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ARTICLE

Predictive testing for hereditary breast and ovarian cancer: a psychological framework for pre-test counselling

Marleen Decruyenaere[1], Gerry Evers-Kiebooms[1], Lieve Denayer[1], Myriam Welkenhuysen[1], Erna Claes[1], Eric Legius[1] and Koen Demyttenaere[2]

[1]*Centre for Human Genetics and* [2]*Department of Psychiatry, University of Leuven, Leuven, Belgium*

Since the identification of two breast–ovarian cancer susceptibility genes (*BRCA1/2*), predictive testing for hereditary breast/ovarian cancer (HBOC) has been available. Given the complexity and uncertainties of HBOC and the potential impact of predictive testing on psychological well-being, we offer the test applicants a combination of information-oriented and psychological counselling. In this paper, we describe the multidisciplinary approach for predictive testing for HBOC as a clinical service in Leuven, hereby focusing on psychological and decision counselling practice. Attention is paid to the theoretical framework used for pre-test psychological counselling in Leuven. We discuss three important interacting dimensions of psychological counselling: individual emotional support, decision counselling and support of the family communication process. Decision counselling consists of an evaluation of the cognitive and the emotional processing of the information given and strategies and resources for coping. This serves as a starting point to facilitate free informed decision making. Scenario development is used as a decision aid. *European Journal of Human Genetics* (2000) **8**, 130–136.

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Introduction

The lifetime risk for women in industrialised countries to contract breast cancer is estimated to be 10–12%.[1,2] Based on the Belgian National Cancer Registry, the cumulative incidence for Belgian women to contract breast cancer is estimated to be 7% before the age of 75. It is estimated that about 5–10% of breast and ovarian cancers might result from a genetic predisposition.[3] A large proportion of the hereditary breast and ovarian cancers can be attributed to a *BRCA1* mutation on chromosome 17[4] or to a *BRCA2* mutation on chromosome 13.[5] However, the penetrance of these *BRCA1* and *BRCA2* mutations is uncertain and variable.[6–9] Women who have inherited a mutant *BRCA1* or *BRCA2* allele have a cumulative risk of 50–80% to develop breast cancer before age 70. The lifetime risk of *BRCA1* mutation carriers developing ovarian cancer is about 20–60% and for *BRCA2* mutation carriers about 10–30%. *BRCA1* mutation carriers have a risk of about 6% of developing colon cancer and of about 8% of developing prostate cancer before the age of 70. Male *BRCA2* mutation carriers have a cumulative risk for breast cancer, estimated to be 6% by age 70. The penetrance estimates must always be used with caution. They are appropriate for counselling in multiple-case families but may not apply to mutation carriers in every family.[9]

Cancer threatens and causes psychological distress. Breast and ovarian cancers are particularly threatening for a woman's self-esteem and body image because they concern parts of the female body connected with fertility, femininity and sexuality.[10,11] Moreover, hereditary breast and ovarian cancer (HBOC) is associated with many threatening uncertainties:[12–14] the risk of being a mutation carrier; the incomplete penetrance among mutation carriers; the influence of other genes and of biological and environmental factors such as hormones and nutrition; the uncertain efficacy of early

Correspondence: Professor Dr G Evers-Kiebooms, Centre for Human Genetics, U.Z. Gasthuisberg, Herestraat 49, B-3000 Leuven, Belgium. Tel: +32 16 34 58 67; Fax: +32 16 34 60 51; E-mail: gerry.kiebooms@med.kuleuven.ac.be
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detection methods, prophylactic surgery and chemo-prevention; the issue of treatment and prognosis of HBOC. For 5 years, predictive testing has increasingly been offered to persons with a family history of breast/ovarian cancer. Given the complexity and uncertainties of HBOC and the potential impact of test results on anxiety, depression levels and self-esteem, it has been recommended[15–20] that predictive testing should be undertaken by a multidisciplinary team. Test participants need fully to understand what the results of the predictive test may mean for them *before* the test is performed. They should receive appropriate information and support before and after predictive testing. Research protocols and follow-up studies have been set up all over the world to evaluate the efficacy and long-term impact of predictive testing for HBOC.

In Leuven, genetic testing for HBOC has been available as a clinical service since the end of 1997. It is the aim of the present paper to describe the multidisciplinary predictive testing service for HBOC in our centre, thereby focusing on the psychological framework at the start of pre-test psychological and decision counselling.

