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Illness Representations, Self-Regulation, and Genetic Counseling: A Theoretical Review

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In this article the Self Regulatory Theory (Leventhal, 1970; Leventhan H., Benyamini, Brownlee, Diefenbach, Leventhal E.A., Patrick-Miller, & Robitaille, 1997) is presented as a framework for conceptualizing genetic counseling. Findings published in the genetic counseling literature are re-interpreted from the perspective of the theory. Clients are seen as active information processors rather than passive receivers of information. Research related to clients' representations of genetic causality of disease and specific genetic conditions is reviewed. Associations of genetic illness representations to self representations, coping and health behaviors are examined, and suggestions are made for applications of the theory in research and clinical practice.

KEY WORDS: genetic counseling; illness representation; self regulatory theory.

INTRODUCTION

According to the Self Regulatory Theory, illness representations, defined as peoples' perceptions of and beliefs about an illness, are important mediating links between health threats and reactions to them (Leventhal, 1970; Leventhal *et al.*, 1997; Leventhal *et al.*, 2003; Leventhal *et al.*, 1998). Those representations are dynamic and changing, and may develop from a variety of sources including direct experience of illness and medical care; experience of illness through family, friends and the media; and ideas embedded in cultural beliefs and language. The theory delineates five core dimensions of illness representations: identity of the threat (its symptoms and label); cause (e.g., infection, genetic, stress); time line (duration and development); consequences (includ-

ing somatic and psychosocial); and controllability in terms of prevention and cure. Variations in these representations will evoke different patient responses to the same illness or condition. The strong connections between illness beliefs and physiological/functional outcome in specific illnesses make consideration of the patient perception framework so important to health-care professionals in terms of prevention and cure (Cooper, 1998). In the context of genetic counseling, variations in these representations may lead to differences in the perceived risks/costs and benefits of taking one action or another when faced with a medical decision.

The aims of the present review article are to delineate the Self Regulatory Theory within a genetic counseling context, re-interpreting published findings in the genetic counseling literature from the theory's perspective, and to suggest further applications of the theory in research and in the clinical genetics context. Guiding the following discussion is a self-regulation model of genetic counseling, depicted in Fig. 1. According to this model, genetic counseling (in the center) is affected by and affects illness cognitions (top), risk perceptions, negative

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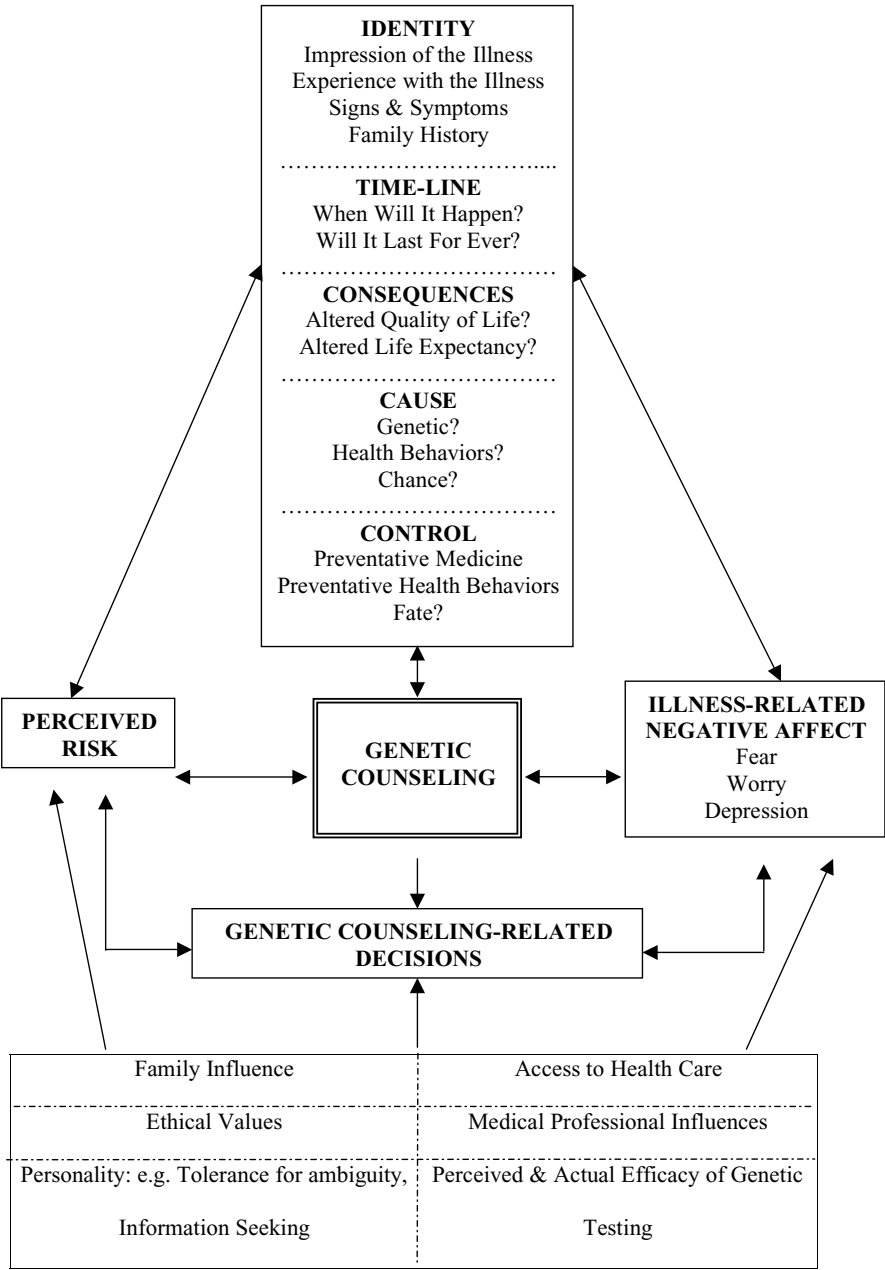


