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Distress facing increased genetic risk of cancer: The role of social support and emotional suppression☆

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ABSTRACT

Objectives: Healthy individuals from hereditary cancer families undergoing genetic testing for cancer susceptibility (GTC) report more distress when they perceive their social support as low and suppress their emotions. This study aimed to explore how suppressing emotions and perceiving others as unsupportive are related with cancer-risk distress.

Methods: We performed a regression-based mediation analysis to assess if expressive suppression mediates or is mediated by perceived social support in the relation with cancer-risk distress. Participants were 125 healthy adults aged over 18 ($M = 36.07$, $SD = 12.86$), mostly female (72,4%), who undergone GTC to assess the presence of hereditary breast and ovarian cancer or Lynch syndromes.

Results: Controlling for age and gender, we found a moderate size indirect effect of social support on cancer-risk distress through expressive suppression ($\beta = -0.095$) and a direct effect of expressive suppression on cancer-risk distress.

Conclusions: When healthy individuals from hereditary cancer families perceive their social network as less responsive, they tend to not express their emotions, which relates to increased distress facing GTC.

Practice implications: Practitioners may assess cancer-risk related distress before the GTC and offer distressed individuals interventions focused on changing emotion regulation strategies in a safe group context.

1. Introduction

Hereditary cancer syndromes are genetic conditions caused by inherited pathogenic variants in specific tumor-suppressor genes, which increase the lifetime risk of developing some kinds of cancer [1]. Two of the most prevalent hereditary cancer syndromes are hereditary breast and ovarian cancer (HBOC), and Lynch syndrome (LS) [1,2]. Women identified with HBOC syndrome have a cumulative lifetime breast cancer risk of over 60% and an ovarian cancer risk of over 40%, which is, respectively, 5 and 25 times higher than the general population [3,4].

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On the other hand, those identified with LS have a lifetime a colorectal cancer risk ranging from 20% to 80%, up to 11 times higher than overall population [2,5]. Plus, women with LS may have to deal with an increased lifetime risk of endometrial and ovarian cancer [2].

Hereditary cancer syndromes can be identified in at-risk individuals – individuals from hereditary cancer families at risk of being carriers, who have not yet been affected by cancer nor subjected to genetic testing - by means of genetic testing for cancer susceptibility (GTC). In the event of a positive result in the GTC, it is possible to monitor cancer risk and prevent cancer onset through personalized prevention programs [6], thus significantly reducing cancer morbidity and mortality [7]. However, undergoing a GTC may also pose significant psychological difficulties for at-risk individuals. Heading to GTC, many at-risk individuals have already experienced the loss of relatives due to hereditary cancer and/or are living through their relatives' negative experiences with cancer prevention measures [8–10]. This proximity and the hereditary nature of the disease may elicit at-risk individuals to feel future losses are inevitable and anticipate they will be next [11,12]. Moreover, at-risk individuals testing positive in the GTC will have to decide between risking to live with increased cancer risk without trying to prevent cancer onset, enhance cancer-screening frequency or undergo organ-removal prophylactic surgeries, which often are the most recommended risk management procedure [13,14]. In this context, when undertaking the GTC some at-risk individuals report increased cancer-related distress, anxiety, worry, depression, and anger [15–17]. Decisional conflicts about taking the GTC and feelings of anticipation of loss may also occur [18], eventually affecting adherence to genetic testing. The level of cancer-related distress before undergoing GTC seems to play a key role in the psychological adjustment to hereditary cancer risk. Previous research found that pre-test cancer-related distress is the main predictor of the long-term cancer related distress independently of the test result [15,19], with important health and quality of life costs. Cancer related distress may be present up to two years after GTC results [20], with negative repercussions for quality of life [21], relationships [22–24], and risk management behavior [25].

One of the most reported variables associated with pre-test cancer related distress is perceived social support (e.g., Lapointe et al., [22]) - the perception of having help from others to meet emotional, informational, and instrumental needs and to manage everyday life stressful situations [26,27]. Social support has long been considered a protective factor against depressive symptoms, particularly in response to stressful events [28]. Also, in the specific context of genetic counseling, previous studies found that fewer sources of social support and less satisfaction with the support network were related with higher pre-test levels of distress and depression in colorectal cancer patients undergoing GTC [29,30]. Moreover, perceived availability of social support was positively associated with health-related quality of life in individuals undergoing genetic counselling for both HBOC and LS [31]. In addition, perceiving others as supportive was found to be associated with more open communication within the family and higher self-esteem which were, in turn, associated with less cancer-risk related distress in women at risk of HBOC [32]. Collectively, these findings point to a clear association of social support with cancer related distress, however the underlying mechanisms by which this association occurs have been less studied.

