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THE INFLUENCE OF FAMILIAL OBLIGATION AND SOCIAL SUPPORT MOTIVES IN DECISIONS TO DISCLOSE GENETIC TEST RESULTS

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ABSTRACT

Genetics differs from other branches of medicine because of the omnipresent and substantial role familial obligation plays throughout the entire genetic testing and disclosure decision-making process. Genetic information may be considered unique from other kinds of health information because it may reveal personal information about one's likelihood of certain medical conditions and information about one's genetic relatives (Annas, Glantz, & Roche, 1995). This thesis uses the disclosure decision-making model (DD-MM, Greene, 2009) as the framework to investigate the factors that predict young adults' decision to disclose genetic test results to a family member. Notably, the DD-MM has never been tested with genetic disclosures. Further, I challenge the assumptions in the DD-MM (and other existing models of decision disclosures) that disclosures are driven solely by self-interested motives, which is predominantly considered as the desire to seek social support. I propose that familial obligation also motivates disclosure, and investigate how a person's reason for disclosure—wanting social support versus familial obligation due to risk relevance— affects the disclosure decision-making process. One hundred seventy-three (N =173) young adults were recruited to report on their hypothetical disclosure experience regarding the genetic health condition, alpha-1 antitrypsin deficiency (AATD). Participants were asked to imagine that they had undergone diagnostic testing and learned that they had a genetic mutation associated with AATD. They were then asked to specify a family member to refer to when completing the scales to measure DD-MM variables, which are used to predict their likelihood of sharing their results with a genetic relative. Participants were asked to report on the degree to which their disclosure decision is motivated by social support and familial obligation. Results indicate that the DD-MM translates to the genetic context and significantly predicts disclosure likelihood. Specific key mechanisms (i.e., a more serious prognosis and more anticipated instrumental support) were significant factors determining disclosure likelihood in the full model, but changed when sub-samples were created based on motive for disclosure. Results show familial obligation is present as a reason for disclosure and that social support does not fully explain the results of the DD-MM.

Table of Contents

LIST OF FIGURES

LIST OF TABLES

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Chapter 1

Literature Review

Mclean (1998) describes a mother and daughter fidgeting nervously in the waiting room of a genetic testing center; one is waiting to be tested for Huntington's disease (HD) while the other refuses to know. It is reasonable to suspect that it was the mother preparing to be tested, taking precautions for herself and her child. Instead, it is the daughter that wants to know "if I'm going to wind up like Uncle Harry. I want to know the chances that my children will inherit Huntington's disease" (para. 1) for when she and her husband start a family. The mother, on the other hand, while regretting that she may be the one causing her daughter's potential diagnosis, has the mentality of "So far, so good…I'm only 42. I want to live my life and make my decisions without a Huntington's diagnosis hanging over my head" (para. 3). This situation is complicated by how HD is passed on. Because HD is a dominant genetic disease (Sturrock & Leavitt, 2010), if the daughter is found to carry the gene for Huntington's disease, her mother must also have the gene. The daughter, then, will know what her mother does not want to know. Mclean ends the article with a simple question: what should the daughter do?

Prior to the Human Genome Project's completion in 2003, people had no way of knowing whether they were a carrier for a genetic condition until symptoms actually appeared (Mclean, 1998). Since the Project's completion, genetic tests a) have become more widely available b) been used for both predictive and diagnostic purposes and c) applied as a means of practicing personalized medicine (Burke & Zimmern, 2004; Evans, Skrzynia, & Burke, 2001; Guttmacher, Porteous, McInerny, 2007; Smith, Parrott, Wienke, Greenberg & Chesnut, in progress). According to Guttmacher et al. (2007), "genomics based knowledge and tools promise

the ability to approach each patient as the biological person he or she is, thereby radically changing our [medical] paradigms" (p.151). Genetic tests offer the promise of confirming suspected diagnoses, predicting the likelihood of future illnesses, detecting the presence of carrier states in unaffected individuals (whose children may be at risk), and predicting responses to therapy (genome.gov, 2013). Therefore, the decision to disclose one's genetic information is a powerful one, as failure to do so is potentially life-threatening (Klitzman, 2009). The implications genetic information has for blood-related family members in particular, raises questions on the legitimacy of the decision to disclose/not disclose genetic information to genetic relatives (Bell & Bennett, 2001; Laurie, 1999; Skene, 1998).

Genetics differs from other branches of medicine because of the omnipresent and substantial role familial obligation plays throughout the entire genetic testing and disclosure decision-making process. Genetic information may be considered unique from other kinds of health information because it may reveal personal information about one's likelihood of certain medical conditions and information about one's genetic relatives (Annas, Glantz, & Roche, 1995). Indeed, scholars (e.g., Hallowell et al., 2003) argue that the familial nature of genetic information is what distinguishes it from other types of medical information. While the family-focus of genetics is recognized in medicine, research on how this distinguishing factor affects peoples' decision to disclose genetic test results has not been considered. This thesis attempts to address this omission.

In this thesis, I will investigate the disclosure decision-making model (DD-MM; Greene, 2009) in the context of sharing genetic test results. Further, I challenge the assumptions in the DD-MM (and other existing models on disclosure decision-making) that disclosures are driven solely by self-interested motives, which is predominantly considered as the desire to seek social support. I propose that familial obligation also motivates disclosure, and investigate how a

person's reason for disclosure—wanting social support versus familial obligation due to risk relevance— affects the disclosure decision-making process.

My thesis is organized in this way. First, I will review the literature on health information disclosure and define key terms. Second, I will describe the genetic condition Alpha-1 Antitrypsin Deficiency, which I have selected as the genetic condition for participants to consider disclosing to a family member. Third, I review the disclosure decision-making model (DD-MM; Greene, 2009). Fourth, I will discuss issues related to disclosure of genetic test results, and the motivations and barriers of disclosing genetic information to family members

Health Information Disclosures

Disclosure research has received considerable attention (see Cozby, 1973; Greene, Derlega, & Mathews, 2006). Disclosure is defined as the process of revealing (or concealing) private information (e.g., Greene et al., 2006; Kelly, 2002; Petronio, 2002). People may reveal private information through both verbal (e.g. "I have…" or "I am considering…" (Chelune, 1979; Cozby, 1973; Derlega & Grzelak, 1979; Derlega, Metts, Petronio, & Margulis, 1993; Greene, Derlega, Yep & Petronio, 2003; Petronio, 2002; Greene, 2009; Omarzu, 2000) and nonverbal behavior (e.g., clothing choice, facial expressions, dancing; Derlega, Winstead, Mathews, & Braitman, 2008; Oster & Gould, 1987). Derlega & Grzelak (1979) elaborate on the concept of personal self-disclosure and explain that it "…includes any information exchange that refers to the self, including personal states, dispositions, events in the past, and plans for the future. It can be objectively defined as any verbal message that formally begins with the word "I" (for instance, "I think," "I feel") or any other verbal message about the self." (p.152). As an information exchange, disclosures are considered a form of interpersonal communication (Greene et al., 2003), which may be intentional (Emlet, 2008), unintentional (Emlet, 2008), and/or indirectly made (e.g., through a third party; Greene, 2009). Notably, disclosers can provide mixed messages

to those they choose to disclose to, despite the disclosure being deliberate and intentional (Greene et al., 2003). Drawing from Greene and colleagues (2006), in this thesis, I define genetic self-disclosures as a voluntary, intentional, deliberate decision to reveal, verbally and/or nonverbally, genetic test results to a blood relative who does not know about the information within an interpersonal, dyadic encounter.

In intentional disclosures, the discloser is in control (Derlega et al., 1993; Greene, 2009; Greene et al., 2006). Those disclosing intentionally are thought to calculate how much they want to tell, when they want to tell it, and to whom (Greene et al., 2003; Petronio, 2002). For those that are privileged to the private information (i.e., target information recipients), their access is regulated by the discloser, either by giving complete access through a full disclosure or giving partial access by selectively disclosing (Kelly, 2002). The deliberate and strategic act of disclosing is often considered voluntary for these reasons (Greene et al., 2003; Greene, 2009; Jourard, 1971).

The voluntary, intentional, deliberate decision to disclose is also the basis for why current models assumed disclosures to be goal-driven (Chaudoir & Fisher, 2010; Davis & Franzoi, 1987; Greene et al., 2006; Omarzu, 2000). How disclosure messages are presented is an important feature of self-disclosure; it allows people to achieve their goal (Greene et al., 2006). The goals in current theoretical models include managing uncertainty (Afifi & Weiner, 2004; Greene, 2009) and attaining social benefits via social support (e.g., self-expression, self-clarification, social validation, relationship development; Altman & Taylor, 1973; Derlega & Chaiken, 1977; Derlega & Grzelak, 1979; Jones & Archer, 1976).

Outcomes of Health Self-Disclosures

Positive outcomes. Perhaps as a function of disclosure being a goal-driven activity, research demonstrates consistent associations between health self-disclosure and positive health

(Derlega et al., 1993; Frattaroli, 2006) and psychological outcomes (Kelly, Klusas, von Weiss, & Kenny, 2001; Pennebaker, 1984). For example, researchers found that verbally discussing or writing about extremely traumatic or upsetting life experiences, which could include a serious health diagnosis, (as opposed to trivial events) is positively associated with lower illness rates (Pennebaker & O'Heeron, 1984), fewer physician visits (Pennebaker, Colder, & Sharp, 1990; Pennebaker, Kiecolt-Glaser, & Glaser, 1988), less immune dysfunction (Pennebaker et al., 1988), and decreased severity of physical symptoms (Kelley, Lumley, & Leisen, 1997). Psychologically, disclosing health information is associated with better psychological adjustment (Lichtman, Taylor, & Wood, 1987), enhanced social and emotional adjustment, and self-esteem (Zemore & Shepel, 1989; Goldsmith, Bute, & Lindholm, 2007). Disclosing can also be cathartic as an emotional release from unburdening one's self from carrying difficult and painful emotional experiences alone (Altman & Taylor, 1973; Greene et al., 2003; Derlega, Winstead, & Folk-Barron, 2000; Derlega, Winstead, Greene, Serovich, & Elwood, 2004; Derlega et al., 2008) and a means of attaining social support and advice (d'Agincourt-Canning, 2001; Derlega, Lovejoy, & Winstead, 1998; Derlega et al., 2004; 2008; Foster, Eeles, Arden-Jones, Moynihan, & Watson, 2004). Thus, sharing personal health information can be rewarding for the discloser.

Negative outcomes. Despite the potential rewards associated with goal achievement via health disclosure, individuals are challenged physically and/or emotionally when initially diagnosed with an illness and as they grapple with the responsibility of managing their health information (Greene et al., 2012). People may experience a range of emotions, including uncertainty, fear, helplessness, and anxiety as they attempt to grapple with their disease-related thoughts and evaluate their self-efficacy about disease self-management (Braitman et al., 2008; Epstein & Street, 2007). These experiences are only heightened when considering to disclose to others or to conceal information. Goffman (1963) described how people with stigmatized

conditions struggle with their personal risk of being "known" and seek to manage the information in order to avoid a negative reaction. The fear of this social rejection can lead to isolation.

Outcomes Associated with Genetic Self-Disclosures

Disclosing genetic test results is associated with some of the same outcomes of general health disclosures. Disclosing genetic test results has been associated with gaining control over one's life and enabling, in the sense that people have reported feeling able to accomplish their goals of helping their selves and other family members to take actions to protect themselves (d'Agincourt-Canning, 2001). Unfortunately, for those receiving genetic test results, genetic testing is not always seen as empowering, but more as a Pandora's box, opening a person to the perceived stigmas related to genetic conditions and genetic discrimination. For example, stigma associated with genetic conditions around life expectancy, lifestyle choices, or decisions about having children are well noted (The Nuffield Council, 2006). These stigmas have been attributed to a lack of understanding and/or misunderstanding of the genetic risk of developing diseases (The Nuffield Council, 2006). As such, the most commonly expressed fear of genetic disclosure is that test results will be used in ways that could cause harm—for example, being denied access to health insurance, employment, education, and loans (Clayton, 2003; Murray & Livny, 1995; Wertz & Fletcher, 1991). As with other health conditions (Smith, 2011), the fear of stigmatization may affect the person diagnosed with the condition and their families. If the diagnosed person chooses not to reveal their diagnosis to others, then, due to the inherent nature of genetic conditions, their genetic relatives may not be aware or prepared for their potential diagnoses. In contrast, those that decide to manage their personal and potentially stigmatizing information by disclosing are putting their blood-related family members at harm of also being stigmatized and/or discriminated against.

Understanding Genetic Self-Disclosures

As just described, there are risks and rewards associated with disclosing one's genetic information (Gallo, Angst, & Knafl, 2009). The current models used to understand health disclosures consider the tested person as an independent decision-maker, judiciously weighing the pros and cons of each treatment decision (d'Agincourt-Canning, 2006) and balancing risks with rewards of disclosing (e.g., Greene et al., 2006; Petronio, 2002). There are many models focused on disclosure decision-making (Chaudoir & Fisher, 2010; Derlega & Grzelak, 1979; Greene et al., 2006; Omarzu, 2000). These models are grounded in social exchange and interdependence theories (Kelley, 1979), which posits that disclosers consider reasons for and against disclosure in order to assess the subjective value of self-disclosure (Derlega et al., 2008; Greene, 2009; Kelly, 2002; Omarzu, 2000; Vangelisti & Caughlin, 1997). Theories that take this approach consider self-oriented rewards to be catharsis, self-clarification, and instrumental support (Derlega & Grzelak, 1979; Derlega et al., 1993), whereas costs of disclosing are rejection, shame and embarrassment. These models assume that there is vulnerability associated with disclosure (Afifi & Olson, 2005; Petronio, 2002), thus individuals make deliberate choices about how, when, and with whom they choose to share their diagnoses (Petronio, Reeder, Hecht, & Mon't Ros-Mendoza, 1996) based on the evaluation of risks and rewards.

Information management models (e.g., disclosure decision model, DDM, Omarzu, 2000; Cycle of Concealment Model, Afifi & Steuber, 2010, Revelation Risk Model, Afifi & Steuber, 2009) outline the process of evaluating the risks and rewards of coming to a disclosure decision. Greene's (2009) DD-MM emerged from theories of information management, but is narrower in scope and focuses on the health disclosure decision process. For this reason, the DD-MM is the most useful theoretical framework for this thesis. However, a dilemma still remains in the testing of this model; specifically, there is a gap in knowledge of how different motivations affect the disclosure decision process and, ultimately, the likelihood to disclose. In this thesis, I focus on the

likelihood of disclosure of genetic information to a genetic relative because I see this as a challenging health context that may include more than one type of motivation guiding a discloser's intentions to disclose. Before I review DD-MM and its predictions related to the likelihood of health disclosures, I will describe the genetic condition studied herein: alpha-1 antitrypsin deficiency.

Genetic Case: Alpha-1 Antitrypsin Deficiency

Alpha-1 antitrypsin deficiency (AATD or Alpha-1) is a hereditary condition passed on from parents to their biological children through genes. It occurs when there is a severe lack of alpha-1 antitrypsin (AAT) protein in the blood, which can lead to serious liver damage that can cause cirrhosis in adults, children and infants and lung disease in adults. AATD is not a rare disorder (American Thoracic Society, 2003); it is one of the most common, potentially lethal heredity disorders (Colp, Pappas, Moran, & Lieberman, 1993). But, AATD has also been described as under-recognized (Stoller et al., 2005). Data collected from studies of 58 counties showed that there are at least 116 million carriers of an abnormal AAT gene and an estimated 3.4 million persons with a severe AAT gene combination coming from both parents (de Serres, 2002). Although the occurrence of Alpha-1 can only be estimated (Kelly, Greene, Carroll, McElvaney, & O'Neill, 2011), approximately 1 out of every 2,500 individuals in the U.S. are affected by the disorder (Alpha-1 Foundation, 2013). This prevalence is comparable to the incidence of cystic fibrosis (1/2500-1/3200) (Ratjen, & Döring, 2003; Rosenstein & Cutting, 1998) and Huntington's disease (30,000) (Ries et al., 2004).

The presentation of AATD-related symptoms mimics other conditions like asthma, which can lead to a five to eight year lag time between onset of symptoms and an AATD diagnosis (Stoller et al., 2005). This delay creates uncertainty regarding the diagnostic process surrounding AATD (Sandhaus, 2010). Additionally, the prognosis for AATD is highly variable. Some people

who have the genetic mutation may not show any symptoms and other people who are carriers may experience symptoms (Wienke, 2012). This further contributes to the uncertainty associated with this disorder. Using the typology presented by Rolland and Williams (2005), AATD fits into the class of genetic conditions in which clinical onset is in adulthood, likelihood of development is variable, and treatment can alter the onset or progression of clinical conditions. Conditions of similar caliber are BRCA mutations for inherited breast and ovarian cancers.

A diagnosis of Alpha-1 requires the patient take a blood test in order to determine the AAT levels in the blood (Alpha-1 Foundation, 2006). Early diagnosis of Alpha-1 is advantageous, because patients can receive counseling for healthier lifestyles and access to relevant treatments (e.g., augmentation therapy). It can facilitate genetic counseling and early detection in other relatives before chronic symptoms arise (Campos, Wanner, Zhang, & Sandhaus, 2005). It is important, therefore, to empower the patient to get tested and to disclose their results with those family members that may be at-risk so that they can consider being tested (Alpha-1 Foundation, 2006).