Fig. 1. Self-regulation model of genetic counseling.

affects, and counseling-related decisions, that are also interconnected and affected by background factors (bottom). The relevance of the Self Regulatory Theory to genetic counselors has already been noted by several authors, predicting that the new discoveries about genetic predispositions resulting from the Human Genome Project would impact on people’s illness representations in general (Marteau and Senior, 1997), and cancer risks in particular (De-

cruyenaere *et al.*, 2000; Diefenbach and Hamrick, 2003; Kelly *et al.*, 2005; Rees *et al.*, 2001).

WHERE DO REPRESENTATIONS OF GENETIC CONDITIONS COME FROM?

The Self Regulatory Theory maintains that illness representations evolve over time, based on both

personal (and vicarious) experiences with diseases and social/cultural information acquired through education, media, social modeling and social comparisons (Leventhal *et al.*, 1997; Leventhal *et al.*, 1998). The genetic counseling literature supports this claim, with evidence that personal experience with a genetic disease is a major determinant of its representation and subsequent decisions. For example, known female carriers of an X-linked condition who had lived with an affected brother were more concrete in their decisions to avoid having an affected child compared to those with less personal experience of the condition (Kay and Kingston, 2002). While this would appear to indicate a more severe representation of the disorder by sisters of an affected individual, personal experience does not necessarily mean that consequences of the disorder are perceived as more severe. In another study, mothers of children affected with fragile-X syndrome (FXS) reported experiencing fewer problems in raising the child than FXS relatives who did not have affected children predicted they would experience (Meryash, 1989).

Thus, personal experience is unique and can lead to different representations among people with seemingly similar experiences. A family history of breast cancer is likely to have a strong impact on women's representations of the disease, particularly beliefs about its consequences, cure and control. But, women with the same objective family history may have had different exposures to the effects of breast cancer and witnessed different consequences of the disease. Experiences may range from exposure to positive role models who survived breast cancer and coped well with the disease, to more negative experiences in which relatives suffered physically and mentally before dying (Rees *et al.*, 2001). Thus, learning about clients' experience with a genetic condition *per se* is not enough to reveal their representations; a more personalized approach to the meaning of the experience is required in order to tell how clients conceptualize the condition.

Cultural beliefs about genetics and genetic conditions are another powerful source of illness representation that has been investigated over the years (Ota Wang, 1998; Weil and Mittman, 1993). For example, Mexican-origin women were found skeptical of genetic testing and reluctant to surrender their own lay theories about the causes of their condition in genetic counseling (Browner *et al.*, 2003). Such cultural differences between professional and lay theories were identified as a major source of miscommunication in genetic coun-

seling. Models of cross-cultural genetic counseling have tried to focus on knowledge of client health beliefs and cultural customs as reasons for failure to accept information provided in counseling. However, cross-cultural genetic counseling has also been criticized for using homogenized group stereotypes without attention to intra-group variation and individual differences, suggesting an evolution from cross-cultural to multicultural genetic counseling that focuses on understanding individuals in groups (Wang, 2001).

Genetic counseling's impact on genetic knowledge has been investigated for years, generally indicating that it increases clients' knowledge about relevant genetic conditions (see reviews by: Braithwaite *et al.*, 2004; Wang *et al.*, 2004; Meiser and Halliday, 2002; Pilnick and Dingwall, 2001). However, while genetic counseling is undoubtedly an important source of disease information, its role may be more modest than we like to think. In a study among relatives of CF⁴ patients, only 9% got their knowledge of the disease from a genetic counselor, the rest coming from family members and family doctors (Denayer *et al.*, 1992). Interviews with clients after genetic counseling showed that they integrated lay and scientific explanations for the occurrence of the condition in the family (Skirton, 2001).