One aspect that has not yet been explored in the association between perceived social support and cancer-risk related distress are emotion regulation processes. Yet, the way individuals deal with their emotions may play an important role in this association. For example, previous research found that effective support stimulates changes in emotion regulation strategies that help to reduce depressive symptoms [28]. One specific emotion regulation process by which social support may relate to pre-test cancer-risk related distress is expressive suppression (ES) - purposefully inhibiting or reducing the behavioral expression of emotions [33]. ES was found to be related with higher levels of general and cancer specific distress in candidates of HBOC susceptibility genetic testing [34]. Moreover, a recent study found that people who have the perception that they will not receive social support tend to suppress their emotions, resulting in higher levels of anxiety before surgery [35]. These preliminary findings point to the hypothesis that ES may have a mediating role in the relation of perceived social support and perceived distress at GCT.

On the other hand, the lack of social support may also mediate the relation between ES and cancer-risk distress. Past research found that people who normally suppress their emotions were more likely to present poorer social support than individuals who use cognitive reappraisal (changing how they think about a given stressor) as a main emotion regulation strategy [33]. Also in breast cancer patients, the relation between difficulties in expressing emotions and distress was found to be mediated by perceived social support [36]. In other words, those who tend to inhibit the expression of their emotions to others may fail to activate the support of their social network, thus ending up perceiving less social support (than individuals who express their emotions more openly), which increases distress [37,38]. The nature of these relationships is still not completely understood due to the scarcity of research and previous studies not assessing simultaneously if expressive suppression and perceived social support are directly or indirectly associated with cancer-risk distress. In fact, both hypotheses are plausible, but they have different impacts on how to organize care to at-risk individuals. If social support mediates ES, screening and intervention should focus on emotional regulation strategies of the at-risk individuals. By contrast, if suppressing emotions mediates social support, then the focus of care should be the evaluation and strengthening of the social network of at-risk individuals.

To our knowledge, this is the first study focusing on the mechanisms by which perceived social support and expressive suppression relate to cancer-risk specific distress prior to undergoing GTC. Specifically, drawing from the scarce literature we will evaluate whether (1) perceived social support is indirectly associated with cancer-risk specific distress through expressive suppression, or (2) expressive suppression is indirectly associated with cancer-risk specific distress through perceived social support.

2. Method

2.1. Participants and procedure

Based on effect sizes described in prior research [32,34], a power analysis was conducted using MedPower [39] to determine adequate sample size. Thus, for a power of $\beta = 0.80$, with α fixed at 0.05 and considering an effect size of 0.25 for both direct and indirect effects, we estimated that 156 subjects would be required. However, due to the COVID-19 pandemic we were only able to collect data from 125 individuals. Participants were at-risk individuals unaffected by cancer aged over 18 ($M = 36.07$, $SD = 12.86$), mostly female (72,4%) (Table 1) enrolled for Genetic susceptibility test for HBOC syndrome (66.7%; n = 83) and (2) LS (33.3%; n = 42), at an oncologic hospital in Portugal. Data were collected in routine clinical settings between November 2018

Table 1
Sociodemographic and clinical characteristics of at-risk individuals.

	Women ($n = 91$)	Men ($n = 34$)	Total ($N = 125$)
Sociodemographic variables	*M (SD)*	*M (SD)*	*M (SD)*
Age in years	36.18 (12.94)	36.03 (12.54)	36.14 (12.78)
Years of formal education	12.47 (3.23)	12.61 (3.55)	12.50 (3.30)
	n (%)	*n (%)*	*n (%)*
Having a partner	52 (57.14)	21 (61.76)	73 (58.4)
Having children	55 (60.44)	18 (52.94)	73 (58.4)
Type of familial syndrome			
HBOC	65 (71.43)	18 (52.94)	83 (66.4)
Lynch Syndrome	26 (28.57)	16 (47.06)	42 (33.6)

Note. n = Frequency; M = Mean; SD = Standard Deviation; HBOC = Hereditary breast and ovarian cancer.