Family Implications of Genetic Tests

The manner in which genetic conditions are inherited has social and psychological consequences for affected families (James, Hadley, Holztman, & Winkelstein, 2006). The knowledge of genetic risk can impact the individual psyche, specific relationships and family identity (McConkie-Rosell & DeVellis, 2000; Rolland and Williams, 2005). Communication literature indicates that evaluation of disclosure risks and rewards (Magsamen-Conrad, 2012) and a person's right to privacy inform decisions on what information is shared (Petronio & Martin, 1986; Petronio, 2002). Although there may be a felt obligation to share genetic information, the discloser is under no real legal obligation to disclose. The potential discloser can evaluate the risks and rewards of disclosing and, afterwards, has a right to decide not to share, or share; and, if

the person does decide to disclose, the amount of information they share is up to them as well. Bioethics literature (Doukas & Berg, 2001; Dupras, & Ravitsky, 2013; Gilbar, 2007; Laurie, 2002; Parker & Lucassen, 2005) has centered on the right of the patient to privacy and confidentiality versus the right of family members to receive information that is clinically relevant to them.

Empirical research (Dancyger, Smith, Jacobs, Wallace, & Michie, 2010; Etchegary et al., 2009) has shown that although patients rarely contest the need for disclosure within the family, they are preoccupied by the ethical dimensions of their genetic responsibility. Parrott, Hong, and Greenberg (in press) found in their analysis of women's narratives on their first blood clot experience that many women felt there was no choice in the situation when one's family history and/or genetic predisposition were involved. There is a desire to protect relatives from the possible adverse effects of disclosure but guidance is needed regarding what, why, to whom, when and how genetic information should be disclosed to mitigate discloser-oriented, receiver-oriented, and relationship-oriented adverse outcomes (Dupras, & Ravitsky, 2013).

The specific risks and rewards associated with disclosing one's AATD diagnosis are psychological and instrumental. Receiving test results for AATD can be shocking and upsetting (Dohany, Gustafson, Ducaine, & Zakalik, 2012; Klitzman, 2009; Lippi, Favaloro, & Plebani, 2011), which can either motivate or discourage disclosure. People may disclose in order to receive instrumental and social support or to release some of the stress they are experiencing from being diagnosed (catharsis). Conversely, people may not disclose due to concern for others, protecting themselves and their family from potential stigma and discrimination. Beliefs about genetic stigma in regards to the control one has over the condition can be held against people diagnosed with AATD (Smith, Wienke, & Baker, 2013). In recent congressional testimony, Dr. Collins (2000) stated:

> While genetic information and genetic technology hold great promise for improving human heath, they can also be used in ways that are fundamentally unjust. Genetic information can be used as the basis for insidious discrimination.... The misuse of genetic information has the potential to be a very serious problem, both in terms of people's access to employment and health insurance and the continued ability to undertake important genetic research.

Unfortunately, Dr. Collins was not far off. Terri Seargent was a victim of genetic discrimination when her employer fired her after hearing that her medical tests determined that she had AATD and she began taking expensive medication for it. Before then, she had had a promising career as a manager and positive performance evaluations. Ms. Seargent filed with the EEOC alleging genetic discrimination and received a determination on November 21, 2000 that the EEOC's investigation supported her allegation of discrimination under the ADA (Jones, 2001). Although there are policies set to safeguard against this type of discrimination, fears of discrimination still persist and impede disclosure.

AATD is a rich case study to examine the disclosure decision-making process in the genetic context, because it is a dominant genetic disorder that is common yet under-recognized. The fact that by itself, AATD is not a disease, but a condition that when exacerbated by environmental factors, such as smoking (Tanash, Nilsson, Nilsson, & Piitulainen, 2010), leads to increased vulnerability to chronic obstructive airway diseases and chronic liver diseases (Alpha-1 Foundation, 2006; American Thoracic Society, 2003) makes this an even more compelling case study. Disclosing to a genetic relative could make them aware of their own health risks, leading to the consideration of different lifestyles, professions, or other decisions that could maintain or improve health (Alpha-1 Foundation, 2010). However, there is risk associated with disclosing

one's alpha-1 antitrypsin deficiency as evidenced by Terri Seargent's genetic discrimination case. Next, I will describe the theory investigated in this thesis: DD-MM.

Disclosure Decision-Making Model

The disclosure decision-making model (DD-MM; Greene, 2009) proposes several testable hypotheses about factors influencing a person's decision to disclose health information. The DD-MM is particularly relevant to the process of making health disclosure decisions (especially disclosure of potentially negatively valenced information) that may relate to physical or psychological health topics or conditions—including family and/or relational health (Greene, 2009; Magsamen-Conrad, 2012). The DD-MM describes three sets of assessments involved in the decision making process related to a) the information to disclose, b) the listener, and c) the discloser's efficacy to disclose the information.

The DD-MM describes how these three assessments are used to predict disclosure likelihood based on the evaluation of risks and rewards throughout. At the end of the disclosure decision-making process, greater likelihood to disclose is associated with fewer risks and greater rewards. The three risk-reward assessments of disclosure are covered in detail in the next few pages, starting with the information assessment.

Information assessment. Considered the first assessment, the information assessment is how people evaluate the health information (e.g., test results) under consideration for disclosure. It is thought to include the five, interrelated components: stigma, preparation, prognosis, symptoms, and relevance. These five aspects are not intended to represent an ordered process, rather, they could be considered in a different order, simultaneously, or not at all (Greene, 2009). *Stigma* is defined as the perception that information could produce negative outcomes (Venetis, Greene, Checton, & Magsamen-Conrad, under review) or feeling ashamed or discredited (Goffman, 1963, cited in Green, 2009). Greater stigma is associated with greater risk (Greene,

2009). *Preparation* is the degree to which the diagnosis was anticipated by the discloser, such as due to a family history. Greater preparation is thought to be moderated by assessment of the receiver (Greene, 2009) and has been associated with lower risk (Greene et al., 2012). *Prognosis* refers to the severity of the diagnosis (i.e., chronic, treatable, or terminal). *Symptoms* refer to visible manifestations of the disease (Greene, 2009). More severe prognosis and more visible symptoms decrease the risk of disclosure and increase its reward, leading to increased intentions. Last, *relevance* is defined as the degree to which the health condition is relevant to the selected target recipient of the disclosure (Greene, 2009). Greater relevance is negatively associated with risk and greater reward. Altogether, disclosure has more reward potential than risk with lower stigma, higher preparation, worse prognosis, more symptoms, and greater relevance.

AATD is a condition with high stigma, potentially low preparation, serious prognosis, visible symptoms and relevance for blood relatives. The stigma that comes with AATD is related to the associated diseases that manifest (e.g., COPD, jaundice; e.g., Berger, Kapella, & Laruson, 2011). Ironically, there often exist five to eight years between onset of symptoms and an AATD diagnosis (Stoller et al., 2005) because the presentation of AATD-related symptoms mimics other conditions like asthma. This delay in diagnosis creates uncertainty regarding the diagnostic process surrounding AATD (Sandhaus, 2010), which is only heightened by the variability in prognosis—some people who have the genetic mutation do not manifest any symptoms while other people who are carriers experience symptoms (Wienke, 2012). Therefore, knowledge of a family history of AATD is ideally the best way to prepare for diagnosis since it is a genetic disorder that can be passed on by one or both parents. Once one family member is diagnosed with the genetic mutation, the condition becomes relevant for blood-related family members. Because family histories of AATD are often not known (Klitzman, 2009), professional groups now specifically recommend testing all individuals diagnosed with COPD for AATD (e.g., American

Thoracic Society, 2003) in order to prevent under or misdiagnosis.

This study limits its scope to low preparation situations, in which college students are asked to consider receiving genetic test results in which they are diagnosed with AATD-related mutations. Realistic to diagnosis, the information provided on AATD to the students addresses both symptom and prognosis uncertainty. Further, the college students will be asked to select a blood relative for the disclosure, so there will be some level of relevance. The following hypothesis is proposed:

Hypothesis 1: Decreased stigma related to one's AATD diagnosis, more serious prognosis, increased symptom visibility, and greater relevance the genetic condition predicts greater likelihood of disclosing AATD genetic test results.

An individual may also repeatedly assess information for various targets or when new pieces of information become available. If the evaluated risk is not considered too great based on the information assessment, the potential discloser will then move forward in the decision-making process to consider the specific potential receiver (Greene et al., 2012).

Receiver assessment. In this next step, called the receiver assessment, the discloser evaluates the quality of the relationship with and anticipated reactions of a specific disclosure target in order to evaluate whether there are more costs or benefits to disclosing to a specific individual(s), especially when there is a specific goal in mind (e.g. attaining social support). Notably, the DD-MM is only intended to explain disclosures in which the discloser has a personal relationship with the target recipient and will be completed through face-to-face or mediated interactions (Greene, 2009). This familiarity the potential discloser has with the target recipient helps to inform the potential discloser's assessment of the receiver. *Relational quality* is most commonly related to greater intentions to disclose (e.g. Altman & Taylor, 1973; Chaiken & Derlega, 1974), even when possible stigma associations with the health information.

Anticipated reactions to the disclosure fall into two categories that are distinguished temporally (Magsamen-Conrad, 2012). *Anticipated response* is the most immediate communicative and/or behavioral reaction (even if that reaction is silence) from the target recipient anticipated by the discloser. *Anticipated outcomes* are the reactions anticipated as occurring after some time has passed (or feedback has occurred) after the initial disclosure and the listener's initial response (Greene et al., 2012; Magsamen-Conrad, 2012). More specifically, anticipated response is directly related to the communications and/or actions of the receiver and can be broken down into subtypes (emotional reaction, avoidance, and support- emotional, informational, and instrumental). Outcomes are evaluated on the implications (positive, negative, or neutral) for the discloser, the receiver, their relationship, and/or the discloser's relationship with others. Theoretically, there is a positive association between relational quality and anticipated reactions. A person's perceptions that the target recipient will respond positively to the disclosure (e.g., offer support) should result in perceptions of more positive relationship outcomes (e.g., stronger relationship) as well as an increased likelihood that the discloser will disclose their diagnosis to the target recipient (Greene et al., 2012).

Finally, the degree to which participants feel confident about how the target recipient would respond (confidence in response), is a variable that is remiss in most models but that is included in the DD-MM in order to have a more complete and telling receiver assessment (Greene et al., 2012). More confidence in response is positively related to anticipated reactions and positively predicts disclosure likelihood.

This study remains consistent with the DD-MM's intended use and requires the participant to choose a family member as a target recipient, ensuring that the discloser has a personal relationship with the target recipient and will be completed through face-to-face or mediated interactions (Greene, 2009). The following hypothesis is proposed:

Hypothesis 2: Better relational quality with a genetically-related family member, positive anticipated reactions (support and outcomes) and more confidence in receiver reactions predict greater likelihood of disclosing AATD results.

If there are more rewards than risks to disclosing to a specific target recipient based on the evaluation of the receiver or if the goal of disclosure can be accomplished by disclosing to a specific individual, the potential discloser will then move forward in the decision-making process to consider their efficacy to disclose their health information (Greene et al., 2012).

Efficacy assessment. Finally, after potential disclosers have completed the information and receiver assessment, and perceived greater rewards than risks for disclosure, they evaluate their own efficacy for disclosing the information. The DD-MM (Greene, 2009) draws on Bandura's (1977) concept for efficacy, and conceptualizes disclosure efficacy as the discloser's perception of his or her ability to disclose a specific message with a target recipient and produce desired results (Greene, 2009; Greene et al., 2012). This is narrower in concept than general efficacy (Bandura, 1977; Makoul & Roloff, 1998) and communication efficacy (Afifi & Steuber, 2009; Afifi & Weiner, 2004) since the particular message or risk related to the disclosure message may be particularly influential to one's assessment of their efficacy (Checton, 2010; Greene, 2009). The DD-MM posits that greater confidence and more skills to disclosure increase the likelihood of disclosure, as they increase the likelihood of achieving the rewards of disclosure and avoid the risks (Greene, 2009). The following hypothesis is proposed:

Hypothesis 3: Stronger disclosure efficacy predicts greater likelihood of disclosure about the AATD genetic-test results.

In summary, the DD-MM posits that the decision to disclose genetic-test results are either encouraged or discouraged by the dialectical dilemma of balancing risks and rewards based on the three assessments (Greene, 2009; Greene et al. 2012; Magsamen-Conrad, 2012). It is

important to note, though, the DD-MM puts forth the notion that people are not simply making a decision about sharing the initial diagnosis; rather, they are in a continual process where decisions have to be made about sharing updates (Greene, 2009). Further, the three assessments may not be equally influential in the decision-making (Checton & Greene, 2012).

Empirical Support for the DD-MM

Although the original publication of the DD-MM (Greene, 2009) did not include measurement information, subsequent studies have operationalized the concepts described in the DD-MM. The DD-MM argues that people base their decision of whether or not to disclose their personal information on the three previously discussed assessments. To date, there is one study examining the DD-MM as a broad theory, testing with general disclosure but not with health information (see Checton & Greene, 2012). The DD-MM has further been applied to several health conditions including HIV/AIDS (Greene et al., 2013), heart disease (Checton & Greene, 2012), nonvisible illness (Checton, Greene, Magsamen-Conrad, & Venetis, 2012), cancer (Venetis et al., under review; Venetis, Magsamen-Conrad, Checton, & Greene, in press), and infertility (Steuber & Solomon, 2011), situations in which self-oriented social support are most applicable since disclosures in these health contexts indirectly affect the target recipient. I will describe the empirical support for DD-MM below.

Greene et al. (2012) tested the DD-MM with both disclosed and undisclosed health information in a sample of 183 individuals with a non-visible health condition. Individuals were asked to report about disclosure scenarios, one where they had disclosed their non-visible health condition to another person and one where they had not yet disclosed that information. Authors tested a "disclosed" and "undisclosed" model of DD-MM. Both models supported the propositions of the DD-MM (Greene, 2009), and there was overlap in the process of disclosure (as explained by the DD-MM) in both decisions about disclosing health information not yet

shared and in examining the process of that same information when it had already been shared.

Checton and Greene (2012) also tested the DD-MM in a health context. They surveyed 203 cardiac patients from a private medical office. Individuals reported about sharing information about a heart-related condition with their partner. Participants had been in the relationship an average of 39 years (ranging from two to 70 years). In this investigation, the DD-MM was applicable to ongoing disclosure decisions between partners managing a heart condition. Their findings suggested that key mechanisms identified in the DD-MM (Greene, 2009; i.e., assessment of information, relational quality, anticipated/actual response, and efficacy) predict the depth, breadth, and frequency of disclosure to a partner about a heart-related condition. Specifically, patients' uncertainty about their heart condition and their perceptions of relational quality both predicted perceived partner support. Prognosis uncertainty also predicted the breadth of communication between partners (from the perception of the patient). Uncertainty about the symptoms of patients' heart condition and perceived partner support both predicted communication efficacy. Symptom uncertainty also predicted frequency of communication about the heart condition between partners (again, from the perspective of the patient). Finally, communication efficacy predicted patients' perspectives of the breadth, depth, and frequency of their communication with their partner about their heart condition.

Venetis et al. (under review) employed the DD-MM to explore cancer-related topic avoidance among cancer patients and their partners. Participants included 95 dyads in which one partner had been diagnosed and/or treated for cancer. Their findings suggest that lack of reciprocity and efficacy are significant predictors of topic avoidance.

While the DD-MM has been applied to many health settings, the genetic context is not one of them. The implications associated with genetic disclosure and a person's responsibility to family may lead to a disclosure decision process that does not replicate findings from those

presented in other studies using the DD-MM, in which family does not play such a pivotal role and self-oriented social support is most applicable. There may be some components of information assessment that greatly influence assessment, while other components may have no bearing. In the next section, I consider the DD-MM's assumed motive for disclosure.

Challenging the DD-MM Framework: Other-Motivations for Disclosure

The DD-MM assumes that the motive driving disclosures, the key reward, so to speak, is social support. The theoretical variable most closely tied to social support is anticipated response, which falls in the second assessment: receiver assessment. As a reminder, while the conceptual definition of anticipated response was in terms of valence (i.e., will the listener respond positively or negatively, Caughlin, Afifi, Carpenter-Theune, & Miller, 2005; Greene et al., 2006), Greene and colleagues operationalize anticipated response as anticipated support: emotional, informational, and instrumental (Magsamen-Conrad, 2012). Empirical tests of the DD-MM (Checton & Greene, 2012; Greene, 2009; Greene et al., 2012) repeatedly show that anticipated response is strongly related to disclosure likelihood, either directly (Greene et al., 2012) or indirectly through efficacy (Checton & Greene, 2012).

In a review of family communication of genetic risk, it was found that when the function of disclosure was to receive advice and support, relationship factors were particularly important (Hughes et al., 2002; McGivern et al., 2004; Ormondroyd et al., 2007); and the need to carefully weigh the decision to tell, the usefulness of the information and how to tell each relative becomes more critical in the decision-making process (Clarke, Butler, & Esplen, 2008; Foster et al., 2004). However, in the genetic disclosure context, the attainment of social support may not be the sole or primary goal underlying a person's disclosure decision process. Though perceptions that being physically and emotionally close to family members may facilitate genetic risk information (Blandy, Chabal, Stoppa-Lyonnet, & Julian-Reynier, 2003; Hughes et al., 2002; Keenan et al.,

2005; Ormondroyd et al., 2007), Greene (2009) explains there are times that the quality of the relationship, even poor, may not influence intentions and the discloser will disclose regardless. Potential disclosers may be impelled by a sense of responsibility or felt obligation to disclose genetic risk information to relatives (d'Agincourt-Canning, 2001; Green, Richards, Murton, Statham, & Hallowell, 1997; Hughes et al., 2002; McGivern et al., 2004; Ormondroyd et al., 2007). The relevance and importance of the genetic health information to family members may supersede other concerns and assessments (d'Agincourt-Canning, 2001; Finlay et al., 2008; Foster et al., 2004; Forrest et al., 2003; Hughes et al., 2002).

