The utility of Leventhal's model in the analysis of the psycho-behavioral implications of familial cancer – a literature review

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Abstract

Introduction: We aim to highlight the utility of this model in the analysis of the psycho-behavioral implications of family cancer, presenting the scientific literature that used Leventhal's model as the theoretical framework of approach.

Material and methods: A systematic search was performed in six databases (EBSCO, ScienceDirect, PubMed Central, ProQuest, Scopus, and Web of Science) with empirical studies published between 2006 and 2015 in English with regard to the Common Sense Model of Self-Regulation (CSMR) and familial/hereditary cancer. The key words used were: illness representations, common sense model, self regulatory model, familial/hereditary/genetic cancer, genetic cancer counseling. The selection of studies followed the PRISMA-P guidelines (Moher *et al.*, 2009; Shamseer *et al.*, 2015), which suggest a three-stage procedure.

Results: Individuals create their own cognitive and emotional representation of the disease when their health is threatened, being influenced by the presence of a family history of cancer, causing them to adopt or not a salutogenetic behavior. Disease representations, particularly the cognitive ones, can be predictors of responses to health threats that determine different health behaviors. Age, family history of cancer, and worrying about the disease are factors associated with undergoing screening. No consensus has been reached as to which factors act as predictors of compliance with cancer screening programs.

Conclusions: This model can generate interventions that are conceptually clear as well as useful in regulating the individuals' behaviors by reducing the risk of developing the disease and by managing as favorably as possible health and/or disease.

Key words: disease, Leventhal's model, family cancer, cancer screening, illness representation.

Introduction

The Self-Regulatory Model (SRM) [1–3] is a cognitive-affective model that highlights the existence of both emotional components as well as cognitive components; both of these components alter the perception of disease threat and influence each other. It is the emotional element that distinguishes the SRM from other theoretical models which explain the perception of disease and treatment and which only take the cognitive and/or behavioral component into consideration (e.g., Health Belief Model [4–6]; Theory of Reasoned Action [7]; Theory of Planned Behavior [8]). This parallel cognitive and emotional processing of the menace implied by the disease recommends using this model in studying cancer, an emotionally challenging disease for the patient and their entire family [3]. The Common Sense Model of Self-Regulation (CSMR) is a dynamic, complex system that highlights self-regulation of health and disease [9].

The cancer diagnosis is a difficult and worrying experience; it is life-disrupting, requires continuous adjustment and generates high levels of psychosocial distress in more than a third of patients [10, 11]. At the same time, the disease can lead to family crisis, changing family dynamics and roles [12]. The patients and their families must continuously adjust to threats to their own identity: at first, when they receive the diagnosis, and later, to the treatment, to various physical symptoms, and to emotional distress. This adjustment is considered by the CSMR, in which the patient with cancer is considered to be actively seeking and processing information about the disease, building his/her own cognitive and emotional representations with regard to the disease, and finally selecting and applying those coping procedures that will help him/her face the threat of disease [13, 14].

Advances in molecular genetics offer individuals the possibility of being tested for their susceptibility to developing certain types of cancer due to gene mutations. Oncogenetics and genetic testing can contribute to reducing the risk of developing the disease, improving health status, and, implicitly, reducing mortality in individuals with hereditary risk of cancer, as well as educating the population by facilitating understanding regarding the implications of genetic, psycho-social and behavioral factors for health and illness [15]. At the same time, it generates different attitudes towards knowing the risk of developing the disease and adopting specific consequent behavior [16]. These choices involve individual psychological, as well as socio-familial risks; therefore, genetic testing from the perspective of stress and adjustment is not simply a health behavior, but a way to cope with the stress caused by the risk of developing the disease, according to the CSMR [17, 18]. From this perspective, the CSMR provides a comprehensive framework for identifying and analyzing the factors affecting decision making, adjustment to the decision to undergo genetic testing, management of the results of genetic tests, and adopting screening actions [19].