and May 2021. Participants were recruited after one genetic counselling session with a medical geneticist. This genetic counselling session aimed to provide genetic information to patients and assess criteria for genetic testing, but it did not include a psychological evaluation. During this session, the medical geneticist also assessed patients' literacy and understanding of the genetic testing context through clinical interview. Participants who fulfilled criteria, decided to undergo genetic testing, and showed sufficient literacy were invited to participate in a project about their thoughts and feelings about hereditary cancer risk as well as family relations. This invitation was first performed by their medical geneticist and then by the author, who presented the study in greater detail. Participants were excluded if they were under 18 or did not have sufficient literacy to understand what was written on the self-report questionnaires. Of the 176 at-risk individuals who were invited, 11 (9.32%) refused to participate and of the 60 at-risk individuals who took the questionnaires home, 36 (60%) did not return them. In addition, 14 participants were recruited after the start of the COVID-19 pandemic. This work is part of an ongoing larger project (project TOGETHER) that aims to study the psychosocial adjustment process of individuals unaffected by cancer who undergo GTC and their families, approved by the hospital's ethical board (Doc. CES-IPOP 04_2017).

2.2. Instruments

2.2.1. Impact of Events Scale

The Impact of Events Scale (IES [40]) is a 15-item 4-point Likert scale (from 0 = "*never*" to 4 = "*almost always*") used to measure distress provoked by a stressor or life event. It comprises two domains: Intrusion, which refers to intrusive thoughts and feelings about the event or stressor (7 items); and Avoidance, which relates to patterns of avoidance in terms of thoughts, feelings, and behaviors (8 items). Summed together, the two dimensions form the total score of distress. The IES has been frequently used with populations undergoing genetic testing and has shown good psychometric properties in the context of hereditary cancer [41]. For this study, we adapted this instrument to measure cancer-risk specific distress (Example item: *"I thought about cancer or cancer-risk when I did not want to"*). In our sample the IES showed excellent internal consistency (α = 0.91, ω = 0.90).

2.3. Multidimensional Scale of Perceived Social Support

The Multidimensional Scale of Perceived Social Support (MSPSS [27]; Portuguese version by Carvalho et al. [42]) comprises 12 items on a 6-point Likert scale (from 1 = "*Strongly Disagree*" to 6 = "*Strongly Agree")* divided in 3 subscales (4 items each) tapping three different sources of social support: significant other (e.g., *There is a special person in my life who cares about my feelings*"), family (e.g., *"I get the emotional help and support I need from my family"*), and friends (e.g., *"My friends really try to help me"*). The mean of all items forms the perceived social support total score, which was the variable used in this study. In the current study, the MPSS demonstrated good internal consistency (α = 0.88, ω = 0.83).

2.4. Emotion Regulation Questionnaire

The Emotion Regulation Questionnaire (ERQ [38]; Portuguese version by Machado Vaz [43]) is a 10-item Likert-Scale (from 1 = "*Strongly Disagree"* to *7* = "*Strongly Agree*") that measures the use of two emotion regulation strategies: cognitive reappraisal (6 items) and expressive suppression (4 items). For this study we used the subscale expressive suppression (e.g., *"I keep my emotions to myself"*) (α = 0.83, ω = 0.83), which measures the extent participants inhibit the expression of both pleasant and unpleasant emotions.

2.5. Data analysis

The analyses were conducted with SPSS version 27. Data were normally distributed except for perceived social support, which was negatively skewed (*Sk* = −1.85 (SE = 0.22), *Ku* = 4.54 (SE = 0.43)). Missing values were missing completely at random according to Little's test ($\chi^2(195) = 226.95$, $p = 0.058$). Because of this, we proceeded with the analysis of missing data patterns and excluded two cases. On the first excluded case, all items from IES questionnaire were missing and on the second case 75% of the items pertaining to the ERQ questionnaire were also not reported. Remaining cases (n = 5) with missing values represented 0.2% of all data and presented no notable patterns, so we also excluded them from further analysis. Since the mediation hypothesis was tested with the ordinary least squares (OLS) regression model provided in the model 4 of the SPSS PROCESS macro by Hayes [44], we verified OLS regression assumptions. Uncorrelatedness of residuals was present (Durbin-Watson = 1.97), and multicollinearity was absent (ES, Tolerance = 0.98, VIF = 1.02, Perceived Social Support, Tolerance = 0.98, VIF = 1.02). A histogram of the dependent variable's residuals showed a normal distribution curve, indicating normality of errors. Also, partial regression plots between independent and dependent variables showed no evidence of a nonlinear relationship between the variables, so linearity was assumed. To assess homoscedasticity, a scatterplot of residuals was plotted, however it was inconclusive in rejecting a pattern. Because of this, we performed the Breusch-Pagan Test (F(5119) = 2.26, $p = 0.053$) and the White test (F(2122) = 2.32, $p = 0.102$), both of which showed homoscedasticity could be assumed.

