Psycho-Oncology
Psycho-Oncology (2012)
Published online in Wiley Online Library (wileyonlinelibrary.com). **DOI**: 10.1002/pon.3072

A self-help coping intervention can reduce anxiety and avoidant health behaviours whilst waiting for cancer genetic risk information: results of a phase III randomised trial

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Received: 25 May 2011
Revised: 15 February 2012
Accepted: 24 February 2012

Abstract

***Objective*: The objective of this study is to evaluate the effectiveness of a self-help coping intervention in reducing intrusive negative thoughts while waiting for cancer genetic risk information.**

***Methods*: Between August 2007 and November 2008, 1958 new referrals for cancer genetic risk assessment were invited to participate in a randomised trial. The control group received standard information. The intervention group received this information plus a written self-help coping leaflet. The primary outcome measure was the intrusion subscale of the Impact of Event Scale.**

***Results*: The intervention significantly reduced intrusive thoughts during the waiting period in those reporting moderate baseline levels of intrusion ($p = 0.03$). Following risk provision, those in the intervention group reporting low and moderate intrusive worries at baseline reported less intrusive thoughts than those in the control group ($p = 0.04$ and $p = 0.03$, respectively). The intervention had no adverse impact in the sample as a whole. Participants in the intervention group with high baseline avoidance and negative affect scores were significantly more likely to remain in the study than those in the control group ($p = 0.05$ and $p = 0.004$).**

***Conclusions*: Findings that the intervention both reduced distress in those with moderate levels of distress and had no adverse effects following notification of cancer genetic risk suggest that this simple intervention can be implemented across a range of oncology settings involving periods of waiting and uncertainty. The intervention may also reduce the number of individuals dropping out of cancer genetic risk assessment or screening. However, those with clinically high levels of psychological distress are likely to require a more intensive psychological intervention.**
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Introduction

From the point of referral into cancer genetics services in the UK, individuals may be faced with a waiting period of several weeks or months before they learn their genetic risk of developing cancer, often with minimal contact from genetic specialists. During and immediately after this time, around one quarter of those involved report high levels of worry and distress [1–4]. This is not only inherently undesirable but also can affect the comprehension of risk information and influence decisions related to surveillance and other clinical options [5–7]. There is also an evidence to suggest that those with high levels of distress may decline or drop out of such assessments [8,9], losing access to specialist support. This paper reports the results of a definitive randomised controlled trial of a written self-help coping intervention, designed to help individuals control unwanted intrusive worries while waiting for cancer genetic risk information. This theory-based intervention was guided by coping theory and empirical evidence suggesting that the structured use of distraction in combination with controlled exposure to any worries or concerns may be an effective coping strategy whilst waiting for genetic risk information. Our intervention was previously found to be acceptable and potentially effective in developmental work including a pilot trial. This paper reports the definitive effectiveness trial of the intervention, which to our knowledge is the first type of intervention to specifically target the early stages of the assessment process rather than following the provision of risk information as in previous studies [10–14].

The transactional model of stress and coping [15] and the related intervention of coping effectiveness training [16] emphasise the importance of matching coping strategies to the specific demands of the situation. According to this approach, when faced with short-term situations involving waiting and uncertainty with limited options for problem-focused coping, the use of distraction can be an effective coping strategy [17,18]. Being instructed to engage in specific cognitive and/or behavioural distraction techniques has been shown to be more effective at controlling intrusive thoughts and physiological distress than receiving general instructions to distract from the stressful situation [19,20]. In addition, encouraging individuals to plan when and how they are going to engage in specific distraction techniques has been found to increase the likelihood of them engaging in that behaviour [21]. However, one of the most documented concerns about distraction is that attempts to suppress intrusive thoughts about a stressful event can subsequently result in more intrusive

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thoughts in the period after suppression than would normally the so-called 'rebound effect' [22].

