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Exploring the course of psychological distress around two successive control visits in women at hereditary risk of breast cancer

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Abstract

In this article we determined the course of psychological distress during a breast cancer surveillance program in women at increased risk of developing hereditary breast cancer (BC). The sample comprised of 357 unaffected women (mean age 40.5 years) adhering to a surveillance programme (MRISC-study). Before and after two successive biannual surveillance appointments, the Impact of Event Scale (BC-specific distress) and the Hospital Anxiety and Depression Scale (general distress) were administered, totalling four measurement moments.

In general, psychological distress remained within normal limits and decreased significantly after a surveillance appointment, except for breast cancer specific distress after the second appointment. Scheduled imaging examinations were not significantly related to distress. The course of BC specific distress differed significantly for risk over-estimators and for young (<40 years) excessive breast self examiners. The course of general distress differed significantly for women closely involved in a sister's BC-process. These more vulnerable subgroups may be in need of extra counselling and care.

Keywords: Hereditary breast cancer; Psychological distress; Surveillance; BRCA1/2; Course; Increased risk

1. Introduction

Women at increased risk of developing breast cancer due to a genetic or familial predisposition may opt for regular surveillance outside of population screening to aim at early detection of an eventual tumour. Regular surveillance programmes for this group of women mostly consist of recommended monthly breast self examination, biannual clinical breast examination (CBE), and yearly mammography [1].

In the literature, evidence can be found indicating increased levels of both general and breast cancer specific distress in high-risk women [2–4], especially on the day of a surveillance appointment [5,6]. Additionally, in earlier studies it has been suggested that there exists a negative linear association between adhering to surveillance and distress, resulting in withdrawal from screening [7,2]. These studies, however, were performed at a time when genetic counselling and testing were not

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yet available and/or were of poor standard, and therefore it is important to obtain more data on this topic from current practice. Further, studies that are more recent provided evidence that greater worry is associated with a higher uptake of health care behaviour [8,9]. So far, few data exist on the course of psychological distress after successive surveillance appointments. In this respect, it has been reported that screening appears to be reassuring for women between the age of 50 and 64 with a family history, as compared to women without a family history [10]. In addition, a study on the psychological impact of population based breast cancer screening in the Netherlands (including women aged 50 years and older) provided no evidence for adverse psychological effects after the surveillance appointment [11]. In a group of 52 younger high-risk women, a reduction in breast cancer specific distress was observed 4 months after the women's initial surveillance appointment [12]. Several limitations about these studies can be raised. First, most of these studies have been published before 1998, while the field of hereditary breast cancer has tremendously evolved over the last years. Second, the numbers of women studied were mostly small, and mainly consisted of women above the age of 50 (except for the study of Gagnon and colleagues [12]), which is not the group of interest in hereditary breast cancer. Thus, data from younger women adhering to breast cancer surveillance are scarce or lacking. Further, the magnetic resonance imaging (MRI)-scan as an additional screening tool may increase the burden of the surveillance process in this group of women. Preliminary findings from a study about health-related quality of life in women at increased risk for breast cancer showed that 37% experienced some degree of anxiety about undergoing the MRI-scan, *versus* 27% of women undergoing mammography and 22% undergoing CBE [13].

Our study aimed to examine psychological distress in a large group of genetically predisposed women adhering to a breast cancer screening programme, and participating in the observational MRISC (Magnetic Resonance Imaging SCreening). The MRISC study started in November 1999, and aimed to evaluate the efficacy of a surveillance programme for women at increased risk of breast cancer due to a genetic or familial predisposition (MRISC – part A). The surveillance schedule in the study consisted of recommended monthly breast self-examination, biannual CBE, and annual imaging by means of both a MRI-scan and mammography [14,15]. A psychological follow-up study being part of this study started in September 2000 (MRISC – part B).

In this article, we describe the course of psychological distress, breast cancer specific and general, around two successive surveillance visits for the total group of women. Furthermore, we report on the course of distress in earlier identified subgroups [16–18] revealing significantly more breast cancer specific distress at the baseline measurement moment. These subgroups are: younger (<40 years), excessive breast self-examiners [16]; breast cancer risk over-estimators [17]; and women closely involved in the breast cancer process of a sister [18]. Additionally, the possible extra burden of scheduled imaging examinations including MRI, was compared to that of physical examination only.