Risk information provided in genetic counseling is interpreted using personal mental models of disease causation and inheritance. Findings demonstrate that following counseling, clients reconstruct their risk perceptions in terms of their beliefs about inheritance, their family experience and their life style (Emery *et al.*, 1998; Peters and Biesecker, 1997). A systematic review of qualitative research about lay understanding of familial risk of common chronic diseases (Walter *et al.*, 2004) found that contributors to one's sense of vulnerability included number of affected relatives, together with other more personal factors like perceived patterns of illness relating to gender and age, and perceived similarities to the affected relative. Quantitative research found that parents' subjective recurrence risk perceptions were partly correlated with the information provided in counseling together with other factors like motivations to have more children and risk expectations before counseling (Shiloh and Saxe,

⁴Cystic fibrosis (CF) is a genetic disease in which a defective gene causes the body to produce abnormally thick, sticky mucus that clogs the lungs and obstruct the pancreas, leading to life threatening lung infections and digestive problems.

1989). Subjective representations are indeed believed to be flexible and not detached from reality (Leventhal *et al.*, 1998). Thus, the “objective” information communicated in genetic counseling is not likely to be ignored, but is modified to fit and integrate into the person’s cognitive representation, producing a new and more complex representation. The impact of genetic counseling on clients’ beliefs is expected to increase in the future with the development of more testing technologies that people perceive as powerful and reliable. An indication of this trend was reported in a study among daughters of Alzheimer’s patients in which risk estimates incorporating APOE⁵ susceptibility testing affected perceptions of disease susceptibility more strongly than identical estimates based on family history alone (LaRusse *et al.*, 2005).

Genetic knowledge is related to representations, but is not identical to them. Representations are subjective and tied to wider belief systems, while knowledge reflects the “objective” information that was learned in counseling, and is usually measured by tests that determine accuracy and precision in recall. It is not unusual to see individuals who respond correctly on a knowledge test, and at the same time express personal beliefs and understandings of the genetic condition that deviates from that knowledge. This was demonstrated in a study with mothers of affected children (Weil, 1991): Besides the specific information provided in genetic counseling, there was a cluster of highly personal causes of the disorder—personal attributes, being selected and blessed, and God’s actions. Medical genetic attributions co-existed with idiosyncratic explanations for the child’s condition, and mothers with the two belief patterns (medical and idiosyncratic) were equally knowledgeable about the genetics of the disorder. The personal belief pattern did not appear to interfere with acceptance or understanding of the information provided in genetic counseling. A mixture of scientific and nonscientific beliefs indicated a sense of personal involvement in the cause of the child’s disorder, which was interpreted as enhancing coping and satisfying the need to find meaning (Weil, 1991). These personal meanings may also serve as a coping strategy, aimed to bolstering a sense of personal control, which will be discussed in more detail later.

THE NATURE OF REPRESENTATIONS OF GENETIC CAUSES

Among the five core illness representation components (identity, time-line, cause, consequences and control/cure), genetic health conditions call attention to the cause of the illness. The literature on causal beliefs about illness in general shows that they frequently encompass beliefs about inheritance (Marteau and Senior, 1997), so genetics is widely acknowledged by people as a potential cause of illness. But, the meanings attached to genetic causes and how they relate to other components of illness representations, including other perceived illness causes, need further examination.

Early studies exploring individual representations of genetic factors as causes of disease resulted in inheritance and genes categorized as “external,” “internal,” “other” and “chance” causes in different studies (e.g. Affleck *et al.*, 1987; Klonoff and Landrine, 1994; Landrine and Klonoff, 1994; Murray and McMillan, 1993; Tennen *et al.*, 1984). Their methodologies were criticized as obscuring how people really conceptualize genetic causes of disease (Marteau and Senior, 1997). A study using direct similarity judgment followed by cluster analysis identified three clusters of illness causes. These were behavioral, environmental, and a third cluster that included three distinct sub-categories—genetic/biologic, mystical and psychosocial illness causes (Shiloh *et al.*, 2002). This last cluster was evaluated by participants as significantly less controllable than the other clusters and was named by the authors “hidden causes,” conveying their shared sense of wonder, uncertainty, and little perceived control. Thus, genetic causes of disease seem to be conceptualized as abstract, uncontrollable and inexplicable. This interpretation concurs with the term “genetic exceptionalism” describing public views of genetic information as more mysterious and powerful than other types of health information (Murray, 1997).

