Empirical Validation of the Genetic Psychosocial Risk Instrument – French Version (GPRI-F)

Validation empirique de la version française de l'instrument de dépistage psychogénétique (GPRI-F)

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Abstract

Objective: This study sought to validate a French version of the Genetic Psychosocial Risk Instrument (GPRI), an instrument used to screen for psychosocial risk in individuals undergoing genetic testing for adult-onset hereditary disease (AOHD). Methods: The French translation of the GPRI (GPRI-F) was first validated linguistically with ten French individuals undergoing genetic testing for AOHD. Then, a psychometric evaluation of the GPRI-F was carried out with a separate cohort of 132 individuals. Convergent validity was assessed with the Impact of Event Scale (IES) and the Perceived Risk Scale (PR), while divergent validity was assessed with a quality of life measure, the SF-8. Results: The 20-item GPRI-F obtained a Cronbach’s α of .81, indicating a good level of internal consistency. Factor analysis yielded a psychometrically sound four-factor solution. The optimal GPRI-F cut-off was determined to be 55 and resulted in an area under the curve of .769. The GPRI-F also had improved sensitivity and specificity relative to the onco-genetic assessment of the treating medical doctor. The GPRI-F was also found to have satisfactory internal validity. Discussion and conclusions: Overall, the psychometric properties obtained in this study support the reliability and validity of using the GPRI-F with French-speaking individuals scheduled to receive genetic counselling and testing to identify those who are at heightened psychological risk and who might benefit from added psychosocial support.

Résumé

Objectif : Cette étude visait à valider une version française de l’instrument de risque psychosocial génétique (GPRI) initialement développé en anglais. Cet instrument sert pour le dépistage du risque psychosocial chez les personnes subissant un test génétique pour une maladie héréditaire de l’adulte. Méthodes : Le GPRI a été traduit en français (GPRI-F) et validé linguistiquement avec dix individus francophones soumis à ce test génétique. Après cette validation, une cohorte distincte de 132 personnes a participé à l’évaluation psychométrique avec le GPRI-F. La validité convergente a été évaluée avec l’échelle d’impact d’événements (IES) et l’échelle de risque perçue (PR) tandis que la validité divergente a été évaluée en utilisant une mesure de la qualité de vie, SF-8. Résultats : L’alpha de Cronbach (α) du GPRI-F à 20 items était de 0,81 indiquant un bon niveau de cohérence interne. L’analyse factorielle a été utilisée pour déterminer la structure factorielle sous-jacente dans les variables et une solution psychométrique à quatre facteurs a été obtenue. Le seuil optimal de GPRI-F a été calculé à 55 et représente une aire sous la courbe de 0,769. Le GPRI-F avait également une sensibilité et une spécificité améliorées par rapport à l’évaluation onco-génétique du médecin traitant. Le GPRI-F présente également une validité interne satisfaissante. Discussion et conclusions : Dans l’ensemble, les propriétés psychométriques obtenues dans cette étude confirment la fidélité et la validité de l’utilisation du GPRI-F avec des individus francophones devant recevoir une consultation génétique afin d’identifier ceux qui présentent un risque psychologique accru et qui pourraient bénéficier d’un soutien psychosocial supplémentaire.

Keywords
- genetic testing;
- distress;
- tool validation;
- psychosocial risk

Mots-clés
- dépistage génétique;
- détresse génétique;
- validation d’instrument;
- risque psychosocial
INTRODUCTION

Individuals with a family history of hereditary diseases carry a burden of stress from being at higher risk of developing the diseases (Baum, Friedman, & Zakowski, 1997; Dorval et al., 2005; Hamilton, Lobel, & Moyer, 2009; Lerman & Schwartz, 1993). In the case of cancer, genetic testing can be used to estimate the chances of developing the disease and, for people at high risk, appropriate detection and prevention measures can be applied. Unfortunately, there are psychological risks associated with genetic testing itself (Bjørnslett, Dahl, Sørebø, & Dørum, 2015; Eijzenga, Hahn, Aaronson, Kluijt, & Bleiker, 2014; Hirschberg, Chan-Smutko, & Pirl, 2015). While the majority of individuals suffer little or only short-term psychological effects from receiving genetic testing (Hamilton et al., 2009), the impact on some subgroups can be considerable, regardless of test results. The negative psychosocial impact of genetic testing includes distress (Gritz et al., 2005), more specifically, distress associated with pressure to inform family members of a mutation they may share, isolation from other family members (McInerney-Leo et al., 2005), survivor’s guilt if found not to carry the mutation, depression, anxiety disorders (van Dijk et al., 2006), and cancer-related worries after receiving BRCA test results (Dagan & Shochat, 2009). Early identification of individuals potentially at higher risk of suffering distress and negative psychological effects following genetic testing makes it possible to allocate valuable psychosocial resources to those most in need. Esplen et al. (2013) recently developed a screening instrument to identify individuals undergoing genetic testing for adult-onset hereditary disease (AOHD) potentially at higher risk for adverse psychosocial effects. The Genetic Psychosocial Risk Instrument is a 20-item scale comprising three subscales. Its score range runs from 13 to 101 and it has a cutoff score of 50 (Esplen et al., 2013). The original English-language GPRI was validated on a sample of 800 individuals with a recognized family AOHD, including cancer. The first subscale of the GPRI is composed of 12 items that examine the person’s anticipated or experienced impact of being at high risk for AOHD. These items include the following: “My worries about the disease affect my daily mood” and “The disease for which I am at risk is currently causing a significant disruption in my family life”. The second subscale comprises five items regarding a person’s history or vulnerability in the area of mental health. These include the following: “I have been diagnosed with a depressive or anxiety disorder in the past.” Lastly, the third subscale comprises three items that deal with the person’s or their family’s experience with the genetic disease in question. These include the following: “I have taken care of a very ill parent or another family member.”