However, in several other studies this model is criticized or considered to be too limited. For example, Marteau and Weinman [17] recommend further developing the model so that it includes the already existing cognitive representations of health threats. At the same time, the behaviors related to salutogenesis and disease control imply different motivational processes. These processes can be generated by cognitions and emotions that are not directly related to illness perception. Therefore, it has been recommended to combine Leventhal's model with a motivational one [20–22]. DiMateo [23] suggests including social support as a global concept in the analysis, with the purpose of accurately measuring the perceptions – including the factors involved in their genesis – of disease and treatment.

The emotional impact of cancer on the patient and his/her family when there is a family history of cancer, the psychosocial and ethical issues raised by genetic testing – these are the factors that motivate us to research the way the CSMR is used in their analyses.

This review aims to summarize the literature that used the CSMR as the theoretical framework of approach in order to highlight the way in which this model, through its specificity (dynamic, individual-centered, self-regulating, oriented towards elaborating personalized therapy plans), can be used in analyzing the psycho-behavioral implications of familial cancer. The review is the first one in the field conforming to Marteau's theoretical analysis regarding the theory of self-regulation. It is proved that the utility of this theory has been ignored when it is about the understanding of choices made by individuals regarding the lack of reaction when they get information about health risks. Further studies, presented in our work, reach the common conclusion that individuals with high genetic risks regarding cancer disease who believe that healthy behavior could reduce the risk of developing cancer when there is a genetic component are less willing to have sanogenetic behavior.

This work shows the impact of CSMR on the construction of psycho-educational programs built to adopt a preventive and sanogenetic lifestyle.

Material and methods

We conducted a systematic search in six databases (EBSCO, ScienceDirect, PubMed Central, ProQuest, Scopus, and Web of Science) with the purpose of identifying any empirical studies pub-

lished between 2006 and 2015 in English with regard to the CSMR and familial/hereditary cancer. The key words used in the search were: illness representations, common sense model, self regulatory model, familial/hereditary/genetic cancer, genetic cancer counseling. The selection of studies followed the PRISMA-P guidelines [24, 25], which suggest a three-stage procedure: in the first stage, studies were selected based on the title, in the second stage, the abstracts of the remaining studies were read and excluded, and, in the final stage, the results section was read (Figure 1). Each member of the research team analyzed all selected titles, abstracts, and then the integral text in order to establish the eligibility of the articles included in the analysis. The disagreements between the reviewers regarding the final selection were resolved by reaching a consensus through discussion.

The studies that we considered relevant were those that used the Common Sense Model (CSM) and/or the Self-Regulatory Model (SRM) as the theoretical background for the analysis of familial cancer (perception/representation of the disease, perceived risk of cancer, worries about cancer, impact of cancer family history, genetic counseling and testing, screening). We included studies focusing on the issue of familial/genetic cancer and those whose results correlate with the CSM and/or SRM theoretical models, as well as studies in which the CSM and SRM theoretical models are used in genetic counseling for familial/genetic cancers.

We excluded articles structured as reviews, debates, case studies, meta-analyses, posters, conference abstracts, PhD theses/dissertations, pure science, exclusively cellular/laboratory tests, studies published in journals other than the journals ranked by Web of Science, studies where the keywords were found only in the bibliography, studies which included the exclusive analysis of cancers with no hereditary/familial risk, of blood (hematologic) cancers, of other chronic diseases with no hereditary risk, and studies conducted exclusively on health care providers.

The goals, the study population, the theoretical model involved and the main results of each study included in the analysis are presented (Table I).

Results

The search yielded a total of 1 904 241 articles. 1 904 173 of these were excluded when the title/abstract was analyzed; the following were also excluded: theses, duplicate abstracts of conferences/congresses, books and articles which could not be accessed in extenso, which led to a remainder of 68 potentially relevant articles. After excluding the reviews, the studies which included several theoretical models without separately analyzing them,