One way of reducing this potential 'rebound effect' in the present context is to encourage individuals to actively consider issues of threat or risk prior to being informed of their level of risk. This should facilitate adaptation or habituation to the idea of risk [23–25] and has been shown to have psychological and physical benefits [26–28]. In an attempt to gain the advantages associated with both distraction and active processing of risk, the intervention encouraged individuals to restrict thinking about issues related to their risk to a set time each day and to use active distraction techniques to control intrusive thoughts at all other times. However, as this combination of distraction and focus on worries has not been previously reported, it is not clear whether this optimum outcome could be achieved.

Phases I and II developmental work [29,30] informed by the UK Medical Research Council's framework for the development of complex interventions [31] suggested that the intervention was acceptable and potentially effective. However, the pilot trial was not fully powered and was too small to explore the impact of the intervention following risk provision, including assessment of a potential rebound effect. It was also restricted to women at risk of breast and/or ovarian cancer. The aims of Phase III fully powered trial were therefore to (i) confirm previous findings that the intervention helped reduce distress whilst waiting for genetic risk information in a larger sample and (ii) explore the longer term psychological impact of the intervention following the provision of genetic risk information, in particular, whether the intervention resulted in greater distress following risk provision for those that had used the distraction techniques whilst waiting for risk information.

Methods

Participants

Eligible participants were male and female referrals into the Cancer Genetics Service for Wales (CGSW) between August 2007 and November 2008 for assessment of their cancer genetic risk (average, moderate or high). CGSW provides appropriate information and support to individuals concerned about their family history of cancer with the majority of referrals being for breast and/or ovarian cancer, followed by colorectal cancer. Upon referral, the first step in the assessment process is the completion of a detailed family history questionnaire that is then verified by specialist professionals within CGSW to estimate an individual's risk of developing cancer. This assessment process usually takes at least 4 weeks and is often longer before individuals receive their risk assessment and are offered appropriate support and advice. In the case of CGSW referrals for breast/ovarian cancer risk for example, the majority of individuals are identified through a letter as being at either average risk (<15% lifetime risk) or moderate risk (15%–24% lifetime risk). Only those at high risk would usually attend a genetic clinic to explore the option of predictive genetic testing. During the trial, all new referrals into the three regional bases of CGSW in South East Wales, South West Wales and North Wales were all eligible for participation. The only exclusion criteria were any urgent referrals or rare cancer syndromes identified upon referral by the clinical team.

Trial design

The trial received full National Health Service (NHS) ethical approval from the Multisite Research Ethics Committee for Wales, and all participants provided written informed consent. Participants in the control arm of the trial represented the 'standard care' group who received the standard information sent to all new referrals upon referral: a letter, family history questionnaire and general information leaflet providing basic information about CGSW. These participants did not receive any additional intervention. In addition to the standard information received by all new referrals, participants in the intervention arm of the trial also received the self-help leaflet. The leaflet was sent out at the point of referral along with the standard information as it was important that participants made use of the intervention at the earliest possible point in the process.

The intervention

A copy of the leaflet (in portable document format or digital format) can be downloaded from http://www.cancergeneticsstorybank.co.uk/howdoicope.htm and is also available as an e-appendix. The leaflet describes how distraction techniques may be useful during the waiting period and provided specific examples of cognitive and behavioural distraction techniques, for example thinking of a calm or favourite place or absorbing yourself in something fun or interesting. The leaflet encouraged individuals to write down any distraction techniques they planned to use [32–34]. It also encouraged readers to allow themselves a set period within the day to think about the future and their concerns, if they wished to do so. However, while encouraging these set periods of emotional processing, the leaflet encouraged readers to engage in distraction at all other times when experiencing unwanted intrusive thoughts.

Recruitment procedure

For both arms of the trial potential, participants were targeted upon referral into CGSW and were sent a sealed information pack about the study enclosed within the information sent out from CGSW to all new referrals. A letter attached to the information pack briefly described the study and indicated that individuals only needed to open the pack and read about the study if they were interested in participating. All information packs included a letter of invitation, consent form, participant information sheet, study instructions and a freepost envelope. The information pack also included the Time 1 baseline assessment questionnaire. For those in the intervention group, the intervention was contained in a separate envelope within the study pack. Those who agreed to participate in the trial were asked to return their completed consent form and baseline questionnaire to the research team in the freepost envelope provided before opening the intervention envelope.