Developed specifically for the context of genetic testing, the GPRI has the added value of focusing not just on symptoms of anxiety, like general measures such as the Impact of Event Scale (IES) do (Horowitz, Wilner, & Alvarez, 1979), but also on variables associated with the genetic testing process (Esplen et al., 2013). As the IES was not designed to examine variables associated with heritable disease or genetic testing or risk, it is not clinically specific, and it may pose barriers for use in the context of genetic testing. Furthermore, whereas the IES is used to investigate a person’s current psychological state, the GPRI was developed to identify individuals liable to experience significant difficulty adjusting in future to the genetic information they receive. As such, the GPRI appears to be a better choice, at face value, than general anxiety measures for clinicians who wish to determine whether clients need additional professional support in the wake of genetic testing (Esplen et al., 2013). Such a screening tool can help reduce future distress by identifying at-risk individuals that might benefit from preventative interventions to contain their level of distress. The GPRI has also been modified to test the psychosocial risk of receiving a diagnosis of autosomal genetic dominant polycystic kidney disease (Simms, Thong, Dworschak, & Ong, 2016).

To date, only the original English-language version of the GPRI has been validated. A French-language version of the GPRI (GPRI-F; see
Appendix A) could meet the needs of the large French-speaking population worldwide. To date, there are no validated French measures that specifically assesses genetic testing distress. The number of French speakers in the world is estimated at 274 million, which makes French the fifth most spoken language. Consequently, it is essential that a measure of genetic testing distress be available in French and that such an instrument possess psychometric properties comparable to those of its English-language counterpart.

Against this background, we undertook a study to collect initial evidence of the validity of the GPRI-F and to characterize the instrument’s psychometric properties. We asked the following question: What are the psychometric properties of the GPRI-F in terms of reliability and of convergent, divergent, and construct validity? We hypothesized that the GPRI-F would demonstrate good validity and reliability. In addition, a cut-off needed to be determined to establish clinical guidelines for the purpose of referrals for further assessment.

METHODS

DESIGN

The research design selected was that of a cohort study.

PATIENT RECRUITMENT

This study was conducted with French-speaking individuals recruited at a genetic testing centre in the Marseille region of France. The service provider received a description sheet of the study and was informed of the recruitment process and of the project’s rationale through clinical educational rounds. The most frequently requested genetic tests at the centre were for adult-onset inherited cancers, which include breast/ovarian cancer and colon cancer. The individuals had to meet the following inclusion criteria: 1) be undergoing genetic testing for BRCA1 and BRCA2 (BRCA1/2) breast/ovarian cancer susceptibility gene mutations; 2) fluent in French, and 3) at least 18 years of age at time of study. Exclusion criteria included a current psychiatric disorder identified by the treating medical onco-geneticist and the inability to complete the French-language version of the questionnaires. As was the case when the original scale was validated, both individuals with and without the disease (cancer) were included, as previous work has shown disease status not to be associated with post-genetic testing distress levels (Bish et al., 2002; Hamilton et al., 2009). The research ethics board of the principal investigator’s primary affiliated institution (York University, Toronto, at time of study) approved the study and issued certificate number #2011-078.

TRANSLATION OF GPRI TO GPRI-F AND LINGUISTIC VALIDATION

The original GPRI’s psychometric properties were established and validated with 800 individuals tested for AOHD (Esplen et al., 2013). The authors of the original GPRI consented to have the scale translated to French and validated. The first author of the original scale was a co-author of this study. In accordance with the guidelines of the EORTC Quality of Life Study Group (de Haes et al., 2000), the GPRI was translated into French by four individuals in order to minimize translation bias. Specifically, the GPRI was first translated independently by a bilingual native English speaker and a bilingual native French speaker. Then, two authors of the present study, CM (residing in Canada) and CJR (residing in France), both bilingual content experts, reviewed the two independent translations to arrive at a final version by consensus. All the items were subjected to this process to ensure that nothing from the original scale was lost in translation. The translation were then pilot-tested on 10 French-speaking patients scheduled for genetic testing for BRCA1/2. These patients were approached prior to their genetic testing appointment with their genetic counsellor in the waiting room of the testing site. After completing the GPRI-F as per the EORTC guidelines for building structured interviews (de Haes et al., 2000), the patients took part in cognitive interviewing and were asked if any of the items were: a) difficult to answer; b)
confusing; c) difficult to understand; and d) upsetting/offensive. Patients were also asked whether they would have formulated items differently. Two participants found three items to be confusing. These were reviewed and refined for clarity by CM and CJR.

