

# Development of the Informing Relatives Inventory (IRI): Assessing Index Patients' Knowledge, Motivation and Self-Efficacy Regarding the Disclosure of Hereditary Cancer Risk Information to Relatives

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## Abstract

**Background** Despite the use of genetic services, counselees do not always share hereditary cancer information with at-risk relatives. Reasons for not informing relatives may be categorized as a lack of: knowledge, motivation, and/or self-efficacy. **Purpose** This study aims to develop and test the psychometric properties of the Informing Relatives Inventory, a battery of instruments that intend to measure counselees' knowledge, motivation, and self-efficacy regarding the disclosure of hereditary cancer risk information to at-risk relatives.

**Method** Guided by the proposed conceptual framework, existing instruments were selected and new instruments were developed. We tested the instruments' acceptability, dimensionality, reliability, and criterion-related validity in consecutive index patients visiting the Clinical Genetics department with questions regarding hereditary breast and/or ovarian cancer or colon cancer. **Results** Data of 211 index patients were included (response rate=62 %). The Informing Relatives Inventory (IRI) assesses three barriers in disclosure representing seven domains. Instruments assessing index patients' (positive) motivation and self-efficacy were acceptable and reliable and suggested good criterion-related validity. Psychometric properties of instruments assessing index patients knowledge were disputable. These items were moderately accepted by index patients and the criterion-related validity was weaker.

**Conclusion** This study presents a first conceptual framework and associated inventory (IRI) that improves insight into index patients' barriers regarding the disclosure of genetic cancer information to at-risk relatives. Instruments assessing (positive) motivation and self-efficacy proved to be reliable measurements. Measuring index patients knowledge appeared to be more challenging. Further research is necessary to ensure IRI's dimensionality and sensitivity to change.

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## Abbreviations

FA factor analysis  
IRI Informing Relatives Inventory  
RMSEA root mean square error of estimation

## Introduction

Identification of hereditary or familial breast or colorectal cancer risks not only has implications for counselees but also for their relatives. According to international [1] guidelines, when a mutation is identified in a counselee, DNA testing in relatives becomes feasible. Also, surveillance, i.e., regular breast or colon screening, and prophylactic surgery may become indicated [2]. When no mutation can be identified, cancer risks are calculated based on the pedigree [3] and surveillance may still be indicated for counselees' relatives.

Genetic counselors encourage counselees to inform at-risk relatives about their genetic test result and the availability of surveillance measures [4]. This holds in particular for the first person within a family who attends genetic counseling: 'the index patient'. Index patients are central in sharing information and facilitating informed decision-making concerning risk management options for relatives [4].

Counselees indeed feel generally obliged to disclose genetic risk information to relatives [5–7]. However, they do not always succeed in correctly informing all relevant relatives [8–12]. As a result, relatives lack the opportunity to make a well-informed decision about pursuing genetic counseling, DNA-testing, and/or surveillance activities [1, 13].

If counselees' ability to be a competent, motivated, and confident informant could be enhanced, this may lead to more relatives being properly informed. On the other hand, literature shows that counselees' may wish not to inform relatives [14–17] or to discharge the responsibility of dissemination to other relatives [4]. These preferences have to be respected. The same goes for relatives' wish not to know [14, 15].

To date, several, mostly qualitative studies, have addressed counselees' barriers for informing their relatives [11, 12, 18, 19]. These may be categorized as a lack of: knowledge, motivation, and/or self-efficacy.

When counselees have a *lack of knowledge*, their understanding of which family members ought to be informed may be insufficient [19, 20]. Moreover, a lack of knowledge may lead to incorrect disclosure. Many studies have shown that genetic risk information is often poorly understood and retained [21–23]. A *lack of motivation* may be due to the desire to protect the relative or oneself, e.g., from negative reactions by the relative [7]. Counselees may consider a relative to be too old or too emotionally fragile to burden them with genetic cancer information [20]. In addition, complex family relations may prevent counselees from disclosing information [7, 12, 19]. A *lack of self-efficacy* may lead to counselees feeling unable to inform relatives. The counselee does not deliberately withhold information but may not be able to reach them or does not feel confident to inform them correctly since genetic information is complex or burdensome [24].