Data collection

All data were collected using postal questionnaires. The first baseline questionnaire was included in the

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DOI: 10.1002/pon

information pack sent out to new referrals. The Time 2 questionnaire was sent out 1 month after referral into CGSW, providing information about the effectiveness of the intervention during the waiting period between referral into CGSW and notification of risk. The Time 3 questionnaire was sent out 1 month after participants had been notified of their risk, allowing time for participants to have processed its implications. At Time 3, one reminder questionnaire was sent out 1 month after the initial questionnaire to ensure maximum data capture for this final time-point.

Measures

At Time 1, basic demographic data (age, gender, qualifications and children) were collected. The primary outcome measure was the intrusion subscale of the Impact of Event Scale (IES) [35]. The IES adapted for familial cancer comprises of 15 items capturing the frequency of participants' intrusive thoughts about their risk of developing cancer and attempts to avoid these thoughts. It allows the calculation of separate intrusion and avoidance subscales (with a possible range of 0–35 and 0–40, respectively) and has been used as a measure of cancer-specific distress in a number of studies [36–38], demonstrating good internal consistency and test–retest reliability among women at increased risk of breast cancer [39]. Participants were asked to indicate the degree to which each of the statements on the IES was true for them in relation to their cancer genetic risk assessment *since their referral* (Time 1) and *in the last week* (Times 2 and Time 3). Secondary outcome measures were the IES-avoidance subscale and the Positive and Negative Affect Schedule (PANAS) [40]. The PANAS consists of 20 adjectives used to describe overall positive affect (PA) and negative affect (NA). In the current study, the PANAS at each time-point was anchored to '*how you have felt in the last week*'. Total PA and NA scores range from 10 to 50 with high scores indicating high levels of PA or NA. Finally, the two-item short form of the Life Orientation Test-revised [41] was used at Time 1 only to measure dispositional optimism.

Sample size

The trial was powered for the main comparison of scores on the IES between the control and intervention groups. Sample calculations were based on an anticipated effect size on IES scores of 0.3, derived from the pilot randomized controlled trial [30]. It was estimated that analysing Time 3 questionnaires from 175 participants in each group would give 80% power at a 5% significance level to detect a change in IES score of 5 points (standard deviation = 17.88). A revised sample size calculation was calculated in March 2008 on the basis of calculating pooled variances for those recruited at each time point at this time to obtain a small extension to the trial because of a lower response rate at Time 1 than expected: Time 1, 183 per group; Time 2, 152 per group; and Time 3, 137 per group.

Randomisation

All randomisation sequences were generated by the South East Wales Trials Unit using random permuted blocks of four and stratified by CGSW region. Pre-randomisation allowed all potential participants to be targeted, ensuring that intervention was available and baseline assessment completed upon referral. CGSW staff remained blind as to group allocation of participants, and the research team had no information about participants (other than anonymised referral number) until the receipt of the consent form and baseline questionnaire. Participants were not informed of their group allocation in the study information pack.

Statistical analysis

Initial analysis assessed participation bias at baseline and differential dropout over time. The primary analyses were intention-to-treat and involved analyses of differences in IES intrusion between groups across the follow-up assessments. Because of the large floor effect of the IES, categorical variables were created from the IES-intrusion subscale by placing all zero scores into one category, placing the top 25% of the scores into a 'high distress' category and splitting the remaining scores around the mean. Ordinal regression was conducted using the four IES-intrusion categories as the dependent variable: no (baseline score = 0), low (baseline score = 1–5), moderate (baseline score = 6–12) and high (baseline score = 13 or over) as the dependent variable. In this way, the impact of the intervention upon membership of these categories was analysed at both Times 2 and Time 3, although controlling for variables found to differ proportionately across trial groups. Ordinal regression was also used for secondary analyses to explore the impact of the intervention on the IES-avoidance subscale (see Table 1 for categorical cut-offs). For the secondary analysis, analysis of covariance was used to assess the significance of between-group differences on the Times 2 and Time 3 PA and transformed NA scales, with their respective baseline scores acting as covariates. At Times 2 and 3, a descriptive analysis of responses to the questions measuring usage and perceived effectiveness of the coping leaflet was conducted and, where possible,