**Empirical Validation of GPRI-F**

**Sample size**

Once the GPRI-F’s cross-linguistic sensitivity was established, the instrument was administered to a large sample of patients scheduled for genetic testing or genetic counselling for AOHD, specifically BRCA1/2. We intended to recruit 200 patients, a minimum of 10 participants per item, as recommended in the guidelines for conducting confirmatory analysis (Anthoine, Moret, Regnault, Sébille, & Hardouin, 2014). However, as recruitment proved harder and longer than anticipated, we settled for a sample size of 132.

**Instruments**

To determine whether a measurement procedure provides a valid assessment of a given construct, the procedure’s convergent and divergent validity must be established (Messick, 1979; Wainer & Braun, 1988). This is done by assessing the strength of the relationship between the scores from two different measurement procedures, for example, by measuring the correlation between two scales (Esplen et al., 2013). A positive correlation indicates convergent validity and a negative correlation indicates divergent validity. To examine the convergent validity of the GPRI-F, participants were asked to complete a questionnaire package containing the GPRI-F (some items rated on a 5-point scale and others dichotomized) with two other French-language measures related to the construct under study. Specifically, to establish convergent validity, the 15-item IES (Horowitz et al., 1979) was used. The IES is a general measure of subjective information processing formulation of stress response, alternating between phases of intrusion and avoidance (Joseph, 2000). Respondents must indicate frequency of given events in the past week on a four-point scale (0 = not at all; 1 = rarely; 3 = sometimes; 5 = often). Scores can range from 0 to 75, with higher scores indicating higher frequency of intrusive thoughts (score range of 0-35) and attempts at avoidance (score range of 0-40). Individuals respond to the items based on an anchored distressing event, in this case, going through genetic testing. A total IES score of 23 or more was the cut-off used to indicate clinical concern (Devilly & Spence, 1999), as was used with a similar study population (Maheu et al., 2015). The psychometric properties of the IES scale are satisfactory: internal consistency of .84–.91 and test-retest reliability of .83 (Thewes, Meiser, & Hickie, 2001). One item regarding percentage perceived risk (PR) of developing cancer (Esplen et al., 2013) was also used for convergent validity, illness perception being a known predictor of distress (Arran, Craufurd, & Simpson, 2014).

As higher anxiety is generally associated with lower health-related quality of life (HRQL; Li et al., 2016), the SF-8 licensed version of the HRQL Questionnaire was used to establish divergent validity (Ardern-Jones, Kenen, Lynch, Doherty, & Eeles, 2010; de Haes et al., 2000). The SF-8 measures eight HRQL domains summarized into physical components (PCS) and mental components (MCS) to produce a summarized score representing the respondent’s overall general perception of their physical and mental health (Ware, Kosinski, Dewey, & Gandek, 2001).

The two subscales have been shown to have good internal consistency (Cronbach’s alpha .61-.68; Roberts, Browne, Ocakes, Oyok, & Sondorp, 2008). Lastly, we further established construct validity by examining the correlation between GPRI-F total score and other common distress measures, including the IES, PR and the SF-8. All the scales mentioned above have been documented extensively in the genetics literature and used with populations at inherited risk to assess the impact of genetic testing on the general physical and mental health of individuals.

Additional predictive scoring was used whereby the treating MD, an onco-geneticist (FE), was asked to note whether, in his opinion, a patient might benefit from additional psychosocial support. The IES measure was used as the
benchmark to determine the sensitivity and specificity of both the GRPI-F and the MD’s evaluation.

**Baseline procedures**

Patients received their questionnaire packages from their treating MD at the end of the genetic consultation. Each package contained a letter describing the study, two copies of a consent form, and one copy of each questionnaire (GPRI-F, IES, PR, and SF-8). The first author provided an explanation of the study procedures to interested individuals, and the consent form was reviewed before obtaining their signature. All participants completed the questionnaire package on site in an average time of 15 minutes.

**Data analysis: Scale reliability and validity**

The GPRI-F was examined to confirm that it met the criteria for internal consistency, which requires a Cronbach’s $\alpha$ close to .80 for the scale and each item and inter-item correlations in the range of .20 to .40, as suggested by Briggs and Cheek (1986). Confirmatory factor analysis (CFA) was performed to evaluate the three-factor structure derived from the English version of the GPRI. The non-normed fit index, the adjusted goodness of fit index, the standardized root mean squared residual, and the chi-squared statistic were used to determine goodness of fit. Since the total score was used to determine a patient’s psychological condition, verifying the scale’s internal structure was essential to ensure the scale’s reliability. A factor analysis was carried out to understand the underlying relationship between the measured variables. Knowing the factor structure helps relate individual items to latent constructs and interpret the results for individual items. As the GPRI’s three-factor structure did not provide a good fit for the GPRI-F, an exploratory factor analysis was carried out to determine the underlying factor structure of the variables. A psychometrically sound four-factor solution was obtained. The construct validity of the GPRI-F was verified by assessing its correlations with the other study measures (IES, PR, and SF-8). The predictive power of the GPRI-F was evaluated by analyzing the receiver operating characteristic (ROC) curve using the IES cut-off score as the benchmark.

**RESULTS**

**Participant characteristics**

A total of 132 individuals undergoing genetic testing for BRCA1/2 took part in the GPRI-F validation study. All received in-person genetic counselling from their treating MD, an onco-geneticist (FE). Most participants were female (79%) with an average age of 49 years, and more than half recently experienced a significant event, either the reception of a cancer diagnosis or the loss of a significant other (Table 1). The average GPRI-F score was 52.9+13.32 (Table 2). Females had a significantly higher score than males (54.2+12.8 vs. 46.2+12.8, p-value < .005).