Other studies on health disclosure (not using the DD-MM) have identified several motivations for disclosure and nondisclosure, which have been organized into self-, other-, relationship-focused and situational-environmental categories (Derlega, Winstead, Wong, & Greenspan, 1987; Derlega et al., 2000, 2004, 2008; see also Fitzpatrick, 1987; Greene et al., 2003, 2006; Greene & Magsamen-Conrad, 2010). Self-focused reasons for disclosure are related to tangible and psychological benefits of disclosure for the discloser, including catharsis, seeking help/support, and self-clarification. This category is similar to the motivation assumed in the DD-MM. An example of this would be disclosing one's AATD diagnosis in order to access emotional or tangible help in coping with the condition. Other-focused reasons for disclosure are centered on how sharing information creates benefits or risks to others (i.e., a sense of duty to inform and a desire to educate) (Derlega et al., 2000; 2004; 2008). In the genetic context, an other-focused reason for disclosure would be based on a "duty to inform" a spouse or sibling about one's alpha-1 antitrypsin deficiency due to relevant risk and/or for making family planning decisions. The dialectical dilemma of risks and rewards may motivate the potential discloser to compromise between individuals' needs and their concerns for the other (Afifi & Steuber, 2009; Greene et al., 2003; Greene et al., 2006).

Multiple Motivations Involved in Genetic Disclosures

The decision to share genetic knowledge is influenced by how tested people consider the benefits and burdens of potentially intervening in one's own health and that of others (Williams & Schutte, 1997). Risks of disclosing include emotional distress and loss of hope (Blandy et al., 2003; Wagner et al., 2003; Williams & Schutte, 1997). Patients have expressed concerns of putting information recipients at harm by delivering difficult information (e.g., stigmatization and insurance discrimination) (Green et al., 1997; Forrest et al, 2003; Ormondroyd et al., 2007) and worries associated with being the discloser (e.g., feelings of guilt and blame associated with passing genetic diseases) (d'Agincourt-Canning, 2001; Forrest et al., 2003; James et al., 2006).

There has also been documented fear from the potential discloser of causing tension within the family regarding decision making (e.g., family planning, treatment options and testing, and financial) (James, et al., 2006; Mellon, Berry-Bobovski, Gold, Levin, & Tainsky, 2006). The implications of disclosing genetic information also threaten concepts of individual privacy (Clarke et al., 2005) and Kantian perspectives of autonomy (Rhodes, 1998; Takala & Hayry, 2000). Using an example similar to the one provided in the introduction, if a daughter chooses to disclose her alpha-1 antitrypsin deficiency to her mother against her mother's wishes of genetic ignorance, it can be argued that the daughter is violating her mother's privacy and right to decide. On the other hand, Rhodes (1998) would argue that if the mother chooses to remain ignorant of relevant information, then she is choosing to leave whatever happens to chance and may not be aware of the moral implications of ceding such information though the ramifications are there, nevertheless.

Therefore, if autonomy is the grounds for determining a course of action, then it cannot also be the ground for not determining a course of action; and, as "…a sovereign over myself I am obligated to make thoughtful and informed decisions without being swayed by irrational

emotions, including my fear of knowing significant genetic facts about myself…and when the relevant information is obtainable with reasonable effort" (Rhodes, 1998, p.18). It is this predicament that makes genetic disclosure an ethical issue and a potential deterrent from disclosing. The morals of the discloser and where he or she places most value, the self vs. the other, privacy vs. autonomy, guides the decision to disclose or not.

The fact that the other being considered in the dialectical dilemma is typically a blood-related family member makes the disclosure decision process more personal and value-based. The genetic status of one person has biological, psychological and relational implications for his or her family members as well (Dancyger et al., 2010). A genetic test result reveals information about the tested person and also potential risks for relatives (Claes et al., 2003). People who carry genetic mutations may feel the need to inform relatives of the test results. They may also inform relatives of the availability of screening and predictive genetic testing so that family members can act responsibly in relation to their genetic risk (Etchegary et al., 2009). By disclosing one's genetic information and sharing the availability of genetic testing, a person can play a central role in facilitating informed decision making and risk management options for others. Thus, the value and harm of testing can extend beyond the person being tested.

While the DD-MM may benefit from exploring other relational and situational-environmental motivations for disclosure, for genetic disclosures, other-focused motivations, specifically duty to inform blood relatives, are particularly relevant. Genetic-based conditions have serious implications for related family members, and their risk for also experiencing the genetic-based condition may be a specific drive for genetic disclosures (Nycum et al., 2009). Indeed, the specific nature of genetic information that makes its disclosure more of a question of obligation (Rhodes, 1998) rather than a completely voluntary act. The responsibility for the management and prevention of risk no longer simply falls on the individual, but is viewed as a

familial obligation (Arribas-Ayllon, Sarangi, & Clarke, 2008; Kenen, 1994; Lupton, 1995; Petersen, 1998; Sommerville & English, 1999; Weijer, 2000) because it is assumed that the rights and duties as part of a family is to protect family. For this reason, the broad notion of duty to inform is narrowed in this thesis to *familial obligation*.

Family Obligation

Familial obligation is defined as family members' responsibilities to one another to warn of a genetic mutation (Forrest, Delatycki, Skene, & Aitken, 2007; Nuffield Council on Bioethics, 1993; Nycum, Avard, & Knoppers, 2009b). The argument that genetic information is not only personal information, but also familial (Nycum et al., 2009b) has become more prominent and the various obligations for disclosure to the entire family have become more carefully scrutinized. This is a result of technology and knowledge advancements and the term "new genetics" becoming more commonly used post-Human Genome Project (Guttmacher, 2000) to refer to genomic medicine and the ubiquity of genetic tests being used for predictive and diagnostic purposes (Burke & Zimmern, 2004; Evans, et al., 2001; Smith et al., in progress). The Nuffield Council of Bioethics (1993) states that both individuals and family members have responsibilities to be willing to give and receive genetic information. Other publications encourage people to "act ethically" (Genetic Interest Group, 1998) by sharing the genetic information with family members. An individual's moral obligation to their at-risk family members is presented as a reason for testing in the first place (Hallowell et al., 2005).

This assumed desire to protect family (Forrest et al., 2007) bleeds into legal obligations as well (Weijer, 2000). Not only is nondisclosure of genetic information to family members viewed as "morally condemnable" (National Consultative Ethics Committee for Health and Life Sciences, 2003) it can be argued as a violation of legal violation of the limits dictated by the case Tarasoff v Regents of University of California (1976). In response to this legal case and the

growing focus on genetics, the American Society of Human Genetics created a policy statement on disclosure of genetic information that is consistent with those of the American Society of Clinical Oncology (ASCO) and the American Medical Association (AMA), in that the duty to warn does not extend to genetic risks; however the policy does acknowledge that it may be acceptable for health professionals to breach genetic confidentiality under certain conditions, including if the harm is serious and foreseeable, the at-risk individuals can be identified, the disease is preventable or treatable, or early monitoring is medically accepted to reduce risk or avert harm (as cited in Storm, Agarwal, & Offit, 2008). Although the duty to warn is widely recognized as not extending to genetic risks, there are people that argue that one's right to confidentiality should be breached in cases of genetic disease due to the risks associated with certain conditions and certain treatments and health behaviors and lifestyles can prevent harm (Weijer, 2001). The limitation of this legal justification is that it applies to a set of moral rules for strangers rather than family (Weijer, 2001). Policies only touch on obligation, the motivation appears to be more intrinsic and moral.

Families may be involved both at the outset and in the aftermath of testing (Nycum et al., 2009b). If genetic testing is predictive rather than diagnostic, the patient's account of family history may give clues about the possibility of a genetic risk. Information about genetic relatives is typically collected as part of a pre-test consultation, and a family history may be needed to supplement test results, to make them meaningful, or to confirm a diagnosis. If diagnostic, a treating physician generates genetic information on behalf of an individual and the diagnosis may have health implications for other genetic relatives and future generations (e.g., trends in symptoms or reproductive implications) (d'Agincourt-Canning, 2000). This means that direct family involvement is needed to make testing more effective and disclosure is the first step.

Most people who undergo genetic testing report feeling some familial obligation to communicate their results with family members (See Wiseman, Dancyger, & Michie, 2010). The decision to disclose is ultimately highly context specific and will be shaped by many factors, including the type of genetic condition at issue (i.e., a single-gene or multifactorial genetic condition), familial relationships, individual personalities, and perceptions of what is in the family's best interest (Nycum, Avard, & Knoppers, 2009a).

Notably, perceptions of who is "family" and the obligation to share a diagnosis with identified family members are often determined by social relationship rather than by biological relationship (Nycum et al., 2009a). There is a lack of moral impulse in the absence of a social relationship that compares such that people often feel less sense of obligation to disclose to family members with whom there is no, or a distant, relationship (Gilbar, 2005). A possible exception is that family members may feel responsibility through self-blame and guilt (Arribas-Allyon et al., 2009). For instance, parents feel guilt when their children are affected or test positive (Chapple, May, & Campion, 1995).

Although Rhodes' (1998) approach of family obligation is more complex and contextually dependent, the argument is similar to the biosocial approach. Based on Kant's concept of autonomy, it is not just a moral right not to know about our own genetic disorders, because without all the relevant information we cannot make autonomous choices; however:

> If genetic similarity were the source of our moral relationships, genetic maps could identify our most similar sibling, or even some distant DNA matching stranger, as the one to whom we owed the most. But if anyone maintained that we had different degrees of responsibilities to different siblings it is not likely that they would attribute that distinction to our degrees of genetic matching. More likely reasons would be related to the intimacy and dependency of our

> previous relationship, or the strength of our feelings, or the history of our interactions, or something about our relative wherewithal and neediness. These reasons, however, point to social relations, not blood or genes, as relevant features for moral responsibility (Rhodes, 1998, p.21)

This means that someone genetically close does not have more moral claim than others we have relationships with. Bloodlines do not determine moral responsibility.

Empirical Support for Familial Obligation and Self-disclosure

There is evidence, both quantitative and qualitative, that familial obligation exists as a reason for disclosure. Derlega et al. (1998; 2004; 2008) consistently found that loyalty to and concern for others were important reasons for disclosure to family members. In 1998, Derlega et al. conducted an interview study among persons with HIV about reasons for and against disclosing their HIV status. Responses indicated that reasons for disclosure included catharsis, seeking help, the duty to inform, education, testing others' reactions, being in an emotionally close relationship, and similarity with the target. Derlega et al. (2004; 2008) further explored the concept of duty to inform and people often conveyed that the target recipient has a right to know or that they wanted to disclose in order to have an open and honest relationship. Specifically, family members were often told because of a sense of "duty" or "loyalty" (Derlega & Winstead, 2001; Greene et al., 2003). Several other studies have interviewed patients and found that patients thought it was their responsibility to inform family members of their risk and that these family members had a right to know (d'Agincourt-Channing, 2001; Foster et al., 2004; McGivern et al., 2004; Ormondroyd et al, 2007; van den Nieuwenhoff, Mesters, Gielen, & de Vries 2007). Dancyger et al. (2010) found that those committed to genetic testing described their primary motivations for testing as arising from an obligation to others, which may override the perception of the value of testing for oneself. This obligation to others can be considered in terms of genetic

responsibility, to do what is morally right for the family. The evidence demonstrates a common human desire to protect family members (Klitzman, Thorne, Williamson, Chung, & Marder, 2007; Etchegary & Fowler, 2008; Dupras & Ravitsky, 2013). This leads to the following research question:

Research Question 1: Do people feel motivated to disclose genetic test results out of a familial obligation?

Depending on the goal of the discloser and the motivations driving the disclosure process, applying the genetic context may shift the discussion of disclosures from privacy, justice, equality, equity, and autonomy to reciprocity, mutuality, solidarity, citizenry, and universality (Nycum et al., 2009b). It also may affect to whom we disclose and how assessments are made in the process. Testing the DD-MM for genetic disclosures, then, may provide insights into "close" relationships and the nature of different personal relationships. Examining the DD-MM with genetic disclosures could also lead to a better understanding of what specific variables play a bigger role when familial obligation is reported as the main motivation for disclosure. When familial obligation is motivating tested persons to disclose, the relationships articulated in the DD-MM may no longer hold. The dialectical dilemma of risks and rewards to motivate the potential discloser (Afifi & Steuber, 2009; Greene et al., 2003; Greene et al., 2006) and the achievement of managing uncertainty (Afifi & Weiner, 2004; Greene, 2009) may be less prominent in the genetic disclosure decision-making process. If familial obligation is the driving motivation, then assessments of prognosis, severity, and relevance for others may be more important predictors of disclosure likelihood, because these are the risks and rewards associated with their family members' wellbeing. While tested persons may still make information, relational, and efficacy assessments, the quality of the information and relationships relevant for

getting support (and avoiding negative reactions) may not be strong predictors. The following hypothesis is proposed:

Hypothesis 4: For those reporting a familial obligation to disclose, greater relevance, more prognosis certainty and more severity predict greater disclosure likelihood.

Young Adults and Genetic Tests

The experience of young people disclosing to adults, in general, remains under-researched (Smith, Greenberg, & Parrott, 2014). Most studies have focused on the genetic information disclosure patterns of siblings or parents communicating to children (Claes et al., 2003; Lerman, Peshkin, Hughes, & Isaacs, 1998; Klitzman et al., 2007; Ratnayake et al., 2011; Wiseman et al., 2010). This pattern seems ironic given that young adults (18-26) are just old enough to undergo genetic testing, receive their test results, and make decisions based off the information provided (Feero, Guttmacher, & Collins, 2008). Young adults are still young enough that family members (e.g., parents, siblings, and grandparents) are still alive and available (Aquilino, 2005; Eggebeen, 2005; Fingerman, Miller, Birditt, & Zarit, 2009; Furstenberg, 2000). Family members may still play an active role in their lives and be major sources of social support (Aquilino, 2005; Eggebeen, 2005; Fingerman et al., 2009; Furstenberg, 2000). And though the research has not centered on this specific population, campaigns to encourage genetic-testing have (Alford et al., 2011; McBride, Wade, & Kaphingst, 2010). Therefore, this is a compelling population to study in relation to the genetic disclosure decision-making process because they are newly autonomous decision makers but may still heavily rely on their family for support and advice. Self-oriented motivations (e.g., social support) and other-oriented motivations (e.g., family obligation) may be particularly salient in young adults.

The Present Study

In this thesis, I use the DD-MM (Greene, 2009) to predict what variables influence participants' likelihood to disclose a hypothetical Alpha-1 diagnosis to genetic relatives. The DD-MM presumes a self-oriented motive for disclosure: a desire for social support. I argue that the relevance of a genetic condition to a blood-related family member plus the uncertainty of prognosis may invite another motive for disclosing, familial obligation. The goal of this thesis is to understand how familial obligation influences the disclosure decision-making process in the genetic context, if at all. The results will provide insights into information management models such as the DD-MM and genetic disclosures.

Chapter 2

Methods

Participants

The total sample included 191 undergraduate participants recruited from a multiple-section, required course at a large, eastern university. Volunteers needed to be at least 18 years of age to participate. Upon reviewing their answers, 18 participants did not complete multiple items on the survey, leading to no information at all for at least one or more variables. These 18 people were removed from the sample.

The final sample included 173 individuals. Of these participants, 89 (51%) identified as female, 83 (48%) as male, and 1 (1%) unidentified. Participants on average were 20 years old (*Mode*=20, *SD*=1.13, *Minimum* = 18, *Maximum* = 23 or older) and held a junior class standing (51%). Participants self-identified as Caucasian (84%), Asian (10%), Hispanic (6%), African-American (4%), and American Indian or Alaskan (2%). Most participants reported that they were not adopted (98%), and had at least one sibling (92%). Participants lived in different social environments, including living alone (8%), with other students (83%), other roommates that were not students (2%), with parents/relatives/guardians (5%) or with a spouse/significant other (2%).

Design

The research question and hypotheses were tested by conducting a cross-sectional study in which participants provided self-report data about their disclosure decision-making process regarding the sharing of positive genetic test results for AATD.

Procedures

An institutional review board approved the study. I contacted prospective participants provided by the participant-pool via email and included a link to the online survey. In the email, I identified myself as a Penn State researcher that is conducting a study for research purposes and

invited each person to complete my online survey. After accessing the survey and providing consent online (see Appendix A), participants were presented with the full survey. The survey opened with a hypothetical scenario, which remained the same for each participant (see Appendix B). They were asked to imagine that they had recently been diagnosed with a genetic condition called alpha-1 antitrypsin deficiency (AATD) and provided a simple factsheet on the condition (see Appendix C). Sara Wienke, a genetic counselor for the Alpha-1 Association's Genetic Counseling Program by the Medical University of South Carolina (MUSC), reviewed both the hypothetical scenario and fact sheet. Even though I made efforts to ensure medical accuracy, participants were asked to report on how believable and informative the scenario and factsheet were after reading it.