Table 1. Psychological profile of sample at baseline

Variable	Intervention *N* = 289 *n* (%)	Control *N* = 301 *n* (%)	χ^2	*p*
Baseline intrusion category				
(Score range)	48 (16.9)	57 (19.5)	1.68	0.64
No intrusion (0)	81 (28.5)	71 (24.2)		
Low intrusion (1–5)	68 (23.9)	75 (25.6)		
Moderate intrusion (6–12)	87 (30.6)	90 (30.7)		
High intrusion (13 or over)				
Baseline avoidance category				
(Score range)	67 (23.7)	69 (23.5)	0.09	0.99
No avoidance (0)	69 (24.4)	74 (25.3)		
Low avoidance (1–6)	75 (26.5)	78 (26.6)		
Moderate avoidance (7–15)	72 (25.4)	72 (24.6)		
High avoidance (16 or over)				
Positive affect	Mean = 25.89	Mean = 26.28	$t = 0.54$	0.59
(Score range 10–50)	SD = 8.70	SD = 8.49		
Negative affect	Median = 15.00	Median = 16.00	$t = -0.63$	0.53[a]
(Score range 10–50)	IQR = 9	IQR = 9		
LOT optimism score	Mean = 11.00	Mean = 12.00	$t = -0.20$	0.84
(Score range 0–30)	SD = 4.33	SD = 4.23		

SD, standard deviation; IQR, interquartile range; LOT, life orientation test [41].
[a]On the basis of transformed variable due to non-normal distribution.

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DOI: 10.1002/pon

compared with similar responses from the control group relating to the standard information.

Results

Participant flow

Between August 2007 and November 2008, 1958 patients referred into CGSW for cancer genetic risk assessment were invited to take part in the trial. The final Time 3 questionnaires were sent out at the beginning of May 2009, allowing sufficient time for the completion of the genetic risk assessment. Figure 1 details the flow of participants through the trial. Of the 980 patients pre-randomised to the intervention arm and the 978 pre-randomised to the control arm, 289 (29%) and 301 (31%) patients, respectively, agreed to participate in the study by returning their Time 1 questionnaire and completed consent form. Although this is a low response rate when calculated from the total number of new referrals targeted, it is important to note here that in previous studies using similar cohorts, the proportion of new referrals who proceeded with their risk assessment ranged from 41% [4] to 57% [30], and thus, this figure is likely to underestimate response rates on the basis of those who proceeded with their referral.

Of the 289 in the intervention group and the 310 in the control group who did consent into the trial and completed their Time 1 questionnaire, 203 (72%) and 226 participants (77%), respectively, returned their Time 2 questionnaires. At Time 3, 152 participants in the intervention group and 179 participants in the control group returned the questionnaires (a follow-up response rate of 67% and 69%, respectively, of those who consented into the trial at Time 1).

For the Time 1–Time 2 analysis, 18 participants in the intervention group and 25 participants in the control group were excluded from the analysis because of protocol deviations. The final Time 1–Time 2 trial sample comprised 185 participants in the intervention group and 201 in the control group. Seven participants in the intervention group and 11 participants in the control group were excluded from the Time 3 trial analysis because they received their risk before the final follow-up questionnaire, leaving a final Time 1–Time 3 trial sample of 145 participants in the intervention group and 168 participants in the control group.