2. Materials and methods

2.1. Participants

In this psychological follow-up study, 357 women at risk of hereditary breast cancer and adhering to regular surveillance were included. At entry, the participants did not have a history of breast cancer, and had a cumulative life time risk (CLTR) of developing breast cancer of at least 15%, based on modified risk tables by Claus [19,14]. Of the 357 women, 328 were included in the MRISC-part A-study. The other 29 women were adhering to regular surveillance as well, but did not have the MRI-scan incorporated in their surveillance programme for different reasons. Thus, screening in this group consisted of recommended monthly BSE, biannual physical examination, and annual mammography, which is in accordance with the national guidelines for this group of women. One hundred and nine women from MRISC-A were not included in the psychological follow-up study.

2.2. Measures

2.2.1. Background variables

Age and the number of years adhering to regular surveillance were measured. Educational level was divided into lower, medium, and higher level. Lower level consists of primary or lower vocational education; medium level included lower or higher general secondary education or intermediate vocational education; higher level included higher vocational education or (pre-) university.

Having a partner and having children were both dichotomised into yes and no.

Women were categorised into three risk categories by the MRISC-study team. Women in risk category 1 were identified as BRCA1 or BRCA2 mutation carriers, with a CLTR of developing breast cancer between 60% and 85%. Women in category 2 had a CLTR between 30% and 50%, and were first degree family members of a proven BRCA1/2 mutation carrier, who did not opt for the test themselves, or first degree relatives from a breast cancer patient from a non BRCA1/2 mutation family or from a family where genetic testing was not performed. Women in category 3 had a CLTR of

15–30%. These women belonged to families with an increased frequency of breast cancer, or were 25% risk carriers in a proven BRCA1/2 mutation family [14,15,20].

2.2.2. Breast cancer specific distress

Intrusion and avoidance, two common responses to stressful situations, were assessed at each measurement moment (see design) using the Impact of Event Scale (IES). This questionnaire developed by Horowitz *et al.* [21] comprises 15 items and can be tailored to a specific event, namely 'breast cancer' in this study. Avoidance is measured in 8 items and intrusion in 7 items, and each item has four answer categories: not at all (score 0), seldom (score 1), sometimes (score 3), and often (score 5). The summation score for the total scale ranges between 0 and 75, with a higher score meaning more breast cancer specific distress. Since the Impact of Event Scale has been used in many settings, study samples and cultural contexts cut-off scores are of limited value. However, as an indication for high distress a total score of 26 or more is given [22]. Reliability analysis on the baseline measurement of the current study revealed Cronbach α's of 0.84 for the subscale avoidance, of 0.86 for the subscale intrusion, and of 0.91 for the entire scale.

2.2.3. General distress

The Hospital Anxiety and Depression Scale (HADS) is a 14-item questionnaire, assessed anxiety (7 items) and depression (7 items) at each measurement moment (see design). Each subscale has a score range between 0 and 21. Cut-off scores per subscale indicative of clinical anxiety and depression are 11 or higher. Obtaining a score of 8–10 on each subscale is representative of doubtful cases of anxiety and depression [23]. By combining the scores of the two subscales, a measure for general distress can be obtained. A cut-off score of 18 (on the total HADS) showed the best sensitivity and specificity in a sample of women with advanced breast cancer [24]. Reliability analysis on the baseline measurement of the current study revealed Cronbach α's of 0.83 for anxiety, of 0.86 for depression, and of 0.90 for the entire scale.

2.2.4. Independent variables

From the baseline measurement, performed two months before a surveillance appointment in the clinic (m0, see design), we identified three variables that were found to be associated with elevated levels of psychological distress [16–18]. The first variable addressed the frequency of breast self-examination and was measured with one question: Do you perform breast self-examination regularly in order to detect possible anomalies? The question had six answer possibilities: no, never; yes, approximately once a year; yes, approximately once every six months; yes, approximately once every three months; yes, approximately once a month; and yes, at least once a week. The answers were recoded into excessive self-examination (at least once a week) *versus* otherwise. This was further combined with the median age resulting in two categories: (1) young (<40 years) excessive breast self examiners and (2) others [16]. This question was asked at the baseline measurement moment (m0).

The second variable concerned risk perception [17], and was measured asking for the women's own risk estimate of developing breast cancer during life in terms of "1 in x" in combination with a percentage. The answer to this question was compared to the objective risk status as assigned by the MRISC study team, and recoded into underestimation, accurate and overestimation. This question was asked twice, namely two months before the first, as well as two months before the second surveillance appointment (m0 and m3, see design).

