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The "Psychosocial Aspects in Hereditary Cancer" questionnaire in women attending breast cancer genetic clinics: Psychometric validation across French-, German- and Spanish-language versions

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Abstract

Background: We performed a comprehensive assessment of the psychometrics of the "Psychosocial Aspects in Hereditary Cancer" (PAHC) questionnaire in French, German and Spanish.

Methods: Women consecutively approached in Cancer Genetic Clinics completed the PAHC, distress and satisfaction questionnaires at pre-testing (T1) and after test result disclosure (T2). In addition to standard psychometric attributes, we assessed the PAHC ability to respond to change (i.e. improvement or deterioration from T1 to T2) in perceived difficulties and computed minimal important differences (MID) in PAHC scores as compared with self-reported needs for additional counselling.

Results: Of 738 eligible counselees, 214 (90%) in France (Paris), 301 (92%) in Germany (Cologne) and 133 (77%) in Spain (Barcelona) completed the PAHC. A six-factor revised PAHC model yielded acceptable CFA goodness-of-fit indexes and good all scales internal consistencies. PAHC scales demonstrated expected conceptual differences with distress and satisfaction with counselling. Different levels of psychosocial difficulties were evidenced between counselees' subgroups and over time (p-values < .05). MID estimates ranged from 8 to 15 for improvement and 9 to 21 for deterioration.

Conclusion: The PAHC French, German and Spanish versions are reliable and valid for evaluating the psychosocial difficulties of women at high BC risk attending genetic clinics.

KEYWORDS

breast cancer, cross-cultural, genetics, minimal important difference, psychometrics

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1 | INTRODUCTION

The hereditary breast and ovarian cancer (HBOC) syndrome is one of the most common hereditary cancer syndromes (Hiraki, Rinella, Schnabel, Oratz, & Ostrer, 2014), suggested by a family history of breast or ovarian cancer, and caused by alterations in major *BRCA1* or *BRCA2* susceptibility genes (Couch, Nathanson, & Offit, 2014). Recently, other breast or ovarian cancer susceptibility genes have been identified (Taylor et al., 2018). Simultaneous tests of multiple genes (gene panels) are now available and are currently being implemented in oncology practice (Domchek, Bradbury, Garber, Offit, & Robson, 2013).

With these advances, genetic counselling has become increasingly complex. The information imparted may be difficult to interpret by counselees and potentially lead to false reassurance, inappropriate cancer risk management decisions and psychosocial distress (Bradbury et al., 2018). There is a need to monitor the psychosocial difficulties that counselees may experience in this evolving genetic practice, identify their risk and protective factors, their impact on well-being, health behaviours and familial communication, and design and assess alternative models of genetic service delivery (Albada, Dulmen, Lindhout, Bensing, & Ausems, 2012; Bradbury et al., 2018).

When tested for a highly penetrant cancer syndrome such as the HBOC and after careful pre-test counselling, most individuals do not appear to experience prolonged, clinically significant anxiety (Hamilton, Lobel, & Moyer, 2009). However, the majority of counselees experience psychosocial difficulties that are specific to the genetic context (Eijzenga, Aaronson, et al., 2014), which may then accentuate psychological distress (Farrelly et al., 2013) and elicit further counselling needs (Eijzenga, Aaronson, et al., 2014). Addressing unmet psychosocial needs during the genetic consultation may reduce distress levels (Eijzenga, Bleiker, et al., 2014) and potentially result in better understanding of information provided, higher perceived control and satisfaction (Pieterse, Ausems, Dulmen, Beemer, & Bensing, 2005).

Based on the European Organisation for Research and Treatment of Cancer Quality of Life Group guidelines (Johnson et al., 2011), the "Psychosocial Aspects in Hereditary Cancer" (PAHC) questionnaire was recently developed to screen for the presence of genetic-specific psychosocial difficulties in clinical practice (Eijzenga, Aaronson, et al., 2014; Eijzenga, Bleiker, et al., 2014). Items of the PAHC address psychosocial concerns organised in six conceptually distinct domains. As this questionnaire may also be useful in observational studies and intervention trials, its dimensionality should also be demonstrated empirically.

The primary aim of the current study was to empirically test the PAHC questionnaire's conceptual structure. In addition, we also assessed the questionnaire in terms of internal consistency, convergent and divergent validity, known-groups validity, responsiveness to change and interpretability (minimal important difference (MID)) (Mokkink et al., 2010).