Baseline sample characteristics

There were no differences in response rates between those in the intervention group and those in the control group

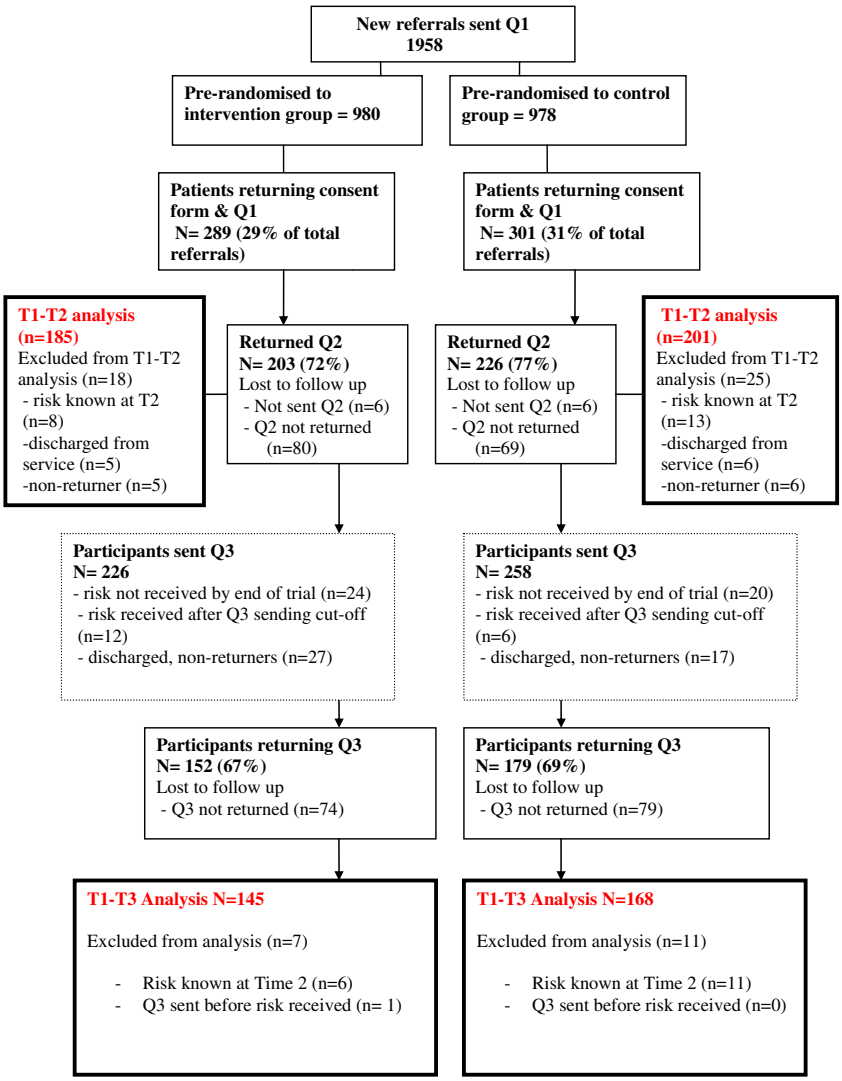

Figure 1. Consort diagram

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DOI: 10.1002/pon

across the three recruitment sites and no significant differences between participants in the intervention group and the control group on baseline psychological variables (see Table 1) or demographic variables. The majority of participants across both groups were female (86.9%), married or cohabiting (72.5%), had at least one child (82.5%) and were educated to at least secondary school level (76.6%).

Participation bias

There was no significant difference in dropout rates from baseline to follow-up between the intervention and control groups. Although it was not possible to explore differences between those consenting to the trial and those who did not consent because of the method of pre-randomisation necessary, analyses of differences between those who remained in the trial compared with those who dropped out after the baseline assessment revealed two interesting findings. As shown in Table 2, participants in the control group who dropped out of the study at Time 2 reported significantly higher levels of baseline IES-avoidance than those who remained in the study ($p = 0.05$). No such difference was found for those in the intervention group. Additionally, those in the control group who dropped out of the study at Time 2 had significantly higher levels of NA at baseline than those who remained in the study ($p = 0.004$). Again, no such difference emerged for those in the intervention groups, suggesting a potential moderating effect of the intervention. No other differences in baseline psychological variables between dropouts and those who remained in the trial were found.