Given the complexity characterizing genetic causality of disease, it is not surprising that many beliefs about genes and heredity are riddled with misconceptions. Many people have a ‘holistic’ belief that genes are inherited in groups, so an individual who shows more resemblance in personality or physical appearance to a relative with the disease is perceived as more likely to develop the disease (Kessler and Bloch, 1989; Richards, 1996). Sometimes the degree of perceived genetic connection is more a function

⁵Apolipoprotein E gene (APOE). The inheritance of the epsilon 4 allele of the APOE gene is a risk factor for Alzheimer’s disease.

of the social closeness with the family member than the genetic closeness (Richards and Ponder, 1996). People believe that ‘female diseases’, such as breast and ovarian cancer, run in the family only through mother-daughter transmission and they fail to recognize the significance of parental family history of the disease (Green *et al.*, 1997; Richards, 1999). The health status of carriers of recessive disease genes is also often misunderstood. Individuals identified as carriers in screening for Tay-Sachs viewed their future health with less optimism than non-carriers, although they did not differ in their present and past estimates of their health (Marteau *et al.*, 1992). In another study, myths like associating resemblance to the affected sib with carrier status were identified as barriers for testing among adult cystic fibrosis sibs (Fanos and Johnson, 1995).

Qualitative analysis of interview data with lay individuals (first year undergraduate volunteers) identified three representational models of inheritance (Henderson and Maguire, 2000): (1) Constitutional—being aware of genetic particles, but having no specific knowledge about their location or function. Belief that a person’s whole constitution (make-up) is inherited and diseases are inherited as opposed to specific genetic material. (2) Mendelian—being aware of the phenotype/genotype distinction, and that genes and genetic predispositions are inherited, not just diseases. Understanding that genetic mutations are the actual cause of disease and having some knowledge of the nature of genes. Understanding dominant, recessive and sex-linked disorders, and awareness that two forms of the same gene have to be present for a recessive gene to influence a person’s characteristics. (3) Molecular—awareness of the specificity of genes, that they are made of DNA, are located in cells, and control the overall functioning of cells and physiological processes in the body. Comprehension of the role of the environment in whether a disease actually manifests in a person. Also, some awareness of developmental issues, such as that genetic disease can develop in later life and is not just present at birth.

The model of heredity held by a client is important to his/her understanding of information provided in genetic counseling. While at least a Mendelian model is required for understanding single gene conditions, multi-factorial diseases cannot be understood unless a more complex molecular level model of inheritance is first acquired. Since the client’s model is motivating action, it is especially important when health behaviors intended to

reduce disease risk are indicated. For example, patients diagnosed with familial hypercholesterolemia (FH) who attributed FH to diet and stress in addition to genes, consequently were more likely to engage in behavioral strategies to reduce risk (Senior *et al.*, 2002).

In addition to general beliefs about genetic contribution to health and illness, people have representations about specific genetic conditions. Illness perceptions are held for all varieties of illness, and a number of illnesses for which this has been empirically verified include Marfan syndrome (Peters *et al.*, 2001), Huntington’s disease (Helder *et al.*, 2002), haemophilia (Llewellyn *et al.*, 2003), cystic fibrosis (Chapman, and Bilton, 2004), hereditary non-polyposis colorectal cancer (Claes *et al.*, 2004), and hereditary breast and ovarian cancer (Claes *et al.*, 2005). These perceptions have been found to differ quite considerably from illness to illness. These specific beliefs—correct or incorrect, adaptive or maladaptive—were related to individuals’ reactions to and coping with the relevant diseases. For example, Huntington’s disease patients’ perceptions of strong illness identity combined with beliefs about a long duration, perceived negative consequences for their daily lives, and little hope for cure or improvement in symptoms were significant predictors (along with using coping strategies) of poorer well-being (Helder *et al.*, 2002).

Educational approaches may not be enough to help clients correct misperceptions. In addition to clarifying genetics and heredity, genetic counselors need to explore how a particular genetic condition is represented in a client’s (patient and non-patient) mind, before attempting to modify interpretations of genetic information. The foundations of clients’ beliefs need to be explored before the counseling process can address misconceptions and fears (Diefenbach and Hamrick, 2003).

BELIEFS IN GENETIC CAUSES AND OTHER ASPECTS OF ILLNESS REPRESENTATIONS

Perceiving a disease as genetic can affect its representation in many ways, including components of the illness representation other than cause. For example, genetic diseases may be regarded as more severe (the “consequences” component) based on the fact that they are rarer. Indeed, perception that a condition is prevalent among others can reduce inferences of its severity (Croyle and Jemmott, 1991).

Self-protective cognitive processes such as minimization (a belief that a self-relevant condition is not severe) can also explain perceptions. For example, once women undergoing genetic screening tests for carrying the fragile-X mutation received test results, non-carriers perceived fragile-X syndrome to be more serious than before testing and than carriers (McConkie-Rosell *et al.*, 2001).