Predictive testing for HBOC in Leuven

In Leuven, the genetic testing team for HBOC consists of a clinical geneticist, a social nurse, a psychologist (staff of the Centre for Human Genetics) and a number of medical specialists at the University Hospital of Leuven (oncologist, gynaecologist, surgeon, psychiatrist, etc.). In this paper, we describe the predictive test service for asymptomatic persons from a family in which a BRCA1/2 mutation has been detected (Table 1). The genetic testing of affected persons is the topic of continuing retrospective study.

Table 1 Outline of the predictive test protocol for HBOC[1]

Session 1 (clinical geneticist + social nurse)
- Information/educational phase
- Emotional support
- \+ Informational text

Clinical examination of breasts and ovaries by oncologist/gynaecologist

Session 2 (psychologist/psychiatrist)
- Emotional support
- Decision counselling
- Guidance of the family communication process

Team meeting

Session 3 (clinical geneticist + social nurse)
- Discussion of the relevant elements in the decision
- Blood sample

Communication of the result (clinical geneticist + social nurse)

Medical follow-up (oncologist/gynaecologist)
For mutations carriers
- Regular screening for early detection *or*
- Preventive surgery (preceded by a consultation with psychologist and psychiatrist)

For non-carriers
- Screening for early detection according to general population level recommendations

Psychological follow-up (social nurse + psychologist)
For mutation carriers and non-carriers:
- After 1 month, 1 year and 5 years

[1]For asymptomatic persons of a family in which the BRCA1/2 mutation has been identified.

First counselling session: medical/educational counselling and emotional support

The individuals to be tested receive accurate and balanced information and education about genetics in general, HBOC and its heritability, cancer risks, the predictive test and its implications, the alternatives to prevention and early detection and the benefits and limitations of these alternatives. This information and education phase is intended to increase the participant's knowledge and understanding of genetic testing for HBOC and its implications. Discussion of the family history of cancer during this session generally triggers negative emotions (grief, anxiety, depression, unresolved loss). Alleviation and management of these emotions are very important. Because of the complexity and the uncertainty associated with HBOC, this session is intensive and time-consuming.

After the first session, the test participants receive an educational leaflet that summarises the information given. An outline of the predictive test programme, including the post-test follow-up, and the address and telephone number of the genetic centre are also provided. This information is not only for personal use but can be distributed to other relatives to inform them about HBOC and the predictive test.

Second counselling session: psychological counselling

Psychological counselling is conducted by a psychologist and perhaps a psychiatrist. The psychiatrist is a member of the Institute for Familial and Sexuological Sciences. The major objectives of the second session are to: provide individual emotional support; facilitate the decision making process; and discuss family communication. The decision counselling includes an evaluation of the test participant's cognitive and emotional processing of the information given and their strategies and resources for coping. A helpful method in decision counselling is the development of 'scenarios'.[21] This means that we ask the test participants to explore the possible outcomes of predictive testing as well as the consequences of declining the test. They are asked to anticipate what could happen in each situation and how they would react in behaviour and emotionally. In this we pay special attention to the expected effect of the test result on specific areas: individual psychological functioning (including body image), the partner relationship (including sexual relationship), family planning, adherence to early detection procedures, timing of preventive measures and

relationships with other family members. Scenario development facilitates the decision making by structuring the problems of making decisions and by stimulating the expression of beliefs, experiences, emotions, motivations and values, taking into account the social context. It enhances feelings of personal control by preparing the test participant to cope behaviourally and emotionally with the test result. At this stage, it is important to identify any lack of social support and inappropriate coping strategies, such as denial or hypervigilance. Reduction of extreme levels of anxiety by cognitive intervention may be appropriate. Other problems, such as a collusive partner relationship or intergenerational conflict,[22] are addressed if needed. In the case of generalised anxiety, major depression or other psychiatric problems, test participants are advised to have additional psychological counselling sessions.

During discussion of the family communication process, the counsellor stimulates and facilitates dissemination of information among the family. Possible communication problems are identified and discussed with the counsellee. What type of information are they going to give their relatives? What kind of reactions can they get from relatives when information is given and why are these individuals reacting in that way? Why do they not want to give information to some relatives? Are family attitudes, myths or conflicts, interfering with information transmission? Special attention is paid to cognitive or emotional processes which hinder the dissemination of information within the family.

The privacy of the counsellee is an important matter during the first and second sessions. Confidentiality and privacy are complex issues in genetic counselling.[23–25] On the one hand, individuals are encouraged to share genetic information with their partner, family members and close friends because it is a crucial factor in building a social support network. On the other hand, the fact that information can be misused by insurance companies or employers, and also by relatives, is discussed.