Analysis of the primary outcome: intrusion

Baseline NA was entered into the ordinal regression model because of its association with loss to follow-up described previously. The potential effect of trial centre (Swansea, North Wales and Cardiff) was also explored by including them in the first ordinal regression. No interaction between centre and group emerged, and there was no main effect of centre. Accordingly, this variable was excluded from further analysis.

As shown in Table 3, ordinal regression revealed no significant differences in level of intrusive thoughts between trial groups for the sample as a whole at Time 2. However, there was a significant intervention effect on Time 2 intrusion for participants with moderate levels of intrusive worries at baseline ($p = 0.03$), with those in the intervention group being significantly more likely to fall into a lower distress category at Time 2 than those in the control group.

At Time 3, baseline NA was again entered into the ordinal regression model. However, there was no difference between control and intervention group on either duration of the waiting period or risk outcome (average, moderate or high), and therefore, these variables were not included in any Time 3 analyses. As shown in Table 4, ordinal regression revealed that there was a significant effect of the intervention on Time 3 intrusion for those participants with low or moderate levels of distress at baseline, that is, intervention group participants with either low or moderate intrusion at baseline were significantly more likely to be in a lower distress category at Time 3 than those in the control group ($p = 0.04$ and $p = 0.03$, respectively).

Table 2. Differential dropout analysis on Time 2 completers and non-completers on psychological variables

	T2 completers		T2 dropouts			
	419		158			
Mann Whitney U	Median	IQR	Median	IQR	*z*	*p*
IES-intrusion T1 (score range 0–35)						
Intervention	6.0	13.0	9.5	14.0	−0.89	0.37
Control	6.0	13.0	8.1	15.0	−1.49	0.14
IES-avoidance T1 (score range 0–40)						
Intervention	7.0	13.0	6.0	17.0	−0.11	0.91
Control	6.0	14.0	10.5	16.0	−1.92	0.05
t-test	Mean	SD	Mean	SD	*t*	*p*
Positive affect (score range 10–50)						
Intervention	25.43	8.39	27.03	9.34	−1.37	0.17
Control	25.77	8.68	27.87	7.70	−1.82	0.07
Negative affect (score range 10–50)						
Intervention	17.96	7.88	18.32	8.10	−0.28	0.78
Control	16.91	7.27	19.94	8.67	−2.85	0.005[a]
LOT optimism score (0–30)						
Intervention	12.11	4.37	12.44	4.25	−0.47	0.67
Control	12.16	4.14	11.94	4.51	0.32	0.75

IES, Impact of Event Scale; SD, standard deviation; IQR, interquartile range; LOT, life orientation test [41].
[a]On the basis of transformed variable.

Secondary analyses

Ordinal regression using categorical cut-offs of the IES-avoidance subscale revealed no effect of the intervention at either Time 2 or Time 3. Analysis of covariance revealed that at Time 2, the intervention had no effect on either PA ($F(1, 366) = 0.67$, $p = 0.41$, partial eta-squared $= 0.03$) or NA using the transformed scores ($F(1, 369) = 0.40$, $p = 0.53$, partial eta-squared $= 0.001$), while controlling for baseline scores on the respective measures.

Intervention usage during the waiting period

Of the 203 participants in the intervention group who completed and returned their Time 2 questionnaire, 187 (92.1%) reported that they had read the coping leaflet. Fifty-three participants (28.3% of those who had read the leaflet) reported that they had used the distraction techniques at least once at Time 2 with 67.9% of these individuals reporting that it had helped them cope with worries about cancer genetic risk assessment. Seventeen participants (32.1%) did not feel that the distraction techniques helped them cope, whereas 75 participants (40.1% of those who had read the leaflet) reported that other information in the coping leaflet helped them cope with worries about cancer genetic risk assessment.

