The Experiences of Young People with Inflammatory Bowel Disease

By:

Oonagh O'Hare

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Faculty of Science
Clinical Psychology Unit, Department of Psychology

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Declaration

I declare that this work has not been submitted for any other degree at the University of Sheffield, or any other institution. The work presented is original and all other sources have been references accordingly.

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Structure and word counts

Section One: Literature Review

Excluding references and tables: 7042

Including references and tables: 11,123

Section Two: Empirical Paper

Excluding references and tables: 8031

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General Abstract

Inflammatory Bowel Disease (IBD) is a chronic health condition that is often diagnosed in youth. Living with a chronic condition can have far reaching implications for the patient and for those around them. However, health research tends to focus on the negative aspects of living with a health condition and may overlook the fact that despite adversity, people can and do adjust and can even grow and develop positively through that adversity. The life experiences of young people with inflammatory bowel disease has been examined in two different contexts.

The first part is a systematic literature review. Here, family functioning in the context of youth IBD has been examined. Family functioning refers to the levels of conflict and cohesion in families and requires family members to have clear roles. In the face of a chronic illness such as IBD, it is possible that family functioning may be put under pressure. Alternatively, it may be that families are well equipped to deal with stressors such as a member having a chronic illness. A systematic search was conducted, and 18 studies included for review. Studies were of reasonable quality although they were weakest in their recruitment procedures leaving questions about the validity of the results. Overall, the findings of the studies were mixed which means that it is still unclear whether there is a relationship between IBD and family functioning. Some families experienced little disruption when accommodating IBD whereas others did experience poorer family functioning. However, a stronger finding was that family functioning was no worse in families of young people with IBD than in families of healthy children or families of children with other chronic conditions.

The second part is a qualitative exploration of post-adversarial growth (PAG) in young people with IBD. PAG refers to a profound personal change in which individuals feel they have positively benefitted from adversity. This can mean different things for different people but can include a new outlook on life, new goals or paths, deeper

relationships or positive cognitive changes. Interviews were transcribed and analysed using Template Analysis. Results suggested that young people with IBD did report changes as a result of their illness that were consistent with the concept of PAG. Despite the adversity, they reported deeper relationships, taking back control of their lives from the illness, and positive psychological change. The results suggest that young people can experience PAG.

Taken together, the two pieces of work suggest that whilst there are indeed challenges to living with IBD, the picture may not be all negative. Some families are able to cope with the challenge of supporting a member with IBD and young people with IBD are able to experience positive growth.

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I would like the thank the charitable organisations who supported the recruitment for this project. I would also like to thank the fourteen participants who shared their stories with me and without whom this research would have been impossible. Their stories were incredibly moving, inspiring, humbling and fascinating. Their collective wisdom carried me through when I thought I would never finish!

I would like to thank my supervisor Professor Gillian Hardy for her support - we didn't think it would be quite so much work when you inherited me, but you supported me nonetheless and I am very grateful.

I would like to thank my partner Dov for his patience and for helping me to keep the fire alive (TSC!). I would also like to thank my fantastic family, particularly my mum and dad who went above and beyond to help me and ensure I was eating more than just percy pigs and colin caterpillars!

Finally, when the completion of this research project felt impossible there were two creatures who sat by my side night after night. They made me laugh, take breaks, offered comfort and affection. They will never know how grateful I am – to Giles and Beatrix.

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Section 1. Literature Review

Family Functioning in the Context of Inflammatory Bowel Disease

Abstract

Objectives. The objective of the current review was to explore the concept of family functioning in the context of youth IBD. It aimed to synthesise what is currently known about the relationship between family functioning and youth IBD.

Methods. A systematic search was conducted through four databases; PsycInfo, Medline, Scopus and Web of Science. Additional grey literature searches were also carried out. Inclusion criteria were that studies involved both families and young people with IBD and utilised a standardised measure of family functioning as well as some description of IBD activity.

Results. Eighteen studies were included for review. Study quality was appraised using the AXIS quality appraisal tool for cross sectional studies. Studies met the majority of criteria but were weakened by poor justification of sample sizes, poor descriptions of recruitment and did not include sufficient information about non-responders. The findings of the studies were mixed; three studies suggest there was a significant link between IBD and poorer family functioning whereas four found no such link. However, two of these studies did find a non-significant trend. Other studies compared family functioning in the context of IBD to healthy controls or other conditions. These studies found that families of young people with IBD were no more likely to experience family dysfunction than families of young people with other conditions or healthy controls.

