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Title: Illness Perceptions, Fear of Cancer Recurrence and Mental Health in Teenage and Young Adult Cancer Survivors

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Abstract

Background: The Common-Sense Model of illness self-regulation is suggested to underpin illness specific cognitions (including both illness perceptions and a fear of cancer recurrence; FCR). There is evidence in adults of associations between FCR, illness perceptions and mental health in adult cancer survivors. However, there is limited empirical research examining these constructs within the developmentally distinct population of adolescent and young adult (AYA) survivors of cancer. The current study aimed to bridge that gap to inform potentially modifiable treatment targets in this population.

Methods: A cross-sectional, correlational design was used to examine the associations between illness perceptions, FCR and mental health. A web-based survey was completed by a convenience sample of AYA survivors. Regression and mediation analysis were performed.

Results: Overall, more negative illness perceptions were associated with more severe FCR and greater depressive and anxiety symptomatology. Higher FCR was predictive of worse overall mental health. More negative overall illness perceptions predicted the relationship between FCR-Depression, mediating 24.1% of the variance. Contrastingly, overall illness perceptions did not predict or mediate the relationship between FCR-Anxiety. However, the specific illness perceptions regarding timeline, personal control, and emotional representation, were predictive of the FCR-Anxiety relationship.

Discussion: Illness perceptions and FCR were predictive of mental health outcomes. Identifying and therapeutically targeting negative illness perceptions in those young adults who have survived adolescent cancer could therefore be a means of reducing anxiety and depressive symptomatology. Limitations and future directions are discussed.

Key Words: Adolescent and Young Adults; Fear of Cancer Recurrence; Illness Perceptions; Depression; Anxiety

Introduction

In the UK, around 2,500 adolescent and young adults (aged 15-24) are diagnosed with cancer every year (Cancer Research UK, n.d) and incidence and survival rates are increasing. Experiencing and surviving cancer can impact on developing identities and cognitive processes, e.g., illness understanding and metacognition (Tutelman & Heathcote, 2020). However, relatively little is known about the psychological aspects of survivorship for this developmentally distinct group into adulthood.

Whilst navigating the unique challenges associated with their cancer, AYAs are also undergoing considerable normative psychological, emotional, physical, and cognitive development (Abrams, Hazen & Penson, 2007; Blakemore & Choudhury, 2006). Compared with children, AYAs are expected to become more autonomous (Zimmer-Gembeck & Collins, 2008) and take increasing responsibility for their own healthcare (Paone, Wigle & Saewyc, 2006). Compared with adults, AYAs have significantly different psychosocial needs (e.g., experiencing higher emotional, informational, physical, and financial distress; Smrke, 2020) and different perceptions towards life (Arnett, 2001). It is, therefore, unsurprising that facing cancer as an adolescent is associated with significant distress (Sansom-Daly & Wakefield, 2013), poorer mental health and other negative psychosocial outcomes (e.g., missing school or work; Zebrack, 2011; Lauer, 2015; Park & Rosenstein, 2022). For some, the period following treatment is associated with unfavourable psychosocial outcomes (Parsons et al., 2012; Tai et al., 2012; Moss et al., 2021), and up to 34% experience posttraumatic stress disorder (PTSD), 13% clinical depression, and 8% anxiety (Kosir er al, 2019).

'Fear of Cancer Recurrence' (FCR) describes a fear or worry that cancer will return or progress (Vickberg, 2003) and is common in AYA cancer survivors, affecting approximately two in three (Thewes et al., 2018). It is the primary psychological concern of young adult cancer survivors (Li & Cheng, 2021) and can negatively affect the transition from 'patient' to 'survivor' identity (Jones, Parker-Raley & Barczyk, 2011) or from paediatric to adult services (Granek et al., 2012). High FCR in AYAs is associated with previous illness recurrence and worse mental health (Fonseca et al., 2010; Lane et al., 2019; Sun et al., 2019), cancer type (e.g., higher in breast cancer and malignant melanoma survivors; Vandraas et al., 2019), lower health related quality of life (Thewes et al., 2018) and increased surveillance scan anxiety (Heathcote et al., 2022). For some survivors, FCR can become chronic and disabling (Simard & Savard, 2007). It has negative effects on social relationships and quality of life (Parsons et al., 2012), is associated with higher clinical costs (e.g., increased use of health services (Lebel et al., 2013; Vachon et al., 2021) and does not decrease in severity over time in cancer survivors of \geq 5 years (Koch et al., 2013).

Illness perceptions influence how people respond to illness (Petrie & Weinman, 2006) and can range from negative (e.g., I have no control over my illness) to positive (e.g., I have control over my illness). Illness perceptions are a potentially malleable treatment target which are known to be associated with FCR within child and adult survivor populations (Fonseca et al., 2010; Kaptein et al., 2015), but have not previously been investigated specifically within AYA cancer survivors. In adult cancer survivors, more negative illness perceptions can be associated with worse FCR, increased cancer worry and other negative psychosocial outcomes (Corter et al., 2013; Phillips et al., 2013; Kaptein et al., 2015; Freeman-Gibb et al., 2017).

The Common-Sense Model (CSM) of illness self-regulation (Leventhal, Meyer & Nerenz, 1980; Leventhal, 1984) provides a strong theoretical rationale for investigating FCR and illness perceptions. The CSM suggests that cognitive beliefs and emotional representations/responses to illness influence health outcomes and coping strategies

(McAndrew et al., 2019). The model proposes five core dimensions of illness beliefs (influenced by past experiences etc): cause; identity; perceived control; severity of illness consequences; and timeline (e.g., perceived risk of recurrence), as well as emotional representations (e.g., the level of worry that the cancer will return; Lee-Jones et al., 1997; Fardell et al., 2016; illness-related distress; Hagger & Orbell, 2003; see supplementary information for clinical example). CSM predicts that more negative cognitive representations, alongside more negative emotional representations lead to more negative illness perceptions and increased FCR. Although seemingly important to AYA cancer survivors based on their narrative accounts (e.g., around control and emotional representations of cancer; Kameny & Bearison, 2002; and how they identify with it; Jones, Parker-Raley & Barczyk, 2011), no prior research has empirically investigated the association between illness perceptions and FCR in this developmentally distinct population.

Like FCR, illness perceptions have also been linked to mental health outcomes (e.g., negative illness perceptions to increased stress; Miceli et al., 2019; and negative illness perceptions to depression; Sansom-Daly et al., 2018; Thong et al., 2018, Wroot et al., 2020). Husson et al., (2020) found significant differences in illness perceptions of AYAs compared to older populations of thyroid cancer survivors, although regardless of age, more negative illness perceptions (particularly illness timeline) were associated with more distress.