Third session: decision

After the second session, the predictive testing team discusses the predictive test request to identify possible problems or pitfalls. In the third session, relevant aspects are discussed with the test participant and a blood sample is taken for DNA analysis. Two to six weeks elapse between the final consent and *availability of results*. This allows test applicants to reconsider their decision to proceed. After disclosure of the predictive test result, relevant risk information and guidelines for prevention and screening are reviewed.

Medical follow-up Tested individuals are followed up by a medical specialist of their choice.

Psychological follow-up counselling This is offered 1 month, 1 year and 5 years after the test result. Special attention is paid to emotions and uncertainties of carriers and non carriers, to the impact of the test result on the relationship with the partner, the children and relatives, to the communication process in the family and to preventive or surveillance behaviour. If a woman is considering preventive mastectomy, additional counselling is provided before and after surgery.

The predictive test approach is summarised in Table 1. If necessary additional counselling is offered. The partner of the test participant (or another support person) is invited to attend all sessions. Pre-test sessions of male family members who apply for predictive testing are usually planned on one day, mainly because the medical risks are smaller. Male applicants know that they can reconsider their decision to proceed in the period before communication of results; additional counselling is available if necessary.

Psychological research is embedded in clinical service. During the pre and post-test period, psychometric tests and questionnaires are administered (Table 2). The main aim of the pre-test psychological assessment is to establish a baseline evaluation of each individual and to predict emotional and behavioural adjustment to the test result. The assessment is also important in the context of a longitudinal study to investigate psychological consequences of genetic risk notification. The subjects are free to participate in the study.

Pre-test counselling: a psychological framework

This section is an explanation of the psychological framework which underlies the counselling that is offered in the pre-test period.

Emotional support

The importance of emotional support has been demonstrated by several studies. Lerman *et al*[32] found that 33% of first-degree relatives of breast cancer patients reported impairment in daily functioning due to worry about breast cancer, and 20% reported sleep disturbances. Moreover, those who were most interested in HBOC testing were the most anxious and distressed.[33] The burden and anxiety over cancer are

Table 2 Psychometric tests

General knowledge of HBOC (multiple choice)[a,b]
Perceived severity of HBOC[a]
Risk perception[a]
Cancer-specific anxiety: impact of event scale[a 26–27]
General anxiety: STAI (Spielberger State Trait Anxiety Inventory[a 28]
Social support questionnaire[a]
Symptom check list[29]
Utrecht coping list (adaptation of the Westbrook Scale)[30]
Questionnaire on body image and sexuality (unpublished questionnaire, constructed in the Netherlands by L Lodder and P Frets)
Martial intimacy questionnaire[31]

[a]Completed during the sessions; the other tests are completed at home.
[b]Incorrect answers are detected and explained after completion of the questionnaire.

linked to the age of the individual at the time the mother was diagnosed or died, the number of affected and/or deceased relatives and their age at diagnosis, the perceived severity and course of the cancer in these family members and the recency of newly diagnosed cases or deaths in the family. Especially daughters of breast-cancer patients, who were children or adolescents when their mother was diagnosed, are at risk of adverse emotional reactions.[34] At this stage of ego and sexual development, identification with the mother and the female body is of utmost importance. Breast/ovarian cancer in the mother, combined with her own genetic susceptibility, is a threat to a daughter's body image, emotional growth, self-esteem and identity.

Decision counselling

An individual's behaviour in respect of health in a threatening genetic context depends on interaction between the individual's genetic knowledge and the process of coping with the threatening information.[35,36] Coping is a dynamic process which changes over time, depending on the cognitive and subjective–emotional perception of the threatening situation and the strategies and resources for coping with the threat. An individual's experiences, beliefs, goals, values, personality and social aspects and the broader cultural context influence these perceptions.

Cognitive processing of the information Based on the information given and on personal experiences and beliefs, the participant constructs a cognitive picture of the disease.[35] Considerable differences exist between individuals in the cognitive representation of HBOC. Research has shown that some counsellees fail to acquire, understand or recall the information given during genetic counselling.[37–39] Therefore, it is important to check how the test participants have interpreted the information (perception of risk, perceived severity, advantages and limitations of predictive testing, etc).