Conclusions. No consistent link between IBD in youth and family dysfunction can be concluded from these studies. This may be attributable to recruitment issues or

alternatively family functioning may be too complex and dynamic a concept for cross-sectional studies to uncover.

Practitioner Points

- Despite the inconclusive results of the current review, clearly some families do struggle to adapt to a young member who has IBD. Under such pressures, IBD outcomes may be affected. Practitioners working in paediatric gastroenterology settings may wish to pay closer attention to the family context and consider referrals to appropriate services if necessary.
- Using purely questionnaire-based assessments however may not be sufficient to uncover the entire picture of family life in the context of IBD. Repeated assessments to capture fluctuations of family functioning at different time points may be helpful. Alternatively, more qualitative methods may prove useful.

Limitations

- The current review is limited by the broad inclusion criteria which did not specify that studies primary aim should be to investigate the direct relationship between family functioning and IBD. This resulted in a wide variety of studies and limited the ability to draw firm conclusions.
- The definition of family functioning included in this review meant that the views of specific family members, such as siblings, may have been overlooked.

Introduction

What is Inflammatory Bowel Disease

Inflammatory Bowel Disease (IBD causes inflammation of the digestive system and refers to two main illnesses; Ulcerative Colitis (UC) and Crohn's Disease (CD). Symptoms vary depending on severity of the disease and site of inflammation but can include stomach pain, frequent bowel movements, skin and joint problems, anaemia and extreme fatigue (Crohn's and Colitis Foundation of America, 2014). Although it can occur at any age, large scale studies have found that onset is typically between 15 and 29 years of age (Johnston & Logan, 2008). IBD is characterised by periods of relapse and remission. It cannot be cured but treatments aim to bring sufferers into and stay in remission. This can include a range of medications including immunosuppressants or steroids (Crohn's and Colitis Foundation of America, 2014). Unfortunately, many of these medications cause significant side effects which many people with IBD report can be worse than the condition (de Rooy et al., 2001). In some cases, sufferers will require surgery. This can include removing parts of or entire affected sections of the digestive system such as the colon and rectum (Crohn's and Colitis Foundation of America, 2014). In these cases, sufferers will be required to have an external pouch connected to their small intestines in order to collect waste (Crohn's and Colitis Foundation of America, 2014)

The impact of living with IBD can be significant; people with IBD report concerns with feeling unclean, and fatigue interfering with attempts to live everyday life (de Rooy et al., 2001). Due to the relapsing and remitting nature of IBD, it can be difficult to predict what symptoms will appear and when. Planning activities can be difficult and people with IBD report difficulties in engaging in social situations due to the constant fear that they

may need to rush to a bathroom or have an embarrassing accident (Kemp, Griffiths, & Lovell., 2012). A meta-synthesis exploring living with IBD found that many sufferers had lived experience of incontinence and the fear of reliving the humiliation was constant (Kemp, Griffiths, & Lovell, 2012).

IBD in Youth

Adolescence is a time of great change; physical, psychological and social. This is a time of rapid physical growth, prioritising of peer relationships over familial ones, increasing independence, sexual development as well as cognitive maturation (Holmbeck, 2002; Sales & Irwin, 2013). Adolescence is also a time of risk taking and pushing boundaries (Sales & Irwin, 2013). Some of these behaviours may involve increased risks to health such as drinking or substance use (Sales & Irwin, 2013). Not surprisingly, adolescence is also a developmental period that is associated with greater risk of developing mental health problems in the general population (Beesdo, Knappe, & Pine, 2009). Given the challenges that accompany life with IBD, it is unsurprising that research has found that young people with IBD can be particularly at risk for low self-esteem, stress and poorer quality of life (Nicholas, Otley, Smith, Avolio, Munk, & Griffiths., 2007). For example, one meta-analysis found that of several different chronic conditions, young people with IBD seemed to experience the poorest mental health outcomes (Lavigne, & Faier-Routman, 1992). Young people with IBD may experience delays in some of the typical developments of adolescence due to their condition. For example, delayed sexual development, delayed or inhibited growth and delayed puberty (Mamula, Markowitz, & Baldassano, 2003). Further, young people with IBD experience high rates of absenteeism from school (Nicholas et al., 2007) which can affect academic attainment and occupational goals in later life (Suris, Michaud, & Viner, 2004). Given these extra stressors on an adolescent with IBD effective use of coping strategies are