Despite the theoretical rationale and evidence of associations between FCR, illness perceptions and mental health in other age groups, and indications that both FCR and illness perceptions are important to AYAs, limited empirical research has examined these constructs. We aimed to bridge that gap by investigating the associations between FCR, illness perceptions and mental health in this developmentally distinct population. We hypothesised that:

- 1. High FCR would be associated with more negative illness perceptions.
- 2. High FCR and more negative illness perceptions would be associated with poorer mental health (depression and anxiety).
- 3. The association between high FCR and poorer mental health (depression and anxiety) would be mediated by illness perceptions.

Method

Design and Sample

Cross-sectional, correlational design. A web-based survey was administered via Qualtrics Survey Software®. Participants were a convenience sample of AYA cancer survivors, recruited over 14 months (November 2020 to January 2022), and were included/excluded based on specific criteria (aged between 16-30, treated for cancer between 13-24 years, having completed active cancer treatment and been in remission for 6+ months (see supplementary information for further details). We used the CHERRIES checklist to improve the quality of the web-based survey (Eysenbach, 2004; see supplementary information).

Measures

We gathered demographic and clinical data, and used validated measures to assess the variables of interest:

- Illness Perceptions: Brief Illness Perception Questionnaire (BIPQ; Broadbent et al, 2006)
- FCR: Fear of Cancer Recurrence Inventory Short Form (FCRI-SF; Simard & Savard, 2009)

Mental health outcomes: Patient Health Questionnaire (PHQ-9; Kroenke & Spitzer, 2002) assessed depression and the Generalised Anxiety Disorder Scale (GAD-7; Spitzer et al, 2006) captured anxiety.

See supplementary information for descriptions of measure validity and rationale.

Procedure

Social media study adverts were posted on Twitter, Facebook, Instagram, and TikTok which included a video of the researcher and a person with personal experience of cancer (PPE) discussing the research or a picture of the project poster. Adverts were also shared via several charities (e.g., Shine) and support groups (e.g., Trekstock). Those who were interested could directly follow an anonymous Qualtrics link from the advertisement or email the research team. Clicking on the link took participants to the online research platform, where they were presented with information about the study, the research team's contact details in case of questions, and a consent form. Subsequently, they filled out study measures. The study took 15-30 minutes to complete, although participants could complete this within a week (using unique IP addresses ensured participants could only participate once).

Demographic and clinical questions were presented first and ensured inclusion/exclusion criteria were met before proceeding to remaining questionnaires. Qualtrics was set to ensure all questions were completed and participants could review and change responses before submission. Participants could opt into a prize draw to receive an online voucher (one of five £20 amazon vouchers) and could email the researcher to ask for a summary of the research findings.

Ethical approval was gained from the [removed for blinding]. The research was anonymous (with an email address recorded only if participants chose to leave it) and confidential with all information relating to research participants and their data obtained and stored in line with EU General Data Protection Regulations (2018). It was not expected that completion of the questionnaires would have any negative effects for participants. However, following responses to question 9 on the PHQ-9 (which refers to suicidal/self-harm ideation) and at study completion, participants were signposted to various mental health resources (e.g., Mind, Samaritans, and Macmillan cancer support). Throughout each stage of the research (e.g., project development, recruitment, data collection and analysis), we consulted with people with personal experience to discuss the research and ensure changes reflected service-user perspectives.

Statistical analysis

The data analysis plan was pre-registered on 16th December 2021 (https://osf.io/xec57/). Statistical analyses were done using SPSS Version 27. Descriptive statistics were performed for participant demographic information, clinical characteristics and all outcome measures. A hierarchical multiple linear regression, with mental health as the dependent variable, and FCR and illness perceptions as the independent variables, including covariates (age and gender) then examined the proportion of the variance in mental health accounted for by FCR and illness perceptions. Gender and age were first added to the regression models, followed by FCR and finally illness perceptions. Additional to the preregistered protocol, post-hoc linear regressions were completed, which included the illness perception sub-scales, instead of overall illness perception scores. Sub-scales were added to the regression models one at a time. Mediation analyses (using the PROCESS macro) finally investigated if any associations found between FCR, and depression/anxiety were mediated by illness perceptions. The final dataset was retrieved from Qualtrics on 18th January 2022.

Results

Participant Characteristics

Of the 214 participants who accessed the survey, 124 did not meet inclusion criteria or were excluded for not fully completing the survey. The final sample consisted of 90 (42% completion rate) AYA cancer survivors, aged 16-30. The mean age was 22.42 years (SD = 2.98), most participants identified their ethnicity as from the UK (91.1%), the majority identified as single (67.8%), and the most frequent highest education achieved was A-Levels (41.1%). Additionally, participants were most frequently recruited via Instagram (53.3%; see supplementary information for more detail). 93.3% of participants showed clinical levels of FCR on the FCRI-SF, 38.9% showed moderate or severe anxiety on the GAD-7, and 35.6% showed moderate or moderately severe depression on the PHQ-9. Further descriptive statistics for illness perceptions, FCR, anxiety, depression, quality of life and functioning impairment can be found in table 1.

Table 1

	Total (N=90)
BIPQ, mean (SD) scores	
Consequences	5.47 (2.48)
Timeline	4.82 (3.26)
Personal Control	6.92 (2.48)
Treatment Control	2.39 (2.42)
Identity	4.71 (2.62)
Illness Concern	6.28 (2.42)
Coherence	3.29 (2.44)
Emotional Representation	7.34 (2.23)
Total score	41.22 (12.76)
FCRI-SF, mean (SD) scores	
Total score	22.57 (5.62)
Clinical Levels, yes (%)	84 (93.3%)
GAD-7, mean (SD) scores	
Total score	8.76 (5.47)

Descriptive Statistics of Key Study Variables

GAD-7 clinical level	
Minimal anxiety, n (%)	21 (23.3)
Mild anxiety, n (%)	34 (37.8)
Moderate anxiety, n (%)	16 (17.8)
Severe anxiety, n (%)	19 (21.1)
PHQ-9, mean (SD) score	
Total score	9.61 (6.21)
PHQ-9 clinical level	
Minimal depression, n (%)	19 (21.1)
Mild depression, n (%)	30 (33.3)
Moderate depression, n (%)	26 (28.9)
Moderately severe depression, n (%)	6 (6.7)
Severe depression. n (%)	9 (10)
WHOQOL-BREF, mean (SD) scores	
Physical QoL	12.17 (1.90)
Psychological QoL	12.11 (2.42)
Social Relationships QoL	13.60 (3.36)
Environmental QoL	15.07 (2.35)
WSAS, mean (SD) scores	
Total Impairment score	12.53 (7.63)

Key: BIPQ – Brief Illness Perception Questionnaire; FCRI-SF – Fear of Cancer Recurrence Inventory-Short Form; GAD-7 - Generalised Anxiety Disorder Assessment; PHQ-9 – Physical Health Questionnaire; WHOQOL-BREF - World Health Organization Quality of Life Assessment Brief; QoL – Quality of Life; WSAS – Work and Social Adjustment Scale.

