

An exploration of the factors that associate with psychological outcomes in
chronic illness and Inflammatory Bowel Disease populations

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A thesis submitted in partial fulfilment of the requirements for the Doctorate in Clinical
Psychology

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Submission date: June 2020

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Declaration

This thesis has not been submitted for any other qualification at the University of Sheffield or any other institution

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Structure and word counts

Section 1: Literature review

Excluding references and tables
7981

Including references and tables
13,712

Section 2: Research report

Excluding references and tables
7999

Including references and tables
12,670

Total word count

Excluding references and tables
15,980

Including references and tables
26,382

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Overall abstract

Chronic illnesses, including Inflammatory Bowel Disease (IBD), present individuals with an array of physical and psychological challenges. To successfully address psychological distress or foster resilience in those living with chronic illness, it is important to understand the factors that underpin such outcomes. This research aimed to: 1) understand the association between illness uncertainty and psychological distress in those with chronic illness, and; 2) develop a theoretical and empirically informed understanding of resilience in those with IBD.

A systematic review and narrative synthesis examined the association between illness uncertainty and psychological distress in people with chronic illnesses. A systematic search of three electronic databases yielded 23 studies. Specific inclusion/exclusion criteria were applied. All studies used self-report measures and all but one study, employed a cross-sectional design. Overall, low to moderate levels of illness uncertainty were present across the study samples. The synthesis illuminated the association between illness uncertainty and psychological distress (measured as depression, anxiety, negative mood states and trauma symptoms). This association remained regardless of individual or contextual factors, time across the illness trajectory; and when confounding variables were controlled for. Appraisals of illness uncertainty need further exploration, but studies have begun examining control-beliefs and intolerance of uncertainty. Limitations of the studies are discussed and further recommendations for future research are presented.

Recent research in the IBD field has begun exploring beliefs about illness and disease-related variables and their association with resilience and other outcomes. This exploratory research employed a cross-sectional, sequential mixed methods design. Therefore, findings from a scoping literature search and qualitative phase, eliciting

personal, voiced resilience experiences via interviews, informed a quantitative examination of factors and their association with resilience, using an online survey. Those over 18 years and with self-reported IBD diagnoses participated in the study. Five people participated in interviews. Potential factors comprising an important part of resilience were identified from the scoping review. The thematic analysis of the interview data supported these and the construct of grit additionally emerged (perseverance towards long-term goals). Next, eighty-five participants completed an online survey to test measures of social support, coping-efficacy and illness-acceptance (illness cognitions and coping resources); control-beliefs and intolerance of uncertainty (beliefs related to controllability); grit; time since diagnosis, disease activity and disease subtype (disease related variables); and their association with resilience (dependent variable).

Correlational analyses and a hierarchical regression analysis were conducted. Disease activity, illness-acceptance, social support, coping efficacy, control-beliefs and intolerance of uncertainty contributed 67.1% of the variance in resilience. Non-significant associations were found between grit, time since diagnosis and resilience. Daily persistence and endurance may be more characteristic of resilience in those with IBD. Limitations of the study are acknowledged, and recommendations for future clinical practice and research are discussed.

Acknowledgements

Thank-you to those who gave their time to participate in this research and to my research supervisors Dr Georgina Rowse, Dr Rebecca Yeates and Dr Fuschia Sirois for your guidance, expertise and steering me back on track when I lost my way.

My thanks also go to my family and friends. Not only did your encouragement and unwavering belief in me drive me to undertake clinical training under much less than favourable circumstances, but your emotional and practical support made the challenges and emotional rollercoaster of clinical training possible. To my life long best friends, my sisters; Anna, my academia guru; Claire, my guru in many other challenging life matters; you're both inspirational; my parents- for so much support, for being you, and wonderful grandparents; my friends - my chosen family, for so much, for being Sienna's extended family and for the much needed fun times; to Sandra for keeping me healthy and Charalene, because ashtanga yoga has kept me physically and emotionally grounded. Combined, you are my resilience and without you all this would not have been possible.

Lastly for my daughter, Sienna, for reminding me of what really matters, for helping me compartmentalise so that this work did not consume me, for your boundless love, joy, acceptance, and making sure there has never been a dull moment. This was for us.

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Section 1: Literature review

How does illness uncertainty relate to psychological distress in chronic illness populations? A systematic narrative review

Abstract

Objectives

Illness uncertainty (IU) arises when illness related events have indeterminable meaning and adequate cognitive schema cannot be formed to interpret the meaning of such phenomena. IU has been associated with negative outcomes. This review aimed to synthesise papers examining the IU/psychological distress association in chronic illness populations; to understand it in relation to care-context, chronic illness type or form of psychological distress. This intended to draw conclusions to inform future research and clinical practice.

Method

Three electronic databases were systematically searched for studies that met pre-defined inclusion criteria, including use of an IU measure based on Mishel's conceptualisation. The 'Observational Cohort and Cross-sectional Studies' tool was employed to assist quality appraisal and papers were narratively synthesised.

Results

The search yielded 23 papers (total participants, N=3126). Twenty-one studies demonstrated positive and significant associations between IU and psychological distress irrespective of gender, care-context, time across the illness trajectory, chronic illness type or form of psychological distress. The ambiguity element of Mishel's IU conceptualisation yielded more significant associations and larger effect sizes than other facets of IU (complexity, inconsistency, and unpredictability). There was a tendency for greater IU to associate with subjective rather than objective illness severity. Intolerance of uncertainty and control beliefs as appraisal processes associated with IU and psychological distress.

Conclusions

IU associates with psychological distress regardless of individual or contextual differences. It is inferred from existing theoretical frameworks that appraisals mediate the association, but research is needed to explore causal pathways and specific appraisal processes linked with IU in chronic illness populations.

Practitioner points

- The presence of IU is ubiquitous in chronic illness populations and should be routinely assessed and normalised.
- Healthcare professionals should assess and target the different facets of IU using Mishel's original measure and varying manifestations of psychological distress in those with chronic illnesses.
- Healthcare professionals should follow current National Institute for Health and Care Excellence guidance on the psychological treatment of emotional disorders (cognitive behavioural therapy) but assess for appraisal processes associated with IU.

Limitations

- The cross-sectional designs employed meant that causal associations could not be determined.
- Findings were not generalisable to all chronic illness populations and 65.2% of studies were conducted in the United States of America, limiting the cross-cultural validity of the findings.
- Few studies conducted power analyses; thus, it was difficult to determine if studies were sufficiently powered to avoid type 1 or II errors. Therefore, findings should be interpreted with caution.

Illness uncertainty: An overview of definitions and models

Uncertainty has been defined as a complex cognitive stressor and perceptual state or attitude of doubt or not knowing that changes over time (Mast, 1995; Wiener & Dodd, 1993). It is an ubiquitous human experience but is particularly poignant for those living with chronic illness (CI, Hilton, 1994; Mishel, 1990), because they present individuals with prolonged physical and psychological challenges. One potential challenge is IU which occurs when “adequate cognitive schema cannot be formed with which to interpret the meaning of illness-related events” (Mishel, 1997, p.225). Insufficient coping to buffer negative emotional responses can lead to protracted or unresolved psychological distress which can precede the need for clinical services. It is thus the focus of this review to develop an understanding of the association between IU and psychological distress in those living with CI.

IU has been conceptualised in many ways. One such conceptualisation suggests its synonymity to emotions evoked by illness-related events that can be positive or threatening (Hilton, 1994). Conversely, in the uncertainty in illness theory (UIT, Mishel, 1981), IU is proposed to be a “neutral cognitive state” (Mishel, 1997, p.58) distinct from its emotional outcomes (McCormick, 2002). The UIT has become the most established and researched framework and differing versions of measures based upon Mishel’s conceptualisation have been cited as the most widely used (Wright et al., 2009). The original Mishel Uncertainty in Illness Scale-Acute (MUIS-A, Mishel, 1981) captures four facets of IU. These include *ambiguity*; meaning information (symptoms or causes) can be vague and interpretable in several ways: *complexity*; uncertainty about treatment and the medical system; *inconsistency*; the symptom pattern presenting as variable and in discord with the disease process; and *unpredictability*, the inability to predict future course; such as about relapsing/remitting symptoms or prognosis (Mishel, 1988).

Contentions underpin the presumed causes and influencing factors of IU. Hilton (1992) suggested that lack of information underlies uncertainty, whereas Mishel (1997) placed emphasis on indeterminable meaning which comprises a multitude of factors, acknowledging IU is a multifaceted concept (McCormick, 2002). Four major components of IU are proposed, including: antecedents generating uncertainty; the appraisal of uncertainty, coping and affect control strategies; and adaptation to the illness. Poor cognitive abilities, low education levels and limited social support have also been associated with high levels of IU in CI populations (Liao et al., 2008; Lien et al., 2009; Mast, 1998). Thus, contextual, and individual factors influence IU.

Mishel's (1981) uncertainty in illness concept and theoretical underpinnings.

The UIT drew upon Lazarus and Folkman's (1984) stress and coping model, which illuminated the importance of the appraisal generated from uncertainty. Appraisal processes and the ways in which individuals cope with IU are variable (Alschuler & Beier, 2015). IU is not necessarily aversive until negatively appraised. Negative appraisals include thoughts related to danger (a threat to well-being) or hopelessness (Mishel, 1990). Mishel (1981, 1984) postulates that adaptive, promising outcomes are borne out of positive appraisals, such as that illness experiences provide opportunity (Mishel, 1981; Mishel, 1984). Conversely negative appraisals lead to the adoption of maladaptive coping strategies to manage affect, leading to outcomes of psychological distress (Mishel, 1988).

The UIT was originally applied to acute illness experiences (Mishel, 1988), but reconceptualised to address the phenomenology of continuous uncertainty in those with CI (Mishel, 1990). Within the theory it is suggested that living with IU chronically, destabilises pre-existing cognitive models of life as predictable and controllable, and thus IU becomes accepted as part of reality (Alligood, 2014). There is henceforth a shift

of perspective from danger to opportunity which may lead to positive psychological changes (Mishel, 1990). However, this has been critiqued because how meaning is assigned to IU and transformed to opportunity appraisals is an idiosyncratic process that lacks empirical backing (Alligood, 2014). Furthermore, Bailey and Neilson (1993) found no association between illness duration (mean duration was 17 years), IU, and its appraisal in those with rheumatoid arthritis.

The UIT shares the conceptual underpinnings of Beck's (1967) cognitive theory of emotional disorders arguing that appraisals (about self, world, and future) are pertinent to the development of psychopathology; thoughts/appraisals impact on emotions which influence coping behaviours. Hence, appraisals of uncertainty in the context of a CI, may lead to undesirable outcomes. Intolerance of uncertainty which is defined as negative reactions to uncertainty on emotional, cognitive, and behavioural levels (Buhr & Dugas, 2009) has been extensively researched. Intolerance of uncertainty has been conceptually and empirically linked with generalised anxiety disorder (Dugas et al., 1998; Dugas & Robichaud, 2007; Freeston et al., 1994) and other emotional disorders (Gentes & Ruscio, 2011).

Empirical support for Mishel's (1981) uncertainty in illness theory.

It has been argued that much of the research supporting the UIT dedicated its focus to the association between IU and psychosocial outcomes rather than the mediating roles of appraisals (McCormick, 2004). Hence, only extant research scaffolds the IU, appraisal link. For example, higher uncertainty associates with danger appraisal (Bailey & Neilsen 1993). The extensive research on IU and outcomes has been conducted with varying populations, including paediatrics (Fortier et al, 2013) and chronic pain (Wright et al., 2009). A large tranche of empirical research on acute and CI populations, links IU with reduced Quality of Life (QOL, Chen et al., 2018; Fedele

et al., 2009; Padilla, 1992), lower adjustment (Christman, 1990; Mishel & Baden, 1987), negative mood states (Christman et al., 1988; Lin et al., 2013; Lutze & Archenholtz, 2007; Wineman, 1990; Wineman et al., 2003), poorer emotional well-being and depressive symptoms (Bailey et al., 2009; Bang et al., 2013; Hoth et al., 2013; Mishel, 1981; Wang et al., 2014). Accordingly, higher levels of certainty have correlated with better QOL and psychosocial adaptation (McNulty et al., 2004; Niv et al., 2017).

Some internal resources have been found to mediate between IU and outcomes, including optimism (Christman, 1990; Mishel et al., 1984; Mishel & Sorenson, 1991), hope (Christman, 1990; Hilton, 1994), mastery (Mishel et al., 1991) and learned resourcefulness (Rosenbaum, 1983). Furthermore ‘grit’, or persistence despite challenge (Duckworth et al., 2007), has been linked with lower IU and thus decreased distress (Sharkey et al., 2017).

Chronic Illnesses

The Department of Health (DOH, 2017) defines CIs as “complex... multiple causes... generally long-term and persistent, and often lead to a gradual deterioration of health and loss of independence. While not usually immediately life threatening, chronic conditions are the most common and leading cause of premature mortality” (p.6). Moreover, they are not passed from person and person and are of long duration with generally slow progression (World Health Organisation, WHO, 2016). CIs range from medication-controlled asthma or diabetes; to neurodegenerative diseases such as Multiple Sclerosis or Parkinson’s disease which present individuals with significant experiences of pain and the threat of unpredictable but gradual decline. Though distinct CI’s vary in their clinical profile, they share common characteristics, including uncertain aetiology, symptom ambiguity, uncontrollable or fluctuating trajectories and

uncertain prognosis. Uncertainty can be exacerbated by the ever-present potential for decline in health functionality (Mishel, 1990).

Psychological distress

Psychological distress is frequently referenced in health-care literature, but as a distinct concept, it has not been clearly defined and articulated (Ridder, 2004). It is conceptually linked with stress and distress, considered the “unique discomforting, emotional state experienced by an individual in response to a specific stressor” (Ridder, 2004, p.539). It has also been characterised by symptoms of depression and anxiety (Drapeau et al., 2012), which have been named as leading causes of morbidity and disability (Prince et al., 2011). Those with CI’s have higher rates of depression and anxiety than physically healthy controls and approximately 20% of those with a CI have depression (NICE, 2010). Existing systematic reviews have measured psychological distress multidimensionally (Gong et al., 2016; Kuswanto et al., 2018), capturing depression, anxiety, trauma symptomatology, mood states and perceived stress symptoms. Research included in this review captures these wider measures of psychological distress.

The current review

Existing reviews have focused on IU and outcomes in specific populations; including caregiver and paediatric uncertainty in relation to young people with CI (Szulxzewski et al., 2017) and chronic pain (Wright et al., 2009). Szulxzewski et al. (2017) conducted a meta-analysis and found associations between child and caregiver uncertainty and outcomes of depression, anxiety, and psychological distress in young people, with medium effect sizes. Wright et al’s. (2009) concept review was not systematically conducted which may have introduced several biases. However, they found IU to be associated with maladaptive coping and higher psychological distress. A

plethora of individual quantitative studies that have examined the IU and psychological distress association in adult CI populations exist, however no study has synthesised findings.

This timely review carries clinical and empirical importance. A synthesis offers a nuanced understanding of the association between IU and psychological distress according to CI-type and care-context. Furthermore, the NICE (2010, guideline 91) recommended cognitive behaviour therapy treatment of depression is based on a standardised manual, rather than being tailored to addressing the psychological processes associated with CI. A nuanced understanding of IU, psychological distress and associated variables could facilitate an understanding of what psychological processes health care professionals (HCP's) might assess and target in CI populations; and highlight those more vulnerable to moving towards a trajectory of negative outcomes. This could be valuable for those working in primary care, hospital settings, health, and psychology departments and psychological therapists.

The review question was developed using the PICO (population, intervention, comparator, and outcome) framework (Moher et al., 2015), culminating in the final review question: How does illness uncertainty relate to psychological distress in chronic illness populations?

This review had five aims: 1) Examine the commonality of IU and the general trend across all studies examining the association between IU and psychological distress across CI populations; 2) consider the association between IU and psychological distress across differing care-contexts; 3) examine the association between IU and varying types of psychological distress; 4) elucidate how this association might differentiate across CI types; 5) identify other variables that have been examined in relation to IU.

Method

Design

The review protocol was registered with the international Prospective Register of Systematic Reviews (PROSPERO, Registration number: CRD42020166475). This review followed the ‘Preferred Reporting Items for Systematic Reviews and Meta-Analyses’ (PRISMA; Moher et al., 2009) guidance. Systematic narrative reviews involve methodically selecting, summarising and critically appraising available research (Moher et al., 2009). In line with narrative synthesis guidance (Popay et al., 2006) a preliminary synthesis of study findings was conducted, and patterns elicited. Next the studies were summarised, and consideration given to the direction and magnitudes of effects. Subsequently, exploration of the similarities, differences and relationships within the data was undertaken, accounting for quality appraisal (Lisy & Poritt, 2016).

Search strategy

Three electronic databases, Scopus, Psycinfo and MEDLINE (all titles, abstracts, keywords) were searched on 20th February 2020, from database inception, with no date restrictions. This enabled the search to capture and synthesise all available research. Only English language papers were included. Search concepts were developed using PICO (Moher et al., 2015); no search concepts were included regarding comparator or study design. Search terms were developed (See Appendix A) from preliminary scans of the literature, thesaurus searching the key constructs and informed by previous reviews studying CI populations (Amo-Setien et al., 2019; Cal et al., 2015; Snippen et al., 2019; Szulczewski et al., 2017). Search-terms included “chronic* ill*”, illness uncertainty”, “Mishel* illness uncertainty scale” and “psychological distress*”.

Boolean operators were utilised to connect terms and combine specificity and sensitivity to yield eligible studies (meeting the inclusion/exclusion criteria).

A PRISMA diagram is presented in figure 1 and provides a summary of the search and selection process (Moher et al., 2009). Where full text articles were not accessible, main authors were contacted which enabled all papers to be successfully obtained. To enable a comprehensive search, reference lists of identified papers elicited by the above systematic search were hand searched; one additional paper was identified and included in the final review using this method (Detprapon et al., 2004). A forward and backward citation search was conducted on all included papers; two papers were identified which were unavailable in English language and thus excluded. Screening resulted in duplicates being removed initially and eligibility subsequently being ascertained by title, and then abstract inspection. If it was unclear if papers met inclusion/exclusion criteria, full articles were inspected.

Inclusion/exclusion criteria

Papers were included according to inclusion and exclusion criteria (see Appendix B). Papers needed to include a CI or form of psychological distress cohering with the definitions discussed in this review (DOH, 2017; Ridder, 2004; WHO, 2016). They were also required to employ an IU measure based on Mishel's conceptualisation so that there was some homogeneity pertaining to what constituted IU in terms of definition and measurement (e.g. Hagen et al., 2015; Mishel 1981; Mishel, 1986).

Where it was unclear if a paper met inclusion, this was reconciled through discussions with research supervisors, with consideration of the papers content and relevance to the review question. Illustratively, two papers were included studying survivors of childhood cancer (Lee, 2006; Santacroce & Lee, 2006). The key rationale for inclusion was that recurrent disease has been found to be 4.4%, 5.6%, and 6.2% at

10, 15, and 20 years post-paediatric cancer survival respectively (Wasilewski-Masker et al., 2009); survivor death rates have been found to exceed age-matched controls, with death from subsequent cancer being the most frequent cause (Mertens et al., 2015). Exclusion-criteria included IU measurement pre-post major surgery or drug trial participation as this was deemed to capture something phenomenologically distinct. However, papers were included for those with a CI undergoing protracted treatments such as haemodialysis (e.g. B. Kim & J. Kim, 2019) or chemotherapy (e.g. Kurita et al., 2013).

Measures of illness uncertainty using Mishel's conceptualisation of IU

Mishel (1983) advocated the development of measures for specific illness and diagnostic groups. Hence several versions have been developed since the original 33-item MUIS-A (Mishel, 1981). The measures employed by papers in this review are described. All measures have demonstrated robust psychometric properties (See Appendix C for psychometric properties).

The MUIS-A measures all facets of IU thus containing subscales of ambiguity, complexity, inconsistency, and unpredictability. It includes questions related to acute illness and hospitalisation. A 28-item two factor MUIS version (Mishel, 1997) including subscales ambiguity (16 items) and complexity (12 items) was subsequently developed. The 23-item Mishel Uncertainty in Illness Scale-Community (MUIS-C, Mishel, 1991) was adapted from the MUIS-A and thus contains similar items. However, those items related to hospitalisation of acutely ill patients were removed; it was designed for community-dwelling individuals, unlikely to be undergoing medical interventions and differed in its use of a unidimensional scale. B. Kim & J. Kim (2019) used the Korean translation (Jung et al., 2005). The 5-item Mishel Uncertainty in Illness Scale-Short

Form (MUIS-SF, Hagen et al., 2015) captures ambiguity about and controllability of the illness; and complexity of treatment/system of care as a unitary construct.

Data extraction

The following data were extracted and tabulated (see Table 1): author(s), publication year, country of publication, sample characteristics (CI, sample size, percentage female, age ranges, mean age), recruitment setting, study design, sampling method, measures of IU and psychological distress adopted by each study, and finally the correlation coefficients (r) between the IU and psychological distress measure; which indicated the effect size (Ellis, 2010). Effects sizes were interpreted according to Cohen's (1988) guidelines; $r=0.10$ to 0.29 (small), $r=0.30$ to 0.49 (medium) and $r=0.50$ to 1.00 (large). Where r -square value was used Cohen's (1992) guidance was followed; <0.12 (low association with outcome), 0.13 to 0.25 (medium); and >0.26 (large).

In papers where the association between IU and psychological distress was not the predominant focus, bivariate correlational findings (with correlation coefficients) were extrapolated. Where possible, partial correlations were also reported.

Quality appraisal

The quality of papers included in a review can impact on the validity of its results (Centre for Reviews & Dissemination, 2008). Assessment of study quality was conducted using the 14-item Observational Cohort and Cross-sectional Studies tool (National Institutes of Health, 2014, see Appendix D for items). A checklist is provided allowing for analysis of transparency, consistency, and validity; enabling papers to be rated as 'poor', 'fair', or 'good'. Items are rated as 'yes', 'no', 'cannot-determine', 'not reported' or 'non-applicable'. No guidelines offer categorisation of numerical ratings and some items were not applicable to the study design. Thus, for the purpose of this review, non-applicable items were excluded from the scoring and a percentage rating

was calculated to facilitate comparisons of study quality (total score/applicable items x 100). The author applied the principles that <50%= poor; 50%-70%=fair and >70%=good. Table 1 presents percentage and quality ratings.

An independent final-year clinical psychology doctoral trainee with experience of quality appraisal and who was blind to the main author's ratings, rated 5 randomly selected papers. This intended to confirm inter-rater reliability of the quality assessment. Any disparities in ratings were discussed until a consensus was reached (the final agreed ratings are presented in Table 1). Overall, the quality of the studies included in this review was deemed fair (N=15) or good (N=8) (see Appendix E). No studies received a 'poor' rating. Discussion of individual aspects of study quality are additionally embedded within the review findings.

Results

In total the systematic search culminated in the inclusion of 23 studies (see figure 1). Table 1 summarises the key study characteristics and extracted data from the reviewed studies.

Figure 2

PRISMA diagram (PRISMA; Moher et al., 2009)

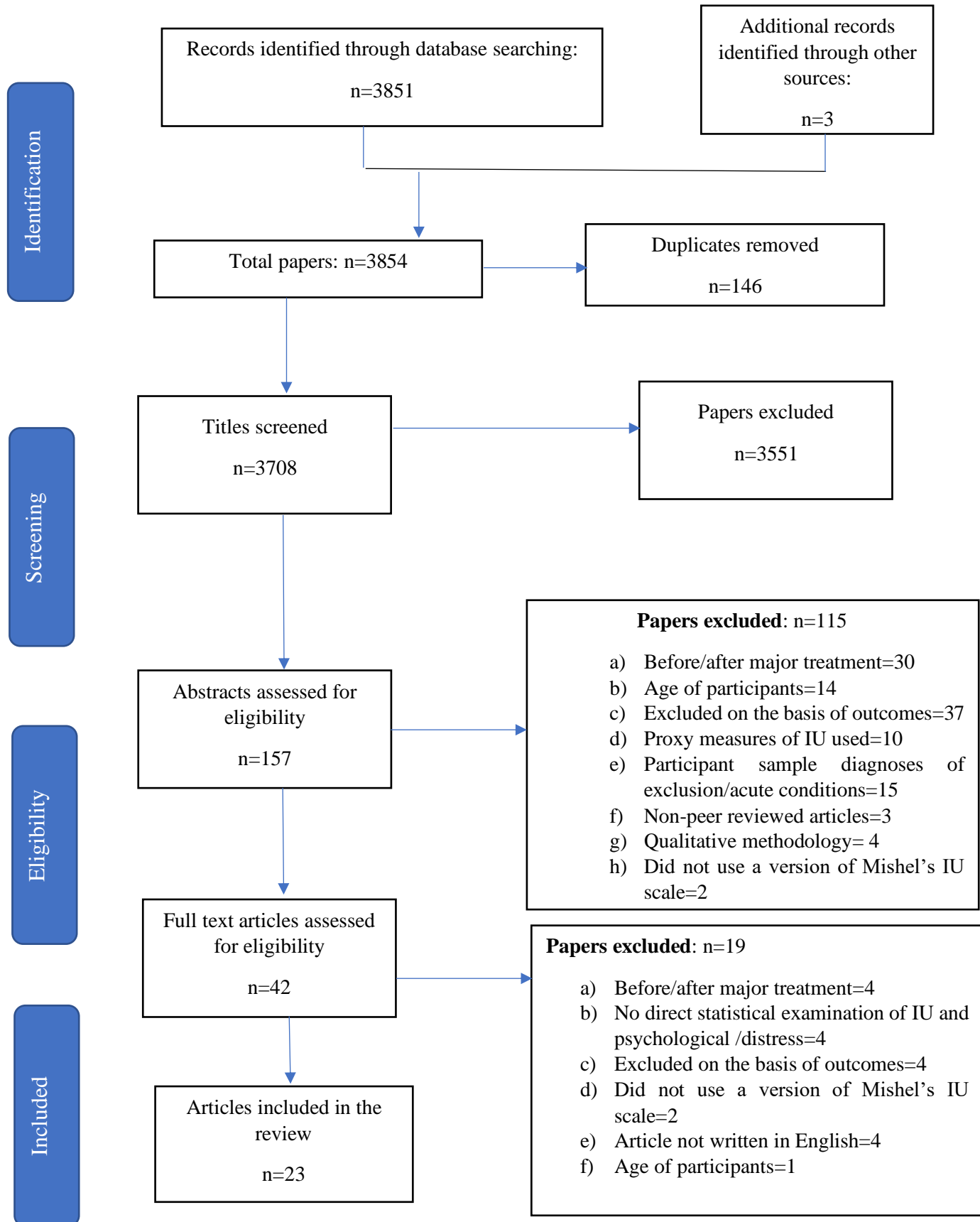


Table 1*Study characteristics*

Author (year) Country	Chronic Illness	Sample characteristics Mean age (years, standard deviation, if reported) Age ranges, % female	Study design	Outcome variables (and measure used)	MUIS Measure	Correlation coefficient	Significant association	Quality score (%) and rating
Ahn et al. (2017) South Korea	Parkinson's disease Outpatient	120 participants from a neurology outpatient department Mean age= 65.01 (9.08) Female= 49.2%	Cross- sectional	Depression (SF-GDS)	MUIS-A (Korean version, 33 items)	Bivariate r=0.46***	Y	70 Good
Bailey et al. (2009) England	Hepatitis C Outpatient	126 patients undergoing watchful, waiting protocol- medical centre Mean age = 53.1 (9.4) Range= 27-78 Female= 50.8%	Cross- sectional	Depression (CES-D)	MUIS-A (33- item)	Bivariate: Ambiguity: r=0.51** Complexity: r=0.23* Inconsistency: r=0.39** Unpredictability: r=0.06 Partial: r=0.36** (ambiguity subscale only)	Y Y Y N Y	72.7 Good
Barberis et al. (2019) Italy	Chronic kidney disease Inpatient	50 patients enrolled at the unit of nephrology of a hospital Female= 26%	Cross- sectional	Depression and anxiety (HADS)	MUIS-A (33- item)	Bivariate Depression r=0.40*** Anxiety r=0.54***	Y Y	54.5 Fair

Author (year) Country	Chronic Illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables	MUIS Measure	Correlation coefficients	Significant association	Quality score (%) and rating
Carpentier et al. (2007). America	Childhood onset asthma Community	121 college students from a large midwestern public university Mean age = 19.7(1.62) Range=18-22 Female= 62.8% 121 healthy control subjects	Cross-sectional	Psychological distress/anxiety and depression (BSI) Illness-induced interference (IIRS)	MUIS-C	Hierarchical regression analysis IU significantly predicted depression, anxiety and full BSI (p<0.001)	Y	80 Good
Colagreco et al. (2014). America	Chronic Hepatitis C Outpatients	92 participants recruited from a single centre, following watchful waiting protocol. Mean age=56.10(7.40) Range=24-74 Female= 35.9%	Cross-sectional	Depression (CES-D)	MUIS-A (33-item)	Bivariate: Total MUIS: r=0.49** Ambiguity: r=0.51** Inconsistency: r=0.36** Complexity: r=-0.07 Unpredictability: r=-0.01	Y Y Y N N	50 Fair
Detprapon et al. (2009) Thailand	Head and neck cancer Outpatients	240 head and neck cancer patients recruited from an outpatient clinic Mean age=55.17 Range= 19-89 Female=29.6%	Cross-sectional	Depression (CES-D) Symptom experience (MSES)	MUIS-C	Bivariate r=0.82***	Y	55.6 Fair

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS Measure	Correlation coefficient	Significant association	Quality score (%) and rating
Hommel et al. (2003)	Childhood onset asthma	56 participants were recruited from undergraduate classes at a large midwestern university.	Cross sectional	Depression (IDD)	MUIS-C	Bivariate Depression r=0.48**	Y	75 Fair
America	Community	Mean age = 19.11(0.97) Range= 18-21 Female=48.2%		Anxiety (BAI)		Anxiety r=0.56**	Y	
Hoth et al. (2013)	COPD Outpatients	407 people with alpha-1 antitrypsin deficiency-associated COPD (From Alpha-1 Foundation Research Registry) Mean age=59.6(9.5) Range= 32-84 Female= 47.9%	Prospective correlational	Illness severity (Illness severity assessment) Depression and anxiety (HADS)	MUIS-A (28-item, measuring ambiguity and complexity)	Partial Depression r=0.19 Anxiety r=0.34* Linear mixed models, controlling for demographic and illness related variables.	N Y	54.5 Fair
America						Ambiguity: b=0.13*** Complexity: b=-0.01	Y N	
B. Kim & J. Kim (2019)	End-stage renal disease	152 patients receiving hemodialysis across 5 hospitals	Cross-sectional	Depression (PHQ-9)	MUIS-A (33-item) Korean version	Bivariate r=0.31***	Y	60 Fair
Korea	Inpatients	Mean age=53.28(11.17) Range=20-79 Female=36.2%		Perceived social support (MSPSS)				

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS Measure	Correlation coefficient	Significant association	Quality score (%) and rating
Kurita et al. (2013) America	Lung cancer Outpatients	49 participants diagnosed with lung cancer at least 6 months prior to enrolment Mean age=64.2(11.0) Range=37-86 Female=71.4%	Cross-sectional	Depression (CES-D) Intolerance of uncertainty (IUS)	Ambiguity subscale of MUIS	Bivariate b=0.18	N	70 Good
Lee (2006) Taiwan	Childhood survivors of cancer Community	45 participants from a pediatrics department database Mean age=27.4(5.54)	Cross-sectional	Post-traumatic stress symptoms (PTSDI)	MUIS-C	Bivariate r=0.40*	Y	60 Fair
Lemaire (2004) America	Endometriosis Community	Female=62.25% 298 women attending an endometriosis conference Mean age=34(7.10) Female=100%	Cross-sectional	Emotional distress (FARS)	MUIS-C	Bivariate r=0.48**	Y	60 Fair

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS Measure	Correlation coefficient	Significant association	Quality score (%) and rating
Lin et al. (2013)	Primary brain tumours (PBT's)	186 patients with PBT's from a cancer centre and brain and spine Centre Clinic Mean age=44.2 Range=19-80 Female=46.8%	Cross-sectional	Mood states (POMS-SF) Presence and severity of symptoms (MD Anderson Symptom Inventory-Brain Tumour Module)	MUIS-BT	Bivariate Anger subscale r=0.40*** Confusion subscale r=0.49*** Depression subscale r=0.48*** Fatigue subscale r=0.35*** Tension subscale r=0.56***	Y Y Y Y	63.6 Fair
Mullins et al. (2000)	Childhood onset asthma	40 college students from two midwestern universities with diagnosed childhood asthma. Mean age=19.67(1.77) Range=18-25 Female=55%	Cross-sectional	Depression (IDD) Illness intrusiveness (IIRS)	MUIS-C	Partial correlations (controlling for age and socioeconomic background) r=0.63***	Y	66.7 Fair
Mullins et al. (2001)	Multiple Sclerosis	78 participants from regional support groups and local neurologists Mean age=46.3(9.1) Range=30-73 Female=70.5%	Cross-sectional	Psychological distress (SCL-90-R) Illness intrusiveness (IIRS)	MUIS-C	Bivariate full scale Global PD $\beta=0.47^{**}$ Hierarchical regression analysis $\beta=0.29^*$	Y Y	70 Good

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS measure	Correlation coefficient	Significant association	Quality score (%) and rating
Mullins et al. (2017) America	Allergies, asthma Other chronic illnesses (type 1 diabetes cystic fibrosis, epilepsy, sickle cell disease, migraines, IBD, obesity) Community	364 college students with self-reported allergies and/or asthma Mean age=19.62(2.09) Range=18-26 Female=65.1% 148 college students with self-reported other chronic illnesses Mean age =20.38(1.70) Female=69.6%	Cross-sectional	Depression (CES-D) Anxiety (SAS) Illness intrusiveness (IIRS)	MUIS-C	Bivariate Depression Asthma/allergies $\beta=0.36^{***}$ Other CI's $\beta=0.25^{**}$ Anxiety Asthma/allergies $\beta=0.46^{***}$ Other CI's $\beta=0.27^{**}$	Y Y Y Y	60 Fair
Pahlevan (2017a) Malaysia	Breast cancer Outpatients	901 healthy students 118 Malaysian women with breast cancer from a private hospital Mean age =50.95(9.35) Female=100% January 2016 to April 2016	Cross-sectional	Depression and anxiety (HADS) Locus of control (Locus of control scale -short form)	SF-MUIS	Partial correlation (controlling for age, cancer stage, time since diagnosis and education) Depression $r=0.32^{**}$ Anxiety $r=0.29^{**}$	Y Y	80 Good

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS measure	Correlation coefficient	Significant association	Quality score (%) and rating
Pahlevan et al. (2017b) Malaysia	Breast cancer Outpatients	135 Malaysian women with breast cancer from a medical centre Mean age =51.18(9.48) Female=100% November 2015 to January 2016	Cross-sectional	Depression and anxiety (HADS) Coping strategies (Brief COPE)	MUIS-SF	Partial correlation (controlling for age, cancer stage, time since diagnosis and education) Depression r=0.32** Anxiety r=0.27**	Y Y	60 Fair
Sanders-Dewey et al. (2001) America	Parkinson's disease Outpatients	44 participants 66 years (SD = 7.9 years) Mean age = 72.8 (7.2) Female=29.5%	Cross-sectional	Psychological Distress (90-SCL-R) Depression and anxiety subscales	MUIS-C	Bivariate Depression r=0.21 Anxiety r=0.20 GSI r=0.14 Multiple regression analysis IU no significant variance to global distress	N N N	70 Good
Santacroe & Lee (2006) America	Young adult survivors of childhood cancer Community	45 participants: a clinical database of a childhood cancer long-term follow up programme Mean age=24.5(5.5) Range=22-40 Female=62.2%	Cross-sectional	Post traumatic symptoms (PTSDI)	MUIS -C	Bivariate r=0.40*	Y	60 Fair

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS measure	Correlation coefficient	Significant association	Quality score (%) and rating
Sharkey et al. (2018) America	Chronic conditions (including asthma, allergies, inflammatory bowel disease, type 1 diabetes, epilepsy, obesity, juvenile rheumatoid arthritis) Community	120 college students Mean age =21.13(5.45) Female=73.3%	Cross-sectional	Depression (CES-D) Anxiety (SAS) Grit (SGS) Illness intrusiveness (IIRS)	MUIS-C	Bivariate Depression r=0.57*** Anxiety r=0.63***	Y Y	60 Fair
Small & Graydon (1992) Canada	COPD Inpatient	26 participants from five large teaching hospitals Mean age=69(8.5) Range=53-86 Female=42.3%	Cross-sectional	Transient mood states (negative mood) Physical symptoms	MUIS-A (28-item) POMS Somatic scale - Bronchitis Emphysema symptom checklist	Bivariate correlation r=0.37 Hierarchical regression analysis (controlling for physical symptoms) IU accounted for 9% of variance (and prediction not significant)	N	70 Good

Author (year) Country	Chronic illness	Sample characteristics Mean age (standard deviation, if reported) (years) Age ranges % female	Study design	Outcome variables (and measures)	MUIS measure	Correlation coefficient	Significant association	Quality score (%) and rating
Wolfe-Christensen et al. (2008)	Childhood onset asthma Community	102 young adult college students recruited from undergraduate psychology and marketing classes at a major Midwestern university Mean age=19.70(1.25) Range=18-22 Female=63.7%	Cross-sectional	Psychological distress (BSI) Symptom severity (Ratings of asthma severity)	MUIS-C	Global severity index of BSI (all nine scales) Bivariate correlation r=0.53**	Y	60 Fair

Note. BAI=Beck Anxiety Inventory (Beck et al., 1988), Brief COPE, (Carver et al., 1989), BSI=Brief Symptom Inventory (Derogatis, 1993), CES-D=Centre for Epidemiological Studies-Depression (Radloff, 1977), et al., 1997), FARS=Feelings and Reactions Scale (Lemaire, 2004), HADS=Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983), IDD=Inventory to Diagnose Depression (Zimmerman & Coryell, 1987), IIRS=Illness Intrusiveness Ratings Scale (Devins et al., 1983), IUS=Intolerance of Uncertainty Scale (Carleton et al., 2007), Locus of control scale-short form (Levenson, 1974), MAX-PC=Memorial Anxiety Scale for Prostate Cancer (Roth et al., 2003), MUIS-A=Mishel Uncertainty In Illness-Acute (Mishel, 1981), MUIS-C=Mishel Uncertainty In Illness-Community (Mishel, 1986), MSES=Modified Symptom Experience Scale (Detprapon et al., 2009), MSPSS=Multidimensional Scale of Perceived Social Support (Zimet et al., 1988), PHQ-9=Patient Health Questionnaire (Kroenke et al., 2001), POMS=Profile of Mood State (Biehl & Landauer, 1975), PTSDI=Post Traumatic Stress Disorder Index (Pynoos et al., 1998, cited in Santacroce & Lee, 2007), SCL-90-R=Symptom Checklist-90-Revised (Derogatis, 1992), SAS=Zung Self-rating Anxiety Scale (Zung, 1971), SF-GDS=Short Form-Geriatric Depression Scale (Cho et al., 1999), SGS=Short Grit Scale (Duckworth & Quinn, 2009). *p < 0.05; **p < 0.01; ***p < 0.001

Study characteristics

Most of the studies were conducted in the United States of America (N=15). Others took place in Canada (N=1), England (N=1), Italy (N=1), Malaysia (N=2), South Korea (N=1), Taiwan (N=1) and Thailand (N=1). Three studies recruited participants from inpatient settings and the remaining studies from outpatient departments (N=11) or community settings (N=9).