**Data screening and outlier analysis**

The data were examined for univariate outliers, normality and multivariate outliers in the preliminary analyses. Univariate outliers were defined as data points falling either 3.29 standard deviations (p-value < .001; two-tailed test) above or below the mean (Tabachnick & Fidell, 2013). No outliers emerged on this basis. Normality was evaluated on the basis of distribution skewness and kurtosis. The criteria used were the following: skewness/S.E. < |0.5| and kurtosis/S.E. < |5| (given n > 100). All total scale scores were within skewness and kurtosis tolerances. The Mahalanobis distance was computed for each record to check for multivariate outliers by entering into a multiple regression with an arbitrary numerical subject code as the dependent variable. No multivariate outliers emerged with the chi-squared cut-off and a p-value < .01.

**Confirmatory Factor Analysis**

The structural equation program (SEQ) was applied to assess the validity of the three GPRI subscales. A CFA of the GPRI-F was performed using the maximum likelihood estimation procedure to test the three-subscale structure.
reported in the literature (Esplen et al., 2013). The item factor loadings of the model were relatively low. Overall, the statistical results did not support the three-subscale factor model for the GPRI-F. The chi-square value was calculated to evaluate overall model fit and assess the discrepancy between the sample and fitted covariance matrices. A good model fit would yield an insignificant result at a .05 threshold (Barrett, 2007; Hu & Bentler, 1995). The calculated chi-square value of 502.18 with 165 degrees of freedom was statistically significant, indicating that the correlation matrix reproduced by the proposed model differed significantly from the actual observed correlation matrix. The Bentler comparative fit index was .61, and the Bentler-Bonett non-normed index was .55, both lower than the suggested cut-off value of .95 (Hu & Bentler, 1995). The standardized root mean square residual was .12, which was greater than the criterion of .05 (Byrne, 1998; Diamantopoulos & Siguaw, 2000) and the root mean square error of approximation (RMSEA) was .13. These results all suggested that the three-factor model was not a good fit. The lead author of the GPRI (Esplen et al., 2013) suggested that the factor structure of the English version might differ from that of the GPRI-F as a function of the symptom severity of

| Table 1 |
| Study Participants’ Characteristics (n = 132) |

<table>
<thead>
<tr>
<th>Variables in GPRI-F</th>
<th>M</th>
<th>SD</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age, in years</td>
<td>48.88</td>
<td>±14.76</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Gender</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>23</td>
<td>17.4%</td>
</tr>
<tr>
<td>Female</td>
<td>104</td>
<td>78.8%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Marital status</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Married/with partner</td>
<td>91</td>
<td>68.9%</td>
</tr>
<tr>
<td>No partner</td>
<td>30</td>
<td>22.7%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Genetic testing status</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Blood sample given, awaiting test results</td>
<td>15</td>
<td>11.4%</td>
</tr>
<tr>
<td>Blood sample not yet given</td>
<td>68</td>
<td>51.5%</td>
</tr>
<tr>
<td>Consulting to determine testing eligibility</td>
<td>7</td>
<td>5.3%</td>
</tr>
</tbody>
</table>

Recent significant event:

- Diagnosis of cancer | 79 | 60.8% |
- Loss of significant others to disease being tested | 54 | 42.5% |

<table>
<thead>
<tr>
<th>Daily mood affected by disease worries (strongly or somewhat agree)</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Daily mood affected by disease worries (strongly or somewhat agree)</td>
<td>39</td>
<td>29.6%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Sad in past month (often or almost all the time)</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sad in past month (often or almost all the time)</td>
<td>30</td>
<td>22.7%</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Anxious in past month (often or almost all the time)</th>
<th>n</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Anxious in past month (often or almost all the time)</td>
<td>45</td>
<td>34.0%</td>
</tr>
</tbody>
</table>

Note. Due to missing data, the count in some GPRI-F categories does not add up to 132; SD = standard deviation.
the respective study samples. Nonetheless, an exploratory factor analysis was undertaken to determine whether alternative models might better fit the data.

**Exploratory Factor Analysis of the GPRI-F**

To identify the factor structure of the GPRI-F, an exploratory factor analysis was performed using a principal component analysis (PCA) algorithm with varimax rotation on all 20 items. Factor loadings for the model showed that five factors had an eigenvalue greater than 1. We next performed a scree test to determine the number of factors. The scree plot revealed a large first factor (eigenvalue = 5.1, explaining 26% of the variance), followed by three other factors explaining 9% to 12% of the variance (Table 3). The four factors had eigenvalues of 5.11, 2.35, 1.89 and 1.74, respectively, and together explained 56% of the variance.