One explanation for poor understanding and/or retention of genetic information is that information about HBOC is very complex and difficult to explain and understand. An individual's intellectual ability, prior knowledge and experience play a crucial role in comprehension and recall of information and should be taken into account in the communication process. A leaflet with key information, which can be read at home, significantly improves understanding and recall of information.[39,40]

Moreover, presentation of risk information influences information processing and subsequent decision making.[41,42] Risk can be presented in distinctive ways: as a percentage or a proportion, in a numerical or verbal manner, as a single figure or as a relative risk, stressing the positive or negative consequences. Presenting risk information in more than one way during genetic counselling can reduce the effect of presentation.

There are well known cognitive biases that may also play a part in processing information, for example the easier it is to imagine, recall or conceptualise an event, the more likely it will seem to arise. For example, a woman from a BRCA1/2 family, whose sister has recently been diagnosed with breast cancer, may overestimate her risk of being a gene carrier. Misunderstandings and confusions should be discussed and corrected during the counselling session.

Emotional appraisal of the situation Emotional reactions are in the first place generated by concrete personal experience and perception of the threat, rather than by verbal statements about it.[35] Intense emotions may be significant barriers to information processing, decision making and surveillance behaviour. Lerman *et al*[43] demonstrated that risk counselling did not produce improved comprehension in a group of female first-degree relatives of breast cancer patients if the women had high levels of anxiety about breast cancer. The literature has reported inconsistent findings about the relation between threat and health behaviour (such as breast cancer screening). In some studies, anxiety about cancer was associated with less undertaking of screening,[32,44] whilst other research found that anxiety was unrelated to the use of health screening.[37] Other studies have reported that distress over cancer was a stimulant to involvement in health practices.[45,46] These contrasting figures may reflect differences in the conceptualisation or measurement of anxiety, in the nature of the behavioural outcome examined, in the health care setting and/or in the sample characteristics.

Strategies for coping Individuals react by coping differently, depending on the level of threat perceived.[47] Extremely low and high levels of distress give rise to defective patterns of coping, whilst moderate levels are more likely to produce a more efficient response. This curvilinear relationship between stress level and coping reaction offers a possible explanation for the above inconsistent findings in the literature.

Coping by focusing on a problem (action plans for managing the threat, such as seeking information, visiting a general practitioner, using a screening service) and coping by focusing on an emotion (such as self-favouring evaluations, defensive pessimism, minimisation of the problem, avoidance/denial) can be either mutually destructive or facilitating. Defensive avoidance and denial are especially likely to interfere with behaviour where the focus is on the problem.[48] Denial/avoidance is destructive if it undermines actions such as acquisition of information, discussion of that information with the family or surveillance behaviour. On the other hand, denial/avoidance is constructive if it reduces extreme emotional distress and allows the individual to direct his internal energy to processing information and making decisions. Especially in the early stages of coping, denial or avoidance has merits as a temporary protection against overwhelming feelings, but in the long run it can undermine actions focused on problems. Moreover, when denial is partial or minimal, it does not necessarily undermine simultaneous coping by focusing on problems.

Subjective perception that time is of the essence might lead to hypervigilance: impulsive reactions, reduced memory span and less reflection on the implications of a decision. This may result in extreme surveillance behaviour, such as daily self-examination of the breasts, or a poorly informed decision to perform a bilateral mastectomy.[49]

Resources for coping Evaluation and enhancement of the test participant's coping resources is another essential component of decision counselling. Familial disease and genetic risk can undermine self-esteem and identity. This is potentially more serious in daughters of breast-cancer patients, who were in adolescents or children when their mother's condition was diagnosed.[34] Self-esteem and feelings of personal control prove to be associated with an increase in adaptive coping such as information seeking and problem solving. Lack of faith in one's own ability to cope proves to be associated with negative reactions, such as feeling hopeless or depressed and giving up.[50,51]

Guidance and support of the damily communication process

Another important aim of HBOC psychological counselling is the guidance and support of the family communication process on the subject of hereditary cancer and discussion of the implications of withholding information.[17,18,52] We are of the opinion that it is the moral responsibility of family members to inform other relatives about the genetic risks in the family, and it is the geneticist's task to remind the counsellee of this responsibility and to stimulate and facilitate the dissemination of such information within the family. We agree with the ethical view that medical confidentiality should not be broken by clinicians when family members do not wish their relatives to be informed about the genetic disease in the family.[53,54] Richards and Green[54] have argued that individuals may have good reason not to pass on information to relatives and that to intervene might disrupt family relationships. This contrasts with the view[55] that medical confidentiality might be broken under certain conditions.