Although instruments are available to measure knowledge [25–27], motivation [28] and self-efficacy [29, 30], these are

either non-specific for a genetic population [25, 30] or the psychometric properties are insufficiently known [25, 28]. To the best of our knowledge, there are no instruments available that measure all foregoing elements of counselees' barriers (not) to inform relatives. Yet, if a valid inventory is available, the presence of these barriers could be assessed, their influence on information dissemination determined and the impact of interventions on barriers established.

The aim of this study was to develop and test the psychometric properties (i.e., acceptability, dimensionality, reliability, and first indication of criterion-related validity) of the Informing Relatives Inventory (IRI), a battery of instruments that intends to measure counselees' knowledge, motivation, and self-efficacy regarding the disclosure of hereditary cancer risk information to at-risk relatives.

## Methods

### Construction of Instruments

Since concise definitions of the barriers lack of knowledge, motivation, and self-efficacy are lacking, we propose a conceptual framework, subdividing the three barriers into seven domains, based on theoretical [11, 12, 18, 19] and clinical considerations (see Table 1). This conceptual framework guided the selection of instruments and the development of the IRI.

As we did not want to add to the number of instruments already available, we tried to use existing instruments where possible. Instruments phrased in English were translated into Dutch by two researchers separately. Afterwards, a bilingual psychologist translated the Dutch version back to English again to check for translation differences. New items had to be developed for the domains 'insight into which relatives need to be informed', 'knowledge about surveillance measures', and 'self-efficacy'. All instruments were combined and presented as the IRI. Two versions were developed for colon and breast cancer separately.

The questionnaire was pre-tested in five counselees, visiting the Department of Clinical Genetics of an academic hospital (one man, four women, two with colon cancer, three with breast and/or ovarian cancer, one mutation carrier, and four counselees with an inconclusive test result). Counselees commented upon the wording and difficulty of items. After minor adjustments, the questionnaire was sent to 163 participants (batch 1). Because of considerable non-response (response rate: 55 %), the questionnaire was subsequently shortened. To make completion easier, adjustments in presentation were made to items of the instruments 'insight into which relatives need to be informed' and 'motivation'. This new version of the questionnaire was tested in three counselees and subsequently sent to 180

**Table 1** Overview of barriers, domains and corresponding instruments

Barriers	Domain	No. of items	Response scale	Description/example of question
Knowledge	Insight into which relatives need to be informed	16	Percentage of correct insight into which relatives need to be informed	Which relatives has the department of clinical genetics recommended you to inform?
	Knowledge about surveillance measures	7	Yes, no, I don't know; scored as percent correct	My relatives are advised now or in the future, to get a mammography every year
	Risk perception	2	Item 1: Perceived own lifetime cancer risk: in percent and words	Item 1: According to your own thoughts and feelings, What is your risk to develop cancer (again)?
			1–7 scale: no risk—complete risk; scored as correct or false Item 2: Perceived compared cancer risk: 1–5 scale: very strongly lowered—very strongly heightened	Item 2: Comparing to other women/persons of your age, what is your risk to develop cancer (again)?
	General knowledge about hereditary cancer	6	Correct, false, I don't know; scored as percent correct	Physical examination is required only if you have symptoms.
Motivation	Positive motivation	13	1–5 scale: no role—a large role	I felt obligated to share information
	Negative motivation	17	1–5 scale: no role—a large role	I did not want to upset my relatives
Self-efficacy	Self-efficacy	7	1–4 scale: not sure at all—very sure	If you would like to inform your family, how confident are you that you have the time to inform them?

participants (Batch 2), resulting in a response rate of 69 %. Batches 1 and 2 were combined to present the results.<sup>1</sup>

### Participants

Consecutive counselees visiting the Clinical Genetics' department of three Dutch academic hospitals were invited to participate in the study between 3 weeks and 4 months after receiving a letter from the department summarizing the information provided during genetic counseling.