This study was undertaken within the "Breast Cancer Risk after Diagnostic Gene Sequencing" (BRIDGES) consortium (https://bridges-research.eu), which provided the opportunity to investigate the PAHC's psychometric performance in women attending genetics clinics for familial breast cancer in France, Germany and Spain.

2 | METHODS

This study protocol was approved in France by the *Comité consultatif sur le traitement de l'information en matière de recherche dans le domaine de la santé* (CCTIRS: Consultative committee for information management in health research—No 16.314) and by the *Commission Nationale Informatique et Libertés* (CNIL: French Information Technology and Privacy Commission), in Germany by the Ethics Committee of the University Hospital of Cologne (No 16-098) and, in Spain by the Ethics Committee of the Instituto Catalán de Oncología of Barcelona (No—PR111/16). All recruited women provided written informed consent.

2.1 | Participants and procedure

From October 2016 to April 2018, women over the age of 18 years, unaffected or affected with a primary non-metastatic breast cancer, and eligible for BC gene panel or targeted testing, were consecutively recruited in the Cancer Genetic Clinic of Institute Curie (France), the University Hospital of Cologne (Germany) and the Instituto Catalán de Oncología of Barcelona (Spain).

The primary analysis focused on evaluating the hypothesised scale structure of the 26-item PAHC. For confirmatory factor analysis, it is recommended to have a minimum of 5 cases per item in order to establish if there is a stable scale structure to the questionnaire

(Tabachnik & Fidell, 2001). Thus, we needed a minimum of 130 participants per country.

The study objectives were explained to the women on the day of the pre-test counselling visit (T1) and, when they agreed to participate, they were given questionnaires to complete at home, either on paper or online, and to return within the next two weeks. If necessary, one reminder was made by telephone call. Questionnaires not completed or received within 2 months after each genetic consultation were considered missing. Two months after the genetic test result disclosure consultation (T2) (i.e. between 1 and 8 months after T1), women were contacted again to complete the PAHC questionnaire following the same procedure.

2.2 | Core measure

2.2.1 | Psychosocial difficulties

The PAHC questionnaire comprises six domains of psychosocial difficulties (i.e. hereditary predisposition, practical issues, family and social issues, living with cancer, general emotions and children-related issues) (Eijzenga, Aaronson, et al., 2014). All items are scored on a four-point Likert scale with response options scored 1 (not at all), 2 (a little), 3 (quite a bit) and 4 (a lot). A non-applicable response option is also provided to items that may not be relevant in some circumstances (e.g. depending on the assessment time—pre- or post-testing, personal or familial actual cancer diagnosis). Additional PAHC items address counselees' needs for further counselling (yes/no) in relation to the domains and overall.

The PAHC was translated, adapted and pilot-tested into French, German and Spanish languages according to international guidelines (Kuliś et al., 2017). A report on this process is available on the BRIDGES website (https://bridges-research.eu/wp-content/uploads/2018/02/D52-PAHC-PU-v1.0.pdf/).

PAHC scale scores were transformed on a 0–100 scale, with higher scores for increasing difficulties. All questionnaire scale scores were computed replacing missing data by the mean score of the scale if a response was provided for more than 50% of the items in the scale; for children-related items, response frequencies were computed only on women with children (Fayers & Machin 2000).

2.3 | Comparative measures

2.3.1 | Psychological distress

The French (Razavi, Delvaux, Farvacques, & Robaye, 1990), German (Herrmann-Lingen *et al.* 2011) and Spanish (Herrero et al., 2003) versions of the Hospital Anxiety and Depression (HADS) were used to assess symptoms of anxiety and depression.

2.3.2 | Specific distress

The Impact of Event Scale-Revised (IES-R) (Weiss & Marmar, 1997), available in French (Brunet, St-Hilaire, Jehel, & King, 2003), German (Maerker & Schützwohl, 2000) and Spanish (Báguena et al., 2001) measures psychological reactions to a stressful event (i.e. in this case, referring to cancer risk).