Clinical Characteristics

Most participants had experienced either Lymphoma (46.7%) or Leukaemia (20%).

The most common stage of cancer at diagnosis was stage 2 (35.6%), with most participants

having received chemotherapy as treatment (93%). Time from diagnosis to remission was

most commonly ≤ 2 years (86.7%), while time in remission at study participation was

variable (e.g., > 1 year = 21.1%; 1-2 years = 20%; 2-3 years = 16.7%). See supplementary information for more detail.

Question 1: What are the associations between FCR, Illness Perception and Mental Health?

i) Correlation Analysis

Tests for normality of distribution were conducted, which indicated that the data was not normally distributed. Therefore, Spearman's correlations were performed to analyse the associations between the study's target variables. Significant associations were seen between all the target variables. Specifically, high FCR associated with more negative overall illness perceptions ($r \ge 0.6$), with the specific perceptions of consequences, illness concern, and emotional representation being particularly strongly associated with high FCR ($r \ge 0.5$). Both a high FCR and more negative illness perceptions were also associated with poorer mental health on the GAD-7 and PHQ-9 ($r \ge 0.4$; Table 2).

Table 2Spearman's Correlation Analysis

	1	2	3	4	5	6	7	8	9	10	11	12	13	14
1. Age	1.00													
2. Gender	.12*	1.00												
3. BIPQ - Consequences	.37**	.09	1.00											
4. BIPQ – Timeline	12	05	.52**	1.00										
5. BIPQ – Personal Control	00	05	.31**	.21*	1.00									
6. BIPQ – Treatment Control	.01	17	.28**	.42**	.21*	1.00								
7. Identity	26*	.13	.64**	.54**	.01	.22*	1.00							
8. Illness Concern	31**	.05	.63**	.26*	.23*	.22*	.42**	1.00						
9. Coherence	09	13	.06	.10	.12	.50**	04	.16	1.00					
10. Emotional Representation	32**	.19	.53**	.05	.31*	.10	.22*	.58**	.04	1.00				
11. BIPQ Total	26*	04	.79**	.70**	.45**	.62**	.64**	.68**	.39**	.50**	1.00			
12. FCRI-SF Total	18	.08	.62**	.28**	.37**	.20	.33**	.59**	.12	.62**	.62**	1.00		
13. GAD-7 Total	32**	.13	.53**	.06	.36**	.06	.23*	.45**	.11	.68**	.44**	.60**	1.00	
14. PHQ-9 Total	30**	.20	.55**	.16	.32**	.21*	.29**	.47**	.10	.60**	.49**	.57**	.82**	1.00

Key: BIPQ – Brief Illness Perception Questionnaire; FCRI-SF – Fear of Cancer Recurrence Inventory-Short Form; GAD-7 - Generalised Anxiety Disorder Assessment; PHQ-9 – Physical Health Questionnaire; * Correlation is significant at the 0.05 level; ** Correlation is significant at the 0.01 level.

Hierarchical multiple regression analyses were conducted to investigate the potential interaction of illness perceptions on the FCR–mental health association. The data was checked and met the eight assumptions needed for completing a multiple hierarchical linear regression (see supplementary information).

Anxiety. The first set of regression models explored the potential interaction of overall illness perceptions on FCR–Anxiety (GAD-7). Model 2, which included total FCR score, age, and gender, explained 41.1% of the variance in total GAD-7 scores. However, adding overall illness perception scores did not significantly increase the amount of variance explained (Model 3; Table 3).

Table 3

Hierarchical Linear Regression for Anxiety, including Age, Gender, FCR Scores and Total Illness Perception Scores.

Model	R ²	F change	Sig. F Change
Model 1. Age; gender	.11	5.46	.006**
Model 2. FCRI-SF Total	.41	43.83	.000**
Model 3. BIPQ Total	.42	.80	.373

Key: BIPQ – Brief Illness Perception Questionnaire; FCRI-SF – Fear of Cancer Recurrence Inventory-Short Form; ** Significance at the 0.01 level.

Model 3 (which included overall illness perception scores, total FCR scores, age, and gender) was a significant predictor of anxiety scores, F (4,85) = 15.20, p = .000. Within this, FCR (β = .49, p = .000) and age (β = -.32, p = .045) showed a significant main effect, with gender (β = 1.18, p = .246) and BIPQ total not showing significance (β = .04, p = .373). This indicates that FCR predicted anxiety scores above and beyond gender and overall illness perceptions.

Despite the total illness perception score not being a significant predictor of the anxiety and FCR association, the correlational analysis showed associations between the illness perception sub-scales and both anxiety scores and FCR scores. A post-hoc regression was therefore completed to investigate if any of the specific illness perception sub-scales explained any variance in the FCR-Anxiety relationship. Model 9 (which included the 8 individual BIPQ sub-scales scores, total FCRI scores, age, and gender) was significant and explained 57.1% of the variance in total GAD-7 scores (Table 4).

Table 4

Model	R ²	F change	Sig. F Change
Model 1. Age; gender	.11	5.46	.006**
Model 2. FCRI-SF Total	.41	43.83	.000**
Model 3. Consequences	.43	2.09	.152
Model 4. Timeline	.48	8.69	.004*
Model 5. Personal	.51	5.55	.021*
Control			
Model 6. Treatment	.51	.11	.895
Control and Identity			
Model 7. Illness Concern	.51	.02	.880
Model 8. Coherence	.52	1.49	.226
Model 9. Emotional	.57	8.73	.004*
Representation			

Hierarchical Linear Regression for Anxiety, including Age, Gender, FCR Scores and Individual Illness Perception Sub-Scale Scores.

Key: FCRI-SF – *Fear of Cancer Recurrence Inventory-Short Form;* * *Significance at the* 0.05 *level;* ** *Significance at the* 0.01 *level.*

Model 9 (which included the 8 individual BIPQ sub-scales scores, total FCRI scores,

age, and gender) was a significant predictor of anxiety scores, F (11,78) = 9.42, $p \le .000$.