therefore essential and maladaptive coping can be associated with poorer physical outcomes in young people with IBD (Mamula et al., 2003). However, younger people have less developed coping skills and the coping strategies of their parents and the family as a whole can heavily influence the coping of the children in the family (Mamula, et al., 2003). Indeed, MacPhee, Hoffenburg and Feranchak (1998) found that young people more heavily rely on the coping strategies of their parents than their own following a recent diagnosis of IBD.

The Family Impact of Chronic Illness

In addition to influencing how the young person will respond to their illness, the family must adapt and respond. Not all families experience a detrimental impact of supporting a child with a chronic condition; indeed, some families report improved relationships and closeness (MacPhee et al., 1998). However, a repeated finding in research is that parents of children with a chronic condition frequently experience stress and burnout (Lindström, Aman, & Norburg., 2010). Indeed, Lindström et al. (2010) found that parents of children with type one diabetes or IBD experienced higher levels of stress and burnout than parents of children without chronic conditions. Other factors that can exacerbate the impact of child chronic illness include having a low family income, being a single-parent household or having more children in the home (Herzer et al., 2010; Drotar & Bonner, 2009).

Family functioning encompasses levels of cohesion or conflict within families, members having clear roles, positive relationships, communication within families, organisation and adaptability (Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010). Family functioning can be affected by chronic illness. For example, conflict can increase if a sibling feels jealous of the increased attention the ill young person receives (Mamula et al., 2003). Further, families supporting a young person with a long-term

illness can experience difficulties in relationships, family structure and cohesion (Herzer et al., 2010). Similarly, a young person's health can be influenced by family functioning. For example, poorer family functioning has been found to increase experience of pain for young people with gastrointestinal conditions (Reed-Knight et al., 2018). Thus, family functioning has repeatedly been found as a strong predictor of psychological outcomes in young people with chronic health conditions (Drotar, 1997).

Researcher have attempted to summarise the patterns of responses families may typically exhibit in the context of a paediatric chronic condition. They have highlighted a consistent finding that after an initial period of adjustment, families' main response is to attempt to return to normal (Knafl, Deatrick, Knafl, Gallo, Grey, & Dixon., 2013). Whilst normalisation is not it itself an indicator of success, it does appear to be associated with good family management in the context of chronic childhood illness (Knafl et al., 2013). Knafl and colleagues (2013) assessed families managing a range of conditions and found four different patterns; family focussed, condition focussed, somewhat family focussed, and somewhat condition focussed. Condition focused represents families who have had the most difficulty incorporating the condition into family life whereas family focussed represents those who perceive the illness as successfully incorporated into family life and therefore not problematic. Few family characteristics seemed associated with a particular response type; only single parent and low-income families were more likely to be classed as condition focused. Further, condition focussed responses were significantly associated with child behaviour problems and poorer daily functioning. However, the majority of families fit within the family or somewhat family focussed patterns whereas only 8% of families fit within a condition focused pattern.

The Family Systems Illness Model (Rolland, 1987) suggests several factors that may influence the family system in the context of illness. For conditions such as IBD

which occur suddenly, the emotional and practical changes can take place quickly and require rapid problem solving and crisis management. It also suggests that relapsing and remitting conditions such as IBD can be particularly challenging for families due to the constant uncertainty and need for flexibility to accommodate the changing illness picture. This uncertainty may also apply to the outcome of IBD. Whilst in itself it is fatal in only extreme circumstances, it is associated with risks of other potentially fatal conditions such as colon cancer, blood clots and liver disease (Crohn's and Colitis Foundation of America, 2004). Further, the severity of the condition varies wildly from person to person and the trajectory can be difficult to predict (Crohn's and Colitis Foundation of America, 2004). This model also purports that a family's response to an illness cannot be understood without taking account of the family beliefs, current coping strategies and attempts to create meaning from events. Families who are able to cope with an illness such as IBD can do so by maintaining hope whilst accepting losses and being flexible in terms of typical family events such as children leaving home and planning around the illness (Rolland, 1987).