Discussion

Although the intervention had no impact upon the sample as a whole, it did result in lower levels of intrusive worries amongst participants with moderate levels of intrusive thoughts at baseline during the waiting period, as well as leading to lower levels of distress in those with low or moderate intrusive worries at baseline following the provision of risk information. Importantly, the findings

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DOI: 10.1002/pon

Table 3. Time 2 intrusion subgroup analysis controlling for baseline NA and intrusion

	Base model *Beta (SE)*	Main model *Beta (SE)*	Interaction model *Beta (SE)*
Threshold			
1	**2.34 (1.09)**	**2.50 (1.10)**	**1.98 (1.13)**
2	**4.28 (1.10)**	**4.45 (1.12)**	**3.96 (1.14)**
3	**5.91 (1.12)**	**6.07 (1.14)**	**5.65 (1.16)**
PANAS (baseline)	**2.03 (0.36)**	**2.06 (0.36)**	**2.10 (0.37)**
Intrusion (baseline)			
No	**−3.12 (0.40)**	**−3.12 (0.40)**	**−3.46 (0.55)**
Low	**−2.18 (0.32)**	**−2.16 (0.32)**	**−2.88 (0.44)**
Moderate	**−1.43 (0.30)**	**−1.44 (0.30)**	**−2.53 (0.46)**
High	**0 (Ref)**	**0 (Ref)**	**0 (Ref)**
Control group	-	0.17 (0.20)	−0.30 (0.50)
Intrusion by control	-	-	
No			0 (Ref)
Low			0.74 (0.61)
Moderate			**1.40 (0.64)**
High			−0.55 (0.65)
Pseudo-R sq	0.476	0.477	0.498

Values in bold have $p < 0.05$.

revealed no evidence that engaging in distraction during the waiting period led to higher levels of distress 1 month following the receipt of cancer genetic risk information, even amongst those individuals with higher levels of distress who did not benefit from engaging with the intervention during the waiting period. These findings provide encouraging evidence that such a simple intervention can be rolled out with ease without concern that it may have any detrimental effect on those unlikely to benefit from it.

Given the low response rate into the trial and the aforementioned finding, a key limitation of this study is that, as in similar studies [1,4], the most anxious individuals may be those who never proceed with their referral. Nevertheless, the level of intrusive thoughts reported at baseline was similar to that reported in other studies of similar cohorts [3,30] with around a quarter of individuals experiencing high or clinical levels of psychological distress. In this study, the only evidence of differential drop-out between trial and intervention group was for intrusion and NA: those in the control group with high scores on these variables were more likely to drop out of the study than those in the intervention group. This has clearly identified the risk that those with high levels of avoidance may not proceed with important cancer risk assessments/screening, thereby identifying a need for early intervention with these individuals where possible.

Another important finding with clear implications for the provision of psychological support to those undergoing cancer genetic risk assessment is that the intervention did not reduce intrusive thoughts in individuals with the highest levels of distress. Although it is possible that further refinement of the intervention in its present form may benefit people with higher levels of worry, it is also important to acknowledge that those with high levels of distress may be in need of more intensive psychological intervention. Within the current intervention format, more specific instructions to regularly practise and use the distraction techniques may have increased the use and benefits of this type of intervention as suggested by other studies [12,42]. Alternatively, providing additional coping

Table 4. Time 3 intrusion subgroup analysis controlling for baseline negative affect and intrusion

	Base model *Beta (SE)*	Main model *Beta (SE)*	Interaction model *Beta (SE)*
Threshold			
1	1.39 (1.21)	1.14 (1.23)	0.81 (1.26)
2	**3.27 (1.22)**	**3.02 (1.23)**	**2.70 (1.27)**
3	**4.80 (1.24)**	**4.56 (1.25)**	**4.25 (1.28)**
PANAS (baseline)	**1.42 (0.40)**	**1.38 (0.40)**	**1.35 (0.40)**
Intrusion (baseline)			
No	**−2.50 (0.42)**	**−2.53 (0.42)**	**−3.46 (0.55)**
Low	**−1.48 (0.34)**	**−1.53 (0.35)**	**−2.88 (0.44)**
Moderate	**−0.60 (0.32)**	**−0.60 (0.32)**	**−2.53 (0.46)**
High	**0 (Ref)**	**0 (Ref)**	**0 (Ref)**
Control group	-	−0.21 (0.22)	**−1.16 (0.52)**
Intrusion by control	-	-	
No			0 (Ref)
Low			**1.36 (0.65)**
Moderate			**1.56 (0.70)**
High			0.58 (0.67)
Pseudo-R sq	0.342	0.344	0.361

Values in bold have $p < 0.05$.