The links between a genetic cause and the “control/cure” component of representation are of special interest. While a disease can be controlled either by prevention or treatment, findings indicate that only the prevention component is related to genetic causes. Among the general population, hereditary breast cancer was perceived as less preventable but as similarly treatable as non-genetic breast cancer (Welkenhuysen *et al.*, 2000). In general, a condition is seen as less preventable when a genetic cause is emphasized (e.g., Senior *et al.*, 2000). Participants in an analogue study who imagined that they were at increased risk for a disease based on genetic testing perceived the condition as less preventable than participants who imagined that they were at increased risk for the same disease based on unspecified testing (Senior *et al.*, 1999). This tendency can result in an inappropriate sense of fatalism when multi-factorial diseases are considered. For example, if parents receiving a positive screening test result that their child was at risk for having familial hypercholesterolemia confused the genetic test as detecting actual raised cholesterol levels, the condition was perceived as familiar, dietary in origin, controllable and less threatening. Yet, when the test was seen as detecting a genetic problem, the condition was perceived as uncontrollable and more threatening (Senior *et al.*, 1999). The authors raised concerns that these perceptions may adversely affect motivation to change behavior and reduce risks.

While genetically caused late onset diseases are generally perceived as unpreventable (even incorrectly in multi-factorial diseases), the birth of a child with a genetic disease is nowadays perceived as more preventable. In a study conducted in three European countries among pregnant women, general samples of men and women, geneticists and obstetricians, genetic screening was the single most important factor influencing attributions of control and blame following the birth of a child with Down's syndrome (Marteau and Drake, 1995). A mother who declined the offer of testing was seen as having control over having a child with Down's syndrome, and was blamed in part for it. This repre-

sents an important shift in views of birth defects and parental responsibilities that may lead to unintended implications. A study among family physicians/obstetricians and a university employee sample indicated that women who chose not to use prenatal diagnosis, or who chose to continue the pregnancy following a diagnosis, were judged more responsible, more to blame, and less deserving of both sympathy and social aid subsequent to giving birth to a disabled child than were women to whom testing was not made available (Lawson, 2003). These developments of blaming mothers were foreseen and criticized by Lippman (1993), who questioned the use of prenatal genetic testing that both reflects and generates the process of ‘geneticization’ (the belief that most biology and behavior are located “in the genes”) that increasingly orients western world views of health and disease. The clear benefits of prenatal screening and diagnosis to families versus the psycho-social costs attached to it will most likely continue to stir controversies and public discussions in the future.

BELIEFS IN GENETIC CAUSES AND SELF REPRESENTATIONS

The interplay between disease representations and self representations is another component of the Self Regulation Theory (Leventhal *et al.*, 1999; Leventhal *et al.*, 1998). Some representations of disease overlap with representations of the self (“it's like me”), and others are external to the self and perceived as personally irrelevant. Therefore, the way people perceive themselves can either enhance or diminish acceptance of genetic information, and genetic information, if accepted, can potentially influence on self representations.

Data on the effects of self representations on representation of genetic diseases and genetic risks are sparse. Support for such effects can be seen in findings among siblings of CF patients, where perception of carrier status was strongly related to higher sibling resentment and guilt (Fanos and Johnson, 1995). This can be interpreted as indicating that individuals who perceived themselves as guilty for resenting their sick sibling were more ready to accept carrier status. But, it is also possible that the healthy sibling resented the sick sibling because he/she perceived him/herself as having a higher risk of CF due to their genetic ties. Among women tested for BRCA1/2 mutations, other components of self-system factors, like being optimistic, influenced

perceived risk for breast cancer. Higher levels of self-reported optimism were associated with lower perceived risk (Kelly *et al.*, 2005). Perceiving oneself as maintaining a healthy life style has also been suggested to affect genetic risk perceptions following genetic counseling (Emery *et al.*, 1998). These sporadic findings should encourage further research on how self representations (e.g., self-concept, self-esteem) moderate and influence representations of genetic diseases and acceptance of information received in genetic counseling.

The effects of genetic information on self representations have been more directly investigated in several studies. When a randomly selected group of high school students received sufficient basic information about the nature and the mode of inheritance of CF, 10% of them reported concern about negative impacts of the carrier status on self-image, expressing worries such as: "I will feel abnormal, sick, very depressed . . ." (Welkenhuysen *et al.*, 1996). In another study, carriers of recessive disease genes were more concerned about their future health (Marteau *et al.*, 1992). And, there is also evidence that carriers may be stigmatized by others (Evers-Kiebooms *et al.*, 1994). Women that were found non-carriers for inheriting the fragile X mutation reported in follow-up improvement in feelings about self, with no change in feelings about self found in the carriers (McConkie-Rosell *et al.*, 2000). Responses from structured interviews indicated that the feelings regarding self in the context of genetic testing are not related to global self-concept, but result from concerns regarding the implications of a positive carrier test for themselves and their families. However, in one study the more the condition was seen as inherited compared to other diseases, the more willing participants were to interact with people with the condition, an effect mediated by the lower perceived contagiousness for such conditions (Bishop, 1991). Thus, there might be both positive and negative impacts on the self of having a genetic condition, which are assumed to relate to specific representations of the condition.