Twenty-two studies employed a cross-sectional design. One employed a prospective longitudinal design (Hoth et al., 2013) measuring variables across 2-years. All studies used self-report measures. Two studies recruited participants from the same medical centre but across different time frames (Pahlevan, 2017a; Pahlevan et al., 2017b), therefore no overlapping datasets were identified.

Clinical characteristics

Demographics

There was a combined total of 3162 participants across studies and a large variation in sample-size (26 to 407, median=118). Mean ages ranged from 19.11 to 65.1 years with 1919 females and 1243 males. Females constituted 60.7% of the overall sample.

Chronic illnesses

Twenty-one studies included one CI and 14 CI's in total were researched across the studies, including: childhood onset asthma (Carpentier et al., 2007; Hommel et al., 2003; Mullins et al., 2017; Mullins et al., 2000; Sharkey et al., 2018; Wolfe-Christensen et al., 2008); breast cancer (Pahlevan, 2017a; Pahlevan et al., 2017b), head and neck cancer (Detprapon et al., 2009); lung cancer (Kurita et al., 2013); brain tumours (Lin et al., 2013); chronic renal patients (Barberis et al., 2019), end-stage renal disease (B.

Kim & J. Kim, 2019); chronic lung disease or COPD (Hoth et al., 2013; Small & Graydon, 1992), endometriosis (Lemaire, 2004); hepatitis C (Bailey et al., 2009; Colagreco et al., 2014), Multiple sclerosis (Mullins et al., 2001), Parkinson's Disease (Ahn et al., 2017; Sanders-Dewey et al., 2002) and young adult survivors of childhood cancer (Lee, 2006; Santacroce & Lee, 2006). Two studies researched mixed CI populations (see Table 1, Mullins et al., 2017; Sharkey et al., 2018). Mullins et al. (2017) compared asthma/allergies, mixed CI, and non-CI groups.

Methodological quality

All the studies clearly defined their research question, objectives, hypotheses, sample population and inclusion/exclusion criteria. Across all studies, independent and dependent variables, and their corresponding measures, were clearly defined and accompanied by clear reporting of their psychometric properties. The main critique pertained to the cross-sectional design (N=22) and limitations associated with this such as the inability to make causal inferences and the unmeasurable influence of confounding variables on findings. Furthermore, it was only possible to determine that >50% of the source population were recruited in 8/23 studies (34.8%). Therefore, for most studies, samples were not representative of the source populations. Convenience sampling also introduced risks pertaining to self-selection bias amongst the studies. Only seven studies (30.4%) reported a power analysis calculation. Therefore, despite most studies recruiting large samples, it was difficult to determine if they were sufficiently powered to avoid type 1 or II errors.

Synthesis of review findings

Studies used a heterogeneous array of measures based on Mishel's conceptualisation of IU, measures of psychological distress; and recruited participants across various care-contexts and CI types. To synthesise findings, consideration was

given firstly to the overall levels of IU; the general trends across studies of the association between IU and psychological distress: and next according to care-context; measures of psychological distress; CI types; and ‘other’ variables researched in relation to IU. Table 1 presents the correlation coefficients for all the included studies and will be used to facilitate comparisons amongst the measures and groups. Tables 3-5 present means and effect size ranges of the groups or measures to further facilitate comparison of findings.

1. What do the studies suggest about the commonality of illness uncertainty in those with chronic illnesses and its association with psychological distress?

Table 2 presents descriptive statistics for IU scores.

Table 2.

The means, standard deviations (SD) and ranges of scores for studies reporting the most used MUIS-C and MUIS-A

Measure	Mean (SD)	Minimum/maximum score range
MUIS-C (possible range 23-115)	56.14(7.9)	45.95-70.60
MUIS-A (possible range 33-165)	88.9(6.39)	81.78-99.03

Note. Nine out of twelve studies using the MUIS-C reported mean IU scores for their samples and all five studies using the original 33-item MUIS-A (or variation of) reported their samples’ mean scores. No MUIS measure provides categorical cut-off scores.

Totalled mean scores across the study samples were subjectively interpreted and deemed low to moderate. Raw scores from the remaining studies using alternate measures (see Appendix F) mirror these findings, suggesting that low to moderate levels of IU are consistent across studies.

Twenty-one studies (91.3%) found statistical significance between IU (or a facet of IU) and form of psychological distress in a positive direction, thus the greater the IU, the greater the psychological distress. Effect sizes were wide-ranging ($r = 0.14-0.82$).

2. Are there differences in the association between illness uncertainty and psychological distress according to the care-context?

The IU measure used reflected the care-context. Illustratively the MUIS-C (Mishel, 1991) was used predominantly with community (N=9) and outpatient samples (N=2). The MUIS-A (Mishel, 1981) was used in inpatient (N=3) and outpatient samples (N=5).

All nine studies researching community samples found significant associations between IU and a form of psychological distress. These studies researched CI's ranging from childhood onset asthma (N=4), endometriosis (N=1), childhood cancer survivors (N=2) and mixed CI's (N=2) (see Table 1). Across studies varying forms of psychological distress were researched, including depression (N=8), anxiety (N=5), general psychological distress (N=5) and post-traumatic stress symptoms (N=2).

Carpentier et al. (2009) conducted hierarchical regression analyses and found that IU significantly predicted depression, anxiety, and psychological distress ($p < 0.001$). Their study was rated as 'good' and included a largely representative population of college students (92% participant rate) who rated their asthma severity as relatively mild and somewhat controllable. From examination of bivariate correlations alone in community samples, effects sizes were either medium (Lemaire, 2004; Lee,

2006; Mullin et al., 2017; Santacroce & Lee, 2006), or large (Hommel et al., 2003; Sharkey et al., 2018; Wolfe-Christensen et al., 2009). These studies shared the limitation of not controlling for confounding variables in their examination of the association, however for those who conducted partial correlations (Hommel et al., 2003; Mullins et al., 2000) significant associations remained.

There was a similar trend towards a significant association between IU and psychological distress for inpatient samples using the MUIS-A. Those researching chronic end of life renal disease (Barberis et al., 2019; B. Kim. & J. Kim., 2019) both found medium effect sizes. Conversely, Small and Graydon (1992) found that IU accounted for only 9% of the variance (non-significant) of negative mood in a sample with COPD. The authors postulated that the mean length of time since diagnosis (10.6 years) led to the development of coping, however length since diagnosis was comparable to other studies within this review where significant associations were found (see Appendix G). However, they used a small sample and did not report a power calculation. Thus, the study might have lacked sufficient power to detect an association (type 1 or 11 error).

Outpatients samples (N=11) included those living in the community but receiving treatment from a medical centre (inclusive of Parkinson's disease, multiple sclerosis, types of cancers, COPD and Hepatitis C). There was a similar trend towards a significant association between IU and psychological distress. Of the significant associations found across outpatient samples, the effect sizes were wide-ranging (0.13-0.82). Two studies found non-significant associations (Kurita et al., 2014; Sanders-Dewey et al., 2002). However, in Sanders-Dewey et al.'s. (2002) study with individuals with Parkinson's disease, the MUIS-C was found to have low internal consistency calling into question the applicability of this measure for their Parkinson's disease

population and thus validity of their findings. Table 3 presents the mean effects sizes for all population groups to further aid comparisons. Mean effect sizes for all care-contexts fell within the medium range.

Table 3.

Effect size ranges, means and standard deviations (SD's) for the association between IU according to care context

Sample	Number of studies	Mean effect size (SD)	Range
Community	9	0.39 (0.20)	0.01-0.63
Outpatients	11	0.41(0.19)	0.07-0.82
Inpatients	3	0.36(0.46)	0.31-0.40

Note: Bivariate correlation coefficients were included in the calculations, however if a study only reported partial correlations these were included in the mean effect size calculation.

3. Are there differences in associations between illness uncertainty and psychological distress according to the type of measurement of psychological distress?

Depression was the most widely measured form of psychological distress (N=16), Within these studies the CES-D (Radloff, 1977, N=6) was the most widely used measure, containing frequency-based questions regarding an array of depressive symptoms. Others used subscales of the HADS (Zigmond & Snaith, 1983, N=4), containing items related to symptoms of anhedonia. See table 1 for other depression measures used.

Measures of depression and associated mood states. Across all 16 studies researching IU and depression there was a general trend towards significant and positive associations, with wide-ranging effect sizes ($r=0.05-0.82$). Several large effect sizes were found between IU and CES-D (Radloff, 1977) (Bailey et al., 2009; Colagreco et al., 2014; Detprapon et al., 2009; Sharkey et al., 2018).

Three of the studies researching depression used the MUIS-A which illuminated the elements of IU within Mishel's conceptualisation more associated with the development of low mood. Ambiguity correlated with depression across several studies (Bailey et al., 2009; Colagreco et al., 2014; Hoth et al., 2013). A strength of Bailey et al.'s. (2009) study was that they controlled for the other IU subscales. Conversely neither Bailey et al. (2009) nor Colagreco et al. (2014) controlled for confounding variables, meaning other unknown factors could have impacted the association between IU and depression. Hoth et al. (2013) also found in their longitudinal investigation that depression was associated with ambiguity across a 2-year time-period and not at any point in the illness trajectory. This indicates that IU plays a causal role. The authors also recruited a geographically diverse sample. Non-significant associations were found between the unpredictability subscale and depression in two studies researching people with Hepatitis C (Bailey et al., 2009; Colagreco et al., 2014).

All other studies using depression measures examined the relationship with MUIS-total scores. Positive and significant associations were consistent across CI types and care-contexts. This was synonymous for negative mood states (Lin et al., 2013), however this finding was not consistent (Small & Graydon, 1992). Hommel et al. (2003) found that once gender, age, subjective and objective illness severity (asthma) were controlled for, the association between IU and depression became non-significant.

Measures of anxiety. Eight studies measured anxiety using anxiety subscales of the HADS (Zigmond & Snaith, 1983, N=4), and BSI (Derogatis, 1993, N=2); the SAS (Zung, 1971, N=1), and BAI (Beck et al., 1988, N=1). Effect sizes ranged from small to large (0.01-0.56). Most of the studies found positive significant relationships between IU and anxiety. Anxiety also remained stable over a 2-year period and correlated with the ambiguity aspect of IU (Hoth et al., 2013), suggesting that anxiety about symptom

ambiguity/vagueness of symptom profile does not abate. In both Mullins et al's. (2017) and Carpentier et al's (2009) studies levels of anxiety were significantly higher in the CI groups compared with the healthy controls (no CI diagnosis).

Eight of these studies examined both anxiety and depression. Interestingly, of these eight studies, anxiety had larger correlation coefficients than depression in five studies (Barberis et al., 2019; Hommel et al., 2003; Hoth et al., 2013; Mullins et al., 2017; Sharkey et al., 2018). In Hommel et al's. (2003) study IU contributed significant variance to anxiety once gender, age, disease variables and depressive symptomatology were controlled for.

Measures of psychological distress. Synonymous with low mood and anxiety a positive significant association was found across six of the seven studies using measures of general distress. All these studies used the MUIS-C (Mishel, 1991) thus conclusions could not be drawn pertaining to individual facets of IU. The effect sizes ranged from small to large with a medium overall mean effect size (see Table 4). Two studies researched childhood survivors of cancer (Santacrose & Lee, 2006) assessing post-trauma symptomatology and found medium significant effect sizes. This supports that IU remains relevant and associated with trauma symptoms for those who are no longer actively unwell but live with an ongoing threat of re-occurrence. Table 4 presents the mean effect sizes for all forms of psychological distress. Overall, they all fell within the medium ranges.

Table 4.

Effect size ranges and means for the association between IU and forms of psychological distress.

Psychological distress categories	Number of studies	Mean effect size (SD)	Effect size ranges
Depression	17	0.38 (0.17)	0.06-0.82
Anxiety	8	0.36(0.2)	0.01-0.63
General psychological distress	5	0.33(0.22)	0.04-0.53
Post-trauma symptoms	2	0.40(0)	0.40

Note: Bivariate correlation coefficients were included in the calculations, however if a study only reported partial correlations these were included in the mean effect size calculation.

4. Are there differences in associations between illness uncertainty and psychological distress according to chronic illness type?

One study directly compared CI groups (see Table 1, Mullins et al., 2017). Their group with mixed CI's had significantly higher levels of IU and anxiety than those with asthma/allergies or those with an absence of a CI diagnosis. Four studies researched childhood onset asthma (Carpentier et al., 2007; Hommel et al., 2003; Mullins et al., 2000; Wolf-Christensen et al., 2009); and two additional studies researched mixed asthma/allergy samples (Mullins et al., 2017; Sharkey et al., 2018). All six of these studies recruited young adult college students limiting generalisability to other populations. Nevertheless, borderline large or large effect sizes were found in most of the studies researching solely asthma or medium or large effect sizes were observed for those including mixed asthma/allergy samples. The only study that researched endometriosis found IU to be associated with emotional distress with a medium effect size (Lemaire, 2004).

Positive significant associations were consistently found for those with cancers, with at least medium effect sizes (0.27-.0.82). The largest effect size was observed for those with head and neck cancer, an illness that threatens parts of the body that are visibly prominent and serve important communicative functions. One out of three studies researching degenerative conditions (Sander-Dewey et al., 2002) found a non-significant association. It was possible that those individuals were clearer about their prognosis and impending outcome. However more studies are needed on IU and degenerative conditions so that robust conclusions can be drawn.

Table 5 presents the mean effect sizes across CI groups. The mean effect size was slightly larger for those with cancers overall, which may reflect the threatening nature of the condition and corresponding treatments. However, all groups fell within the medium effect size range.

Table 5.

Effect size ranges, means and standard deviations (SD'S) for the association between IU according to CI types.

Sample	Number of studies	Mean effect size (SD)	Effect size ranges
Cancers	6	0.43 (0.17)	0.18-0.82
Childhood onset asthma/allergies	6	0.36 (0.23)	0.01-0.63
Degenerative conditions	3	0.30(0.16)	0.14-0.47

Note: Bivariate correlation coefficients were included in the calculations, however if a study only reported partial correlations these were included in the mean effect size calculation.

5. What is known about other variables in relation to illness uncertainty?

Gender. Eighteen studies used mixed gender samples, and seven examined IU according to gender. Women reported significantly higher levels of IU in two studies on

participants with mixed CI's and childhood onset asthma (Sharkey et al., 2018; Wolfe-Christensen et al., 2008). There was no significant difference in IU scores between genders in five studies (Ahn et al., 2017; Bailey et al., 2009; Carpentier et al., 2009; Kurita et al., 2013; Mullins et al., 2001).

Illness intrusiveness. Illness intrusiveness was assessed across five studies (Carpentier et al., 2007; Mullins et al., 2000; Mullins et al., 2001; Mullins et al., 2017; Sharkey et al., 2018). Only one study (Sharkey et al., 2018) found that IU and illness intrusiveness combined influenced anxious and depressive symptomatology. The other studies found that IU and illness intrusiveness independently predicted depressive and anxious symptomatology (Mullins et al., 2000; Mullins et al., 2001; Mullins et al., 2017). Illness intrusiveness did not mediate the relationship (Mullins et al., 2000). Mullins et al. (2017) found greater effects sizes for IU than illness intrusiveness for both depressive ($\beta=0.36$, $p < .001$) and anxiety symptoms ($\beta=0.46$, $p < .001$). .

Grit. Sharkey et al. (2018) found an inverse relationship between grit and IU ($\beta= -0.21$, $p < .05$). Their overall path-analysis demonstrated that IU mediated the relationship between grit and outcomes of anxiety and depression. Thus, grit was associated with reduced IU which in turn was related to decreased distress.

Coping styles. Pahlevan et al. (2017b) found a significant positive relationship between IU and avoidant coping ($r = 0.218$, $p < 0.05$), and the inverse for active emotional coping ($r = -0.297$, $p < 0.01$). Kurita et al. (2013) examined avoidance (e.g. of uncertain situations threatening images or thoughts) and its relationship with intolerance of uncertainty. Avoidance was found to mediate between intolerance of uncertainty and non-somatic depressive symptoms.

Locus of control. Pahlevan (2017a) examined LOC and found that individuals with breast cancer with higher internal LOC (ILOC, and lower external LOC)

experienced lower anxiety (-0.374, $p < 0.01$) and depression (-0.269, $p < 0.01$).

Uncertainty mediated the relationship between LOC and depression, but not anxiety.

Disease-related variables

Symptom severity. Subjective symptom severity was measured across seven studies and objective illness severity was additionally measured in three of those studies (see Table 5). The measures used to examine illness severity differed in accordance with differing clinical profiles. Overall, IU was more frequently positively associated with subjective ratings, thus the more one perceived their symptoms to be severe the greater the IU. A limitation of Wolfe-Christensen et al's. (2008) study was there use of a single item created by the research team, thus calling into question the comprehensiveness and reliability of it.

Table 5.

The Correlation coefficients (r) between subjective and objective illness severity and IU.

Study	Subjective illness severity rating	Objective illness severity rating
Carpentier et al. (2007)	0.01	-0.06
Detprapon et al. (2009)	0.69***	
Hommel et al. (2003)	0.17	0.23
Mullins et al. (2000)	0.38*	
Mullins et al. (2017)	0.106**	
Lin et al. (2013)	Anger 0.26*** Tension 0.18* Depression .24** Fatigue 0.24*** Confusion Non-significant	
Wolf-Christensen et al. (2008)	0.40**	0.07

Note. *p = < 0.05; **p = < 0.01; ***p = < 0.001

Illness duration. Illness durations varied greatly across studies and are tabulated in Appendix G. Three out of seven studies that examined illness duration found a non-significant association with IU. In two studies ‘time since diagnosis’ was used as a control and the significant relationship between IU and PD remained (Pahlevan, 2017a; Pahlevan et al., 2017b). In the only prospective study, ambiguity, and psychological distress were not impacted by number of years since diagnosis (Hoth et

al., 2013). Only one study found that an earlier point in the illness trajectory was associated with greater IU in those with brain tumours (Lin et al., 2013).

Discussion

This is the only systematic review to synthesise and examine the current state of quantitative research investigating the association between IU and psychological distress in adult CI populations. The review comprised 23 papers spanning almost three decades (1992 to 2019), with a total number of 3126 participants. Overall low to moderate levels of IU were observed across the included studies, confirming that IU is an integral aspect of the CI experience (Mishel, 1990).

This synthesis garners strong support for the association between IU and psychological distress, supported by 21 studies. While conclusions cannot be drawn regarding the direct or causal link between IU and psychological distress, findings suggest IU impacts on psychological distress in adults with CI. This is consistent with the meta-analytic evidence found in relation to paediatric IU in which medium effect sizes were observed between IU and outcomes of depression and anxiety (Szulxzewski et al., 2017).

IU was ubiquitously linked with psychological distress, regardless of gender, care-context, form of psychological distress or CI type. Some findings suggest that ambiguity; symptoms or causes that can be vague and interpretable in several ways, might be more pertinent to the development of psychological distress than other facets of IU. Furthermore, slightly higher associations were found overall for those with cancers, with at least medium effect sizes (0.27-.0.82). This might reflect the threat of mortality cancer poses (American Cancer Society, 2008). More research is needed researching inpatient samples.

This synthesis found that IU associated with diverse manifestations of distress. Depression and anxiety were frequently comorbid which reflects findings in the general population (Wu & Fang, 2014); and overall, they were consistently associated with IU. Clark and Watson's (1991) tripartite model argues that anxiety and depression which are physiologically and phenomenologically distinct (anxiety is characterised by hyperarousal and depression by anhedonia), are both underpinned by negative affectivity. Comorbidity can complicate the clinical picture, leading to treatment resistance (Wu & Fang, 2014). However, these findings suggest that IU is an underlying transdiagnostic factor elevating negative affectivity in those with CI's. Furthermore, trauma symptoms were observed to associate with IU in two childhood cancer survivor populations (Lee, 2006; Santacrose & Lee, 2003). Post trauma symptomatology has also been identified in a chronic heart disease population (Moreland & Santacrose, 2018), which suggests this finding is generalisable to other CI populations and underpinned by IU related challenges. However more research is needed assessing post trauma symptomatology in those with other CI's.

Theoretical frameworks

Considering Mishel's (1990) framework it can be inferred from these findings that negative appraisal and maladaptive coping strategies are employed in response to the experience of IU, leading to outcomes of psychological distress. Indeed, in Wright et al's. (2009) review IU was found to associate with maladaptive coping strategies. Most of the studies did not assess appraisal processes connected to IU, mirroring a previous critique of the empirical literature guided by Mishel's UIT (McCormick, 2004). However, research has begun exploring intolerance of uncertainty (Kurita et al., 2013) and ILOC (Pahlevan et al., 2017); the appraisal that one is responsible for the events that occur/perceptions of control. These are important areas of research because

how in control a person feels influences uncertainty (McCormick, 2002). Furthermore, ILOC has previously been found to inversely associate with anxiety, and depression (Fan et al., 2010; Park & Gaffey, 2007) and better health outcomes (Green & Murdock, 2013). A large body of research has studied intolerance of uncertainty as an influential transdiagnostic factor underlying anxiety disorders and depression (Boswell et al., 2013; Carleton et al., 2012). Hence, a fruitful area for future research pertains to the exploration of intolerance of uncertainty and ILOC as specific appraisal process in the context of CI and IU.

This synthesis indicates that IU correlates with psychological distress at any point in the illness trajectory and post recovery, into survivorship (Lee, 2006; Santacroce & Lee, 2006). This evidence and the longitudinal investigation (Hoth et al., 2013) refutes Mishel's (1990) UIT reconceptualization that, over time, a perspective shift from a danger appraisal to opportunity may lead to positive psychological changes. However, psychological adaptation to CI and chronic IU is a complex, evolving and possibly cyclical process (Mishel, 1988, 1990). Qualitative research in this area would enrich the understanding of how IU is experienced and evolves over time.

The cognitive behavioural model can also be used to understand the association between IU and psychological distress. The theory argues that appraisals, emotions, and behaviours (such as maladaptive coping strategies) interact and influence each other. Pahlevan et al. (2017b) found that IU was positively associated with avoidant coping (such as denial or self-distraction), anxiety and depression, a finding that was consistent with previous findings in a sample with prostate cancer (Guan et al., 2020). Pahlevan et al. (2017b) adds the importance of IU in this association. The Cognitive Behavioural Therapy (CBT) model suggests that the ways in which one behaves (coping strategies) can perpetuate negative affect and appraisal. Thus, CBT advocates targeting coping

strategies, management of affect and cognitive reframing (Beck, 1976). A previous study demonstrated that applying CBT and shifting control-beliefs from external to internal (ILOC) led to positive outcomes in a CI population (Mehrtak et al., 2017).

Limitations of the studies

The limitations of the included studies restrict the validity and generalisability of the findings. The predominant limitation concerned the cross-sectional design which meant causality could not be deduced. Henceforth it was unclear whether IU contributed directly to psychological distress or whether those with higher psychological distress experienced greater IU. It might be that there is a reciprocal and mutually reinforcing relationship between these variables. Future longitudinal research should utilise designs that elucidate causation and explore the temporal relationships between variables.

A cross-sectional design also introduces risks pertaining to uncontrolled, confounding variables (Setia, 2016). IU and psychological distress are dynamic and likely to be confounded by external events. Therefore, caution should be exercised when attributing psychological distress solely to IU. Additionally, in relation to examination of the direct association between IU and psychological distress, only nine studies used statistical methods that allowed for known confounding variables to be controlled for. Finally, only six studies reported a power calculation, thus despite most studies recruiting large samples, it was difficult to determine if studies were sufficiently powered (Nayak, 2010).

The studies were also limited in terms of their generalisability. Most studies were conducted in the USA; thus, the cross-cultural validity of the findings is spurious. Additionally, convenience sampling was used across studies. This method can reduce sample bias by generating a more representative sample of the population of interest (Hulley et al., 2013), however, in half the studies less than 50% of the available sample

were recruited. Thus, in comparison to random sampling methods this design lacks rigour. Furthermore, all studies researching childhood asthma or mixed asthma/allergies samples used young adult college students and thus future research would need to diversify samples to generalise findings. As significant differences between asthma and allergy groups have been observed (Hullman et al., 2013), future research should delineate groups. Replicated studies of specific CI's might add to the understanding of IU and psychological distress for each CI, which vary naturally in their challenges, particularly with regards to the condition's controllability.

While a strength of the studies was that they utilised valid and reliable measures, Carleton et al. (2013) argued that the validity and psychometric properties of the CES-D have been questioned. For example, items assessing somatic concerns may artificially inflate scores for chronic pain populations. Considering that the phenomenology of many CI's involves the experience of pain or other somatic complaints, it must be acknowledged that this may have conflated findings in the six studies in which it was employed in. Future research might consider the use of depression measures that do not include items related to somatic complaints. In general, the extensive use of self-report measures may have increased the chance of shared method variance which can lead to a higher probability of significant relationships between variables (Podsakoff et al., 2003). With further consideration of measurement, future studies should delineate MUIS subscales to assist understanding of which aspects may be a target for psychological intervention.

Limitations of the review

A narrative synthesis approach was chosen on the basis the studies were not homogenous enough to conduct a meta-analysis; it was deemed unfeasible to divide papers into homogenous clusters to conduct subgroup statistical analyses. Therefore,

this review did not use meta-analytic statistics to quantify associations and test moderators, however the narrative synthesis enabled examination of the IU, psychological distress association across settings, CI types and forms of psychological distress. The conclusions may have been undermined by publication bias meaning that quantitative studies that demonstrate significant findings are more likely to be published or those that did not are more likely to remain unpublished (Jooper et al., 2012). Furthermore, the exclusion of grey literature and non-English language articles from the review may have increased this bias. Future reviews could include correspondence with experts in the field to pursue unpublished data (Rosenthal, 1979). Finally, mean effects sizes were used to supplement comparison of groups, however this simple statistical calculation does not calculate the precision of individual studies (Borenstein et al., 2007), thus these findings should be interpreted with caution.

Strengths of this review included the breadth of electronic databases searched, with no publication-date restrictions, enabling a comprehensive search of eligible studies. This increased the probability they were all retrieved. This review followed a systematic process and employed a critical appraisal tool, second-rated, which increased its robustness and would enable other reviewers to accurately replicate. Furthermore, the PRISMA (2009) checklist was used to facilitate quality and comprehensiveness (see Appendix H).

Clinical implications

This synthesised body of evidence does not refute current practices and guidance. An intervention employing cognitive-behavioural strategies in line with NICE (2010) guidance, suggests that IU can be modified by targeting a patient's cognitions, knowledge, and coping skills and that this can have positive effects for those with a CI (Gil et al., 2006). These findings suggest that assessing and targeting IU as a

transdiagnostic factor underpinning many forms of psychological distress is relevant in diverse care settings, across genders, and at any time across the illness trajectory.

HCP's should routinely assess for the presence of IU and forms of psychological distress at the point of diagnosis, throughout the illness trajectory and into survivorship (Decker et al., 2007). Use of the MUIS-A would enable assessment of the distinct aspects of IU, however when used in community samples, consideration ought to be taken with regards to non-applicable items. HCP's should assess for the idiosyncratic meaning of IU, appraisals (including of intolerance of uncertainty and control beliefs) and coping strategies. Where psychological distress reaches a clinical level, the above assessment information can guide the development of a CBT case formulation (Jacqueline & Lisa, 2015) and guides target areas for intervention that account for IU. Given the prevalence of IU in CI populations and potential for negative appraisal or intolerance of uncertainty, the experience of IU should be normalised. In line with Mishel's (1981) theory and the pertinence of ambiguity to psychological distress, HCP's should endeavour to provide clear information and effective communication.

Conclusions

A synthesis of all available studies strongly suggests an association between IU and psychological distress, regardless of individual or contextual differences. Findings support current guidance recommending CBT to address psychological distress in those with CI. IU should be considered as a transdiagnostic factor, and appraisals processes targeted. HCP's should normalise IU in those with CI's at any point along the illness trajectory. Future research is needed to establish appraisal processes linked to IU, causal pathways and how to investigate how IU evolves over time.

References

* = included in this review

- *Ahn, S., Lee, J., Chu, S., & Sohn, Y. (2017). Uncertainty and depression in people with Parkinson's disease: A cross-sectional study. *Nursing & Health Sciences, 19*(2), 220-227. <https://doi:10.1111/nhs.12332>.
- Alligood, M. (2014). *Nursing theorists and their work*. Elsevier Health Sciences.
- Alschuler, K., & Beier, M. (2015). Intolerance of uncertainty: Shaping an agenda for research on coping with multiple sclerosis. *International Journal of MS Care, 17*(4), 153-8. <https://doi:10.7224/1537-2073.2014-044>.
- Amo-Setien, F., Abajas-Bustillo, R., Torres-Manrique, B., Martin-Melon, R., Sarabia-Cobo, C., Molina-Mula, J., & Ortego-M, C. (2019). Characteristics of nursing interventions that improve the quality of life of people with chronic diseases. A systematic review with meta-analysis. *PLoS One, 14*(6). <https://doi:10.1371/journal.pone.0218903>.
- Bailey, J., & Neilson, B. (1993). Uncertainty and appraisal of uncertainty in women with rheumatoid arthritis. *Orthopaedic Nursing, 12*(2), 63- 67. <https://doi:10.1097/00006416-199303000-00011>.
- *Bailey, D., Landerman, L., Barroso, J., Bixby, J., Mishel, M., Muir, A., Strickland, L., & Clipp, E. (2009). Uncertainty, symptoms, and quality of life in persons with chronic hepatitis C. *Psychosomatics, 50*(2), 138-146. <https://doi:10.1176/appi.psy.50.2.138>.

- Bailey, D., Wallace, M., Latini, D., Hegarty, J., Carroll, P., Klein, E., & Albertsen, P. (2011). Measuring illness uncertainty in men undergoing active surveillance for prostate cancer. *Applied Nursing Research, 24*(4), 193-199. <https://doi:10.1016/j.apnr.2009.08.001>.
- Bang, J., Seongwoo, Jo., Beomkim, G., Sangkwon, B., Jungbae, E., Illnoh, C., & Yunchoi., J. (2013). The mental health and quality of life of adult patients with congenital heart disease. *International Journal Cardiology, 170*(1), 49–53. <https://doi.org/10.1016/j.ijcard.2013.10.003>.
- *Barberis, N., Gitto, S., Larcana, L., Buemi, R., & Santoro, D. (2019). Role of emotional intelligence as a mediating factor between uncertainty and anxiety hospital in chronic renal patients. *Illness, Crisis & Loss, 27*(2), 71-86. <https://doi.org/10.1177/1054137316667595>.
- Beck, A. (1967). *Depression: Causes and treatment*. University of Pennsylvania Press.
- Beck, A.T. (1976). *Cognitive Therapy and the Emotional Disorders*. Penguin Books.
- Beck, A., Epstein, N., Brown, G., & Steer, R. (1988). An inventory for measuring clinical anxiety: Psychometric properties. *Journal of Consulting and Clinical Psychology, 56*(6), 893-7. <https://doi:10.1037//022-006x.56.6.893>.
- Biehl, B., & Landauer, A. (1975). *Das Profile of Mood States (POMS)*. Mannheim.
- Borenstein, M., Hedges, L., & Rothstein, H. (2007). Introduction to meta-analysis. <https://www.meta-analysis.com/downloads/Meta%20Analysis%20Fixed%20vs%20Random%20effects.pdf>

- Boswell, J., Thompson-Hollands, J., Farchione, J., & Barlow, D. (2013). Intolerance of uncertainty: A common factor in the treatment of emotional disorders. *Journal of Clinical Psychology, 69*(6). <https://10.1002/jclp.21965>.
- Buhr, K., & Dugas, M. (2009). The role of fear of anxiety and intolerance of uncertainty in worry: An experimental manipulation. *Behavioural Research Therapy, 47*(3), 215-23. <https://doi:10.1016/j.brat.2008.12.004>.
- Cal, S., Ribeiro de Sa, L., Glustak, M., & Santiago, M. (2014). Resilience in chronic diseases: A systematic review. *Cogent Psychology, 2*(1), 1024928. <https://doi?10.1080/23311908.2015.1024928>.
- Carleton, R., Norton, M., & Asmundson, G. (2007). Fearing the unknown: a short version of the Intolerance of Uncertainty Scale. *Journal of Anxiety Disorders, 21*(1), 105–117. <https://doi:10.1016/j.janxdis.2006.03.014>.
- Carleton, R., Mulvogue, M., Thibodeau, M., McCabe, R., Antony, M., & Asmundson, G. (2012). Increasingly certain about uncertainty: intolerance of uncertainty across anxiety and depression. *Journal of Anxiety Disorders, 26*(3), 468–479. <https://doi:10.1016/j.janxdis.2012.01.011>.
- Carleton, N., Thibodeau, M., Welch, P., Abrams, M., Robinson, T., & Asmundson, G. (2013). The Centre for epidemiologic studies Depression Scale: A review with a theoretical and empirical examination of item content and factor structure. *Plos One, 8*(3) <https://doi.org/10.1371/journal.pone.0058067>.
- *Carpentier, M., Larry, L., Mullins, J., & Van Pelt, J. (2007) Psychological, Academic, and Work Functioning in College Students with Childhood-Onset Asthma. *Journal of Asthma, 44*(2), 119-124. <https://doi:10.1080/02770900601182418>.

- Carver, C., Scheier, M., & Weintraub, J. (1989). Assessing coping strategies: A theoretically based approach. *Journal of Personality and Social Psychology*, 56(2), 267-283.
<http://doi.www.psy.miami.edu/faculty/ccarver/documents/p89COPE.pdf>.
- Centre for reviews & dissemination (2009). *Systematic reviews: CRD's guidance for undertaking reviews in healthcare*. CRD University of York.
https://www.york.ac.uk/media/crd/Systematic_Reviews.pdf
- Chen, T., Kao, C., Cheng, S., & Chang, Y. (2018). Uncertainty and depressive symptoms as mediators of quality of life in patients with heart failure. *PLoS ONE*, 14(3), Article e0214825. <https://doi.org/10.1371/journal.pone.0214825>.
- Cho, M., Bae, J., Suh, G., Hahm, B., Kim, J., Lee, D., & Kang, M. (1999). Validation of Geriatric Depression Scale, Korean version (GDS) in the assessment of DSM-III-R major depression. *Journal of Korean Neuropsychiatry Association*, 38(1), 48–63.
- Christman, N., McConnell, E., Pfeiffer, C., Webster, K., Schmitt, M., & Ries, J. (1988). Uncertainty, coping, and distress following myocardial infarction: transition from hospital to home. *Research Nursing Health*, 11(2), 71-82.
<https://doi.org/10.1002/nur.4770110203>.
- Christman, N. (1990). Uncertainty and adjustment during radiotherapy. *Nursing Research*, 39, 17–20.
- Cohen, M., Mansoor, D., Langut, H., & Lorber, A. (2007). Quality of life, depressed mood, and self-esteem in adolescents with heart disease. *Psychosomatic Medicine*, 69(4), 313-8.
<https://doi:10.1097/PSY.0b013e318051542c>.

Cohen, J. (1988). *Statistical power analysis for the behavioural sciences (2nd ed.)*. Lawrence Erlbaum Associates.

*Colagreco, P., Bailey, D., Fitzpatrick, J., Musil, C., Afdgal, N., & Lai, M. (2014). Watchful waiting: role of disease progression on uncertainty and depressive symptoms in patients with chronic Hepatitis. *Journal of Viral Hepatitis*, *21*(10), 727–733.
<https://doi:10.1111/jvh.12207>.

Department of Health (DOH) (2017). Chronic conditions.

<https://www1.health.gov.au/internet/main/publishing.nsf/Content/chronic-disease>.

Derogatis, L. (1992). *SCL-90-R: Administration, Scoring & Procedures Manual-II, for the R (Revised) Version and Other Instruments of the Psychopathology Rating Scale Series*. 2nd Edition, Clinical Psychometric Research, Inc.

Derogatis, L. (1993). *Brief Symptom Inventory: Administration, Scoring, and Procedures Manual*. National Computer Systems, Inc.

*Detprapon, M., Sirapo-ngam, Y., Mishel, M., Sitthimongkol, Y., & Vorapongsathorn, T. (2009). Testing of Uncertainty in Illness Theory to predict quality of life among Thais with head and neck cancer. *Journal of Nursing Research*, *13*(1), 1-15.

Devins, G., Binik, Y., Hutchinson, T., Hollomby, D. J., Barre, P., & Guttman, R. (1983). The emotional impact of end-stage renal disease: importance of patients' perceptions of intrusiveness and control. *International Journal of Psychiatric Medicine*, *13*(4), 327–343. <https://doi:10.2190/5dcp-25bv-u1g9-9g7c>.

Dibley, L., Norton, C., & Whitehead, E. (2018). The experience of stigma in inflammatory bowel disease: An interpretative (hermeneutic) phenomenology study. *Global Nursing Research, 74*(4), 838-851. <https://doi.org/10.1111/jan.13492>.

Department of Health (2017). Chronic conditions.

<https://www1.health.gov.au/internet/main/publishing.nsf/Content/chronic-disease>.

Dorrian, A., Dempster, M., & Adair, P. (2009). Adjustment to inflammatory bowel disease: The relative influence of illness perceptions and coping. *Inflammatory Bowel Diseases, 15*(1), 47-55. <https://doi:10.1002/ibd.20583>.

Drapeau, A., Marchand, A., & Beaulieu-Pre´vost, D. (2012). Epidemiology of psychological distress. *Mental illnesses - understanding, prediction and control* (pp. 105). www.intechopen.com: InTech. Doi:10.5772/30872.

Duckworth, A., Peterson, C., Matthews, M., & Kelly, D. (2007). "Grit: Perseverance and passion for long-term goal," *Journal of Personality and Social Psychology, 92*(6), 1087–1101. <https://doi:10.1037/0022-3514.92.6.1087>.

Duckworth, A., & Quinn, P. (2009). Development and validation of the Short Grit Scale (GRIT–S). *Journal of Personality Assessment, 91*(2), 166–174. <https://doi.org/10.1080/00223890802634290#>.