We detected multivariate outliers using the Mahalanobis' distance (n = 2). After examination of these outliers, they were considered model fit outliers because the exclusion of those rendered different results [45]. Moreover, upon closer inspection, no inconsistent patterns of responses were found. Still, we decided to report the results with and without outliers as per best-practices recommended by Aguinis et al. [45]. Confidence interval (at 95%) to test for direct and indirect effects was plotted with the bootstrapping method (5000 samples).

To explore the relationship between perceived social support, expressive suppression and cancer-risk related distress, two mediation models were tested. Model 1 included perceived social support as an independent variable, expressive suppression as a mediator variable, and cancer-risk specific distress as a dependent variable. Model 2 was composed by expressive suppression as the independent variable, perceived social support as the mediator variable, and cancer-risk related distress as the dependent variable. In both models, we used age and gender as covariates. We included these covariates because past research has consistently shown that age and gender may be associated with cancer-related distress [46,47].

3. Results

Intercorrelations between all continuous variables and descriptive statistics can be found in Table 2. Mean values of expressive suppression, perceived social support, and cancer-risk specific distress were 3.69, 5.42 and 17.85, respectively. Since the independent variable perceived social support presents a non-normal distribution, we also reported the median and interquartile ranges as these are better indicators for non-parametric distributions [48]. As so, the social support scale median was 5.7, while the family and significant other subscales' median were both 6, and the friends subscale median was 5.25. Moreover, the interquartile range of the significant other subscale is lower (0.25) than the family (1.00) and the friends (1.25) subscales, which suggests that, in general, participants rated their significant other as the main source of social support.

Spearman correlations between all the study continuous variables showed a significant correlation ($r_s = -0.26$, $p = 0.003$) between perceived social support and expressive suppression, as well as between expressive suppression and cancer-risk specific distress ($r_s = 0.40$, $p <$

Table 2
Intercorrelations[†] and descriptive statistics of study's continuous variables.

Variable	1	2	**2.1**	**2.2**	**2.3**	3	4	Range	*M*	*SD*	***Med***	***IQR***
1. Age	–	-0.057	**-0.163***	**-0.012**	**0.003**	-0.12	-0.004	NA	36.14	12.78	**35.00**	**23.00**
2. Social Support (**PSS[‡]**)	-0.057	–	**0.768*****	**0.700*****	**0.615*****	-0.259***	-0.162*	1–6	5.42	.67	**5.67**	**.92**
2.1 PSS From Friends	**-0.163***	**0.768*****	–	**0.365*****	**0.319*****	**-0.233*****	**-0.167***	**1–6**	**5.18**	**1.00**	**5.25**	**1.25**
2.2. PSS From Family	**-0.012**	**0.700*****	**0.365*****	–	**0.388*****	**-0.197****	**-0.057**	**1–6**	**5.47**	**.87**	**6.00**	**1.00**
2.3 PSS From Significant Other	**.0003**	**0.615*****	**0.319*****	**0.388*****	–	**-0.093**	**-0.104**	**1–6**	**5.60**	**.96**	**6.00**	**.25**
3. Expressive Suppression	-0.12	-0.259***	**-.0233*****	**-0.197****	**-0.093**	–	0.398***	1–7	3.69	1.47	**3.75**	**2.50**
4. Cancer-risk specific distress	-0.004	-0.162*	**-0.167***	**-0.057**	**-0.104**	0.398***	–	0–60	17.85	11.38	**18.00**	**17.50**

Note. [†] = Spearman rank correlation was used due to violation of normality by the perceived social support variable; [‡] = Perceived Social Support; *M* = Mean; *SD* = Standard Deviation; *Med* = Median; *IQR* = Interquartile range NA = Not applicable; * $p < .1$, ** $p < .05$, *** $p < .01$.