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DOI: 10.1002/pon

strategies such as mindfulness or cognitive challenge may prove of benefit. Although psychological interventions for individuals in the later stages of the cancer genetic assessment/testing process involving multiple group therapy sessions [10,11] or individualised coping skills training [12] have been shown to be effective, these have involved substantial therapist time and are unlikely to be practical or cost-effective options for NHS service providers where hundreds of patients are accessing a service across a large geographical region. Additionally, these more complex interventions may be more difficult to teach and implement, and their use by people with no defined psychopathology or stated desire to learn psychological coping techniques cannot be ensured. Accordingly, more research is necessary to determine whether the provision of alternative or additional coping strategies is likely to be of benefit to those with the highest levels of distress. Of course, the question of whether NHS tertiary services such as CGSW should be responsible for providing any interventions for individuals who are likely to have pre-existing high levels of general anxiety/neuroticism [4] is an important one to contemplate.

Of particular relevance to the effectiveness of the intervention post-risk information is that the coping leaflet emphasises that once informed of their risk, individuals will receive information and support to help them make decisions about the future. The combination of distraction techniques and reassuring information may well benefit those individuals who have concerns about the implications of their risk and who doubt their ability to cope with learning their risk. Previous studies have indicated that increasing an individual's perceived personal control and ability to cope can help them adapt to genetic risk information [43,44], and this combined aspect of the intervention may represent an important active component of the leaflet.

Previous studies have suggested that high levels of distress may act as a barrier to undergo cancer genetic risk assessment and genetic testing [3,8,9]. Although the intervention appears to offer a potential means of support for individuals who may not otherwise choose to proceed with their referral, the question of how best to access those individuals who decline referral or discontinue risk assessments because of high levels of distress continues to be problematic. The intervention reported here may well be one solution to this problem, and its effectiveness in this context could be evaluated through exploring whether implementing the intervention into normal service delivery also improves engagement with and retention to such services.

Despite the apparent limitations of the intervention, there is sufficient evidence to suggest that this brief self-help coping leaflet does reduce intrusive thoughts in a number of individuals undergoing cancer genetic risk assessment both during and following the waiting period, without any subsequent negative effect. The cost of the leaflet (one sheet of A4) was negligible and is therefore likely to be a highly cost-effective intervention for NHS services with potential roll-out to other cancer services involving periods of waiting and uncertainty. Such an intervention could also be easily adapted to an on-line information resource and, within CGSW, is currently being piloted as a digital story. This paper has revealed that our simple, easy to implement intervention, which is clearly aligned to coping theory and coping effective training, can provide people with a valuable 'toolkit' of coping skills from which they can make use of at appropriate times in their journey, whether that be as an affected cancer patient or someone living with the fear of being at increased genetic risk of cancer.

Supporting information

Supporting information may be found in the online version of this article.

Acknowledgements

This study was funded by the Wales Office for Research and Development Health and Social Care Funding Scheme and sponsored by Cardiff University. We thank all the men and women who agreed to participate in this trial. We acknowledge the support received from the clinical and administrative staff of CGSW throughout this trial. We would like to thank Helen Jones and Jennifer Hilgart for their Research Assistant support, the Trial Steering Committee and John Evans (SEWTU) for designing and supporting the secure database. The South East Wales Trial Unit is funded by the Wales Office for Research and Development in Health and Social Care. There are no financial and personal relationships between any authors and others that might bias their work.

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DOI: 10.1002/pon

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DOI: 10.1002/pon