The third variable addressed the degree of involvement in the breast cancer process of a sister [18]. This item was measured by means of a question having four answer-categories: completely involved; closely involved; involved at some distance; not at all involved. Based on the answer to this question the women were divided into two groups: (1) completely or closely involved and (2) involved at some distance, not at all involved or not having a sister diagnosed with breast cancer.

As a fourth factor that could be associated with psychological distress during the surveillance process we considered the type of examination at the surveillance appointment. This appointment either consisted of solely physical examination or of physical examination in combination with scheduled mammography and MRI-scan. For organisational reasons these latter imaging examinations could take place on a separate day.

2.3. Design

This study is part of a larger observational study [15] consisting of 6 assessments around two consecutive biannual surveillance appointments in the clinic during a breast cancer screening programme. Data were collected between January 2001 and May 2003. The assessments were performed on the following moments: two months prior to a surveillance visit (twice, m0 and m3), the day of the surveillance visit (twice, m1 and m4) and one to four weeks after the surveillance visit (twice, m2 and m5). The assessments m2 and m5 were planned one week after the clinic visit in case of solely physical examination, and four weeks after an appointment consisting of physical examination in combination with imaging examinations (mammography and MRI).

For the analysis in this study four assessments were used, the two assessments performed on the day of the surveillance appointment (m1 and m4), and the two assessments administered after communication of the results of the surveillance moment (m2 and m5).

2.4. Procedure

The enrolment-procedure of the women in the psychological part of the study has been described elsewhere [16–18]. Before each assessment moment, women received their questionnaire at home together with a reply-paid envelope. Women who did not return their m2 or m5 questionnaire within 4 weeks were sent a reminder. Since the questionnaires on m1 and m4 had to be filled in on the day of the surveillance visit at the outpatient clinic, no reminders were sent when women did not return one of these questionnaires.

The Medical Ethical Committee of the Erasmus MC in Rotterdam approved the study.

2.5. Statistical analyses

The characteristics of the study sample were tested for differences between the three risk categories by the method of one-way analysis of variance in case of continuous data and by the method of chi-square in case of ordinal data. These analyses were performed using SPPS 11.0. Missing values in the questionnaires Impact of Event Scale and Hospital Anxiety and Depression Scale were handled as follows: for women who filled in more than 75% of the questions per subscale a total score was computed, corrected for the total number of questions of the subscale. For women who filled in less than 75% of the questions per subscale no total score was computed. The total scores on the Impact of Event Scale and on the Hospital Anxiety and Depression Scale were used as a measure of breast cancer specific and general distress, respectively.

Differences between the four assessment moments as well as differences in the course of both types of psychological distress for the distinguished subgroups were tested for significance with the MIXED procedure of SAS 8.2. The significance levels, including the *F*-ratios were calculated for the different measurement moments, the three distinguished factors (excessive breast self-examination, risk perception, close involvement in the breast cancer process of a sister), and the interactions of the measurement moments and the three above-mentioned factors. The following confounding variables were entered into the analysis: risk category, number of years adhering to surveillance, educational level, having a partner, having children and age. It has to be noted that in the model about the impact of excessive breast self-examination age was not included as a confounding factor, because age was already incorporated into the variable excessive breast self-examination. This vulnerable subgroup namely existed of younger women (below the age of 40) who examined their breasts more frequent than recommended [16].

3. Results

3.1. Sample characteristics

The number of participants included in the analysis per measurement moment is shown in Table 1. The differences in the number of women participating per measurement moment can be attributed to the fact that inevitably there are missing questionnaires and that in the time that the statistical analyses were done not every woman was completely assessed yet. Characteristics of the participants, distinguished by risk category, are shown in Table 2. Most characteristics were not significantly different between the three risk categories, except for the period of adherence, which was lower in the group of mutation carriers. The 29 women who did not undergo a MRI-scan as part of the surveillance programme did not differ significantly from the other participants with respect to the demographic variables as presented in Table 2. The women who did not want to participate in the psychological follow-up study did not differ significantly from the women who did, with respect to age and risk status.

In Table 3 the classification of the women into the different subgroups reporting higher psychological distress at baseline is given. Breast cancer risk perception differed significantly between the three risk categories on both the occasions of measurement. In Table 4 an overview of the type of examination on the two surveillance-moments (m1 and m4) is shown.

3.2. Breast cancer specific distress

In Table 5 the mean scores and standard errors on the Impact of Event Scale as a measure of breast cancer specific distress are displayed, for the total group as well as for the distinguished subgroups. The differences across time and/or several subgroups by means of *F*-values and *P*-values are given in Table 6.