Another model of family functioning in the context of illness is the Family Adjustment and Adaptation Response (FAAR; Patterson, 1988). According to this model, following a disruption such as an illness, families may initially try and resist any major changes in the face of a disruption, but if the situation escalates to the point that the stress outweighs resources a family may experience a crisis (Patterson, 1988). The FAAR model however suggests that crises are opportunities to learn to adapt to the new challenges and adjust (Patterson & Garwick, 1994). How successfully a family adapts depends on whether they can develop new coping strategies or whether they can adjust their beliefs and the meaning they have made of their situation. Families who have high levels of conflict and dysfunction will find this process of adjustment difficult and may experience

prolonged crises (Patterson & Garwick, 1994). From this perspective, families of adolescents with IBD could result in a family facing on-going and repeated crises due to the chronic and unpredictable nature of the illness. However, an alternative view could be that families under these conditions will develop high resilience to on-going stressors (Patterson & Garwick, 1994). However, from this perspective, what is clear is the importance of viewing adolescent illness in the context of family functioning.

Family Functioning and IBD

Family functioning is an important area of research given that studies on adults with CD have found that they are more likely to report insecure attachment patterns than healthy peers (Agostini et al., 2010). At a time when young people could be developing independence, their health condition could result in the opposite; increased dependence on their families (Suris et al., 2004). Indeed, young people with IBD rely more heavily on a smaller number of people for social support and that social support is more commonly provided by family members than people outside of the family (Haller et al., 2003). Quality of life in young people with IBD has also been related to perceived social support and that social support is less likely to include non-family members than their peers (MacPhee et al., 1998). Further, this relationship is more prominent if diagnosis is recent (MacPhee et al., 1998).

However, our understanding of the impact of IBD on family functioning has been largely derived from studies of families of children and adolescents with chronic conditions rather than IBD specifically. However, Herzer et al. (2010) has highlighted that disease characteristics can influence the relationship between the illness and the impact on the family. Models of family life cycles and chronic illness have highlighted the onset, course and outcome of an illness as key to understanding the family response

to the illness (Rolland, 1987). Given some of the unique characteristics of IBD such as its relapsing and remitting nature, the idiosyncratic nature of how it impacts individuals physically, the difficulties with treatment and the challenges it poses to continuing with everyday life, it may be particularly challenging for a family to manage. Understanding the unique challenges that IBD poses to a family is key to developing effective support for families. Although research is emerging on the relationship between IBD and family functioning, this research has yet to be synthesised. Such a review would help to identify factors that may be pertinent in the development of support for families supporting a young person with IBD. The current review aims to address this by synthesising what is known about the relationship between IBD and family functioning as well as providing a commentary on the quality of the research used to examine this relationship.

Method

Defining the Question

The main topic of interest in the current review is family functioning in the context of IBD. The aim of the current review was to synthesise the literature exploring the concept of family functioning in the context of IBD. Thus, it was important that the terms used were broad enough to include all studies that included assessments of family functioning in IBD without assuming a direction or excluding studies that included assessments of family functioning as part of a broader aim. Using Shaw's (2010) CHIP framework the question was defined as follows:

Table 1. Question definition

Component	***Question***	***Search Terms***
Context	Inflammatory Bowel Disease	IBD, Inflammatory Bowel Disease
How	Observational	None specified
Issues	Family functioning	Family functioning, family, parents, siblings, social support, family relations, family members
Population	Children &Adolescents	Paediatric, pediatric

Search Strategy

Searches took place in September 2018. Potential studies for inclusion were identified primarily through searches in four databases; Web of Science, Medline, PsycInfo and Scopus (accessed via Ovid). In all databases the terms for the search string for the context was *paediatric OR pediatric AND "inflammatory bowel disease" OR IBD*. In all databases the search term for issues was "family functioning". In PsycInfo *"family functioning"* included mapped terms; *family relations, family, parents, social support, emotional adjustment, family members and "family functioning"* as a keyword. Terms were combined using OR. In Medline *"family functioning"* included the terms *family, family relations, parents, siblings, family health and "family functioning"* as a keyword.