Concerns about self-concept have influenced the development of policies on conducting carrier testing on minors and on the informed-consent process (American Society of Human Genetics and American College of Medical Genetics, 1995; American College of Medical Genetics, 1997). Four mechanisms have been identified through which self-concept can be affected by genetic information: altered perception of genetic identity (the individual's perception of defects in his/her inherited

endowment), diminished social identity (the perception of self as less adequate and worthy in the interaction with others), altered perception of health (self perceptions as defective through the connections between the self, health and illness) and threat to the parental role (perceived barriers to attain the valued normative role of parenthood) (McConkie-Rosell and Devellis, 2000). These findings led the authors to recommend that genetic counselors clarify in counseling the role of self-concept, the threat to self-concept related to genetic knowledge, and coping behaviors activated in response to this threat. Adaptation, development and implementation of such coping efforts were called to be taken into account in developing genetic counseling interventions (McConkie-Rosell and Devellis, 2000).

THE INFLUENCE OF ILLNESS REPRESENTATIONS ON COPING

Central to the Self Regulatory Theory are the following: illness representations guide the selection of procedures to cope with and to eliminate illness threats (Leventhal *et al.*, 1997); coping procedures are embedded in "if-then" rules that determine which coping procedures follow from specific illness representations; and coping procedures have a "natural" perceived relevance to perceived cause of disease, as people tend to fit therapy to the source of the threat (Leventhal *et al.*, 1998). Since lack of control is an important attribute of genetic causes of disease (Shiloh *et al.*, 2002), it is not surprising that taking active means to cope with genetic diseases and risks have been found to be associated with perception of regaining control over them. Genetic counselees with higher perceived control used fewer emotion-focused coping strategies, possibly because their increased perceived control reduced their need to use the emotion-focused coping that is activated when people are unable to exert control over their outcomes (Shiloh *et al.*, 1997).

While regaining control is an important goal of coping, control is also associated with undesirable feelings of responsibility and self blame. Guilt and responsibility have been commonly expressed by carriers of genetic mutations about issues surrounding pregnancy (Kay and Kingston, 2002). Since emphasizing a genetic cause may imply lack of control simultaneously with lack of blame, it can either be accepted or denied by individuals, depending on their specific situational needs and coping styles. Findings show that genetic counselees try to

balance between these two conflicting desires (to regain control and to avoid blame) and to cope with the threat by using cognitive coping mechanisms through which they alter their illness representations. For example, genetic clients considered inheriting a gene as not sufficient for occurrence of disease (familial adenomatous polyposis, a Mendelian disease with 100% penetrance), and regarded life-style as also important (Michie *et al.*, 1996). This can be interpreted as a defensive attempt to perceive the cause of the disease as less deterministic and regain a sense of control (Emery *et al.*, 1998). It can also indicate an attempt for reassurance, because people are more likely to perceive genetic factors as increasing disease risk, and behavioral factors as decreasing their risk (Ponder *et al.*, 1996). In another study, interviews with patients post myocardial infarctions (MI) revealed that many of them had complex ideas about what caused their MI and could name several causes (including heredity). A key process in attributing cause appeared to be an attempt to avoid blaming themselves or others for their MI, while simultaneously seeking to assert control over future recurrence. Patients' attempts to avoid blaming and assert control were interpreted by the authors as behaviorally adaptive for their emotional adjustment, even if not necessarily precise (French *et al.*, 2005).

Subjective representations are assumed to be more closely related to reactions than the objective information (Leventhal *et al.*, 1997; Leventhal *et al.*, 1998). This claim of the Self Regulatory Model has been supported in the genetic counseling arena. In a study of women at high risk for delivering a child affected with fragile-X syndrome, the perceived burden associated with having an affected child varied according to personal experience. Further, those who perceived Fragile-X syndrome as more serious were more likely to opt for selective abortion (Meryash, 1989). In another study, couples' reproductive plans were found correlated with subjective perceptions of recurrence risks (for a variety of genetic conditions), but not with the objective risk information received in counseling, even when this information was correctly recalled (Shiloh and Saxe, 1989).

Another concept within the Self Regulation Theory is that reality continues to affect people's representations as outcomes of coping behaviors unfold. If coping behaviors based on specific representations don't work, the representations are invalidated and expected to be reassessed and changed. An outcome consistent with genetic information (like detecting

an affected fetus early in pregnancy by genetic diagnosis) should therefore strengthen the weight of this information in clients' representations, while an outcome that is incompatible with the information (like a non-carrier in a family developing the disease) may challenge the validity of genetic information in forming illness representations. These theoretical predictions demand further empirical investigation in genetics.