Table 1
Number of participants per measurement moment

Assessment moment	*n* (study sample)[a]	Missing	Dropout since previous assessment	*n* (used in analyses)
m1[b]	357	12	3	342
m2[b]	354	12	9	333
m4[b]	329	18	13	298
m5[b]	295	13	1	281

[a] Differences in *n* (study sample) per assessment moment is attributed to the fact that still a number of women has to be assessed; 357 − 295 = 61 women not yet completely assessed.

[b] m1 represents the day of the first surveillance appointment during this study; m2 represents 1–4 weeks after this first appointment; m4 represents the day of the second surveillance appointment during this study (6 months after the first); and m5 represents 1–4 weeks after this second appointment.

Table 2
Patient characteristics distinguished by risk category[a]

Variable	Risk category 1 CLTR[b] 60–85% (n = 42)	Risk category 2 CLTR 30–50% (n = 201)	Risk category 3 CLTR 15–30% (n = 114)	Total (n = 357)
Age mean (sd)	40.4 (10.2)	41.1 (8.8)	39.6 (8.3)	40.5 (8.8) range: 21–63 years
Number of years of adherence mean (sd)[e]	3.1 (2.4)	5.6 (4.5)	5.6 (4.7)	5.3 (4.4) range: 0–3 years
Educational level[c]				
Lower level	7 (17%)[d]	34 (17%)	21 (19%)	61 (17%)
Middle level	23 (56%)	106 (53%)	59 (53%)	188 (54%)
Higher level	11 (27%)	59 (30%)	32 (28%)	102 (29%)
Having a partner (yes)	38 (95%)	173 (87%)	97 (87%)	308 (88%)
Having children (yes)	26 (63%)	147 (74%)	79 (71%)	252 (72%)

[a] The numbers may vary due to missing values.
[b] CLTR = cumulative life time risk.
[c] Lower level consists of primary or lower vocational education; medium level included lower or higher general secondary education or intermediate vocational education; higher level included higher vocational education or (pre-)university.
[d] Percentages indicate column percentages.
[e] Significantly different between the three risk categories (two-tailed).

Table 3
Distribution into the identified subgroups reporting more psychological distress (at baseline)[a]

Variable	Risk category 1 CLTR[b] 60–85% (n = 42)	Risk category 2 CLTR 30–50% (n = 201)	Risk category 3 CLTR 15–30% (n = 114)	Total (n = 357)
Young hypervigilant breast self examiners	6 (15%)[c]	10 (5%)	10 (9%)	26 (7%)
Others	35 (85%)	188 (95%)	102 (91%)	325 (93%)
Cognitive risk perception[d]				
Overestimators C1	–	18 (9%)	45 (42%)	63 (18%)
Accurate estimators C1	24 (58%)	86 (44%)	36 (33%)	146 (42%)
Underestimators C1	17 (42%)	92 (47%)	27 (25%)	136 (40%)
Overestimators C2	–	19 (11%)	42 (42%)	61 (20%)
Accurate estimators C2	18 (50%)	68 (39%)	27 (26%)	113 (36%)
Underestimators C2	18 (50%)	88 (50%)	32 (32%)	138 (44%)
Close involvement in BC process of a sister	11 (27%)	50 (25%)	33 (30%)	94 (27%)
Others	30 (73%)	148 (75%)	79 (70%)	257 (73%)

NB: The number of women belonging to more than one of the abovementioned categories was as follows:
Belong to all three: n = 1.
Belong to overestimators and excessive breast self examiners: n = 4.
Belong to overestimators and closely involved in BC process of a sister: n = 19.
Belong to excessive breast self examiners and closely involved in BC process of a sister: n = 3.
[a] The numbers may vary due to missing values.
[b] CLTR = cumulative life time risk.
[c] Percentages indicate column percentages.
[d] Significantly different between the three risk categories both for C1 (first surveillance appointment) and C2 (second surveillance appointment) (two-tailed).

In Fig. 1, the courses of breast cancer specific distress around both surveillance appointments are shown for the total sample and for each of the examined subgroups. For the total group breast cancer specific distress was higher before the surveillance appointments, and dropped thereafter. The difference in breast cancer specific distress before and after the control visit was statistically significant for the first surveillance appointment only (m1–m2) (F = 20.38, P < 0.00). When

Table 4
Type of examination on the two surveillance appointments (m1 and m4)

	m1 (n)[a]	m4 (n)
Physical examination only	196	188
Physical examination in combination with mammography and MRI scan[b]	160	159

[a] Due to missing values the numbers do not add up to 357.
[b] For organisational reasons the MRI scan was not always performed on the day of the clinic appointment.