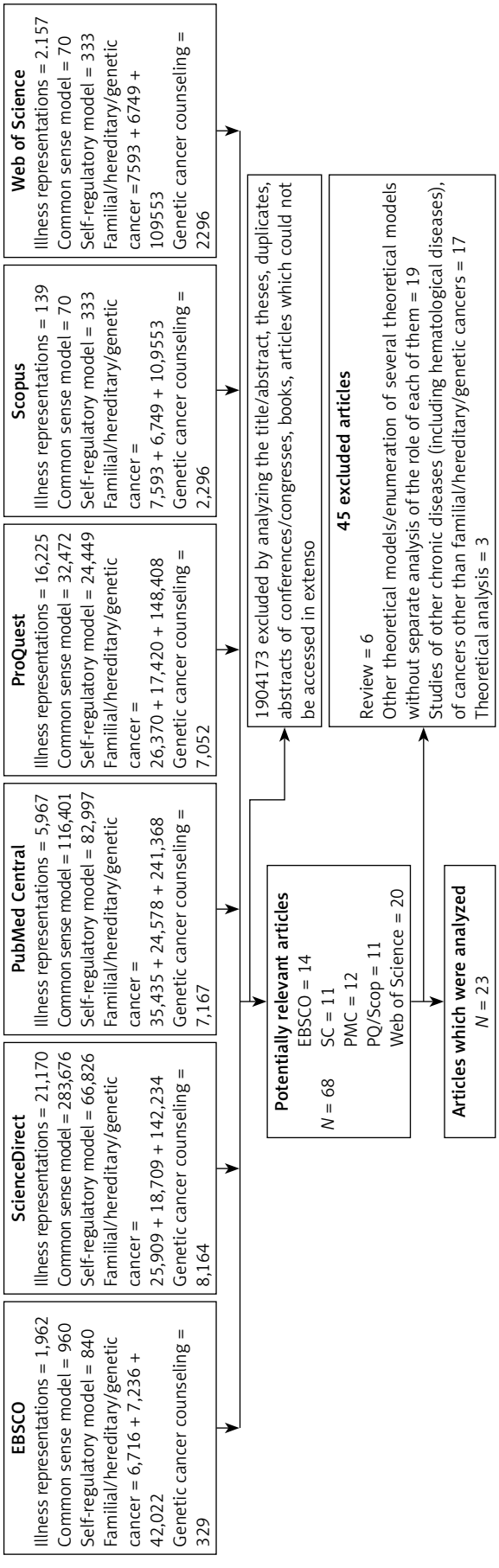

Figure 1. Study selection process

Table I. Overview of CSM/SRM studies

Authors	Study objectives	Design, population	Involvement/theoretical model
Anagnostopoulos *et al.*, 2012	Examination of associations between illness perception, self-efficacy, perceived benefits and barriers related to mammographic screening and its predictive value	Cross-sectional *N* = 408 (female, age group ≥ 40 years, no personal history of cancer)	The Common Sense Model of Self Regulation (CSM + SRM) – theoretical standard for conducting research on health beliefs, disease representations and women's habits to undergo mammograms
Bradbury *et al.*, 2008	Research on the impact of being informed about the history of the parents related to BRCA mutations during adolescence or early adulthood	Retrospective N = 22 adults descendants from parents carrying BRCA1/2 mutations who were informed about mutations in breast cancer before the age of 25	SRM – semi-structured interview guidance (selection of key areas)
Cameron and Reeve, 2006	Assessment of unique associations between risk perceptions and worries and attitudes towards genetic testing for breast cancer detection	Cross-sectional *N* = 303 (62 1st degree relatives with a history of breast cancer in the family, 152 nurses, 89 students with no 1st degree relatives diagnosed with breast cancer)	Common Sense Model of Self Regulation (CSM + SRM)
Cameron *et al.*, 2012	Testing the effectiveness of a communication strategy	Experimental *N* = 749 adults	CSM – used in testing the effectiveness of the strategy
Del Castillo *et al.*, 2011	Comparison of cancer representations among adults who had not suffered from cancer and who had lived/were living or were not living with patients suffering from the disease	Cross-sectional *N* = 130 adults with no personal history of cancer who had lived/were living or not with a relative diagnosed with cancer	SRM – theoretical framework
DiLorenzo *et al.*, 2006	Investigation of a hereditary disease specific model of worry about cancer	Cross-sectional *N* = 628 (> 18 years without diagnosis of breast cancer, colon cancer, prostate cancer, heart disease, diabetes, with family history of chronic disease)	SRM (the findings of the study are consistent with the model)
Hadley *et al.*, 2011	Analysis of the link between colonoscopy following detection of the mutation for Lynch syndrome and depression.	Prospective *N* = 134 mutation carriers (36 index cases, 98 family members) from 47 families	SRM – theoretical framework
Fantini-Hauwel *et al.*, 2011	Assessment of the impact of genetic test results for hereditary cancer from a multifactorial perspective on psychological health	Prospective *N* = 77 adults genetically tested for gene mutations involved in colorectal cancer	SRM – the findings of the study are consistent with the model
Kaphingst, Lachance, Condit, 2009	Analysis of the correlation between beliefs about inheriting cancer and searching for information for own health and protective behaviors	Cross-sectional *N* = 5813 adults	CSM – adapting the model in the analysis