Dugas, M., Freeston, M., & Ladouceur, R. (1997). Intolerance of uncertainty and problem orientation in worry. *Cognitive Therapy and Research, 21*(6), 593–606. <https://doi.org/10.1023/A:1021890322153>.

Ellis, P. (2010). *The essential guide to effect sizes: Statistical power, meta-analysis, and the interpretation of research results*. Cambridge University Press.

- Fan, S., Eiser, C., & Ho, M. (2010). Health-related quality of life in patients with hepatocellular carcinoma: a systematic review. *Clinical Gastroenterology Hepatology*, 8(7), 559-564. <https://doi.org/10.1016/j.cgh.2010.03.008>.
- Fedele, D., Mullins, L., Eddington, A., Ryan, L., Junghans, A., & Hullman, S. (2009). Health-related quality of life in college students with and without childhood-onset asthma. *Journal of Asthma*, 46(8), 835-840. <https://doi:10.1080/02770900903184229>.
- Fortier, M., Batista, M., Wahi, A., Kain, A., Strom, S., & Leonard, S. (2013). Illness uncertainty and quality of life in children with cancer. *Journal of Pediatric Hematology Oncology*, 35(5), 366-370. <https://doi:10.1097/MPH.0b013e318290cfdb>.
- Freeston M. H., Rhéaume J., Letarte H., Dugas M. J., Ladouceur R. (1994). Why do people worry? *Personality Individual Differences*, 17, 791–802. [https://10.1016/0191-8869\(94\)90048-5](https://10.1016/0191-8869(94)90048-5).
- Gentes, E., & Ruscio, A. (2011). A meta-analysis of the relation of intolerance of uncertainty to symptoms of generalized anxiety disorder, major depressive disorder, and obsessive-compulsive disorder. *Clinical Psychology Review*, 31(6), 923-933. <https://doi:10.1016/j.cpr.2011.05.001>.
- Gong, Y., Palmer, S., Gallacher, J., Marsden, T., & Fone, D. (2016). A systematic review of the relationship between objective measurements of the urban environment and psychological distress. *Environment Int*, 96, 48-57. <https://doi:10.1016/j.envint.2016.08.0109>.
- Green, C., & Murdock, K. (2013). Multidimensional control beliefs, Socioeconomic status, and health. *American Journal of Health Behaviour*, 37(2), 227-37. <https://doi:10.5993/AJHB.37.2.10>.

- Hagen, K, Aas, T., Lode, K, Gjerde, J., Lien, E., Terje, J., Lash, T., Soiland, H., & Lind, R. (2015). Illness uncertainty in breast cancer patients: validation of the 5-item short form of the Mishel Uncertainty in Illness scale. *European Journal of Oncology Nursing*, 19(2), 113-119. <https://doi:10.1016/j.ejon.2014.10.009>.
- Hilton, B. (1992). Perceptions of uncertainty. Its relevance to life-threatening and chronic illness. *Critical Care Nurse*, 12(2), 70-73. <https://doi:10.4037/ccn1992.12.2.70>.
- Hilton, B. (1994). The uncertainty stress scale: Its development and psychometric properties. *Canadian Journal of Nursing Research*, 26(3), 15-30.
- *Hommel, K., Chaney, J., Wagner, J., White, M., Hoff, A., & Mullins, L. (2003). Anxiety and Depression in Older Adolescents with Long Standing Asthma: The Role of Illness Uncertainty. *Children's Health Care*, 32(1), 51-63, https://doi.org/10.1207/S15326888CHC3201_4.
- *Hoth, K., Wamboldt, F., Strand, M., Ford, D., Sandhaus, R., Strange, C., Bekelman, D., & Holm, K. (2013). Prospective Impact of Illness Uncertainty on outcome in chronic lung disease. *Health Psychology*, 32(11), 1170-1174. <https://doi:10.1037/a0032721>.
- Hulley, S., Cummings, S., Browner, W., Grady, D., & Newman, T. (2013). *Designing Clinical Research*. Lippincott Williams and Wilkins.
- Hullman, S., Eddington, A., Molzon, E., & Mullins, L. (2013). Illness appraisals and health-related quality of life in adolescents and young adults with allergies and asthma. *International Journal of Adolescent Medicine and Health*, 25(1), 31–38. <https://doi.10.1515/ijamh-2013-0004>.

- Jacqueline, B., & Lisa, S. (2015). Developing and using a case formulation to guide cognitive-behaviour therapy. *Journal of Psychology and Psychotherapy*, 5(3), 179.
<https://doi:10.4172/2161-0487.1000179>.
- Joober, R., Schmitz, N., Annable, L., & Boksa, P. (2012). Publication bias: What are the challenges, and can they be overcome? *Journal Psychiatry Neuroscience*, 37(3), 149-152. <https://doi:10.1503/jpn.120065>.
- Jung, C., Kim, M., Rhee, M., & Do, H. (2005). Functional status and psychosocial adjustment in gynecologic cancer patients receiving chemotherapy. *Korean Journal of Women Health Nursing*, 11(1), 58–66. <https://doi:10.4069/kjwhn.2005.11.1.58>.
- *Kim, B., & Kim, J. (2019). Influence of Uncertainty, Depression, And Social Support on Self-Care Compliance in Hemodialysis Patients. *Therapeutics and Clinical Risk Management*, 15, 1243-1251. <https://doi:10.2147/TCRM.S218934>.
- Kroenke, M., Spitzer, R., & Williams, J. (2001). The PHQ-9 validity of a brief depression severity measure. *Journal of General Internal Medicine*, 16(9), 606-613.
<http://dx.org/10.1046/j.1525-1497.2001.016009606.x>.
- *Kurita, K., Garon, E., Stanton, A., & Meyerowitz, B. (2013). Uncertainty and psychological adjustment in patients with lung cancer. *Psychooncology*, 22(6), 1396–1401.
<https://doi:10.1002/pon.3155>.
- Kuswanto, C., Stafford, L., Sharp, J., & Schofield, P. (2018). Psychological distress, role, and identity changes in mothers following a diagnosis of cancer: A systematic review. *Psychooncology*, 27(12), 2700-2708. <https://doi:10.1002/pon.4904>.
- Lazarus, R., & Folkman, S. (1984). *Stress, Appraisal, and Coping*. Springer.

- * Lee, Y. (2006). The relationships between uncertainty and posttraumatic stress in survivors of childhood cancer. *Journal of Nursing Research, 14*(2), 133-142.
<https://doi:10.1097/01.jnr.0000387571.20856.45>.
- *Lemaire, S. (2004). More than just menstrual cramps: Symptoms and uncertainty among women with endometriosis. *Clinical Research, 33*, 71-79.
<https://doi:10.1177/088421750326105>.
- Levenson, H. (1974). Activism and powerful others: Distinctions within the concept of internal-external control. *Journal of Personality Assessment, 38*(4), 377-383.
<https://doi.org/10.1080/00223891.1974.10119988>.
- Li, X., He, L., Wang, J., & Wang, M. (2019). Illness uncertainty, social support, and coping mode in hospitalized patients with systemic lupus erythematosus in a hospital in Shaanxi, China. *PLoS ONE, 14*(2). <https://doi:10.1371/journal.pone.0211313>.
- Liao, M., Chen, M., Chen, S., & Chen, P. (2008). Uncertainty and anxiety during the diagnostic period for women with suspected breast cancer. *Cancer Nursing, 31*(4), 274-83.
<https://doi:10.1097/01.NCC.0000305744.64452.fe>.
- Lien, C., Lin, H., Kuo, I., & Chen, M. (2009). Perceived uncertainty, social support and psychological adjustment in older patients with cancer being treated with surgery. *Journal of Clinical Nursing, 18*(16), 2311-9. <https://doi:10.1111/j.1365-2702.2008.02549.x>.
- Lin, L., Acquaye, A., Vera-Bolanos, E., Cahill, J., Gilbert, M., & Armstrong, T. (2012). Validation of the Mishel's Uncertainty in Illness Scale-Brain Tumor Form (MUIS-BT). *Journal of Neurooncology, 110*, 293-300. [https:// DOI: 10.1007/s11060-012-0971-8](https://doi:10.1007/s11060-012-0971-8)

- *Lin, L., Acquaye, A., Vera-Bolanos, E., Gilbert, M., & Armstrong, T. (2012). Uncertainty, Mood States, and Symptom Distress in Patients with Primary Brain Tumours Analysis of a Conceptual Model Using Structural Equation Modelling. *Cancer, 119*(15), 2796-806. <https://doi:10.1002/cncr.28121>.
- Lisy, K., & Porritt, K. (2016). Narrative synthesis. Considerations and challenges. *International Journal of Evidence-Based Healthcare, 14*(4), 201. <https://doi:10.1097/01.XEB.0000511348.97198c>.
- Lutze, U., & Archenholtz, B. (2007). The impact of arthritis on daily life with the patient perspective in focus. *Scandinavian Journal of Caring Sciences, 21*(1), 64-70. <https://doi:10.1111/j.1471-6712.2007.00443x>.
- Mast, M. (1995). Adult uncertainty in illness: a critical review of research. *Scholarly Inquiry for Nursing Practice, 9*, 3–24; discussion 25–29.
- Mast, M. (1998). Survivors of breast cancer: Illness uncertainty, positive reappraisal, and emotional distress. *Oncology Nursing Forum, 25*(3), 555-62.
- McNulty, K., Livneh, H., & Wilson, L. (2004). Perceived Uncertainty, Spiritual Well-Being, and Psychosocial Adaptation in Individuals with Multiple Sclerosis. *Rehabilitation Psychology, 49*(2), 91–99. <https://doi.org/10.1037/0090-5550.49.2.91>.
- McCormick, K. (2002). A concept analysis of Uncertainty in Illness. *Journal of Nursing Scholarship, 34*(2), 127-131. <https://doi.org/10.1111/j.1547-5069.2002.00127.x>.
- Mehrtak, M., Habibzadeh, S., Farzaneh, E., & Rjaei0Khiavi, A. (2017). Effectiveness of teaching cognitive-behavioural techniques on locus of control in hemodialysis patients. *Electron Physician, 9*(10), 5631-5637. <https://doi:10.19082/5631>.

- Mertens, A., Yasui, Y., Neglia, J., Potter, J., Nesbit, M., & Ruccione, K. (2015). Late mortality experience in five-year survivors of childhood and adolescent cancer: The Childhood Cancer Survivor Study. *The Journal of Clinical Oncology*, *19*(13), 3163-3172. <https://doi:10.1200/JCO.2001.19.13.3163>.
- Mishel, M. (1981). The measurement of uncertainty in illness. *Nursing Research*, *30*, 258–263. <http://dx.doi.org/10.1097/00006199-198109000-00002>.
- Mishel, M. (1983). Parents' perception of uncertainty concerning their hospitalized child: Reliability and validity of a scale. *Nursing Research*, *32*(6), 324–330.
- Mishel, M. (1984). Perceived uncertainty and stress in illness. *Research for Nursing Health*, *7*, 163–171. <https://doi.org/10.1002/nur.4770070304>.
- Mishel, M., Hostetter, B., & Graham, K. (1984). Predictors of psychosocial adjustment in patients newly diagnosed with gynecological cancer. *Cancer Nursing*, *7*(4), 291-9.
- Mishel, M., & Baden, C. (1987). Uncertainty as a mediator between support and adjustment. *Western Journal of Nursing Research*, *17*(9), 43–57. <https://doi:10.1177/019394598700900106>.
- Mishel, M. (1988). Uncertainty in illness. *Image: Journal of Nursing Scholarship*, *20*(4), 225-232. <https://doi:10.1111/j.1547-5069.1988.tb00082.x>.
- Mishel, M. (1990). Reconceptualization of the uncertainty in illness theory. *Image – The Journal of Nursing Scholarship*, *22*(4), 256–262. <https://doi:10.1111/j.1547-509.1990.tb00225.x>.

- Mishel, M., Padilla, G., Grant, M., & Sorenson, D. (1991). Uncertainty in illness theory: A replication of the mediating effects of mastery and coping. *Nursing Research, 40*, 236–240.
- Mishel, M., & Sorenson, D. (1991). Uncertainty in gynecological cancer: A test of the mediating functions of mastery and coping. *Nursing Research, 40*, 161–171.
- Moher, D., Liberati, A., Tetzlaff, J., Altman, D., & Group, T. (2009). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA Statement. *Public Library of Science Medicine, 6*(7), Article e1000097.
<https://doi.org/10.1371/journal.pmed.1000097>.
- Moher, D., Shamseer, L., Clarke, M., Ghersi, D., Liberati, A., Petticrew, M., Shekelle, P., Stewart, L. (2015). Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Systematic Reviews, 4*(1), 1-9.
<https://doi.org/10.1186/2046-4053-4-1>.
- Moreland, P., & Santacroce, S. (2018). Illness uncertainty and posttraumatic stress in young adults with congenital heart disease. *Journal of Cardiovascular Nursing, 33*(4), 356-362. <https://doi:10.1097/JCN.0000000000000471>
- *Mullins, L., Chaney, J., Balderson, B., & Hommel, K. (2000). The Relationship of Illness Uncertainty, Illness Intrusiveness, and Asthma Severity to Depression in Young Adults with Long-Standing Asthma. *International Journal of Rehabilitation and Health, 5*(3), 177-186. <https://doi.org/10.1023/A:1012999205376>.
- *Mullins, L., Cote, M., Fuemmeler, B., Jean, V., Beatty, W., & Paul, R. (2001). Illness intrusiveness, uncertainty, and distress in individuals with multiple sclerosis. *Rehabilitation Psychology, 46*(2), 139–153. <https://doi:10.1037//0090-5550.46.2.139>.

*Mullins, A., Gamwell, K., Sharkey, K., Bakula, C., Tackett, D., Suorsa, A., Chaney, K., & Mullins, J. (2017). Illness uncertainty and illness intrusiveness as predictors of depressive and anxious symptomology in college students with chronic illnesses. *Journal of American College Health, 65*(5), 352- 360.
<https://doi:10.1080/07448481.2017.1312415>.

National Heart Lung, and Blood Institutes (NHLBI) (2014). *Study Quality Assessment Tools*. Quality assessment tool for Observational Cohort and Cross-sectional Studies tool.
<https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools>.

National Institute for Health and Care Excellence. (2010). *Depression in adults with a chronic health condition (guideline 91)*. <https://www.nice.org.uk/guidance/cg91/evidence/full-guideline-243876061>

Nayak, B. (2010). Understanding the relevance of sample size calculation. *Indian Journal of Ophthalmology, 58*, 469-470. <https://doi: 10.4103/0301-4738.71673>.

Niv, G., Josef, B., Bassat, B., Avni, I., Lichtenstein, L., Niv, Y., & Barnoy, S. (2017). Quality of life and uncertainty in Crohn's disease. *Quality of Life Research, 26*(6), 1609-1616.
<https://doi:10.1007/s11136-017-1509-5>.

Padilla, G., Mishel, M., & Grant, M. (1992). Uncertainty, appraisal, and quality of life. *Quality of Life Research, 1*(3), 155-165. <https://www.jstor.org/stable/4034464>.

*Pahlevan, S. (2017a). Locus of control, quality of life, anxiety, and depression among Malaysian breast cancer patients: The mediating role of uncertainty. *European Journal of Oncology Nursing, 27*, 28-35. <https://doi:10.101/j.ejon.2017.01.005>.

- *Pahlevan, S., Ahadzadeh, A., & Perdamen, H. (2017b). Uncertainty and quality of life of Malaysian women with breast cancer: Mediating role of coping styles and mood states. *Applied Nursing Research*, 38, 88-94. <https://doi:10.1016/j.apnr.2017.09.012>.
- Park, C., & Gaffey, A. (2007). Locus of control, quality of life, anxiety and depression among Malaysian breast cancer patients: The mediating role of uncertainty. *European Journal of Oncology Nursing*, 27, 28-35. <https://dx.doi.org.10.1016/j.ejon.2017.01.005>.
- Podsakoff, P., MacKenzie, S., Lee, J., & Podsakoff, N. (2003). Common method biases in behavioural research: A critical review of the literature and recommended remedies. *Journal of Applied Psychology*, 88(5), 879-903. <https://doi.org/10.1037/0021-9010.88.5.879>.
- Popay, J., Arai, L., Rodgers, M., & Britten, N. (2006). *Guidance on the conduct of narrative synthesis in systematic reviews*. A product from the ESRC Methods programme. [file:///C:/Users/Katie/Downloads/download%20\(1\).pdf](file:///C:/Users/Katie/Downloads/download%20(1).pdf).
- Prince, M., Patel, V., Saxena S., Maj, M., Maselko, J., Phillips, M., & Rahman, A. (2011). Global Mental Health 1. No health without mental health HM government. *Lancet*, 370, 859–77 [https://doi:10.1016/S0140-6736\(07\)61238-0](https://doi:10.1016/S0140-6736(07)61238-0).
- Pynoos, R., Rodriguez, N., Steinberg, A., Stuber, M., & Frederick, C. (1998). *UCLA PTSD index for DSM-IV: Adolescent version*. Los Angeles: University of California at Los Angeles Trauma Psychiatry Service.
- Radloff, L. (1977). The CES-D Scale: A self-report depression scale for research in the general population. *Applied Psychology Measurement*, 1, 385-401. <https://doi.org/10.1177/014662167700100306>.

Ridder, S. (2004). Psychological Distress: Concept Analysis. *Journal of Advanced Nursing*, 45(5), 536-45. <https://doi:10.1046/j.1365-2648.2003.02938.x>.

Rosenbaum, M. (1983). Learned resourcefulness as a behavioural repertoire for the self-regulation of internal events: Issues and speculations. In Rosenbaum, C., Franks, & Jaffe, Y. (Eds.). *Perspective on behaviour therapy in the eighties* (pp. 51-73). Springer.

Roth, A., Rosenfeld, B., Kornblith, A., Gibson, C., Scher, H., Curley-Smart, T., Holland, J., & Breitbart, W. (2003). The memorial anxiety scale for prostate cancer. Validation of a new scale to measure anxiety in men with prostate cancer. *Cancer*, 97(11), 2910-8. <https://doi.org/10.1002/cncr.11386>.

*Sanders-Dewey, N., Mullins, L., & Chaney, J. (2001). Coping style, perceived uncertainty in illness, and distress in individuals with Parkinson's disease and their caregivers. *Rehabilitation Psychology*, 46(4), 363-381. <https://doi:10.1037/0090-5550.46.4.363>.

*Santacroce, J., & Lee, Y. (2006). Uncertainty, Posttraumatic Stress, and Health Behaviour in Young Adult Childhood Cancer Survivors. *Nursing Research*, 55(4), 259-266. <https://doi:10.1097/00006199-200607000-00006>.

Setia, M. (2016), Methodology series module 3: Cross-sectional studies. *Indian Journal of Dermatology*, 61, 261-264. <https://doi:10.4103/0019-5154.182410>.

Sharkey, C., Bakula, D., Gamwell, K., Mullins, A., Chaney, J., & Mullis, L. (2017). The role of grit in college student health care management skills and health-related quality of life. *Journal of Pediatric Psychology*, 42(9), 952-961. <https://doi:10.1093/jpepsy/jsx073>.

- *Sharkey, M., Bakula, D., Baraldi, A., Perez, M., Suorsa, K., Chaney, J., & Mullins, L. (2018). Grit, Illness-Related Distress, and Psychosocial Outcomes in College Students with a Chronic Medical Condition: A Path Analysis. *Journal of Pediatric Psychology, 43*(5), 552-560. <https://doi.10.1093/jpepsy/jsx145>.
- Sirois, F., & Hirsch, J. (2017). A longitudinal study of the profiles of psychological thriving, resilience, and loss in people with inflammatory bowel disease. *British Journal of Health Psychology, 22*(4), <https://doi:10.1111/bjhp.12262>.
- *Small, S., & Graydon, J. (1992). Perceived uncertainty, physical symptoms, and negative mood" in hospitalized patients with chronic obstructive pulmonary disease. *Heart Lung, 21*(6), 568-574.
- Snippen, N., Vries, H., Burg-Vermeulen, S., Hagedoorn, M., & Brouwer, S. (2019). Influence of significant others on work participation of individuals with chronic diseases: a systematic review. *British Medical Journal Open, 9*(1), e021742. <https://doi:10.1136/bmjopen-2018-021742>.
- Szulczewski, L., Mullins, L., Bidwell, S., Eddington, A., & Pai, A. (2017). Meta-analysis: Caregiver and youth uncertainty in pediatric chronic illness. *Journal of Pediatric Psychology, 42*(4), 395–421. <https://doi:10.1093/jpepsy/jsw097>.
- Wang, Q., Hay, M., Clarke, D., & Menahem, S. (2014). Associations between knowledge of disease, depression and anxiety, social support, sense of coherence and optimism with health-related quality of life in an ambulatory sample of adolescents with heart disease. *Cardiology Young, 24*(1), 126–133. <https://doi.org/10.1017/S1047951113000012>.

- Wasilewski-Masker, K., Yasui, Q., Leisenring, W., Meacham, L., Hammond, S., Meadows, A., Robison, L., & Mertens, A. (2009). Later recurrence in pediatric cancer: A report from the childhood cancer survivor study. *Journal of National Cancer Institute, 101*(24), 1809-1720. <https://doi:10.1093/jnci/dip417>.
- Wineman, N. (1990). Adaptation to multiple sclerosis: The role of social support functional disability, and perceived uncertainty. *Nursing Research, 39*(5), 294-299. <https://doi.org/10.1097/00006199-199009000-00013>.
- Wineman, N., Schwetz, K., Zeller, R., & Cyphert, J. (2003). Longitudinal analysis of illness uncertainty, coping, hopefulness and mood during participation in a clinical drug trial. *Journal of Neuroscience Nursing, 35*(2), 100–106. <https://doi:10.1097/01376517-200304000-00006>.
- Wiener, C., & Dodd, M. (1993). Coping amid uncertainty: an illness trajectory perspective. *School Inquiry Nursing Practice, 7*, 17–31; discussion 33–35.
- *Wolfe-Christensen, C., Isenberg, J., Mullins, L., Carpentier, M., & Almstrom, C. (2008). Objective versus subjective ratings of asthma severity: Differential predictors of illness uncertainty and psychological distress in college students with asthma. *Children's Health Care, 37*, 183. <https://doi:10.1080/02739610802151514>.
- World Health Organisation, WHO (2016). Noncommunicable diseases <https://www.who.int/westernpacific/health-topics/noncommunicable-diseases>.
- Wright, L., Afari, N., & Zautra, A. (2009). The illness uncertainty concept: A review. *Current Pain and Headache Reports, 13*(2), 133-8. <https://doi:10.1007/s11916-009-0023-z>.

- Wu, A., & Fang, Y. (2014). Comorbidity and depressive and anxiety disorders: challenges in diagnosis and assessment. *Shanghai Arch Psychiatry, 26*(4), 227-231.
<https://doi:10.3969/j.issn.1002-0829.2014.04.006>.
- Yarcheski, A. (1988). Uncertainty in illness and the future. *Western Journal of Nursing Research, 10*(4), 401-413. <https://doi:10.1177/019394598801000404>.
- Zigmond, A., & Snaith, R. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica, 67*(6), 361–370. <https://doi:10.1111/j.1600-0447.1983.tb09716.x>.
- Zimet, G., Dahlem, N., Zimet, S., & Farley, G. (1988). The multidimensional scale of perceived social support. *Journal of Personality Assessment, 52*(1), 30–41.
https://doi:10.1207/s15327752jpa5201_2.
- Zimmerman, M., & Coryell, W. (1988). The validity of a self-report questionnaire for diagnosing major depressive disorder. *Archives of General Psychiatry, 45*(8), 738–740.
<https://doi.org/10.1001/archpsyc.1988.01800320050006>.
- Zung, W. (1971). A rating instrument for anxiety disorders. *Psychosomatics, 12*(6), 371–379.
[https://doi:10.1016/S0033-3182\(71\)71479-0](https://doi:10.1016/S0033-3182(71)71479-0).

Appendix A

Search strategy: search terms

Search terms entered into all search engines Scopus, Psycinfo and MEDLINE

Population	Target variable	Psychological outcome
“chronic* ill*” OR	“illness uncertainty” OR	“psychological distress*”
“chronic condition*” OR	“Mishel* illness	OR “psychological stress”
“chronic* disease*” OR	uncertainty theory” OR	OR depression OR
“chronically critically ill”	“Mishel* illness	“depressive symptom*”
OR “chronic patient*” OR	uncertainty scale” OR	OR “depressive disorder”
“noncommunicable	MUIS OR uncertainty	OR “emotional distress”
disease*” OR NCD		OR “mood state*” OR
OR illness OR disease OR		“negative mood” OR
“disabled persons” OR		“negative affect” OR
LTC OR “long term		anxiety OR anxious OR
condition*” OR arthritis		loneliness OR PTSD OR
OR asthma OR “brain		“posttraumatic stress
injuries” OR cancer OR		disorder” OR distress OR
“cardiovascular disease*”		“psychological well-
OR “chronic obstructive		being” OR “psychosocial
pulmonary disease*” OR		adjustment” OR
COPD OR CVD OR		“psychological
“diabetes mellitus” OR		adjustment” OR “common
diabetes OR epilepsy OR		mental disorder”
HIV OR		
hypercholesterolemia OR		

Population	Target variable	Psychological outcome
hypertension OR “inflammatory bowel disease” OR “interstitial cystitis” OR lupus OR “musculoskeletal disorder” OR “multiple sclerosis” OR “parkinsons disease” OR “sickle cell disease” OR stroke		

Note. COPD=chronic obstructive pulmonary disease; CVD=cardiovascular disease; HIV=Human immunodeficiency virus; LTC=long term condition; MUIS=Mishel’s illness uncertainty scale; NCD=noncommunicable disease.

Appendix B

Search strategy: Inclusion/exclusion criteria

Table presenting inclusion and exclusion criteria

Inclusion	Exclusion
<p>Over the age of 18 years and individuals with a diagnosed chronic health condition defined by DOH (2017) and WHO (2016) (and defined diagnostically by the medical profession)</p> <p>The paper includes the measurement of PD as defined in previous research as a broad concept (Gong et al., 2016; Kuswanto et al., 2018)</p> <p>A Quantitative methodology was used including cross-sectional, correlational, prospective and longitudinal studies.</p> <p>The direct relationship between IU and PD is statistically examined.</p>	<p>Populations with conditions that do not fit the definition of a diagnosable chronic health condition, such as medically unexplained symptoms, chronic pain conditions or diagnoses of exclusion (e.g. fibromyalgia, chronic fatigue syndrome);</p> <p>Papers that are studying an acute self-limiting condition, other non-chronic physical health illness or chronic mental illnesses</p> <p>Papers studying participants under age 18 or where the data is not separate for the adult age group and the child/adolescent age group</p>

Inclusion criteria	Exclusion criteria
The paper is published in a peer-reviewed journal.	The paper uses proxy measures of individual uncertainty (e.g. child illness uncertainty, parent uncertainty or carer uncertainty)
The paper is written in the English language.	Outcomes measures that do not constitute PD (see introduction)
IU is measured using a version of or subscale of the MUIS (e.g. Hagen et al., 2011; Mishel, 1981; Mishel, 1986)	Measurement of IU immediately before and/or after participants are undergoing a major medical procedure/surgery/drug trial.
The full-text paper is possible to obtain following reasonable means to obtain it	Non-peer reviewed papers such as grey literature, conference abstracts, case studies or unpublished theses
	Studies employing a qualitative methodology

Appendix C

Psychometric properties of MUIS measures used in the studies included in this review

MUIS-Acute (MUIS-A, Mishel, 1981)

The MUIS-A which has been available since 1981 has undergone repeated psychometric evaluation (Bailey et al., 2009). Cronbach's alphas of the four subscales, ambiguity (13 items), complexity (7 items), inconsistency (7 items) and unpredictability (5 items) were 0.86, 0.81, 0.78 or 0.65 respectively (Mishel, 1997); and a Cronbach's alpha of 0.87 for the whole scale (Mishel, 1997). The full scale MUIS-A was found to be 0.90 in a study with those with Hepatitis C (Bailey et al., 2009). The Cronbach's alpha was 0.84 in another study researching those with Hepatitis C (Colagreco et al., 2014). The Cronbach's alpha was 0.86 in a study on a population of people with chronic renal disease (Barberis et al., 2019). The Cronbach's alphas for the ambiguity and complexity subscales were 0.88 and 0.79 respectively in a study with individuals with chronic lung disease (Hoth et al., 2013).

The 33-item MUIS-brain tumour form (MUIS-BT, Lin et al., 2012).

Six out of 33 items in the original MUIS-A were modified to better suit the experience of people with primary brain tumours and uncertainty. Cronbach's alphas of the four subscales, ambiguity/inconsistency, unpredictability of disease prognosis, unpredictability of symptoms and complexity were 0.90, 0.77, 0.75 and 0.65, respectively for those with brain tumours (Lin et al., 2012).

The 33-item Korean MUIS (Jung et al., 2005)

The Korean MUIS was found to have a Cronbach's alpha on 0.91 in people with gynaecologic cancer patients (Jung et al., 2005) and 0.79 in a study researching people with chronic renal disease on haemodialysis (B. Kim & J. Kim, 2019).

MUIS-Community (MUIS-C, Mishel, 1997)

MUIS-C scores across 18 adult samples with CI's (total n=1068) were examined and Cronbach's alpha ranged from 0.53-0.92; values exceeded 0.85 in the majority of the samples (Mishel, 1997), comparable to the MUIS-A. Bailey et al. (2011) conducted a secondary analysis examining the reliability of the MUIS-C for use with men undergoing active surveillance for prostate cancer and the Cronbach's alpha for the full MUIS-C was .908. The Cronbach's alphas collected from 20 studies with people with CI's (Mishel, 1997, cited in Carpentier et al., 2009) ranged from 0.74 to 0.92.

MUIS-short form (MUIS-SF, Hagen et al., 2015)

The 5-item MUIS-SF was found to have a Cronbach's alpha of 0.70 in patients with breast cancer. In two studies researching women with breast cancer (Pahlevan, 2017a; Pahlevan et al., 2017b) the Cronbach's alpha were found to be 0.50 and 0.56, respectively.

Appendix D
Quality appraisal tool

The Observational Cohort and Cross-sectional studies tool (National Institutes of Health (2014). Observational Cohort and Cross-sectional studies tool.

<https://www.nhlbi.nih.gov/health-topics/study-quality-assessment-tools>

Criteria	Yes	No	Other (CD, NR, NA)*
1. Was the research question or objective in this paper clearly stated?			
2. Was the study population clearly specified and defined?			
3. Was the participation rate of eligible persons at least 50%?			
4. Were all the subjects selected or recruited from the same or similar populations (including the same time period)? Were inclusion and exclusion criteria for being in the study prespecified and applied uniformly to all participants?			

Criteria	Yes	No	Other (CD, NR, NA)*
5. Was a sample size justification, power description, or variance and effect estimates provided?			
6. For the analyses in this paper, were the exposure(s) of interest measured prior to the outcome(s) being measured?			
7. Was the timeframe sufficient so that one could reasonably expect to see an association between exposure and outcome if it existed?			
8. For exposures that can vary in amount or level, did the study examine different levels of the exposure as related to the outcome (e.g., categories of exposure, or exposure measured as continuous variable)?			
9. Were the exposure measures (independent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
10. Was the exposure(s) assessed more than once over time?			

Criteria	Yes	No	Other (CD, NR, NA)*
11. Were the outcome measures (dependent variables) clearly defined, valid, reliable, and implemented consistently across all study participants?			
12. Were the outcome assessors blinded to the exposure status of participants?			
13. Was loss to follow-up after baseline 20% or less?			
14. Were key potential confounding variables measured and adjusted statistically for their impact on the relationship between exposure(s) and outcome(s)?			

Quality Rating (Good, Fair, or Poor)

Rater #1 initials:

Quality Rating (Good, Fair, or Poor)

Rater #2 initials:

Additional Comments (If POOR, please state why):

*CD, cannot determine; NA, not applicable; NR, not reported

Appendix E

Quality appraisal of all studies

Table of Quality appraisal ratings for each item on the observational cohort and cross-sectional studies appraisal tool (items 1-14, see Appendix D).

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Score (%) and rating
Ahn et al. (2017)	Green	Green	CD	Green	Green	Red	Red	NA	Green	NA	Green	NA	NA	Green	70% GOOD
Bailey et al. (2009)	Green	Green	Green	Green	Red	Red	Red	Green	Green	NA	Green	NA	NA	Green	72.7% GOOD

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Score (%) and rating
Barberis et al. (2019)			NR		NR			NA		NA			NA		54.5% FAIR
Carpentier et al. (2007)								NA		NA		NA	NA		80% GOOD
Colagreco et al. (2014)								NA				NA			50% FAIR
Detprapon et al. (2009)			NR					NA		NA		NA	NA		55.6% FAIR

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Score (%) and rating
Hoth et al. (2013)								NA				NA			75% GOOD
Hommel et al. (2003)			CD							NA		NA	NA		60% FAIR
B. Kim & J. Kim (2019)			CD					NA		NA		NA	NA		60% FAIR
Kurita et al. (2013)								NA		NA		NA	NA		70% GOOD
Lee (2006)								NA		NA		NA	NA		60% FAIR

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Score (%) and rating
Lemaire (2004)								NA		NA		NA	NA		60% FAIR
Lin et al. (2013)			CD		CD			NA		NA			NA		63.6% FAIR
Mullins et al. (2000)			CD			NA	NA			NA		NA	NA		66.7% FAIR
Mullins et al. (2001)								NA		NA		NA	NA		70% GOOD
Mullins et al. (2017)			CD					NA		NA		NA	NA		60% Fair

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Score (%) and rating
Pahlevan (2017a)								NA		NA		NA	NA		80% GOOD
Pahlevan et al. (2017b)			CD					NA		NA		NA	NA		60% FAIR
Santacroce & Lee (2006)								NA		NA		NA	NA		70% GOOD
Sanders-Dewey et al. (2002)			CD					NA		NA		NA	NA		60% FAIR

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	Score (%) and rating
Sharkey et al. (2018)	Green	Green	CD	Green	Red	Red	Red	NA	Green	NA	Green	NA	NA	Green	60% FAIR
Small & Graydon (1992)	Green	Green	Green	Green	Red	Red	Red	NA	Green	NA	Green	NA	NA	Green	70% GOOD
Wolfe-Christensen et al. (2008)	Green	Green	CD	Green	Red	Red	Red	NA	Green	NA	Green	NA	NA	Green	60% FAIR

Note. Numbers 1 to 14 correspond to quality appraisal tool questions (see Appendix D); Green = Yes Red = No White = NA/NR/CD. A “good” study has the least risk of bias; the results are considered to be valid. “Fair” constitutes some susceptibility to bias but not sufficient to invalidate the result.