Satisfaction with the genetic counselling visit (Pieterse, Dulmen, Beemer, Bensing, & Ausems, 2007) comprises eight items addressing expertise, communication, client-centeredness, information, responses to preferences, access, service organisation and general satisfaction rated on a 10-point rating scale ranging from 1 (very bad) to 10 (excellent). This questionnaire was translated using standard EORTC procedures (Dewolf, Velikova, Johnson, Scott & Bottomley, 2009).

All comparative scales presented adequate internal consistencies with Cronbach's alphas above the 0.70 threshold, except for the HADS-Depression French-version scale ($\alpha = .67$).

Needs for additional counselling were assessed by the specific PAHC items related to the six domains (as described above).

Additional data on socio-demographic characteristics were provided by the patient and clinical data were obtained from the medical record, including BC diagnosis status or type of genetic test result, that is positive (a pathogenic variant is identified) versus true negative (in women who underwent predictive targeted testing) or non-informative (in women who underwent diagnostic gene panel testing) results.

2.4 | Statistical analyses

Baseline socio-demographic and clinical characteristics of the participants were described using mean (standard deviation) or median (range) for continuous variables and number (percentage) for categorical variables. We used the Kruskal–Wallis test or Student's *t* test for quantitative data and the chi-square statistic for categorical. Effect sizes (Cohen's d) for group differences are given where values of 0.2 are considered small, 0.5 of moderate and 0.8 of large (Cohen, 1988).

Confirmatory factor analysis (CFA) was used to evaluate empirically the scale structure of the PAHC. We first tested a predefined conceptual model (Eijzenga, Aaronson, et al., 2014). Goodness of fit was evaluated using the chi-square goodness-of-fit statistics and fit indexes: Root Mean Square Error of Approximation (RMSEA), Comparative Fit Index (CFI) and Tucker-Lewis Index (TLI). Values <0.08 or <0.05 for the RMSEA, >0.90 or >0.95 for the CFI and TLI are indicative of acceptable or good fit between the hypothesised model and the observed data; the smallest likelihood ratio (χ^2/df) and Akaike information criteria (AIC) were sought (Jackson, Gillaspy, & Purc-Stephenson, 2009). Alternative measurement models were subsequently evaluated, where necessary.

Internal consistency reliability was calculated using Cronbach's coefficient alpha, with values >0.70 used as the criterion for acceptable internal consistency at the group level (Cronbach & Warrington, 1951).

Correlation analyses were performed for subscales of the PAHC with scores of HADS, IES-R scales and "Satisfaction with the consultation" scales; correlations >0.50 were considered indicative of convergent validity and correlations r < 0.35 of divergent validity. Spearman or Pearson correlations were used, depending on the score distributions. We expected the distress scales to be highly correlated with the

PAHC emotional scale (Brédart *et al*. 2005; Farrelly et al., 2013) (convergent validity) and moderately correlated (divergent validity) with other PAHC scales (Eijzenga et al., 2015), and the PAHC scales and the "satisfaction with the consultation" scale to be moderately correlated (Brédart *et al*. 2005; Pieterse et al., 2005) (divergent validity).

Known-group validity was evaluated based on hypothesised differences in PAHC scale scores as a function of socio-demographic and clinical characteristics. We hypothesised that younger counselees (Farrelly et al., 2013), those with a lower education, having children (Eijzenga et al., 2015) or affected with BC (van Roosmalen et al., 2004) will present higher levels of difficulties; differences between country settings were also expected due to case mix and counselling specificities (known-group differences).

Responsiveness was evaluated by calculating mean changes in PAHC scale scores between the two assessment points and by genetic test result. We hypothesised that the PAHC scale scores would change significantly over time (Bennett et al., 2012) overall, and according to positive (*BRCA1/2*) or negative test results (i.e. no pathogenic variant was found; the result is negative non-informative and true negative; we excluded women who received variant of uncertain significance result to compare two clear categories of women) (Esteban et al., 2018; Lumish et al., 2017; Oberguggenberger et al., 2016) (responsiveness to change).