0.001). However, we could not find a significant correlation between perceived social support and cancer-risk specific distress ($r_s = -0.16$, $p = 0.072$). Despite this, we advanced with the mediation analysis, based on the argument that a significant correlation between the independent and dependent variables is not required to perform mediation analysis [49].

We tested two models in our analysis. Model 1 included five variables: perceived social support as the independent variable, cancer-risk specific distress as the dependent variable, and expressive suppression as the mediator variable. As depicted in Fig. 1, the standardized regression coefficient between perceived social support and expressive suppression was statistically significant ($\beta = -0.23$, $t(121) = -2.57$, $p = 0.011$, CI [−0.878, −0.114]), as well as between expressive suppression and cancer-risk specific distress ($\beta = 0.42$, $t(120) = 4.97$, $p < 0.001$, CI [0.129, 0.300]). The total effect of perceived social support on cancer-risk specific distress was also significant ($\beta = -0.18$, $t(121) = -2.00$, $p = 0.047$, CI [− 0.399, − 0.002]), but this effect did not remain significant when the covariates were added ($p = 0.122$). Likewise, the direct effect of perceived social support on cancer-risk specific distress was not significant ($\beta = -0.08$, (t(120) = −1.00, p = 0.318 CI [− 0.281, 0.919]). Importantly, the completely standardized indirect effect of perceived social support on distress was significant ((−0.23) (0.42) = −0.095, CI [−0.184, −0.019]), as well as the unstandardized effect ((−0.50) (0.21) = −0.11, CI [−0.225, −0.021]). These results mean that for each increase in standard deviation in perceived social support, cancer-risk distress decreases for 0.095 standard deviations, and when perceived social support increases for one total value, cancer risk-distress decreases 0.11 values.

To test if expressive suppression is mediated by perceived social support in the relation with cancer risk specific distress, we tested the inverted model. As such, model 2 was composed of expressive suppression as the independent variable, perceived social support as the mediator variable and cancer-risk specific distress as the dependent variable. Results showed a direct effect of expressive suppression on cancer-risk specific distress ($\beta = 0.42$, $t(120) = 4.97$, $p < 0.001$, CI [0.129, 0.300]). However, no statistically significant indirect effect of expressive suppression on cancer-risk specific distress through perceived social support was found ((−0.23) (0.42) = −0.02, CI [−0.018, 0.075]).

To check if outliers would impact the results significantly, we also tested both models without the outliers. In model 1, the only significant difference we encountered was regarding the total effect of social support on cancer-risk specific distress, that became not statistically significant ($\beta = -0.143$, $t(119) = -1.57$, $p = .119$, CI [−0.430, 0.049]). In model 2, no significant differences occurred. Results with and without outliers are displayed on Table 3.

There was a significant indirect effect between perceived social support and cancer-risk specific distress through expressive suppression,

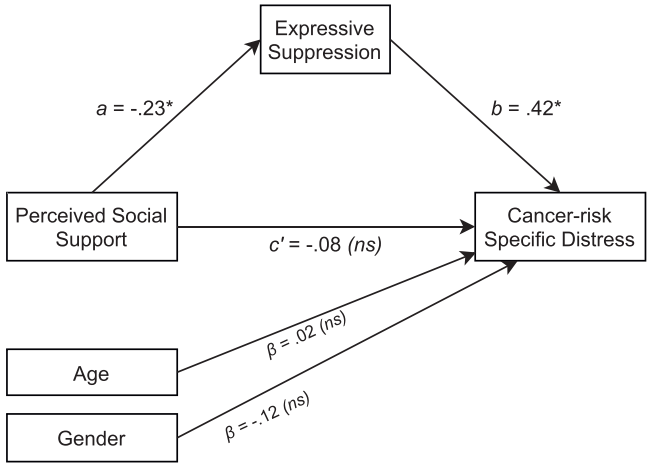

Fig. 1. *Final Mediation Model:* A mediation model of expressive suppression mediating the relation of perceived social support and cancer-risk distress, with age and gender as covariates. *a* = standardized regression coefficient of perceived social support on expressive suppression; *b* = standardized regression coefficient of expressive suppression on cancer-risk specific distress; *c'* = standardized direct effect of perceived social support on cancer-risk specific distress; * = effect is significant; *ns* = effect is not significant; β = standardized regression coefficient of covariates.