Table 5
Psychological distress scores per measurement moment distinguished by subgroup

	Breast cancer specific distress mean[a] (s.e.)				General distress mean[a] (s.e.)			
	m1	m2	m4	m5	m1	m2	m4	m5
Total sample	9.6 (0.6)	7.3 (0.6)	8.1 (0.7)	7.5 (0.7)	7.9 (0.4)	6.8 (0.4)	7.3 (0.4)	6.6 (0.4)
Young excessive breast self examination								
Yes	12.9 (2.3)	14.3 (2.4)	10.5 (2.5)	10.4 (2.4)	9.2 (1.4)	7.5 (1.4)	8.1 (1.4)	6.3 (1.4)
No	9.3 (0.7)	6.7 (0.7)	7.9 (0.7)	7.3 (0.7)	7.8 (0.4)	6.7 (0.4)	7.2 (0.4)	6.6 (0.4)
Risk perception								
Underestimation	9.6 (0.9)	7.4 (0.9)	6.6 (0.9)	6.8 (0.9)	7.9 (0.5)	7.0 (0.5)	7.0 (0.5)	6.4 (0.5)
Accurate estimation	9.3 (0.8)	6.5 (0.8)	7.6 (0.9)	7.2 (0.9)	7.9 (0.5)	6.3 (0.5)	7.0 (0.5)	6.5 (0.5)
Overestimation	10.2 (1.2)	8.8 (1.2)	12.9 (1.2)	9.9 (1.2)	7.9 (0.6)	7.5 (0.6)	8.7 (0.7)	7.3 (0.7)
Closely involved in sister's BC process								
Yes	10.1 (1.3)	8.4 (1.3)	9.3 (1.3)	8.7 (1.3)	7.9 (0.7)	7.1 (0.7)	6.9 (0.7)	7.1 (0.7)
No	9.4 (0.8)	6.9 (0.8)	7.7 (0.8)	7.1 (0.8)	7.9 (0.4)	6.7 (0.4)	7.5 (0.4)	6.4 (0.4)
Imaging examination on the surveillance visit								
Yes	10.6 (0.8)	8.0 (0.8)	8.9 (0.9)	7.9 (0.9)	8.3 (0.5)	7.1 (0.5)	7.6 (0.5)	6.3 (0.5)
No	8.7 (0.8)	6.6 (0.8)	7.4 (0.8)	7.2 (0.9)	7.6 (0.4)	6.6 (0.4)	7.0 (0.4)	6.8 (0.4)

[a] Means were adjusted for risk category, number of years adhering to surveillance, educational level, having a partner, having children and age; s.e. = standard error of the mean.

Table 6
Differences in course of breast cancer specific and general distress for the total sample and the different subgroups[a]

	Breast cancer specific distress		General distress	
	F	*P*-value <	*F*	*P*-value <
Total sample				
m1–m2	20.38	0.00	18.54	0.00
m4–m5	1.28	0.26	6.92	0.01
(m1–m2)–(m4–m5)	5.16	0.02	1.11	0.29
Excessive breast self examiners *versus* others				
m1–m2	4.12	0.04	0.44	0.51
m4–m5	0.05	0.82	1.24	0.27
(m1–m2)–(m4–m5)	1.45	0.23	0.13	0.72
Accurate- and under-estimators *versus* over-estimators of breast cancer risk				
m1–m2 (under-. *versus* over-estimators)	0.35	0.55	0.52	0.47
m1–m2 (accurate-. *versus* over-estimators)	0.98	0.32	2.58	0.11
m4–m5 (under- *versus* overestimators)	4.47	0.03	1.13	0.29
m4–m5 (accurate- *versus* over-estimators)	2.87	0.09	1.13	0.29
(m1–m2)–(m4–m5)(under- *versus* over-estimators)	3.73	0.05	1.59	0.21
(m1–m2)–(m4–m5)(accurate- *versus* over-estimators)	3.67	0.06	3.49	0.06
Closely involved in BC process of a sister *versus* others				
m1–m2	0.55	0.46	0.79	0.38
m4–m5	0.00	0.99	3.88	0.05
(m1–m2)–(m4–m5)	0.27	0.61	0.66	0.42
Mammography/MRI imaging *versus* physical examination only				
m1–m2	0.23	0.63	0.18	0.67
m4–m5	0.76	0.38	2.96	0.09
(m1–m2)–(m4–m5)	0.09	0.76	0.92	0.34

[a] Analyses were adjusted for risk category, number of years adhering to surveillance, educational level, having a partner, having children and age.

investigating the course of distress around both surveillance appointments, the slope was significantly different between the first and the second appointment, indicating a higher distress level before the first appointment as compared to the second appointment ($F = 5.16$, $P < 0.02$) (see also Fig. 1).