The MID was assessed for each PAHC scale based on an anchor-based counselee-derived approach (Jaeschke, Singer, & Guyatt, 1989), using the six domain-specific counselee's self-reported needs for additional counselling at T1 and at T2 as anchors. A correlation of 0.30 or higher was considered as an acceptable association between the anchor and each scale at T1 (Revicki, Hays, Cella, & Sloan, 2008). Each anchor was constructed considering three categories: no change (no need or persistent need at both measurement points), deteriorated (developed need at T2 not reported at T1) and improved (need reported at T1 not reported at T2). Subsequently, differences in PAHC scale scores were reported for each category of the anchor. To control for change observed for stable patients according to the anchor, MID estimates were obtained using the difference in two consecutive categories of the anchor, that is "improvement" versus "no change" and "no change" versus "deterioration" (Cella et al., 2002). The 95% confidence intervals (CI) for the differences in mean change scores are reported. Anchor-based MID estimations were compared with the statistical distribution based method, based on fractions of 0.2, 0.3 and 0.5 of the PAHC scales' standard deviation (*SD*) at T1 and at T2, and where estimates <0.2 *SD* were considered too small (Maringwa et al., 2011).

3 | RESULTS

3.1 | Patients' characteristics and completion rate

Among 239 counselees in France (Paris), 326 in Germany (Cologne) and 173 in Spain (Barcelona) consecutively approached, who accepted to participate in the study, 214 (90%), 301 (92%) and 133 (77%), respectively at T1, and 167 (70%), 221 (73%) and 67 (39%), respectively at T2, returned the PAHC questionnaire.

As shown in Table 1, the mean age (standard deviation) of the French, German and Spanish respondents was 47.9 (11.9), 47.4 (10.7) and 47.7 (11.8) years, respectively, and, 172 (80%), 253 (84%) and 86 (65%), respectively, were affected by BC.

There were no statistically significant differences observed between the 648 respondents and the 90 non-respondents in mean age (47.6 vs. 46.6 years), having children (75% vs. 73%) or being affected by BC (79% vs. 70%).

3.2 | Questionnaire acceptability and item descriptive statistics

Among the 648 respondents, 533 (83%) completed all PAHC items. The level of missing data per item was below 5% in all three countries. On a item scale score from 1 to 4, the mean item scores range from 1.17 to 3.29 (Supplementary material S1).

3.3 | Confirmatory factor analyses

The data did not fit the originally hypothesised PAHC model well (i.e. the 6 originally hypothesised factors by Eijzenga et al. (2014)). Three successive, revised models were tested based on conceptual meaning and factor loadings. The best six-factor model yielded acceptable fit (χ^2/df = 4.64; RMSEA = 0.075 [90% CI: 0.071–0.079]; CFI = 0.870, TLI = 0.848, AIC = 34,358.863), which could be improved (Supplementary materials S2 and S3).

Examination of residuals evidenced pairs of items within the same factor with higher observed correlations than reproduced correlations. These pairs appeared semantically redundant [i.e. items 8 and 9 (r observed = 0.65 and r reproduced = 0.60), 19 and 20 (r observed = 0.56 and r reproduced = 0.33) and 23 and 26 (r observed = 0.43 and r reproduced = 0.30)]. This model was thus modified allowing correlations between residuals of three redundant pairs of items within a same factor. This modified exploratory model provided an improved, more acceptable fit (χ^2/df = 3.64, RMSEA = 0.061 [90% CI: 0.057–0.066], CFI = 0.91 and TLI = 0.90).

Two new scales distinguish issues related to personal and familial cancer from the original "Living with cancer" domain. The original "Practical issues" domain was removed as one of the two items (item 7) had a low factor loadings (0.30). A table comparing the original hypothesised and newly proposed structure is provided in the Supplementary material S2.

The six dimensions pertain to 1) "Hereditary predisposition" (items 1–4, 6); 2) "Familial and social issues" (items 8–11, 13); 3) "Emotions" (items 14–18); 4) "Familial cancer" (items 12, 19, 20, 23); 5) "Personal cancer" (items 21, 22); 6) "Children-related issues" (items 24–26).

3.4 | Reliability

The internal consistency of the PAHC scales was adequate, with Cronbach's alpha coefficients ranging from 0.71 ("Personal cancer") to 0.87 ("Emotions") (Supplementary material S1).