Inclusion criteria were: (1) first person in the family to visit the Genetics department for hereditary colorectal or breast and/or ovarian cancer: 'index patient', (2)  $\geq 1$  relative at increased risk, i.e., eligible for genetic testing and/or surveillance, (3)  $\geq 18$  years of age, and (4) ability to read Dutch. The sample comprised: index patients with conclusive test results (i.e., a pathogenic mutation had been found), an inconclusive test result, and those where DNA testing was not possible as no permission had been given. For the latter two groups, there

had to be an increased cancer risk for one or more relatives based on the family history.

Screening of summary letters resulted in 343 eligible participants. All eligible index patients received a home-sent introductory letter along with an informed consent form and a form to indicate the wish to decline participation. Questionnaire completion was performed by a web-based questionnaire system. When preferred, the questionnaire was available as a paper-and-pencil version, presented in the same layout. This study was formally exempted from formal approval by the Medical Ethics committee of the Academic Medical Center in Amsterdam, since the Medical Research Involving Human Subjects Act (WMO) does not apply.

### Measures

#### Knowledge

We considered knowledge to consist of four domains (see Table 1). First, index patients' *insight into which relatives need to be informed* was assessed by asking them to indicate the number of at-risk relatives for each of the following categories: parents, children, siblings, nephews, nieces, aunts, uncles, cousins, grandparents, and grandchildren. In the Netherlands, it is considered standard care for the Clinical Genetics department to identify at-risk relatives based on index patients pedigree [31]. The genetic counselor discusses with the index patient whose relatives are at risk and mentions these relatives

<sup>1</sup> To address problems of non-response emerging in batch 1, the instruments for positive and negative motivation were adjusted. First, six items were removed based on high inter-correlations ( $>0.3$ ). Next, deviating answer categories of ten items (makes informing harder/easier) were adjusted so they were comparable with the remaining items (plays a role in disclosure decision). No differences in score distribution (Kolmogorov-Smirnov test) and means (Mann-Whitney *U* test) were found between batches 1 and 2 on these items. Therefore, data were combined.

in the summary letter, which is sent to the index patient after the last counseling session.

Index patients' answers were compared with the advice mentioned in the summary letter, to calculate a percentage 'correct knowledge' indicating the correctly remembered relative number of at-risk family members (range 0–100 %). Only index patients who were aware that they were advised by the genetic counselor to inform at-risk relatives were asked to answer these questions ( $n=147$ ).

Secondly, index patients' *knowledge about surveillance measures for relatives* was assessed with seven items representing regular surveillance measures for breast and/or ovarian or colon cancer (e.g. regular mammography or colonoscopy). Index patients were asked to indicate, for each surveillance of the seven measures, whether it applied to any of their relatives or not (categories: yes, no, I do not know). Answers were compared with the advice mentioned in the summary letter of the index patient to calculate a percentage 'correct knowledge about surveillance options'. This indicates the relative number of screening options for family members, correctly remembered by the index patient (range 0–100 %). These items were only completed by index patients who were aware that they were advised by the department to inform their relatives about surveillance measures ( $n=142$ ).

Thirdly, index patients' *risk perception* was measured with two items: perceived lifetime breast and/or ovarian or colon cancer risk for which the respondent could indicate a percentage and perceived breast and/or ovarian or colon cancer risk as compared to the average Dutch women or person answered on a 5-point Likert scale (1 = very strongly lowered, 2 = moderately lowered, 3 = equal, 4 = moderately heightened, 5 = very strongly heightened). Answers were scored as correct (1) or incorrect (0), resulting in a range of 0–2. The accuracy of the answer was based on the difference between the index patients' perceived risk and the objective risk as indicated by the genetic counselor allowing for a 5 % difference on the first item. For the second item, response options 1 and 2 were scored as incorrect, unless the index patient had a double-sided mastectomy (in case of breast cancer), 3 as incorrect, and options 4 and 5 as correct. Finally, index patients' level of *correct knowledge about hereditary breast or colon cancer* was assessed using a selection of six items developed by Pieterse et al. [25], based on the work by Claes et al. [11]. Items were worded as statements with three response categories (correct, incorrect, or I do not know). The score for knowledge about hereditary cancer was calculated as the number of correct answers on the relevant items (range 0–6). A higher score indicated more knowledge about hereditary cancer.