Table I. Cont.

Kelly *et al.*, 2008	Analysis of the subjective change of ovarian cancer risk as a response to genetic counseling and testing	Prospective *N* = 78 women with significant personal or family history of inherited predisposition to breast and/or ovarian cancer	CSM – the basis for the development of new ways of conceptualizing the perceived risk of ovarian cancer
Kowalkowski *et al.*, 2012	Examination of different opinions about cancer in groups of people with different histories of cancer	Cross-sectional *N* = 7172 adults	CSM – in building regression models for evaluating the effect of different stimuli on cancer perception
Lancastle, Brain, Phelps, 2011	Assessment of psychological effects of screening for ovarian familial cancer	Cross-sectional *N* = 1,999 women with high risk of ovarian cancer due to family history of cancer	SRM – the theoretical framework for understanding the emotional responses in ovarian familial cancer screening
Lifford *et al.*, 2012	Withdrawal from screening for ovarian familial cancer in the light of surgical intervention	Cross-sectional *N* = 1,999 women with high risk of ovarian cancer due to family history of cancer	SRM – theoretical model for understanding the range of women's reactions to the risk of familial cancer
Lykins *et al.*, 2008	Analysis of the link between personal and familial history of cancer and beliefs about the causes and prevention of malignant diseases	Cross-sectional *N* = 6,369 adults	CSM – in disease representations
Patrick-Miller *et al.*, 2013	Assessment of the model for communication of the results of BRCA1/2 clinical tests by telephone	Prospective *N* = 167 women undergoing BRCA 1/2 testing	SRM – assessment of responses to the information communicated immediately and remotely in the new models of providing genetic services
van Oostrom *et al.*, 2007a	Study of differences between individuals with a family history of BRCA1/2 mutation or modified genes for HNPCC who opted for having their susceptibility to cancer tested	Prospective *N* = 271 adults with a family history of HNPCC or BRCA genetic mutation	SRM – theoretical framework
van Oostrom *et al.*, 2007b	Examination of predictor variables for distress caused by hereditary cancer	Prospective *N* = 271 adults with a family history of HNPCC or BRCA1/2 genetic mutation	SRM – theoretical framework
van Oostrom *et al.*, 2007c	Exploration of the links between cognitive representations of the disease, adjustment and psychological distress related to genetic testing	Prospective *N* = 235 unaffected individuals undergoing predictive testing for HNPCC or BRCA genetic mutations	CSM + SRM – theoretical framework
Rabin, Pinto, 2006	Understanding the situation in which some cancer survivors and their relatives are motivated by the cancer-related experience to make positive changes in their health behavior	Prospective *N* = 65 breast cancer survivors and 33 1st degree relatives who had never been diagnosed with cancer (sister, mother, daughter)	CSM – theoretical framework
Shiloh *et al.*, 2009	Testing predictions of the Self-Regulatory Model (SRM) whose variations in representation will lead to different responses to the same health threat	Cross-sectional, case-control *N* = 249 adults with risk and with no risk of cancer	SRM – theoretical framework

Table I. Cont.