Separate keyword searches were conducted in all databases. For both Medline and PsycInfo, separate searches were carried out for the context and issues and were then combined in the search history using AND. In Web of Science, the term '*paediatric*' was removed as this appeared to limit the results to just two articles.

Screening and Selection

Figure one below illustrates the screening and selection process for the current review. The searches returned a total of 1241 papers. 553 papers were duplicates which left 688 for screening. Initially screening of titles and abstracts resulted in a further 510 papers eliminated due to not being relevant to the aims of the review. The remaining 178

were then assessed in more detail and the inclusion/exclusion criteria were applied. An additional paper was identified through grey literature searches. At this stage the main reason for exclusion was not including a validated measure of family functioning. The studies meeting the inclusion criteria were included for review (N=18).

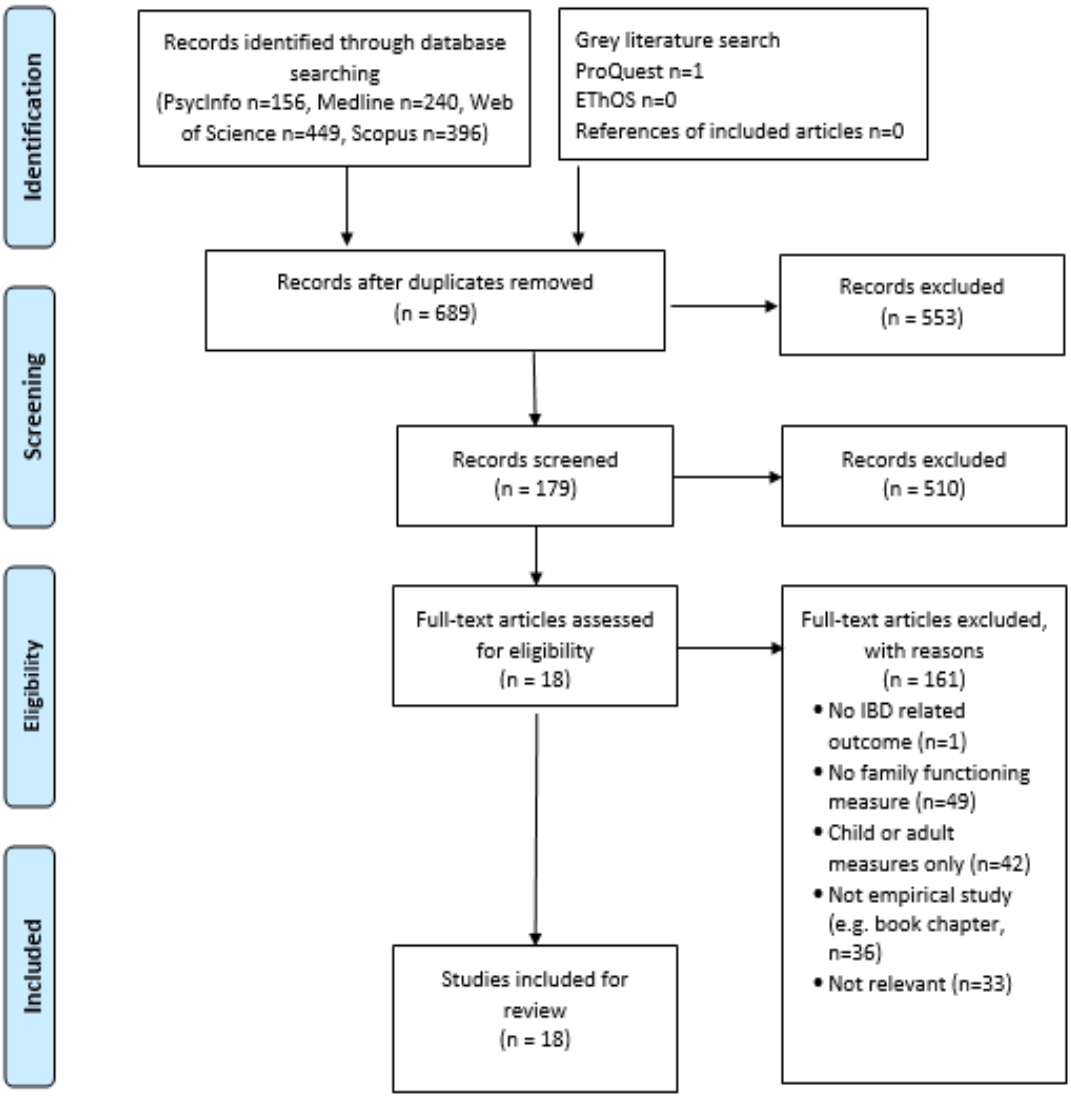

Figure 1. PRISMA Flow Diagram

Inclusion/Exclusion Criteria

An inclusion/exclusion criterion were applied to potentially relevant articles. Articles were included that fulfilled the following criteria:

- Observational studies

- Utilised a validated measure of family functioning
- The children/adolescents had a diagnosis of IBD (comorbid physical or mental health conditions were permitted)
- There was some measure of IBD activity/status

Articles were excluded if:

- The study design was not observational
- Participants were either adults or children/adolescents only
- The child/adolescent participants did not have a diagnosis of IBD
- There were no measures of IBD activity/status
- A validated measures of family functioning was not included

Quality appraisal

A common complaint when conducting systematic reviews is the lack of a quality appraisal tool specifically designed for appraising cross-sectional studies. Historically, studies wishing to appraise and synthesise literature from cross-sectional studies have relied on tools that have been developed for use with other research designs but can be used for multiple designs (Centre for Reviews and Dissemination, 2009). A commonly used tool in such cases is Downs and Black (1998) and this was considered for use in the current review. Downs and Black (1998) is a widely used tool which has been validated (Centre for Reviews and Dissemination, 2009; Research Triangle Institute, 2002).

The advantage of using Downs and Black (1998) is that it can be used to appraise multiple research designs in one review (Centre for Reviews and Dissemination, 2009). However, in the case of the current review, all except one study were cross-sectional thereby removing the advantage of using a multi-design tool and increasing the risks involved in using such tools; the use of multi-design tools has been criticised for being