CD=cannot determine, NR= not reported NA=not applicable. In total Good=9, Fair=14, Poor=0

Appendix F

Illness uncertainty scores

Table presenting IU raw scores for each paper

Study	MUIS-C (range 23-115)	MUIS-A (original 33 item) (range 33- 165)	MUIS-A modified (28- item) (range 28- 140)	“other” *SF-MUIS (range 5-25) ** BT-MUIS
Ahn et al. (2017)		99.03(13.04)		
Bailey et al. (2009)		87.30(17.6)		
Barberis et al. (2019)		81.78 (16.8)		
Carpentier et al. (2007)	NR			
Colagreco et al. (2014)		86.45 (13.84)		
Detprapon et al. (2009)	53.9(13.05)			
Hommel et al. (2003)	47.63(12.19)			
Hoth et al. (2013)			Ambiguity 35.4(10.5) Complexity 25.6 (6.9)	
B. Kim & J. Kim (2019)		90.00(11.63)		
Kurita et al. (2013)			NR	
Lee (2006)	51.3(14.5)			
Lemaire (2004)	65.03(15.90)			

Study	MUIS-C (range 23-115)	MUIS-A (original 33 item) (range 33-165)	MUIS-A modified (28-item) (range 28-140)	“other” *SF-MUIS (range 5-25) ** BT-MUIS
Lin et al. (2013)**				Ambiguity/inconsistency (range 16-66) Score: 34.03 (10.84) Unpredictability of disease prognosis (range, 7-32) Score: 17.97
Mullins et al (2000)	45.95(12.74)			
Mullins et al (2001)	NR			
Mullins et al. (2017)	Asthma/allergies: 53.91(13.89) Other CI's: 60.34 (15.26)			
Pahlevan (2017a)*				*NR
Pahlevan et al. (2017b)*				*11.28
Sanders-Dewey et al. (2001)	70.6(10.7)			
Santacrocce & Lee (2006)	51.3(14.5)			
Sharkey et al. (2018)	61.45(8.14)			
Small & Graydon (1992)			77.23(13.89)	
Wolf-Christensen et al. (2008)	NR			

Note. NR= not reported

Appendix G

Time since diagnosis/illness duration figures

Table presenting TSD figures reported from each paper

Study	Time since diagnosis - mean (years) or related information
Ahn et al. (2017)	7.12
Bailey et al. (2009)	NR
Barberis et al. (2019)	NR
Carpentier et al. (2007)	6.8(SD=3.85).
Colagreco et al. (2014)	4.45(range 1.08–8.59)
Detprapon et al. (2009)	The authors reported: Almost half (42.5%) had been treated using a combination of surgery and radiotherapy for 1 - 12 months (mean = 5.27 months).
Hommel et al. (2003)	13.63(SD=3.65)
Hoth et al. (2013)	12.1(SD=7.5)
B. Kim & J. Kim (2019)	NR
Kurita et al. (2013)	NR but inclusion “diagnosed for a minimum of 6 months with cancer of the lung”
Lee (2006)	The mean time since diagnosis was 16 (SD = 6.4) years; range 5-31 years
Lemaire (2004)	NR
Lin et al. (2013)	NR
Mullins et al. (2000)	The authors reported: Age range of participants (18-25, 19.67, SD = 1.77) – they received a diagnosis of asthma prior to 12 years of age
Mullins et al. (2001)	13.7(SD=7.3)

Study	Time since diagnosis - mean (years) or related information
Mullins et al. (2017)	NR
Pahlevan (2017a)	Means NR
Pahlevan et al. (2017b)	Means NR
Sanders-Dewey et al. (2001)	6.5 years (SD = 3.8 years).
Santacroce & Lee (2006)	14.2(SD=6.1) *time since treatment completion, childhood cancer survivors
Sharkey et al. (2018)	NR
Small & Graydon (1992)	10.62(SD=12.02).
Wolfe-Christensen et al. (2008)	All participants received a diagnosis of asthma during childhood (age at diagnosis ranged from 1 to 12 years old) Mean age - 19.70 (SD= 1.25)

Note. NR= not reported, SD=standard deviation

Appendix H

Prisma checklist

The PRISMA checklist was retrieved from the weblink <http://prisma-statement.org/prismastatement/Checklist.aspx>

Section/topic	#	Checklist item	Reported on page #
TITLE			
Title	1	Identify the report as a systematic review, meta-analysis, or both.	p.1
ABSTRACT			
Structured summary	2	Provide a structured summary including, as applicable: background; objectives; data sources; study eligibility criteria, participants, and interventions; study appraisal and synthesis methods; results; limitations; conclusions and implications of key findings; systematic review registration number.	P's. 2-3
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known.	P's 8-9
Objectives	4	Provide an explicit statement of questions being addressed with reference to participants, interventions, comparisons, outcomes, and study design (PICOS).	p.9-10
METHODS			
Protocol and registration	5	Indicate if a review protocol exists, if and where it can be accessed (e.g., Web address), and, if available, provide registration information including registration number.	p.10
Eligibility criteria	6	Specify study characteristics (e.g., PICOS, length of follow-up) and report characteristics (e.g., years considered, language, publication status) used as criteria for eligibility, giving rationale.	P's.9-12
Information sources	7	Describe all information sources (e.g., databases with dates of coverage, contact with study authors to identify additional studies) in the search and date last searched.	p.10
Search	8	Present full electronic search strategy for at least one database, including any limits used, such that it could be repeated.	p.10(Appendix A)
Study selection	9	State the process for selecting studies (i.e., screening, eligibility, included in systematic review, and, if applicable, included in the meta-analysis).	p.11 p.15
Data collection process	10	Describe method of data extraction from reports (e.g., piloted forms, independently, in duplicate) and any processes for obtaining and confirming data from investigators.	p.13

Data items	11	List and define all variables for which data were sought (e.g., PICOS, funding sources) and any assumptions and simplifications made.	p.15
Risk of bias in individual studies	12	Describe methods used for assessing risk of bias of individual studies (including specification of whether this was done at the study or outcome level), and how this information is to be used in any data synthesis.	p.15 *quality appraisal for narrative synthesis
Summary measures	13	State the principal summary measures (e.g., risk ratio, difference in means).	P's. 27-37
Synthesis of results	14	Describe the methods of handling data and combining results of studies, if done, including measures of consistency (e.g., I ²) for each meta-analysis.	p.10 *narrative synthesis guidance
Section/topic	#	Checklist item	Reported on page #
Risk of bias across studies	15	Specify any assessment of risk of bias that may affect the cumulative evidence (e.g., publication bias, selective reporting within studies).	n/a *quality appraisal undertaken
Additional analyses	16	Describe methods of additional analyses (e.g., sensitivity or subgroup analyses, meta-regression), if done, indicating which were pre-specified.	n/a

RESULTS			
Study selection	17	Give numbers of studies screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally with a flow diagram.	p.14-15 *PRISMA
Study characteristics	18	For each study, present characteristics for which data were extracted (e.g., study size, PICOS, follow-up period) and provide the citations.	P's. 14-25
Risk of bias within studies	19	Present data on risk of bias of each study and, if available, any outcome level assessment (see item 12).	P. 26
Results of individual studies	20	For all outcomes considered (benefits or harms), present, for each study: (a) simple summary data for each intervention group (b) effect estimates and confidence intervals, ideally with a forest plot.	P's. 26-39

Synthesis of results	21	Present results of each meta-analysis done, including confidence intervals and measures of consistency.	n/a
Risk of bias across studies	22	Present results of any assessment of risk of bias across studies (see Item 15).	n/a
Additional analysis	23	Give results of additional analyses, if done (e.g., sensitivity or subgroup analyses, meta-regression [see Item 16]).	n/a
DISCUSSION			
Summary of evidence	24	Summarize the main findings including the strength of evidence for each main outcome; consider their relevance to key groups (e.g., healthcare providers, users, and policy makers).	P's. 38-44
Limitations	25	Discuss limitations at study and outcome level (e.g., risk of bias), and at review-level (e.g., incomplete retrieval of identified research, reporting bias).	P's. 41-43
Conclusions	26	Provide a general interpretation of the results in the context of other evidence, and implications for future research.	P's. 43-44
FUNDING			
Funding	27	Describe sources of funding for the systematic review and other support (e.g., supply of data); role of funders for the systematic review.	

Factors that associate with resilience in people with IBD

Section Two: Research Report

Factors that associate with resilience in people with IBD

Abstract

Objectives

In response to an under-developed research field in relation to Inflammatory Bowel Disease (IBD) and resilience, this study aimed to develop a theoretical and empirically informed understanding of resilience in people with IBD by exploring the factors that associate with it.

Design

A sequential mixed-method cross-sectional design was employed.

Method

Participants over 18 years with self-reported IBD diagnoses were recruited via social media advertisements, a volunteer research database at the University of Sheffield, and the Crohn's and Colitis UK website. Five participants were interviewed about their resilience experiences and data were thematically analysed. Eighty-five participants completed an online survey comprising measures of illness cognitions; social support, coping-efficacy, and illness-acceptance; control beliefs and intolerance of uncertainty; grit; time since diagnosis, IBD-subtype, disease activity and resilience.

Results

Grit was interpreted from the qualitative data, but correlation analyses revealed a non-significant association with resilience. Time since diagnosis and IBD-subtype also did not correlate with resilience. A hierarchical regression revealed that disease activity negatively predicted and explained 15.7% of the variance in resilience. Illness-

Factors that associate with resilience in people with IBD

acceptance, social support and coping-efficacy explained a further 54.1%. Control-beliefs and intolerance of uncertainty predicted a further 10.8% of the variance in resilience.

Conclusions

Illness cognitions and beliefs about controllability predict resilience over and above the negative influence of disease activity. Rather than perseverance towards long-term goals (grit), daily persistence or endurance might be more pertinent to resilience in those with IBD. Future research should examine causal pathways and the mediating influences of the constructs explored in this study between disease activity and resilience.

Practitioner points

- Targeting illness cognitions using cognitive behaviour therapy may foster resilience. Professional support could be particularly beneficial when disease-activity is high.
- Consideration could be taken to target intolerance of uncertainty and third-wave therapies could be considered to foster acceptance.

Limitations

- The qualitative phase comprised a homogenous sample limiting transferability of the findings.
- A cross-sectional design excludes causal explanations.
- The validity of the scales measuring control-beliefs and coping-efficacy were compromised and their findings should be interpreted with caution.

Keywords: Inflammatory Bowel Disease, illness-cognitions, control-beliefs, disease-activity, grit, mixed-methods.

Factors that associate with resilience in people with IBD

IBD is a chronic illness (CI) and umbrella term for inflammatory conditions of the digestive system. Subtypes include Ulcerative Colitis (UC) and Crohn's Disease (CD) which affect the large intestine and other parts of the digestive tract respectively (NICE, 2015). Indeterminate IBD is diagnosed where diagnostic tests are indicative of IBD, but when CD or UC are indistinguishable (Guindi & Riddell, 2004). An estimated 620,000 people in the UK (0.5-1%) have IBD (Molodecky et al., 2012) and 6.8 million globally (Global Burden Disease Collaborators [GBD], 2020). IBD is incurable and debilitating, presenting individuals with psychological challenges associated with unpredictable relapsing/remitting symptomatology (Moum et al., 1996). IBD subtypes share a similar symptom profile, including diarrhoea, faecal incontinence, fatigue, and abdominal pain (Bielefeldt et al., 2009; Larsson et al., 2008).

Research about people with IBD confirms it associates with negative psychopathological outcomes including depression and anxiety (Goodhand et al., 2012; Todorovic, 2012; Walker et al., 2008), psychological distress (Larsson et al., 2008; Nordin et al., 2002) and deleterious effects on quality of life (QOL, Bennebroek et al., 2012; Casellas et al., 2002). IBD is episodic in nature and is known to disrupt social activities, employment, relationships (Kemp et al., 2012; Restall et al., 2016), personal goals and functioning (Graff et al., 2009).

Resilience as a reported and desired state in those with IBD (Fourie et al., 2018; Luo et al., 2019) has been less researched (Luo et al., 2019; Sirois & Hirsch, 2017) and is under theorised in IBD populations. Research theorising resilience in the context of other CI's has measured the construct as absence of psychopathology and preservation of QOL (Stanton et al., 2007; Stewart & Yuen, 2011). An IBD study by Kiebles et al.

(2010) theorises resilience as retaining psychological/emotional functioning; but in this study a specific measure of resilience was not used. As a desired outcome at both an individual and clinical level, this research sought to empirically explore resilience factors in an IBD population to develop a theoretical understanding of resilience specific to IBD.

Resilience factors in chronic illness and IBD

Definitions of resilience vary substantially (Davydov et al., 2010), however, there is some consensus that it constitutes a return to pre-illness functioning (Gheshlagh et al., 2016; Carver, 1998). Carver's (2010) generalised model of resilience in contexts of trauma and adversity distinguishes between deterioration (loss, depression), 'bouncing back' (a return to pre-illness functioning) and thriving (benefit and growth). A return to pre-illness functioning is influenced by illness cognitions (beliefs about illness and coping resources) that can mediate between the condition and individual well-being (Evers et al., 2001; Heijmans & De Ridder, 1998; Scharloo et al., 1998). One such illness cognition is self-efficacy and has been posited as one of four functions of resilience (Rutter, 1987). Pioneer of self-efficacy, Albert Bandura (1977; 1997) posited that it is one's perception of their capability to execute the necessary behaviours to manage situations.

Other key psychological factors are identified as theoretically and empirically linked with resilience in the literature. Carver (1998) argued that those who thrive do so because they develop efficacious strategies for coping. Coping-efficacy which associates with thriving in those with arthritis and IBD (Sirois & Hirsch, 2013; Sirois & Hirsch, 2017) pertains to appraisals of how successfully one copes (Gignac et al., 2000). Furthermore, illness-acceptance; the recognition and willingness to adapt and tolerate

the unpredictable, uncontrollable nature of the disease (Evers et al., 2001) is an illness cognition found to positively correlate with QOL (Kurpas et al., 2013; Lewko et al., 2012); adjustment and resilience in IBD populations (Kiebles et al., 2010; Sirois & Hirsch, 2017).

Irrespective of IBD's association with a lower sense of control (Graff et al., 2009), research pertaining to control-beliefs, IBD and outcomes is limited. The locus of control (LOC) construct (Rotter, 1975) refers to the expectancy belief about control over an outcome (Green & Murdock, 2013). The belief in individual responsibility for events that occur is termed internal LOC (ILOC, Rotter, 1954). The modified social learning theory (Wallston, 1992) purports that health behaviour is contingent on one's perceptions of control over health, synonymous to the construct of coping-efficacy. Bandura's (1977) social cognitive theory additionally argues that one intrinsically seeks to exert control over their coping behaviour. ILOC has been linked to better psychological outcomes in those with health conditions (Lenze et al., 2008; Panagiotou et al., 2014) and decreased anxiety and depression (Fan et al., 2010; Park & Gaffey, 2007). Qualitative findings report control issues and powerlessness as central tenets of the IBD experience (Devlen et al., 2014; Dibley et al., 2017; Dudley-Brown, 1996; Pihl-Lesnovska et al., 2009).

How in control one feels also influences uncertainty (McCormick, 2002).

Uncertainty is pertinent for those with IBD given its unknown aetiology, fluctuating and unpredictable symptomatology, thus the ability to tolerate uncertainty is an important factor. Intolerance of uncertainty (IOU) refers to the propensity to react negatively to uncertain situations (Buhr & Dugas, 2009) and has been conceptually and empirically linked to generalised anxiety disorder (GAD, Dugas et al., 1997) and lower

Factors that associate with resilience in people with IBD

psychological adjustment in those with lung cancer (Kurita et al., 2013). Higher perceived certainty has been linked to better QOL in one IBD study (Niv et al., 2017), however, the relationship between IOU and resilience has never been examined in an IBD population and thus is not understood.

Social support has been found to lead to positive outcomes in those with IBD. Received support is distinct from perceived support (Kamp et al., 2019), which is the realisation of being accepted and cared for (Strom & Egede, 2012); another illness cognition. The protective function of social support has been well documented (Ozbay et al., 2008). Theoretical models of social support acknowledge the importance of both emotional (receiving love and empathy) and practical components (Charney, 2004) through exerting positive effects on multiple neurobiological pathways and fostering effective coping (Ozbay et al., 2008). Social support also positively correlates with illness acceptance (Janowski et al., 2012) and self-efficacy (Wang et al., 2015).

In IBD populations, research demonstrates a link between increased social support, improved QOL (Janke et al., 2004; Katz et al., 2016); and adjustment in patients with IBD cross-sectionally (Gick & Sirois, 2010) and longitudinally (Oliveira et al., 2007). However, the relationship with outcomes in IBD is complex as tensions such as unwanted confrontation and undesirable reactions can occur (Palent & Himmel, 2019). This suggests more research is needed to understand the relationship between social support and resilience in those with IBD.

Setbacks are inherent within IBD population given its relapsing/remitting symptom profile. Hence, grit may be important to resilience in those with IBD, conceptualised as a dimension of resilience (Stoffel & Cain., 2018), which comprises perseverance towards long term goals with sustained commitment despite setback and

adversity (Duckworth et al, 2007). Those individuals possessing grit approach threatening situations with assurance they can exercise control over them (Duckworth et al., 2007).

Disease-specific factors

Disease-specific factors also associate with resilience but research outcomes present mixed findings. The relationship between IBD subtypes and psychological functioning are variable (Graff et al., 2006; Rubin et al., 2004). Little is known about what the impact of time following IBD diagnosis is on resilience, however the time-period around diagnosis has been associated with higher rates of anxiety and depression (Munkholm et al., 1995). Furthermore, distress levels fluctuate in parallel with disease activity (Casellas et al., 2005; Porcelli et al., 1996). Greater illness severity has been associated with poorer psychological adjustment (Voth & Sirois, 2009) which may link to its influence on sense of mastery, the sense that one has control of one's life (Graff et al., 2009). Those in remission were found to have greater perceived control and exhibited less depression (Gandhi et al., 2014). In a review of disease activity in CD, ten studies revealed an inverse relationship between health related QOL and disease activity (Vander-Have, 2014). Research suggests greater disease activity negatively affects resilience in IBD populations, however more research is needed.

Despite constituting a relatively small field, the relationship between IBD and resilience is supported by the literature. One IBD study using a small sample found that illness-acceptance and coping associated with emotional and psychological functioning (Kiebles et al., 2010). A recent study (Sirois & Hirsch, 2017) based on Carver's (1998) resilience framework measured loss, resilience, or thriving. Higher levels of coping-efficacy, illness-acceptance and social-support were associated with resilience and

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lower levels of depression across domains of life satisfaction, personal growth, and relationship quality. However, this study did not use a specific measure of resilience or incorporate examination of disease activity.

Study aims

This study aimed to develop an understanding of resilience in an IBD population based on the key psychological constructs discussed, informed by Carver's (1998) conceptual resilience framework. This research used a mixed-methods design to elicit qualitative and quantitative evidence to underpin a robust theoretical model. This study aimed to address gaps in current knowledge by examining additional variables in relation to resilience, including IBD subtype, time since diagnosis (TSD), IOU, control-beliefs and understanding of the role of disease activity. The experience of thriving (Carver, 1998) was not incorporated because outcomes of 'bouncing back' better reflect the objectives of recovery focused clinical services aiming to foster resilience (pre-illness functioning) in the context of limited fiscal resources.

Clinical implications

The new empirically informed, theoretical understanding developed in this study has important clinical implications. NICE (2010, guideline 91) recommend cognitive behavioural therapy (CBT), for the treatment of depression in those with CI's. However, there remains a lack of understanding of how psychological processes strengthen resilience (Timmer et al., 2011) as a desired outcome, particularly in IBD populations. These findings will enable the tailoring of intervention strategies so important factors may be nurtured in those with IBD who are in need (Luo et al., 2019).

Methods

Design

A sequential mixed methods design was employed (Creswell et al., 2003), whereby the personal, lived accounts of resilience experience, elicited via interviews (phase A), confirmed, and extended findings in the existing literature and informed additional constructs to be examined statistically in an online survey (phase B). Interviewed participants also commented on the usability of the survey (Appendix A). The survey contained self-report measures enabling statistical examination of the associations between the variables and resilience. A mixed-methods approach draws on the strengths of each data type (Halcomb & Hickman, 2015), by foregrounding both participant's voice and statistical examination of associations between constructs.

Epistemological position

The researcher and study approach were oriented towards critical realism as a philosophical and methodological framework (Fletcher, 2017). Aligning with the mixed-methods design, critical realism combines components of positivist and constructionist paradigms but deviates from both in its position that ontology (the nature of reality) is not reducible to epistemology (knowledge of reality) (Fletcher, 2017). Empirically, events can be measured and observed but only understood through the filter of human experience, thus, the researcher's interpretation was influenced by her own unique experiences, beliefs, and values. Therefore, subjective interpretations can differ according to individual data-analysts (Madill et al., 2000).

The researcher arrived at this positioning from her previous experiences of training in the Clinical Psychology field and learning to deliver Cognitive Analytic

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Therapy (CAT, Ryle, 1999) which is underpinned by attachment and cognitive, behavioural theories (Beck, 1976; Bowlby, 1969; Freud, 1900; Kernberg, 1975; Watson, 1913; Winnicott, 1960). CAT focuses on how emotional and relational patterns underlie one's current experiences or difficulties. The researcher was concurrently immersed in other psychological theories relevant to resilience and related constructs which underpinned the analysis (Bandura, 1977; Carver, 1998; Duckworth et al., 2009; Rotter, 1987). Indeed, the researcher understood theoretical and evidence informed knowledge of IBD but had no lived experience of it. Combined, these experiences exerted an influence on the data being interpreted through a relational lens and with preconceived ideals related to positive psychological processes and outcomes.

Ethics

The study was designed in accordance with the British Psychological Society Ethical Guidelines (2018) and the British Psychological Association Code of Human Research Ethics (2014). Ethical approval for the study was obtained from the Sheffield University Ethics Committee who provided research governance sponsorship (Appendix B).

Phase A: Qualitative enquiry

Recruitment

Purposive sampling methods were employed intending to recruit enough participants that met specified inclusion criteria, to attend a focus-group. A group size of 4-8 has been argued to be suitable for exploring a range of participant experiences (Krueger & Casey, 2015). Advertisements were disseminated electronically via social media platforms, Twitter and Facebook and a database containing email addresses from volunteer research participants held by the University of Sheffield. Only two

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participants attended a joint interview due to participant constraints and logistical challenges thus a design amendment was subsequently developed to elicit participant views via individual interviews; a methodology that enabled study aims to be achieved. The amendment was ethically approved. Three participants who had expressed interest in participation in the focus group but who could not attend were invited to participate in individual interviews. One was face-to-face and two via telephone interviews.

Participants

To be eligible to participate, participants needed to be over 18 years with a confirmed diagnosis of IBD, including IBD indeterminate.

Procedure

Participants who agreed to be interviewed were electronically sent an information sheet (Appendix C) and consent form (Appendix D). Signed consent forms were collated prior to the interviews. Three participants were interviewed on University of Sheffield premises, conducted by the main researcher. A semi-structured interview schedule was developed by the researcher, guided by thematic analysis (TA) and qualitative IBD literature (Braun & Clarke, 2013; Luo et al., 2019). Questions were designed to be flexible, with suggested prompts including questions like “are there any psychological factors that you feel have affected your resilience?” (see Appendix E for interview schedule).

Those who opted for telephone interviews provided audio-recorded verbal consent and consent forms were electronically signed by participants. Interviews lasted 37-64 minutes (average 54.7 minutes) and were conducted in August/September 2019. Participants were offered a £10 voucher for their participation. Another clinical psychology doctoral trainee observed the focus group and took notes to aid accurate

transcription and offer additional insights. All interviews were audio-recorded and anonymously transcribed (using pseudonyms) verbatim by the researcher.

Qualitative analytic method

TA was employed and data were analysed in an inductive way, enabling themes to emerge from the data (Braun & Clark, 2006; Patton, 1990). TA is a flexible approach to data-analysis and can be conducted within both realist/essentialist and constructionist paradigms, conducive with the critical realist epistemological position (Braun & Clarke, 2006). Indeed, the researcher supports the realist idea that some forms of “truth” exist and interview data offered opportunity for participants to share part of their perceptual reality. However, it was acknowledged that one’s understanding of others’ realities cannot be fully achieved (Willig, 2008). Braun and Clarke’s (2006) 6-step process was undertaken and revisited in a recursive process (see Appendix F), leading to the culmination of themes.

Reflexivity

The researcher recognised herself as an active part of the research process. To bring to conscious awareness the influence of the researcher’s assumptions on the interpretation of data (Brocki & Wearden, 2006), the transcripts were repeatedly revisited to ensure the codes and themes were rooted in the data. Input was also derived from another researcher who had observed and given her interpretations of the focus group data. Furthermore, research supervisors (who are also Clinical Psychologists) were involved in theme generation and the researcher’s reflections were discussed to facilitate consideration of her influence on the interpretation.

Results

An analysis and interpretation of data is presented. All participants identified as white British females and considered themselves in remission. Their ages ranged from 22-49 years (mean 34) and disease duration ranged from 2-28 years (mean 10.8). They reported diagnoses of CD (n=2) and UC (n=3). They discussed times that their condition caused distress and felt difficult to endure. Remission was frequently associated with reduced distress and a sense of increased resilience. The perception of themselves as resilient was evident, however it was a dynamic state.

Two superordinate themes emerged, each comprising three subordinate themes (Table 1). Themes are discussed and accompanied by illustrative quotes (see Appendix G for further supporting quotes). Resilience was characterised by ambivalent states; defined as simultaneously holding conflicting feelings or reactions (Schneider & Swartz, 2017). Themes comprised intrapersonal (within the mind) and interpersonal processes (occurring relationally). How participants managed the inherent tensions either positively or negatively affected their sense of resilience.

Table 1

Summary of superordinate and subordinate themes

Superordinate themes	Subordinate themes
1. Intrapersonal ambivalence: Grit	1.1 “I quite like the control” 1.2 “An invisible illness” 1.3 Carry on regardless”.
2. Interpersonal ambivalence: Social support	2.1 “The support really helps” 2.2 “Nobody really understood” 2.3 “brave face”

Themes

1. Superordinate theme: Intrapersonal ambivalence, grit

‘Grit’ permeated the discourse; namely the need for diligent engagement with long-term goals (social and occupational) to maintain resilience. Participants reclaimed control by coping independently which boosted resilience, but a conflict occurred when this led to the legitimacy of the condition and its severity being questioned. Endurance, a regulatory strength, persisting diligently when presented with difficulty (Hamby et al., 2013) was adopted. This created mixed emotions and either positively or negatively impacted resilience.

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1.1 “I quite like the control” For all participants, IBD challenges were buffered through reclaiming perceived control. This was achieved by managing the condition autonomously in variable ways. This prevented a potential deterioration in their mental state which boosted resilience. One way of regaining control was through positive risk-taking and making disparate decisions to medical professionals to stay on track with valued goals. The ability to manage independently and work towards goals (social and occupational) generated feelings of satisfaction and pride:

“If I had no way of helping me control it myself I would feel a bit hopeless, I quite like the control.....I think without being able to manage it for myself I would struggle to be okay about it.” (Harriet)

“the satisfaction that I’m managing it myself and actually its working really well.” (Annie)

1.2 “An invisible illness” The internal conflict around autonomous management was that the legitimacy of the condition and its severity came under question. One participant discussed relief at the point of diagnosis as it validated the legitimacy of her symptoms. The perception that others might think the symptoms to be spurious or exaggerated threatened identity, causing difficult emotions, negatively impacting resilience.

Grit encompassed personal goals related to maintaining integrity, and a premonitory identity as a fully functioning, able-bodied, genuine individual. This conflict was variably managed. Most of the participants subscribed to the non-disclosure view due to anxieties about looking weak or burdening others. While this temporarily boosted resilience, the lack of openness about the impact of the condition meant that participants endured their symptoms alone, lessening resilience. Conversely, one participant opted

for full public disclosure, motivated by anger. She self-preserved by attempting to prove or demonstrate teleological evidence of the severity of the experience.

“The frustration.....that some people are questioning whether it’s as bad as you’re making out.” (Annie)

“I was pissed off at people, when you’ve got an invisible illness, you tend to get people thinking she’s making it up or exaggerating.....that was horrendous and I genuinely think that was the worst part” (Sarah)

“No one’s asking me if I’m alright but I’m just thinking none of these people around me understand what I’m going through right now and that can be hard.” (Claire)

1.3 “Carry on regardless”. Endurance was illuminated by phrases such as *“not let it beat me”* and *“bouncing back you’ve just got to”*. These participants strived towards an imagined life trajectory prior to their IBD diagnosis, demonstrating commitment to activities valued by them and wider society. This was underpinned by a fear of loss in some cases. Differing internal coping strategies were employed to achieve endurance, including *“ignorance”*, and following a period of loss and grief; *“acceptance”*.

“I made myself do everything and I still went to work everyday.....mentally really didn’t wanna be there but, I force myself.... I’m ignorant because I’m like no I’m fine, just gotta carry on” (Claire)

“If I’m having a flare up it’s not going to stop me from going out there, doing my socialising, having plans that I’ve already made.....I just keep pushing through.” (Harriet)

“When you reach the acceptance you have to look at how you live your life from now on.....keep looking forward so, you might end up in a better place than you thought you were headed anyways.” (Sarah)

2. Superordinate theme: Interpersonal ambivalence, social support

Actual or perceived social support impacted resilience. Participants desired support that was empathic and boundaried, which enhanced resilience. However, a conflict arose from the fear or reality of receiving negative reactions, being misunderstood, judged, or stigmatised. To avoid this eventuality, participants put on a “*brave face*” which simultaneously benefited and disadvantaged resilience. This was interpreted as stoicism; “silent endurance, lack of emotion.....a ‘stiff upper lip’ (Moore et al., 2012, p.159-160).

All participants described parents, family, and close friends as their main sources of support. In line with autonomous management of the condition, there were mixed feelings towards medical professionals who were viewed as either helpful or a hindrance, contingent on their availability and expertise.

2.1 “The support really helps” All participants desired support in the form of an inherent understanding, thus others who had a realistic understanding and ability to provide empathic care were valued. This enabled frank conversations about their experiences and reciprocal support. This enhanced resilience by helping emotional management of the condition. However, it was important that the care received had limits so that it was not a threat to their identity. When participants felt “*pandered too*” (Claire) or wrapped “*in cotton wool*” (Harriet) this undermined their perceived competence. Empathy was desired but an encouraging approach that honoured their self-efficacy was imperative.

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“I think the support really helps me being able to manage...the negative emotional effects the disease can have and that’s the importance of support.” (Harriet)

“ I feel like I handle it quite well, I don’t want someone to pander to me..... they’re supportive, they’re not feeling sorry for me because that’s not what you want.” (Claire)

2.2 “Nobody really understood”. Their need for closeness was accompanied by a fear of being misunderstood or dismissed. Others lacked understanding or knowledge or there was a felt stigma about being ill and a taboo around IBD. The impact of enduring an experience misunderstood by others had profound effects on the ways in which participants related to others. The perceived or real experience of others misunderstanding created difficult emotions which led to a reduced sense of resilience. Anger that one participant felt at experiencing severe pain and fatigue that was not acknowledged by those around her, led her to develop an online support forum. However, participants also managed this by dismissing others in varying ways. This included choosing to not “*let them in*”; to initiate intimacy with new people or share experiences. Creating this ‘safe’ distance temporarily supported resilience, however in the long-term this compromised intimacy and restricted a social network of supportive individuals, hindering resilience. For others, an intolerance of others’ lack of understanding was manifest or a minimisation of others’ experiences of suffering. A self-perception as being comparatively ‘strong’ temporarily boosted resilience as it defended against the perception of themselves as vulnerable. However, this perpetuated a sense of distance from others which depleted their sense of resilience.

“Nobody was listening, or nobody really understood what I was going through and that really pissed me off.” (Sarah)

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“Brought out a less tolerant side of me.....others might say, you know, my goldfish died (laughs) and that’s where I have less empathy with that situation.” (Sally)

“Some people wallow in being a victim of something.” (Annie)

2.3 “Brave face”. The interpersonal conflict was resolved through the concealment of their struggles, stoicism. This enabled resilience as it facilitated self-preservation, however, it brought about conflicted feelings. An underlying anxiety and guilt about letting others down led to avoidance of self-care such as taking time off sick from work (linking to endurance). Participants engaged with stoicism with resentment and frustration that perpetuated their aloneness. This reinforced the lack of acknowledgement from others, which negatively impacted resilience in the long term. There was recognition that stoicism was unsustainable and when expected by others it brought about anger, lessening a sense of resilience.

“I’d be fine on the outside to most people then internally it’d be really shit....”
(Claire)

“Tend to just brush it off and go with I’m fine.” (Sarah)

“I had a fury and said I’ve got a life-long life changing chronic condition you might be allowed to feel a bit sorry for yourself.” (Annie)

Data analysis summary

The data revealed key factors that have been previously associated with resilience in those with CI’s and IBD. Central tenets of resilience pertained to coping independently, perceived control and social support. There was also reference to uncertainty, acceptance, and the influences of disease activity; higher disease activity

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was associated with reduced resilience. TSD evoked mixed feelings. The construct of 'grit' also emerged.

Phase B: Quantitative enquiry

Recruitment and Procedure

The recruitment methods were the same as phase A. This phase was additionally advertised through Crohn's and Colitis UK's website (see Appendix H). Participants from Phase A or expressed interest but could not participate for logistical reasons were emailed a weblink to the Qualtrics online survey. The information sheet and consent were contained within the weblink (Appendix I). Data were collected from 30th January until 15th March 2020. Data collated after this was deemed to be influenced by Covid-19 circumstances and was thus excluded from the analysis. Participants were able to enter a £25 vouchers prize draw.

Quantitative measures

Screening and demographics information.

The online survey contained screening questions pertaining to age and confirmation of IBD diagnosis. The survey contained demographic and IBD-related questions (including age of diagnosis, subtype, relapse/remission status) and the following self-report measures (see Appendix J). For all measures higher scores indicate greater levels of the constructs. A full-scale, total score was analysed for all measures except the Illness Cognition Questionnaire (ICQ; Evers et al., 2001). The internal consistency scores (α) for each measure in the current study ranged from acceptable to excellent (.78 - .93, see Table 4).

Connor-Davidson Resilience Scale (CD-RISC, Connor & Davidson, 2003).

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The 25-item CD-RISC was designed to assess the personal characteristics that embody resilience, including self-efficacy, patience, optimism, faith, personal competence, trust/tolerance/strengthening effects of stress, acceptance of change and secure relationships and evaluates items on a 5-point Likert scale from 0 “not true at all” to 4 “true nearly all of the time” (scoring 0-100). The internal consistency was found to be good ($\alpha = 0.89$) in a sample of individuals with generalised anxiety disorder and post-traumatic stress disorder (Connor & Davidson, 2003).

Short Health Scale (SHS, Hjortswang et al., 2006, in Mcdermott et al., 2013).

The 4-item SHS is a proxy measure of disease activity capturing health-related QOL. The SHS comprises 4x100mm analogue scales assessing symptom burden, activities of daily life, disease related worry and general well-being. The scale was previously validated in a Swedish and Norwegian population (Hjortswang et al., 2006) and later in an English IBD population, with test-retest reliabilities revealing correlations from 0.70-0.89 (Mcdermott et al., 2013).

Illness Cognitions Questionnaire

The ICQ comprises the subscales, helplessness, acceptance, and perceived benefits. The 6-item acceptance subscale was used in this study which assesses acceptance of one’s CI and uses a 1-4 Likert scale from 1 “not at all” to 4 “completely” (scoring 6-24). The acceptance subscale has demonstrated good internal consistency ($\alpha = 0.91$, Evers et al., 2001) and $\alpha = .92, .89$ at two time points (Sirois & Hirsh, 2017).

Coping Efficacy Scale (CES, Gignac et al., 2000).

The 3-item CES assesses the extent that individuals feel that they are coping effectively with symptoms, emotional aspects and daily challenges of their

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condition measured on a Likert scale from 1 “strongly disagree” to 5 “strongly agree”. As in previous research (Sirois & Hirsch., 2017) ‘IBD’ replaced the term ‘illness’. The scale was created in error for this study and thus was scored on a 7-point Likert scale with the additional scale items “somewhat-disagree” and “strongly-agree” (scoring 1-21). The CES has demonstrated good internal consistency in arthritis ($\alpha = .80$; Sirois & Hirsch, 2013), and IBD sample ($\alpha = .90$) (Sirois & Hirsch, 2017).

Duke-UNC Functional Social Support Questionnaire (FSSQ, Broadhead et al., 1988).

The 8-item FSSQ measured perceived social support, covering receipt of emotional and practical support, on a 5-point Likert scale ranging from 1 “much less than I would like” to 5 “as much as I would like”. The measure is scored by averaging all items, resulting in scores ranging from 1-5. It has demonstrated good internal consistency at two time points (alphas= .91 and .93) (Sirois & Hirsch, 2017).

Control Beliefs Inventory (CBI, Sirois, 2002).

The 26-item CBI measures four specific health related control beliefs: General control, chance control, symptom control and mastery/health self-efficacy. The CBI was designed for use with CI populations and each subscale has demonstrated acceptable or good internal consistency ($\alpha = 0.70-0.91$, Sirois, 2003). The original scale assessed items on a 6-point Likert scale, from 1 “strongly disagree” to 6, “strongly agree”. The scale was created in error for this study and thus was scored on a 1-7 Likert scale with “neither agree nor disagree” (scoring 7-182).

The Intolerance of Uncertainty Scale-Short Form (IU-SF, Carleton et al., 2007).

The 12-item IU-SF assesses prospective anxiety and inhibitory anxiety through assessing reactions to uncertainty, ambiguous situations, and the future. It demonstrated

excellent internal consistency ($\alpha = .91$) in a sample of American undergraduate students (Carleton et al., 2010), large community sample ($\alpha = .92$) (Carleton et al., 2010, cited in Hale et al., 2016) and demonstrated good reliability and validity in a general population Chinese sample ($\alpha = .0.86$).

The Short Grit Scale (SGS, Duckworth & Quinn, 2009).

The 8-item SGS measures trait-level perseverance and passion for long-term goals. It includes elements of diligence, enduring, being hard-working and not discouraged by setbacks, assessed on a 5-point Likert scale, 1 “not like me at all” to 5, “very much like me”. The measure is scored by averaging all items, resulting in a grit score ranging from 1 to 5. The SGS has demonstrated good internal reliability, test-retest reliability, and criterion reliability in studies of adolescents and young adults (Duckworth & Quinn, 2009; Sharkey et al., 2017). In Traino et al.’s. (2019) study on college students with CI’s the reliability was good ($\alpha = 0.81$).

Sample characteristics

Eighty-five participants completed the survey. Most of the participants were female, in remission and UK based (see Table 3). A priori power analysis determined the sample size needed to prevent type II errors when conducting regressions analyses. Applying a cautious assumption that all nine predictor variables correlated with resilience, a sample size of 135 was required to achieve 0.80 power at a p-value of 0.05 (Field, 2009). A post-hoc analysis based on the six variables that significantly correlated with resilience and entered into the hierarchical regression analysis was conducted. The rule of thumb that between 10-15 participants per predictor were needed to achieve 0.80 power at the 0.05 level was used (Field, 2009), hence, 60-90 participants were required.

Results

Data analytic plan

Statistical Package for Social Sciences (SPSS) version 23 was used to analyse the data. Correlation analyses enabled examination of the associations between the continuous variables and the outcome. Categorical variables were examined via comparison of mean resilience scores and independent t-tests. A hierarchical regression analysis examined the amount of variance in resilience that could be explained by illness-acceptance, coping-efficacy, social support, control-beliefs, and intolerance of uncertainty (Field, 2009). Disease activity was entered into block 1 as a covariate; at block 2 illness-acceptance, coping-efficacy and social support were grouped together as coping resources. At block 3, control-beliefs and intolerance of uncertainty were grouped together as beliefs related to controllability.

The data were examined to check for assumptions of multivariate analyses which involved examination of missing data, outliers, and parametric assumptions of normality (Tabachnik, & Fidell, 2014). Histograms, Q-Q plots, skewness, and kurtosis values were assessed and checked against values demonstrating normal distribution (see Appendix K, Stevens, 2002). The Shapiro-Wilks test (Shapiro & Wilk, 1965) was used because it has more power to detect differences than alternate tests (Field, 2009). Pearson correlation and Spearman's rho coefficients were interpreted for normally distributed and non-normally distributed data, respectively. No outliers were identified from visual inspection of scatterplots. Finally, the absence of multicollinearity was assumed because the correlation coefficients for all independent variables were not too highly correlated ($r < .90$) (Field, 2013).

Missing data

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There was a small amount of missing data values (n=15). A statistical test indicated that data were missing completely at random (Little, 1988; $p = 1.00$). Thus, it was appropriate to use case mean substitution (Hanna, & Dempster, 2012); reported in previous research (Downey & King, 1998; Eekhout et al., 2012; FoxWasylyshyn & El-Masri, 2005; Raymond, 1986; Roth et al., 1999).

Descriptive data

Table 2 summarises participant and disease-related characteristics. The majority were female, UK based and considered themselves to be in remission.

Table 2

Participant and disease characteristics

Participant characteristics	
Gender % (n) (female)	72.94 (62)
Age (mean, <i>SD</i>)	39.49 (11.45)
Country:	
United Kingdom % (n)	87.1(74)
Europe % (n)	3.5(3)
Canada % (n)	2.4(2)
USA % (n)	4.7(4)
Australia % (n)	1.2(1)
Other % (n)	1.2(1)
Disease characteristics	
Age of diagnosis (mean, <i>SD</i>)	26.53 (11.1)
Illness duration (mean, <i>SD</i>)	13.04 (10.99)
Diagnostic subtype:	
Crohn's disease % (n)	49.4(42)
Ulcerative Colitis % (n)	44.70(38)
Other: % (n)	5.9(5)
Disease activity status:	
Relapse (n, %)	35.29(30)
Remission (n, %)	64.71(55)

Note. *SD* = standard deviation

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Descriptive scoring data from all measures are presented in Table 3. None of the scales had categorical cut-offs, but for all, greater scores indicated a greater presence of each construct.

Table 3

Cronbach's alphas, and descriptive statistics for all measures

Variables	A	Possible range	Minimum in sample	Maximum in sample	Mean	SD
TSD (years)		-----	0	45	12.96	11.02
Disease activity	.84	0-400	38	400	201.4	90.87
Social support	.88	1-5	1.38	5.00	3.87	0.88
Acceptance	.89	6-24	6	24	16.41	4.18
Grit	.78	1-5	1.25	3	2.24	0.37
Control beliefs	.87	26-182	65	153	116.60	18.71
Coping-efficacy	.88	3-21	3	21	15.32	4.23
Intolerance of uncertainty	.89	12-60	15	60	32.47	9.3
Resilience	.93	0-100	29	96	66.07	15.30

Note. TSD=Time since diagnosis.