## Table 2
**GPRI-F Score Distribution**

<table>
<thead>
<tr>
<th>Gender</th>
<th>n</th>
<th>M</th>
<th>SD</th>
<th>Minimum</th>
<th>Maximum</th>
</tr>
</thead>
<tbody>
<tr>
<td>Female</td>
<td>100</td>
<td>54.2</td>
<td>12.8</td>
<td>27</td>
<td>85</td>
</tr>
<tr>
<td>Male</td>
<td>23</td>
<td>46.2</td>
<td>12.8</td>
<td>28</td>
<td>69</td>
</tr>
<tr>
<td>Total</td>
<td>127</td>
<td>52.92</td>
<td>13.32</td>
<td>27</td>
<td>86</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Marital status</th>
<th>n</th>
<th>M</th>
<th>SD</th>
<th>Minimum</th>
<th>Maximum</th>
</tr>
</thead>
<tbody>
<tr>
<td>Married/with partner</td>
<td>91</td>
<td>48.80</td>
<td>12.29</td>
<td>27</td>
<td>80</td>
</tr>
<tr>
<td>No partner</td>
<td>30</td>
<td>51.43</td>
<td>14.07</td>
<td>23</td>
<td>85</td>
</tr>
<tr>
<td>Total</td>
<td>121</td>
<td>49.45</td>
<td>12.74</td>
<td>23</td>
<td>85</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Genetic testing status</th>
<th>n</th>
<th>M</th>
<th>SD</th>
<th>Minimum</th>
<th>Maximum</th>
</tr>
</thead>
<tbody>
<tr>
<td>Blood sample given, awaiting test results</td>
<td>15</td>
<td>48.80</td>
<td>14.93</td>
<td>27</td>
<td>85</td>
</tr>
<tr>
<td>Blood sample not yet given</td>
<td>68</td>
<td>48.75</td>
<td>12.32</td>
<td>23</td>
<td>81</td>
</tr>
<tr>
<td>Consulting to determine testing eligibility</td>
<td>7</td>
<td>45.29</td>
<td>12.78</td>
<td>27</td>
<td>62</td>
</tr>
<tr>
<td>Total</td>
<td>90</td>
<td>48.49</td>
<td>12.70</td>
<td>23</td>
<td>85</td>
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</table>

<table>
<thead>
<tr>
<th>Diagnosis of cancer</th>
<th>n</th>
<th>M</th>
<th>SD</th>
<th>Minimum</th>
<th>Maximum</th>
</tr>
</thead>
<tbody>
<tr>
<td>Yes</td>
<td>79</td>
<td>51.25</td>
<td>13.46</td>
<td>27</td>
<td>85</td>
</tr>
<tr>
<td>No</td>
<td>51</td>
<td>48.35</td>
<td>11.89</td>
<td>23</td>
<td>76</td>
</tr>
<tr>
<td>Total</td>
<td>130</td>
<td>50.12</td>
<td>12.90</td>
<td>23</td>
<td>85</td>
</tr>
</tbody>
</table>

**Reliability Assessments**

Table 3 show that the Cronbach’s α was above .74 for the first three factors and .52 for the fourth factor. These results were consistent with the item-total correlation. The inter-item correlation was over .36 for the first three factors and factor 4 had an inter-item correlation of .27. These correlations fell within the range of .20 to .40 based on the internal consistency criteria suggested by Briggs and Cheek (1986). The standardized Cronbach’s α for the 20 items of the GPRI-F was .81, which was in line with the suggested criterion of .80 for good internal consistency. The item-total correlation was above .25 for all items, showing acceptable range for an exploratory study (Table 3).
### Table 3
Exploratory Factor Analysis: GPRI-F Factor Solutions and Factor Loadings

<table>
<thead>
<tr>
<th>Item # - description</th>
<th>Loading</th>
<th>Community</th>
<th>Item-total</th>
<th>Item-mean</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Factor 1: Anticipated or experienced impact of having a disease risk or genetic mutation: 6 items, Cronbach’s α = .82, inter-item correlation = .43, variance explained = 26%</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>8. My worries about the disease affect my daily mood</td>
<td>.62</td>
<td>.64</td>
<td>.61</td>
<td>2.55</td>
</tr>
<tr>
<td>9. I worry often about my risk of getting the disease</td>
<td>.79</td>
<td>.69</td>
<td>.73</td>
<td>3.08</td>
</tr>
<tr>
<td>10. I am concerned about my risk of getting the disease</td>
<td>.74</td>
<td>.62</td>
<td>.68</td>
<td>3.21</td>
</tr>
<tr>
<td>13. I have generally felt nervous and anxious in the past month</td>
<td>.75</td>
<td>.58</td>
<td>.61</td>
<td>2.92</td>
</tr>
<tr>
<td>12. I have generally felt sad in the past month</td>
<td>.73</td>
<td>.56</td>
<td>.55</td>
<td>2.54</td>
</tr>
<tr>
<td>4A. If I learn that I have a genetic mutation, I believe that I will have more problems in my life</td>
<td>.41</td>
<td>.43</td>
<td>.37</td>
<td>2.61</td>
</tr>
<tr>
<td><strong>Factor 2: Anticipated or likely external impact from having a disease risk or genetic mutation: 6 items, Cronbach’s α = .79, inter-item correlation = .39, variance explained = 12%</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4C. If I learn that I have a genetic mutation, I believe that I will have difficulties in my family relationships</td>
<td>.67</td>
<td>.48</td>
<td>.56</td>
<td>2.31</td>
</tr>
<tr>
<td>4B. If I learn that I have a genetic mutation, I believe that I will change plans for my career</td>
<td>.77</td>
<td>.61</td>
<td>.61</td>
<td>1.92</td>
</tr>
<tr>
<td>5. The disease for which I am at risk is currently causing a significant disruption in my family life</td>
<td>.52</td>
<td>.52</td>
<td>.53</td>
<td>2.58</td>
</tr>
<tr>
<td>6. I am worried that my test result will impact on my relationship with my significant other</td>
<td>.83</td>
<td>.72</td>
<td>.69</td>
<td>2.22</td>
</tr>
<tr>
<td>7. I am worried about talking to my children about the heritable nature of the disease for which I am being tested</td>
<td>.7</td>
<td>.54</td>
<td>.52</td>
<td>2.38</td>
</tr>
<tr>
<td>11. I feel guilty that I might pass on the disease risk to my children</td>
<td>.57</td>
<td>.46</td>
<td>.42</td>
<td>2.95</td>
</tr>
</tbody>
</table>
### Factor 3: Personal history of or vulnerability to mental health issues or symptoms: 5 items, Cronbach’s α = .74, inter-item correlation = .36, variance explained = 9%