Also, excessive breast self examiners showed an increased level of distress as compared to the others, which did not decrease after the surveillance appointment (as compared to the others (Table 5)). The course of breast cancer specific distress in younger breast self examiners differed from the other women around the

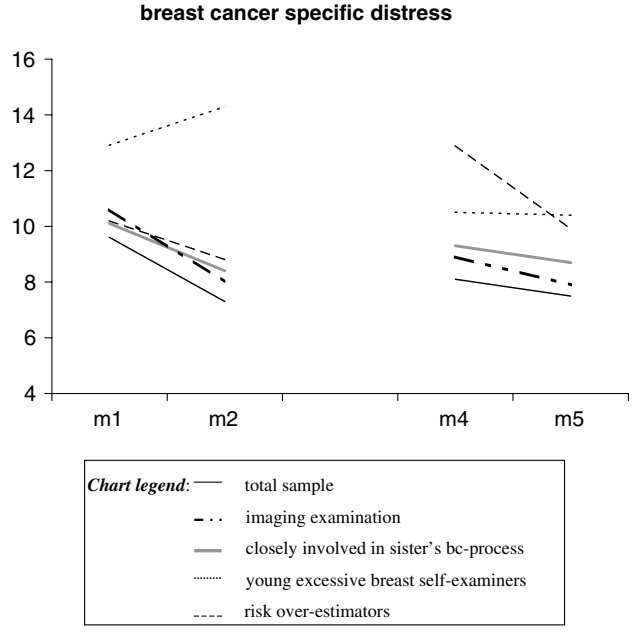

Fig. 1. The course of breast cancer specific distress for the total sample and the different subgroups around two successive surveillance appointments.

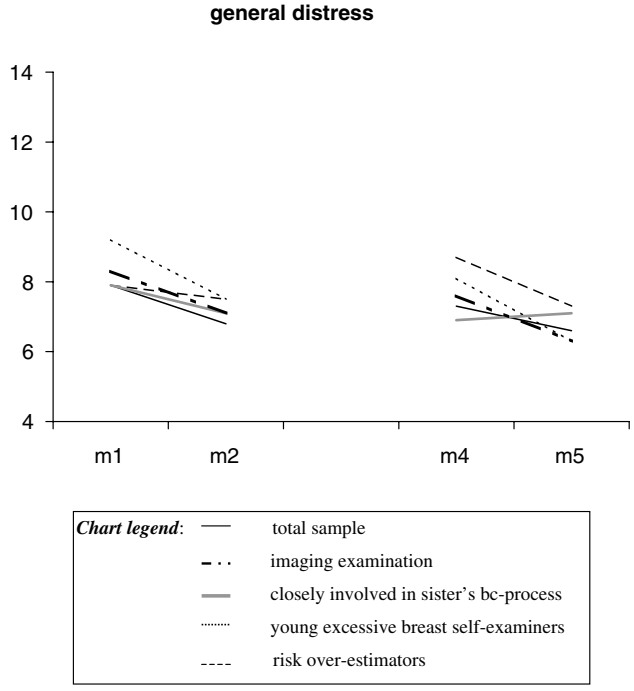

Fig. 2. The course of general distress for the total sample and the different subgroups around two successive surveillance appointments.

first surveillance appointment. The first group reported significantly higher breast cancer specific distress both before and after the appointment while the others had a lower level of distress before the clinic visit that decreased further after the appointment ($F = 4.12$, $P < 0.04$) (Table 5 and 6, Fig. 1). The course of breast cancer specific distress around the second surveillance appointment did not differ for the excessive self-examiners *versus* the others, but the degree of breast cancer distress remained higher in the group over-examiners (ns).