TABLE 1 Socio-demographic and clinical characteristics across samples

	French respondents (*N* = 214)	German respondents (*N* = 301)	Spanish respondents (*N* = 133)
Age (years)			
Mean (*SD*)	47.9 (11.9)	47.4 (10.7)	47.7 (11.8)
Median (Q1-Q3)	48 (39–56)	48 (41–54)	48 (41–55)
Range	21–78	18–77	19–80
Education level (%)			
Medium education level or below	6 (2.83)	37 (12.37)	46 (34.59)
Secondary or superior education	206 (97.17)	262 (87.63)	87 (65.41)
Missing data	2	2	0
Marital status (%)			
Married/partnered	150 (70.4)	212 (71.1)	102 (77.3)
Others (widowed, separated/ divorced, single/never married)	63 (29.6)	86 (28.9)	30 (22.7)
Missing data	1	3	1
Having children (%)			
Yes	171 (79.9)	213 (70.8)	103 (77.4)
Personal breast cancer (%)			
Yes	172 (80.4)	253 (84.1)	86 (64.7)
Breast cancer lifetime risk			
BOADICEA estimates			
Mean (*SD*)	19.6 (11.9)	18.1 (9.2)	–
Median (range)	16.5 (0.7–81.1)	16.5 (0.7–81.1)	–
Type of genetic test *n* (%)*			
Gene panel test	179 (84.0)	242 (82.0)	61 (62.2)
Targeted test	34 (16.0)	53 (18.0)	37 (37.8)
Genetic test result *n* (%)*			
BRCA1/2 or other high/moderate-risk pathogenic variant	33 (15.6)	66 (22.3)	21 (21.4)
Uninformative/True negative	163 (77.3)	190 (64.4)	55 (56.1)
Variant uncertain significance (VUS)	15 (7.1)	39 (13.2)	22 (22.4)

Abbreviations: *SD*, standard deviation; Q1-Q3: first-third quartile.
*1/3, 6, 35 genetic test type/results unavailable at time of study analysis for the French, German and Spanish samples respectively.

3.5 | Convergent-divergent validity

As expected, the PAHC "Emotions" correlated above 0.50 with the HADS and the IES-R scales. Correlations of other PAHC scales with HADS and IES-R ranged from 0.13 for "Familial cancer" and HADS-Depression to 0.49 for "Hereditary predisposition" and IES-R-Intrusion. All *p*-values were below .001. The almost null correlations between the PAHC and the satisfaction scales indicate that these scales address different concepts (Table 2).

3.6 | Known-group differences

Except for "Emotions", all scales of the PAHC were able to discriminate between counselees according to one or more of the clinical (BC diagnosis status), socio-demographic characteristics (age, level of education) or country criteria (*p*-values < .05) (Table 3).

3.7 | Responsiveness to change

Except for "Familial and social issues" and "Emotions", changes in mean scores of the PAHC scales over time were all statistically significant, ranging from −2.05 (p = .034) ("Familial cancer") to −8.51 (p < .0001) ("Hereditary predisposition") (Table 4). Analysis of change over time by genetic test result showed significant results only for ("Familial and social issues"), with a mean difference of 4.92 for women receiving a positive (i.e. pathogenic variant) result compared with 0.20 for those with a negative (i.e. non-informative or true negative) result (p = .04).

TABLE 2 Correlation between the PAHC scales and, HADS, IES-R and Satisfaction with the consultation scales (*N* = 648)

PAHC scales	HADS-Anxiety	HADS-Depression	IES-Hyperarousal	IES-Intrusion	IES-Avoidance	Satisfaction with consultation
Hereditary predisposition (HP)	0.42****	*0.24****	0.44****	0.49****	0.40****	*−0.01 NS*
Familial & social issues (FSI)	0.45****	0.38****	0.41****	0.44****	0.35****	*−0.04 NS*
Emotions (E)	**0.75****	**0.57****	**0.63****	**0.66****	**0.50****	*0.0 NS*
Familial cancer (FC)	*0.31****	*0.13****	*0.24****	*0.32****	*0.27****	*0.06 NS*
Personal cancer (PC)	0.43****	0.36****	0.41****	0.47****	*0.35****	*0.03 NS*
Children-related issues (CRI)	*0.27****	*0.17****	*0.25****	*0.30****	*0.27****	*0.06 NS*

Note: Correlations higher than |0.50| are in bold and lower than |0.35| in italic.
HADS, Hospital Anxiety and Depression Scale subscale scores; IES-R, Impact of Event Scale-Revised; NS, non-significant.
All scales Cronbach's alphas above.70 in the different language versions except for HADS-Depression in French (α = .67).
*p < .05.
**p < .01.
***p < .001.
****p < .0001.