Participants were asked to name one blood-related family member (first name only) and their relation to the participant. Participants were asked to respond to multiple items relating to the disclosure decision-making model (DD-MM; 2009), their reasons for disclosure (Derlega, Winstead, & Folk-Barron, 2000) and their disclosure behaviors (Omarzu 2000). Questions on demographics (e.g., gender, age and race), family, previous experience with genetics, and previous knowledge of AATD were included at the end of survey. It should not have taken the participant more than 60 minutes to complete the entire study, if completed in one sitting (duration mean: 3 hours, 42 minutes; trimmed mean: 32 minutes). Participants were debriefed upon completion of the survey.

Pilot Study

A pilot study was conducted to assess study feasibility, survey adequacy, measure reliability, and overall internal validity. Participants were recruited from a multiple-section required undergraduate CAS 100b course (n=70) and through Amazon's Mechanical Turk (n=73) (N=143, 41% female, 57% male, 2% unidentified). Ages ranged from 18 to 67 years old (M=26.8,

SD=10.9). The majority of the sample identified as white (81%); Asian (13%), Hispanic/Latino (4%), Black/African American (4%), American Indian/Alaskan (1%), Native Hawaiian/Other Pacific Islander (1%). Confirmatory factor analyses were not performed on the pilot-data because the sample was less than 200 participants. Descriptive statistics from the pilot study are presented in Table 1. When composite scores were created, the items were coded so that higher scores represented more of the construct.

Table 1

*Descriptive Statistics for Scales Gathered in Pilot Test (*N = *138)*

Variable	*M*	*SD*	α	Skewness	Kurtosis
Genetic stigma	2.11	0.64	.89	0.47	0.40
Preparation	3.50	0.77	.71	-0.14	-0.61
Prognosis	4.09	0.68	.80	-0.85	0.72
Symptoms	3.21	0.88	.60	0.07	-0.44
Relevance	3.23	0.73	.83	0.45	-0.24
Relational quality	4.28	0.75	.89	-1.50	2.78
AS: emotional	1.81	0.73	.77	0.98	0.85
AS: informational	2.00	0.86	.84	0.95	0.97
AS: instrumental	1.76	0.85	.86	1.22	0.81
AR: discloser-oriented outcomes	1.81	0.85	.90	0.95	0.60
AR: receiver-oriented outcomes	1.81	0.85	.95	0.95	0.60
AR: relationship-oriented outcomes	1.84	0.69	.80	0.98	0.50
AR: other relationship-oriented outcomes	1.80	0.75	.82	1.14	1.87
Confidence in response	4.12	0.72	.89	0.68	-0.36
Disclosure efficacy	3.92	0.82	.87	-0.44	-0.37
Likelihood of future disclosure	3.37	1.19	.70	-0.41	-0.74
Disclosure for social support	4.00	0.99	.88	-1.12	0.84

Disclosure as family obligation	4.15	0.72	.86	-1.00	1.65

Notes. AS = Anticipated support. AR = Anticipated reactions.

The pilot study was able to address a number of logistical issues regarding measure construction and reliability. The results showed that the Cronbach's alphas for the measures preparation, symptoms, anticipated receiver-oriented outcomes, and likelihood of disclosure were small. In an effort to improve measure reliability, these measures, and others, were revised for clarity, directness, and specificity. The pilot measure for symptoms was heavily revised for the study. Each item was made more specific to AATD and referenced particular symptoms (i.e., wheezing, shortness of breath, chronic cough, and liver and/or lung depreciation) associated with the genetic condition. Further, only four of the seven items were used for analysis. The pilot measure for preparation included six items, which were reduced to four for the current study. Similarly, three of the seven items included in the original measure for prognosis were removed for the present study. For this study, relevance was measured with five of the eight items used in the pilot study. Stigma will be analyzed with 11 of the 12 items tested in the pilot study. Relational quality was minimally revised with only one item being eliminated from the pilot measure. One item from the pilot's measure on discloser-oriented outcomes and two items from receiver-oriented outcomes were edited out for the present analysis. Finally, for this study, several items were added to the likelihood of disclosure measure, but, as it turns out, the original four items tested in the pilot were most reliable.

In addition, before the participant was presented with the hypothetical scenario, a question was added asking the participants to report which living blood-relatives they have interacted with in their lifetime. A list of biological relatives is provided for the participant to consider. The list included the following: *biological mother, biological father, biological brother, biological sister, biological aunt, biological uncle, biological female cousin,* and *biological male*

cousin. When asked to consider one family member when responding to survey questions assessing a potential disclosure recipient, the current study prompted participants to select one of the blood-relatives the participant had previously reported interacting with from the list provided at the beginning of the survey. This had not been the case for the pilot study. A measure to check one's knowledge of AATD after being presented with the scenario and fact sheet was also added to the current survey. A question on the participant's primary motive for disclosure was added to the survey as well.

Measures

Measures from the survey appear in the Appendices (D-H). Variables measured include *information valence* (stigma, relevance, symptoms, prognosis, and preparation), *relational quality, anticipated support* (emotional, informational, and instrumental)*, anticipated outcomes* (discloser-, receiver-, relationship-, and other relationship-oriented), *confidence in response*, *disclosure efficacy, likelihood of disclosure,* and *reasons to disclose.*

Collected data were screened for normality and outliers. All variables were below the guidelines for skewness and kurtosis (< 3 and < 10, respectively) recommended by Kline (2005). The data were also screened for missing data. For each measure, if a participant was missing less than 5% of the data, then the missing values were imputed by group-mean substitution. As stated in the participants' section, participants who had data missing for at least one variable were deleted from the dataset.

Once the data was screened, confirmatory factor analyses (CFA) were used evaluate the multi-item scales for factor structures and if they met the criteria of face validity, internal consistency, and parallelism (Hunter & Gerbing, 1982). Three tests were used to assess the goodness of fit for the estimated hypothesized model, including the Comparative Fit Index (CFI), Standardized Root Mean Squared Residual (SRMR), and the Root Mean Squared Error of

Approximation (RMSEA). CFI values range from 0 to 1, with better overall fit indicated by higher values. Specifically, Hu and Bentler (1999) recommend CFI values of .95 or higher as an indication of a good fitting model. To the contrary, Holbert and Stephenson (2002) recommended, that for a sample of less than 250, a cutoff value close to .09 for the *SRMR* should be used to evaluate model fit and the *RMSEA* should be close to .06 or less. Unfortunately, the *RMSEA*, which accounts for errors of approximation in the population, tends to over-reject true population models when the sample size is below 250; therefore, I used an *RMSEA* value of .10 or less as a cutoff of acceptable model fit (Browne & Cudeck, 1993). Further, Hu and Bentler (1999) suggested that when there is a small size of less than 250, a combinational rule which dictates that a cutoff value of .96 for CFI in combination with *SRMR* > .09 should be used. This rule is more preferable since it results in the least sum of Type I and Type II error and combinational rules based on *RMSEA* and *SRMR* tend to reject more simple and complex models under due to robustness, or lack thereof.

While all of the measures passed the *SRMR* criteria, many of them had CFIs and *RMSEA*s that did not. I inspected the inter-correlation matrices. I did not identify particular items that did not fit well with the data. Instead, the inter-correlation matrices appeared to be flat, but often not very strong. For scales with sub-components, a second-order factor structure or more complex tests may have improved the goodness-of-fit indices. With the *SRMR* as the ultimate cut-off, composite scores were created for every measure. The findings should be taken with caution.

Cronbach's alphas were calculated as summary estimates of reliability for composite measures. Descriptive statistics of all measures, including means, standard deviations, skewness, kurtosis, and Cronbach's alpha can be found in Table 2. When composite scores were created, the items were coded so that higher scores represented more of the construct.

Genetic stigma. Eleven items (adapted from Link, Cullen, Struening, Shrout, & Dohrenwend, 1989) were used to index perceptions of genetic stigma (e.g., *Most people would not want their children to marry me because I have AATD*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were coded afterwards for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .83, *SRMR* = .08, *RMSEA* = .16 (90% CI, .14, .18). The items were summed and averaged to form a scale (see Appendix D).

Preparation. Four items (one from Greene et al., 2012 and three from Greenberg & Smith, 2013) were used to index one's level of preparation for the genetic diagnosis (e.g., *I would not be expecting genetic tests for breathing problems*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .99, *SRMR* = .03, *RMSEA* = .07 (90% CI, .00, .18). The items were summed and averaged to form a scale (see Appendix D).

Prognosis. Four items (three adapted from Checton & Greene, 2012 and Greene et al., 2012 and one from Greenberg & Smith, 2013) were used to measure the perceived level of severity of the genetic condition (e.g., *AATD is a serious health condition*). The response options were be *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were coded afterwards for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .99, *SRMR* = .02, *RMSEA* = .07 (90% CI, .00, .18). The items were summed and averaged to form a scale (see Appendix D).

Symptoms. Four items (one adapted from Greene et al., 2012 and three adapted from Checton, 2010) were used to measure symptoms with regards to visibility and uncertainty to relate to the genetic context (e.g., *It would be not be difficult for others to notice my AATD*

symptoms). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .88, *SRMR* = .07, *RMSEA* = .17 (90% CI, .09, .27). The items were summed and averaged to form a scale (see Appendix D).

Relevance. The five-item measure (adapted from Greenberg & Smith, 2013) was used to index perceptions of relevance (e.g., *My AATD diagnosis has no real implications for others* (R)). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were coded afterwards for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .95, *SRMR* = .05, *RMSEA* = .09 (90% CI, .02, .16). The items were summed and averaged to form a scale (see Appendix D).

Relational quality. Five Likert-type quality (four items adapted from Checton and Greene and one from Greenberg & Smith, 2013) items were used to measure overall relational quality (e.g., *I enjoy spending time with this family member*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which will be coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .97, *SRMR* = .04, *RMSEA* = .12 (90% CI, .06, .18). The items were summed and averaged to form a scale (see Appendix E).

Anticipated support. Three types of anticipated responses were measured: emotional, informational, and instrumental support. Emotional support was measured with five items (adapted from Magsamen-Conrad, 2012) (e.g., *My family member would immediately offer emotional support*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were coded afterwards for analysis (1 = *strongly disagree* to 5 = *strongly*

agree). Informational support was measured with four items adapted from Magsamen-Conrad (2012) (e.g., *Initially, my family member would help me look for information*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). Instrumental support was measured with four items adapted from Magsamen-Conrad (2012) (e.g., *My family member would initially offer instrumental support (accompany to doctor, loan money for treatments*)). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The best overall anticipated support fit was obtained from three correlated latent factors (*r*= .49, 59, 71).

For emotional support, the CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .92, *SRMR* = .06, *RMSEA* = .15 (90% CI, .09, .21). For informational support, the CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .96, *SRMR* = .05, *RMSEA* = .17 (90% CI, .08, .27). For instrumental support, the CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .96, *SRMR* = .04, *RMSEA* = .17 (90% CI, .09, .27). The items for each factor were summed and averaged to form respective scales (see Appendix E).

Anticipated outcomes. Four types of anticipated outcomes were measured: discloser-oriented, receiver-oriented, relationship-oriented, and other relationship-oriented. Anticipated discloser-oriented outcomes were measured with three items (adapted from Afifi & Steuber, 2009, 2010; Derlega, 2002; Vangelisti & Caughlin, 1997; Vangelisti, Caughlin, & Timmerman, 2001) (e.g., *Down the road, sharing my AATD diagnosis would negatively affect how my family member would feel about me*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were coded afterwards for analysis (1 = *strongly disagree* to 5 = *strongly agree*). Anticipated receiver-oriented outcomes were measured with two items (adapted

from Afifi & Steuber, 2009; Derlega, 2002; Vangelisti & Caughlin, 1997) (e.g., *It would ultimately hurt my family member's feelings if s/he knew the information*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). Anticipated relationship-oriented outcomes were measured with four items adapted from Derlega et al. (2002) and Afifi & Steuber (2009) (e.g., *Telling my genetic test results to my family member would ultimately hurt our relationship*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). Anticipated other relationship-oriented outcomes were measured with five items adapted from Vangelisti & Caughlin (1997) (e.g., *Telling my genetic test results to my family member would ultimately hurt my relationship with other family members*). The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The best overall anticipated outcomes fit was obtained from three correlated latent factors (r=.78, 65, 66).

The items for anticipated discloser-oriented outcomes and receiver-oriented outcomes were highly correlated. For the combined measure for discloser- and receiver-oriented outcomes, the CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .97, *SRMR* = .05, *RMSEA* = .14 (90% CI, .08, .20). For relationship-oriented outcomes, the CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .96, *SRMR* = .03, *RMSEA* = .13 (90% CI, .07, .19). For other relationship-oriented outcomes, the CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = 1.00, *SRMR* = .03, *RMSEA* = .04 (90% CI, .00, .12). The items were summed and averaged to form a scale so that higher scores indicate more negative outcomes (see Appendix E).

Confidence in response. The degree to which participants felt confident about how the target recipient would respond was measured by five items (e.g., *I can accurately predict how my family member will react*) adapted from Greenberg and Smith (2013). The response options were strongly disagree, disagree, neutral, agree and strongly agree, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .99, *SRMR* = .03, *RMSEA* = .07 (90% CI, .00, .14). The items were summed and averaged to form a scale (see Appendix E).

Disclosure efficacy. Three items were developed to apply to the genetic context and were tested (e.g., *I would have trouble finding the right words if I tried to share my diagnosis with my family member* (R)). The response options were *not at all*, *slightly*, *somewhat*, *moderately* and *completely*, which will be later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .84, *SRMR* = .07, *RMSEA* = .16 (90% CI, .14, .17). The items were summed and averaged to form a scale (see Appendix F).

Likelihood to disclose. A four-item measure (adapted from Greene et al.'s (2012), which was originally adapted from Vangelisti et al., 2001 and Caughlin et al., 2005), was used to measure the likelihood of disclosing (e.g. *I am confident that I could share my diagnosis with my family member if I decide to*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .97, *SRMR* = .03, *RMSEA* = .14 (90% CI, .06, .24). The items were summed and averaged to form a scale (see Appendix G).

Reasons for disclosure. Twenty items (based on Derlega et al., 2000) were used to capture participants' anticipated motivations for disclosing genetic information to their selected blood relative. Participants were asked to rate how much various self-, other-, and relationship-oriented reasons would influence their decision to disclose the genetic test results to a blood-

relative. The response options were *strongly disagree*, *disagree*, *neutral*, *agree* and *strongly agree*, which were later coded for analysis (1 = *strongly disagree* to 5 = *strongly agree*). Of the 20 items, five were used to measure social support as a motive for disclosure (e.g., *My family member would give me the comfort I need*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .99, *SRMR* = .02, *RMSEA* = .07 (90% CI, .00, .14). Another five items were used to measure familial obligation (e.g., *If there is a chance that my family member might have the genetic mutation I want them to find out*). The CFA for a single-latent variable model produced these goodness-of-fit indices: *CFI* = .92, *SRMR* = .07, *RMSEA* = .17 (90% CI, .12, .23). The items for each motive factor were summed and averaged to form respective scales (see Appendix H). The two latent factors were significantly correlated (*r*= .65).

Primary motive. In addition, participants were also asked to identify their primary reason for disclosure based broadly on Derlega et al.'s (2000) measure of self-, other-, and relationship-oriented reasons for disclosure. Participants were asked to choose a primary motive from the following list: *to attain support or help in managing your diagnosis, a duty to inform and/or protect blood-related family members, a desire to educate other of your diagnosis, to vent or share feelings of distress, the family member would find out anyways, it seemed like the right thing to do,* and *the family member is available to me.*

Table 2

*Descriptive Statistics for Scales (*N = *173)*

Variable	*M*	*SD*	α	Skewness	Kurtosis
Genetic stigma	2.21	0.70	.92	0.27	-0.50
Preparation	2.55	0.79	.76	0.47	0.09
Prognosis	3.98	0.63	.78	-0.73	2.26
Symptoms	3.76	0.55	.62	-0.49	0.43
Relevance	3.42	0.83	.71	-0.54	-0.03

Relational quality	4.48	0.57	.84	-1.17	1.23
AS: emotional	4.38	0.59	.77	-0.73	-0.40
AS: informational	4.18	.69	.75	-0.69	.26
AS: instrumental	4.47	0.63	.81	-1.26	1.70
AR: discloser/receiver-oriented outcomes	2.07	0.65	.77	0.96	1.64
AR: relationship-oriented outcomes	1.75	0.79	.76	0.79	-0.14
AR: other relationship-oriented outcomes	1.99	0.57	.94	0.67	0.38
Confidence in response	4.08	0.64	.85	-0.13	-0.62
Disclosure efficacy	3.70	0.94	.85	-0.31	-0.57
Likelihood of future disclosure	4.36	0.64	.81	-0.59	-0.76
Disclosure for social support	4.34	0.70	.91	-0.94	0.45
Disclosure as family obligation	4.17	0.71	.80	-0.56	-0.39

Analysis Plan

Data were analyzed by multiple correlations, independent sample *t* tests, one-way ANOVAs, and hierarchical multiple regressions using SPSS 22.0 and AMOS 22.0. With a sample size of 194 and presuming an $\alpha = .05$, the most complex regression (for H1-H3), which has 14 independent variables [three of which are subcomponents of anticipated support and three of which are subcomponents of anticipated outcomes], needed to produce a medium effect size ($f^2 = .15$) to have power of .95. Results are presented by research question and hypothesis in Chapter 3.