Within this, FCR had a significant main effect ($\beta = .28$, p = .008), as did the illness

perceptions of timeline ($\beta = -.39$, p = .023), personal control ($\beta = .41$, p = .040), and

emotional representation ($\beta = .80, p = .004$). This indicates that these 3 specific illness

perceptions predicted the relationship between FCR-Anxiety.

Depression. The second regression models explored the potential interaction of

illness perceptions on the FCR-Depression (PHQ-9) link. Model 3 (which included total FCR

scores, overall illness perception scores, age, and gender) was significant and explained

41.9% of the variance in total PHQ-9 scores (Table 5).

Table 5

Hierarchical Linear Regression for Depression, including Age, Gender, FCR Scores and Total Illness Perception Scores.

Model	R ²	F change	Sig. F Change
Model 1. Age; gender	.11	5.56	.005**
Model 2. FCRI-SF Total	.39	38.69	.001**
Model 3. BIPQ Total	.41	4.52	.036*

Key: BIPQ – *Brief Illness Perception Questionnaire; FCRI-SF* – *Fear of Cancer Recurrence Inventory-Short Form;* * *Significance at the 0.05 level;* ** *Significance at the 0.01 level*

Model 3 (which included total FCR scores, total BIPQ scores, age, and gender) was a significant predictor of depression scores, F (4,85) = 15.34, p = .036. Within this, FCR (β = .45, p = .000), BIPQ total (β = .11, p = .036) and gender (β = 2.35, p = .044) showed a significant main effect, with age (β = -.25, p = .173) not showing significance. This indicates that both overall illness perceptions and gender predicted the relationship between FCR-Depression.

A post-hoc regression was completed to investigate whether any specific illness perceptions were significant predictors of FCR-Depression. Model 3 (which included total FCR scores, the consequence illness perception subscale, age, and gender) significantly explained 42.3% of the variance in total PHQ-9 scores. However, individually adding the other 7 illness perception sub-scales did not significantly increase the amount of variance explained (Model 9; Table 6).

Table 6

Model	R ²	F change	Sig. F Change
Model 1. Age; gender	.11	5.56	.005**
Model 2. FCRI-SF Total	.39	38.69	.001**
Model 3. Consequences	.42	5.14	.026*
Model 4. Timeline	.44	2.42	.124
Model 5. Personal Control	.46	2.53	.115
Model 6. Treatment	.47	.78	.460
Control and Identity			
Model 7. Illness Concern	.47	.21	.647
Model 8. Coherence	.47	.00	.983
Model 9. Emotional	.48	1.92	.170
Representation			

Hierarchical Linear Regression for Depression, including Age, Gender, FCR Scores and Individual Illness Perception Sub-Scale Scores.

Key: FCRI-SF – *Fear of Cancer Recurrence Inventory-Short Form;* * *Significance at the* 0.05 *level;* ** *Significance at the* 0.01 *level*

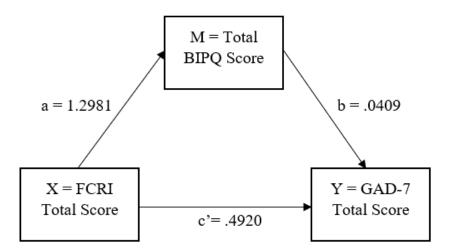
Model 9 (which included the 8 individual BIPQ sub-scales scores, total FCRI scores, age, and gender) was a significant predictor of depression scores, F (11,78) = 6.56, p = .000. However, within this, only FCR had a significant main effect (β = .31, p = .021), with no significant effects for gender, age, or any of the BIPQ subscales. Thus, indicating that none of the specific illness perception domains, predicted the relationship between FCR-Depression.

Question 2: Do illness perceptions mediate the association between high FCR and poorer

mental health?

Anxiety. A mediation analysis was completed to investigate whether there was an indirect effect of illness perception scores, within the total effect seen between FCRI scores and GAD-7 scores (Figure 1).

Figure 1 Illness Perceptions Mediation Model of FCR-Anxiety.

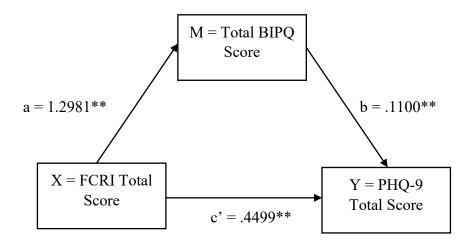


The indirect effect of illness perceptions on the FCR-Anxiety interaction was not significant, due to the bootstrap confidence intervals including 0 [Effect = .05, 95% C.I. (-.07, .19)]. Illness perceptions therefore do not significantly mediate the total effect (β = .55) seen between FCRI total scores and GAD-7 total scores.

Depression. A mediation analysis was completed to investigate whether there was an indirect effect of illness perception scores, within the total effect seen between FCRI scores and PHQ-9 scores (Figure 2).

Figure 2

Illness Perceptions Mediation Model of FCR-Depression.



The indirect effect of illness perceptions on the FCR-Depression interaction was significant, due to the bootstrap confidence intervals not including 0 [Effect = .14, 95% C.I. (.02, .28)]. Illness perceptions therefore significantly mediate the total effect (β = .59) seen between FCRI total scores and PHQ-9 total scores. Specifically, FCRI scores account for 75.9% of PHQ-9 scores in AYA cancer survivors, but 24.1% is accounted for through the illness perceptions of these AYAs.

Discussion

In AYA cancer survivors, we found that more negative illness perceptions were associated with higher FCR and more severe anxiety and depression. Specifically, illness concern, consequences, and personal control were most strongly associated with all outcome variables. High FCR was predictive of worse mental health outcomes; total illness perceptions predicted and mediated the relationship between FCR and depression. Contrastingly, total illness perceptions did not predict the relationship between FCR-Anxiety, but the specific illness perceptions of timeline, personal control, and emotional representation did.