imprecise (Research Triangle Institute, 2002). For this reason, a tool more appropriate for use with cross-sectional studies was sought. Unfortunately, very few tools have been designed specifically for appraising cross-sectional studies (Downes, Brennan, Williams & Dean., 2016).

Recently a tool has been developed for use specifically with cross-sectional studies. AXIS (Downes et al., 2016) was developed using research evidence, research experience and a multi-disciplinary Delphi process and is comparable to the development of other quality appraisal tools (Crowe & Sheppard, 2011). Quality appraisal of cross-sectional studies has been defined as high-functioning if it includes the following areas; comparability of subjects, exposure or intervention, outcome measurement, statistical analysis, and funding or sponsorship (Research Triangle Institute, 2002). AXIS addresses each of these issues (Downes et al., 2016). Further, many quality appraisal tools address either poor reporting or poor design quality but rarely both. One of the advantages of AXIS is that it addresses study design and quality as well as quality of reporting (Downes, et al., 2016).

It could be considered that a limitation of AXIS is that it does not provide a scoring system to quantify the quality of the studies being reviewed. However, using any appraisal tool still involves a degree of subjectivity and for this reason, the use of tools that assign scoring or categorisation of study quality has been advised against (Centre for Reviews and Dissemination, 2009). Further, another limitation of the AXIS tool is that is has not yet been validated. However, many widely used tools have not been validated (Centre for Reviews and Dissemination, 2009; Crowe & Sheppard, 2011; Research Triangle Institute, 2002) and this issue is therefore not exclusive to the AXIS tool. Considering the above, the AXIS tool was used in the current review (see appendix A).

Independent Verification of Quality Appraisal

When using a quality appraisal tool, a degree of subjectivity may influence ratings and therefore the synthesis of the review (Centre for Reviews and Dissemination, 2009). Consequently, a final year clinical psychology trainee repeated the quality appraisal on a sample of four of the included studies, chosen at random. There was substantial inter-rater agreement (kappa=.74, p=.001; Landis & Koch, 1977). There was a total of eight disagreements; two per study. The items where disagreements arose were different for each paper. Discussions were held until total agreement was reached over any discrepancies in quality ratings.