<table>
<thead>
<tr>
<th>Item</th>
<th>Factor 3 Loadings</th>
<th>Factor 4 Loadings</th>
<th>Total Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>14. I have had emotional problems in the past</td>
<td>.69</td>
<td>.55</td>
<td>.47</td>
</tr>
<tr>
<td>16. I have been diagnosed with a depressive or anxiety disorder in the past</td>
<td>.78</td>
<td>.62</td>
<td>.66</td>
</tr>
<tr>
<td>15. I have had counselling with a counselor and/or a mental health professional in the past</td>
<td>.75</td>
<td>.56</td>
<td>.55</td>
</tr>
<tr>
<td>17. I have had emotional problems that led me to have thoughts about suicide</td>
<td>.65</td>
<td>.50</td>
<td>.48</td>
</tr>
<tr>
<td>18. I am now seeing a counselor for one or more of these emotional concerns</td>
<td>.57</td>
<td>.46</td>
<td>.40</td>
</tr>
</tbody>
</table>

### Factor 4: Personal or family history of the genetic disease being tested in the clinic: 3 items, Cronbach’s α = .52, inter-item correlation = .27, variance explained = 9%

<table>
<thead>
<tr>
<th>Item</th>
<th>Factor 3 Loadings</th>
<th>Factor 4 Loadings</th>
<th>Total Score</th>
</tr>
</thead>
<tbody>
<tr>
<td>2. I have taken care of a very ill parent or another close family member</td>
<td>.73</td>
<td>.57</td>
<td>.32</td>
</tr>
<tr>
<td>3. I lost a close family member (e.g. parent/ sibling) to the disease for which I am receiving counseling/testing</td>
<td>.75</td>
<td>.59</td>
<td>.42</td>
</tr>
<tr>
<td>1. I have/had a personal diagnosis of the disease for which I am receiving counseling/testing</td>
<td>.54</td>
<td>.39</td>
<td>.27</td>
</tr>
</tbody>
</table>

*Note. Loadings < .4 were omitted.*

### Construct Validity

The correlation was assessed between the GPRI-F and three common distress measures: the IES, PR, and the SF-8 (Table 4). The correlation between PR and the GPRI-F total score was .23. The correlation between the GPRI-F total score and the IES, the IES-intrusion and the IES-avoidance was .53, .54 and .46, respectively. Regarding divergent validity, the Pearson's correlation between the SF-8 and the GPRI-F total score was -.23 and the correlation between the GPRI-F total score and the SF-8 PCS and the SF-8 MCS was -.08 and -.34, respectively.

### GPRI-F Performance

Using the IES score as the benchmark, we identified an optimal GPRI-F score cut-off to determine which individuals should receiving psychological counselling following genetic testing. An IES score less than 23 meant individuals were not at risk; otherwise, they were considered to be at risk. The cut-off value for the GPRI-F was determined by identifying the maximum Kolmogorov-Smirnov statistic (KS), which is the best differentiator for establishing an, at-risk and a no-risk group. Risk and no-risk separation analysis revealed the optimal GPRI-F cut-off to be 55 with a maximum KS = .65, where KS is a measure of distance between two populations. The KS value ranges from 0 to 1,
where 1 implies that the model is perfectly accurate in predicting default accounts or separating two populations. The cut-off score obtained for the GPRI-F was similar to the cut-off score of 50 reported by Esplen et al. (2013) for the GPRI. Next, ROC was used to assess the sensitivity and specificity of the GPRI-F using the IES as the benchmark. The area under the curve (AUC) of the GPRI-F with a score cut-off of 55 was found to be .769. Based on this cut-off, sensitivity and specificity were .667 and .762, respectively.

Also using the IES measure as the benchmark, we evaluated the MD’s assessment as to whether the patient required additional counselling. The sensitivity of the MD’s assessment was .593 and its specificity was .248.

**DISCUSSION**

This study aimed to validate a French translation of the GPRI (Esplen et al., 2013). The GPRI is a unique screening instrument that assesses the specific anticipated psychosocial impacts associated with genetic testing. It is unlike other psychological measures used to assess general anxiety or impacts.