Our novel finding that high FCR was associated with more negative illness perceptions in AYA cancer survivors (hypothesis 1 supported) fits with similar findings in adult populations (e.g., Phillips et al., 2013; Kaptein et al., 2015). This is perhaps unsurprising given the shared theoretical underpinning of CSM for both illness perceptions and FCR. We found significant positive correlations between individual illness perceptions and FCR scores, suggesting that the cognitive representations underpinning FCR (e.g., around cancer timeline or perceived control) might conceptually overlap with those underpinning illness perceptions (e.g., around illness timeline or personal control). Expanding on the existing evidence (Thong et al., 2018; Sun et al., 2019; Wroot et al., 2020), we found that both illness perceptions and RCT are important contributory factors in the mental health of AYA cancer survivors (second and third hypotheses supported). This is particularly important considering the increased vulnerability AYAs have for mental health difficulties following cancer treatment (Kosir er al, 2019; Moss et al., 2021), at a time when they are also undergoing distinct psycho-emotional changes (Abrams, Hazen & Penson, 2007; Blakemore & Choudhury, 2006). It is unsurprising that anxiety was associated with FCR, given that FCR is fear or worry that cancer will return (Vickberg, 2003), while general anxiety is defined as the experience of persistent, excessive, and unfocused worry and anxiety (Tyrer and Baldwin, 2006; NICE, 2011). However, FCR and anxiety need exploring further to understand how these constructs interact: Do AYAs who are anxious fear most things, including cancer recurrence? Or is the experience of anxiety related to FCR different and so need specific understanding and informed care?

Whilst overall illness perceptions did not predict the relationship between FCRanxiety, the individual illness perceptions of timeline, personal control and emotional representation, were shown to be predictive (more negative perceptions predicting a more negative interaction). The CSM suggests that someone's cognitive and emotional representations of their illness precede and influence their physical and emotional coping responses (McAndrew et al., 2019). It is therefore possible that these individual cognitive and emotional illness perceptions (i.e., thinking you have no control; thinking that cancer will last for longer; experiencing a greater emotional impact due to having cancer), may be more directly associated with the emotional response of experiencing anxiety and uncertainty. However, while these individual perceptions may play a predictive role and could be used to predict AYA FCR-Anxiety levels, overall illness perceptions did not significantly mediate the total effect seen between FCR and anxiety. This suggests that while a potential clinical marker, targeting overall illness perceptions may not reduce FCR and anxiety, and these may need more direct targeting, through evidence-based approaches, such as cognitivebehavioural therapy (CBT) (NICE, 2014).

Contrastingly, we found that more negative overall illness perceptions were predictive of the association between high FCR-depression, mediating the relationship seen between FCR and depression, and accounting for 24.1% of the variance. It may be that more negative illness perceptions associate directly with factors associated with depression. For example, more negative cognitive and emotional perceptions may contribute to a negative thinking bias or a feeling of hopelessness - common symptoms and perpetuating factors of depression (Beck, 1979). The CSM would suggest that more negative illness perceptions and higher FCR (both underpinned by negative cognitive and emotional representations of cancer), influence depressive thinking styles and feelings to predict depression (i.e., they influence worse emotional coping responses). However, the reverse interaction could be true - a negative thinking bias could influence the development of more negative illness perceptions, or the interaction might be bi-directional. Overall illness perceptions might be important in the association between FCR and depression, acting as a potentially modifiable treatment target, reducing FCR and improving depression. However, further longitudinal research is needed to develop a causal understanding.

Strengths and Limitations

A pre-registered data analysis plan increased replicability of findings and prevented ad-hoc decisions, minimising risk of bias or false positives (Moore, 2016). Important covariates (gender and age) were controlled for, decided a priori, to further minimise risk of bias. The potential conceptual overlap of worry/fear was considered across all three constructs of anxiety, FCR and emotional representations (within illness perceptions), and tests conducted for multicollinearity to increase reliability of analyses. The data did not show

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multicollinearity, suggesting distinct functions in each construct. However, future research might explore the conceptual similarities and differences of these constructs. The use of the CHERRIES checklist (Eysenbach, 2004) additionally improved the quality of the web-based survey.

The cross-sectional nature of the research makes causality and direction of relationships difficult to establish (Fiedler, Schott & Meiser, 2011). While illness perceptions were found to mediate the relationship between FCR-depression, which variable precedes another is unclear (e.g. negative illness perceptions preceding FCR). Mediation analysis did allow inferences to be made about potential dual roles of illness perceptions, as both a potential cause and effect of high FCR and lower mood. The lack of prior research on how illness perceptions, FCR and mental health interact in TYA cancer survivors means this initial investigation provides useful information to inform future longitudinal research.

An online UK-wide survey was used as a convenient and cost-effective way of reaching young populations from a broad range of regions - intended to increase generalisability. However, online surveys are at greater risk of self-selection bias (Bethlehem, 2010), and our sample predominantly identified as British (91.1%) – somewhat higher than the national figure, according to Census Data (2011). Recruiting only in the UK meant a western, educated, industrialised, rich and democratic cultural sample (WEIRD; Henrich, Heine & Norenzayan, 2010). As such, if illness perceptions are culturally and contextually specific, we cannot necessarily generalise to other cultures.

Implications for research and clinical practice

Our findings suggest that interventions targeted at improving illness perceptions may be a way to target both FCR and depression in AYAs, in line with previous literature in other populations (e.g., diabetes; Keogh et al., 2007; myocardial infarction; Petrie et al., 2002). Discovering effective and efficacious ways of improving illness perceptions, while also improving FCR levels and mental health for young people with cancer, would be clinically important and time and cost effective.

Our findings highlight the importance of all health professionals holding FCR and illness perceptions in mind when working with AYA cancer survivors. Illness perceptions and FCR can help clinicians understand and formulate AYAs' experiences of anxiety and depressive symptomatology. For young people who are anxious, the illness perceptions around cancer timeline, personal control, and emotional representation might be prioritised. For young people who are depressed, it may be helpful to explore overall illness perceptions. Clinicians routinely measuring FCR (using the FCRI-SF) and illness perceptions (using the BIPQ), could aid in assessment, formulation and intervention related to illness perceptions, FCR and mental health. For example, nursing staff could use measures to discuss with AYA cancer survivors how they perceive their cancer and the impact it is having on their mood. This initial exploration could inform appropriate stepped-care interventions -from basic psychoeducation and support to signposting / referral for targeted additional support (e.g. psychology). It appears to be important that high FCR and poor mental health be taken into account when planning service delivery. For example in considering that when AYAs enter remission, the frequency of clinical contact significantly reduces, minimising their access to support with these ongoing challenges.

Future longitudinal research should explore the direction of the associations between FCR and illness perceptions and their effect on mental health. Building an understanding of the processes of influence will help inform both where and when clinical intervention should be targeted.

Longitudinal research could explore and further conceptualise FCR and its relationship to mental health outcomes. A low level of anxiety around recurrence might help

AYAs make helpful health related decisions (e.g., attend check-ups). However, questions remain as to whether there is a point at which FCR transitions from adaptive to maladaptive, when it begins to predict poor mental or physical health outcomes. Understanding these processes could inform future clinical practice as to when additional support is helpful.