### Motivation

Index patients' motivation to inform relatives was assessed by using 30 items based on the questionnaire developed by Finlay et al. [28]. All items concerned potential determinants of disclosure of information to relatives and were answered using a 5-point Likert scale (1 = factor plays no role in disclosure, 5 = factor plays a large role in disclosure). Items were presented to participants as: 'Reasons to inform relatives' (13 items) and 'Reasons for not informing relatives' (17 items). Negative motivation items were only completed by index patients who had indicated not having informed all at-risk relatives ( $n=71$ ), since these questions could only be completed by participants who were aware that they did not inform a relative. A higher total score indicated more positive (range 13–65) or negative motivation (range 17–85) to inform relatives.

### Self-efficacy

Self-efficacy is defined as an individual's perceived ability to perform a specific behavior [32]. A list of seven possible obstacles for informing at-risk relatives was derived from the literature [33–35]. Based on a scale developed by Smith and Fishbein [36], for each barrier, index patients were asked to indicate how sure they were that they would be able to overcome this particular obstacle using a 4-point Likert scale (1 = not sure at all, 4 = very sure). A higher score indicated more self-efficacy (range 0–28). For the purpose of assessing validity of the self-efficacy instrument, overall self-efficacy was assessed with one question: 'If you wanted to, how sure are you that you are able to reach all relatives who need to be informed?'

### Background Characteristics

Socio-demographic characteristics comprised index patients' age, gender, education level, and marital status. Clinical characteristics included the genetic test result and being diagnosed with cancer (yes/no). For the purpose of assessing criterion-related validity of the instruments, index patients were asked to report the number of (already) informed relatives using the same categories to assess index patients' insight into which relatives need to be informed. Answers were compared with the advice mentioned in the summary letter, resulting in a percentage 'correctly informed relatives'. Finally, index patients reported their current intention to inform (remaining) relatives on a 5-point Likert scale (1 = no, not at all, 5 = yes, definitely).



## Statistical Analyses

We assessed *acceptability*, the ease of use of an instrument [37], by examining missing data frequencies for individual items. Item non-response of more than 30 % was considered as questionable, 15–30 % as moderate and <15 % as adequate. For more insight in the characteristics of the different instruments, we assessed score distribution at instrument level (mean, percentage of minimum and maximum scores, skewness and kurtosis).

To confirm the *one-dimensional structure* of each of the domains, positive motivation ( $n=204$ ), negative motivation ( $n=71$ ), and self-efficacy ( $n=204$ ), confirmatory factor analysis (FA) was used. A  $\chi^2 > 0.05$  was used as an indication that the one-dimensional structure fits adequately. The root mean square error of estimation (RMSEA) was used as a supplementary statistic. Values <0.05 suggested an adequate fit, those between 0.05 and 0.08 indicated a moderate fit, and values >0.08 indicated a questionable fit of the one-dimensional model [38].

Possibilities to analyze the dimensionality of the instruments' assessing 'knowledge' were limited. First, it is because the instruments for 'insight into which relatives to inform' and 'knowledge about surveillance measures' are count variables. Secondly, risk perception is only measured with two items. Correspondence with the author who developed the items about 'knowledge about hereditary cancer' [25] led us to conclude that these items were not designed to measure one underlying construct.

The *reliability* of the instruments was determined by calculating the instruments' internal consistency using Cronbach's alpha. Inter-item and corrected item-total correlations were also calculated. The average inter-item correlation should ideally fall between 0.15 and 0.50 [39]. In a reliable instrument, items correlate with the overall score of the instrument, and we considered corrected item-total correlations to be at least >0.3 [40]. The reliability of the instruments comprising the domain knowledge was not assessed.