Shiloh *et al.*, 2013	Examination of the cognitive, emotional and behavioral impact of BRCA1/2 testing among men who were genetically tested	Cross-sectional *N* = 81 men with a family history of cancer (51 were carriers of BRCA1/2 mutations, and 30 men were non-carriers)	SRM – comparing the findings of the study against the model
Shedlosky-Shoemaker *et al.*, 2010	Examination of the factors associated with perceived accuracy of genetic testing	Prospective *N* = 120 adults with high risk of BRCA1/2 mutations	SRM – theoretical basis
Rubinstein *et al.*, 2011	Determination of specific components of family history and personal characteristics related to the perception of the disease in breast, colon and ovary cancer	Cross-sectional *N* = 2,505 women	SRM + CSM – theoretical framework

and the study of other chronic diseases (including hematological diseases) and of cancer types without familial/genetic associations, 23 articles remained, which met all the inclusion and exclusion criteria (Figure 1).

Seven (30.4%) studies included only female subjects, 1 study included only male subjects, while the rest of the studies (69.5%) included both sexes. All the selected studies included adult subjects (> 18 years of age). Regarding the characteristics of the subjects included in the study, most studies (69.5%) included individuals with heightened risk of cancer due to their family history of cancer/cancer genetic mutation, 2 studies included both individuals with this risk of cancer and individuals with no such risk of cancer, and 5 studies were conducted on subjects from the general population.

Eleven (47.8%) studies analyzed breast and ovarian cancer (6 of them involved BRCA1/2 mutations), 2 studies analyzed exclusively colorectal cancer, and 3 studies covered both BRCA1/2 mutations and gene mutations involved in HNPCC (hereditary nonpolyposis colorectal cancer).

Following the analysis of the 23 articles included in the study, several key themes emphasizing the utility of the CSMR in the psycho-behavioral analysis of familial cancer and genetic testing were identified:

1. Identification of the factors and predictors involved in: compliance with the screening and genetic testing program [26–28]; choosing the response to factors that threaten health and in adopting a healthy lifestyle [29, 30]; emotional distress caused by hereditary cancer [31, 32].
2. Assessment of the psycho-emotional impact, of illness perception and of the perceived risk of cancer in case of screening for genetic cancer [33, 34]; genetic testing and counseling [32, 35–39]; family history of cancer, family history of gene mutations for cancer [30, 40–46].
3. Analysis of particular models for: communication of genetic testing results [36, 47]; worrying about genetic diseases [48].

Factors and predictors involved in the compliance with screening and genetic testing

Three studies used the CSMR as a theoretical framework for identifying the variables which lead a patient to follow a cancer screening program. Anagnostopoulos [26] and Lifford [28] concluded that individuals, when their health is threatened, develop a cognitive and emotional representation of the disease that causes them to get involved or not in a health monitoring program. In the case of mammographic screening, young age and a family history of breast cancer

as well as worrying about developing cancer were factors associated with undergoing repeated mammograms throughout life [26]. In the case of screening for ovarian familial cancer, previous experience with cancer, specific distress related to the illness and the belief that aging is a cause of familial cancer were associated with refusal of surgical intervention as a screening procedure for ovarian familial cancer [28].

Worrying about cancer was strongly associated with interest in genetic testing, and with positive beliefs about the benefits of screening, of genetic testing and of prophylactic surgery [27].

Factors that may influence the response to health-threatening factors or the type of adherence to healthy behaviors

Knowing the familial/genetic history of cancer, which is considered to be a factor that could reduce cancer risk, leads the individuals to seek significantly more cancer-related information compared to the average population [29]. In the same study it is shown that heavy smokers who believe that the main cause of lung cancer is genetic are significantly more likely to smoke. In another study conducted on people with heightened risk of cancer, the results showed that smokers were unrealistically optimistic about their health status and cancer-related worries [30].

Predictors of emotional distress caused by familial cancer

Two prospective studies conducted on subjects undergoing genetic testing, using as a theoretical framework of approach the CSMR, identified predictors of emotional distress caused by hereditary cancer: emotional distress before the testing, hopelessness, the number of first degree relatives affected by cancer and powerful emotional representations of the disease [31].

Both studies [31, 32] show that disease coherence and passive adjustment are predictors of emotional distress caused by hereditary cancer, these factors being useful in anticipating the emotional reactions that the individual may manifest in the case of genetic testing.