Conclusion

The findings indicate that illness perceptions and FCR are significantly associated, and both predict mental health outcomes. Overall illness perceptions predict depression, while the specific illness perceptions of timeline, personal control, and emotional representation predict anxiety. Additionally, overall illness perceptions mediate the relationship between FCR and depression, but do not mediate the relationship between FCR and anxiety. This suggests that FCR and illness perceptions are important in the experience of AYA cancer survivors, could help explain vulnerability to mental health difficulties and should be considered within clinical interventions and services. Further longitudinal and cross-cultural research into these relationships could improve our understanding and inform future clinical practice, policy and guidelines.

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Online Supplementary Materials

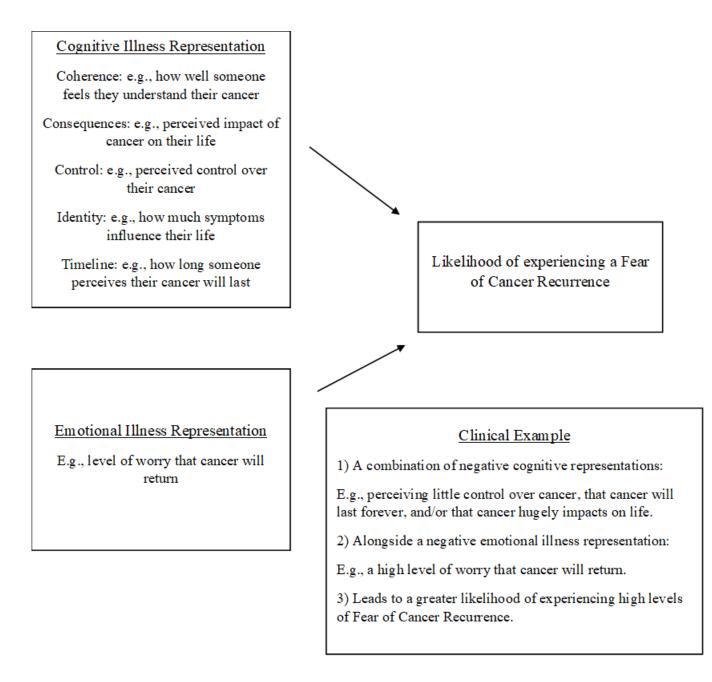
Material 1

Example of the Common-Sense Model Applied to the Experience of FCR. The model

highlights how the interaction between someone's cognitive illness representations and their

emotional illness representations influence a cancer survivors experience of a Fear of

Cancer Recurrence.



Study Inclusion/Exclusion Criteria and Rationale

Inclusion Criteria	Rationale
1) Aged between 16-30 years old.	 To avoid capturing part of the 'pre-pubescent children' population (i.e., 13-16; Košir, 2020). Developmentally 16-24 is a unique period when adolescents are individuating from their families of origin and becoming autonomous adults, including taking responsibility for their healthcare. 24–30-year-olds were also included because of the impact cancer can have on several developmental and life-stage factors (e.g., identity development and cognitive development; Tutelman & Heathcote, 2020).
2) Completed active cancer treatment (e.g., chemotherapy), are in remission from any type of cancer and/or are receiving maintenance treatment (e.g., hormone therapy).	 6+ months post treatment, in remission and finished 'active' treatment to ensure FCR occurred post treatment. Individuals using maintenance treatments aimed at reducing relapse or managing related symptoms were included within this post active treatment bracket. Any type of cancer was included due to the exploratory nature of the research (controlled for during analysis).
3) Treated for cancer as a TYA (aged 13-24).	 To ensure inclusion of participants who may have been diagnosed as a child but continued to receive active cancer treatment as a TYA. Important due to the potentially long-term nature of cancer that some individuals experience.
Exclusion Criteria 1) Currently receiving active treatment for cancer.	• Including treatments actively aimed at removing or stopping a cancer (e.g., chemotherapy, radiotherapy, or surgery).
2) Intellectual or physical disabilities which would prevent participants from completing the questionnaires.	• Unable to complete the research questionnaires.

• Unable to complete the research questionnaires.

3) Insufficient English language ability precluding participants from completing the questionnaires.

Description of Measures Used

Construct	Measure	Number of Items and Administration	Reliability and Validity	Additional Information
Demographic/ Clinical Information	Demographic Information and Clinical Characteristics	 whether they are in education Clinical characteristics: cance treatments participants receiv 	er type and stage at diagnosis, durat	ion of cancer since diagnosis, what g active treatment, if participants were
Illness Perceptions	The Brief Illness Perception Questionnaire (BIPQ; Broadbent, Petrie, Main & Weinman, 2006).	 Nine-item scale developed to assess the cognitive and emotional representations someone holds about their illness. Rate a 0-10 Likert scale: e.g., 'no affect at all' to 'severely affects my life'. 	 Suggested to show good test–retest reliability. Concurrent validity with other relevant measures. Good predictive validity in terms of recovery. Good discriminant validity between conditions (Broadbent et al, 2015). This study: α = 0.77. 	 Each item captures a different aspect of someone's illness perceptions: 'illness consequences'; 'timeline'; 'personal control'; 'treatment control'; 'identity'; 'coherence'; 'emotional representation'; 'illness concern'. Participants completed the BIPQ when thinking of their cancer.
Fear of Cancer Recurrence	Fear of Cancer Recurrence Inventory - Short Form (FCRI-SF; Simard & Savard, 2009).	 Nine-item FCRI-SF is comprised of the 'severity' subscale from the FCRI full scale measure. Measures the presence and severity of intrusive thoughts associated with FCR. 	 Suggested to show strong correlation with the total FCRI score (r = .84). High internal consistency (α = 0.89). Good convergent validity with other measures of FCR 	 Short form version chosen to reduce question load. Clinical cut off suggested at ≥ 13.