Following translation, the GPRI-F was first subjected to linguistic validation and then its internal consistency, convergent validity and divergent validity was assessed with French-speaking patients slated for genetic counselling and testing for BRCA1/2. The psychometric properties obtained in this study support the reliability and validity of the GPRI-F with such patients. The GPRI-F total score obtained a Cronbach’s alpha of .81, which is similar to that obtained by the total score of the original English version (Esplen et al., 2013).

Whereas the GPRI was shown to have a three-factor structure, the GPRI-F showed an additional factor. The additional factor found in the GPRI-F partitioned Factor 1 of the original scale into two six-item subscales. That is, the 12 items of the original Factor 1 were split as follows: Six items reflected an anticipated or experienced impact on self of being at risk of having a disease risk or genetic mutation and six items reflected an external anticipated impact on family, friends, and work of having a disease risk or genetic mutation (see Table 3). Thus, Factor 1 of the GPRI was partitioned into Factors 1 and 2 of the GPRI-F. Factors 2 and 3 of the GPRI became Factors 3 and 4 of the GPRI-F. Why the same items loaded onto two factors (Factors 1 and 2) instead of just one (Factor 1) could have to do with symptom severity differences between the populations used in the validation studies and between the two populations in terms of the regulations and laws protecting individuals from genetic discrimination. More specifically, the GPRI-F was tested in France where laws prohibiting genetic discrimination may not be as publicized as in North America (Laedtke, 2016).
O’Neill, Rubinstein, & Vogel, 2012). However, given that the tool is meant to be used in its entirety and that its total score used and not its separate subscale scores, the fact that a four-factor scale in the GPRI-F was found instead of a three-factor structure like the GPRI has no overall impact on its use with European and French-speaking populations in general. Still, when viewed separately, each factor could provide a reason for expanding the discussion between patients and healthcare providers when assessing the impact to self and others of the perceived risk of having a disease risk or genetic mutation. In the end, Factor 1 of the GPRI-F was composed of six items (8, 9, 10, 13, 12, 4A) and explained 26% of the variance; Factor 2 was composed of six items (4C, 4B, 5, 6, 7, 11) and explained 12% of the variance; Factor 3 was composed of five items (14, 16, 15, 17, 18) and explained 9% of the variance; and Factor 4 was composed of three items (2, 3, 1) and explained 9% of the variance. These last three items (2, 3, and 1) focus on personal or family history of genetic diseases rather than on psychosocial factors like the rest of the items.

Genetic testing, especially predictive testing for AOHD, is likely to increase dramatically in the next decade. Recently, tools have been developed that can measure and predict distress in individuals receiving genetic testing (Cella et al., 2002; Chung et al., 2009). Given the number of French-speaking individuals in the world, it is essential that we have linguistically equivalent measures with adequate psychometric properties to assist genetic counsellors. The GPRI-F can help in this regard, as it was shown to identify patients that might benefit from additional psychosocial support. On the basis of sensitivity and specificity, the GPRI-F was shown to perform much better than an MD’s assessment alone. Our findings suggest that clinical implementation of the GPRI-F could provide a cost-effective and efficient means of identifying individuals that might need additional psychosocial support. Approximately 25% of individuals receiving genetic counselling are likely to experience clinically relevant levels of psychological distress after counselling (Eijzenga, Aaronson, et al., 2014). Unfortunately, the problem goes undetected in most cases. Therefore, the GPRI-F could be a very valuable tool. It can raise awareness and facilitate the management of psychosocial problems during and after genetic counselling and testing. The GPRI-F is a brief screening tool that patients found to be acceptable. It can potentially reduce healthcare costs by helping to target psychosocial services for patients who most need them, instead of offering psychosocial services to all patients undergoing genetic testing. A recent study conducted in the Netherlands made use of patient-reported outcomes to help identify psychosocial problems experienced by individuals undergoing genetic counselling for cancer with the intent to improve communication and management of psychosocial problems between the counselees and counsellors (Eijzenga et al., 2015). Using their own brief psychosocial screening tool, the Psychosocial Aspects of Hereditary Cancer (PAHC) questionnaire, organized around six problem domains, the authors assessed whether the use of this questionnaire improved awareness and management of the counselees’ psychosocial issues (Eijzenga et al., 2015). Their results showed that prior awareness of individuals’ issues in comparison to the control group did not result in an increase number of problems discussed at one-month post-testing. However, the authors note that the prevalence of problems reported at baseline, including psychosocial problems, was low (Eijzenga et al., 2015). It need to be pointed out, however, that their study is marked by certain limitations that might have prohibited proper identification of specific psychosocial problems by means of a non-validated genetic distress tool developed for the purpose of their study. Moreover, the study might have been underpowered to detect smaller yet relevant differences, as noted by the authors themselves. In light of these controversial results from the Eijzenga et al. (2015) study, the availability of the GPRI and the GPRI-F is critical as valid and reliable tools for assessing psychosocial risks in individuals undergoing genetic testing for AOHD. Its used can easily be inserted into the daily genetic counselling practices to screen for psychosocial risks.
METHODOLOGICAL LIMITATIONS