In summary, illness representations have an important role in clients' coping efforts. Cognitive coping strategies are often used by genetic counselees, especially if behavioral acts to control the condition are not available. Therefore, holding on to misconceptions in the face of contradictory information provided by counselors may result from defensive coping functions that these beliefs serve (Richards, 1996). According to the theory of engagement (McAllister, 2003)⁶, 'personal theories of inheritance' (explanatory systems specific to individuals that draw upon lay models of inheritance and information specific to their own family), form part of a coping process and coming to terms with risk. The approach of genetic counselors to clients' misconceptions should, therefore, be practical. The self-protective role of misconceptions should be weighted against possible interference with appropriate health protective behaviors and informed choices that need to be made. Counselors should weigh the costs and benefits of changing misconceptions against the costs and benefits of keeping them for clients' health and well-being, before undertaking to change them. And, when they undertake to change them, they should appreciate how difficult it may be to disabuse someone of an attribution that may serve as a defense mechanism, and try to facilitate adaptation of substitute coping mechanisms.

GENETIC REPRESENTATIONS AND HEALTH BEHAVIORS

Another important issue concerns the potential effects of genetic illness representations on health behaviors. The findings in this area are inconsistent and often contradict prior hypotheses. For example, beliefs about the severity of a genetic condition were investigated in several studies among samples of genetic clients for conditions like Tay-Sacks

⁶Engagement is defined as the degree of cognitive and emotional involvement with one's increased risk of developing cancer as a result of one's family history (McAllister, 2003).

(Rosenstock, 1979), cleft lip/palate (Sagi *et al.*, 1992), and sickle cell and beta-thalassemia (Rowley *et al.*, 1991). A few (but not all) intentions and actual behaviors related in predicted ways to disease severity perceptions. For example, among carriers of sickle cell and beta-thalassemia counselees, a greater perceived seriousness of the disease predicted that the partner will be tested, but did not predict utilization of prenatal diagnosis (Rowley *et al.*, 1991). In another study, CF carrier testing was offered to a reproductive-age, non-pregnant population. Among respondents planning to have children, testing decisions were not associated with the perceived burden of a child with CF or with the likelihood of aborting for it, but had more to do with implications of being a carrier *per se* (Tambor *et al.*, 1994).

Interest in genetic testing for increased susceptibility for late onset diseases has attracted attention in recent years, prompting the study of the relationships between various illness representations and interest in testing. The perceived preventability of the disease was found especially important for interest in predictive genetic testing (Shiloh *et al.*, 1999). Interestingly, in another study, first-degree relatives of colorectal cancer patients who believed that less than 50% of colorectal cancers were caused by heredity⁷ were more likely to be interested in genetic testing than were participants who believed that 50% or more of colorectal cancers were caused by heredity (Kinney *et al.*, 2000). This may indicate participants' motivation to be reassured by test results, other beliefs about the utility and implications of screening, or the fact that the more accurate group (<50%) is more likely to seek information to begin with.

Other results that are considered surprising without the use of Self-Regulation Theory, can be easily explained with the incorporation of the theory. For example, studies found that 42% adults from high-risk families who received negative results following predictive genetic testing for familial adenomatous polyposis (FAP) were still interested in attending physical bowel screening (Michie *et al.*, 2002). Those not reassured by negative genetic test results indicated in interviews that preference to continue physical examination was related to several beliefs about the disease and the test: perception of the genetic cause of the disease as transient, so a

present result cannot predict the future; holding a behavioral model of the cause of FAP, or at least that the condition is being caused by other factors in addition to genes; perceiving the test result to be more uncertain, abstract and threatening; and disbelief that a genetic test based on a blood sample can truly detect a disease in the bowel (Michie *et al.*, 1996; Michie *et al.*, 2003; Michie *et al.*, 2002). Such beliefs may result in increasing numbers of "worried-well" (Miller *et al.*, 1988), overuse of medical services, and increased medical risks resulting from unnecessary examinations.

Another concern raised about offering genetic information and testing for late onset diseases was that they would increase fatalistic illness representations, causing a decline in protective health behaviors (e.g., Evers-Kiebooms *et al.*, 2000). But, a survey of the literature turned up no evidence to support these predictions (Lerman *et al.*, 2002). In a study conducted among relatives of individuals affected with alcoholism (Gamm *et al.*, 2004), participants' strong belief in a genetic cause was associated with a significantly increased risk perception, but not with a fatalistic outlook towards developing alcoholism, due to a strong sense of personal control. In another study among people with familial hypercholesterolemia (FH), illness perceptions (risk of raised cholesterol, greater control over FH, and belief that both genes and cholesterol level contribute to risk of a heart attack) were found to be associated with adherence to medication regimen. But, the associations were small, and the authors argued that representations may be a consequence rather than a cause of greater adherence (Senior *et al.*, 2004). They therefore suggested that intervention strategies aimed at helping patients to establish routines for medication taking may be more effective in increasing adherence than interventions designed to alter perceptions related to medications.