		• Items rated on a 0-4 Likert scale: e.g., 'not at all' or 'never' to 'a great deal' or 'all the time'.	 (r = .5977; Simard & Savard, 2009). This study: α = 0.81. 	
Depressive Symptoms	PHQ-9 (Kroenke & Spitzer, 2002). Continuous variable.	 Nine-item measure of depressive symptoms. Maps closely with the DSM-IV's depressive symptom criteria (Kroenke & Spitzer, 2002). Statements rated on a 0-3 Likert scale: e.g., 'not at all' to 'nearly every day', with higher total scores suggesting more depressive symptoms. 	 Suggested to have good reliability (α = 0.89) and validity. Good sensitivity in measuring depressive symptoms (Kroenke, Spitzer & Williams, 2001). This study: α = 0.90. 	 Used commonly in clinical practice (e.g., within IAPT; Clark, 2011) Clinical cut offs in this research: minimal (≤4), mild (5-9), moderate (10-14), moderately severe depression (15-19), severe (≥20) depression (Permanente, 2016).
Anxiety Symptoms	GAD-7 (Spitzer et al, 2006).	 Seven-item measure of generalised anxiety symptoms. Statements rated on a 0-3 Likert scale: e.g., 'not at all' to 'nearly every day', with higher scores suggesting more anxiety symptoms. 	 Suggested to have good reliability (α = 0.92) and validity (Spitzer et al, 2006). This study: α = 0.92. 	 Used commonly in clinical practice (e.g., within IAPT; Clark, 2011). Clinical cut offs in this research: minimal (≤4), mild (5-9), moderate (10-14), severe (≥15) anxiety (Plummer et al, 2016).
Quality of Life	World Health Organization Quality of Life Assessment Brief (WHOQOL-	 26-item measure of quality of life (QOL). From the full-scale WHO-QOL-100 quality of life assessment. 	 Suggested to correlate highly (0.89 or above) with the WHOQOL-100 domain scores. Good discriminant validity, content validity, internal 	 Short form version chosen to reduce question load. Measures QOL across four domains: 'physical health', 'psychological', 'social

	BREF; Whoqol Group, 1998).	• Statements rated on a range of 1-5 Likert scales: e.g., 'very poor' to 'very good'.	 consistency, and test–retest reliability. This study: α = 0.93. 	relationships' and 'environment'.Provides an overall quality of life and general health score.
Functioning	Work and Social Adjustment Scale (WSAS; Mundt, Marks, Shear & Greist, 2002).	 Simple measure of functioning. Rates five areas of functioning on how impaired they are. Uses a 0-8 Likert scale: e.g., 'not at all' to 'very severely'. 	 Suggested to show good internal reliability (α = 0.70 to 0.94). Good test-retest reliability (0.73). This study: α = 0.83. 	• Sensitive to patient differences in disorder severity and treatment-related change.

Note: It is acknowledged as a limitation that measures were validated for participants aged 18+, and not 16+.

	Total (N=90)	
Age, mean (SD; range) years	22.42 (2.98; 16-30)	
Gender women, n (%)	65 (72.2)	
Ethnic Group, n (%)		
UK or Irish	82 (91.1)	
White (other)	1 (1.1)	
Indian	2 (2.2)	
Pakistani	3 (3.3)	
Asian (other)	2 (2.2)	
Marital Status, n (%)		
Single	61 (67.8)	
Married	1 (1.1)	
With a partner	28 (31.1)	
Highest Education Status, n (%)		
No qualification	2 (2.2)	
GCSE's	10 (11.1)	
A-Levels	37 (41.1)	
Undergraduate	30 (33.3)	
Postgraduate	5 (5.6)	
Other	6 (6.7)	
Current Occupation, n (%)		
School student	6 (6.7)	
University student	24 (26.7)	
Full time employment	27 (30)	
Part time employment	22 (24.4)	
Unemployed	11 (12.2)	
Recruited from, n (%)		
Charity	6 (6.7)	
Twitter	4 (4.4)	
Facebook	9 (10)	

Participant Demographic Characteristics

Instagram	48 (53.3)
Other	7 (7.8)
Missing	16 (17.8)

Participant Clinical Characteristics

	Total (N=90)
Type of cancer, n (%)	
Brain and other CNS tumours	4 (4.4)
Breast	1 (1.1)
Cervical	4 (4.4)
Germ Cell Tumours	4 (4.4)
Leukaemia	20 (22.2)
Lymphoma	42 (46.7)
Melanoma	1 (1.1)
Bone	2 (2.2)
Soft Tissue Sarcoma	3 (3.3)
Uterine Sarcoma	1 (1.1)
Testicular	2 (2.2)
Thyroid	2 (2.2)
Other	4 (4.4)
Stage at diagnosis, n (%)	
Stage 1	14 (15.6)
Stage 2	32 (35.6)
Stage 3	13 (14.4)
Stage 4	19 (21.1)
Other/unknown	12 (13.3)
Time from diagnosis to remission, n (%)	
Under 1 year	52 (57.8)
1-2 years	26 (28.9)
2-3 years	9 (10)
3-4 years	1 (1.1)
4-5 years	0 (0)
Over 5 years	2 (2.2)
Types of treatment received, n (%)	
Chemotherapy	84 (93.3)

Immunotherapy	9 (10)
Radiation therapy	29 (32.2)
Stem cell transplants	10 (11.1)
Surgery	36 (40)
Targeted therapy	2 (2.2)
Hormone therapy	6 (6.7)
Other	3 (3.3)
Length of time received active treatment,	
n (%)	
Under 1 year	58 (64.4)
1-2 years	24 (26.7)
2-3 years	5 (5.6)
3-4 years	2 (2.2)
4-5 years	0 (0)
Over 5 years	1 (1.1)
Age during treatment, n (%)	
13-24 years	87 (96.7)
\geq 12 years and between ages 13-24	3 (3.3)
Approx. when treatment finished, n (%)	
6-12 months ago	19 (21.1)
12-24 months ago	23 (25.6)
Over 24 months ago	48 (53.3)
Time in remission, n (%)	
Under 1 year	19 (21.1)
1-2 years	18 (20)
2-3 years	15 (16.7)
3-4 years	12 (13.3)
4-5 years	10 (11.1)
Over 5 years	16 (17.8)
Other physical health conditions, n (%)	
Yes	21 (23.3)
No	69 (76.7)

Note: Participants could choose multiple responses for types of treatment received. Additionally, participants had to state any other physical health conditions.

CHERRIES Checklist

Checklist for Reporting Results of Internet E-Surveys (CHERRIES)