Results

Characteristics of studies and participants

All studies included in the current review were cross-sectional except one longitudinal study (Wojtowicz, 2014). Sample sizes varied, ranging from 15 to 183. Studies focused exclusively on IBD although one study included other long term paediatric conditions (Herzer et al., 2010). All studies specified that one caregiver also took part but did not specify which caregiver. Three studies specifically addressed mothers and/or fathers. The most commonly used measures of family functioning were the McMaster Family Assessment Device (n=9; FAD; Epstein, Baldwin, & Bishop, 1983) and the Paediatric Quality of Life Scale – Family Impact Module (n=5; PedsQL-FI; Varni, Seid, & Kurtin, 2001). Other studies utilised the Family Inventory of Life Events (n=4; FILE; McCubbin & Thompson, 1987). Child outcome measures varied however the most common IBD assessment tools were the PDCAI (Hyams et al., 1991) and the PUCAI (Turner et al., 2007) and the LCAI (Lichtiger et al., 1994). All studies included adolescents, but seven studies also included children; the mean age of participants ranged

from 11.6 years to 15.7 years. The majority of studies used a mix of statistical analyses and investigated both relationships as well as comparing differences (n=12) whilst the remaining studies looked at either relationships (n=3) or comparing differences (n=3). The studies are summarised in table three below.

Quality of the studies

The quality appraisal revealed that all studies were similar in terms of quality (see table two below). Studies had clear aims and objectives (n=17), used appropriate methods for the aims (n=17), had a clear target population (n=18), used an appropriate sampling frame (n=17), measured appropriate outcomes (n=17), used appropriate measures (n=18), clearly specified what statistical significance was based on (n=17), described the data adequately (n=18), had internally consistent results (n=18), presented data for all analyses (n=16), drew reasonable conclusions (n=18), acknowledged limitations (n=15), and addressed ethics and consent (n=17). Weaknesses in quality were similar across all papers; studies consistently failed to justify their sample size (n=17) and to provide information about non-responders (n=13).

Strengths and Limitations of Studies

Recruitment and Procedures

Studies consistently failed to justify their sample size. Most studies recruited through hospital outpatient clinics and the sampling method was to approach all eligible participants. As most studies were limited to one or two recruitment sites it is likely that sample sizes were determined by pragmatics rather than methodology. Only one paper commented on the statistical power of their study and it seems likely that many studies were underpowered limiting the ability of studies to detect a relationship between family functioning and IBD should one exist. The sample sizes varied greatly, and many

acknowledged the limitations of using a small sample size without justifying why it had been so.

Another common weakness across papers was that many papers did not included an account of how many potential participants were approached or how many were excluded for missing data. Similarly, even where it was clear that there were non-responders or those who declined to take part, most studies failed to describe who the non-responders were and address whether they were likely to be different from those who did respond. Generally, the description of recruitment across papers was poor and in certain studies it was impossible to tell whether the sampling procedure would have been resulted in sample bias. For example, one study merely combined data from other studies without full reporting of the different studies recruitment procedures. Another study recruited from a therapy trial without exploring whether this would have resulted in sample bias nor reporting the original recruitment procedure.

Participants

Studies recruited almost exclusively from outpatient clinics. It was noted during the review of the studies that the experience of young people with IBD who were in hospital was missing. Further, several studies acknowledged that their sample was biased towards less severely afflicted, thus the conclusions may not generalise to the whole population of young people with IBD.

Measures

Overall, most of the studies included the basic components of adequate research. Given that one of the inclusion criteria in the current review was to have used validated measures of family functioning, it is unsurprising that all of papers included for review used validated measures of family functioning and other variables of interest. However, where this becomes a weakness is the heavy reliance on self-reported measures; none of

the papers used an objective measure of family functioning and most of the other outcomes, such as child or parent mental health or quality of life, also relied on self-report measures.

Design

A strength of the studies was that they all chose appropriate designs to meet their aims. However, the abundance of cross-sectional designs reflects the studies available exploring family functioning and IBD. This limited variety in the study design could be considered a weakness as the pool of knowledge from which conclusions can be drawn is homogenous. This topic could be explored in depth through qualitative research. Further, given the relapsing and remitting nature of IBD, longitudinal studies would be ideally placed to explore whether the relationship between paediatric IBD and family functioning is static or changing.