Chapter 3

Results

Descriptive Statistics

At the start of the study, participants reported if the following blood-related family members were alive and known: mother (99%), father (98%), brother (59%), sister (59%), aunt (92%), uncle (94%), female cousin (93%), and/or male cousin (93%). When asked to choose one of the above listed family members as the recipient of the hypothetical disclosure, a little more than half of the participants chose their mother (54%); whereas the other half of the sample selected their father (20%), brother (11%), sister (10%), cousin (3%), aunt (1%), or uncle (1%). A minority of participants were familiar with genetics, either by taking a course in college (32%) or through genetic testing (7% had personally been tested and 19% had a family member undergo testing). Only 7 individuals (4%) had previously heard of AATD.

On average, participants agreed that it would be easy for others to notice AATD-related symptoms ($M = 3.77$, $SD = 0.55$), perceived AATD as a serious prognosis ($M = 3.98$, $SD = 0.63$), and believed that AATD test results would be relevant to a blood-relative ($M = 3.42$, $SD = 0.83$). Participants did not report a genetic stigma ($M = 2.21$, $SD = 0.70$) and did not feel prepared for the diagnosis based on the scenario described ($M = 2.55$, $SD = 0.79$). Participants reported, on average, positive relationships with their specified blood-relative ($M = 4.48$, $SD = 0.57$) and anticipated their relative to give emotional support ($M = 4.38$, $SD = 0.59$), informational support ($M = 4.18$, $SD = 0.69$), and instrumental support ($M = 4.47$, $SD = 0.63$). Furthermore, on average, participants anticipated less negative discloser/receiver-oriented outcomes ($M = 2.07$, $SD = 0.65$), relationship-oriented outcomes ($M = 1.75$, $SD = 0.60$), and other relationship-oriented outcomes ($M = 1.99$, $SD = 0.57$). Participants reported some efficacy to disclose ($M = 3.70$, $SD = 0.94$) and agreed that they were likely to disclose the results to their relative ($M = 4.36$, $SD = 0.64$).

Demographic variables were tested with the variables within the DD-MM for mean differences. One-way ANOVAs and independent sample t-tests were used to test differences among participants' class status (e.g., freshman or sophomore), age, gender, knowledge and experience with genetics, and number of siblings.

Class status. The ANOVA tests with class status as the independent variable were only statistically significant for condition relevance, F (3, 167) = 2.12, $p < .05$. Tukey post-hoc comparisons of the four class groups indicate that freshman (M = 3.00, SD = 0.86) thought AATD was significantly less relevant to their selected family member than juniors (M = 3.57, SD = 0.84), $p < .05$. All other comparisons between the class status groups were not statistically significant at $p < .05$.

Age. There was a statistically significant correlation between age and anticipated other relationship-oriented outcomes, r (172)= -.17, $p < .05$. ANOVAs were also used to test for differences based on age (treating age as a categorical variable). Assessments on condition preparation differed with statistical significance across the six age groups, F (5, 166) = 2.51, $p <$ .05. Tukey post-hoc comparisons of the six groups indicate that 18 year olds (M = 3.00, SD = 0.88) felt statistically significantly more prepared than 19 year olds (M = 2.08, SD = 0.64), $p <$ .05. All other comparisons between the age groups were not statistically significant at $p < .05$.

Gender. Males and females showed statistically significant differences for a number of DD-MM related variables. Females (M = 4.59, SD = 0.59) reported closer relationships with the family relative than males did (M = 4.36, SD = 0.59); t(170) = -2.66, $p < .05$. Females reported more anticipated informational support (M = 4.32, SD = 0.57) and instrumental support (M = 4.60, SD = 0.54) from their chosen family member than males did (M = 4.02, SD = 0.78 for informational), t(170) = -2.84, $p < .05$ and (M = 4.31, SD = 0.68 for instrumental) t(170) = -3.08, $p < .05$ respectively. Females reported less anticipated negative outcomes for their relationship

with their blood relative (M = 1.62, SD = 0.55) and for their relationship with others (M = 1.89, SD = 0.55) than males did (M = 1.90, SD = 0.63 for relationships with blood relative), $t(170)$ = 3.08, $p < .05$ and (M = 2.10, SD = 0.58 for relationship with others), $t(170) = 2.42$, $p < .05$ respectively. Finally, females (M = 4.46, SD = 0.62) reported greater likelihood to disclose than males did (M = 4.26, SD = 0.66), $t(170) = -2.01$, $p < .05$.

Previous Genetic Experiences and Siblings. There were no statistically differences between those that have taken a college course on genetics and those that have not across any of the variables within the DD-MM. Confidence in response differed statistically significantly between those that have personally undergone genetic testing (M = 4.47, SD = 0.48) and those that have not (M = 4.06, SD = 0.64), $t(170) = 2.14$, $p < .05$. There were, however, no statistically significant differences between those that have had a family member undergo genetic testing and those that have not across any of the variables within the DD-MM. There were no statistically significant correlations or differences among the number of siblings a participant had and any of the variables within the DD-MM at $p < .05$.

Hypothesis Testing of the DD-MM

Hypotheses 1-3 tested hypotheses posed by the DD-MM, corresponding to the three assessments predicting a person's likelihood to disclose their AATD-related genetic test results to a blood-related family member. A three-block, hierarchical, regression analysis was performed with likelihood to disclose as the dependent variable. The information assessment variables (stigma, preparation, prognosis, symptoms, and relevance) were entered in the first block, the receiver assessment variables (relational quality, anticipated emotional/instrumental support, anticipated informational support, anticipated outcomes, and confidence in response) were entered in the second block, and disclosure efficacy was entered in the third block of independent variables. All independent variables were examined for collinearity based on Menard's (1995)

criteria that a tolerance of less than 0.10 (or a variance inflation factor (VIF) greater than 10) indicates a serious collinearity problem. Results of the collinearity tolerance (all greater than .35) and VIF (all less than 3.0) provided support that collinearity was not a concern and suggested that the estimated βs are well established in the regression model.

The full DD-MM in which information assessment, receiver assessment, and disclosure efficacy predict disclosure likelihood (Model 3) was statistically significant, $R^2 = .48$, $F(14, 158) = 10.22$, $p < .05$. The regression estimates appear in Table 3.

Hypothesis 1 predicted that decreased stigma, more serious prognosis, increased symptom visibility, and greater relevance predicted a greater likelihood of disclosing AATD genetic test results. As a reminder, while preparation is predicted to facilitate disclosure in DD-MM, preparation was expected to be small and not to vary in this sample, given the surprise, hypothetical scenario; it was very small and not statistically significant. The beta coefficient was in the predicted direction, nonetheless. The beta coefficients for stigma, prognosis, and symptoms were statistically significant, in the predicted direction, and showed a strong association to disclosure likelihood. Relevance did not. The beta coefficient for relevance was neither statistically significant nor in the right direction. As such, the results of the regression analysis provided partial support for Hypothesis 1.

Hypothesis 2 predicted that better relational quality, positive anticipated reactions (support and outcomes) and more confidence in receiver response predict greater likelihood of disclosing AATD results. The beta coefficients for emotional, informational and instrumental support, confidence in response, and anticipated discloser/receiver- and other relationship-oriented outcomes were all in the predicted direction; interestingly, the beta coefficients for relational quality and anticipated relationship-oriented outcomes were unrelated. The beta coefficient for anticipated instrumental support was statistically significant with strong

association to disclosure likelihood. The beta coefficients for relational closeness, anticipated emotional support, anticipated informational support, confidence in response, and all factors of anticipated outcomes were not statistically significant and did not indicate strong associations to disclosure likelihood. The results of the regression analysis provided partial support for Hypothesis 2.

The third and final assessment, assessment of disclosure efficacy, added disclosure efficacy to complete the model. Hypothesis 3 predicted that stronger disclosure efficacy predicted greater likelihood of disclosure. The beta coefficient for disclosure efficacy was in the predicted direction but was not statistically significant. Hypothesis 3 was not supported.

Table 3

*Summary of Hierarchical Regression Predicting Likelihood of Disclosure (*N = *173)*

	Likelihood of Disclosure								
	Model 1			Model 2			Model 3		
Variable	*B*	*SE*	β	*B*	*SE*	β	*B*	*SE*	β
Block 1									
Stigma	-.33	.06	-.36*	-.10	.07	-.11	-.05	.05	-.06
Preparation	-.05	.06	-.06	-.05	.05	-.06	.04	.08	.04
Prognosis	.20	.08	.20*	.15	.07	.14*	.16	.07	.16*
Symptoms	.23	.09	.20*	.08	.08	.06	.04	.08	.12
Relevance	-.01	.06	-.01	-.06	.05	-.08	-.05	.05	-.06
Block 2									
Relational Quality				-.01	.09	-.01	.00	.08	.00
AS: emotional				.21	.11	.19	.17	.11	.25
AS: informational				.01	.07	.01	-.00	.07	-.00
AS: instrumental				.24	.10	.23*	.27	.10	.26*
AR: discloser/receiver-oriented outcomes				-.08	.08	-.08	-.07	.08	-.07
AR: relationship-oriented				.03	.12	.03	.01	.12	.01

outcomes						
AR: other relationship-oriented outcomes	-.14	.11	-.13	-.13	.11	-.11
Confidence in Response	.11	.09	.11	.08	.09	.08
Block 3						
Disclosure Efficacy				.08	.05	.11

Note. First block: change in $R^2_{change} = .22$, $F_{change}(5, 167) = 9.66$, $p < .05$; Second block: change in $R^2_{change} = .24$, $F_{change}(8, 159) = 9.01$, $p < .05$; Third block: change in $R^2_{change} = .01$, $F_{change}(1, 158) = 2.70$, *ns*. Full model*:* $R^2 = .48$, $F(14, 158) = 10.22$, $p < .05$.
* $p<.05$

Research Question 1: Familial Obligation as a Motive to Disclose

Research question 1 explores the role familial obligation has, if any, on motivating people to disclose their genetic test results. To address this question, participants were asked (a) to choose their primary motivation for potentially disclosing their AATD status and (b) to fill out scales related to motives.

For the primary motivation question, over a quarter (29%) of participants indicated that a duty to inform and/or protect blood-related family members was their main motive for disclosure and 24 (14%) participants reported that their primary motivation was that it seemed like the right thing to do. Other primary motivations included to attain support or help in disease management (44%), as a means to vent or share feelings of distress (7%); to fulfill a desire to educate others about AATD (2%); because the family member would find out anyways (2%); and because the family member was available to them (2%).

For the scales, on average participants agreed with social support motives ($M = 4.34$, $SD = 0.70$); they also agreed with familial obligation motives ($M = 4.17$, $SD = 0.71$). Table 4 shows the zero-order correlations among the two motivations and the likelihood of disclosure. The zero-order correlations showed that a familial obligation motive to disclose is positively related to

disclosure. These findings provide a preliminary answer to Research Question 1: familial obligation is positively related to disclosure likelihood.

In addition, the correlations (see Table 4) show that social support motives and disclosure likelihood are also positively related. The direction of the correlation coefficient was in the predicted direction and there appeared to be a strong association between the two variables. The two motives were also strongly correlated, suggesting that these motives may be mutually supportive. Figure 1 shows the motives in the same plot.

Table 4.

*Correlations Among Reason for Disclosure and Likelihood of Disclosure (*N = *173)*

	1.	2.	3.	4.
1. Social support motive	--			
2. Obligation motive	.65*	--		
3. Primary motive (S-O)	.40*	-.44*	--	
4. Likelihood of disclosure	.59*	.47*	.13	--

Notes. No missing data.
$p < .05$ level

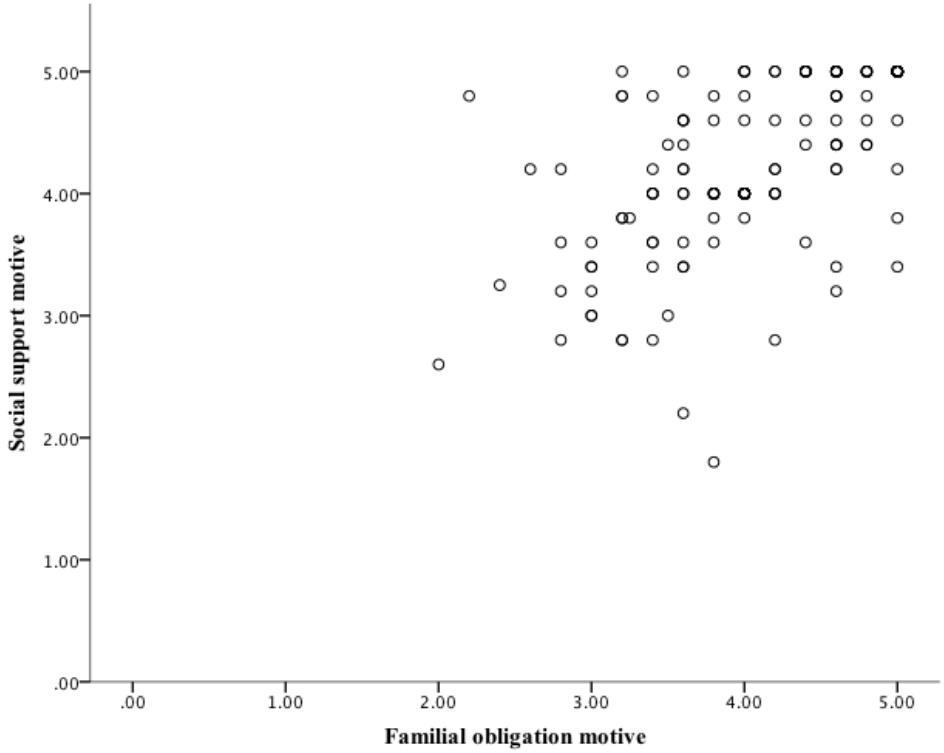

Figure 1. Scatterplot of participants' composite social support score by composite familial obligation composite score.

Finally, a discrepancy score was created, subtracting familial obligation scores from social support scores ($M = 0.17$, $SD = 0.59$). The correlation shows a small positive relation between relatively stronger social support motive and disclosure likelihood, but the relation was not statistically significant with disclosure likelihood. This finding suggests that, as a whole, participants are highly motivated and that these two motives for disclosure are not mutually exclusive. Furthermore, the discrepancy score may not be statistically significantly associated with disclosure likelihood because the difference between motive scores is minimal, indicating that either or both motives may be associated with disclosure. Figure 2 plots participants' primary motive with their disclosure likelihood; the pattern suggests that a person's likelihood to disclose is determined at random rather than by one disclosure motive over another (or something curvilinear).

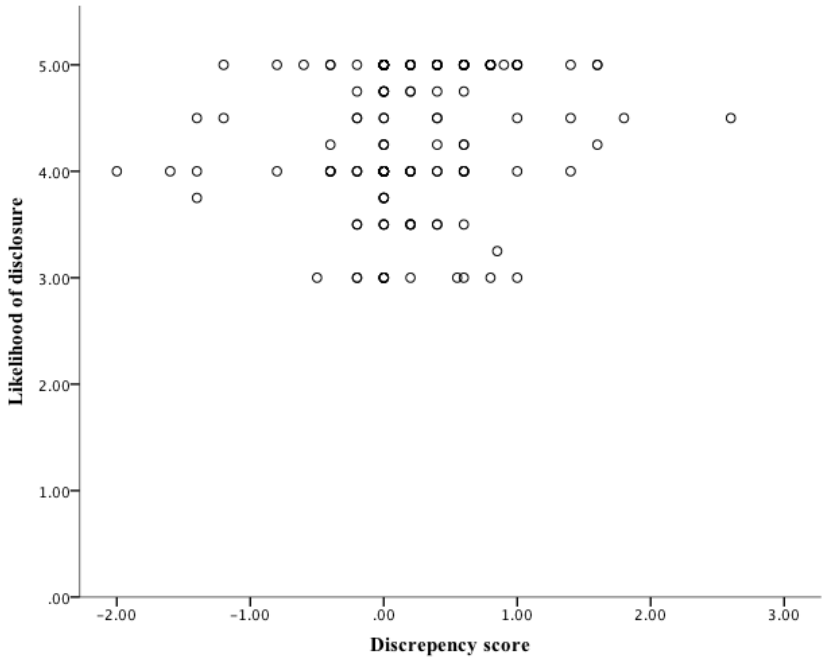

Figure 2. Scatterplot of participants' composite likelihood of disclosure score by their motivation discrepancy score.

Hypothesis Testing of Familial Obligation

Hypothesis 4 predicted that for those reporting a familial obligation to disclose, greater relevance and a more serious prognosis predict greater disclosure likelihood. To test Hypothesis 4, the sample was separated into those with a greater familial obligation motive for disclosure from those with a greater social-support motive for disclosure. Effect codes were derived from the motive-discrepancy score: those with a low score (≤ 0) were coded as -1 (stronger familial obligation motives, n =98) and those with a high score (> 0) coded as 1 (stronger social support motives, n =75). Importantly, these regressions have less power. The power to detect a large effect size (f^2 = .15) when α = .05, for a regression with 14 independent variables is .44 for the social support sample, .60 for the familial obligation sample.

The three-block, hierarchical multiple regression analysis with likelihood to disclose as the dependent variable was recalculated for the separate, motive sub-samples (see Table 5).

