for hereditary colon cancer. *Health Psychology, 16*, 73-86.

Virtanen, S. M., & Aro, A. (1994). Dietary factors in the aetiology of diabetes. *Annals of Medicine, 26*, 469-478.

Virtanen, S. M., Laara, E., Hypponen, E., Reijonen, H., Rasanen, L., Aro, A., Knip, M., Ilonen, J., & Akerblom, H. K. (2000). Cow's milk consumption, HLA-DQB1 genotype, and type 1 diabetes: A nested case-control study of siblings of children with diabetes. Childhood diabetes in Finland study group. *Diabetes, 49*, 912-917.

Vitaliano, P. P., Russo, Carr, J. E., Maiure, R. D., & Becker, J. (1985). The Ways of Coping Checklist: Revision and psychometric properties. *Multivariate Behavioral Research, 20*, 3-26.

Wald, N. J., & Morris, J. K. (1998). Neonatal screening for cystic fibrosis. *British Journal of Medicine, 316*, 404-405.

Wallston, K. (2000). Conceptualization and operationalization of perceived control. In A. Baum, T. A. Revenson, & J. E. Singer (Eds.), *Handbook of Health Psychology* . Mahwah (NJ): Lawrence Erlbaum.

Wardle, J. (1995). Women at risk for ovarian cancer. *Journal of the National Cancer Institute- Monographs, 17*, 81-85.

Waters, D. L., Wilcken, B., Irwig, L., VanAsperen, P., Mellis, C., Simpson, J. M., Brown, J., & Gaskin, K. (1999). Clinical outcomes from newborn screening for cystic fibrosis in New South Wales. *Archives of Diseases in Childhood, 80*, F1-7.

Weinstein, N.D. (1993) Testing four competing theories of health-protective behavior. *Health Psychology, 12*, 324-333.

Wertz, D. C. (1994). Genetic testing for children: Who decides? *JAMA, 272*, 875-881.

Wiggins, S., Whyte, P., Huggins, M., Adam, S., Theilmann, J., Bloch, M., Sheps, S. B., Schecter, M. T., & Hayden, M. R. (1992). The psychological consequences of predictive testing for Huntington's disease. *New England Journal of Medicine, 327*, 1401-5.

Wilcken, B., Towns, S. J., & Mellis, C. M. (1983). Diagnostic delay in cystic fibrosis: Lessons from newborn screening. *Archives of Diseases of Childhood, 58*, 863-866.

Wilcken, B., & Travert, G. (1999). Neonatal screening for cystic fibrosis: Present and future. *Acta Paediatrica Supplement, 431*, 33-35.

Yu, L., Robles, D. T., Abiru, N., Kaur, P., Rewers, M., Kelemen, K., & Eisenbarth, G. S. (2000). Early expression of antiinsulin autoantibodies of humans and the NOD mouse: Evidence for early determination of subsequent diabetes. *Proceedings of the National Academy of Sciences of the United States of America, 97*, 1701-1706.

Yu, M. S., Norris, J. M., Mitchell, C. M., Butler-Simon, N., Groshek, M., Follansbee, D., Erlich, H., Rewers, M., & Kingensmith, G. J. (1999). Impact on maternal parenting stress on receipt of genetic information regarding risk of diabetes in newborn infants. *American Journal of Medical Genetics, 86*, 219-226.

Zerres, K., & Stephan, M. (1986). Attitudes to early diagnosis of polycystic kidney disease. *Lancet, 13*, 1395.

Ziegler, A. G., Hummel, M., Schenker, M., & Bonifacio, E. (1999). Autoantibody appearance and risk for development of childhood diabetes in offspring of parents with type 1 diabetes: The 2-year analysis of the German BABYDIAB study. *Diabetes, 48*, 460-468.

BIOGRAPHICAL SKETCH

Amy E. Baughcum is currently in the doctoral program through the Department of Clinical and Health Psychology at the University of Florida, specializing in child clinical/pediatric psychology. Her primary research interests to date include the psychological impact of genetic screening, early childhood feeding practices, and childhood obesity. Her primary clinical interests are focused on working with chronically and terminally ill children and their families.

Ms. Baughcum completed her master's degree in clinical psychology in 2001 in the Department of Clinical and Health Psychology at the University of Florida. Her master's thesis was titled Maternal Feeding Practices and Child Overweight in Preschool Children.

Ms. Baughcum completed her bachelor's degree in psychology at Williams College, in Williamstown, Massachusetts in 1997. Ms. Baughcum is originally from the Washington, DC metropolitan area.

In the fall of 2003, Ms. Baughcum began her clinical internship year specializing in pediatric psychology at Columbus Children's Hospital in Columbus, Ohio. After internship, she plans to complete a postdoctoral fellowship and pursue an academic career as a pediatric psychologist.

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy.

Suzanne Bennett Johnson, Chair
Professor
Medical Humanities and Social Sciences
Florida State University, Tallahassee

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy.

Alexandra L. Quittner, Cochair
Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy.

Samuel F. Sears
Associate Professor of Clinical and Health Psychology

I certify that I have read this study and that in my opinion it conforms to acceptable standards of scholarly presentation and is fully adequate, in scope and quality, as a dissertation for the degree of Doctor of Philosophy.

Fonda D. Eyler
Professor of Psychology

This dissertation was submitted to the Graduate Faculty of the College of Agricultural and Life Sciences and to the Graduate School and was accepted as partial fulfillment of the requirements for the degree of Doctor of Philosophy.

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Dean, College of Health Professions

Dean, Graduate School