Assessment of the emotional impact of screening for hereditary cancer

In the case of screening for Lynch syndrome, done on carriers of gene mutations, a monitoring colonoscopy may serve as an adjustment strategy in moderating the emotional distress caused by the identification of the susceptible mutation of cancer [33].

However, in the case of screening for ovarian familial cancer, the results of the study conducted on a sample of women with risk of ovarian cancer due to family history of cancer did not reach a conclusion about the predictive usefulness of the variables related to the illness perception in explaining the emotional distress during screening [34]. Almost a fifth of the women were extremely anxious, and more than a quarter reported high levels of emotional distress specific to the ovarian cancer, although they were taking part in a screening program for ovarian cancer [34].

The impact of genetic testing and counseling at a psycho-emotional level and on the perceived risk of cancer

Fantini-Hauwel *et al.* [35] measured anxiety as a state comparatively in two groups of patients – carriers and non-carriers of the gene mutation involved in colorectal cancer, before and after the genetic testing. The results of the study revealed that, in the case of non-carriers, the anxiety before the test was strongly correlated with the anxiety after the test. In the case of carriers, no association was found between pre-test and post-test anxiety. Moreover, for the carriers, the communication of the results generated a reorganization of psychological functioning, which was independent of their previous emotional state and mood. In the case of the subjects with alexithymia the difficulty of expressing emotions before the test determined a similar difficulty after the test. Alexithymia, according to the study, is the only predictor of post-test emotional distress for both carriers and non-carriers. For mutation carriers who suffered from alexithymia, the capacity to recognize and express emotions was negatively affected after they received the test results [35].

Patrick-Miller *et al.* [36] measured anxiety as a state, general depression and anxiety before the test and after the communication of test results for BRCA1/2 by telephone. Anxiety as a state decreased significantly after the communication of test results, but no significant difference could be found in the general anxiety and in the depression levels, and the general anxiety decreased significantly after clinical monitoring.

Van Oostrom *et al.* [32] evaluated hereditary cancer distress and cancer-related worries in a group of healthy individuals undergoing predictive testing for BRCA1/2 or HNPCC mutations, in a longitudinal study before and after genetic testing. The perception of the disease (IPQ-R [49]) predicted the emotional distress caused by hereditary cancer, as well as cancer-related worries. Emotional distress linked to hereditary cancer increased immediately after they received the results of the testing; yet, 6 months later, they experienced a lower level of worrying than the original one. In terms of cancer-related worries, both carriers and

non-carriers showed lower levels of worrying 6 months after the disclosure of the results, compared to the original level.

However, in an evaluation done after a maximum of 4 years following genetic testing, Shiloh [37] found that male carriers of BRCA1/2 were significantly more stressed after the testing, that they perceived breast cancer as having fewer emotional consequences and effects on the carrier, and that it was easier to treat (Brief IPQ, [50] compared to males who did not carry the BRCA1/2 mutation. After the disclosure of the genetic testing results, 48% of the carriers reported that testing increased the perception of their own risk and 74% of them intensified the screening for cancer.

Nevertheless, Kelly *et al.* [39] found that in the case of women, especially those with a history of breast cancer, there is an improvement in the accuracy of the evaluation of one's risk for ovarian cancer after the genetic counseling and before the genetic testing (before the genetic counseling, the risk was greatly underestimated). The disclosure of the genetic testing results had only a moderate influence on the accuracy of the evaluation of one's risk for ovarian cancer, as the tendency of underestimating the risk was maintained, especially in women carrying the BRCA1/2 mutation. The percentage of estimated risk for ovarian cancer declined over time, regardless of testing results or history of breast cancer [39].

In a group of people with increased risk for BRCA1/2 gene mutations, Shedlosky-Shoemaker *et al.* [38] studied in a longitudinal study the change in the perceived accuracy of the cancer risk depending on the perceived accuracy of the genetic testing, which was influenced by genetic counseling. The perceived risk of developing cancer was lower in subjects who showed increased perceived precision of genetic testing after counseling.