Table 3
Results of the total effect regression model**s** and the tested mediation models with and without multivariate outliers.

	With multivariate outliers			Without multivariate outliers		
	R^2	*p-value*	*Sig.*	R^2	*p-value*	*Sig.*
Total effect model 1[†]	0.047	0.122	No	0.033	0.266	No
Total effect model 2[Δ]	0.203	< 0.001	Yes	0.191	< 0.001	Yes
Mediation model 1[‡]	*β*	*C.I.*	*Sig.*	*β*	*C.I.*	*Sig.*
Total Effect of Social Support	-0.178	[−0.399, −0.002]	Yes	-0.143	[−0.430, 0.049]	No
Direct Effect of Social Support	-0.083	[−0.281, 0.092]	No	-0.046	[−0.288, 0.165]	No
Indirect Effect of Social Support[¶]	-0.095	[−0.178, −0.016]	Yes	-0.097	[−0.185, −0.022]	Yes
Mediation model 2[§]						
Total Effect of Expressive Suppression	0.438	[0.141, 0.308]	Yes	.425	[0.135, 0.304]	Yes
Direct Effect of Expressive Suppression	0.418	[0.129, 0.300]	Yes	.415	[0.126, 0.301]	Yes
Indirect Effect of Expressive Suppression[¶]	0.019	[−0.018, 0.074]	No	.011	[−0.034, 0.063]	No

Note. [†] = Total effect of Social Support on Cancer-risk Specific Distress while considering Sex and age as covariates; [Δ] = Total effect of Expressive Suppression on Cancer-risk Specific Distress while considering Sex and age as covariates; [‡] = Perceived Social Support effect on Cancer-risk Specific Distress through Expressive Suppression; [§] = Expressive Suppression effect on Cancer-risk Specific Distress through Perceived Social Support; [¶] = Completely standardized indirect effect; R^2 = Coefficient of determination; *β* = Standardized regression coefficient; *C.I.* = Confidence Interval at 95%; *Sig.* = Presence of a statistically significant effect.

which means that expressive suppression plays a mediating role in this association. To finalize, we conducted a power analysis using the joint significance test provided by MedPower [39]. Given our sample and effect sizes, we estimate the probability of having found a true indirect association of social support with distress through expressive suppression to be 74%, both with and without outliers. Moreover, according to Kenny [39] categorization, this effect can be considered of moderate size.

4. Discussion and conclusion

To our knowledge, this was the first study that aimed to explore the mechanisms underpinning the association between perceived social support, emotion regulation and cancer-risk specific distress in healthy at-risk individuals from families with HBOC and Lynch syndrome. Results confirm the mediating role of ES, i.e., less perceived social support relates to an increase in expressive suppression, which is associated with higher cancer-risk specific distress. Moreover, we could also find a direct effect of ES on cancer-risk distress, in the absence of an indirect effect of ES on cancer-risk distress through perceived social support. This means that ES is uniquely and atemporally associated with cancer-risk distress [50]. Although correlational, these findings fall in line with Gross's theory of emotional suppression, which associates suppression with negative affective outcomes [33].

Findings also show that the main drivers of perceived social support are the significant others and the family. These results reinforce previous research [51,52,53] that suggests that the family is the main source of emotional and informational support and that communal coping processes within the family occur to facilitate adaptation to hereditary cancer syndromes. Moreover, results showed that perceived social support is atemporally associated with cancer-risk distress through expressive suppression. This means that at-risk individuals who feel less supported by their support network, tend to also inhibit the expression of their emotions, and that inhibition is related to cancer-risk specific distress at the time of GTC. This finding is in line with previous research by Marroquín [28], who suggests that support from others allows individuals the opportunity to change how they regulate their emotions, either by contributing to shifting attention from stressors or to changing how they think about them. Based on our results we add that support from others, particularly significant others and family, may also be associated with less inhibiting of emotional expression as a way to regulate emotions. Our results also add to the findings from Den Heijer et al. [51]. These authors reported that open communication about hereditary cancer within the family mediates the effects of perceived social support on distress experienced by at-risk individuals from families with HBOC. Our conclusions reinforce this, adding that it seems to be important not only to communicate openly, but also to promote an interpersonal environment where at-risk individuals feel that they have someone who will support them and with whom they can share their emotions.