From a methodological standpoint, our study was limited by the relatively small nonprobability, homogenous sample of individuals receiving genetic testing for inherited susceptibility to cancer. Yet, given that our analysis had a high variable-to-factor ratio of 20:4 and that our sample size met the suggested criterion of at least five subjects per variable (Gorsuch, 1983), our sample size was sufficient to establish the psychometric properties of the GPRI-F. As for having a homogenous sample, in many ways, other inherited diseases can have similar psychological effects on persons obtaining genetic testing, as the literature and clinical practice suggest (Esplen et al., 2013). These include the distress of passing down a mutated gene to the next generation. As the purpose of our study was to validate a scale and not to generalize results to a larger population, this limitation is tolerable. Still, given that our study population was comprised mostly of women and that psychologic functioning tends to differ by gender, we recommend future validation of the GPRI-F with a predominantly male population at risk for AOHD. We also recommend that future studies seek to recruit participants at a ratio of 10 per item tested in order to further assess the psychometric properties of the GPRI-F and its capacity to screen for the negative psychological impact of genetic testing for other inherited diseases.

CONCLUSION

The GPRI-F has been shown to be a valid measure to screen for psychosocial risk among individuals receiving genetic testing for breast and ovarian cancer susceptibility (BRCA1/2). The instrument’s psychometric properties indicate it would be appropriate for use in clinical settings and that it could be tested for predictive power in longitudinal studies.

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Conflict of interest statement: All authors declare they have no conflict of interest.


Research involving human participants and/or animals: All procedures performed in studies involving human participants were done in accordance by the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from each study participant. Research Ethics Board approval was obtained from the PI’s primary affiliated institution, which at the time of the study was York University.

REFERENCES


Appendix A

Instrument de Dépistage Psychogénétique (GPRI-F)

Le but de ce questionnaire est d’aider à identifier les individus qui pourraient nécessiter d’un soutien additionnel au cours de leurs consultations génétiques. Les questions portent sur vos expériences de vie et sur vos attentes face à la maladie pour laquelle vous ayez avoir une consultation génétique ou un test. Veuillez noter que “maladie” fait référence à la maladie pour laquelle vous ayez assisté à une consultation génétique ou vous ayez soumis aux tests génétiques. Veuillez lire chaque question attentivement et répondez en cochant la case la plus appropriée.

<table>
<thead>
<tr>
<th>Nom :</th>
<th>Date (jour-mois-année)</th>
</tr>
</thead>
</table>

1. J’ai ou j’ai déjà eu une maladie cancrosuse pour laquelle je vais avoir une consultation génétique ou un test? (5) Oui (1) Non

2. J’ai pris soin d’un parent ou d’un autre membre de la famille très malade (ex. sœur ou frère) (5) Oui (1) Non

3. Si oui, la maladie de cette personne est liée à la raison pour laquelle je vais avoir une consultation génétique ou un test? (5) Oui (1) Non

4. J’ai perdu un membre de ma famille (ex. parent/sœur/frère) de la maladie pour laquelle je vais avoir une consultation (5) Oui (1) Non

5. Veuillez indiquer que(s) est/sont le(s) membre(s) de votre famille qui est/sont décédé(s) (0) un parent (0) une sœur (0) un frère (0) Autre (précisez) ____________________________

<table>
<thead>
<tr>
<th>Si j’apprends que je suis porteur(e) d’une mutation génétique, je crois que:</th>
<th>Absolument d’accord</th>
<th>Assez d’accord</th>
<th>Neu en accord ni en désaccord</th>
<th>Presque jamais d’accord</th>
<th>Non concerné(e)</th>
</tr>
</thead>
<tbody>
<tr>
<td>a. J’aurais davantage de problèmes dans ma vie</td>
<td>5</td>
<td>4</td>
<td>3</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>b. Je changerais de plan de carrière/profession</td>
<td>5</td>
<td>4</td>
<td>3</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>c. J’aurais des difficultés dans mes relations familiales</td>
<td>5</td>
<td>4</td>
<td>3</td>
<td>2</td>
<td>1</td>
</tr>
</tbody>
</table>

6. Si je perds une journée de travail en raison de cette maladie, je suis sûr(e) que l’assurance maladie me remboursera (5) Oui (1) Non

7. Si je perds une journée de travail en raison de cette maladie, je suis sûr(e) que l’assurance maladie me remboursera (5) Oui (1) Non

8. Si je perds une journée de travail en raison de cette maladie, je suis sûr(e) que l’assurance maladie me remboursera (5) Oui (1) Non

9. Je suis préoccupé(e) par mon risque de développer la maladie | 5 | 4 | 3 | 2 | 1 | 0 |

10. Je me sens coupable de pouvoir transmettre la maladie à mes enfants | 5 | 4 | 3 | 2 | 1 | 0 |

11. Au cours du mois dernier, je me suis généralement senti(e) triste | Presque toujours | Souvent | Parfois | Presque jamais | Pas du tout |
| 5 | 4 | 3 | 2 | 1 |

12. Au cours du mois dernier, je me suis généralement senti(e) triste | 5 | 4 | 3 | 2 | 1 |

13. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

14. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

15. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

16. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

17. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

18. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

19. J’ai eu des problèmes d’ordre émotionnel dans le passé | (5) Oui | (1) Non |

POUR USAGE EXTERNE

Instruction pour calculer le pointage : somme additionner tous les items de 1 à 19 et mettre le total ici _________________

Si le total est 55 ou plus, et si l’item 19 est oui, donc il est recommandé de demander une consultation ou psychosociale.