The impact of family history of cancer and of gene mutations on the psycho-emotional state of the individual and on the perceived illness risk

Family history of cancer is associated with different illness perceptions, as revealed by the results of the analyzed studies that used the CSMR as a theoretical framework [41, 42].

In a study conducted on healthy adults living with a relative diagnosed with cancer [41], it was the cancer-related family experience that had the greatest impact both on the emotional status and on the content of cancer-related representations. These results are supported by Kowalkowski *et al.* [42], who, in his survey, found significant correlations between cancer history and perceptions about cancer, and that having a family history of cancer was more likely to lead to worries about developing the disease in the future. Moreover, family history of cancer led individuals to believe that the disease was most often not caused by their behavior or lifestyle. This belief is present in studies conducted by Bradbury *et al.* [40], Kaphingst *et al.* [29] and Shiloh *et al.* [30], where some of the smokers with cancer risk considered that healthy behavior would not reduce cancer risk when there was genetic susceptibility. However, in Lykins's *et al.* randomized study [43], for individuals with a personal history of cancer (survivors), the presence of family history of cancer did not affect their tendency to believe in the role that controllable factors (smoking, unhealthy diet, stress, alcohol, lack of exercise) have in influencing the risk of cancer, contrary to the beliefs of individuals with no personal history of cancer, but with a family history of this disease [43]. Rabin and Pinto [44] found no significant difference between breast cancer survivors and their first-degree relatives in the perception of the role of controllable factors in developing cancer.

Rubinstein *et al.* [45] found that the most significant predictive factor for the perceived risk of breast cancer was having first-degree relatives with breast cancer. The total number of relatives with breast cancer was associated with the perception of a high risk for ovarian cancer and the perception of a reduced capacity of preventing the ovarian cancer. Family history of a particular type of cancer was associated with different perceptions of risk for another type of cancer [40, 42]). Regarding the family history of genetic mutations susceptible to cancer, Bradbury *et al.* [40], in a retrospective qualitative study conducted on adult descendants from parents who carried BRCA1/2 mutations, found that most descendants believed that the disclosure of information related to genetic mutation history had a significant impact on their emotional status, and that for some individuals it could even lead to a change in their health behavior. Most descendants reported that their interest in genetic counseling or testing increased when they discovered cancer genetic mutations in their parents [40]. Associations between the history of genetic mutations of cancer and the perception of the disease were also found by van Oostrom *et al.* [46] in a survey of adults with a family history of BRCA1/2 or HNPCC genetic mutation. Individuals from families with BRCA1/2 mutations perceived hereditary cancer as more serious and tended to perceive less control over the disease; at the same time they more often felt significantly overwhelmed by the genetic risk and unable to cope with this risk compared to individuals from families with mutations involved in HNPCC.

Analysis of models of communication in the context of genetic testing and of models of worry specific to genetic diseases

In an experimental study having the CSMR as a theoretical framework, Cameron *et al.* [47] tested the effectiveness of a strategy for communicating the information referring to genetic testing and its implications in adhering to an adaptive behavior in the case of a hypothetical test for colon cancer. According to the results of the study, disclosure of information referring to risk actions led to improved risk beliefs about coherence regarding health promoting behaviors. The disclosure of information reduced cancer risk projections compared to those who did not receive this information. Explaining through short messages how the action can reduce the genetic risks may foster beliefs that motivate individuals to take protective measures [47].

In the search for effective models for the communication of genetic testing results, Patrick-Miller *et al.* – also having the CSMR as a theoretical framework of approach – studied the effect of communicating the results via telephone. The results of the study show that this method cannot be associated with negative emotional and cognitive responses, which could support the inclusion of telephone communication in providing genetic services [36].

Using the CSMR, DiLorenzo *et al.* [48] investigated a model of worrying specific to colon cancer. According to this model, family history of cancer, the specific risk, and the overall risk of disease influence the level of worry about the disease.

In the systematic analysis of the 23 studies we have highlighted the utility of the CSMR in the analysis of the psycho-behavioral implications of familial cancer for individuals affected by the disease or with high risk for the disease in the screening for familial cancer and in the genetic testing.