Hypothesis 4 posited that those reporting a familial obligation to disclose, greater relevance and a more serious prognosis predicted greater disclosure likelihood. The findings showed the beta coefficients for relevance and prognosis were positively predicted, indicating a greater relevance and a worse prognosis, but only prognosis was highly associated with one's likelihood to disclose The beta coefficient for prognosis was statistically significant, but those for relevance, stigma, symptoms, preparation, anticipated emotional support, anticipated informational support, anticipated discloser/receiver-oriented outcomes, anticipated relation-oriented outcomes, anticipated other relationship-oriented outcomes, relational quality, and disclosure efficacy were not. Unexpectedly, anticipated instrumental support and confidence in response were statistically significant. As such, these findings partially support Hypothesis 4.

For those reporting a greater want for social support as a motivation to disclose, three variables predicted disclosure likelihood with statistical significance: anticipated emotional support, anticipated instrumental support, and anticipated discloser/receiver-oriented outcomes were statistically significant. As a reminder, more serious prognosis and greater anticipated instrumental support, predicted disclosure likelihood with statistical significance with the full sample (Table 3). The differences between the entire sample and the sub-sample who had primarily a social support motive were many. Those in the sub-sample did not indicate that greater prognosis was a statistically significant predictor of likelihood to disclose. On the other hand, only in the sub-sample were anticipated emotional support and anticipated discloser/receiver-oriented outcomes statistically significant predictors of disclosure likelihood. These findings suggest that social support as a motive for disclosure, while prevalent, is not the sole reason for disclosing genetic test results and does not fully explain the results of the entire sample.

Table 5

Parameters from the Hierarchical Multiple Regression Predicting Likelihood of Disclosure Based on Disclosure Motive

Variable	Likelihood of Disclosure					
	Familial Obligation (n = 69)			Social Support (n = 57)		
	B	*SE*	β	*B*	*SE*	β
Stage 1						
Stigma	-.09	.10	-.09	.09	.11	.09
Preparation	-.04	.07	-.06	-.05	.08	-.06
Prognosis	.28	.11	.30*	-.11	.12	-.09
Symptoms	.01	.13	.01	-.17	.15	.13
Relevance	.07	.08	.09	-.14	.08	-.17
Stage 2						
Relational Quality	-.10	.14	-.09	-.14	.15	-.11
AS: emotional	.03	.21	.02	.57	.17	.41*
AS: informational	-.09	.13	-.02	.07	.12	.07
AS: instrumental	.30	.15	.30*	.56	.18	-.45*
AR: discloser/receiver -oriented outcomes	.23	.16	.25	-.41	.15	-.38*
AR: relationship-oriented outcomes	-.31	.21	-.33	.45	.24	.35
AR: other relationship-oriented outcomes	.03	.18	.03	-.36	.18	-.30
Confidence in Response	.29	.15	.30*	-.11	.14	-.11
Stage 3						
Disclosure Efficacy	.02	.08	.03	.06	.07	.09
R^2	.52*			.72*		

Note. $F(14, 54) = 4.23$, $p < .05$ for familial obligation. $F(14, 42) = 7.82$, $p < .05$ for social support.
* $p<.05$

Post-Hoc Analyses: Predicting Motives

Post-hoc analyses were performed to test for any differences in motives by demographic variables. None of the differences were statistically significant. For continuous variables, correlations were used. The predominant motive was not related with statistical significance to

age, $r = .12$, *ns*., or participants' number of siblings, $r = -.12$, *ns*.

Chi-square tests were used for categorical variables; the tests were not statistically significant for class status, X^2 (3, 171) = 2.69, *ns*, *Phi* = .13, gender, X^2 (1, 172) = 0.70, *ns*, *Phi* = .06, identifying as white, X^2 (1, 173) = .57, *ns*, *Phi* = -.06. The differences also were not statistically significant for living with other students (the most prevalent living situation reported in this study), X^2 (1, 173) = .1.47, *ns, Phi* = -.09, the relationship the participants' had to the blood-related family member selected for the potential disclosure (mother, sibling, etc.) were not statistically significant, X^2 (6, 173) = 10.40, *ns*, *Phi* = .24, previous enrollment in a college course covering genetics, X^2 (1, 172) = .74, *ns*, *Phi* = -.03, personal experience with genetic testing, X^2 (1, 172) = .22, *ns*, *Phi* = -.04, or the participant's family's experience with genetic testing, X^2 (1, 170) = .01, *ns*, *Phi* = -.01.

Chapter 4

Discussion

The goal of this thesis was threefold. First, the study investigated whether variables included in the DD-MM (Greene, 2009) would predict participants' likelihood to disclose an Alpha-1 diagnosis to blood-related, family relatives. Second, this project sought to explore whether familial obligation would be a motive for disclosure of test results for a genetic condition like AATD. Third, I wanted to understand how familial-obligation motives, if present, would influence which DD-MM variables would predict disclosure. The findings showed that three of the five information assessment factors—stigma, prognosis, and relevance—statistically significantly predicted likelihood of disclosure, partially supporting Hypothesis 1. Hypothesis 2, which focused on the factors of one's relational assessment, was also partially supported since the sub-factor of anticipated support, anticipated instrumental support, is a statistically significant predictor of disclosure likelihood. Hypothesis 3 was not supported, because disclosure efficacy was not a statistically significant predictor. However, the full DD-MM, in which information assessment, receiver assessment, and disclosure efficacy were tested together, appears to fit the genetic context and statistically significantly predicts disclosure likelihood.

The findings also showed that familial obligation was reported as a reason for disclosure and as a primary motivator for disclosure. When participants were split based on how much the agreed on a particular motive, familial obligation or social support, the relative importance of certain DD-MM variables changed. Those in the familial obligation sub-sample reported that prognosis, anticipated instrumental support and confidence in response predict likelihood to disclose; whereas those in the social support sub-sample indicated that anticipated emotional and instrumental support and anticipated discloser/receiver-oriented outcomes are statistically significant predictors of disclosure likelihood. The proceeding section will reflect on how

Greene's (2009) DD-MM fits the genetic context and how this study's results compare with previous work on the DD-MM.

DD-MM and Disclosing Genetic Test Results

Genetic conditions, such as alpha-1 antitrypsin deficiency, are an important context for studying disclosure decision-making because it raises the question of how ethics, morality, power, the self and others' protection are all factors that may influence how people and their families manage private information about health. The DD-MM (Greene, 2009) was used as a framework for identifying the components of the disclosure decision-making process that might predict disclosing genetic test results. Overall, the results showed mixed support—not all key mechanisms identified in the DD-MM influenced disclosure likelihood. Assessing genetic test results for information relating to a poor prognosis and assessing the potential target recipient for the anticipated amount of instrumental support he or she is able to offer showed to be statistically significant predictors of disclosure likelihood. Disclosure efficacy, which is the central focus of the third assessment, never appeared to influence the likelihood of disclosure. Each of these three assessment stages of the DD-MM are described, below, in further detail.

Information Assessment. There was general support for Hypothesis 1, which represented the first assessment stage: information assessment. The model posits that perceptions of stigma, prognosis uncertainty, and/or disease progression and symptom visibility decrease intentions to disclose. In contrast, when the diagnosis is perceived to be relevant to others, people are more likely to disclose, especially if the diagnosis can be passed to others via transmission or genes (Greene, 2009). The model is uncertain, however, about how preparation is to influence intentions to disclose. Greene (2009), when developing the model, did not assume all five aspects (stigma, preparation, prognosis, relevance, and symptoms) were intended to represent an ordered process, occur simultaneously or at all. The present findings support the latter notion since three

of the five information assessment factors, stigma, prognosis, and relevance, were statistically significant predictors of likelihood of disclosure, but only stigma resulted in the direction Greene (2009) hypothesized. Greater perceptions of stigma negatively predicted likelihood of disclosure. In contrast, perceptions of a more severe prognosis and increased symptom visibility positively predicted disclosure likelihood.

These findings are particularly interesting since they conflict with what Greene et al. (2012) found when testing the DD-MM with nonvisible illness. They found that those factors specifically referring to the severity of the health information (i.e., stigma and prognosis) predicted decreased disclosure efficacy and speculated that it may be due to the difficult nature of sharing about a disease that is progressing poorly or is stigmatized with others. Sharing depressing information or information that may threaten one's identity may not be done in hopes of protecting others and/or themselves (e.g., Derlega et al., 1987; Goldsmith, 2009). This explanation does not fully apply to the current findings. Although genetic stigma negatively predicted likelihood of disclosure, which partially speaks to this proposed explanation of results, perceptions of a more serious prognosis positively predicted disclosure likelihood. This, of course, could be due to the context of genetic information. Greene (2009) has previously suggested that when a person is assessing information about a condition, there is significant concern that the symptoms and prognosis will passively "out" the person. This may explain why prognosis and symptoms positively predict disclosure likelihood. Disclosure is a reaction to the manifestation of the genetic condition and "a sense of inevitability" (Greene, 2009) that disclosers may feel.

Preparation showed little relationship with disclosure likelihood; and, across the regressions, relevance showed virtually no relationship with disclosure likelihood. Both of these findings may be explained by the design: the study focused on a low preparation situation, in

which college students were asked to consider their test results after visiting their doctor for chronic wheezing, and participants were asked to select a blood relative for the disclosure, fostering relevance to the target recipient. However, it is important to note that Venetis et al. (under review) found similar results for these information assessment factors.

Receiver assessment. Hypothesis 2, representing the variables in the second stage of assessment, receiver assessment, was partially supported. Greene et al. (2012) tested a hypothesized DD-MM for undisclosed information that broke down receiver assessment into several factors, including: relational quality, anticipated support, anticipated outcomes, and confidence in response. Magsamen-Conrad (2012), when testing the implications anticipated response has on sharing personal information in relationships, further refined receiver assessment by defining three dimensions of anticipated support (i.e., emotional, informational, and instrumental) and operationalized anticipated outcomes to include four categories (i.e., discloser-oriented outcomes, receiver-oriented outcomes, relationship-oriented outcomes, and other relationship-oriented outcomes). It is expected that more positive assessments of relationship quality, anticipated emotional, instrumental, and informational support, anticipated discloser-, receiver-, relationship-, and other relationship-oriented outcomes, and confidence in response predict greater likelihood of disclosure. More specifically, Greene (2009) hypothesized that anticipated support, if positive, would lead to disclosure even if the information assessed were stigmatized.

The present study tested measures of relational quality, anticipated support (emotional, informational, and instrumental), three types of anticipated outcomes (discloser/receiver, relationship, and other relationship) and confidence in response. Unlike Greene et al. (2012) and Magsamen-Conrad (2012), results of the study showed that only anticipated instrumental support was a statistically significant sub-factor of receiver assessment that predicted disclosure

likelihood. This indicates that genetic conditions such as AATD may directly affect genetic relatives and, as a result, not require a positive relationship with the information recipient in order for disclosure to happen. It could also be that the other assessments of receiver, in addition to instrumental support, are superfluous to the genetic disclosure decision-making process. For instance, informational support may not be needed from others since the concept of a surrogate information-seeker (a person who seeks health information on behalf of others) works differently in the genetic health context. Since AATD could be a threat to both the discloser and the receiver, a surrogate information-seeker, in this context, does not necessarily embody the qualities typical of someone in this role such as being likely to be both married and a parent, having good or excellent health, being a caregiver of an adult relative, and having someone close with a serious medical condition (Sadasivam et al., 2013). Therefore, informational support is not always a viable option within the family.

Furthermore, Magsamen-Conrad (2012) points out that there is an assumption in literature that disclosers want "positive" responses and that there are certain forms of responses that are inherently positive. An example of this is that many assume that positive signs of emotional, instrumental, or informational support directly predict positive disclosure decisions. However, people could simultaneously expect an initial negative emotional reaction as well as instrumental support and, as a result, may feel less efficacy to disclose and/or decreased inclinations of disclosure likelihood (Magsamen-Conrad, 2012). The current study's results that only positive anticipated instrumental support statistically significantly predicts disclosure likelihood is indicative that these assumptions of support need to be further tested to determine if they are wrong or just context-specific.

Efficacy assessment. Further support that these assumptions need to be tested is that disclosure efficacy was one of the key mechanisms not statistically significant in the genetic

disclosure decision-making process. Hypothesis 3, which focused on the third assessment—disclosure efficacy, was not supported. The model posits that disclosure efficacy positively predicts likelihood of disclosure (Greene et al., 2012), although greater uncertainty about one's health condition may decrease the importance of disclosure efficacy (Checton & Greene, 2012). The present findings did not support this hypothesis.

This statistically non-significant result in this thesis contrasts with previous research testing the DD-MM. Essentially all previous studies testing the model showed that disclosure efficacy was a statistically significant predictor of disclosure intentions and/or behaviors when applied to several health conditions, including HIV/AIDS (Greene et al., 2013), heart disease (Checton & Greene, 2012), nonvisible illness (Checton et al., 2012), cancer (Venetis et al., in press), and infertility (Steuber & Solomon, 2011). More notably, the pilot study (Greenberg & Smith, 2013), which tested many of the current study's measures in the same genetic context, found that disclosure efficacy positively predicted one's breadth of disclosure. The contrast this study's findings have pertaining to disclosure efficacy with all other studies suggests that further research must be done, focusing on the disclosure of genetic test results. As of now, it is unclear whether disclosure of genetic information is a unique context from the other health settings in which increasing disclosure efficacy is not a method for increasing disclosure likelihood. It could be that the statistical significance of prognosis and instrumental support are enough to motivate a person to disclose genetic information, information that is applicable and has direct bearing on both the discloser and the target recipient.

These results indicate that the DD-MM takes on a different form and structure when applied to genetic information disclosures, which may be associated with the inclusion of other motives for disclosure in addition to want of social support. The next section will discuss the presence of another motive in the genetic disclosure decision-making process, familial obligation.

Familial Obligation as Motivation to Disclose

As previously discussed, there is debate about who has the right to share information about a genetic test in families at risk for genetic disease. The debate is not only centered on ownership rights to the genetic information, but is also about the ethical and moral obligations related to revealing or concealing information that could impact the health of family members. It was hypothesized that the results of testing the DD-MM in this new health context may be guided by the appearance of other motivations or reasons for disclosure other than for self-oriented needs. Research Question 1 inquired whether these other types of motivation, namely familial obligation, did exist and were present in participants' responses and assessments. The data provided support that familial obligation did exist as a motivation and, as it happens, was the second most common answer participants gave when asked to choose their primary motivation.

Although familial obligation has not been previously examined in relation to the DD-MM, or any other disclosure model, it may very well be that the special nature of genetic information makes this context particularly inviting for familial obligation as a motivator. The profound and direct effect the information from genetic test results has on both the person receiving them and their blood-related family member(s) introduces other reasons for disclosure that focus on the needs of those other than the discloser (e.g., familial obligation, duty to inform, desire to educate). The presence of familial obligation, in this study, challenges assumptions made by information management theories such as the DD-MM. Although expectations that self-oriented reasons for disclosure (i.e. attainment of help and support) are reinforced, the existence of familial obligation as a driving force for disclosure begs for further exploration both in this health context and others (e.g., infectious, chronic) for further model refinement.

Differences in the DD-MM Associated With Motivation

There was general support for Hypothesis 4, which represented the motive-category, familial obligation. The model assumes that social support and key reward and motive driving disclosures. Hypothesis 4 predicted that for those reporting a familial obligation to disclose, greater relevance and more prognosis certainty and severity would lead to greater disclosure likelihood. The present findings provide evidence that not only does familial obligation exist as a motive for disclosure in the genetic context, but that it is associated with a different disclosure decision-making process than that of social support. When the sample was split by motive-category, results showed that a person's assessment of information only predicted disclosure likelihood when a greater sense of familial obligation was indicated as a reason for disclosure. Specifically, a more serious prognosis was strongly, positively related to disclosure likelihood in the full sample at all stages, and for the familial-obligation sub-sample. Relevance, on the other hand, was not a statistically significant predictor at any stage in the full sample or for the familial-obligation sub-sample. As such, Hypothesis 4 was only partially supported.

In addition to a more serious prognosis, greater anticipated instrumental support and more confidence in response were statistically significant predictors of disclosure likelihood in the familial-obligation sub-sample. The two other information assessment variables, symptom visibility and genetic stigma, showed their predicted effects in the first stage with the full sample, but diminished to virtually non-existent relationships in the other models. Although it was not hypothesized that these two variables would predict disclosure likelihood, it was thought that the factors within information assessment would be more closely tied to familial obligation. Therefore, it is interesting that their statistical significance reduced during the testing of the second and third stage with the full sample and became nonexistent for the familial obligation sub-sample.

However, it is not surprising that the theoretical variable most closely tied to social support, anticipated reactions, which falls in the second assessment (i.e., receiver assessment), was the only statistically significant predictor of disclosure likelihood in the social support sub-sample. Results indicated that more anticipated emotional support, less anticipated instrumental support and less negative anticipated discloser/receiver-oriented outcomes are quintessential for social support and predicting disclosure likelihood. This may be, in part, due to the young adult sample used for analysis. Young adults have been shown to hold essentialist beliefs about genetics (Smith, Greenberg, Parrott, 2014), which means this population may require less informational support since essentialists believe genes determine disease, health, and wellbeing (Parrott, Kahl, Traeder, & Ndiaye, 2012) and participants may find their diagnosis both personal and predetermined. It could also be that the offering of instrumental and/or informational support could be seen as an example of "unhelpful" social support (Barbee, Rowatt, & Cuningham,1998), causing young adults to feel "smothered" by the offering of that type of assistance.