Finally, a behavior that has attracted attention in recent years is communicating about genetic information within the family (e.g., Tercyak *et al.*, 2001; Wilson *et al.*, 2004). The notion that illness representations may affect this behavior has gained some support. Students were asked to imagine that they had just been diagnosed with a disease whose cause was described either as controllable or uncontrollable. Those imagining a disease with a controllable cause thought they would have more negative feelings and be less likely to disclose information about the disease to their family or an acquaintance compared to those imagining a disease represented as less

⁷Approximately 5% of colorectal cancers can be attributed to hereditary alterations in one of several genes, and an additional 10% to 15% may be due to inherited susceptibility to unidentified hereditary gene alterations.

controllable (Senior *et al.*, 2002). These findings are encouraging with regard to clients' disclosure of information about genetic risks, which are perceived as uncontrollable. This should be further investigated, including possible interactions between control beliefs and other facets of illness representations (identity, cause, consequence, time-line) and their effect on willingness to disclose genetic information in families.

Genetic counseling has been shown to have a direct effect on individual perceptions of the costs and benefits of specific health behaviors, and thus, on their implementation. Comprehensive counseling with families at risk for hereditary non-polyposis colorectal cancer resulted in an increase in personal beliefs of control over cancer risk, the perceived efficacy of early detection, and greater anticipated ability to cope with a positive test result (Keller *et al.*, 2002).

In conclusion, it seems that the associations between illness representations and genetic counselors' behavior are more complex than is reflected in most research to date. Future research should include emotional predictors, and look for cognitive-emotional interactions for predicting and explaining behavior. The Self Regulatory Theory can direct such research efforts, because it specifically posits that, parallel to the cognitive representations and coping attempts that follow from them, health threats generate emotional states of fear and distress and a corresponding need for procedures to manage these emotions (fear control). Action plans to satisfy both motivations result in behaviors that combine the needs to reduce danger and to reduce fear, and sometimes incorporate a compromise between them (Leventhal, 1970; Leventhal *et al.*, 1997; Leventhal *et al.*, 2003; Leventhal *et al.*, 1998). The applicability of this approach has already been noted by some authors. Decruyenaere *et al.* (2000) suggested that the level of emotional distress in high-risk persons interacts with perceived control regarding the uptake of DNA testing. Extreme threat without feeling of control leads to stress-induced delay or withdrawal from DNA testing. An empirical study supported these notions by showing that women's interest in predictive genetic testing for breast cancer was indeed predicted by an interaction between cancer-anxiety and perceived vulnerability. Higher perceived risk for developing breast cancer was positively related to interest in testing only among women who were *not* highly anxious about breast cancer (Shiloh and Ilan, 2005).

CLINICAL IMPLICATIONS

The relevance of the Self Regulatory theoretical approach to genetic counseling practice was noted throughout the article. Recommendations made to genetic counselors included: (1) to examine understanding of genetics and heredity in general before any educational attempt is initiated; (2) to explore specific representations that clients have about particular genetic conditions before predicting or trying to modify their knowledge and reactions; (3) to investigate clients' prior experiences with genetic conditions using a personalized approach that would disclose individual meaning of the experience; (4) to clarify the role of self-representations, types of threat to self-concept and coping behaviors activated in response to these threats and direct counseling to deal with these issues; (5) to evaluate the costs and benefits of clients' misconceptions before trying to change them; and (6) to consider interactions and compromises between cognitive and emotional motivations as predictors of counselees' action plans for coping with genetic conditions and risks.

Similar recommendations were previously published with regard to reproductive decision-making (Welshimer and Earp, 1989) and undergoing genetic testing for cancer susceptibility (Diefenbach and Hamrick, 2003; Miller *et al.*, 1996; Miller and Diefenbach, 1998). These recommendations may be practiced by many genetic counselors in their daily interactions with clients, but process studies documenting it are still missing (Biesecker and Peters, 2001). This article is meant to put such clinical practices into a theoretical framework that makes understandable why specific interventions are desirable, and how to interpret counseling outcomes and research findings. The choice of the Self Regulation theory as a framework for conceptualizing genetic counseling is especially pertinent because it provides not only clinical implications and research hypotheses, but also a wider point of view about clients as active information processors rather than passive receivers of information. Such a viewpoint seems especially compatible with professional values guiding genetic counseling.

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