3.8 | Minimal important difference

The mean change in scores of the PAHC scales by categories of the anchor is reported in Table 5. The deterioration category (i.e. occurrence of need for further counselling after the test result disclosure) comprised only few patients (ranging from 6 to 14). Differences in mean change between "improvement" (i.e. resolution of need for further counselling after the test result disclosure) and "no change" (i.e. no need or persistence of need at both time points) were statistically significant for each PAHC scale, with MID values varying from 8.42 for "Personal cancer" to 14.86 for "Emotions". Differences in mean change between "deterioration" and "no change" were significant for "Family and social issues" (21.05), "Emotions" (9.07) and "Personal cancer" (16.19).

Distribution of PAHC scores at T1 and T2 were similar (Supplementary material S4). The observed anchor-based MID values were close to 0.3 and 0.5 *SD*; the non-significant MID anchor-based values for deterioration in "Familial cancer" and "Children" were also reflected in more restricted score distribution, as below 0.2 *SD*.

4 | DISCUSSION

Our results provide evidence of adequate psychometric properties for the PAHC scales in women at high BC risk attending genetics clinics in France, Germany and Spain. The questionnaire proved to be acceptable to patients, with a low number of missing item responses across countries.

The PAHC conceptual model originally hypothesised was partially confirmed empirically in the current study. Our factor analysis led to make a distinction between items related to personal and familial cancer issues originally imbedded in the "Living with cancer" domain; this reflects the difference between the impact of a hereditary cancer syndrome on those already affected by cancer versus on those who are unaffected at the time of genetic testing.

The new PAHC scale structure exhibits satisfactory index-of-fit and good internal consistency estimates. As expected (Brédart *et al*. 2005; Eijzenga et al., 2015; Farrelly et al., 2013), apart from the PAHC "Emotions" scale, most PAHC scales exhibited only moderate correlations with the distress scales. All PAHC scales evidenced almost no correlations with satisfaction with the consultation. This may be explained, in our sample, as has been observed in other studies (Pieterse et al., 2005, Oberguggenberger et al., 2016), by a very high mean score (8.9 on a 1–10 scale) and little score variability (1.7) in the satisfaction scale scores.

Most PAHC scale scores were able to discriminate between counselees who differed in terms of age, education level, parental status, BC diagnosis or country in the hypothesised direction. In addition, most PAHC scales evidenced significant change over time.

Differences in counselees' reported need for additional counselling at pre-test and after genetic test result disclosure served as the anchor to appraise the minimal important difference. The mean change in PAHC scores was in the expected direction. Counselees classified as having improved by the anchor tended to present lower PAHC scores while those classified as having worsened by the anchor tended to present higher PAHC scores, and those classified as having not changed by the anchor generally presented PAHC scores close to 0. MID estimates varied between PAHC scales as has been reported for other patient-reported outcome measures (Maringwa et al., 2011). The range of the anchor-based MID was 8 to 15 points for improvement, which is in line with the 5%–10% range of the instrument (5–10 points on 0–100 scale; Maringwa et al., 2011). For most PAHC scales, the anchor-based MID for improvement was close to 0.5 *SD* estimates, which reflects a "moderate" effect size.