Descriptive analyses

Independent t-tests assessed differences between categorical variables and resilience. There was no significant effect for gender, $t(83) = -0.11$, $p = 0.917$, despite females ($M = 66.2$, $SD = 15.2$) scoring greater than males ($M = 65.8$, $SD = 15.9$). There was no significant effect for disease activity status (relapse/remission), $t(83) = -1.8$, $p = 0.917$, despite those reporting to be in remission scoring greater ($M = 68.3$, $SD = 14.6$) than those in relapse ($M = 62.1$, $SD = 15.9$). There was no significant effect for disease subtype, $t(78) = 0.84$, $p = 0.622$, despite those with CD reporting greater resilience scores ($M = 67.6$, $SD = 14.9$) than those with UC ($M = 64.7$, $SD = 16.3$). The “other” subtype was omitted from this analysis due to the small number in this subgroup ($N = 4$). Finally, correlational analysis revealed that age and resilience were not significantly correlated, $r(-0.54)$, $p = 0.63$.

Correlational analyses

All variables except grit and TSD were significantly correlated with resilience ($p < 0.001$). Therefore, grit and TSD were not entered into the hierarchical regression analysis. As expected, negative correlations were found between disease activity and IOU and resilience (see Table 4).

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Table 4

Bivariate analyses of the relationship between independent variables and resilience.

	TSD	Disease activity	Acceptance	Coping- efficacy	Social support	Control beliefs	IOU	Grit	Resilience
TSD	1	-.123	.323**	.188	.181	-.021	.030	.101	.197
Disease activity		1	-.518**	-.558**	-.285**	-.390**	.429**	.123	-.391**
Acceptance			1	.736**	.400**	.492**	-.330**	-.234*	.705**
Coping- efficacy				1	.408**	.348**	-.400**	-.082	.625**
Social support					1	.028	-.204	.111	.456**
Control beliefs						1	-.221*	-.270*	.494**
IOU							1	.028	-.474**
Grit								1	-.176
Resilience									1

Note. TSD=Time since diagnosis. Greater scores on all scales equate to higher amount of that construct. Pearson Product-moment correlations coefficients

are presented for disease activity, acceptance, control-beliefs, IOU, grit and resilience. Spearman's Rho correlation coefficients are presented for TSD, social-support and coping-*efficacy*. * $p < 0.05$ ** $p < 0.01$.

Regression analyses

A hierarchical regression analysis was conducted to analyse the data. The model explained 67.1% of the variance and disease activity, social-support, coping-efficacy, illness-acceptance, IOU and control-beliefs were significant predictors, $R^2 = .671$, $R^2_{\text{adjusted}} = .646$, $F(2,78)=12.874$, $p<0.001$. Table 5 reports the regression analysis. In step 1, disease activity explained 15.7% of the variance in resilience, $R^2=.157$, $R^2_{\text{Adjusted}} = .147$, $F(1, 83) = 15.434$, $p = < .001$. The addition of illness-acceptance, coping-efficacy and social support explained a further 40.6% of the variance in resilience, $\Delta R^2 = .563$, $R^2_{\text{Adjusted}} = .541$, $F(3, 80) = 24.783$, $p < .001$, with only social support and illness acceptance making a significant contribution to the model. The addition of control-beliefs and IOU at step 3 explained a further 10.8% of the variance in resilience, $\Delta R^2 = 0.671$, $R^2_{\text{Adjusted}} = .646$, $F(2, 78) = 12.874$, $p < .001$, making a significant contribution to the model.

Table 5

Summary of regression analyses predicting resilience

Block	Variables	Unstandardised Coefficients		Standardised Coefficients	95.0% Confidence interval for β	
		B	Std. Error	B	Lower bound	Upper bound
1	Disease activity	-.067	.017	-.396***	-.100	-.033
2	Disease activity	.008	.015	.048	-.022	.039
	Social support	3.099	1.429	.177*	.255	5.944
	Coping efficacy	.906	.419	.251*	.073	1.739
	Acceptance	1.764	.413	.482***	.941	2.587
3	Disease activity	.036	.15	.215*	.007	.065
	Social support	4.147	1.29	.237**	1.579	6.716
	Coping efficacy	.568	.376	.157	-.179	1.316
	Acceptance	1.478	.378	.404***	.725	2.232
	Control-beliefs	.218	.063	.267**	.092	.344
	IOU	-.458	.123	.278***	-.704	-.213

Note. N=85. Note. N = 85. Block 1 $\Delta R^2 = .147^{***}$, Block 2 $\Delta R^2 = .541^{***}$, Block 3 $\Delta R^2 = .646^{***}$

* $p < .05$, ** $p < .01$, *** $p < .001$

Discussion

This exploratory study, informed by Carver's (1998) conceptual resilience framework, developed a nuanced understanding of resilience in IBD populations, by extending existing research exploring social support, coping-efficacy and illness-acceptance. Grit was interpreted as a feature of resilience from the qualitative data. The study then conducted the first statistical examination of grit, IOU, control-beliefs, and resilience in an IBD population, with consideration of disease activity.

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Resilience was not differentiated according to gender, age, or disease subtype. The regression lends support to existing studies finding that social-support, coping-efficacy and illness-acceptance associate with resilience in IBD populations (Sirois & Hirsch, 2017). Sirois and Hirsch (2017) found that coping-efficacy distinguished thriving from resilience cross-sectionally. In the current study coping-efficacy was less predictive of resilience than other factors, however the measure's validity was affected and thus comparison with other studies is compromised.

These findings are in line with the supposition that an individual's perceived capability to perform an action (coping-efficacy) influences emotional well-being, linked to agency to exert control over events (Bandura, 1977, 1997). The modified social learning theory (Wallston, 1992) argues that coping behaviour is contingent on one's perceptions of control over health outcomes. These findings indicate resilience is one such outcome. Confirming the association between control-beliefs and positive outcomes in other CI populations (Lenze et al., 2008; Panagiotou et al., 2014), control-beliefs predicted resilience in those with IBD.

Uncertainty is linked to how in control one feels (McCormick, 2002). IOU was negatively correlated with resilience; an anticipated finding because it is predictive of psychopathology in CI and emotional disorder populations (Gentes & Ruscio, 2011; Kurita et al., 2013; Mcevoy & Maloney, 2012). Combined, IOU and control-beliefs contributed 10.7 % of the variance in resilience over and above other factors. In this study, IOU also held negative correlations with control-beliefs, coping-efficacy and illness-acceptance. Hence these positive appraisals (which predict resilience) may

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indirectly influence the association between IOU and resilience. However, future research could investigate how these cognitions mediate the IOU/resilience association.

Grit, the perseverance towards long-term goals (Duckworth et al., 2007) was found not to significantly correlate with resilience. This was unanticipated given previous findings on grit's positive correlation with HRQOL, psychological wellbeing and life satisfaction (Duckworth et al., 2007; Sharkey et al., 2017; Singh & Jha, 2008; Vainio & Daukantaite, 2016). It might be that what participants voiced in this study was a daily form of endurance, persisting diligently when presented with difficulty (Hamby et al., 2013), supported by Carver (2010) who argued the struggle to prevail is a likely associate of resilience. This study suggests that in those with IBD, if one is unable to envisage long-term goals due to limitations imposed by unpredictable disease activity, the character trait of persistence; the tendency to continue striving on a daily basis, may be more apt and has been found to correlate with resilience in a non-clinical sample (Kim et al., 2013). Research examining endurance and resilience in relation to IBD is warranted. Furthermore, those with IBD worry about their education or occupation being affected (Luo et al., 2019). Hence the negative correlation between grit and resilience generated in this study suggests that grit triggers the threat system (LeDoux, 1998) within a sociocultural context whereby value is placed on productivity and occupational success. Acceptance of illness-related limitations may therefore be more pertinent.

Like previous findings with IBD populations, illness-acceptance predicted resilience (Kiebles et al., 2010; Sirois & Hirsch, 2017); perhaps because it positivises IBD's meaning, aiding tolerance of its unpredictable, uncontrollable nature (Evers et al., 2001). Those with greater acceptance also possessed greater coping-efficacy, so it may

be that the implementation of effective coping strategies enables acceptance and resilience (Carver, 1998). It is possible that a reciprocal and mutually reinforcing relationship exist between these variables and resilience, however future longitudinal research would be needed to elucidate causal pathways. Finally, the positive correlation between social support and illness-acceptance mirrors previous findings (Janowski et al., 2012). The qualitative analysis indicated complexities within the relationship between social support and resilience, thus while it was found to predict resilience, the FSSQ (Broadhead et al, 1988) did not capture these complexities, limiting the findings.

Resilience and disease variables

Disease-activity negatively predicted resilience explaining 15.7% of the variance in the model (prior to controlling for the other variables). This was unsurprising because higher disease-activity associates with distress (Graff et al., 2006) depression and anxiety (Tribbick et al., 2017) and lower QOL (Vander-Have, 2014). (Dorrian et al., 2009; Knowles et al., 2011; Knowles et al., 2013; Zhang et al., 2016). These findings suggested that once social support, acceptance, and coping-efficacy were controlled for, combined, they reduced the negative effect on resilience of disease activity, turning it into a small positive effect. This indicates interrelationships between the variables and disease activity and that their effects are not in isolation.

Prior research has elucidated that disease-activity can directly impact perceptions of one's illness influencing a negative mindset and that they mediate the association between disease-activity and negative outcomes (Tribbick et al., 2017). The negative appraisals in these existing studies pertain to chronicity and lack of controllability. The findings in this study indicate a complex relationship between disease activity, the variables under examination in this study and resilience. Thus,

further research could extend these findings by conducting moderator or mediator analyses to diversify the types of illness perceptions under examination and further elucidate the role of disease activity and these variables in relation to resilience.

Acceptance was positively and significantly correlated with TSD. This suggests acceptance may evolve with experience, perhaps because a process of desensitisation to aversive IBD experiences occurs over time (Carver, 2010). Resilience is a dynamic process (Skrastins & Fletcher, 2016; Werner, 1994) and longitudinal research that collects data from the point of diagnosis over the disease course would further understanding of the temporal relations between variables and resilience.

Study limitations

The study limitations should be acknowledged when interpreting the findings. Phase A yielded a small sample size (Braun & Clarke, 2013; Fugard & Potts, 2014). However, the data were deemed adequate to meet the study aims. Selection bias may have influenced who volunteered (Khazaal et al., 2014) towards those perceiving themselves as resilient. The sample was a homogenous population of white, British, educated females in remission. Thus, findings are less transferable to others, particularly those experiencing active disease. Reflective diary keeping would have aided reflection on the researcher's contributions to the interpretation of data (Ortlipp, 2008).

Due to the cross-sectional design, causal inferences could not be determined (Sedgwick, 2014). Furthermore, the design did not enable exploration of the influence of confounding factors including personality traits or emotional disorders which may impact on resilience (Kim et al., 2013). Additionally, self-report measures bear the risk of recall bias and shared method variance; meaning the results could have been artificially inflated (Podsakoff et al., 2003). The measures used were psychometrically

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sound, however the Likert scales for the CES and CBI were entered erroneously, thus findings for those measures should be interpreted with caution. Finally, the study relied upon self-reported diagnoses and disease-activity. While recruitment from clinical settings may have improved the accuracy of this data, arguably, capturing perceived disease severity holds more value and self-reports are comparable to medical reports (Randell et al., 2014).

The complexities of defining resilience are widely recognised (Windle, 2010). Within this study, resilience was measured as resilience-related traits (Connor & Davidson, 2010) and as a dependent variable which is fluid in nature but was captured cross-sectionally. While this research was exploratory and aimed to develop a better conceptual understanding of resilience in those with IBD, these conceptual, measurement and design issues, mean that caution should be applied when interpreting the findings and when comparing these findings with other studies.

The current sample were only partially representative of the general population. Those known to have the highest diagnostic incidence fall within the age-bracket of 20-29 years (Johnston & Logan, 2008) and in this study the mean diagnostic age was 26.53 years. However, approximately three quarters were female and epidemiological studies suggest there is a 1:3 incidence ratio of males to females (Bernstein et al., 2006; Brant & Nguyen, 2008, GDB collaborators 2019).

Nevertheless, the post-hoc analysis indicated that the sample size was adequate to avoid type I or II errors. The bivariate correlations between variables and resilience were at the .01 alpha level which reduces the risk of type 1 errors. Finally, this exploratory research adds to a burgeoning body of literature on resilience in IBD populations.

Implications for clinical practice

These findings indicate that across the illness trajectory fostering one's ILOC, coping-efficacy and illness-acceptance may improve resilience. In line with current guidance (NICE, 2010), CBT enables practitioners to foster these psychological resources and challenge negative appraisals. However, acceptance is more specifically addressed through the application of third-wave therapies, such as Mindfulness-Based Stress Reduction (Kabat-Zinn, 1990), Compassion Focused Therapy (Gilbert, 2009) or Acceptance and Commitment Therapy (ACT, Hayes, 2004), which aim to enable one's ability to embrace the present moment and difficult emotions (Khoury et al., 2013). A review of ACT revealed promising findings for those with long-term conditions (Graham et al., 2016). Additionally, targeting IOU as a transdiagnostic factor in line with CBT for Generalised Anxiety Disorder (NICE, 2011) might foster resilience. Future research could examine the effectiveness of third-wave therapies for those with IBD.

Fostering the aforementioned factors may be particularly important when disease-activity is high. Higher disease activity also leads to a lower perception of social support; thus, practitioners should encourage engagement with health-care services at these times. It may be that professional support is creatively adapted such as utilising phone or video contacts so that it is accessible to individuals where disease activity imposes physical restrictions.

Conclusions

The current study found that illness cognitions pertaining to coping resources and beliefs about controllability predict resilience over and above the negative predictive influence of disease activity. Rather than perseverance to long-term goals, daily persistence or endurance might be more important to resilience in an IBD population. Future research should examine endurance, causal pathways between illness cognitions and resilience, and the mediating influences of these illness cognitions between disease activity and resilience. Finally, research could examine the application and effectiveness of third-wave therapies in those with IBD.

References

- Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioural change. *Psychological Review*, 84(2), 191-215. <https://doi.org/10.1037/0033-295X.84.2.191>.
- Bandura, A. (1997). *Self-efficacy: The exercise of control*. W H Freeman/Times books/Henry Holt & Co. <https://psycnet.apa.org/record/1997-08589-000>
- Beck, A. (1976). *Cognitive therapy and the emotional disorders*. International Universities Press. <https://psycnet.apa.org/record/1976-28303-000>.
- Bennebroek, F., Thijssens, N., Stokkers, P., Grootenhuis, M., Bockting, C., Nieuwkerk, P., & Sprangers, M. (2012). Do inflammatory Bowel Disease patients with anxiety and depressive symptoms receive the care they need? *Journal Crohn's Colitis*, 6(1), 68-76. <https://doi:10.1016/j.crohns.2011.07.006>.
- Bernstein, C., Wajada, A., Svenson, I., MacKenzie, A., Koehoorn, M., Jackson, M., Fedorak, R., & Blanchard, J. (2006). The epidemiology of inflammatory bowel disease in Canada: A population-based study. *American Journal of Gastroenterology*, 101(7), 1559-1568. <https://10.1111/j.1572-0241.2006.00603.x>.
- Bielefeldt, K., Davis, B., & Binion, D. (2009). Pain and inflammatory bowel disease. *Inflammatory Bowel Disease*, 15(5), 778-788. <https://doi:10.1002/ibd.20848>.
- Bowlby, J. (1969). *Attachment and loss. Vol. I. Attachment*. Basic Books.
- Brant, S., & Nguyen, G. (2008). Is there a gender difference in the prevalence of Crohn's disease or ulcerative colitis? *Inflammatory Bowel Diseases*, 14(S2), S2-S3. <https://doi.org/10.1002/ibd.20540>.

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- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3(2), 77-101. <https://dx.doi.org/10.1191/1478088706qp063oa>.
- Braun, V., & Clark, V. (2013). *Successful qualitative research: A practical guide for beginners*. Sage. <http://www.uk.sagepub.com/books/Book233059>.
- British Psychological Association Code of Human Research Ethics (2014). *Code of human research ethics*. <https://www.bps.org.uk/sites/www.bps.org.uk/files/Policy/Policy%20%20Files/BPS%20Code%20of%20Human%20Research%20Ethics>.
- British Psychological Society (2018). *Code of ethics and conduct*. <https://www.bps.org.uk/sites/www.bps.org.uk/files/Policy/Policy%20-%20Files/BPS%20Code%20of%20Ethics%20and%20Conduct%20%28Updated%20July%202018%29.pdf>.
- Broadhead, W., Gehlbach, S. De Gruy, F., & Kaplan, B. (1988). The duke-UNC functional social support questionnaire. Measurement of social support in family medicine patients. *Medical Care*, 26(7), 709-23. <https://www.ncbi.nlm.nih.gov/pubmed/3393031>.
- Brocki, J., & Wearden, A. (2006). A critical evaluation of the use of interpretative phenomenological analysis (IPA) in health psychology. *Psychology and Health*, 21(1), 87-108. <https://doi.org/10.1080/14768320500230185>.
- Buhr, K., & Dugas, M. (2009). The role of fear of anxiety and intolerance of uncertainty in worry: An experimental manipulation. *Behavioural Research Therapy*, 47(3), 215-23. <https://doi:10.1016/j.brat.2008.12.004>.

Factors that associate with resilience in people with IBD

- Carleton, R., Norton, M., & Asmundson, G. (2007). Fearing the unknown: a short version of the intolerance of uncertainty scale. *Journal of Anxiety Disorders, 21*(1), 105-117. <https://doi:10.1016/j.janxdis.2006.03.04>.
- Carleton, R., Gosselin, P., & Asmundson, G. (2010). The intolerance of uncertainty index: replication and extension with an English sample. *Psychology Assessment, 22*(2), 396-406. <https://doi:10.1037/a0019230>.
- Carver, C. (1998). Resilience and thriving: Issues, models, and linkages. *The Journal of Social Issues, 54*, 245–266. <https://doi.org/10.1111/j.1540-4560.1998.tb01217.x>.
- Carver, C. (2010). Resilience and thriving: Issues, models, and linkages-Thriving: Broadening the paradigm beyond illness to health. <https://www.semanticscholar.org/paper/Resilience-and-thriving-%3A-issues-%2C-models-%2C-and-%3A-Carver/5ab2e4a6396edb3ed09fbb827327c790d5fdf2f4>
- Casellas, F., Lopez-Vivancos, J., Casado, A., & Malagelada, J. (2002). Factors affecting health related quality of life of patients with inflammatory bowel disease. *Quality of Life Research, 11*(8), 775– 781. <https://doi.org/10.1023/A:1020841601110>.
- Casellas, F., Arenas, J., Baudet, J., Fabregas, S., Garcia, N., Gelabert, J., Medina, C., Ochtorena, I., Papo, M., Rodrigo, L., & Malagelada, J. (2005). Impairment of health-related quality of life in patients with inflammatory bowel disease: a Spanish multicentre study. *Inflammatory Bowel Disease, 11*(5), 488–96. <https://doi:10.1097/01.MIB.0000159661.55028.56>.
- Charney, D. (2004). Psychobiological mechanisms of resilience and vulnerability: implications for successful adaptation to extreme stress. *American Journal of Psychiatry, 161*, 195-216. <http://dx.doi.org/10.1176/appi.ajp.161.2.195>

Factors that associate with resilience in people with IBD

- Cohen, J. (1992). Quantitative methods in Psychology. A power primer. *Psychological Bulletin*, 112(1),155.<http://www.bwgriffin.com/workshop/Sampling%20A%20Cohen%20tables.pdf>.
- Connor, K., & Davidson, J. (2003). Development of a new resilience scale: The Connor-Davidson resilience scale (CD-RISC). *Depression and Anxiety*, 18(2), 76-82.
<https://pdfs.semanticscholar.org/cdf7/e48f5b42847e1d7cabd7a4fdb3761a40279.pdf>.
- Creswell, J., Plano Clark, V., Gutmann, M., & Hanson, W. (2003). Advanced mixed methods research designs. In A. Tashakkori & C. Teddie (Eds), *Handbook of mixed methods in social and behavioural research* (p.209-240). Thousand Oaks, CA: Sage.
- Davydov, D., Stewart, R., Ritchie, K., & Chaudieu, I. (2010). Resilience and mental health. *Clinical Psychology Review*, 30, 479-95. <https://doi:10.1016/j.cpr.2010.03.003>.
- Devlen, J., Beusterie, K., Yen, L., Ahmed, A., Cheifetz, A., & Moss, A. (2014). The burden of inflammatory bowel disease: A patient-reported qualitative analysis and development of a conceptual model. *Inflammatory Bowel Disease*, 20(3), 545-52.
<https://doi:10.1097/01.MIB.0000440983.86659.91>.
- Dibley, L., Norton, C., & Whitehead, E., (2018). The experience of stigma in inflammatory bowel disease: An interpretative (hermeneutic) phenomenology study. *Global Nursing Research*, 74(4), 838-851. <https://doi.org/10.1111/jan.13492>.
- Dorrian, A, Dempster M., & Adair, P. (2009). Adjustment to inflammatory bowel disease: the relative influence of illness perceptions and coping. *Inflammatory Bowel Disease*, 15(1), 47–55. <https://doi:10.1002/ibd.20583>.

Factors that associate with resilience in people with IBD

- Downey, R., & King, C. (1998). Missing data in Likert ratings: A comparison of replacement methods. *Journal of General Psychology, 125*(2), 175-91.
<https://doi:10.1080/00221309809595542>.
- Duckworth, A., Peterson, C., Matthews, M., & Kelly, D. (2007). "Grit: Perseverance and passion for long-term goal." *Journal of Personality and Social Psychology, 92*(6), 1087–1101. <https://doi:10.1037/0022-3514.92.6.1087>.
- Duckworth, A., & Quinn, P. (2009). Development and validation of the Short Grit Scale (GRIT-S). *Journal of Personality Assessment, 91*(2), 166-174.
<https://doi.org/10.1080/00223890802634290>.
- Dudley-Brown, S. (1996). Living with Ulcerative Colitis. *Gastroenterological Nursing, 19*(2), 60-64. <https://doi:10.1097/00001610-199603000-00004>.
- Dugas, M., Freeston, M., & Ladouceur, R. (1997). Intolerance of uncertainty and problem orientation in worry. *Cognitive Therapy and Research, 21*(6), 593–606.
<https://doi.org/10.1023/A:1021890322153>.
- Eekhout, I., Boer, R., Twisk, J., Vet, H., & Heyman, M. (2012). Missing data: A systematic review of how they are reported and handled. *Epidemiology, 23*(5), 729-32.
<https://doi:10.1097/EDE.0b013e3182576cdb>.
- Evers, A. Kraaimaat, F., Van Lankveld, W., Jongen, P., Jacobs, J., & Bijlsma, J. (2001). Beyond unfavourable thinking: The illness cognition questionnaire for chronic disease. *Journal of Consulting and Clinical Psychology, 69*(6), 1026-1036.
<http://doi.org/10.1037/0022-006X.69.6.1026>.

Factors that associate with resilience in people with IBD

- Fan, S., Eiser, C., & Ho, M. (2010). Health-related quality of life in patients with hepatocellular carcinoma: a systematic review. *Clinical Gastroenterology Hepatology*, 8(7), 559-564. <https://doi.org/10.1016/j.cgh.2010.03.008>.
- Field, A. (2009). *Discovering statistics using SPSS (Third edition) (and sex and drugs and rock 'n' roll)*. file:///C:/Users/Katie/Downloads/Andy%20Field%20(3).pdf.
- Fletcher, P., Schneider, M., Van Ravenswaay, V., & Leon, Z. (2008). I am doing the best that I can! Living with inflammatory bowel disease and/or irritable bowel syndrome (part II). *Clinical Nurse Specialist*, 22(6), 278-285. <https://doi:10.1097/01.NUR.0000325382.99717.ac>.
- Fourie, S., Jackson, D., & Aveyard, H. (2018) Living with Inflammatory Bowel Disease: A review of qualitative studies. *International Journal of Nursing Studies*, 87, 149-156. <https://doi:10.1016/j.ijnurstu.2018.07.017>.
- Fox-Wasylyshyn, S., & El-Masri, M. (2005) Handling Missing Data on Self-Report Measures. *Research in Nursing and Health*, 28, 488-495. <https://doi.org/10.1002/nur.20100>.
- Freud, S. (1900). *The interpretation of dreams. S.E., vols IV and V*. Hogarth Press. <https://psychclassics.yorku.ca/Freud/Dreams/dreams.pdf>
- Fugard, A., & Potts, H. (2015). Supporting thinking on sample sizes for thematic analyses: a quantitative tool. *International Journal of Social Research Methodology*, 18(6), 669-684. <https://doi.org/10.1080/13645579.2015.1005453>.

Factors that associate with resilience in people with IBD

- Gandhi, S., Jedel, S., Hood, M., Mutlu, E., Swanson, G., & Keshavarzian, A. (2014). The relationship between coping, health competence and patient participation among patients with inactive inflammatory bowel disease. *Journal of Crohn's and Colitis*, 8(5), 401-8. <https://doi:10.1016/j.crohns.2013.10.005>.
- Gentes, E., & Ruscio, A. (2011). A meta-analysis of the relation of intolerance of uncertainty to symptoms of generalized anxiety disorder, major depressive disorder, and obsessive-compulsive disorder. *Clinical Psychological Review*, 31(6), 923-933. <https://doi:10.1016/j.cpr.2011.05.001>.
- Gheshlagh, R., Sayehmiri, K., Ebadi, A., Dalvandi, A., Dalvand, S., & Tabrizi, K. (2016). Resilience of patients with chronic physical diseases: A systematic review and meta-analysis. *Iran Red Crescent Medical Journal*, 18(7), e38562. <https://doi:10.5812/ircmj.3856>.
- Gick, M., & Sirois, F. (2010). Insecure attachment moderates' women's adjustment to inflammatory bowel disease severity. *Rehabilitation Psychology*, 55(2), 170–179. <https://doi:10.1037/a0019358>.
- Gignac, M., Cott, C., & Badley, E. (2000). Adaptation to chronic illness and disability and its relationship to perceptions of independence and dependence. *Journals of Gerontology: Series B: Psychological Sciences and Social Sciences*, 55(6), 362-372. <https://doi.org/10.1093/geronb/55.6.P362>.
- Gilbert, P. (2009). Introducing compassion focused therapy. *Advances in Psychiatric Treatment*, 15(3), 199-208. <https://doi:10.1192/apt.bp.107.005264>.

- Global Burden Disease (GBD) collaborators (2020). The global, regional, and national burden of inflammatory bowel disease in 195 countries and territories, 1990–2017: a systematic analysis for the Global Burden of Disease Study 2017. *Lancet Gastroenterol Hepatology*, 5, 17–30. <https://www.thelancet.com/action/showPdf?pii=S2468-1253%2819%2930333-4>.
- Goodhand, J., Wahed, M., Mawdsley, J., Farmer, A., Aziz, Q., & Rampton, D. (2012). Mood disorders in inflammatory bowel disease: Relation to diagnosis, disease activity, perceived stress, and other factors. *Inflammatory Bowel Diseases*, 18, 2301–2309. <https://doi.org/10.1002/ibd.22916>.
- Graff, L., Walker, J., Lix, L., Clara, I., Rawsthorne, P., Rogala, L., Miller, N., Jakul, L., McPhail, C., Ediger, J., & Bernstein, C. (2006). The relationship of inflammatory bowel disease type and activity to psychological functioning and quality of life. *Clinical Gastroenterological Hepatology*, 4(12), 1491-1501. <https://doi:10.1016/j.cgh.2006.09.027>.
- Graff, L., Walker, J., Clara, L., Lix, L., Miller, N., Rogala, L., Rawsthorn, P., & Bernstein, C. (2009). Stress coping, distress, and health perceptions in inflammatory bowel disease and community controls. *American Journal of Gastroenterology*, 104(12), 2959-69. <https://doi:10.1038/ajg.2009.529>.
- Graham, C., Gouick, J., Krahe, C., & Gillanders, D. (2016). A systematic review of the use of Acceptance and Commitment Therapy (ACT) in chronic disease and long-term conditions. *Clinical Psychology Review*, 46, 46-58. <https://doi:10.1016/j.cpr.2016.04.009>.

Factors that associate with resilience in people with IBD

- Green, C., & Murdock, K. (2013). Multidimensional Control Beliefs, Socioeconomic Status, and Health. *American Journal of Health Behaviour, 37*(2), 227–237.
<https://doi:10.5993/AJHB.37.2.10>.
- Guindi, M., & Riddell, R. (2004). Indeterminate colitis. *Journal of Clinical Pathology, 57*(12), 1233-1244. <https://doi:10.1136/jcp.2003.015214>.
- Halcomb, E., & Hickman, L. (2015). Mixed methods research. *Nursing Standard, 29*(32), 41-47. <https://doi:10.7748/ns.29.32.41.e8858>.
- Hale, W., Richmond, M., Bennett, J., Berzins, T., Fields, A., Weber, D., Beck, M., & Osman, A. (2016). Resolving uncertainty about the Intolerance of Uncertainty Scale–12: Application of modern psychometric strategies. *Journal of Personality Assessment, 98*(2), 200–208. <https://doi.org/10.1080/00223891.2015.1070355>.
- Hall, N., Rubin, G., Dougall, A., Hungin, P., & Neely, J. (2005). The fight for “health-related normality”: A qualitative study of the experiences of individuals living with established inflammatory Bowel Disease. *Journal of Health Psychology, 10*(3), 443–455.
<https://doi:10.1177/1359105305051433>.
- Hamby, S., Grych, J., & Banyard, V. (2013). *Life Paths Research measurement packet*. Sewanee, TN: Life Paths Research Program.
- Hanna, D., & Dempster, M. (2012). *Psychology statistics for dummies*. John Wiley & Sons.
- Hayes, S. (2004). Acceptance and commitment therapy, relational frame theory, and the third wave of behavioural and cognitive therapy. *Behaviour Therapy, 47*(6), 869-885.
[https://doi:10.1016/S0005-7894\(04\)80013-3](https://doi:10.1016/S0005-7894(04)80013-3).

- Heijmans, M., & De Ridder, D. (1998). Assessing illness representations of chronic illness: explorations of their disease-specific nature. *Journal of Behavioural Medicine, 21*(5), 485–503. <https://doi.org/10.1023/A:1018788427100>.
- Hjortswang, H., Jarnerot, G., Curman, B., Sandberg-Gertzen, H., Tysk, C., Blomberg, B., Almer, S., & Strom, M. (2006). The SHS: a valid measure of subjective health in ulcerative colitis. *Scandinavian Journal of Gastroenterology, 41*(10), 1196–203. <https://doi.org/10.1080/00365520600610618>.
- Janke, K., Raible, A., Bauer, M., Clemens, P., Meisner, A., Hauser, W., Steder-Neukamm, U., Henrich, G., Herschbach, P., Gregor, M., & Klump, B. (2004). Questions on life satisfaction in inflammatory bowel disease. *International Journal of Colorectal Disease, 19*(4), 343–353. <https://doi.org/10.1007/s00384-003-0522-z>.
- Janowski, K., Steuden, S., Pietrzak, A., Krasowska, D., Kaczmarek, L., Gradus, I., & Chodorowska, G. (2012). Social support and adaptation to the disease in men and women with psoriasis. *Arch Dermatol Research, 304*(6), 421–432. <https://doi.org/10.1007/s00403-012-1235-3>.
- Johnston, R., & Logan, R. (2008). What is the peak age for onset of IBD? *Inflammatory Bowel Diseases, 14*(S2), S4–S5. <https://doi.org/10.1002/ibd.20545>.
- Jones, M., Wessinger, S., & Crowell, M. (2006). Coping strategies and interpersonal support in patients with irritable bowel syndrome and inflammatory bowel disease. *Clinical Gastroenterology Hepatology, 4*(4), 474–81. <https://doi.org/10.1016/j.cgh.2005.12.012>.
- Kabat-Zinn, J. (1990). *Full Catastrophe Living: the program of the Stress Reduction Clinic at the University of Massachusetts Medical Centre*. Dell.

Factors that associate with resilience in people with IBD

- Kamp, K. West, P., Holmstrom, A., Luo, Z., Wyatt, G., & Given, B. (2019). Systematic review of social support on psychological symptoms and self-management behaviours among adults with inflammatory bowel disease. *Journal of Nursing Scholarship, 51*(4), 380-389. <https://doi:10.1111/jnu.12487>.
- Katz, L., Tripp, D., Ropeleski, M., Depew, W., Curtis Nickel, J., Vanner, S., & Beyak, M. (2016). Mechanisms of quality of life and social support in Inflammatory Bowel Disease. *Journal of Clinical Psychology in Medical Settings, 23*(1), 88-98. <https://doi:10.1007/s10880-015-9431-x>.
- Kemp, K., Griffiths, J., & Lovell, K. (2012). Understanding the health and social care needs of people living with IBD: A meta-synthesis of the evidence. *World Journal of Gastroenterology, 18*(43), 6240–6249. <https://doi:10.3748/wjg.v18.i43.6240>.
- Kernberg, O. (1975). *Borderline conditions and pathological narcissism*. Jason Aronson.
- Khazaal, Y., Singer, M., Chatton, A., Achab, S., Zullino, D., Rothen, S., Khan, R., Billieux, J., & Thorens, G. (2014). Does self-selection affect samples' representativeness in online surveys? An investigation in online video game research. *Journal of Medical Internet Research, 16*(7). <https://doi:10.2196/jmir.2759>.
- Khoury, B., Lecomte, T., Gaudiano, B. Brandon, A., & Paquin, K. (2013). Mindfulness interventions for psychosis: A meta-analysis. *Schizophrenia Research, 150*(1), 176-184. <https://doi:10.1016/j.schres.2013.07.055>.
- Kiebles, J., Doerfler, B., & Keefer, L. (2010). Preliminary evidence supporting a framework of psychological adjustment to inflammatory bowel disease. *Inflammatory Bowel Diseases, 16*(10), 1685–1695. <https://doi.org/10.1002/ibd.21215>.

Factors that associate with resilience in people with IBD

Kim, J., Lee, H., & Lee, K. (2013). Influence of temperament and character on resilience.

Comprehensive Psychiatry, 54(7), 1105-1110.

<https://doi.org/10.1016/j.comppsy.2013.005>

Knowles, S., Cook, S., & Tribbick, D. (2013). Relationship between health status, illness

perceptions, coping strategies and psychological morbidity: a preliminary study with

IBD stoma patients. *Journal of Crohn's & Colitis*, 7(10), e471–

e478. <https://doi.org/10.1016/j.crohns.2013.02.022>.

Knowles, S., Wilson, J., Connell, W., & Kamm, M. (2011). Preliminary examination of the

relations between disease activity, illness perceptions, coping strategies, and

psychological morbidity in Crohn's disease guided by the common sense model of

illness. *Inflammatory Bowel Disease*, 17(12), 2551–2557.

<https://doi:10.1002/ibd.21650>.

Krueger, R., & Casey, M. (2015). *Focus groups: A practical guide for applied research*. Sage

publications.

Kurita, K., Garon, E., Stanton, A., & Meyerowitz, B. (2013). Uncertainty and psychological

adjustment in patients with lung cancer. *Psychooncology*, 22(6), 1396–1401.

<https://doi:10.1002/pon.3155>.

Kurpas, D., Mroczek, B., Knap-Czechowska, H., Bielska, D., Nitsch-Osuch, A., Kassolik, K.,

Andrzejewski, W., Gryko, A., & Steciwko, A. (2013). Quality of life and acceptance

among patients with chronic respiratory diseases. *Respiratory Physiology and*

Neurobiology, 187(1), 114-117. <https://doi:10.1016/j.resp.2013.02.009>.

Factors that associate with resilience in people with IBD

- Larsson, K., Loof, L., Ronnblom, A., & Nordin, K. (2008). Quality of life for patients with exacerbation in inflammatory bowel disease and how they cope with disease activity. *Journal Psychosomatic Research, 64*(2), 139-48.
<https://doi:10.1016/j.jpsychores.2007.10.007>.
- LeDoux, J. (1998). *The emotional brain*. Weidenfeld and Nicolson.
- Lenze, E., Shardwell, M., Ferrell, R., Orwig, D., Yu-Yahiro, J., Hawkes, W., Fredman, L., Miller, R., & Magaziner, J. (2008). Association of serotonin-1A and 2A receptor promoter polymorphisms with depressive symptoms and functional recovery in elderly persons after hip fracture. *Journal of Affective Disorders, 111*(1), 61-66.
<https://doi:10.1016/j.jad.2008.02.005>.
- Lewko, J., Zarzycki, W., & Krajewska-Kulak, E. (2012). Relationship between the occurrence of symptoms of anxiety and depression, quality of life, and level of acceptance of illness in patients with type 2 diabetes. *Saudi Medical Journal, 33*(8), 887-94.
<https://pubmed.ncbi.nlm.nih.gov/22886123/>.
- Little, R. (1988). A Test of Missing Completely at Random for Multivariate Data with Missing Values. *Journal of the American Statistical Association, 83*(404), 1198-1202.
<https://doi:10.1080/01621459.1988.10478722>.
- Luo, D., Lin, Z., Shang, X., & Li, S. (2019). "I can fight it!": A qualitative study of resilience in people with inflammatory bowel disease. *International Journal of Nursing Science, 6*(2), 127-133. <https://doi:10.1016/j.ijnss.2018.12.008>.
- Madill, A., Jordan, A., & Shirley, C. (2000). Objectivity and reliability in qualitative analysis: Realist, contextualist and radical constructionist epistemologies. *British Journal of Psychology, 91*, 1-20. <https://doi:10.1348/000712600161646>.