Motivated Young Adults

Results from both motive sub-samples provide evidence that young adults are highly motivated to disclose and that there is more than one reason for disclosure of genetic test results. This strong motivation was surprising. It may result from the hypothetical nature of the experiment; it may reflect how young adults have been socialized to think about genetic testing and its disclosure; or it may reflect the generalized, sociological changing in sharing (e.g., through social media) and salience of reasons to share.

Results also show that the reasons for disclosure, social support and familial obligation, are not mutually exclusive. Derlega et al. (2008) came to similar conclusions when disclosers reported that close relationships are associated with being a ''safe place,'' with trust as an

attribution for disclosure and fear of losing the other's respect and desire for privacy as reason for nondisclosure Consider that concern for discloser/receiver-oriented outcomes was a statistically significant predictor of disclosure likelihood for those in the social support sub-sample. However, when it came down to it, participants rarely shared with Derlega et al. (2008) that putting the relationship at risk was a reason for not disclosing to various partners.

In this study, participants reported that they were more likely to disclose to a blood relative for reasons of social support when they anticipated more emotional and instrumental support, but anticipated relationship- and other relationship-oriented outcomes were not statistically significant predictors of disclosure likelihood for any model tested. The results also indicate that there are other disclosure motives available and significant, since familial obligation, social support, or the combination of the two motives do not fully explains the results of the full model tested with the entire sample. Post hoc analyses indicate that differences in motives by family relation and demographic data are statistically significant.

Possibly as a function of young adulthood, the majority of the sample (74%) chose either their mother or father as a blood-relative to potentially disclose to. Interestingly, familial obligation was the greater motive for disclosure when participants chose either parent. In regards to home life, it was only the subsample of those reporting to live with a husband/wife/significant other/domestic partner that familial obligation was not the greater motive for disclosing. Again, this may be a function of young adulthood. It could also be that the concept of family and feelings of responsibility shift when a person is beginning their own family. The want for social support from family members, instead of feelings of obligation, may increase as the young adult begins a life with a significant other.

In reference to demographics, analyses indicated that all ethnicities (i.e., American Indian or Alaskan, Hispanic, Asian, and White) except Black or African American felt familial

obligation as a greater motive for disclosure to a genetic relative than social support. Studies have consistently shown that disclosure is a gendered responsibility. Women feel responsibility for maintaining the health of children and partners; as well, they negotiate professional health care for their families and communicate health information on their behalf (d'Agincourt-Canning, 2001). In fact, having a female relative has been shown to be a predictor of having a disclosure conversation about family health history (Parrot et al., in press) Post-hoc analyses further support these claims. Females reported being more motivated by a sense of familial obligation than their male counterparts. This would suggest that females do feel a profound sense of responsibility to family such that they would disclose to other blood-related family members in order to warn, protect, and/or foster a greater sense of physical and mental wellness.

This study could offer important insights into DD-MM and how well DD-MM extends to the genetic disclosure context. The rest of this chapter discusses the implications of these findings in greater detail.

Limitations

This project was limited in several ways, including sampling, procedure, and design limitations. Sampling limitations are discussed first.

The quality of a sample is based on both its size and its ability to represent the larger population on the characteristic of interest. In this vein, all efforts were made to obtain a sample size that corresponded to the one calculated from the power analysis (N=194) and was sufficient for analyses. Unfortunately, this sample size could not be obtained, which made measure construction and the detection of statistically significant results more difficult. For instance, certain measures did not sufficiently satisfy all CFA cutoff criteria, which, as previously mentioned, could be due to the small sample size (Hu & Bentler, 1999). Moreover, certain

mechanisms and factors that were approaching statistical significance may play a statistically significant role in the DD-MM when tested in the future with a larger sample.

Another limitation of the sample is generalizability. Ideally, the sample should be representative of the population from which it is drawn so that conclusions from the sample are applicable to the entire population. Although it is hoped that this study's findings are indicative of the disclosure decision-making process of any person with a genetic condition, it is uncertain if they do. The genetic context is rather complex and the evaluation of information regarding one's test results and/or diagnosis and their evaluation of the information recipient may make it so generalizations across genetic conditions cannot be made. It may be that those with sickle cell disorder, in which symptoms can be relatively nonvisible, evaluate information and their efficacy to disclose much differently than those with a deleterious gene mutation for breast cancer, hypercholesterolemia or Huntington's disease. The weight and importance attributed to each assessment or its subfactors based on the specific genetic condition may alter the disclosure process as a whole. Thus, the findings may not generalize to disclosing about other genetic conditions other than Alpha-1.

Furthermore, these data were collected in one state in the northeastern United States and it is unclear whether results would generalize to other areas or countries. Similarly, cultures vary in how strong familial obligation is emphasized in the family unit and families are complicated interactive social units that can vary in norms (Sorenson & Wertz, 1986; Sorenson, Jennings-Grant, & Newman, 2003). Deference to authorities or hierarchies in families (Forrest et al., 2003, p.321) can impact who gets disclosed to and why. For example, a grandmother may be disclosed to first because she is seen as having more authority than an aunt, even though she is not at genetic risk herself while the aunt is. My study is limited in that my measures do not report on these discrepancies and family norms.

In regards to procedure, it should be noted that despite similarities between regression and path analysis, there are limitations by performing the former as opposed to the latter. Regression methods require mutually exclusive categorization of variables, either independent or dependent. This is not the case for path analysis and structural equation modeling (SEM)—variables could be independent and dependent simultaneously. As such, SEM is considered a statistically efficient means to test a model since it requires only a single comprehensive method (Hair, Anderson, Tatham, & Black, 1998). Although SEM would be the preferred method for analyzing this study's data (and has been the most common method for other DD-MM studies), regression was the applied method of analysis due the statistical knowledge and skill-level of the researcher. This means that the validity in this study was tested by separately measuring each construct and discriminant validity was only assumed. Therefore, the use of regression methods makes it difficult to draw conclusions about the full measurement model.

Moreover, several of the scales used in this study were scales developed specifically for the genetic context and had only been tested in the pilot study. As such, these scales had minimal testing or verification before use in this study. Future research utilizing these scales will need to be done to further validate them.

Finally, there are profound limitations of study design that should be noted as well. The study required participants to consider a hypothetical scenario in which they were tested and diagnosed with AATD. Participants were then prompted to respond to questions about their disclosure decision-making process in terms of disclosing to a genetic relative. The use of a hypothetical scenario, however, makes it difficult to capture the true strength of the relationships between variables. Participants were not actually burdened with Alpha-1 or the decision to disclose to a genetic relative and consequently may not respond the same way to the measures as someone who is faced with the reality of being diagnosed with a genetic condition.

Second, this was a cross-sectional study such that data was collected, simultaneously, at only one point in time. However, disclosure within relationships is an ongoing dyadic process (Greene, 2009). Because there was no collection of data before the participant was hypothetically diagnosed with AATD, there is no way of knowing whether the diagnosis changed their disclosure-decision-making patterns. Additionally, because data was collected via survey methods instead of experimental methods, no causal claims can be made for the relationship between disclosure about genetic disease risk and one's motivation to disclose. Therefore, using cross-sectional data limits conclusions to only those of association between variables in the model and one's motivation to disclose since determining temporal precedence is not possible. Furthermore, this type of study design does not provide information on intraindividual change so that information on how one's assessments and motivations to disclose evolve over time. Although found less often in information management studies, a design that allows for tracking disclosure decisions across a period of time may have been more advantageous.

Another potential limitation of the current study is the absence of items measuring the participants' sense of personal responsibility, genetic beliefs, family orientation and communication patterns, and cultural consonance prior to exposure to the AATD scenario and factsheet. These concepts could better inform the profile of those that are likely to disclose genetic risk information and for each motivation. Further, it is possible that any or all of these variables are cofounders that, if measured, could be better labeled as moderating or mediating variables.

Despite these limitations in sampling, procedure, and design, the project offers potential for future research.

Future Research

Studying disclosure is essential for the comprehension of how health information is managed in terms of illness and relationships. Currently, there is limited knowledge on the disclosure decision-making process on the consistency across disclosure decisions, not just if people disclose (or keep secrets or avoid). There is even less research on the disclosure decision making process regarding the disclosure of genetics nor is there any literature on the intergenerational disclosure decision-making process from the young adult perspective. This study attempted to examine both, the young adult's genetic disclosure decision-making process when deciding to disclose to a blood relative such as one's biological mother or father. However, this process is only looked at using a hypothetical scenario, which limits our understanding of how social support and/or familial obligation influence the disclosure decision-making process. Since it is so important to understand what motivates young adults who have been diagnosed with a genetic mutation to communicate their risk to others, future research could recruit young adults that have been tested positive for a genetic mutation such as AATD and examine their genetic disclosure decision-making process, examining the experience either when genetic information has or has not been previously shared.

Furthermore, future research could explore other components of the decision-making process, especially from the other end of the disclosure process. Some concepts to consider are regret and/or satisfaction in relation to the primary motivation(s) stated for disclosure. Studies including these concepts would not only contribute more to the health information disclosure literature but to information management literature, in general. Results could also provide insight for the construction of health intervention materials and decision aids.

It has been previously established that disclosing genetic information is a crucial decision for people, especially in the family context. Such disclosures are considered to be personalized

risk messages as well. However, while the DD-MM does position level of risk as a determinant for why people disclose, risk has only been looked at generally in terms of information assessment and disclosure for health information (or, in this study for genetic information) and not as a means to compare the disclosure experience across different specific health or genetic conditions. Doing so would put in perspective how much information assessment for a specific diagnosis influences the disclosure process.

Similarly, this study only examines the disclosure process of young adults. However, there are many individuals diagnosed with a genetic mutation post young adulthood. The diagnosis experience, motivation to disclosure, and the disclosure decision-making process can be very difficult for a young adult just establishing themselves and their independence versus an older adult that is more established in their career and has a family of their own. The functions served by self-disclosure may vary depending upon the gender and stage in lifespan of the discloser (Parker & Parrott, 1995). Future research should investigate how people of different age groups are motivated to disclose and make disclosure assessments.

Finally, the literature discussed in this study makes prominent several elements that may affect self-disclosure that have not yet been methodically studied. Issues such as ethics, morality, power, and self and other protection are all factors that may influence how individuals and families manage private information about health which need further exploration. These elements could all be investigated in the context of family communication about genetic risk, as each of these elements likely impact how families communicate about genetic disease.

Implications

Many people are managing genetic conditions such as AATD and are making habitual disclosure decisions about sharing information regarding their condition. Little research, however, has explored disclosure decision-making in regards to one's motivation to disclose. The

goal of this research was to provide a better understanding of the factors influencing young adults' patterns based on feelings of familial obligation and/or a need for social support. The findings from this study have several theoretical implications, implications for the study of communication about genetic disease risk, and practical implications. I will discuss each of these three areas of implications in turn in this section. Implications for disclosure and the DD-MM will be considered first.

Theoretical Implications. Theoretical contributions of this study have implications for theory-building and future research in disclosure and the DD-MM. Self-disclosure is an important element in the development and maintenance of interpersonal relationships (e.g., Altman & Taylor, 1973) and has been consistently associated with positive health (Derlega et al., 1993; Frattaroli, 2006) and psychological outcomes (Kelly, Klusas, von Weiss, & Kenny, 2001; Pennebaker, 1984). Results of the present study contribute to the vast body of disclosure literature by examining the role of disclosure between genetically-related people who are managing a genetic mutation for AATD.

More specifically, the current study utilized the DD-MM (Greene, 2009) as a framework for exploring patterns of disclosure for people considering disclosure of genetic test results. Findings provide support for the key components identified in the DD-MM (Greene, 2009) for predicting decisions to disclose information. In the present study, assessment of information (i.e., stigma, prognosis, symptoms) and assessment of a receiver (i.e., perceived emotional and instrumental support, anticipated outcomes) were found to predict likelihood of disclosure of genetic health information. As such, disclosure models (or a portion of them) like the DD-MM can be incorporated into broader theories of information management and self-regulation (Checton, 2010). Reasons for sharing health information (e.g. familial obligation and social support) facilitate the management of information and/or the regulation of their health condition

as has been shown with the health benefits associated with disclosure (Frattaroli, 2006). Therefore, people's motivations for disclosure and their disclosure decision-making process should be situated within more global approaches to health issues, which warrants continued research.

The model also indicates potential patterns for disclosure and the managing of personal health information and helps to explain how communicative environments are fostered. Consequently, this study broadens the heuristic and explanatory power of DD-MM (Greene, 2009) by examining the motivations behind disclosure of genetic information rather than relying on the assumptions made for other health contexts. By understanding disclosure more broadly and based on differing health contexts, an understanding of how motivations impact disclosure decisions can be achieved.

The role of ethics and morality in the management of private information is also brought to light in this study, and in the broader body of literature investigating how families communicate about genetic risk. As discussed previously, there is often debate in families at risk for genetic conditions such as AATD and about who has the right to disseminate information from one's genetic test. This debate is not one simply about ownership rights, but about ethical and moral obligations related to revealing or concealing information that could impact the health of other family members. The incorporation of ethics and morality has not yet been considered in relation to the DD-MM and adds an interesting element to how individuals and families manage personal information about health issues (e.g., genetic risk). More research should be conducted to more fully understand the role ethics and morality play in how individuals and families manage genetic information. Future theory extension and development should investigate the intertwining nature of family communication environment, information management, and ethical ownership of information, especially in reference to health information.

Combining these theoretical implications leads to the possibility of the development of a new communication theory to explain how motivations for disclosure and demographic variables influence how an individual makes decisions to reveal or conceal health information, and specifically genetic risk.

Genetics implications. Results of the current study also have several implications for the study of communicating about genetic risk. First, the addition of theory as a framework from which to explore the genetic disclosure decision-making process is novel and largely missing from the body of literature investigating genetic information management. The successful use of theory in this study provides further incentive for research to move forward with the theoretical application to examine the communicative process people and families go through in making disclosures about genetic risk to those both considered inside and outside the family unit. A potential outlet for future research might be studying the narrative approach to communicating personal and familial genetic risk to understand how narratives change based on motivation, privacy management, relational closeness, or familial communication patterns. Researchers could also further explore the notion on illness uncertainty to grasp how uncertainty related to genetics is communicated or managed. As genetic testing services for late onset disorders become more ubiquitous, so does the body of research examining genetic testing from the viewpoint of the person being tested (Polzer, Mercer, & Goel, 2012). However, the focus of these studies has been centered on the accuracy of the person's perceptions of risk compared with those of experts rather than on the active construction of meaning given to the person's genetic test results and genetic risk.

Additionally, current research has accounted for the identification of how many and which family members are disclosed to (Hughes et al., 2002) and the uptake of genetic testing by family members in response (Claes et al., 2005; McGivern et al., 2004). Unfortunately, this line

of research falls short in providing a full picture of the decision-making process and actual communication used when disclosing about genetic risk. This study sought to investigate the motivation and key mechanisms leading to disclosure of genetic risk with a specified family member so that future association of disclosure breadth, depth, duration, and frequency can be made. Findings from this study and those examining actual disclosure behaviors can add to what we already know about how families communicate about genetic risk, provide a better understanding of what influences in-depth conversations about genetic risk and the associated outcomes, and guide the construction of health campaigns and materials.

Practical implications. The present study offers several practical implications for health care professionals, campaign designers and marketers, and family members. This study sought to understand several family and individual level factors that may influence the in-depth communication about genetic risk an individual has with others.

Results of this study and others testing the DD-MM (Checton, 2010; Greene et al., 2012; Magsamen-Conrad, 2012) indicate that factors within information assessment influence disclosure likelihood. Understanding how patients assess health information (or genetic information) can help health care providers (e.g., physicians and genetic counselors) plan for more effective information giving, sharing, and catering to patients' information needs. By aiding in patients' understanding of their own risk and genetic condition, health care professionals can help patients give meaning to their risk and help construct patients' narratives and stories for disclosure to family members. Being able to more specifically tailor consultations based on patients' uncertainty and family orientation give physicians and genetic counselors the opportunity to help people communicate their genetic risk in more effective ways.

Furthermore, this study offers insights into one's motivations for disclosure of genetic risk and the communication patterns associated one's informational and relational assessments. If

individuals were to become knowledgeable of the motivations behind disclosure and the specific communication habits (perhaps through genetic counseling appointments), patients may utilize this information to increase communication with family members about genetic risk based on what they want to accomplish with their disclosure. For example, if a person chooses to disclose to their mother or father for social support, the disclosure may become more of an event of storytelling fraught with emotion to gain support or as a means of catharsis. A person may be more inclined to communicate about private topics so that the target recipient's knowledge and reactions (e.g., offering of support) and the disclosure outcomes are possibly shaped by the disclosure.

This, of course, leads to the construction of materials, campaigns, and interventions to facilitate disclosure based on one's motivation. The data from this study can guide marketers and campaign designers on what mechanisms and factors to focus on, especially if tailored for a specific reason for disclosure and/or targeted to a specific demographic. Perhaps motivating families to negotiate rules about revealing and concealing private information (Petronio, 2002) is a practical method for increasing genetic disclosure.

Conclusion

This thesis uses the disclosure decision-making model (DD-MM, Greene, 2009) as the framework to accomplish three goals: a) investigate what factors predict young adults' decision to disclose genetic test results to a family member; b) explore whether familial obligation exists as a motive for disclosure of a test results for a genetic condition; and c) understand how familial-obligation motives, if present, influence the DD-MM and predict disclosure in contrast to those of social support. This study offers theoretical, genetic, and practical implications on the disclosure decision-making process regarding genetic test results and offers the opportunity to inform intervention materials to increase disclosure likelihood.