×	Checklist Item	Explanation	Page
ල් ව Item category			
Design	Describe survey design	Describe target population, sample frame. Is the sample a convenience sample? (In "open" surveys this is most likely.)	6
IRB (Institutional Review Board) approval and informed consent process	IRB approval Informed consent	Mention whether the study has been approved by an IRB Describe the informed consent process. Where were the participants told the length of time of the survey, which data were stored and where and for how long, who the investigator was, and	14 14; supplementary material
F	Data protection	the purpose of the study? If any personal information was collected or stored, describe what mechanisms were used to protect unauthorized access.	15; supplementary material
Development and pre-testing	Development and testing	State how the survey was developed, including whether the usability and technical functionality of the electronic questionnaire had been tested before fielding the questionnaire.	Supplementary material
Recruitment process and description of the	Open survey versus closed survey	An "open survey" is a survey open for each visitor of a site, while a closed survey is only open to a sample, which the investigator knows (password protected survey).	15
sample having access to the questionnaire	Contact mode	Indicate whether or not the initial contact with the potential participants was made on the Internet. (Investigators may also send out questionnaires by mail and allow for Web-based data entry.)	15
	Advertising the survey	How/where was the survey announced or advertised? Some examples are offline media (newspapers), or online (mailing lists – If yes, which ones?) or banner ads (Where were these banner ads posted and what did they look like?).	15
		It is important to know the wording of the announcement, as it will heavily influence who chooses to participate. Ideally the survey announcement should be published as an appendix.	
Survey administration	Web/E-mail	State the type of e-survey (eg, one posted on a Web site, or one sent out through e-mail). If it is an e-mail survey, were the responses entered manually into a database, or was there an automatic method for capturing responses?	15
	Context	Describe the Web site (for mailing list/newsgroup) in which the survey was posted. What is the Web site about, who is visiting it, what are visitors normally looking for? Discuss to what degree the content of the Web site could pre-select the sample or influence the results. For example, a survey about vaccination on a anti- immunization Web site will have different results from a Web survey conducted on a government Web site	15

Mandatory/voluntary	Was it a mandatory survey to be filled in by every visitor who wanted to enter the Web site, or was it a voluntary survey?	15
Incentives	Were any incentives offered (e.g., monetary, prizes, or non- monetary incentives such as an offer to provide the survey results)?	15
Time/Date	In what timeframe were the data collected?	10
Randomization of items or questionnaires	To prevent biases items can be randomized or alternated.	N/A
Adaptive questioning	Use adaptive questioning (certain items, or only conditionally displayed based on responses to other items) to reduce number and complexity of the questions.	15; supplementary material

	Number of Items	What was the number of questionnaire items per page? The number of items is an important factor for the completion rate.	Supplementary material
	Number of screens (pages)	Over how many pages was the questionnaire distributed? The number of items is an important factor for the completion rate.	Supplementary material
	Completeness check	It is technically possible to do consistency or completeness checks before the questionnaire is submitted. Was this done, and if "yes", how (usually JAVAScript)? An alternative is to check for completeness after the questionnaire has been submitted (and highlight mandatory items). If this has been done, it should be reported. All items should provide a nonresponse option such as "not applicable" or "rather not say", and selection of one response option should be enforced.	15
	Review step	State whether respondents were able to review and change their answers (e.g., through a Back button or a Review step which displays a summary of the responses and asks the respondents if they are correct).	15
Response rates	Unique site visitor	If you provide view rates or participation rates, you need to define how you determined a unique visitor. There are different techniques available, based on IP addresses or cookies or both.	15; supplementary material
	View rate (Ratio of unique survey visitors/unique site visitors)	Requires counting unique visitors to the first page of the survey, divided by the number of unique site visitors (not page views!). It is not unusual to have view rates of less than 0.1 % if the survey is voluntary.	N/A
	Participation rate (Ratio of unique visitors who agreed to participate/unique first survey page visitors)	Count the unique number of people who filled in the first survey page (or agreed to participate, for example by checking a checkbox), divided by visitors who visit the first page of the survey (or the informed consents page, if present). This can also be called "recruitment" rate.	17
	Completion rate (Ratio of users who finished the survey/users who agreed to participate)	The number of people submitting the last questionnaire page, divided by the number of people who agreed to participate (or submitted the first survey page). This is only relevant if there is a separate "informed consent" page or if the survey goes over several pages. This is a measure for attrition. Note that "completion" can involve leaving questionnaire items blank. This is not a measure for how completely questionnaires were filled in. (If you need a measure for this, use the word "completeness rate".)	17

Preventing multiple entries from the same individual	Cookies used	Indicate whether cookies were used to assign a unique user identifier to each client computer. If so, mention the page on which the cookie was set and read, and how long the cookie was valid. Were duplicate entries avoided by preventing users access to the survey twice; or were duplicate database entries having the same user ID eliminated before analysis? In the latter case, which entries were kept for analysis (e.g., the first entry or the most recent)?	15; supplementary material
	IP Check	Indicate whether the IP address of the client computer was used to identify potential duplicate entries from the same user. If so, mention the period of time for which no two entries from the same IP address were allowed (e.g., 24 hours). Were duplicate entries avoided by preventing users with the same IP address access to the survey twice; or were duplicate database entries having the same IP address within a given period of time eliminated before analysis? If the latter, which entries were kept for analysis (e.g., the first entry or the most recent)?	15
	Log file analysis	Indicate whether other techniques to analyse the log file for identification of multiple entries were used. If so, please describe.	NA
	Registration	In "closed" (non-open) surveys, users need to login first and it is easier to prevent duplicate entries from the same user. Describe how this was done. For example, was the survey never displayed a second time once the user had filled it in, or was the username stored together with the survey results and later eliminated? If the latter, which entries were kept for analysis (e.g., the first entry or the most recent)?	NA
Analysis	Handling of incomplete questionnaires	Were only completed questionnaires analysed? Were questionnaires which terminated early (where, for example, users did not go through all questionnaire pages) also analysed?	15
	Questionnaires submitted with an atypical timestamp	Some investigators may measure the time people needed to fill in a questionnaire and exclude questionnaires that were submitted too soon. Specify the timeframe that was used as a cut-off point and describe how this point was determined.	15
	Statistical correction	Indicate whether any methods such as weighting of items or propensity scores have been used to adjust for the non- representative sample; if so, please describe the methods.	N/A

Regression Assumptions

Assumption	Description of Checks Conducted/Evidence	Assumption met?
1. Dependent variable measured	GAD-7 and PHQ-9 total scores	/
on a continuous scale.	are continuous scales.	\checkmark
2. Presence of two or more independent variables.	Gender, Age, Illness perceptions and FCR.	\checkmark
3. Independence of observations	Durbin-Watson Test completed and between the normal range of 1.5 - 2.5.	\checkmark
4. There is a linear relationship between (a) the dependent variable and each of your independent variables, and (b) the dependent variable and the independent variables collectively.	Visual inspection of scatterplots confirmed linearity.	
5. Data shows homoscedasticity.	Graph of homoscedasticity generated on SPSS – for both anxiety and depression regressions, variances along the line of best fit remain similar as you move along the line.	\checkmark
6. Data does not show multicollinearity	Test for multicollinearity included within SPSS regressions. - For both Anxiety and Depression variables: all VIF values between 1-10 (indicating no multicollinearity).	\checkmark
7. There are no significant outliers, high leverage points or highly influential points.	Visual inspection of histograms confirmed that there were no data points falling away at the extremes.	\checkmark