*Criterion-related validity* was investigated by determining the association between the instruments' scores and intention to inform relatives and the percentage correctly informed relatives. We assume that index patients with high knowledge, motivation, and self-efficacy scores would be more inclined to inform at-risk relatives and would have informed more relatives. Significant associations between the instruments' scores, intention to inform relatives, and percentage correct informed relatives would suggest adequate criterion-related validity. We considered one significant association (either with intention or informed relatives) as moderate criterion-related validity and no significant associations as questionable criterion-related validity.

*Validity of the self-efficacy instrument* was additionally assessed by correlating the total score of the separate items

with overall self-efficacy. All analyses were performed using SPSS 19.0.

## Results

### Descriptives

Of all 339 index patients who received the questionnaire, 278 (82 %) responded, 67 (20 %) of whom declined participation, resulting in a response rate of 62 % ( $n=211$ ). Most frequent reasons to decline were (1) lack of time ( $n=20$ ); (2) too much burden ( $n=20$ ), and (3) assuming not having to inform relatives ( $n=7$ ). Data of seven index patients with high rates of missing data were excluded. Included index patients were significantly older ( $M=54$ ,  $SD=11$ ) than non-responders ( $M=52$ ,  $SD=15$ ), ( $p<0.05$ ), but did not differ in gender and disease type (see Table 2).

### Knowledge

*Acceptability* The instrument assessing 'insight into which relatives need to be informed' was less acceptable with percentages of missings varying between 13 and 33. 'Knowledge about screening options' was moderately accepted with percentages of missings between 6 and 23. The instrument assessing 'risk perception' was well accepted with percentages of missings between 2 and 5. 'Knowledge about hereditary cancer' was also well accepted with percentages of missings between 1 and 2 (see Table 3).

*Criterion-Related Validity* More insight into which relatives need to be informed was significantly related to a higher percentage of informed relatives ( $r(127)=0.74$ ,  $p<0.001$ ) supporting moderate criterion-related validity. No significant associations were found between other 'knowledge' instruments and intention to inform relatives or the 'percentage informed relatives', suggesting that these knowledge instruments' criterion-related validity is not supported.

### Motivation

*Acceptability* The instrument assessing positive motivation was moderately accepted with percentages of missings varying between 10 and 18. The instrument for negative motivation was completely missed-out by 20 index patients. The remaining index patients ( $n=51$ ) accepted it moderately with percentages of missings varying between 2 and 14 (see Table 3). The distribution of the instrument for 'positive motivation' was right-skewed whereas the instrument for 'negative motivation' was left-skewed, indicating that

**Table 2** Demographic characteristics of the sample ( $n=204$ )

Patient characteristics	Included patients	Non-participants ( $N=139$ )	<i>P</i> -value
Gender ( $n=204$ )			0.60
Male	45 (22 %)	34 (25 %)	
Female	159 (78 %)	105 (76 %)	
Age ( $n=204$ ) [mean (SD)]	54 (11)	52 (15)	0.02
Range	18–80	20–86	
Education <sup>a</sup> ( $n=198$ )			
Low	27 (14 %)		
Middle	107 (54 %)		
High	56 (27 %)		
Personal situation ( $n=200$ )			
Married/partner	157 (79 %)		
Widowhood	10 (5 %)		
Single	33 (16 %)		
Counseled for ( $n=204$ )			0.92
Breast and/or ovarian cancer	103 (50 %)	71 (51 %)	
Colon cancer	101 (50 %)	68 (49 %)	
Carrier status ( $n=204$ )			
Mutation	21 (10 %)		
Inconclusive test result	183 (90 %)		
Diagnosed with cancer ( $n=204$ )			
Breast and/or ovarian cancer	79 (39 %)		
Colon cancer	48 (24 %)		
Other	27 (13 %)		
No	65 (32 %)		

<sup>a</sup> *Low*: non/primary school, *middle*: secondary/lower level vocational school, *high*: college/university

reasons to inform relatives play a more important role than reasons not the inform relatives.