Specifically, this research indicates that a limited version of the DD-MM fits the genetic context. Both prognosis and anticipated instrumental support are statistically significant factors of disclosure likelihood. However, when the DD-MM is examined more carefully, based on one's motivation for disclosure, there are stark differences in the DD-MM when one is primarily disclosing out of familial obligation versus a want for social support. Those disclosing out of familial obligation assess the health information and their potential information receiver. Those disclosing primarily for social support consider only factors within the receiver assessment. It is important to recognize that, across all models, disclosure efficacy is never a statistically significant predictor of likelihood of disclosure.

The ability to understand what factors and motivations are at play in one's disclosure decision-making process regarding genetic test results is tremendous as we enter into the genomic era. Not only can health information be better communicated to those receiving genetic test results, but these results can foster better family communication practices and inform campaign and decision aid designs.

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Appendix A

Informed Consent Form

IRB Protocol ID:

INFORMED CONSENT FORM FOR SOCIAL SCIENCE RESEARCH
The Pennsylvania State University

Title of Project: The Influence of Obligation and Social Support Motives in Decisions to Disclose Genetic Test Results

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1. Purpose of the Study: The purpose of this project is to study how people share a genetic-based diagnosis with a family member

2. Procedures to be followed: You will be given a scenario describing a genetic condition. After reading it, you will be asked to answer questions about how you might share these genetic test results with a family member. You will, then, be asked questions about the genetic condition itself, about your relationship with the person to whom you would share the information and about yourself (e.g. demographics).

3. Benefits: Your participation will help us understand the decisions people make about sharing genetic information. You may also gain insights about yourself, and find yourself more prepared if you were to get genetic tests in the future.

4. Risks/Discomforts: You will be asked to reflect on a genetic-based condition and your decisions to share, or not share, the information with others. Thinking about disclosing the test results to a family member, depending on your relationship with him/her, may make you feel temporarily uncomfortable.

5. Duration: It will take less than 60 minutes to complete the study.

6. Statement of Confidentiality: All data obtained from participants will be kept confidential. Only researcher and her advisor will have access to the questionnaires. If this research is published, no information that would identify you will be written. The data collected will be stored in the HIPAA-compliant, Qualtrics-secure database. No guarantees can be made regarding the interception of data sent via the Internet by any third parties. The Pennsylvania State University's Office for Research Protections, the Institutional Review Board and the Officer for Human Research Protections in the Department of Health and Human Services may review records related to this research study. In the event of a publication or presentation resulting from the research, no personally identifiable information will be shared.

7. Right to Ask Questions: Please contact Marisa Greenberg at (916) 342-2046 or msg225@psu.edu with questions, complaints, or concerns about this research. You can also call this number if you feel this study has harmed you. If you have any questions, concerns, or problems about your rights as a research participant or would like to offer input, please contact The Pennsylvania State University's Office for Research Protections (ORP) at (814) 865-1775. The ORP cannot answer questions about research procedures. All questions about research procedures can only be answered by the research team.

8. Compensation: Participants will earn academic credit. If you do not qualify for the study (under 18 years old), or do not want to participate, you can still earn credit in the class by completing an alternative assignment.

9. Voluntary Participation: Participation in this research study is completely voluntary. You have the right to withdraw at anytime or refuse to participate entirely without jeopardy to your academic status, GPA or standing with the university.

You must be 18 years of age or older to consent to participate in this research study.

If you are 18 years or older and you agree to participate in this research study, please click on "I agree". If you do not want to participate in this study, please click on "I disagree".

Appendix B

Survey: Hypothetical Scenario

SITUATION

You have had asthma for several years. Although you try to keep it in check, you find yourself wheezing often and regularly.

You have tried different medications in order to prevent worsening conditions. Unfortunately, you find it increasingly difficult to breath when you exert yourself (like when you walk up a flight of stairs) and you have developed a serious cough.

When you visit the doctor at the University Health Center with your latest concerns, the doctor decides to run several new tests, including a genetic test. The genetic test reveals that you have a genetic mutation that leads to a condition called alpha-1 antitrypsin deficiency (AATD). Your tests indicate that you have a homozygous version of the mutation (you received an abnormal version of the gene that makes alpha-1 antitrypsin from each parent).

The doctor explains that the symptoms you have been suffering from (such as wheezing, chronic cough, and shortness of breath) are typical of AATD. This is the likely cause of your irreversible asthma and other lung symptoms.

The doctor explains that AATD is a condition that results when you have reduced amounts of AAT protein in the blood. The lack of AAT protein predisposes to serious lung disease, which appears in your 50s or 60s.

The only ‘cure’ for AATD is a liver transplant. There is a treatment available for AATD that involves an expensive, weekly infusion of the alpha-1 antitrypsin protein that has been purified from donor plasma. The doctor hands you a fact sheet on alpha-1 and says you should look it over and meet with a pulmonary specialist for further testing. The doctor says that you should share your results with your blood relatives so they can get tested.

Appendix C

Survey: AATD Fact Sheet

WHAT ARE SOME IMPORTANT FACTS ABOUT ALPHA-1 ANTITRYPSIN DEFICIENCY?

Alpha-1 Antitrypsin Deficiency:

- Is also referred to as Alpha-1 or AATD
- Is a hereditary condition passed on from parents to their children through genes
- Occurs when there is a severe lack of alpha-1 antitrypsin (AAT) protein in the blood
- Is easy to find through a blood test
- Can be treated, but cannot be cured without a liver transplant
- May cause lung disease in adults
- May cause liver damage that can lead to cirrhosis in adults, children and infants
- Often goes undetected for years or misdiagnosed

How Is Alpha-1 Antitrypsin Deficiency Inherited?
Each person has two copies of the gene that causes alpha-1 antitrypsin. We inherit one copy of the gene from each parent. A person that inherits an abnormal AAT gene from both parents has the severe deficiency. If they inherit one abnormal AAT gene and one normal AAT gene, then they would be a carrier. In order for a person to have the severe deficiency, both of their parents must be at least carriers.

Risks Associated With ZZ: This individual has two abnormal alpha-1 genes and could develop Alpha-1-related lung and/or liver disease.

What Are Symptoms of Alpha-1 Antitrypsin Deficiency?

- Lung disorders, including asthma-like symptoms, chronic cough, chronic bronchitis, emphysema, COPD, or lung deterioration
- Liver disorders, jaundice, neonatal hepatitis, elevated liver enzymes, chronic liver disease, cirrhosis, and liver cancer.
- Panniculitis

It is important to know that even you may not have signs or symptoms, even if you have the genetic mutations leading to AATD.

Who Should Consider Being Tested?
If you have a family history of AATD or uncontrolled respiratory symptoms, then you should consider being tested. In addition, those with unexplained liver disease should be tested.

What Should I Do With My Results?

- Talk openly with your physician about treatment options and ask questions
- Stop smoking and avoid irritants such as dust, secondhand smoke and fumes
- Avoid drinking alcohol
- Start or maintain health behaviors (exercise, healthy eating)
- Share your results with blood relatives and encourage them to get tested

Appendix D

Measures of Information Assessment (1 of 5)

Genetic Stigma

Now that you've read the hypothetical situation----the presence of genetic mutations leading to AATD—and you have read the fact sheet about AATD, we have a few questions for you about how you feel about AATD. Imagine you have really received these genetic test results----how would you think about this health condition, such as its prognosis, symptoms, relevance for others, etc.

Other people may react to genetic mutations. How much do you agree or disagree with the following statements about how other people will view the news that you have genetic mutations leading to AATD?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. Most people would feel that AATD is a sign of personal failure.					
2. With AATD, most people would not hire me to take care of their children, even if I have no symptoms.					
3. Most people will think less of me due to AATD.					
4. Most employers would pass over my application in favor of someone else without AATD.					
5. Most people would be reluctant to date me because I have AATD.					
6. Once they know that I have AATD, most people will take my opinions less seriously.					
7. Most people will stigmatize me because I have AATD.					
8. People would view me negatively because I have AATD.					
9. Most people will think AATD is disgraceful.					
10. Most people will think that having AATD is my fault.					
11. Most people would respond very positively to me when they find out that I have AATD.					

Appendix D

Measures of Information Assessment (2 of 5)

Preparation

Now that you've read the hypothetical situation----the presence of genetic mutations leading to AATD—and you have read the fact sheet about AATD, we have a few questions for you about how you feel about AATD. Imagine you have really received these genetic test results----how would you think about this health condition, such as its prognosis, symptoms, relevance for others, etc.

If this really happened, how prepared would you be for these genetic test results?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. The genetic test results would be a total surprise. (R)					
2. I had a sense that I was going to be diagnosed with a genetic reason for the irreversible asthma (described in the story).					
3. I would not be expecting genetic tests for breathing problems. (R)					
4. I would not be prepared for a genetically linked health condition. (R)					

Appendix D

Measures of Information Assessment (3 of 5)

Prognosis

Now that you've read the hypothetical situation----the presence of genetic mutations leading to AATD—and you have read the fact sheet about AATD, we have a few questions for you about how you feel about AATD. Imagine you have really received these genetic test results----how would you think about this health condition, such as its prognosis, symptoms, relevance for others, etc.

How much do you agree with the following statements about your health in the future (prognosis) with AATD?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. My health will deteriorate.					
2. I am concerned about my future with AATD.					
3. I consider AATD to be a chronic condition.					
4. AATD is a serious health condition.					

Appendix D

Measures of Information Assessment (4 of 5)

Symptoms

Now that you've read the hypothetical situation----the presence of genetic mutations leading to AATD—and you have read the fact sheet about AATD, we have a few questions for you about how you feel about AATD. Imagine you have really received these genetic test results----how would you think about this health condition, such as its prognosis, symptoms, relevance for others, etc.

How do you feel about AATD symptoms?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. It would be easy for others to notice my chronic cough.					
2. I understand what breathing-related symptoms to expect with AATD.					
3. My shortness of breath will interrupt the natural flow of my day.					
4. My lungs and/or liver will get worse over time.					

Appendix D

Measures of Information Assessment (5 of 5)

Relevance

Now that you've read the hypothetical situation----the presence of genetic mutations leading to AATD—and you have read the fact sheet about AATD, we have a few questions for you about how you feel about AATD. Imagine you have really received these genetic test results----how would you think about this health condition, such as its prognosis, symptoms, relevance for others, etc.

How much do you agree or disagree with the following statements about how relevant Alpha-1 is to others?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. My AATD diagnosis has no real implications for others. (R)					
2. AATD is only significant for me. (R)					
3. My genetic mutations have no real implications for anyone else. (R)					
4. Others may be interested in my AATD diagnosis.					
5. Others are indirectly affected by my AATD diagnosis					

Appendix E

Measures of Receiver Assessment (1 of 8)

Overall Relational Quality

For the remainder of the survey, think only about this family member when responding to our questions.

First, please tell us more about your overall relationship with this family member.	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. I enjoy spending time with this family member.					
2. I am not close to this family member.					
3. This family member's opinions are important to me.					
4. My relationship with this family member is satisfying.					
5. I talk about personal things with this family member.					

Appendix E

Measures of Receiver Assessment (2 of 6)

Overall Response: Emotional

These next items ask more specifically about what you think would initially happen if you decided to share your genetic test results and the AATD diagnosis with this family member.

How do you think your family member would respond if you were to tell him/her your diagnosis?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. This family member would immediately offer emotional support.					
2. My family member would immediately judge me. (R)					
3. First, my family member would emphasize s/he still cares for me.					
4. Initially, my friend would show more concern for him/herself than for me. (R)					
5. My family member would immediately withdraw emotional support. (R)					

Appendix E

Measures of Receiver Assessment (3 of 8)

Overall Response: Informational Support

These next items ask more specifically about what you think would initially happen if you decided to share your genetic test results and the AATD diagnosis with this family member.

How do you think your family member would respond if you were to tell him/her your diagnosis?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. Initially, my family member would help me look for information on AATD.					
2. At first, my family member would offer informational support.					
3. My family member would initially hesitate to help me look for information about AATD. (R)					
4. My family member would immediately help me to search out information on AATD.					

Appendix E

Measures of Receiver Assessment (4 of 8)

Overall Response: Instrumental Support

These next items ask more specifically about what you think would initially happen if you decided to share your genetic test results and the AATD diagnosis with this family member.

How do you think your family member would respond if you were to tell him/her your diagnosis?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. My family member would initially offer instrumental support (accompany to doctor, loan money for treatments).					
2. My family member would soon withdraw instrumental support ("cut me off"). (R)					
3. Initially, my family member would NOT do anything to help me. (R)					
4. The first thing my family member would do is offer to help me.					

Appendix E

Measures of Receiver Assessment (5 of 8)

Overall Response: Anticipated Self- and Other-Oriented Outcomes

The next set of items ask about the more long term outcomes of what you think would happen if you decided to share your AATD diagnosis with your family member.

What do you think would happen down the road (in a few months or so) if you were to tell him/her your genetic test results?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. In the end, revealing my AATD diagnosis would really harm my family member's perception of the person I truly am.					
2. Down the road, sharing my AATD diagnosis would negatively affect how my family member would feel about me.					
3. Revealing the Alpha--1 diagnosis would ultimately harm the way my family member sees me.					
4. It would ultimately hurt my family member's feelings if s/he knew the information.					
5. Ultimately, I think my family member would worry about me if I told him/her.					

Appendix E

Measures of Receiver Assessment (6 of 8)

Overall Response: Anticipated Relationship-Oriented Outcomes

The next set of items ask about the more long term outcomes of what you think would happen if you decided to share your AATD diagnosis with your family member.

What do you think would happen down the road (in a few months or so) if you were to tell him/her your genetic test results?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. Telling him/her about the genetic test results would ultimately hurt my relationship with him/her.					
2. If I told my family member, in a few months we would be even closer than we are now. (R)					
3. I think telling my family member would eventually end or severely alter our relationship.					
4. Ultimately, my family would no longer like me if s/he knew the information.					

Appendix E

Measures of Receiver Assessment (7 of 8)

Overall Response: Anticipated Other Relationship-Oriented Outcomes

The next set of items ask about the more long term outcomes of what you think would happen if you decided to share your AATD diagnosis with your family member.

What do you think would happen down the road (in a few months or so) if you were to tell him/her your genetic test results?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. If I told this family member, I would ultimately lose a bond that I have with other members of the family.					
2. Other members of the family would never trust me in the future if I told this family member my diagnosis.					
3. Telling my diagnosis to this family member would ultimately hurt my relationship with other members of the family.					
4. Revealing my diagnosis (to my family member) would eventually create stress for other members of the family who are important to me.					
5. If I told my family member my diagnosis it would ultimately improve my relationship(s) with other people. (R)					

Appendix E

Measures of Receiver Assessment (8 of 8)

Confidence in Response

You've made many predictions of how you expect this family member to respond to your news. How confident are you in your predictions? Put differently, are you uncertain about how s/he might respond?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. I am confident in my predictions of how my family member will react to my news.					
2. I am uncertain about how my family member will respond to me when I tell him/her about my AATD diagnosis. (R)					
3. I am sure about my expectations of how s/he will respond to me.					
4. I can accurately predict how this family member will react.					
5. I am confident that I know how this person would respond.					

Appendix F

Measure of Disclosure Efficacy (1 of 1)

Disclosure Efficacy

IF you decided to share the AATD diagnosis with this family member, do you think it would be easy or hard for you to tell him/her about it?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. I would have trouble finding the right words if I tried to share my diagnosis with my family member. (R)					
2. I would get tongue--tied if I tried to share my diagnosis with my family member. (R)					
3. I do not know what to say when I try to share information about my diagnosis. (R)					

Appendix G

Measure of Disclosure Likelihood (1 of 1)

Likelihood of Disclosure

Now that you've considered the situation and some possible outcomes of telling this family member about your genetic test results and the AATD diagnosis, how likely you are to tell your family member your diagnosis?	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. I'm likely to tell him/her about my diagnosis in 24 hours after learning about my test results.					
2. I will probably tell him/her about my AATD diagnosis in a week or so after I've gotten them.					
3. I doubt that I will share my diagnosis with my family member in the near future. (R)					
4. I'm pretty sure that I'll tell my family member my diagnosis eventually					

Appendix H

Measure of Motivation to Disclose (1 of 1)

Reason for Disclosure

The next questions focus on how much various reasons may influence your desire to disclose or not disclose your AATD diagnosis to your family member. Whether you decided, ultimately, to tell your family member about your genetic test results or not, how much did the following reasons motivate your decision?

You may have more than one motivation. Different motivations may not affect your decision to disclose at all or were definitely behind your reason to disclose.	Strongly agree	Agree	Neutral	Disagree	Strongly Disagree
1. I "owe it" to my family member to tell them. (FO)					
2. I feel a sense of duty to tell my family member. (FO)					
3. I feel obligated to tell my family member. (FO)					
4. I don't want my diagnosis to come as a surprise to my family member. (FO)					
5. I am concerned about my family member's health and my own. (FO)					
6. My family member would give me the comfort I need. (SS)					
7. This family member would not make me feel alone. (SS)					
8. My family member could provide valuable advice to me. (SS)					
9. My family member would provide me with assistance. (SS)					
10. My family member would be able to provide support. (SS)					

Note. FO = Familial obligation SS = Social support.