TABLE 3 Known-group comparisons

	Age				
	Mean (*SD*)				
PAHC scales*	<40, *N* = 158	41–50, *N* = 231	>50, *N* = 259	*p*-Value	Effect size
Hereditary predisposition (HP)	36.62 (21.65)	35.26 (25.17)	32.27 (24.45)	.036	0.08
Familial & social issues (FSI)	15.13 (15.25)	14.91 (16.05)	15.93 (18.55)	.809	0.009
Emotions (E)	35.01 (24.81)	30.05 (22.09)	30.18 (23.66)	.102	0.08
Familial cancer (FC)	62.56 (27.05)	61.16 (25.90)	64.36 (25.70)	.384	0.06
Personal cancer (PC)	55.84 (29.35)	59.49 (27.37)	56.47 (29.51)	.429	0.05
Children-related issues (CRI)	50.24 (28.03)	50.78 (28.17)	45.80 (25.73)	.234	0.09
	Level of education				
	Medium education level or below, *N* = 89	**Secondary or superior education, *N* = 555**			
Hereditary predisposition (HP)	39.57 (27.48)	33.63 (23.45)		.056	0.23
Familial & social issues (FSI)	12.52 (16.42)	15.71 (16.67)		.100	0.19
Emotions (E)	29.36 (23.78)	31.78 (23.71)		.376	0.10
Familial cancer (FC)	68.36 (27.27)	61.80 (25.68)		.029	0.25
Personal cancer (PC)	56.90 (32.60)	57.25 (28.15)		.916	0.01
Children-related issues (CRI)	55.86 (27.09)	47.15 (26.95)		.010	0.32
	Having children				
	Yes, *N* = 487	**No, *N* = 161**			
Hereditary predisposition (HP)	34.89 (24.79)	33.67 (22.12)		.581	0.05
Familial & social issues (FSI)	16.02 (17.01)	13.70 (16.42)		.134	0.14
Emotions (E)	31.07 (23.61)	33.19 (24.22)		.329	0.09
Familial cancer (FC)	63.97 (25.86)	59.44 (26.12)		.056	0.17
Personal cancer (PC)	57.90 (28.89)	55.42 (28.51)		.344	0.09
Children-related issues (CRI)	48.64 (27.10)	—			
	Affected/unaffected with BC				
	Yes, *N* = 511	**No, *N* = 137**			
Hereditary predisposition (HP)	33.27 (23.74)	39.51 (25.04)		.007	0.26
Familial & social issues (FSI)	15.90 (17.06)	13.74 (16.16)		.187	0.13
Emotions (E)	31.42 (23.78)	32.29 (23.79)		.708	0.04
Familial cancer (FC)	60.63 (25.95)	71.15 (24.42)		<.0001	0.42
Personal cancer (PC)	62.25 (26.28)	38.43 (30.19)		<.0001	0.84
Children-related issues (CRI)	36.79 (30.92)	34.93 (33.94)		.150	0.06
	Country				
	German, *N* = 294	**Spanish, *N* = 129**	**French, *N* = 210**		
Hereditary predisposition (HP)	33.85 (24.12)	45.19 (26.56)	28.99 (20.26)	<.0001	0.26
Familial & social issues (FSI)	16.76 (16.83)	11.84 (15.99)	15.84 (17.26)	.0006	0.11
Emotions (E)	33.89 (24.27)	29.27 (23.23)	29.82 (23.17)	.067	0.09
Familial cancer (FC)	51.94 (24.31)	79.99 (18.70)	67.65 (25.01)	<.0001	0.48
Personal cancer (PC)	58.58 (28.65)	53.66 (31.72)	57.70 (26.97)	.456	0.07
Children-related issues (CRI)	30.61 (29.96)	46.86 (33.68)	38.26 (30.69)	<.0001	0.25

*Among different scales, sample sizes vary depending on the number of missing data; we provide: *N* = minimum sample size among the different scales; all PAHC scores are on a 0–100 scale; higher scores indicate higher expressed difficulties; values of 0.2, 0.5 and 0.8 for effect size are considered small, moderate and large respectively. *p*-Value of *t* tests for two sample or Kruskal–Wallis for three samples.

TABLE 4 Responsiveness to change among patients over T1 and T2 assessment times overall and, by genetic test result

	T1	T2	T2-T1[a]		Positive, *N* = 90	Negative/Uninformative, *N* = 405	
PAHC scales	Mean (*SD*)	Mean (*SD*)	Mean difference	*p*-Value	Mean difference	Mean difference	*p*-Value
Hereditary predisposition (HP)	33.03 (23.36)	24.52 (24.20)	−8.51 (24.52)	<.0001	−3.94 (23.0)	−8.44 (23.35)	.14
Familial & social issues (FSI)	14.72 (16.05)	15.05 (19.18)	0.34 (18.96)	.712	4.92 (17.84)	0.20 (17.73)	.04
Emotions (E)	31.34 (23.49)	29.51 (23.31)	−1.83 (20.52)	.062	−0.12 (18.19)	−2.32 (20.10)	.39
Familial cancer (FC)	62.17 (25.99)	60.12 (27.97)	−2.05 (20.45)	.034	−0.34 (19.58)	−2.12 (20.25)	.50
Personal cancer (PC)	57.90 (28.40)	53.73 (28.17)	−4.18 (24.88)	.0004	0.68 (22.30)	−4.57 (25.67)	.11
Children-related issues (CRI)	46.54 (26.40)	38.86 (26.86)	−5.76 (19.65)	<.0001	−2.56 (19.35)	−8.26 (22.92)	.10