Regarding the relation with breast cancer risk perception, over-estimators have an increased level of distress as compared to the under-estimators at all the assessment moments, but overall the level of distress dropped after the visits. This decrease was not found for under-estimators at the second surveillance visit, but their pre-visit distress level was already low (Table 5). With regard to the course of distress, we found significant differences around the second surveillance appointment between under- and over-estimators ($F = 4.47$, $P < 0.03$) (Table 6). The latter group reported significantly higher breast cancer specific distress, particularly on the day of the second clinic appointment. The course of the distress for both groups also differed significantly from the course around the first appointment in this study ($F = 3.73$, $P < 0.05$). The risk over-estimators showed a fairly steep decrease in distress after the second surveillance appointment, which was a result of the higher distress they displayed on the day of this second surveillance appointment.

Women who have been involved in the breast cancer process of a sister showed higher levels of breast cancer specific distress in comparison with the other women. However, we did not find any significant impact or difference in BC specific distress around the control visits in relation to being involved in the BC process of a sister. Similarly, there were no significant differences between the courses of distress with regard to imaging examinations.

3.3. General distress

Data on the intensity and the course of general distress for the total sample, as well as for the different subgroups, are shown in Tables 5 and 6 and in Fig. 2. For the total group, general distress significantly decreased after both appointments ($F = 18.54$, $P < 0.00$; and $F = 6.92$, $P < 0.01$, respectively) (Table 5 and 6). Further, the course of general distress around both visits did not significantly differ from each other (Table 6, Fig. 2). The only significant difference further observed concerned the women who were closely involved in the breast cancer process of a sister. After the second surveillance appointment, these women reported more general distress than women who were not involved in the breast cancer process of a sister ($F = 3.88$, $P < 0.05$) (Table 6).

4. Discussion

4.1. In general

In this study we investigated the course of psychological distress around two successive, biannual surveillance

appointments in a substantial group of women who were at increased risk of hereditary breast cancer. The main result of our study was that for the total study sample psychological distress remained within normal ranges during the surveillance programme. Further, both types of distress, breast cancer specific and general, decreased significantly after a surveillance appointment, with the exception for breast cancer specific distress after the second appointment.

Remarkably, the fairly steep decrease of breast cancer specific distress after the first measured surveillance appointment appeared to result from the relatively high breast cancer specific distress-score on the day of the clinic appointment (as compared to the lower score on the day of the second assessed visit) (see Fig. 1). This observation suggests an effect of administering emotionally upsetting questionnaires on a probably emotionally upsetting moment (i.e. the threat of being diagnosed with an abnormality or breast cancer later that same day). Inspection of the course of general distress indicates that there is a significant decrease in distress after both clinic visits, showing a similar slope for the first and second visit. Thus, it appears that the possible stress inducing effect of administering a questionnaire is limited to questions about breast cancer rather than questions about general well being.

The pre-visit levels of breast cancer specific distress before the second surveillance appointment were considerably lower than before the first surveillance visit, whereas the post-screening levels were similar after both appointments. We hypothesise that the women prepare themselves to face possibly emotionally upsetting questions about breast cancer in combination with an actual confrontation with breast cancer specific cues.

According to the courses of both general and breast cancer specific distress as shown in Figs. 1 and 2 there is reason to assume that the regular surveillance appointments reassure the women, since almost every line depicted in these figures is decreasing, though not always significantly. Further, a higher level of distress at the day of the surveillance appointment is in fact understandable. First, the distress may be provoked by the actual threat that an abnormality or breast cancer can be diagnosed. In addition, higher levels of psychological distress may be caused by a lack of control about health when undergoing an examination in the cancer clinic. Moreover, our institute may remind the women of their (close) relatives, who learned their breast cancer diagnosis, were treated, hospitalised, or even died in this clinic.

On the other hand elevated distress levels may motivate the women to maintain their need for reassurance and safeguard their participation in a surveillance programme [25].

4.2. Vulnerable subgroups

In line with earlier studies [16,17,26,27] we found that with regard to breast cancer specific distress the younger excessive breast self-examiners and the breast cancer risk over-estimators appeared to be more vulnerable than the other participating women. Around the first measured visit in this study, the excessive breast self-examiners reported higher breast cancer specific distress, and around the second appointment the risk over-estimators did (in comparison with women underestimating their breast cancer risk). Moreover, women who were closely involved in the breast cancer process of a sister reported higher general distress after the second appointment (as compared to women not being involved in the BC process of a sister). The observation that excessive breast self-examiners and risk over-estimators reported higher breast cancer specific distress scores during the surveillance process is not entirely surprising as both groups of women were preoccupied by the fear to get breast cancer. With respect to our data, it is noteworthy to realise that 125 women of the total sample belonged to at least one subgroup, which means that at least 35% of the women in our study sample might be considered more vulnerable to psychological distress [16–18].