- Maxwell, J., & Mittapalli, K. (2010). *Realism as a stance for mixed methods research*. In: SAGE Handbook of mixed methods in social and Behavioural Research. <http://dx.doi.org/10.4135/9781506335193.n6>.
- Mcdermott, E., Keegan, D., Byrne, K., Doherty, G., & Mulcahy, H. (2013). The short health scale: A valid and reliable measure of health quality of life in English speaking inflammatory bowel disease patients. *Journal of Crohn's and Colitis*, 7(8), 616-621. <https://doi.org/10.1016/j-cohns.2012.07.030>.
- McEvoy, P., & Mahoney, A. (2012). To be sure, to be sure: intolerance of uncertainty mediates symptoms of various anxiety disorders and depression. *Behavioural Therapy*, 43(3), 533-545. <https://doi:10.1016/j-beth.2011.02.007>.
- Molodecky, N., Soon, I., Rabi., Ghali, W., Ferris, M., Chernoff, G., Benchimol, E., Panaccione, R., Ghosh, S., Barkema, H., & Kaplan, G. (2012). Increasing incidence and prevalence of the inflammatory bowel diseases with time, based on systematic review. *Gastroenterology*, 142(1), 46-54. <https://doi:10.1053/j.gastro.2011.10.001>.
- Moore, A., Grime, J., Campbell, P., & Richardson, J. (2012). Troubling stoicism: Sociocultural influences and applications to health and illness behaviour. *Health*, 17(2), 159-173. <https://doi:10.1177/136459312451179>.
- Moum, B., Vatn, M., Ekbohm, A., Aadland, E., Fausa, O., Lygren, I., Sauar, J. Schulz, T., & Stray, N. (1996). Incidence of ulcerative colitis and indeterminate colitis in four counties of south-eastern Norway, 1990–93. A prospective population-based study. The Inflammatory Bowel South Eastern Norway (IBSEN) Study Group of Gastroenterologists. *Scandinavian Journal Gastroenterology*, 31(4), 362–366. <https://doi:10.3109/00365529609006411>.

Factors that associate with resilience in people with IBD

- Munkholm, P., Lanholz, E., Davidsen, M., & Binder, V. (1995). Disease activity courses in a regional cohort of Crohn's disease patients. *Scandinavian Journal of Gastroenterology*, 30(7), 699–706. <https://doi:10.3109/00365529509096316>.
- National Institute for Health and Care Excellence (NICE). (2010). *Depression in adults with a chronic health condition (guideline 91)*. <https://www.nice.org.uk/guidance/cg91/evidence/full-guideline-243876061>.
- National Institute for Health and Care Excellence (NICE). (2015). *Inflammatory Bowel Disease. Quality standard. Guidance QS81*. <https://www.nice.org.uk/guidance/cg159>.
- Niv, G., Josef, B., Bassat, B., Avni, I., Lichtenstein, L., Niv, Y., & Barnoy, S. (2017). Quality of life and uncertainty in Crohn's disease. *Quality of Life Research*, 26(6), 1609-1616. <https://doi:10.1007/s11136-017-1509-5>.
- Nordin, K., Pahlman, L., Larsson, K., Sundberg-Hjelm., & Loof, L. (2002). Health-related quality of life and psychological distress in a population-based sample of Swedish patients with inflammatory bowel disease. *Scandinavian Journal Gastroenterology*, 37(4), 450–457. <https://doi.org/10.1080/003655202317316097>.
- Oliveira, S., Zaltman, C., Elia, C., Vargens, R., Leal, A., Barros, R., & Fogaca, H. (2007). Quality-of-life measurement in patients with inflammatory bowel disease receiving social support. *Inflammatory Bowel Diseases*, 13(4), 470–474. <https://doi.org/10.1002/ibd.2007>.
- Ortlipp, M. (2008). Keeping and Using Reflective Journals in the Qualitative Research Process. *The Qualitative Report*, 13(4), 695-705. <https://nsuworks.nova.edu/tqr/vol13/iss4/8>.

Factors that associate with resilience in people with IBD

- Ozbay, F., Fitterling, D., Charney., & Southwick, S. (2008). Social support and resilience to stress across the life span: a neurobiologic framework. *Current Psychiatry Reports, 10*, 304-310. <http://dx.doi.org/10.1007/s11920-008-0049-7>
- Palent, A., & Himmel, W. (2019). Are there also negative effects of social support? A qualitative study of patients with inflammatory bowel disease. *British Medical Journal, 9*, 226-242. <https://doi:10.1136/bmjopen-2018-022642>.
- Panagiotou, I., Tsilika, E., Parpa, E., Patiraki, E., Zygogianni, A., Kouloulas, V., & Mystakidou, K. (2014). Locus of control and distressing symptoms in young cancer patients when assessing depression. *JBUON, 19*(3), 792-798. <https://pubmed.ncbi.nlm.nih.gov/25261669/>.
- Park, C., & Gaffey, A. (2007). Locus of control, quality of life, anxiety and depression among Malaysian breast cancer patients: The mediating role of uncertainty. *European Journal of Oncology Nursing, 27*, 28-35. <https://dx.doi.org.10.1016/j.ejon.2017.01.005>.
- Patton, M. (1990). *Qualitative Evaluation and Research Methods* (2nd ed.). Sage Publications, Inc.
- Pihl-Lesnovska, K., Hjortswang, H., Ek, A., & Frisman, G. (2010). Patients perspective of factors influencing quality of life while living with Crohn disease. *Gastroenterology Nursing, 33*(1), 37-44. <https://doi:10.1097/SGA.0b013e318cd49d0>.
- Podsakoff, P., MacKenzie, S., Lee, J., & Podsakoff, N. (2003). Common method biases in behavioural research: A critical review of the literature and recommended remedies. *Journal of Applied Psychology, 88*(5), 879-903. <https://doi.org/10.1037/0021-9010.88.5.879>.

Factors that associate with resilience in people with IBD

- Porcelli, P., Leoci, C., & Guerra, V. (1996). A prospective study of the relationship between disease activity and psychologic distress in patients with inflammatory bowel disease. *Scandinavian Journal Gastroenterology*, *31*(8), 792–796.
<https://www.ncbi.nlm.nih.gov/pubmed/8858749>.
- Randell, R., Long, M, Cook, S., Wrennall, C., Chen, W., Martin, C., Anton, K., Sandler, R., Kappelman, M. (2014). Validation of an Internet-based cohort of inflammatory bowel disease (CCFA Partners). *Inflammatory Bowel Diseases*, *20*(3), 541–544.
<https://doi.org/10.1097/01.MIB.0000441348.32570.34>
- Raymond, M. (1986). Missing data in evaluation research. *Evaluation & the Health Professions*, *9*(4), 395–420. <https://doi.org/10.1177/016327800900401>.
- Restall, G., Simms, A., Walker, J., Graff, L., Sexton, K., Rogala, L., Miller, N., Haviva, C., Targwnik, L., & Bernstein, C. (2016). Understanding work experiences of people with inflammatory bowel disease. *Inflammatory Bowel Disease*, *22*(7), 1688-97.
<https://doi:10.1097/MIB.0000000000000826>.
- Ris, E. (2015) "Grit: A Short History of a Useful Concept." *Journal of Educational Controversy*, *10*, 1-18. <https://cedar.wvu.edu/jec/vol10/iss1/3>.
- Roth, P., & BeVier, C. (1998). Response rates in HRM/OB survey research: Norms and correlates, 1990-1994. *Journal of Management*, *24*, 97-117.
<https://doi.org/10.1177/014920639802400107>.
- Rotter, J. (1954). *Social learning and clinical psychology*. Prentice-Hall.

- Rotter, J. (1975). Some problems and misconceptions related to the construct of internal versus external control of reinforcement. *Journal of Consulting and Clinical Psychology*, 43(1), 56-67. <http://psycnet.apa.org/buy/1975-11748-001>.
- Rubin, G., Hungin, A., Chinn, D., & Dwarakanath, D. (2004). Quality of life in patients with established inflammatory bowel disease: a UK general practice survey. *Aliment Pharmacological Therapy*, 19(5), 529-535. <https://www.ncbi.nlm.nih.gov/pubmed/14987321>.
- Rutter, M. (1987). Psychosocial resilience and protective mechanisms. *American Journal of Orthopsychiatry*, 57(3), 316-331. <https://doi:10.1111/j.1939-0025.1987.tb03541.x>.
- Ryle, A. (1999). *Cognitive Analytic Therapy: Active participation in change. A new integration in brief Psychotherapy*. John Wiley and sons.
- Scharloo, M., Kaptein, A., Weinman, J., Hazes, M., Willems, L., Bergman, W., & Rooijmans, G. (1998). Illness perceptions, coping and functioning in patients with rheumatoid arthritis, chronic obstructive pulmonary disease and psoriasis. *Journal of Psychosomatic Research*, 44(5), 573-85. [https://doi:10.1016/S0022-3999\(97\)00254-7](https://doi:10.1016/S0022-3999(97)00254-7).
- Schneider, I., & Schwarz, N. (2017). Mixed feelings: the case of ambivalence. *Current Opinion in Behavioural Sciences*, 15, 39-45. <https://doi:10.1016/j.cobeha.2017.05.012>.
- Sedgwick, P. (2014). Cross sectional studies: advantages and disadvantages. *British Medical Journal*, 348, 2276. <https://doi:10.1136/bmj.g2276>.
- Sharkey, C., Bakula, D., Gamwell, K., Mullins, A., Chaney, J., & Mullis, L. (2017). The role of grit in college student health care management skills and health-related quality of life. *Journal of Pediatric Psychology*, 42(9), 952-961. <https://doi:10.1093/jpepsy/jsx073>.

Factors that associate with resilience in people with IBD

- Sharkey, C., Bakula, D., Baraldi, A., Perez, M., Suorsa, K., Chaney, J., & Mullins, L. (2018). Grit, Illness-related Distress, and psychosocial outcomes in college students with a chronic medical condition: A path analysis. *Journal of Pediatric Psychology, 43*(5), 552-560. <https://doi:10.1093/jpepsy/jsx145>.
- Singh, K., & Jha, S. (2008). Positive and negative affect, and grit as predictors of happiness and life satisfaction. *Journal of the Indian Academy of Applied Psychology, 34*(2), 40–45. <https://doi:10.1037/t06070-000>.
- Sirois, F. (2003). Manual for the Control Beliefs Inventory-CBI.
<file:///C:/Users/Katie/Downloads/CBI%20manual%202019%20-%20English.pdf>
- Sirois, F., & Hirsch, J. (2013). Associations of psychological thriving with coping efficacy, expectations for future growth, and depressive symptoms over time in people with arthritis. *Journal of Psychosomatic Research, 75*(3), 279–286. <https://doi.org/10.1016/j.jpsychores.2013.06.004>.
- Sirois, F., & Hirsch, J. (2017). A longitudinal study of the profiles of psychological thriving, resilience, and loss in people with inflammatory bowel disease. *British Journal of Health Psychology, 22*(4), 920-939. <https://doi:10.1111/bjhp.12262>.
- Skrastins, O., & Fletcher, P. (2016) "One flare at a time": adaptive and maladaptive behaviours of women coping with inflammatory bowel disease and irritable bowel syndrome. *Clinical Nurse Specialist, 33*(1), 43-51. <https://doi:10.1097/nur.0000000000000345>.
- Stanton, A., Revenson, T., & Tennen, H. (2007). Health Psychology: psychosocial, adjustment to chronic disease. *Annual Review Psychology, 58*, 565-92. <https://doi:10.1146/annurev.psych.58.110405.085615>.
- Stevens, J. (2002). *Applied multivariate statistics for the social sciences, 4th Edn*. Erlbaum.

Factors that associate with resilience in people with IBD

Stewart, D., & Yuen, T. (2011). A systematic review of resilience in the physically ill.

Psychosomatics, 52(3), 199-209. <https://doi:10.1016/j.psym.2011.01.036>.

Stoffel, J., & Cain, J. (2018). Review of grit and resilience literature within health professions education. *American Journal of Pharm Education*, 82(2), 6150.

<https://doi:10.5688/ajpe6150>.

Strom, J., & Egede, L. (2012). The impact of social support on outcomes in adult patients with type 2 diabetes: A systematic review. *Current Diabetes Report*, 12(6).

<https://doi:10.1007/s11892-012-0317-0>.

Tabachnik, B., & Fidell, L. (2014) *Using multivariate statistics*, (6th Ed), Pearson.

Timmer, A., Preiss, J., Motschall, E., Rucker, G., Jantschek, G., & Moser, G. (2011).

Psychological interventions for treatment of inflammatory bowel disease (Review). *The Cochrane Library*, 8 (2). <https://doi:10.1002/14651858.CD006913.pub2>.

Todorovic, V. (2012). Providing holistic support for patients with inflammatory bowel disease.

British Journal of Community Nursing, 17(10), 466-472.

<https://doi:10.12968/bjcn.2012.17.10.466>.

Traino, K., Bakula, D., Sharkey, C., Roberts, C., Ruppe, N., Chaney, J., & Mullins, L. (2019).

The Role of Grit in Health Care Management Skills and Health-related Quality of Life in College Students with Chronic Medical Conditions. *Journal of Pediatric Nursing*, 46,

72-77. <https://doi.org/10.1016/j.pedn.2019.02.035>.

Factors that associate with resilience in people with IBD

Tribbick, D., Salzberg, M., Connell, W., Macrae, F., Kamm, M., Bates, G., Cunningham, G., Austin, D., & Knowles, S. (2017). Differences Across Illness Perceptions in Inflammatory Bowel Disease and Their Relationships to Psychological Distress and Quality of Life. *Gastroenterology Nursing, 40*(4), 291-299.
<https://doi:10.1097/SGA.0000000000000225>.

Vainio, M., & Daukantaitė, D. (2016). Grit and different aspects of well-being: Direct and indirect relationships via sense of coherence and authenticity. *Journal of Happiness Studies, 17*(9), 2119–2147. <https://doi:10.1007/s10902-015-9688-7>.

Vander-Have, M., Van der Aalst, K., Kaptein, A., Leenders, M., Siersema, P., Oldenburg, B., & Fidder, H. (2014). Determinants of health-related quality of life in Crohn's Disease: A systematic review and meta-analysis. *Journal of Crohn's and Colitis, 8*(2), 93-106.
<https://doi:10.1016/j.crohns.2013.04.007>.

Voth, J., & Sirois, F. (2009). The role of self-blame and responsibility in adjustment to inflammatory bowel disease. *Rehabilitation Psychology, 54*(1), 99-108.
<https://dx.doi.org/10.1037/a0014739>.

Walker, J., Ediger, J., Graff, L., Greenfeld, J., Clara, L., Liz, L., Rawsthorne, P., Miller, N., Rogala, L., McPhail, C., & Bernstein, C. (2008). The Manitoba IBD cohort study: a population-based study of the prevalence of lifetime and 12-month anxiety and mood disorders. *American Journal of Gastroenterology, 103*(8), 1989-97.
<https://doi:10.1111/j.1572-0241.2008.01980.x>.

Factors that associate with resilience in people with IBD

- Wallston, K. (1992). Hocus-pocus, the focus isn't strictly on locus: Rotter's social learning theory modified for health. *Cognitive Therapy and Research*, 16(2), 183-199.
<https://nursing.vanderbilt.edu/projects/wallstonk/pdf/A55.pdf>.
- Wallston, K., Wallston, B., Smith, S., & Dobbins, C. (1987). Perceived control and health. *Current Psychological Research and Review*, 6, 5-25.
<https://link.springer.com/article/10.1007/BF02686633>.
- Wang, C., Qu, H., & Xu, H. (2015). Relationship between social support and self-efficacy in women psychiatrists. *Chinese Nursing Research*, 2(4), 103-106.
<https://doi.org/10.1016/j.cnre.2015.10.002>.
- Watson, J. (1913). Psychology as the behaviourist views it. *Psychological Review*, 20, 158-177.
<http://dx.doi.org/10.1037/h0074428>.
- Werner, E. (1994). Overcoming the odds. *Journal of Developmental Behavioural Paediatrics*, 15(2), 131-136.
https://journals.lww.com/jrnldbpcitation/1994/04000/overcoming_the_odds.12.aspx.
- Willig, C. (2008). *Introducing Qualitative research in Psychology (2nd Ed)*. Open University Press.
- Windle, G. (2010). What is resilience? A review and concept analysis. *Reviews in Clinical Gerontology*, 21(2), 152-169. <https://doi:10.1017/S0959259810000420>.
- Winnicott, D. (1960). The Theory of the Parent-Infant Relationship. *International Journal of Psychoanalysis*, 41, 585-595. <http://icpla.edu/wp-content/uploads/2012/10/Winnicott-D.-The-Theory-of-the-Parent-Infant-Relationship-IJPA-Vol.-41-pps.-585-595.pdf>.

Factors that associate with resilience in people with IBD

Zhang, M., Hong, L., Zhang, T., Lin, Y., Zheng, S., Zhou, X., Fan, R., Wang, Z., Zhang, C., & Zhong, J. (2016). Illness perceptions and stress: Mediators between disease severity and psychological well-being and quality of life among patients with Crohn's disease. *Patient Preference and Adherence*, *10*, 2387-2396. <https://doi:10.2147/PPA.S118413>.

Appendix A

Participant survey comments

*Participants were asked to consider practical aspects of the preliminary online survey (e.g. length of survey and ease of completion) and to identify if any potential emotional issues might arise.

It took them approximately 15 minutes to complete the survey

The feedback was largely positive about the length and utility of the survey; however, they gave their views on the order of the measures within the survey.

The length felt “okay” but advised to consider order of scales so that it alternates short and long scales to keep effort up and willingness to complete until the end;

Some items felt repetitive.

The layout is important, to have the Likert scale visible where each item is;

Comments regarding the wording of the resilience scale (this was because the preliminary scale contained the version of the CD-RISC that was online prior to purchasing). Once purchased the full accurate wording of the scale was amended.

Appendix B

Ethics approval and research governance sponsor letter



Downloaded: 25/04/2019
Approved: 08/04/2019

Katie Boden
Registration number: 170149396
Psychology
Programme: Doctorate Clinical Psychology

Dear Katie

PROJECT TITLE: An exploration of the factors that associate with resilience in individuals with IBD: A mixed methods study
APPLICATION: Reference Number 023134

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 08/04/2019 the above-named project was **approved** on ethics grounds, on the basis that you will adhere to the following documentation that you submitted for ethics review:

- University research ethics application form 023134 (dated 05/04/2019).
- Participant information sheet 1055099 version 2 (26/03/2019).
- Participant information sheet 1055100 version 2 (26/03/2019).
- Participant consent form 1055101 version 2 (26/03/2019).

The following optional amendments were suggested:

Amend the Head of Psychology Department to Glenn Waller and be consistent about the DClin Psy. In addition, Crohn's Disease is sometimes written without the apostrophe in the information and consent form--please amend.

If during the course of the project you need to [deviate significantly from the above-approved documentation](#) please inform me since written approval will be required.

Yours sincerely

Jilly Gibson-Miller
Ethics Administrator
Psychology

Factors that associate with resilience in people with IBD



The
University
Of
Sheffield.

Department Of Psychology. Clinical Psychology Unit.

Doctor of Clinical Psychology (DClin Psy) Programme
Clinical supervision training and NHS research training
& consultancy.

**Clinical Psychology Unit
Department of Psychology
University of Sheffield
Floor F, Cathedral Court
1 Vicar Lane
Sheffield
S1 2LT**

Dr A R Thompson, Clinical Training Research Director
Please address any correspondence to Amrit Sinha
Research Support Officer
Telephone: 0114 2226650
Email: a.sinha@sheffield.ac.uk

24st September 2018

To: Research Governance Office

Dear Sir/Madam,

RE: Confirmation of Scientific Approval and indemnity of enclosed Research Project

Project title: An exploration of the factors that associate with resilience in individuals with IBD: A mixed methods study

Investigators: Katie Boden (DClin Psy Trainee, University of Sheffield); Georgina Rowse; Fuschia Sirois; Rebecca Yeates Academic Supervisors (University of Sheffield)

I write to confirm that the enclosed proposal forms part of the educational requirements for the Doctoral Clinical Psychology Qualification (DClin Psy) run by the Clinical Psychology Unit, University of Sheffield.

Three independent scientific reviewers usually drawn from academic staff within the Psychology Department have reviewed the proposal. Review includes appraisal of the proposed statistical analysis conducted by a statistical expert based in the School of Health and Related Research (SCHARR). Where appropriate an expert in qualitative methods is also appointed to review proposals.

I can confirm that approval of a proposal is dependent upon all necessary amendments having been made to the satisfaction of the reviewers and I can confirm that in this case the reviewers are content that the above study is of sound scientific quality. Consequently, the University will if necessary indemnify the study and act as sponsor.

Given the above, I would remind you that the Department already has an agreement with your office to exempt this proposal from further scientific review. However, if you require any further information, please do not hesitate to contact me.

Yours sincerely

A handwritten signature in blue ink, appearing to be 'A. Thompson', written over a light blue grid background.

Dr. Andrew Thompson
Director of Research Training

Cc.: Katie Boden; Georgina Rowse; Fuschia Sirois; Rebecca Yeates.

Appendix C

Information sheets (focus group and interviews)

Information sheet

An exploration of factors associated with resilience in those with IBD: A focus group



Department of Psychology.

Clinical Psychology Unit

Doctor of Clinical Psychology (DClin Psy) programme

Telephone: 0114 222 6574

Email: kboden@sheffield.ac.uk

You are invited....

To participate in a research study conducted at the University of Sheffield. Before you decide to take part it is important that you understand the research that is being done and what it will involve, so that you can give informed consent. Please take the time to read the following information carefully and discuss it with others if you wish. You are welcome to ask any questions to Katie Boden (lead researcher) if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

*If you are interested in participating, please ensure you could be available on **Friday 31st May or Friday 7th June 2019**. The focus group will last approximately 1 hour to 1 and a half hours and a specific time will be agreed when enough participants have expressed an interest.

The project and aims

The overall aim of the research is to explore the factors that influence resilience in individuals with IBD. The first element (which you are being invited to participate in) is a group interview to discuss what factors are felt to be important in influencing resilience from those with lived experience of IBD. This will contribute to the development of the research question and second part of the study.

Why have I been invited to participate in the study?

You have been invited to participate because you have been given a medical diagnosis of IBD (either Crohn's Disease or Ulcerative Colitis). Approximately seven other people will also be recruited to take part in the group interview for this study.

Factors that associate with resilience in people with IBD

Do I have to take part?

It is up to you to decide whether or not you would like to take part and you are under no obligation to participate. If you decide to take part you will be given this information sheet to read and keep and you can withdraw from the study at any time during the focus group and up until 4 weeks after. You do not need to give a reason.

What will happen once I agree to participate in the study?

You will be asked to attend a group interview at a time that is convenient. You will be asked questions about what you think the important factors are that influence resilience in living with IBD. This will take approximately 60 minutes and it is your choice whether you feel comfortable answering any of the questions. The group interview will be audio-recorded and the files transferred to a password protected computer that is secure and regularly backed up. Data will be destroyed on successful publication of the research.

Will my taking part in this project be kept confidential?

All the information that we collect about you during the course of the research will be kept strictly confidential and will only be accessible to members of the research team. An alternative name (a pseudonym) will be assigned to you on completion of the group interview (you can choose this yourself if you wish) and so you will not be identifiable in any reports or publications.

What are the benefits of taking part?

Whilst there are no immediate benefits to participating in the project, it is hoped that this work will add to our knowledge base around the factors that influence resilience so that it can inform the help individuals receive around managing their condition. It is also hoped that it will stimulate further research in this area.

What are the possible disadvantages and risks of taking part?

Some of the questions will likely prompt thought around how you have managed your condition, which may make you feel uncomfortable in some way. If you feel distressed at any stage, you can leave the interview or withdraw from the project completely.

What if something goes wrong or if I become distressed as a result of taking part in the study?

If after participating, you decide that you would like to withdraw your data from the study please email kboden@sheffield.ac.uk, quoting the pseudonym allocated to you at the end of the study. You do not need to provide a reason for withdrawing from the study.

If you feel distressed after participating, you can contact your GP or a non-statutory organisation such as Crohn's and Colitis UK.

If, after participating in the study, you wish to raise a complaint, you can do this by contacting Dr Glenn Waller (Head of Psychology Department) on 0114 222 6571 or by email on g.waller@sheffield.ac.uk.

What type of information will be sought from me and why is the collection of this information relevant for achieving the research project's objectives?

Information about your age, gender, ethnicity and your diagnosis or IBD will be collated because this will help the researcher see whether these factors impact on levels of resilience

Factors that associate with resilience in people with IBD

What will happen to the results of the research project?

The study results will be written up and submitted as a doctoral thesis. You will not be identifiable in any report or publication of these results. The results of this study may be published in a peer-reviewed scientific journal in the future.

Will I receive any reimbursement of expenses for taking part in this research?

You will receive reimbursement on any travel costs you have incurred so please keep any receipts for public transport travel. You will also be given a £10 gift voucher as a gesture of thanks for your participation.

Who is organising and funding the research?

The University of Sheffield is organising and funding this research

Who has ethically reviewed the project?

The ethics of this research has been reviewed and approved by The University of Sheffield's Research Ethics Committee.

Who is the data controller?

The University of Sheffield will act as the Data Controller for this study. This means that the University is responsible for looking after your information and using it properly.

What is the legal basis for processing my personal data?

According to data protection legislation, we are required to inform you that the legal basis we are applying in order to process your personal data is that 'processing is necessary for the performance of a task carried out in the public interest' (Article 6(1)(e)). Further information can be found in the University's Privacy Notice

<https://www.sheffield.ac.uk/govern/data-protection/privacy/general>

Contact for further information

If you would like any further information you can contact Katie Boden (lead researcher) by email kboden1@Sheffield.ac.uk

Thank-you for taking part in the project.

Information sheet

An exploration of factors associated with resilience in those with IBD: An interview



Department of Psychology.

Clinical Psychology Unit

Doctor of Clinical Psychology (DClin Psy) programme

Telephone: 0114 222 6574

Email: kboden1@sheffield.ac.uk

You are invited....

To participate in a research study conducted at the University of Sheffield. Before you decide to take part it is important that you understand the research that is being done and what it will involve, so that you can give informed consent. Please take the time to read the following information carefully and discuss it with others if you wish. You are welcome to ask any questions to Katie Boden (lead researcher) if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

*The date and time of the interview will be negotiated between you and the researcher. This can take place face to face or via telephone or skype. It is intended to last approximately 1 hour.

The project and aims

The overall aim of the research is to explore the factors that influence resilience in individuals with IBD. The first element (which you are being invited to participate in) is an interview to discuss what factors are felt to be important in influencing resilience from those with lived experience of IBD. This will contribute to the development of the research question and second part of the study.

Why have I been invited to participate in the study?

You have been invited to participate because you have been given a medical diagnosis of IBD (either Crohn's Disease or Ulcerative Colitis).

Do I have to take part?

It is up to you to decide whether or not you would like to take part and you are under no obligation to participate. If you decide to take part you will be given this information sheet to read and keep and you can withdraw from the study at any time during the interview and up until 4 weeks after. You do not need to give a reason.

What will happen once I agree to participate in the study?

Factors that associate with resilience in people with IBD

You will be asked to attend an interview at a time that is convenient. You will be asked questions about what you think the important factors are that influence resilience in living with IBD. This will take approximately 45 to 60 minutes and it is your choice whether you feel comfortable answering any of the questions. The interview will be audio-recorded and the files transferred to a password protected computer that is secure and regularly backed up. Data will be destroyed on successful publication of the research.

Will my taking part in this project be kept confidential?

All the information that we collect about you during the course of the research will be kept strictly confidential and will only be accessible to members of the research team. An alternative name (a pseudonym) will be assigned to you on completion of the interview (you can choose this yourself if you wish) and so you will not be identifiable in any reports or publications.

What are the benefits of taking part?

Whilst there are no immediate benefits to participating in the project, it is hoped that this work will add to our knowledge base around the factors that influence resilience so that it can inform the help individuals receive around managing their condition. It is also hoped that it will stimulate further research in this area.

What are the possible disadvantages and risks of taking part?

Some of the questions will likely prompt thought around how you have managed your condition, which may make you feel uncomfortable in some way. If you feel distressed at any stage, you can terminate the interview or withdraw from the project completely.

What if something goes wrong or if I become distressed as a result of taking part in the study?

If after participating, you decide that you would like to withdraw your data from the study please email kboden1@sheffield.ac.uk, quoting the pseudonym allocated to you at the end of the study. You do not need to provide a reason for withdrawing from the study.

If you feel distressed after participating, you can contact your GP or a non-statutory organisation such as Crohn's and Colitis UK.

If, after participating in the study, you wish to raise a complaint, you can do this by contacting Dr Glenn Waller (Head of Psychology Department) on 0114 222 6571 or by email on g.waller@sheffield.ac.uk.

What type of information will be sought from me and why is the collection of this information relevant for achieving the research project's objectives?

Information about your age, gender, ethnicity and your diagnosis or IBD will be collated because this will help the researcher see whether these factors impact on levels of resilience

What will happen to the results of the research project?

The study results will be written up and submitted as a doctoral thesis. You will not be identifiable in any report or publication of these results. The results of this study may be published in a peer-reviewed scientific journal in the future.

Will I receive any reimbursement of expenses for taking part in this research?

Factors that associate with resilience in people with IBD

You will receive reimbursement on any travel costs you have incurred (if relevant) so please keep any receipts for public transport travel. You will also be given a £10 gift voucher as a gesture of thanks for your participation.

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The University of Sheffield is organising and funding this research.

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The ethics of this research has been reviewed and approved by The University of Sheffield's Research Ethics Committee.

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What is the legal basis for processing my personal data?

According to data protection legislation, we are required to inform you that the legal basis we are applying in order to process your personal data is that 'processing is necessary for the performance of a task carried out in the public interest' (Article 6(1)(e)). Further information can be found in the University's Privacy Notice

<https://www.sheffield.ac.uk/govern/data-protection/privacy/general>

Contact for further information

If you would like any further information you can contact Katie Boden (lead researcher) by email kboden1@Sheffield.ac.uk

Thank-you for taking part in the project.

Appendix D

Consent form (focus group)

Consent Form

Exploring resilience factors in individuals with Inflammatory Bowel Disease

<i>Please tick the appropriate boxes</i>	Yes	No
Taking Part in the Project		
I have read and understood the project information sheet or the project has been fully explained to me. (If you will answer No to this question please do not proceed with this consent form until you are fully aware of what your participation in the project will mean.)	<input type="checkbox"/>	<input type="checkbox"/>
I have been given the opportunity to ask questions about the project.	<input type="checkbox"/>	<input type="checkbox"/>
I agree to take part in the project. I understand that taking part in the project will include participating in a focus-group/interview and that this will be audio-recorded. I understand that audio data will be destroyed once the research has been successfully published.	<input type="checkbox"/>	<input type="checkbox"/>
I understand that my taking part is voluntary and that I can withdraw from the study up until 4 weeks after the focus group. I do not have to give any reasons for why I no longer want to take part and there will be no adverse consequences if I choose to withdraw. I understand that if I do not wish to answer any particular questions, I am free to decline. I am aware I can contact the researcher Katie Boden on kboden1@sheffield.ac.uk to discuss this further if I wish.	<input type="checkbox"/>	<input type="checkbox"/>
How my information will be used during and after the project		
I understand my personal details such as name, phone number, address and email address etc. will not be revealed to people outside the project.	<input type="checkbox"/>	<input type="checkbox"/>
I understand and agree that my words may be quoted in publications, reports, web pages, and other research outputs. I understand that I will not be named in these outputs unless I specifically request this.	<input type="checkbox"/>	<input type="checkbox"/>
I understand and agree that other authorised researchers will have access to this data only if they agree to preserve the confidentiality of the information as requested in this form.	<input type="checkbox"/>	<input type="checkbox"/>
I understand and agree that other authorised researchers may use my data in publications, reports, web pages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form.	<input type="checkbox"/>	<input type="checkbox"/>
I give permission for the focus-group data that I provide to be given to the Research Support Officer at the Clinical Psychology Unit University of Sheffield so it can be used for future research and learning.	<input type="checkbox"/>	<input type="checkbox"/>
So that the information you provide can be used legally by the researchers		
I agree to assign the copyright I hold in any materials generated as part of this project to The University of Sheffield.	<input type="checkbox"/>	<input type="checkbox"/>

Name of participant [printed]

Signature

Date

Factors that associate with resilience in people with IBD

Name of Researcher [printed]

Signature

Date

Appendix E

Interview schedule used for focus group and individual interviews

Core question	Prompts
Ask everyone's name (focus group), how long you have lived with IBD for and (if comfortable to answer) which subtype it is?	N/A
Do you feel this definition of resilience is relevant to your experience of your condition?	Definition given to participants "resilience as the ability to bounce back from adversity or challenges" Clarify many definitions of resilience but for the purpose of the research this one has been chosen. Clarify definition if required
Are there any disease-specific factors that you think have influenced your resilience?	Prompts: Does anything come to mind? e.g. Has the time since diagnosis influenced resilience? Has disease subtype (CD, UC) influenced resilience? Has the level of disease-activity influenced resilience?
Are there any psychological factors that you feel have affected your resilience levels?	Prompts: Explain psychological processes include what is in your mind and may include coping strategies. Does anything come to mind? Do thought processes influence or relate to your resilience? Do your beliefs influence or relate to your resilience? Do your feelings influence or relate to your resilience?

Factors that associate with resilience in people with IBD

Do your views on your condition influence or relate to your resilience?

Core question	Prompts
<p>What areas of your life do you feel your condition has impacted on?</p>	<p>Prompts: Does anything come to mind? Does it impact on your employment? Does it impact on your intimate relationships? Does it impact on your social life?</p>
<p>Do you feel as though your ability to be resilient has changed since you started experiencing IBD symptoms?</p>	<p>Prompt: How might coping with challenges have changed before or after your IBD diagnosis? How have your coping styles or resilience changed over course of the condition?</p>
<p>What are your views on the important factors that enable you to manage your condition?</p>	<p>Prompt: Are there obstacles to managing your condition? Do you think if those factors were taken away it would affect your ability to be resilient?</p>
<p>Have there been times during your condition that you have felt more resilient than others?</p>	<p>Prompt: Can you recall a time when you felt your resilience was low? Can you recall a time when you felt your resilience was high?</p>
<p>What are your main sources of support?</p>	<p>Prompt: Is this support the most important factor to being resilient or are other factors more important?</p>

Appendix F

Braun and Clarke (2006) thematic analysis steps

1. Familiarisation of data.

The main researcher undertook the role of interviewer, transcriber and data-analyst which facilitated immersion in the data and familiarisation. The transcript was checked against the recordings for accuracy. Time was taken to repeatedly read the transcripts and elements of the data that were deemed meaningful were highlighted.

2. Generating initial codes

Line-by-line coding was conducted by electronic notetaking (see line by line coding example below). The researcher was mindful of the surrounding text so that context was not lost. Braun and Clarke (2006) recommend coding multiple times in order to uncover potential multiple meanings, thus the transcripts and codes were read repeatedly, and additional notes made that were based on semantic meaning. The research question and aims were kept in mind throughout. Attention was paid to word choices, repetitions, how experiences were described and use of metaphor. The data-analyst was mindful to give equal attention to each line. Further, all relevant extracts for each theme were collated and either utilised as illustrative quotes in the final report or as part of a separate document (see Appendix G). Some codes considered less relevant to the research question and aims were excluded (e.g. discussion about medication use).