**Dimensionality** Positive motivation: The confirmatory one-factor FA showed that all items loaded substantially on one component, with an eigenvalue of 4.25, explaining 26.91 % of the variance (see Table 4). However, the one-dimensional structure did not fit adequately:  $\chi^2$  (65,  $n=156$ )=240.42 ( $p<0.000$ ) and RMSEA=0.15.

Negative motivation: The confirmatory one-factor FA indicated a questionable fit of the proposed one-dimensional model (eigenvalue of 7.21, explaining 41.35 % of the variance.  $\chi^2$  (119,  $n=43$ )=276.20 ( $p<0.000$ ) and RMSEA=0.18) (see Table 4).

**Reliability** The internal consistency of the instruments was considered good (positive motivation:  $\alpha=0.82$ ; negative motivation:  $\alpha=0.90$ ). The items for positive and negative motivation appeared to be well related to the instrument, indicated by corrected item-total correlations of  $>0.30$ . Inter-item correlations for positive motivation were acceptable ranging from 0.09 to .66. Inter-item correlations for negative motivation ranged from  $-0.06$  to 0.95, exceeding the acceptable range between 0.10 and 0.50.

**Criterion-Related Validity** More positive motivation was significantly related to a stronger intention to inform relatives ( $r(101)=0.34$ ,  $p=0.05$ ) and to a higher percentage of correctly informed relatives ( $r(138)=0.28$ ,  $p=0.001$ ), supporting adequate criterion-related validity. Negative motivation was not related to the intention to inform relatives and the percentage of correctly

**Table 3** Acceptability and score distribution of the instruments

	Missing participant percentage	Missing Medical file percentage	Score range	Mean (SD)	Floor percentage	Ceiling percentage	Skewness (SE)	Kurtosis (SE)
<b>Knowledge</b>								
Correct insight who to inform ( $n=147$ )	13–33	1	0–100	78.54 (30.73)	4.8	54.4	$-1.27^*$ (0.21)	0.40 (0.41)
Correct knowledge about surveillance measures ( $n=142$ )	10–23	2.5	0–100	79.43 (24.65)	0.0	45.1	$-0.67^*$ (0.22)	$-1.03^*$ (0.44)
Correct risk perception ( $n=204$ )	2–5	8–24	0–2	1.01 (0.78)	50.0	25.5	$-0.02$ (0.11)	$-1.37^*$ (0.37)
Correct knowledge about hereditary cancer ( $n=204$ )	1–2	–	0–6	4.23	0.0	14.7	$-0.24$ (0.17)	$-0.59$ (0.34)
<b>Motivation</b>								
Positive motivation ( $n=204$ )	10–18	–	13–65	40.14 (13.76)	0.5	1.0	$-0.78^*$ (0.18)	0.56 (0.35)
Negative motivation ( $n=71$ )	2–14	–	17–85	26.69 (12.93)	11.3	1.4	$1.93^*$ (0.33)	$7.43^*$ (0.66)
Self-efficacy ( $n=204$ )	9–11	–	0–28	20.11 (5.61)	3.4	6.4	$-0.59^*$ (0.18)	$-0.27$ (0.36)

*SD* standard deviation, *SE* standard error

\*  $p<0.05$

**Table 4** Factor loadings three separate confirmatory factor analysis: one-factor solution on the items of positive motivation, negative motivation, and self-efficacy