Ways of handling emotion may complicate discussion of genetic information in the family. Relatives develop their own ways of dealing with the risk. Some refuse to discuss any aspect of the situation, whilst others share their experience and feelings with relatives. Especially individuals who want to tell the family in order to control the threat and their own grief, or who see themselves as the conveyor of information to the family may be at risk and may need additional support and guidance in such a role.[18] Some families develop family secrets and myths about HBOC and its inheritance. An example of such phenomena is patient pre-selection:[56] a sister or brother is pre-selected by the other siblings to be the potential patient, so that they have the illusion that their own risk has been reduced. Processes of identification are used to support the illusion: behavioural or physical resemblances between the pre-selected person and the affected parent are highlighted, which reinforces the pre-selection. Such pretending can be stronger than factual information. An atmosphere of secrets and taboos may thwart the communication process and the social support sought in the family and may even result in conflicts and family rifts.

Julian-Reynier *et al*[57] reported that about 14% of 161 patients who had been asked to contact their relatives, firmly refused to do so, mainly because of problematic familial relationships. Other significant barriers to informing family members are guilt, shame, anxiety or depression. Green *et al*[52] found that 'communication, both obtaining and giving information, was impeded by adoption, divorce and remarriage, family rifts, and large age gaps between siblings'. Moreover, HBOC is generally perceived as merely a woman's problem.[52,58] This, in combination with the taboo surrounding HBOC, results in neglect of the male members in the family communication process. It also prevents women bringing their partners to the counselling sessions.

The discussion about family communication is complex and delicate because of the interplay of individual and family beliefs, attitudes, imaginings and intense emotions. Moreover, several conflicting values play a part in dissemination of information: the right to know, the right not to know, autonomy, privacy and solidarity.[23–25]

Discussion

In this paper, we have described the multidisciplinary predictive testing service for HBOC in Leuven, with special attention to psychological and decision counselling and the underlying psychological concepts. Notwithstanding essential differences in types of disease, the predictive test approach for Huntington Disease (HD)[59,60] and experience with other neurogenetic diseases have been a valuable foundation and starting point for developing predictive testing for HBOC. Unlike HD, this is potentially preventable and can be treated if detected early, although the efficacy of early detection and prevention is uncertain. This results in a need for more counselling and recommendations for surveillance behaviour. The pre-test sessions for HBOC take longer than for HD, because HBOC is associated with greater complexity and uncertainty. Moreover, initial research suggests that people who are most interested in HBOC testing are the most anxious and distressed.[33] This contrasts with the HD findings that predictive test applicants for HD were a self-selected group with significantly higher mean ego-strength and with significantly better means of coping than the general population.[60] Another major difference from HD is that non-carriers of the BRCA1/2 mutation in families with the mutation remain at the general population level of risk of cancer. Lynch *et al*[61] suggested that some women with a favourable result might continue to worry about developing cancer. Because of the far-reaching medical and psychological consequences of predictive testing for HBOC, we offer the applicants a combination of information-oriented and

psychological counselling. Both are prerequisites for free informed decision making. We fully agree that the decision to undergo predictive testing '… should be approached as a multi-step process, and those considering genetic testing must be given every chance to rethink and confirm their final decision'.[19] It is important that people have sufficient time to reflect between receiving the first information on HBOC and their decision to take the test. Although it is not considered a standard of care,[19,20] we offer the applicants a psychological and decision counselling session with a psychologist, and if necessary a psychiatrist, before their final decision. Some (published) test protocols[16,18] also include an additional session with a psychologist.

A specialist multidisciplinary team is required to provide extensive pre- and post-test counselling for HBOC. In our opinion, this team should at least include a clinical geneticist, a psychologist or genetic nurse and relevant specialist physicians (oncology, surgery, gynaecology, psychiatry). The specific organisation of the genetic testing service in a country depends on several factors, such as its health care system and financial resources. It is of the utmost importance that sufficient financial resources are allocated to genetic cancer services so that predictive testing can be offered in a multidisciplinary context.

At present, many questions about HBOC remain unsolved– the impact of genetic testing and efficacy of education, counselling and preventive measures. Further research is needed to reduce these uncertainties. Genetic protocols have been developed all over the world in the form of research projects to evaluate the efficacy of genetic testing in reducing morbidity and mortality from HBOC. Longitudinal studies to evaluate the immediate and long-term psychological impact of genetic testing are also of the utmost importance. This requires sufficient knowledge of methodology for qualitative and quantitative psychological research. Psychologists working in cancer genetics must combine research and psychological counselling skills, and have sufficient insight into psychological and genetic concepts.

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