Factors that associate with resilience in people with IBD

Line-by-line coding examples:

Data extracts from transcripts	Coding (descriptive and semantic)
<p>Sally: Other things that happen in life, if you can get through how bad having IBD is that actually you can cope with a lot more and I think it becomes a bit of a reference that you might say, I'm in real pain, oh but it's not as bad as the pain I might feel, if I had a flare up</p>	<p>Comparing IBD challenges to other adverse events desensitisation</p>
<p>Harriet: yeh I agree with that totally and on my shorter term scale when you are having the flare ups, erm, kind of, I've got to a point now where I can't stop this kind of letting me do everything that I still want to do, if I'm having a flare up its not going to stop me from going out there, doing my socialising, having loads of plans that I've already made, so it's kind of learning how to kind of function around the IBD diagnosis and erm still, kind of, because when I was diagnosed I was only 15 and my friends wouldn't have really understood, like what was going on, erm , so it was kind of like learning to just carry on and staying as I was at that age to like, maintain like, my social life</p>	<p>Sharing experience with peer, Making note of disease activity and effects of time on mindset Determination to function as well as desires Meeting goals, enduring despite symptoms, perseverance- grit Social goals important - grit Adaptation to maintain functioning Age, young friends not understanding-invisible struggle Act normal, brave face to stay connected to friends-stoicism</p>
<p>Researcher: that sounds quite lonely Harriet: yeh its hard, yeh, yeh I just didn't really, well it's not something I'd ever like come across, erm</p>	<p>Emotionally demanding to keep functioning despite IBD challenges and others not understanding New experience for her</p>
<p>Sally: cos you were very young, that's a.. Harriet: yeh, teenage years, I say luckily, that's probably the wrong word, but my brother had it or was diagnosed it's a couple of year before so me and the family sort of understood it so it wasn't as lonely as it could have been but more in terms of my friends being at school and my social life it was kind of trying to maintain where I was at in that part of my life whilst still having the diagnosis, er, yeh</p>	<p>Suggestion of age as an influencing factor Family/other with lived experience understanding helping combat aloneness, aiding resilience Brave face, endure symptoms to maintain social identity Concealing illness</p>

Factors that associate with resilience in people with IBD

<p>Researcher: Yeh, and it sounds like for you (looking at Sally) if I can deal with that then I can deal with other stuff that comes along, like it sounds like a really positive mind frame to have</p> <p>Sally: yeh I think it's really easy to have a positive mind frame when you're in remission</p> <p>Harriet: yes definitely (laughs)</p> <p>Sally: and it's hard when you have a relapse, I think that's the, but I mean I would agree in terms of you adapt your life accordingly and [pause] but definitely and in some ways if I'm honest it's made me less tolerant of other people at times probably brought out a less tolerant side of me because erm [pause] because the excuses that other people might come up with, and you think really? actually that doesn't register on the, erm, on the list of things that might disrupt your life</p>	<p>Disease activity as affecting resilience and mind frame</p> <p>In agreement regarding disease activity</p> <p>Adapting to functioning around disease activity</p> <p>Intolerance of others, dismissiveness of others struggles</p> <p>Questioning legitimacy of others difficulties</p> <p>Comparison of IBD challenges to others, disruption to functioning</p>
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<p>Claire: erm yeh in a way I think that people still don't understand it and people don't appreciate quite how bad it is so it actually I think helps your resilience cos you feel like you can't show it so you almost have to go the other way that you've gotta put the brave face on for other people but then it becomes normal for you because you're acting it, it becomes normal, erm but because people don't quite understand it, or it feels, I think like with my close friends, good friends they get it, but you do still feel a bit of stigma for it, like I don't want to be labelled as the person who's poorly cos of this, how people treat you for it or don't treat you for it cos you don't let them in that much, it's just like when you're tired and people are like why you tired you know like just family say you shouldn't be tired you're young, it's things like that and they don't really know what they're saying and they obviously they</p>	<p>Others not understanding the struggle</p> <p>Struggle invisible</p> <p>Others diminishing the extent of the challenges</p> <p>Helping resilience as its invisible, a pretence to self and others, stoicism</p> <p>Not showing emotions, "brave face" for others- stoicism</p> <p>"brave face" transcending to her</p> <p>Close social circle understanding but felt stigma</p> <p>Fear of being labelled, identity as poorly person</p> <p>Fear of being different, being treated different</p> <p>Avoiding intimacy, dismissing others</p>
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Factors that associate with resilience in people with IBD

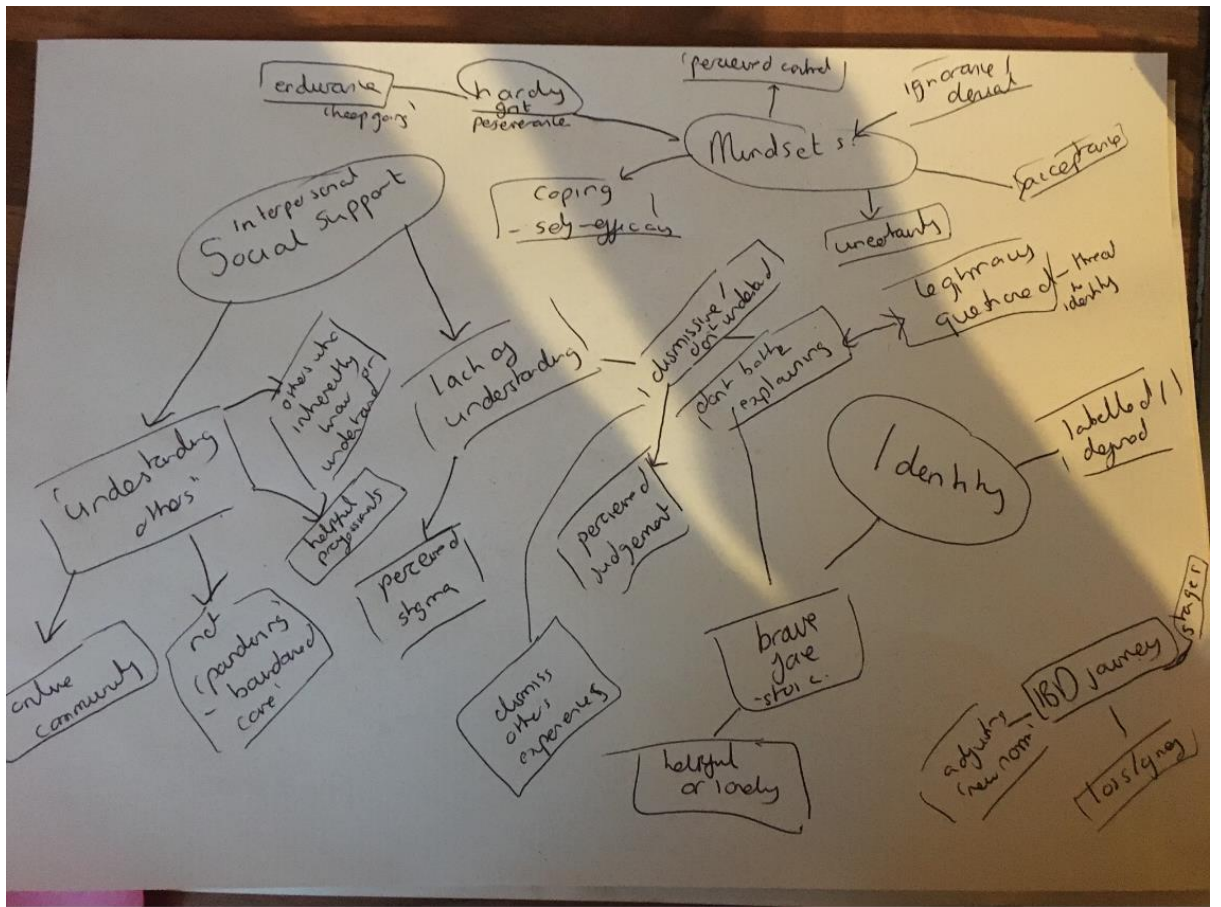
<p>know what I've got but they just haven't thought about what they're saying to you so when someone says something like that to you and you're like [pause]</p>	<p>Others as dismissive and not understanding</p> <p>Dismissive comments, lack of "knowing" from others, insensitivity</p>
<p>Researcher: yeh what's it like hearing comments like that Claire: Well it's not very nice but I don't sort of respond to them cos I just think, especially with older family members it's not gonna make any difference, they won't get it</p>	<p>Unpleasant feeling being misunderstood</p> <p>Choosing not to clarify and explain condition, withholding due to expectation others won't understand</p>

3. Searching for themes

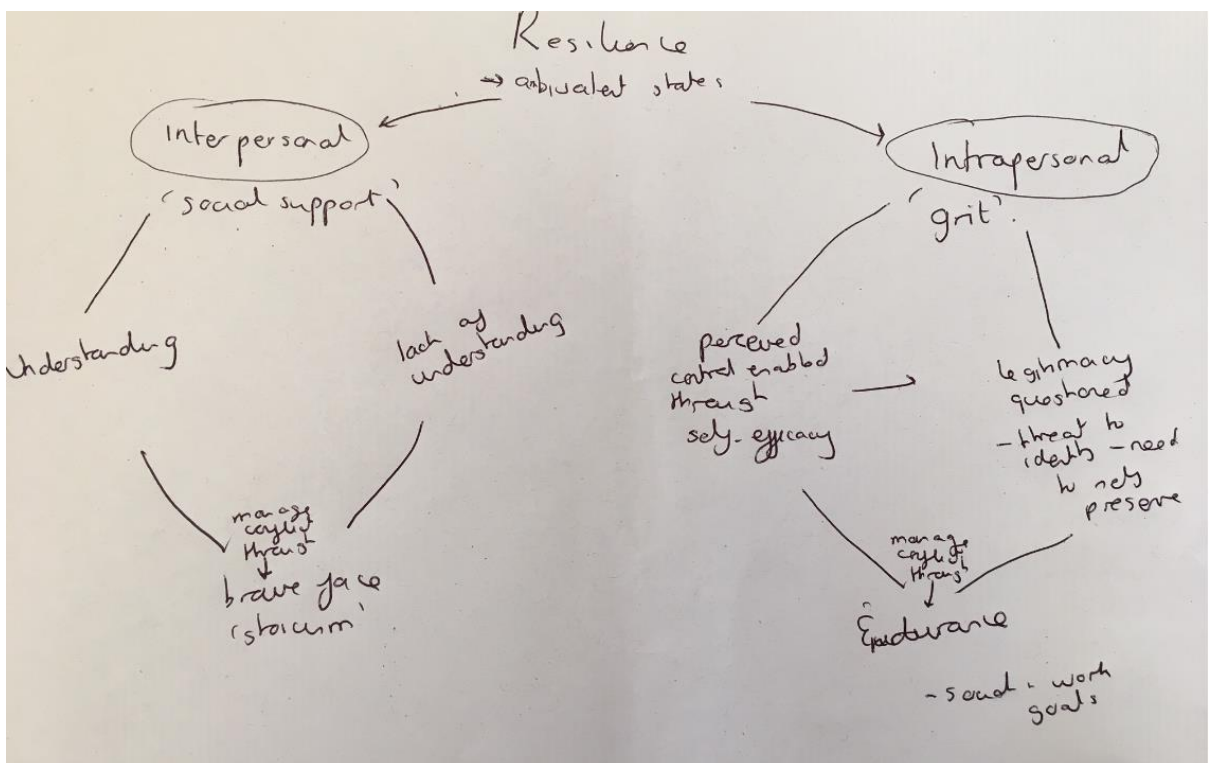
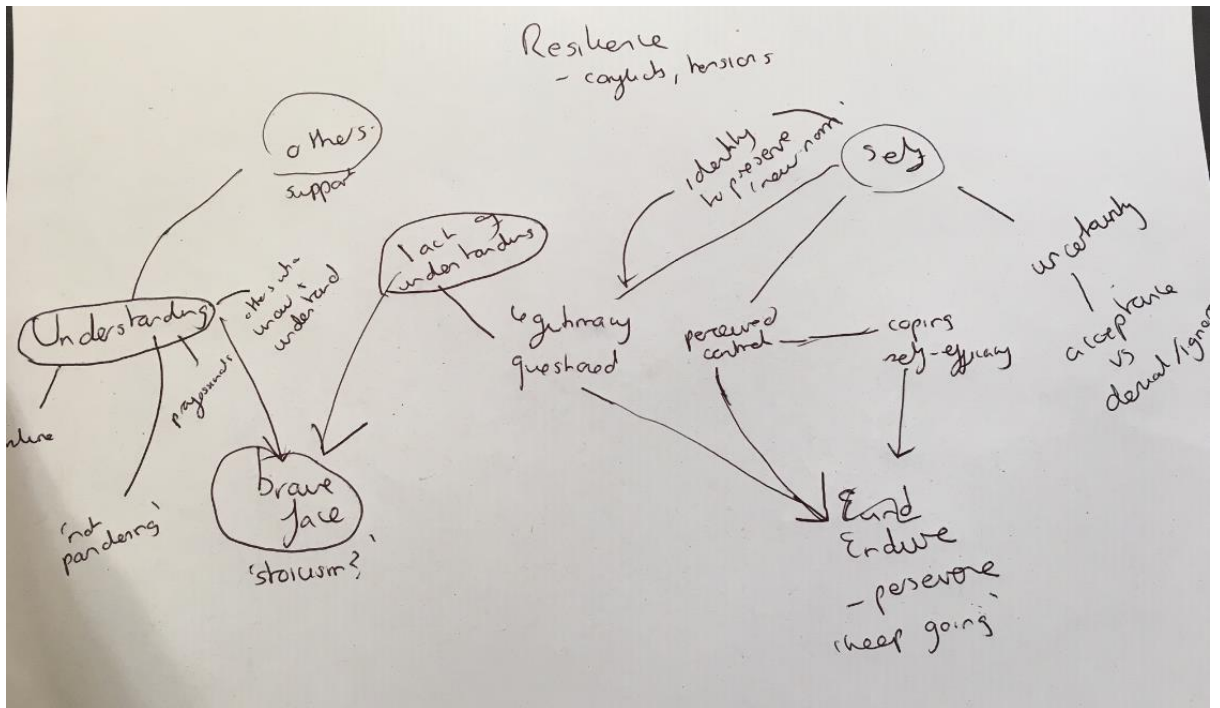
The codes were reviewed for comparisons, similarities, and contrasts. Similar codes enabled identification of broader themes and subthemes and diagrams/maps were constructed to organise the themes. The conceptual maps evolved (below are three examples of maps constructed in chronological order). The research team also read the transcripts and themes/constructs that were apparent within the data were discussed in research supervision.

Conceptual maps examples:

Factors that associate with resilience in people with IBD



Factors that associate with resilience in people with IBD



4. Reviewing themes

Research supervision was used to review the thematic maps and alternative possibilities and perspectives on the data were discussed. Diagrams and supporting quotes

facilitated decision-making and repeatedly reading transcripts ensured that themes reflected the dataset as a whole and represented all participants discourse.

5. Defining and naming themes

Braun and Clarke (2006) recommend describing the central organising features of themes in a succinct and coherent manner, accompanied by quotes to illustrate themes. Research supervision was utilised to discuss the various constructs (and their accompanying definitions) that may be present in the analysis and this aided clarity over themes.

6. Producing the report

The report aimed to provide a clear, concise narrative of the themes and interpretation of the data, giving relatively equal attention to each theme. Braun and Clarke's (2006) checklist of criteria for undertaking a good quality TA was followed.

Process	No.	Criteria
Transcription	1	The data have been transcribed to an appropriate level of detail, and the transcripts have been checked against the tapes for 'accuracy'.
Coding	2	Each data item has been given equal attention in the coding process.
	3	Themes have not been generated from a few vivid examples (an anecdotal approach), but instead the coding process has been thorough, inclusive and comprehensive.
Analysis	4	All relevant extracts for all each theme have been collated.
	5	Themes have been checked against each other and back to the original data set.
	6	Themes are internally coherent, consistent, and distinctive.
	7	Data have been analysed – interpreted, made sense of – rather than just paraphrased or described.
	8	Analysis and data match each other – the extracts illustrate the analytic claims.
Overall	9	Analysis tells a convincing and well-organized story about the data and topic.
	10	A good balance between analytic narrative and illustrative extracts is provided.
	11	Enough time has been allocated to complete all phases of the analysis adequately, without rushing a phase or giving it a once-over-lightly.
Written report	12	The assumptions about, and specific approach to, thematic analysis are clearly explicated.
	13	There is a good fit between what you claim you do, and what you show you have done – ie, described method and reported analysis are consistent.
	14	The language and concepts used in the report are consistent with the epistemological position of the analysis.
	15	The researcher is positioned as <i>active</i> in the research process; themes do not just 'emerge'.

Appendix G

Illustrative quotes for each theme (pseudonyms used)

Superordinate theme: Grit

Subordinate theme: Reclaiming control through autonomous coping

“If there’s a problem, I’m proactive and other people have sort of said that to me, like I admire so much how you’re dealing with it and then how I talk about it so there is a psychological satisfaction to be had from that knowing I’m not gonna let it beat me.”

(Annie)

“You adapt your life accordingly.” (Sally)

“For me it’s about understanding myself, like I had to be very aware of what I was eating, what I was drinking, making sure that I understood my condition, I couldn’t expect other people to understand if I didn’t know what’s going on.” (Harriet)

“Quite a positive person and I’m very erm I don’t just let things just happen to me I will go and sort things out.” (Annie)

“I’d rather deal with it myself erm unless its, I’ve not felt like I really needed to go.” (Claire) (referring to consideration of professional help)

“I’ll try and eat food that’s a bit more carbs, potato based bread cos its thicker and it helps, its heavy and helps everything come out, stay away from veg or anything high fibre, I’ll stay away from things that I know are gonna bother it if it’s really inflamed in there.” (Claire)

Factors that associate with resilience in people with IBD

“And I think cos I feel like I handle it quite well I don’t want someone to panda to me, I don’t want to be pandered too.” (Claire)

“You feel like you literally have no control, you have no control when you want to go to the toilet, you have no control over whether your tummy’s going to hurt today and how that’ll feel you literally it feels like you lost control of anything.” (Sarah) (before feeling in control and developing self-efficacy)

“I think a lot of the time it’s our reaction to stuff we have the control over how we react to something, we don’t have the control over what’s happening to us, we just have to decide how we’re going to deal with that.” (Sarah) (self-efficacy of emotions)

“Kind of recognise what symptoms are telling you that a flare is coming and stuff so to me I think it’s always essential to have a plan,” (Sarah)

Subordinate theme: Legitimacy under question, maintaining identity through self-preservation:

“Like straight away was really helpful cos I couldn’t believe that someone actually believed me straight.” (Claire) (point of diagnosis)

“Like I’ll go on course and stuff in London and I just keep it to myself cos then you don’t have to explain it to anybody.” (Claire)

“People are like why you tired you know like just family say you shouldn’t be tired you’re young, it’s things like that and they don’t really know what they’re saying and they obviously they know what I’ve got but they just haven’t thought about what they’re saying.” (Claire)

Factors that associate with resilience in people with IBD

“I threw the book across the room and I thought I’m going to deal with it how I want to deal with it.” (Annie)

“The frustration from it being an invisible illness where erm you know that some people are questioning whether it’s as bad as you’re making out or erm then with us you’re paranoid if they might be but you’re not sure kind of thing.” (Annie)

“You either lie to them and tell them you’re alright or you can actually tell them how you are and you can literally see their eyes glaze over when you’re telling them that you’re still sick.” (Sarah)

“It’s sometimes just easier not to tell people in the first place but then they’re not watching you and asking these questions.” (Harriet)

“It’s like it’s not so much of a focus for myself in my own life, so it doesn’t have to be something you associate me with, you don’t have to look at me and ask me all these questions just like let that happen in the background.” (Harriet)

“To explain the whole, whole diagnosis and what’s wrong with me, people don’t actually wanna know that realistically.” (Harriet)

“Well my manager knows and people I think people I’m close with at work know but not everybody.” (Claire) (limited disclosure)

Subordinate theme: Endurance (an invisible struggle)

“Yeh helpful, just don’t even go there, I wonder at times if having IBD has made me more independent.” (Sally)

“Trying to still do all the things you need to do.” (Annie)

Factors that associate with resilience in people with IBD

“I’ve got to a point now where I can’t stop this kind of letting me do everything that I still want to do.” (Harriet)

“I’m going to carry on and do my thing and not let it stop me.” (Harriet)

“It was kind of like learning to just carry on and staying as I was at that age to like, maintain like, my social life.” (Harriet)

“Not letting it stop me into doing what I need to do, and for me that’s really important.” (Harriet)

“If I’m having a flare up it’s not going to stop me from going out there, doing my socialising, having loads of plans that I’ve already made.” (Harriet)

“I don’t mind now, the challenge, I kind of don’t mind challenges in my life, whatever the challenges are, I quite like fighting them (all laugh), I quite like seeing a challenge and acknowledging that challenge, how am I gonna beat that.” (Harriet)

“I just keep pushing through, and ensure it doesn’t stop me doing my uni work and doesn’t stop me doing or achieve what I want to achieve” (Harriet)

“Times when I could of shouldn’t have been there, you know times like that when I force myself to do things.” (Claire)

“Sit at home and you’re not at work and you feel really guilty for not being at work like the guilt always kicks in and I think cos you’ve been brought up with the whole you work for a living.” (Claire) (social pressure to endure)

“I think it’s just that but people just get on with things don’t they.’ (Claire)

Factors that associate with resilience in people with IBD

“I’m a bit ignorant to it, so I kind of think well I’ll put it to the back of my mind that I’ve got anything wrong with me because it’s just easier just to, get on with things.”

(Claire)

“If someone else was saying that to me then I’d feel awful for them but when I’m saying it, it’s just everyday life.” (Claire)

“When the symptoms are bad it’s hard to get yourself out of a mental state of I just don’t wanna do anything I don’t wanna see anybody it’s that kind.” (Claire) (endurance hard when symptoms more severe)

“I think just it’s doing the things you like to do that make a massive difference and even though sometimes going out is hard cos you’re scared to do it, it helps a hell of a lot cos you’re doing stuff that you enjoy.” (Claire)

“I have a reference point when I’m ill or when anything’s happened, and I’ll say well actually it’s not as bad as that.” (Sally) (desensitisation - allows endurance)

“I think your brothers got the right attitude, for me, I think he’s got the right attitude, to carry on regardless.” (Sally)

“Students that I’ve had with IBD, I have to say they are not necessarily the ones who have asked for exceptional circumstances.....maybe that’s resilience.” (Sally)

“There was never a question that you wouldn’t get up and go to school or work or anything like that so that carries on that stays with you throughout your life.” (Sally)

“Trying to still do all the things you need to do.” (Annie)

“You either don’t do life or you deal with life on or own terms and take that control back and I think that’s when the resilience comes in.” (Sarah)

Factors that associate with resilience in people with IBD

Superordinate theme: Social support

Subordinate theme: Supportive, understanding others

“Even my friends who understand a lot about it now, but sometimes they’re too nice, like if I’m having a flare up like they wrap me up in cotton wool, which I find a bit patronising like I always when I’m ill I hate people treating me like I’m ill whereas my parents like I’ve mentioned before are very much tough love.” (Harriet)

“I think it’s just easier when people understand what you’re going through cos you don’t have to explain everything, it’s like an unsaid communication between the two of you, you don’t have to say it it’s difficult to put into words how IBD can make you feel sometimes.” (Harriet)

“Well work has been amazing I have to say my boss I told him straight away and erm he said have as much time off as is needed.” (Annie)

“Just having emotional support and having someone to talk to, knowing those nurses are at the end of the line.” (Annie)

“The sheer number of women who came to me and emailed me or sent me a card saying its amazing how you’re dealing with this.” (Annie)

“Yeh sort of friends and family my mum always asks me how I am and she’ll send little cards and presents through the post and obviously my husband has been great but I’d have to say work really cos they’ve just been utterly supportive.” (Annie)

“They were just as flexible as I needed them to be.” (Annie) (about employers)

Factors that associate with resilience in people with IBD

“I feel like I need to support people as well because it gives my entire journey of crapness a purpose if I use it positive ways and me being able to offer people advice or support makes me feel better.” (Sarah)

“I mean it was nice to be able to talk to people who understood anyway so that was helpful.” (Sarah)

“My MD actually had Crohn’s disease which helped monumentally.” (Sarah)

“He’s [husband] probably the only person he actually kind of gets it and he’s very patient with it, to be fair my mum and dad are aswell, my family my close family are and friends.” (Claire)

“Like if I’m on a night out and I don’t feel good and I’ll say to a friend I’m gonna have to go home now and they understand it and things like that I think it must just be the people that you are around.” (Claire)

“And I think cos I feel like I handle it quite well I don’t want someone to panda to me, I don’t want to be pandered too.” (Claire)

“Husband yes, yeh I would say, I mean of course he’s only ever known me with Crohn’s, erm so it’s just not an issue for him.” (Sally)

“I had a very understanding flatmate who I would still consider to be my closest friend and I think she put up with a lot.” (Sally)

Subordinate theme: Misunderstanding/dismissive others:

“When I was really poorly my social life as well and people didn’t understand it, so that had quite a big impact on going out and seeing friends and then friends didn’t necessarily understand it so they’d get frustrated at you.” (Claire)

Factors that associate with resilience in people with IBD

“You can explain it as much as you, whereas it’s not always good enough for some people, because they don’t know what it’s like to be in your shoes.” (Claire)

“It’s just like when you’re tired and people are like why you tired you know like just family say you shouldn’t be tired you’re young, it’s things like that and they don’t really know what they’re saying and they obviously they know what I’ve got but they just haven’t thought about what they’re saying to you.” (Claire)

“I was only 15 so a lot of my friends wouldn’t have really understood.” (Harriet)

“If I did have a flare up initially if I was at uni I’d feel a little like alone, so it kind of made me feel a bit more negative about the whole thing.” (Harriet)

“Part of that independence, because like you’re very aware of what you need to do and how you can manage it, the people who don’t understand it are the barriers.” (Harriet)

“Especially as when you’re younger you can get labels like a flake.” (Harriet)

“It’s sometimes just easier not to tell people in the first place but then they’re not watching you and asking these questions.” (Harriet)

“I think that was out of anger and frustration more than anything else because nobody was listening or nobody really understood what I was going through and that really pissed me off.” (about developing an online support forum) (Sarah)

“A lot of people find it difficult to understand anyway, it was a taboo subject.” (Sarah)

“Like it doesn’t make sense to them and people stop inviting you out to things because you don’t turn up very often because you’re not very well and its very painful and its exhausting to keep having to tell people that you’re still not well and you figure out who actually cares.” (Sarah)

Factors that associate with resilience in people with IBD

“They don’t understand and a lot of the time its upsets people when you’re not very well, like my mum if I actually tell her when I’m ill, how I feel and in what way she gets upset about that and I don’t think it’s worth upsetting her.” (Sarah)

“I don’t have friends that are very dramatic and have a lot of issues that aren’t real issues because that stresses me out aswell.” (Sarah)

“We were sort of sat at the dinner table eating and I was just kind of I was just so upset about the whole thing and this daunting thing ahead of me and he just sort of said no no we’re not doing that don’t let your chin drop.” (Annie)

“Partly because I had a very tough boss that would not have been allowed almost.”
(Sally)

“Those who have the condition, yes, those who don’t have the condition, not necessarily and there’s a sort of spectrum of how supportive they can be.” (Sally)

“Because the excuses that other people might come up with, and you think really? actually that doesn’t register on the, erm, on the list of things that might disrupt your life.” (Sally)

“I purposely avoided forums because they just, I think not to be mean but just some people wallow in being a victim of something.” (Annie)

Subordinate theme: Stoicism/brave face

“I don’t think I’ve ever taken time off for IBD.” (Sally)

“They try their very best but because I know they don’t really get were I’m coming from I tend to just brush it off and go with I’m fine.” (Sarah)

Factors that associate with resilience in people with IBD

“I don’t really tell a lot of people not a lot of people really know, only some of my close friends know some of them didn’t really get it so I don’t think there’s any need for people who aren’t that close to me to know, because it shouldn’t be impacting them.” (Claire)

“There’s chairs and you can sit down but you want people to stand up because it’s better for people to think when they’re stood up and sometimes I’ve got to sit down when I should be the one who’s kind of, practicing what you preach and then you’re sat down and you’re like urgh I shouldn’t be doing this.” (Claire)

“I suppose there’s that thing of sometimes when you are feeling pretty shit about it you don’t feel like there’s that many people to talk to about it and the ones you do speak to you feel like they’ve heard it all before and you don’t want to bog them down so you feel like you don’t want to sound like you’re being pathetic.” (Claire)

“I can get into work every day and I manage to do these things so it’s kind of I shouldn’t really complain with how I am cos of what other people.” (Claire)

“Probably times when I could of shouldn’t have been there, you know times like that when I force myself to do things.” (Claire)

“I think people know there’s something, but they don’t know what it is.” (Claire)

“You don’t have to look at me and ask me all these questions just like let that happen in the background, I do, just don’t worry about it.” (Harriet)

“You just have to like push through it like still when I’m going through that I’m not gonna let anyone else in my life that doesn’t need to know that I’m going through that.” (Harriet)

“Hate letting people down, but I wouldn’t want to take time off work because I’m yeh it’s not I don’t know I’d feel guilt as well.” (Harriet)

Factors that associate with resilience in people with IBD

“It’s sometimes just easier not to tell people in the first place but then they’re not watching you and asking these questions.” (Harriet)

Appendix H

All online advertisements

Crohn's and Colitis UK advertisement:

Project title: Resilience in individuals with Inflammatory Bowel Disease.



What the researchers will look at:

The challenges of living with IBD can generate significant stress which can be difficult to cope with. As a result, many individuals experience periods of low mood, anxiety and low self-esteem. We are interested in identifying what helps individuals adapt to the challenges of living with IBD and reduce distress. Resilience is defined as the ability to “bounce back” from adversity and has been well researched in individuals with other physical health conditions. However, there is very little research and understanding about what factors enhance or hinder resilience in people living with IBD. Further knowledge could enable healthcare professionals to help those with IBD adapt and develop resilience in the face of IBD challenges. Factors that influence resilience could include the severity of the disease, the amount of social support one feels they have, how one copes with the condition or how in control one feels.

Researchers at the University of Sheffield are looking for people aged 18 or over who have a confirmed diagnosis of IBD (this might include Crohn's Disease, Ulcerative

Factors that associate with resilience in people with IBD

Colitis or Indeterminate IBD) to take part in a survey which aims to identify what factors help people feel resilient when living with IBD.

The first part of this study has already been conducted. This was an interview study and identified traits, coping skills and experiences people with IBD thought may influence their own resilience. These findings have informed the development of the second part of the study; this online survey. The survey aims to identify what factors have the most influence on resilience across a wider population of individuals with IBD. Approximately 135 people are needed to complete this online survey so that the relationships between the identified factors and resilience can be statistically analysed.

What do the researchers think this could mean for people with IBD?

This research aims to understand what factors are most likely to enhance resilience in individuals with IBD. An improved understanding and evidence base means that psychological interventions (such as talking therapies) offered to individuals with IBD who are struggling with their mental and emotional health can be tailored to strengthen resilience in this population.

If you are interested and would like more information about the study, please click on this link:

https://sheffieldpsychology.eu.qualtrics.com/jfe/form/SV_af1OR152RMOMwLj

Social media advertisement:

"Do you have IBD? Looking for adults over 18 with IBD to participate in my online research study on resilience in people with IBD. To take part, please follow this link: https://sheffieldpsychology.eu.qualtrics.com/jfe/form/SV_af1OR152RMOMwLj":

Factors that associate with resilience in people with IBD

University volunteer database:

Email title:

Do you have IBD? Take part in research on resilience.

Dear Colleague,

>

>I am researching resilience in people with IBD, as part of a study at the University of Sheffield.

>

>If you are over 18, have IBD and are interested please click on this link which will take you to an online survey:

>

>https://sheffieldpsychology.eu.qualtrics.com/jfe/form/SV_af1OR152RMOMwLj

>

>The research has been approved by the Research Ethics Committee in the Department of Psychology at the University of Sheffield.

>

>Thank you very much,

>

>Katie Boden

>Trainee Clinical Psychologist

>

>Information related to this message is available

at https://sheffieldpsychology.eu.qualtrics.com/jfe/form/SV_af1OR152RMOMwLj.

Appendix I

Online survey information and consent

Q5



You are invited....

To participate in a research study conducted at the University of Sheffield. Before you decide to take part it is important that you understand the research that is being done and what it will involve, so that you can give informed consent. Please take the time to read the following information carefully and discuss it with others if you wish. You are welcome to ask any questions of Katie Boden (lead researcher) if there is anything that is not clear or if you would like more information. Please take time to decide whether or not you wish to take part.

The project

This is an exploratory study of the factors that influence resilience in individuals with IBD.

What is the purpose of the research?

This study aims to explore what factors associate with resilience in people managing IBD.

Why have I been invited to participate in the study?

You have been invited to participate because you have been given a medical diagnosis of IBD (either Crohn's Disease or Ulcerative Colitis). Approximately 130 other people will also be recruited to take part in the online survey.

Do I have to take part?

It is up to you to decide whether or not you would like to take part and you are under no obligation to participate. If you decide to take part, you can withdraw from the study up until 4 weeks after completion of the online survey. You do not need to give a reason. You can also withdraw during completion of the survey by closing the browser.

Do I have to take part?

It is up to you to decide whether or not you would like to take part and you are under no obligation to participate. If you decide to take part, you can withdraw from the study up until 4 weeks after completion of the online survey. You do not need to give a reason. You can also withdraw during completion of the survey by closing the browser.

What will happen once I agree to participate in the study?

You will be asked to complete an online survey which will take approximately 15-20 minutes to complete. In the survey you will be asked questions about psychological factors such as dealing with uncertainty, how in control you feel, how accepting of your illness you are, and questions related to resilience. Following completion of the online survey you will be entered into a prize draw with a chance to receive £25 in amazon gift vouchers.

What are the benefits of taking part?

Whilst there are no immediate benefits to participating in the project, it is hoped that this work will add to our knowledge base around the factors that influence resilience so that it can inform the help individuals receive around managing their condition. It is also hoped that it will stimulate further research in this area.

What are the possible disadvantages and risks of taking part?

Some of the questions in the survey will ask about how you manage uncertainty and feelings in control, which may make you feel uncomfortable in some way. If you feel distressed at any stage, you can pause the survey or withdraw from the project completely.

What if something goes wrong or if I become distressed as a result of taking part in the study?

If after participating, you decide that you would like to withdraw your data from the study please email kboden1@sheffield.ac.uk, quoting the unique survey number allocated to you at the end of the study. You do not need to provide a reason for withdrawing from the study.

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If you feel distressed after participating, you can contact your GP or a non-statutory organisation such as Crohn's and Colitis UK.

If, after participating in the study, you wish to raise a complaint, you can do this by contacting Dr Glenn Waller (Head of Psychology Department) on 0114 222 6568 or by email on g.waller@sheffield.ac.uk.

Protecting your identity

All the information that we collect about you during the course of the research will be kept strictly confidential. You will not be identifiable in any reports or publications.

What type of information will be sought from me and why is the collection of this information relevant for achieving the research project's objectives?

Information about your age, gender, ethnicity and your diagnosis or IBD will be collated because this will help the researcher to understand whether these factors impact on levels of resilience.

What will happen to the results of the research project?

The study results will be written up and submitted as a doctoral thesis. You will not be identifiable in any report or publication of these results. The results of this study may be published in a peer-reviewed scientific journal in the future.

Who is organising and funding the research?

The University of Sheffield is organising and funding this research.

Who has ethically reviewed the project?

This project has been ethically approved via Sheffield ethics committee.




Contact for further information

If you would like any further information you can contact Katie Boden (lead researcher) by email kboden1@Sheffield.ac.uk

Online consent:

Consent Block Options ▾

Q107 **Please Select the appropriate response.**
Taking part in the project

	Yes	No
 I have read and understood the project information sheet dated $\$(date://CurrentDate/SL)$ (If you will answer No to this question please do not proceed with this consent form until you are fully aware of what your participation in the project will mean.)	<input type="radio"/>	<input type="radio"/>
 I have been given the opportunity to ask questions about the project.	<input type="radio"/>	<input type="radio"/>
 I agree to take part in the project. I understand that taking part in the project will include completing a questionnaire.	<input type="radio"/>	<input type="radio"/>
I understand that my taking part is voluntary and that I can withdraw from the study anytime up to the point that I submit my survey. I do not have to give any reasons for why I no longer want to take part and there will be no adverse consequences if I choose to withdraw.	<input type="radio"/>	<input type="radio"/>

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Q108

How my information will be used during and after the project

	Yes	No
<input type="checkbox"/> I understand my personal details such as name, phone number, address and email address etc. will not be revealed to people outside the project.	<input type="radio"/>	<input type="radio"/>
<input type="checkbox"/> I understand and agree that my anonymous words may be quoted in publications, reports, web pages, and other research outputs.	<input type="radio"/>	<input type="radio"/>
<input type="checkbox"/> I understand and agree that other authorised researchers will have access to this anonymous data only if they agree to preserve the confidentiality of the information as requested in this form.	<input type="radio"/>	<input type="radio"/>
<input type="checkbox"/> I understand and agree that other authorised researchers may use my anonymous data in publications, reports, web pages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form.	<input type="radio"/>	<input type="radio"/>
<input type="checkbox"/> I give permission for the anonymous data that I provide to be deposited in the Dept. of Psychology at the University of Sheffield so it can be used for future research and learning.	<input type="radio"/>	<input type="radio"/>

Q109

So that the information you provide can be used legally by the researchers

	Yes	No
<input type="checkbox"/> I agree to assign the copyright I hold in any materials generated as part of this project to The University of Sheffield.	<input type="radio"/>	<input type="radio"/>

Q110

Do you wish to continue? To acknowledge that you have read and understood this information and would like to continue with the research study, please click on "I agree".

I agree

No, thank you

Factors that associate with resilience in people with IBD

Appendix J Copy of all measures

Demographics questions (including questions regarding TSD, relapse/remission status)

Demographics Block Options ▾

Q12 What is your gender?

Male

Female

Other

Q8 What is your current age?

*

Page Break

Q10 In what country/continent do you currently live?

United Kingdom

Australia

Europe

South America

Canada

Other (please list below)

USA

Page Break

Q34 What is your diagnosis?

Crohn's disease

Ulcerative colitis

Other, please list below

Q38 Please list other IBD below:

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<input type="checkbox"/>	How old were you when you were diagnosed with IBD? (in years)
Q14	<input type="text"/>
<input type="checkbox"/>	Would you consider yourself to be in relapse or remission right now?
Q16	<input type="radio"/> Relapse
	<input type="radio"/> Remission

Connor-Davidson Resilience Scale (CD-RISC, Connor & Davidson, 2003).

Removed for copyright reasons

Appendix K

Normality tests

SPSS screenshots of tests of normality demonstrating Shapiro-Wilks tests. The Shapiro-Wilk test has more power to detect differences, therefore the significance level from this test was interpreted (Field, 2009). If the value of this was greater than 0.05 ($p > 0.05$), the data were deemed to be normally distributed. Therefore, disease activity, FSSQ data (social support), SGS (grit), CES (coping-efficacy), TSD and acceptance were deemed to be not normally distributed. The dependent variable of resilience was normally distributed.