	Factor loadings
Items of positive motivation <sup>a</sup>	
a. I feel obliged to provide information	0.50
b. I was encouraged by the health professional	0.49
c. I already promised my relatives to inform them before I got the result	0.55
d. I'm encouraged by other relatives to disclose the information	0.64
e. I think the information can help them to make medical decisions	0.49
f. A relative asked me about the test result	0.54
g. I would like to have my relatives' advice when I make medical decisions	0.62
h. I need emotional support from my family	0.64
i. I would like to encourage my relative to go for genetic services	0.49
j. I would like to encourage my relative to go for regular screening	0.51
k. I understand the test result well	0.34
l. I have a close relationship with some of my relatives	0.44
m. I think relatives' children should know the information	0.43
Items of negative motivation <sup>b</sup>	
a. I do not have any contact with some of my relatives	0.35
b. I do not have a good relationship with some of my relatives	0.33
c. I do not believe the information would be useful to them	0.37
d. I do not think the relative can handle the information emotionally	0.70
e. I am worried about being responsible for causing problems in a relationship or marriage	0.58
f. My relative told me they did not want to know	0.62
g. I do not have time to tell my relatives	0.47
h. It is difficult to reach some of my relatives	0.49
i. I do not want to burden my relative, since he/she is having difficulties	0.71
j. I consider my relative too young to inform	0.69
k. It is emotionally difficult for me to share the information	0.73
l. The information is so complex, I do not know how to share the information	0.75
m. I do not know who has an increased risk for having hereditary cancer	0.76
n. Some relatives do not understand how the information applies to them	0.77
o. I do not want to upset my relatives	0.73
p. I feel guilty or anxious about the test result	0.90
q. I feel the information is too personal to share	0.79
Items of self-efficacy <sup>c</sup>	
a. Find the time to speak them	0.78
b. Contact them	0.85
c. Disclose the information clearly	0.91
d. Cause undesirable turbulence	0.70
e. Explain what the importance of the information is for them	0.88
f. Avoid problems in the relationship	0.79
g. Have enough knowledge	0.66

<sup>a</sup> One-factor solution on the items of positive motivation, capturing 26.91 % of the variance ( $n=156$ )

<sup>b</sup> One-factor solution on the items of negative motivation, capturing 41.35 % of the variance ( $n=43$ )

<sup>c</sup> One-factor solution on the items of self-efficacy, capturing 64.23 % of the variance ( $n=174$ )

informed relatives, suggesting that criterion-related validity is not supporting.

#### Self-Efficacy

**Acceptability** The item non-response for the instrument assessing self-efficacy was acceptable with percentages of

missings varying between 9 and 11 (see Table 3). The distribution was right-skewed, indicating high self-reported self-efficacy in participants.

**Dimensionality** The confirmatory one-factor FA resulted in an unsatisfactory model fit, explaining 64.23 % of the variance ( $\chi^2(14, n=174)=128.92$  ( $p<0.000$ ) and RMSEA=0.21) (see Table 4).

**Reliability** The internal consistency was satisfactory ( $\alpha=0.92$ ). The items of the instrument assessing self-efficacy appeared to be well related to the instrument, indicated by corrected item-total correlations of  $>0.30$ . Inter-item correlations were high ranging from 0.43 to 0.84.

**Criterion-Related Validity** Higher self-efficacy was significantly related to higher intention to inform relatives ( $r(99)=0.30, p<0.05$ ) and a higher percentage of at-risk relatives that needed to be informed ( $r(137)=0.30, p<0.001$ ), supporting criterion-related validity in this self-efficacy instrument.

**Validity of the Self-Efficacy Instrument** All individual self-efficacy items were significantly associated with the item to assess overall self-efficacy ( $r=0.39-0.77$ ), indicating validity of the instrument.

## Conclusions

Sharing genetic cancer information with at-risk relatives is a complex process [13, 23, 41]. To move forward, the process of family communication should be assessed and desirable outcomes and instruments (beyond a one-dimensional assessment of the number of relatives informed) should be defined [41].

This study aimed to establish psychometric properties of such instruments. We presented a first conceptual framework that aimed to improve insight into index patients' barriers regarding the disclosure of genetic cancer information to at-risk relatives. Based on literature and clinical considerations, we defined three barriers in disclosing cancer risk information: lack of knowledge, motivation, and self-efficacy. Subsequently, we developed the IRI, an inventory that provided assessment of those barriers and assessed its' psychometric properties.

The findings show that especially the instruments assessing index patients' positive motivation and their sense of self-efficacy to be reliable and acceptable to index patients. The criterion-related validity is adequate and although both instruments are moderately related to each other, correlations do not appear that high that they must be considered to address one concept. Dimensionality analyses lead us to conclude that the proposed one-dimensional structure of the instruments is questionable. The items loaded substantially and explained quite some variance, but supplementary statistics ( $\chi^2$  and RMSEA) showed that the one-dimensional structure did not fit properly. It is likely that the items have more in common (more covariance between the items) than the proposed one-factor structure can explain. This may also be caused by the skewed distributions of the instruments.