Using the CSMR as a theoretical framework of approach, the results of the analyzed studies supported the idea that individuals create their own cognitive and emotional representation of the disease when their health is threatened; this representation is influenced by the presence of a family history of cancer and causes them to adopt or not a salutogenetic behavior [26, 27, 29, 31, 33, 34, 41, 43, 46, 47]. Disease representations, particularly the cognitive ones, can be predictors of responses to health threats that determine different health behaviors [29, 30]. Age, family history of cancer, and worrying about the disease are factors associated with undergoing screening for cancer [26–28]. At the same time, the disease representation can cause, according to Cameron and Reeve [27], irrational behavior, withdrawal from screening for cancer, or the preservation of unhealthy behavior. For the time being, no consensus has been reached as to which factors act as predictors of compliance with cancer screening programs, which makes it necessary to conduct more longitudinal studies. Associating the CSMR with decision-making in the analysis may generate a better understanding of the way in which individuals decide to engage or not in cancer screening or genetic testing programs [20]. Further research is recommended on whether individuals who perceive disease as having a genetic cause expose themselves more to carcinogenic agents, behavior which then affects their health.

The perceived risk and worrying about the disease are analyzed through the two parallel systems of representation of the disease – the cognitive and the emotional one – according to the CSMR [3, 27]. Many of the studies included in this review support the role of disease representations in risk perception, and the connection between risk perception and worries about the disease, in the context of family history of cancer or of genetic testing or counseling [28, 30, 32, 37, 38, 39, 42, 44, 45, 48]. The dynamics of the CSMR highlight, through the conducted longitudinal studies, the importance of knowing the predictors of emotional distress caused by hereditary cancer, as it is necessary to anticipate the emotional and cognitive reactions that a person might have when being tested for genetic susceptibility to cancer [32], in order to identify the maladaptive representations and their cognitive restructuring [31].

In order to improve the understanding of the connection between the risk for a genetic disease and behavior, Cameron *et al.* [47] developed an experimental model for information communication, yet these results cannot be generalized, as further studies are necessary in order to generate interventions that are conceptually clear and that can prove their efficiency in adopting salutogenetic behaviors.

The CSMR can be considered to be a useful integrative theoretical framework in understanding and analyzing familial cancer and genetic testing, by emphasizing the distinction between the interactive influences of cognitive and emotional representations related to the perceived risk of cancer and worrying about the disease when there is a family history of cancer, with a predictive role in the adoption of preventive behaviors [27] and assuring quality of life for these patients [51].

Limitations of this review

Most of the analyzed studies included some limitations, which in some cases led to limitation of accuracy and of generalizability of the results.

The main general limitation is that only one study is randomized, the rest being studies of series of cases. Therefore, to generalize the results, it is necessary to conduct new studies on larger populations, on individuals from other societies and in various cultural contexts. Another limitation present in 60.8% of the reviewed studies was their cross-sectional design. Thus, longitudinal studies are needed to explore how perceptions about the disease change over time in response to new influences (personal and/or familial experiences related to the disease), changes in the emotional consequences of genetic testing and the adjustment following genetic testing. To analyze the risk-behavior causality, it is necessary to assess longitudinally the impact of educational activities based on genetic susceptibility or on information concerning the family history of cancer on the disease-related beliefs and how these beliefs affect behavior. A prospective analysis of the factors that predict the withdrawal from screening at different times is also necessary.

Clinical implications

The findings of this analysis can be used to develop and implement personalized psycho-educational programs aimed at modifying maladaptive representations and negative emotional responses related to familial cancer, reducing the barriers and enhancing the perceived benefits, which would result in increased compliance with treatment and screening programs [52, 53], and increasing the quality of individual, familial and social life.

In conclusion, choosing the CSMR as a useful tool in conceptualizing the analysis of the psycho-behavioral implications of familial cancer is justified, as it undergoes simultaneous cognitive and emotional processing of the threat posed by the disease and, at the same time, it centers on the individual within a specific familial, social and cultural context. Still, further rigorous, longitudinal research is necessary, in order to standardize and validate the explanatory constructs generated by the CSMR in analyzing the psycho-behavioral implications of familial cancer.

Conflict of interest

The authors declare no conflict of interest.

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