SPSS screenshot of Shapiro-Wilk tests:

	Tests of Normality					
	Kolmogorov-Smirnov ^a			Shapiro-Wilk		
	Statistic	df	Sig.	Statistic	df	Sig.
Totaldiseaseactivity	.109	85	.015	.965	85	.021
TotalFSSQfinal	.110	85	.012	.934	85	.000
TotalGRIT	.134	85	.001	.962	85	.013
TotalCBI	.084	85	.200*	.979	85	.174
Totalcopingeff	.125	85	.002	.943	85	.001
TotalRISC	.086	85	.174	.983	85	.324
TotalIU	.078	85	.200*	.976	85	.117
TSD	.156	85	.000	.898	85	.000
ICQACCsubscale	.111	85	.012	.968	85	.032

*. This is a lower bound of the true significance.

a. Lilliefors Significance Correction

Factors that associate with resilience in people with IBD

SPSS output screenshots demonstrating kurtosis and skewness values, histograms and Q-Q plots for each variable:

Totaldiseaseactivity	Mean		201.3901	9.85562
	95% Confidence Interval for Mean	Lower Bound	181.7912	
		Upper Bound	220.9891	
	5% Trimmed Mean		200.8391	
	Median		197.0000	
	Variance		8256.325	
	Std. Deviation		90.86432	
	Minimum		38.00	
	Maximum		400.00	
	Range		362.00	
	Interquartile Range		157.50	
	Skewness		.092	.261
	Kurtosis		-1.076	.517
	TotalFSSQfinal	Mean		3.8733
95% Confidence Interval for Mean		Lower Bound	3.6845	
		Upper Bound	4.0621	
5% Trimmed Mean			3.9238	
Median			4.0000	
Variance			.766	
Std. Deviation			.87535	
Minimum			1.38	
Maximum			5.00	
Range			3.63	
Interquartile Range			1.25	
Skewness			-.746	.261
Kurtosis			-.113	.517

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TotalGRIT	Mean		2.2408	.03964
	95% Confidence Interval for Mean	Lower Bound	2.1619	
		Upper Bound	2.3196	
	5% Trimmed Mean		2.2516	
	Median		2.2500	
	Variance		.134	
	Std. Deviation		.36543	
	Minimum		1.25	
	Maximum		3.00	
	Range		1.75	
	Interquartile Range		.50	
	Skewness		-.530	.261
	Kurtosis		-.161	.517

ICQACCsubscale	Mean		16.4118	.45322
	95% Confidence Interval for Mean	Lower Bound	15.5105	
		Upper Bound	17.3130	
	5% Trimmed Mean		16.4248	
	Median		16.0000	
	Variance		17.459	
	Std. Deviation		4.17844	
	Minimum		6.00	
	Maximum		24.00	
	Range		18.00	
	Interquartile Range		7.00	
	Skewness		.019	.261
	Kurtosis		-.765	.517

TotalCBI	Mean		116.6042	2.02984
	95% Confidence Interval for Mean	Lower Bound	112.5676	
		Upper Bound	120.6408	
	5% Trimmed Mean		116.8772	
	Median		119.0000	
	Variance		350.223	
	Std. Deviation		18.71424	
	Minimum		65.00	
	Maximum		153.00	
	Range		88.00	
	Interquartile Range		27.91	
	Skewness		-.351	.261
	Kurtosis		-.408	.517

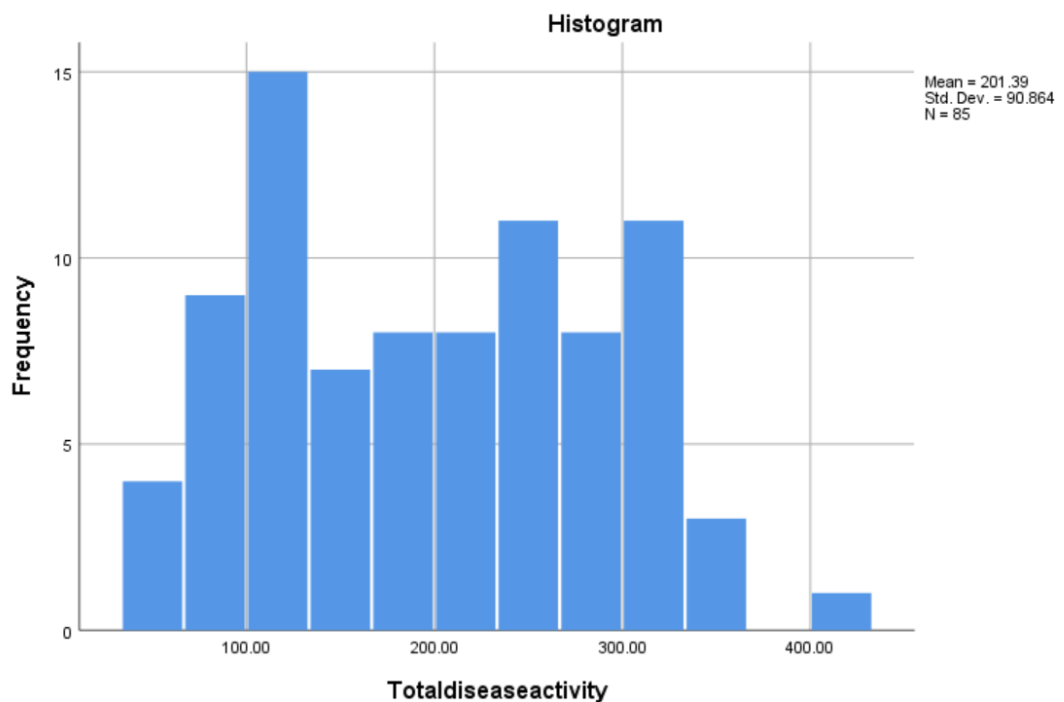
Factors that associate with resilience in people with IBD

Totalcopingeff	Mean		15.3213	.45894
	95% Confidence Interval for Mean	Lower Bound	14.4086	
		Upper Bound	16.2339	
	5% Trimmed Mean		15.5727	
	Median		15.0000	
	Variance		17.903	
	Std. Deviation		4.23118	
	Minimum		3.00	
	Maximum		21.00	
	Range		18.00	
	Interquartile Range		5.00	
	Skewness		-.667	.261
	Kurtosis		.082	.517
	TotalRISC	Mean		66.0706
95% Confidence Interval for Mean		Lower Bound	62.7701	
		Upper Bound	69.3711	
5% Trimmed Mean			66.3693	
Median			67.0000	
Variance			234.138	
Std. Deviation			15.30156	
Minimum			29.00	
Maximum			96.00	
Range			67.00	
Interquartile Range			25.50	
Skewness			-.217	.261
Kurtosis			-.554	.517
TotalIU		Mean		32.4706
	95% Confidence Interval for Mean	Lower Bound	30.4652	
		Upper Bound	34.4760	
	5% Trimmed Mean		32.0948	
	Median		32.0000	
	Variance		86.443	
	Std. Deviation		9.29745	
	Minimum		15.00	
	Maximum		60.00	
	Range		45.00	
	Interquartile Range		11.00	
	Skewness		.559	.261
	Kurtosis		.353	.517

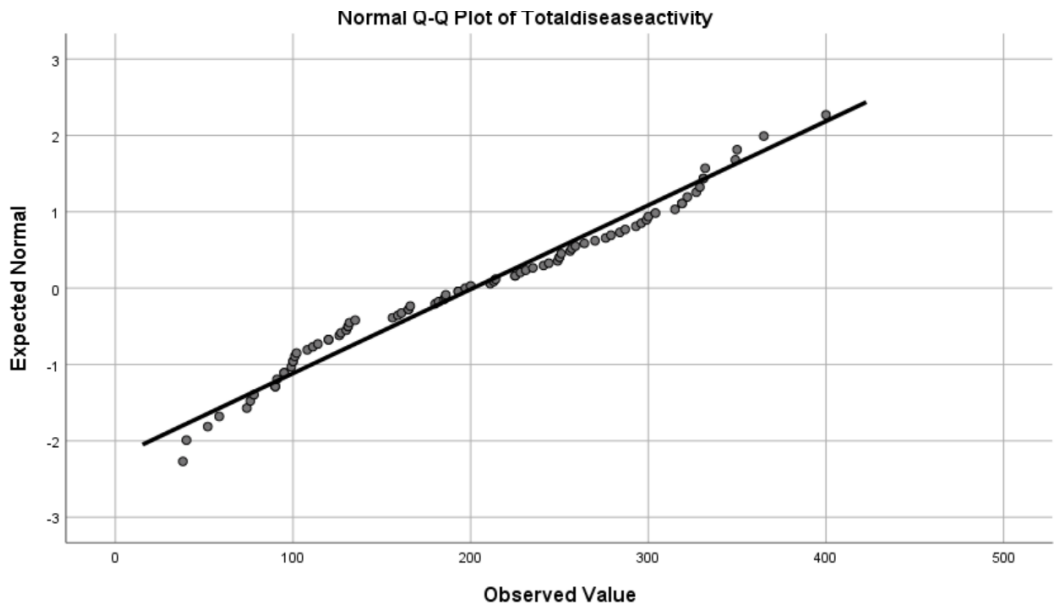
Factors that associate with resilience in people with IBD

TSD	Mean		12.9647	1.19481
	95% Confidence Interval for Mean	Lower Bound	10.5887	
		Upper Bound	15.3407	
	5% Trimmed Mean		12.1046	
	Median		9.0000	
	Variance		121.344	
	Std. Deviation		11.01562	
	Minimum		.00	
	Maximum		45.00	
	Range		45.00	
	Interquartile Range		18.00	
	Skewness		.959	.261
	Kurtosis		.253	.517

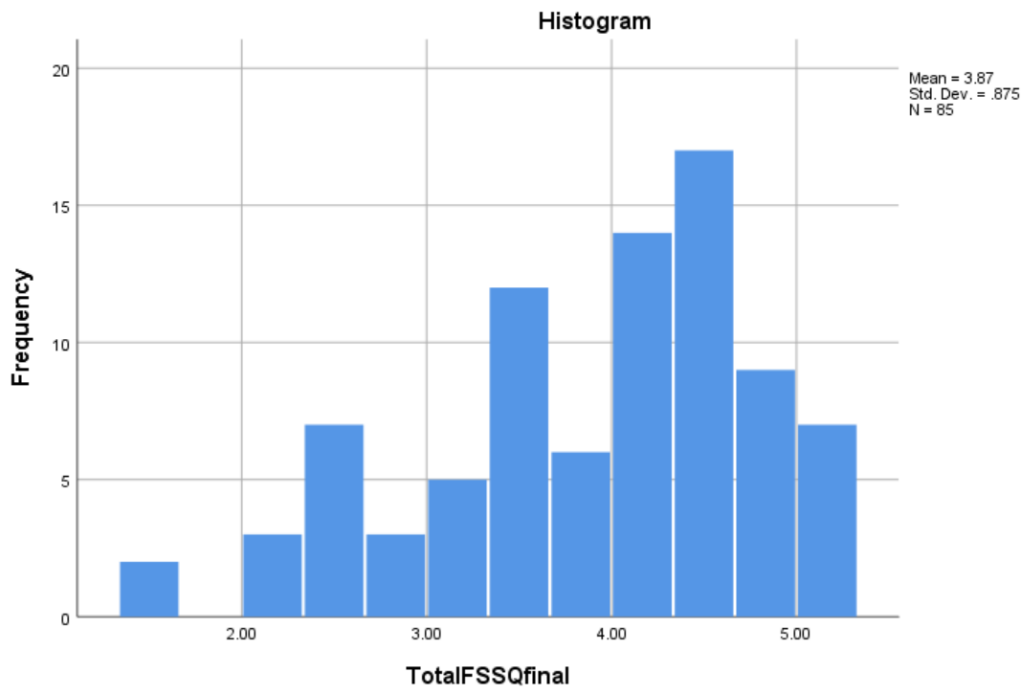
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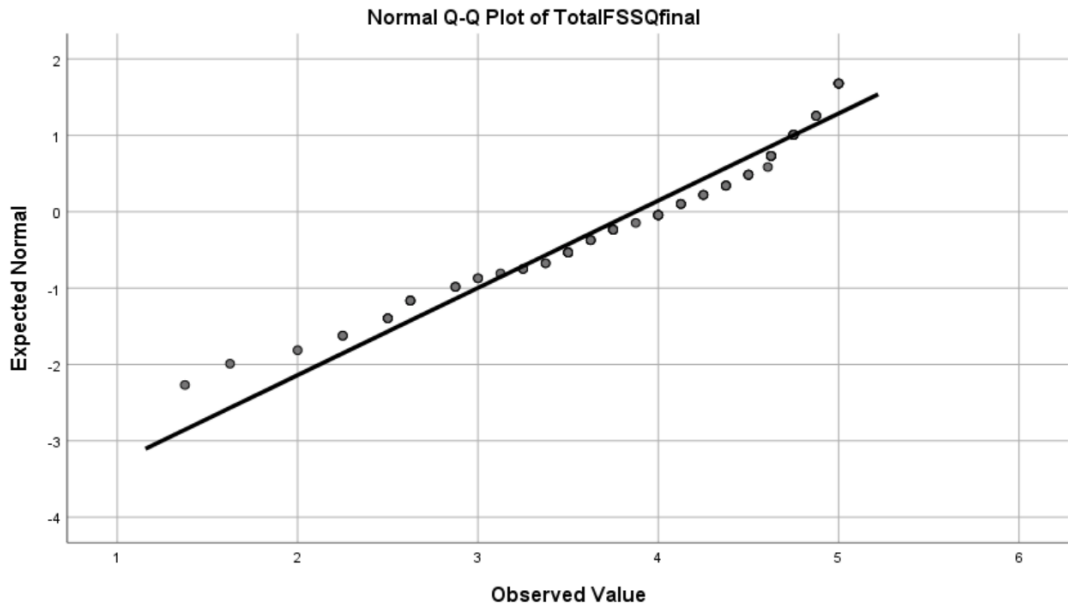
Factors that associate with resilience in people with IBD



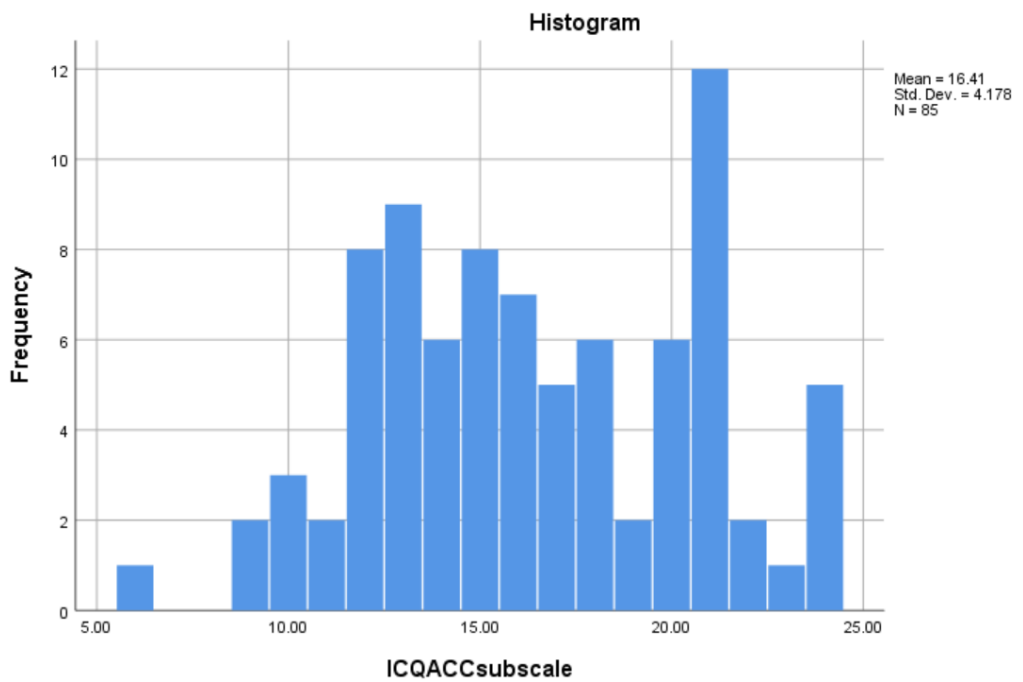
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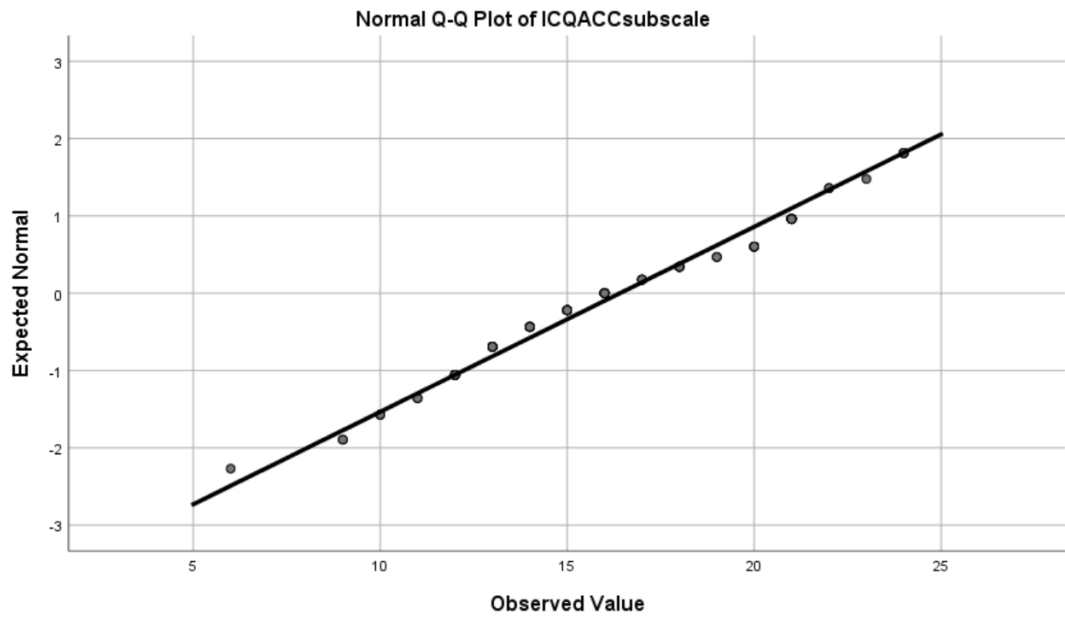
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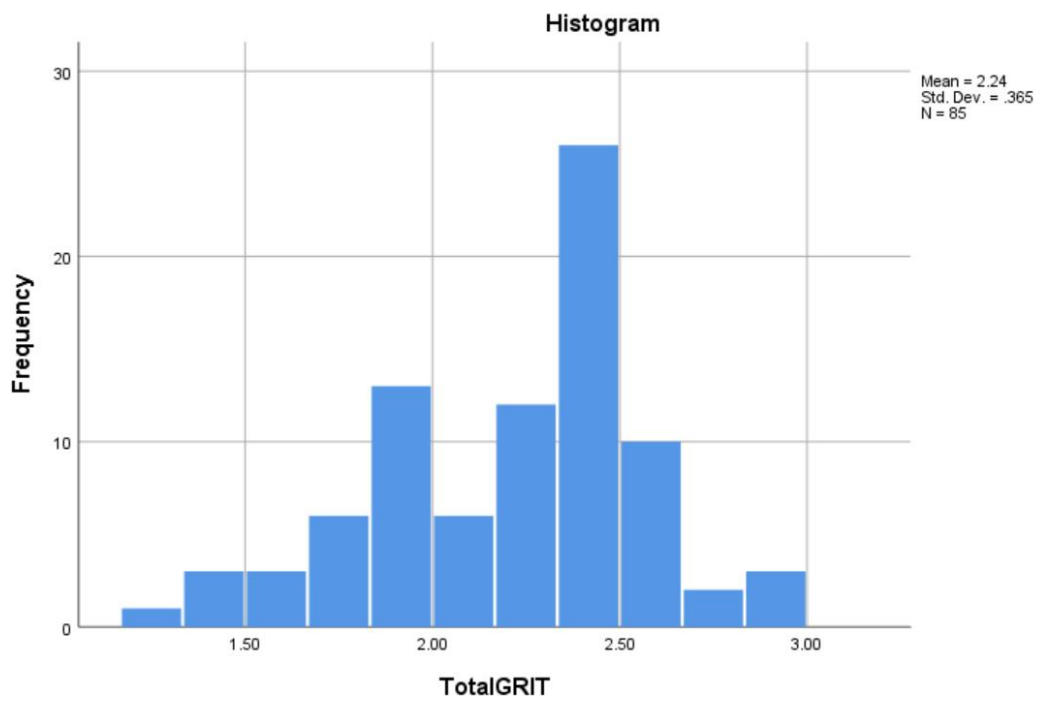
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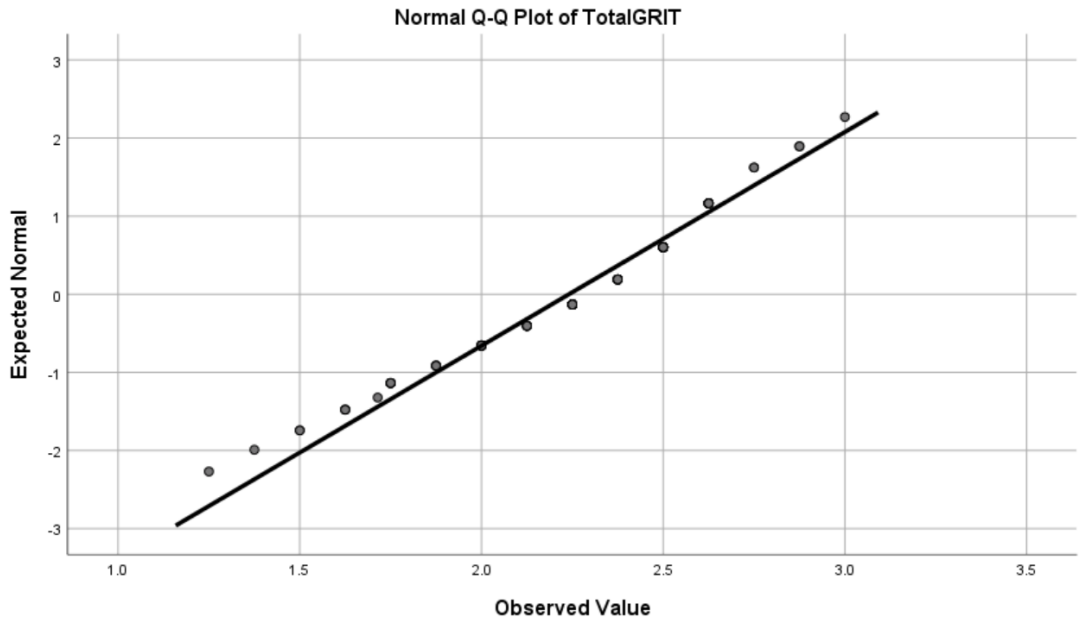
Factors that associate with resilience in people with IBD



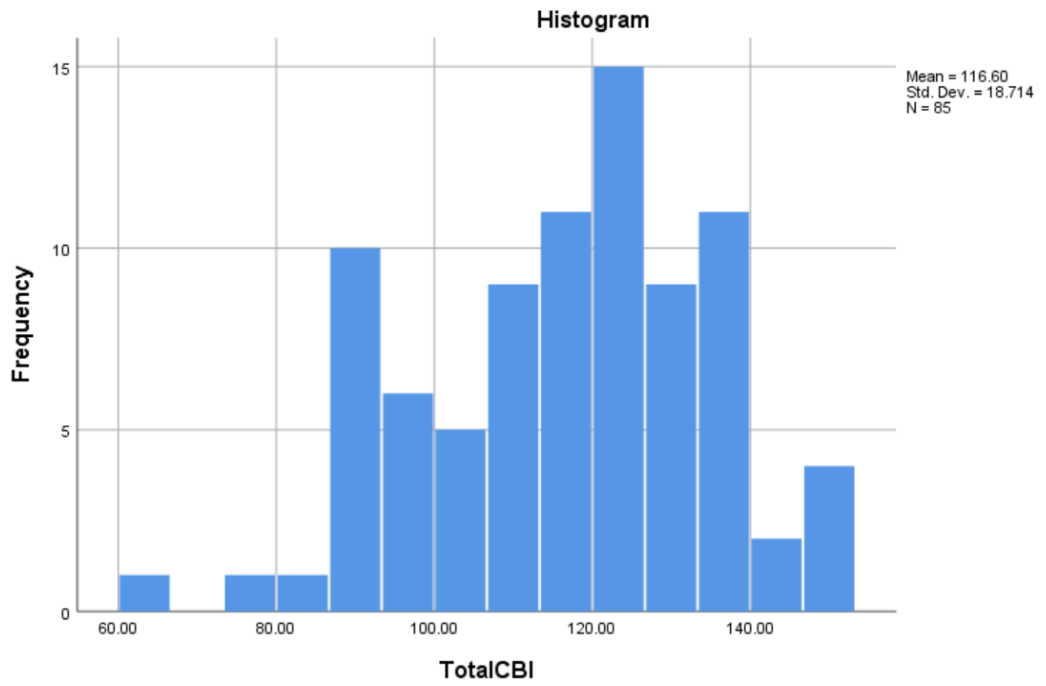
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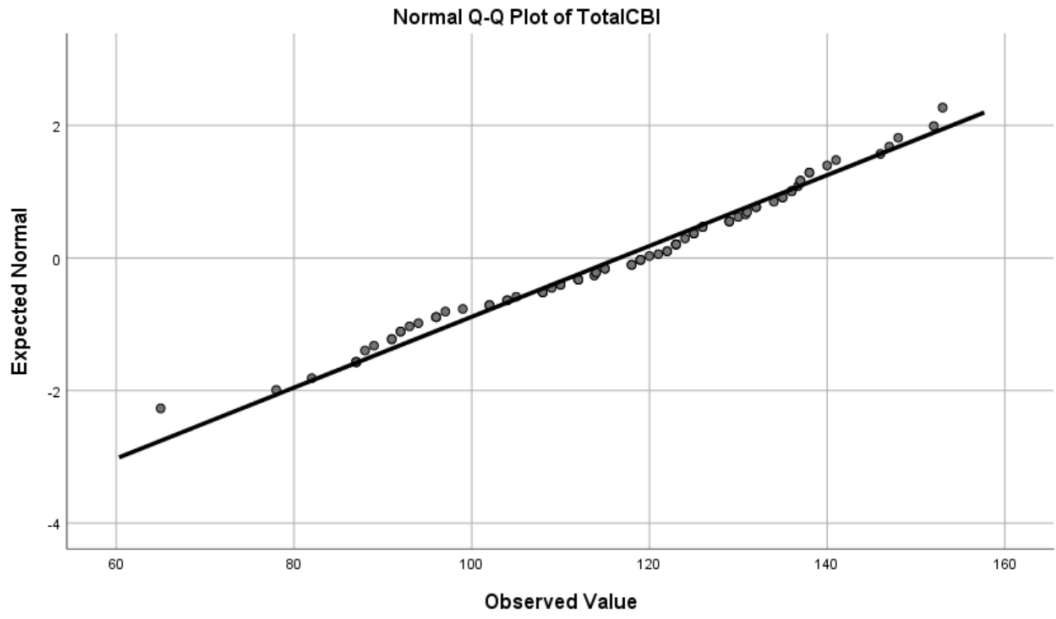
Factors that associate with resilience in people with IBD



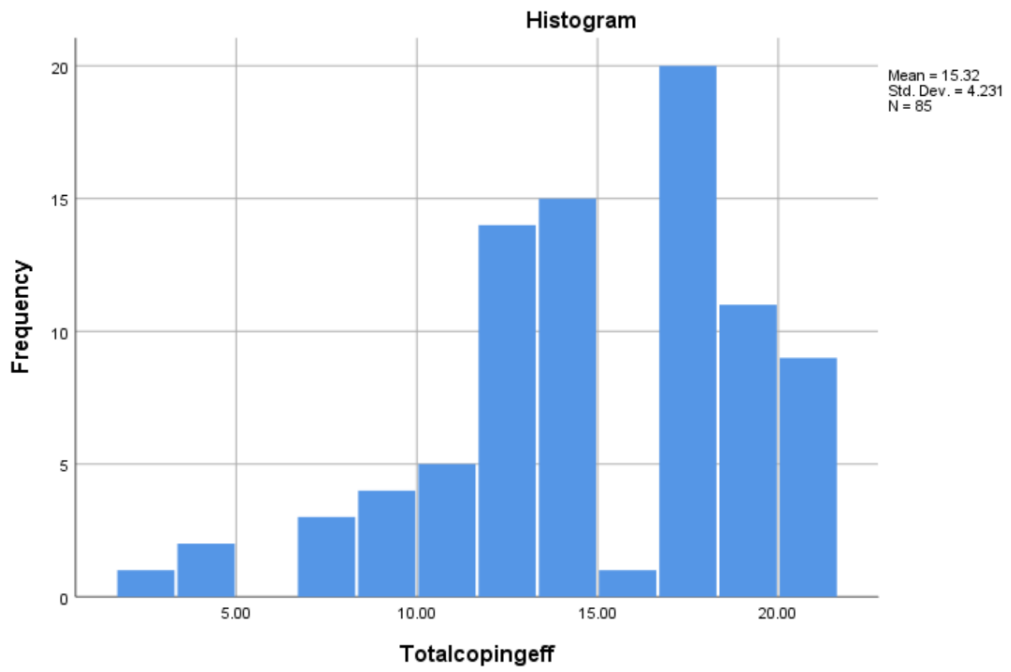
TotalCBI



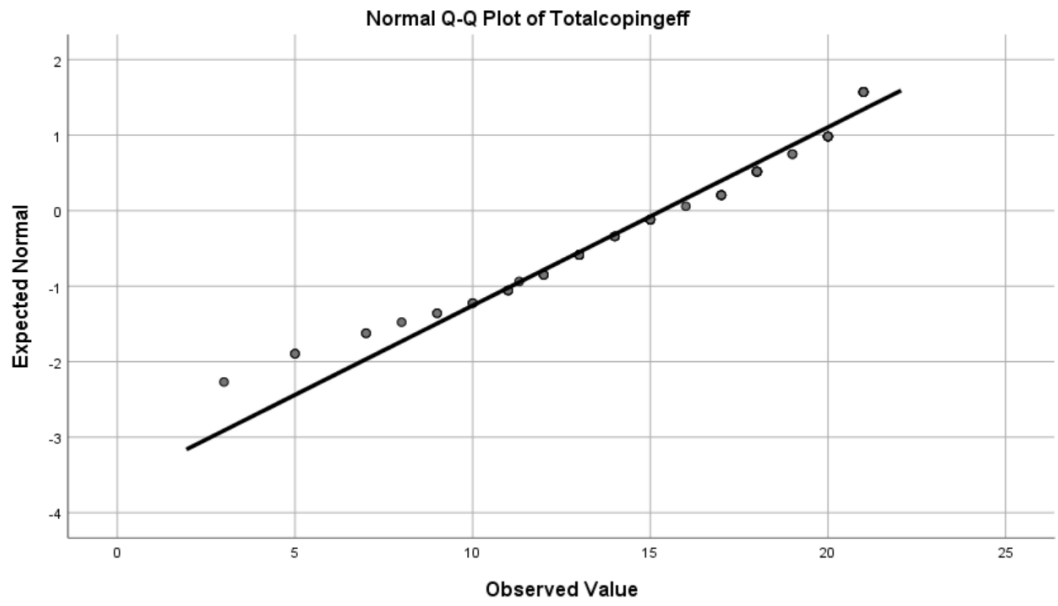
Factors that associate with resilience in people with IBD



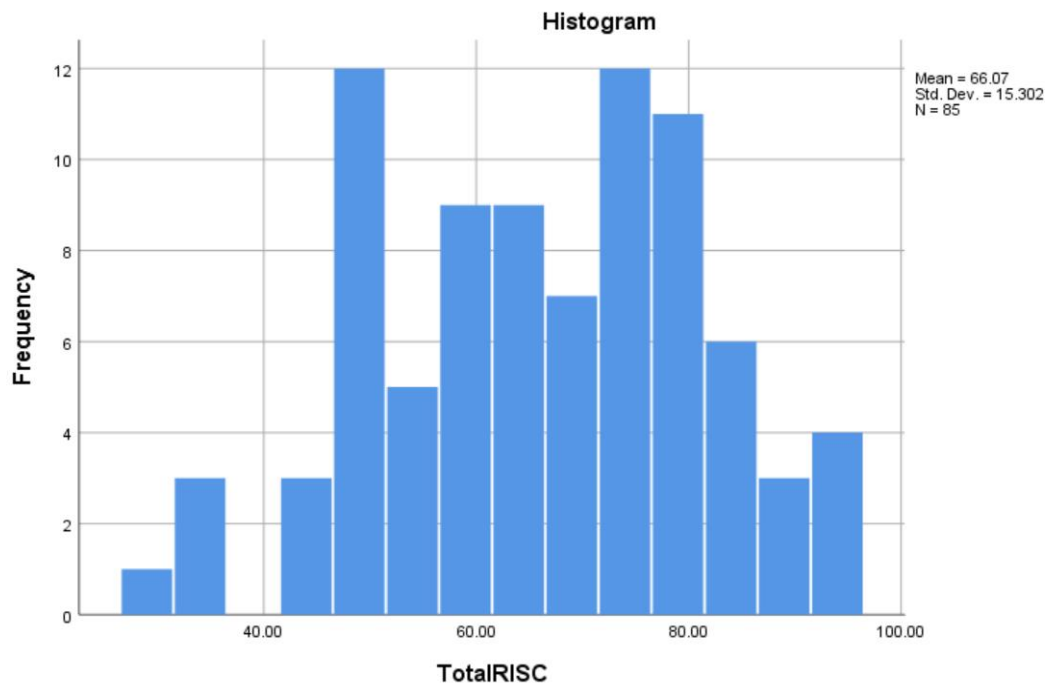
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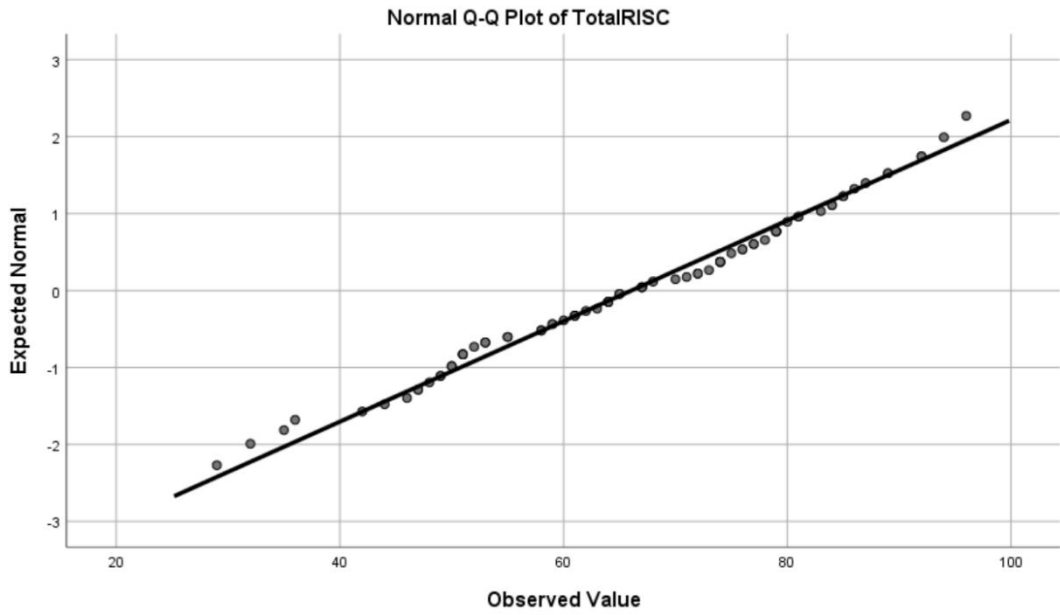
Factors that associate with resilience in people with IBD



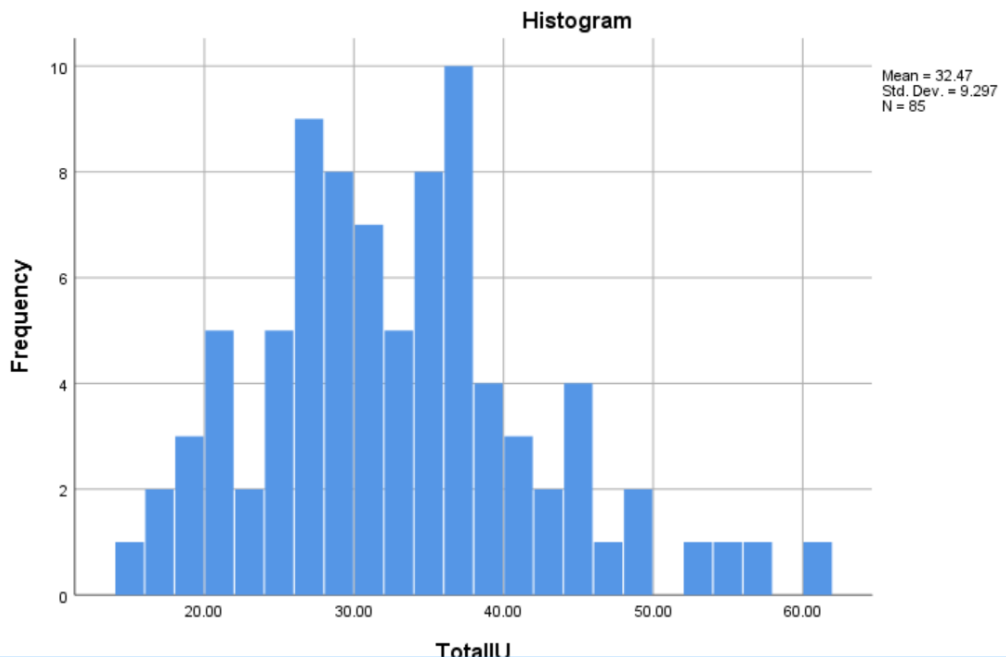
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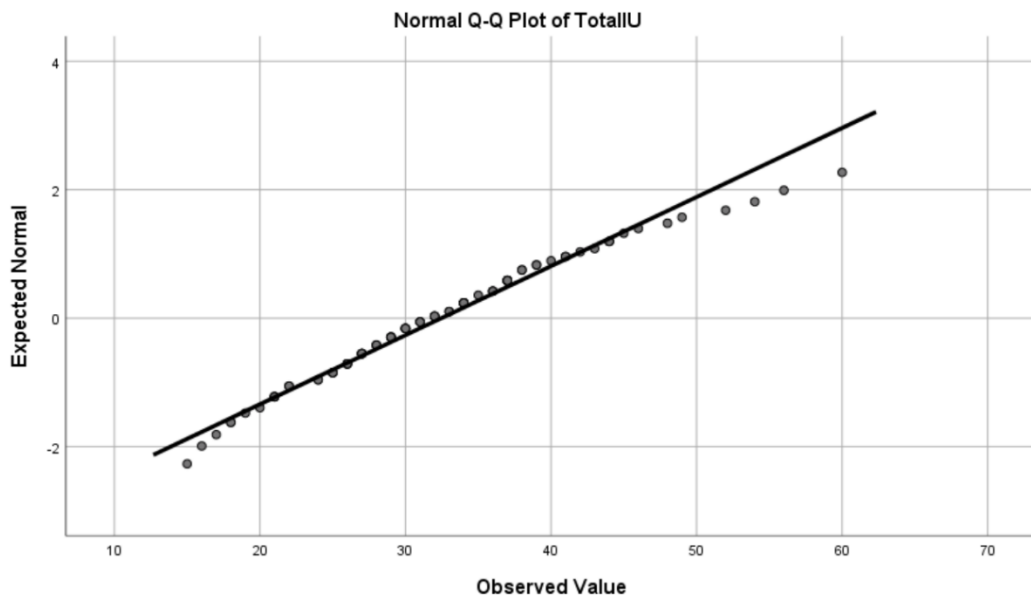
Factors that associate with resilience in people with IBD



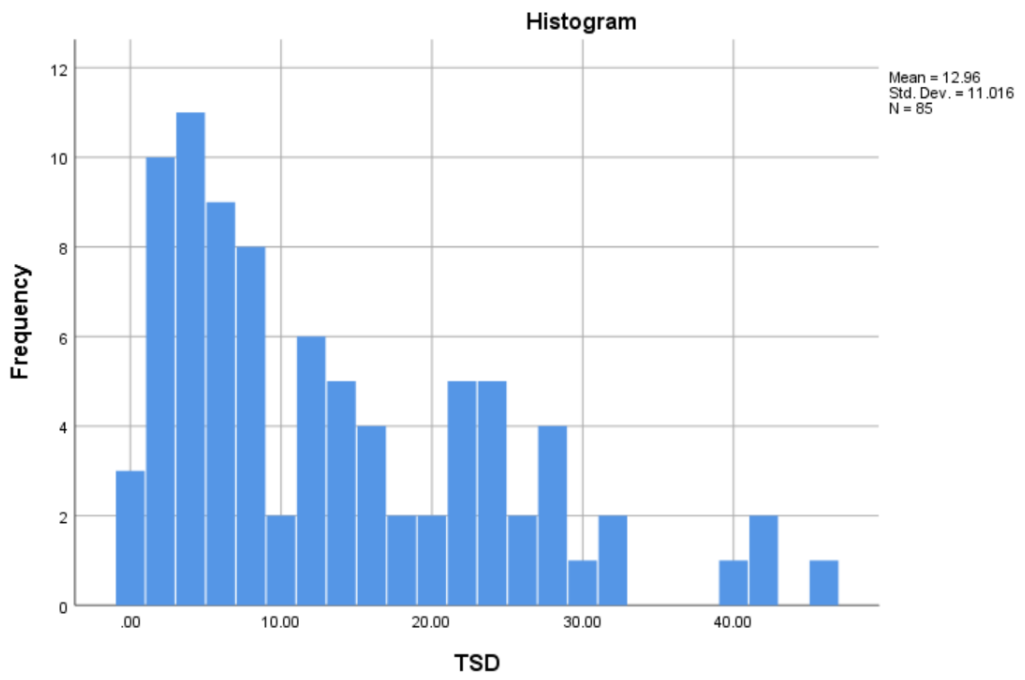
TotalIU



Factors that associate with resilience in people with IBD



TSD



Factors that associate with resilience in people with IBD