4.1. Practice implications

In terms of implications for practice, our findings emphasize the importance for clinicians to be aware of how at-risk individuals feel about the availability and capacity of their social network to support them as they need it. In addition, it is important to gauge whether they feel comfortable expressing their emotions with the members of this network. Research has shown that at-risk individuals do not always feel like they are able to express their worries despite having a seemingly good support network due to protective buffering or survivor guilt behaviors [52]. Therefore, at-risk individuals with lower perceived social support that want to express their emotions but feel inhibited may be in a higher risk of distress if they do not find a place and a time to express their more painful emotions. It is also important to note that the timing of providing social support is crucial for individuals to perceive it as effective, as sometimes support is provided at moments where it is not needed and that can be unwelcomed [54,55]. Practitioners should account for this and stimulate at-risk individuals to effectively communicate their needs regarding when and how they wish to be supported.

Supportive expressive groups where health professionals facilitate the expression of emoticons within a safe group environment could prove useful in improving both emotional expression and perceptions of social support. Supportive Expressive groups have been shown to be effective in reducing intrusion and depression, as well as feasible in the context of women with HBOC syndrome [56]. Also, multifamily discussion groups were shown to help strengthen intra and interfamilial support, by providing a space to share experiences with other hereditary cancer families, thus expanding support network and improving communication about cancer risk within families [57]. One shortcoming in the delivery of these interventions is that they focus mainly on individuals that are already identified as pathogenic variant carriers. However, pre-test distress is the strongest predictor of post-test distress [15,19]. Thus, health professionals could monitor distress and perceived support since the first genetic counseling consultation and offer distressed individuals reporting poor social support the opportunity to integrate one of these groups even before results are known.

4.2. Study limitations

From a theoretical point of view, the lack of a significant total effect between perceived social support and cancer-risk specific distress when controlled for age and gender was unexpected. From a methodological perspective, a few explanations may be advanced. The sample may not have been large enough to reach statistical significance (type II error). Also, the low variability in perceived social support (distribution was negatively skewed) in our sample might be affecting the results. It is plausible that participants supported by others were more willing to enter the study. A third explanation could be related to the specific way we are measuring perceived social support. As Den Heijer et al. [51] advanced, the MSPSS may be too broad to capture specific nuances of social support in the context of hereditary cancer. Perhaps a more specific instrument would have yielded different results.

We have a high percentage of invited participants that did not return the questionnaires and due to the COVID-19 pandemic our sample size is not large enough to confidently exclude the possibility of type I and type II errors, given that statistical power did not reach 80%. Due to a small sample, we also could not include the type of syndrome in our mediation models and due to ethical and data privacy reasons, we could not collect data about family health history. It would be interesting if future research could explore whether the role of expressive suppression and perceived social support remains the same across different syndromes regardless of family history. Finally, the atemporal nature of the study design does not allow for causal relationships to be inferred [50]. Future studies should investigate if causal mediation is occurring through longitudinal study designs.

4.3. Conclusions

This study sheds light on one of the possible mechanisms behind the association of perceived social support and cancer-risk specific distress in at-risk individuals to HBOC and Lynch Syndrome. Our results suggest that this relation is mediated by expressive suppression. At-risk individuals who have lower levels of perceived social support may tend to suppress their emotional expression, and this may result in higher cancer-risk specific distress.

Ethics approval

This work was approved by the IPO-PORTO hospital's ethical board (Doc. CES-IPOP 04_2017) and all procedures used in this study adhere to the tenets of the Declaration of Helsinki.

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CRediT authorship contribution statement

Célia Sales, **Paula Mena Matos** and **Eunice Silva** conceived the study. Data collection and curation was performed by **Pedro Gomes** assisted by **João Silva**. The first draft was written by **Pedro Gomes**. **Pedro Gomes** performed the data analysis with the assistance of **Paula Mena Matos**. **Célia Sales**, **Paula Mena Matos**, **Eunice Silva**, **João Silva** and **Eliana Silva** reviewed the manuscript, provided suggestions and assisted in writing the discussion. **Pedro Gomes** incorporated co-author's suggestions and wrote the final version of the manuscript.

Declaration of Competing Interests

The authors declare no conflict of interests in the elaboration of this work.

Data availability statement

Research data are not shared due to ethical reasons.

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