[a]Among different scales, sample sizes vary depending on the number of missing data;Patients responding at both assessment times; *N* range: 332 (Children)–449 (Hereditary predisposition); Positive test result *N* range: 50 (Children)–73 (Familial Cancer and Personal Cancer); Negative/uninformative test result *N* range: 222 (Children)–296 (Hereditary predisposition). *p*-Value of paired *t* test.

TABLE 5 Change in need for help per PAHC scales over T1 and T2 assessment times and mean (*SD*) of PAHC change scores in three anchor-defined groups and the MID (95% CI) between no need and need

				MID (95% CI)* Difference in mean change	
PAHC scales	Improved, no need anymore, *N* = 48	No change, persistent need or no need, *N* = 263	Deteriorate, developed need, *N* = 6	Improvement No need anymore—no change	Deterioration No change—developed need
Hereditary predisposition (HP)	−20.61 (28.15)	−6.09 (23.03)	1.07 (25.78)	−14.52 (−22.0; −7.04)	−7.16 (−20.88; 6.56)
Familial & social issues (FSI)	−8.24 (26.52)	0.71 (15.73)	21.76 (22.60)	−8.96 (−16.88; −1.04)	−21.05 (−31.62; −10.48)
Emotions (E)	−15.51 (24.30)	−0.65 (19.19)	8.42 (17.33)	−14.86 (−22.02; −7.70)	−9.07 (−17.12; −1.02)
Familial cancer (FC)	−10.91 (24.07)	−0.62 (19.58)	1.19 (20.37)	−10.29 (−16.38; −4.20)	−1.81 (−12.68; 9.06)
Personal cancer (PC)	−11.52 (29.82)	−3.10 (23.80)	13.10 (21.86)	−8.42 (−15.94; −0.90)	−16.19 (−27.92; −4.46)
Children-related issues (CRI)	−15.60 (31.89)	−5.92 (19.92)	−5.56 (18.26)	−9.68 (−19.11; −0.25)	−0.36 (−15.17; 14.45)

Abbreviation: MID, Minimal clinical importance. 95% CI comprising 0 are statistically non-significant.

Excluding values below 0.2 *SD* (too small effect size), anchor-based MID for deterioration ranged from 9 to 21 points; however, further investigation is required considering the small number of counselees classified as having worsened by the anchor in this study.

From these results, clinicians and researchers using the PAHC may consider that an intra- or inter-individual difference of around 10 on the 0–100 PAHC scales may be interpreted as counselees' perceived change in difficulties that may imply a need (negative change) or not (positive change) for additional counselling.

4.1 | Study limitations

We need to acknowledge a number of limitations of our study. First, although the PAHC was developed for any hereditary cancer syndrome, in this study, its psychometric performance was only addressed in women confronted with a HBOC syndrome. Moreover, the samples comprised primarily women opting for genetic testing and who were affected with BC. Although this reflects the current population of counselees in the participating centres (respondents did not differ on key available characteristics), our results may not generalise to broader populations.

Second, while we addressed most of the psychometric properties of the PAHC (Mokkink *et al*., 2010), it was not feasible in the context of this study to assess the temporal reliability (test-retest).

Third, MID was determined based on one anchor only. Although this anchor may be understandable and clinically relevant to clinicians and health managers in terms of estimating the need of personnel for genetic counselling service, a distinction can be made between reported difficulties and perceived need for psychosocial care (Brédart et al., 2013), and there are significant barriers to seeking psychosocial support (Sun et al., 2018).

5 | CONCLUSIONS

The French, German and Spanish language versions of the PAHC questionnaire may be used to better understand counselees' psychosocial difficulties and highlight their needs and potential gaps in

genetic counselling services. This study provided an empirically derived PAHC scale structure, which is valid, reliable and responsive in assessing psychosocial difficulties in women attending genetic clinics for high BC risk.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

ETHICAL APPROVAL

Reference no for ethical approval in France (No 16.314), Germany (No 16–098) and Spain (No PR111/16).

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