For the self-efficacy instrument, we recommend a minor alteration: 'Are you able to communicate...' instead of 'Are you able to reach...' makes it more clear that we intend to measure index patients perceived ability to disclose the information to at-risk relatives.

The instrument assessing index patients' negative motivation appears to be reliable as well. It is moderately accepted by index patients; nevertheless, we qualified the dimensionality and predictive validity of the instrument as weak. Moreover, results have to be interpreted carefully since the sample size for this instrument was small ( $n=51$ ). To prevent this amount of missing data, we recommend adjusting the question of negative motivation from 'Why did you not inform your relative' to 'Reasons for not informing relatives'. In this case, all participants will be able to complete this items.

We are, to our knowledge, the first to measure index patients' 'insight into which relatives to inform' and 'knowledge about surveillance options' and compare index patients' answers to the advice given by the Department of Clinical Genetics. This enabled us to classify index patients' knowledge as correct or not. However, assessing the psychometric properties of the 'knowledge' instruments was challenging. Whereas instruments assessing risk perception and knowledge about hereditary cancer were well accepted, and knowledge about surveillance options was moderately accepted, the scoring of 'insight into which relatives need to be informed' showed many missing items, indicating poor acceptability. It remains unclear how to interpret these missings: either as a lack of insight into which relatives to inform, or as an indication that a relative does not need to be informed or as 'carelessness' of the respondent. An explanation for the large number of missing data may be the large proportion of participants with an inconclusive test result (90 %). Index patients may only have disclosed information which they perceived as relevant to their relatives [42]. Other reasons may be low education, black or white thinking (a mutation means bad news, no mutation means good news), or having an own opinion about cancer risks and hereditary likelihood [42]. Despite several attempts to provide as clear instructions and/or format as possible, we may not have succeeded in this respect. We feel that a structured personal interview is needed for a valid assessment of index patients' insight into which relatives to inform.

Some methodological issues need to be addressed. First, sample size differed per instrument, due to (1) adaptations made to the instruments (batch 1 vs. batch 2) and (2) the fact that—interestingly—27 % of the samples ( $n=57$ ) were not aware that they were advised by the genetic counselor to inform at-risk relatives or that their relatives had an advice for surveillance measures ( $n=62, 29\%$ ). The latter finding emphasizes the need for strategies that assist index patients to communicate with at-risk relatives [15, 41, 42]. *Post hoc* analyses showed no significant differences between participants who were and were not aware of the advice given by the



genetic counselor in age, gender, educational level, carrier status, type of counseling (breast and/or ovarian or colon cancer), hospital, and time between receiving the summary letter and completing the questionnaire.

Second, despite precautionary measures like personalization of the letter, an enclosed return envelope with stamp, ensured anonymity, and an invitation of the clinical geneticist and reminders sent at three time points, the response rate was lower than expected (i.e., 61 %, respectively). The length and complexity of the questionnaire (127 items and 15 pages) might explain this. Participants' comments in the survey suggested problems with understanding the items about which relatives to inform in particular. Yet, other Dutch psychosocial studies in the field of Clinical Genetics report comparable response rates [23, 43], suggesting these generally healthy users of this medical service to be less likely to contribute to research than populations of (chronically) ill patients.

The instruments we developed should not be considered as the final version. Although a first indication of criterion-related validity was addressed, future research is necessary to ensure the validity of the new instruments, in particular construct validity. Moreover, test-retest reliability should be examined to confirm found results and the instrument should be tested on sensitivity to change.

In conclusion, the current study has contributed to the possibility of assessing index patients' barriers regarding the disclosure of cancer risk information to at-risk relatives. The instruments assessing positive motivation and self-efficacy are reliable measurements to assess these barriers in index patients. However, future research is necessary to further ensure reliability, validity, and sensitivity to change of the new instruments.

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**Competing interests** The authors declare that they have no competing interests.

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