The observations from our study that young excessive breast self-examiners differed significantly from the remaining women around the first measured surveillance visit and the risk over-estimators did around the second surveillance visit cannot be explained clear-cut. Further studies should focus on the identification more specifically of the determinants for increased distress, i.e. young excessive examiners, over-estimators, and women involved in the breast cancer process of a sister.

In addition, we want to underscore that the women participating in this study not only represent a unique, but also perhaps also a selective group of women. Although in the analyses we adjusted for the period of adherence to regular surveillance, we cannot ignore that these women adhered to breast cancer surveillance for a mean of 5.5 years, with a range from first- or second time attendees to adhering for over 10 years. Thus, for a substantial part of these women, the surveillance appointments have become a routine check-up, and we assume that over the years the women probably have found a useful way for themselves of dealing with their high-risk status as well as the regular surveillance appointments at the clinic. Also of importance is the fact that these women participated in a surveillance programme at a Family Cancer Clinic, intended to provide surveillance for all relevant family members. In practice, this means that women may have a feeling of social support from their relatives with the same experiences regarding surveillance. The experience of clinicians and interviews with a random subgroup of study participants led us to conclude that women feel distressed

when coming to the clinic appointment. However, the reassurance afterwards is of most importance, after which most women indicate to immediately "go on with their lives" and return to daily activities and routine. We realise that a longer follow-up of these women is warranted in order to study the course of psychological distress around further surveillance appointments.

4.3. Clinical implications

There has been some debate on how extensive and customary additional psychological support should be offered. Coyne and colleagues have underlined that the mental health issues may be overemphasised, since in a lot of studies regarding psychological distress in high-risk women the results do not reach clinical significance [28–30]. Esplen *et al.* [31] provided data about favourable effects of supportive group therapy for women with BRCA1/2 mutations, although the true effect can only be demonstrated using a randomised controlled study design. In the present study, the mean distress scores were not indicative of persevering psychological distress. This suggests that most women in the surveillance process do not need extensive psychological treatment. Moreover, the elevated distress scores that we observed may very well be analogous to distress levels of women randomly drawn out of a primary care waiting room. However, the observed associations between subgroups that are more vulnerable and distress in this rather large study group should not be discarded. Consequently, some suggestions for providing additional support for these women can be given. We suggest that physicians just directly ask for breast self check more than once a week, risk estimation, and involvement in a sister's cancer process. Another approach would be the use of a one-page questionnaire with regard to the specific issues to be completed by the women. We will develop such a questionnaire as part of a forthcoming intervention study.

With regard to the need of specific professional attention for the identified vulnerable subgroups we suggest that the young excessive breast self-examiners might benefit from additional psychological counselling to learn to better cope with their fear. The kind of counselling will be dependent on the dynamics of the fear, the learning capacities of the woman, and her willingness to have professional support. The risk over-estimators may be offered additional genetic counselling to explore why they overestimate their risk. If relevant, additional psychological consultation may be considered. In view of our earlier results showing that besides the cognitive representation of one's own breast cancer risk an affective component of risk perception also has to be considered as an important indicator of psychological distress, it is of importance to pay attention to this component as well [17].

The women who are closely involved in their sister's cancer process may benefit from general psychological support to learn to adequately manage this type of distress.

4.4. Limitations of the study

Because this study was conducted in a clinical practice there was a variation in the intervals between having the examination in the clinic and receiving the results of the examination and thus in the intervals of collecting our data. In case of a physical examination the interval between m1 and m2, or m4 and m5 was one week, and in case of having additional imaging examination this interval was 4 weeks. In the analyses conducted in this study we did not consider these intervals as being different from each other, simply because our point of interest was the level of psychological distress after receiving the results.

Another comment concerns the number of statistical analyses. In light of the phenomenon of multiple testing it would have been more appropriate to fix the *P*-value at 0.01 (two sided). However, we applied a *P*-value at 0.05 (two sided) because this study was merely an exploration of the course of distress.

In conclusion, we found that breast cancer surveillance in a group of high-risk women does not induce pathological distress over time, and that distress decreases after the control visits. Further, the earlier identified vulnerable subgroups score higher with respect to both breast cancer and general distress during the surveillance programme, and notably risk over-estimators do not show a decrease of distress after the control visit. Our findings implicate that it is worthwhile to discuss extra support with the identified more vulnerable women, focussing on their own distress determinants. It should be further investigated whether this is beneficial in the long run.

Conflict of interest statement

None declared.

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