Table 2. Summary of Quality Assessment

	Burke, Neigut, Kocoshis, Chandra, & Sauer (1994)	Engström (1991)	Giannakopoulos et al. (2016)	Gray, Graef, Schuman, Janicke, & Hommel (2013)	Gumidyala & Greenley, (2014)	Herzer et al (2010)	Herzer, Denson, Baldassano, & Hommel (2011)	Jelenova et al. (2015)	Jelenova et al. (2016)	Kunz, Greenley, & Howard (2011)	Mackner & Crandall (2006)	Odell, Sander, Denson, Baldassano & Hommell, (2011)	Reed-Knight, et al., 2018	Schuman, Graef, Janicke, Gray, & Hommel (2013)	Szajnberg, Krall, Davis, Treem & Hyams (1993)	Tojek, Lumley, Corlis, Ondersman, & Tolia (2002)	Werner et al. (2015).	*Wojtowicz* (2014)
Were the aims/objectives of the study clear?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✗	✓	✓	✓
Was the study design appropriate for the stated aim(s)?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✗	✓	✓	✓
Was the sample size justified?	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✗	✓
Was the target/reference population clearly defined?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓

	Burke, Neigut, Kocoshis, Chandra, & Sauer (1994)	Engström (1991)	Giannakopoulos et al. (2016)	Gray, Graef, Schuman, Janicke, & Hommel (2013)	Gumidyala & Greenley, (2014)	Herzer et al (2010)	Herzer, Denson, Baldassano, & Hommel (2011)	Jelenova et al. (2015)	Jelenova et al. (2016)	Kunz, Greenley, & Howard (2011)	Mackner & Crandall (2006)	Odell, Sander, Denson, Baldassano & Hommell, (2011)	Reed-Knight, et al., 2018	Schuman, Graef, Janicke, Gray, & Hommel (2013)	Szajnberg, Krall, Davis, Treem & Hyams (1993)	Tojek, Lumley, Corlis, Ondersman, & Tolia (2002)	Werner et al. (2015).	*Wojtowicz* (2014)
Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	?	✓	✓	✓	✓	✓	✓	✓
Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?	?	✓	✓	✓	✓	?	✓	?	?	✓	?	✓	✗	✓	✓	✗	✓	✓
Were measures undertaken to address and categorise non-responders?	?	✓	✓	✗	✓	✗	✗	?	?	✓	?	✗	✗	✗	✗	✗	✓	✗

	Burke, Neigut, Kocoshis, Chandra, & Sauer (1994)	Engström (1991)	Giannakopoulos et al. (2016)	Gray, Graef, Schuman, Janicke, & Hommel (2013)	Gumidyala & Greenley, (2014)	Herzer et al (2010)	Herzer, Denson, Baldassano, & Hommel (2011)	Jelenova et al. (2015)	Jelenova et al. (2016)	Kunz, Greenley, & Howard (2011)	Mackner & Crandall (2006)	Odell, Sander, Denson, Baldassano & Hommell, (2011)	Reed-Knight, et al., 2018	Schuman, Graef, Janicke, Gray, & Hommel (2013)	Szajnberg, Krall, Davis, Treem & Hyams (1993)	Tojek, Lumley, Corlis, Ondersman, & Tolia (2002)	Werner et al. (2015).	*Wojtowicz* (2014)
Were the risk factor and outcome variables measured appropriate to the aims of the study?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✗	✓	✓	✓
Were the risk factor and outcome variables measured correctly using instruments/measurements that had been trialled, piloted or published previously?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Is it clear what was used to determined statistical significance and/or precision estimates? (e.g. p-values, confidence intervals)	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✗	✓	✓	✓

	Burke, Neigut, Kocoshis, Chandra, & Sauer (1994)	Engström (1991)	Giannakopoulos et al. (2016)	Gray, Graef, Schuman, Janicke, & Hommel (2013)	Gumidyala & Greenley, (2014)	Herzer et al (2010)	Herzer, Denson, Baldassano, & Hommel (2011)	Jelenova et al. (2015)	Jelenova et al. (2016)	Kunz, Greenley, & Howard (2011)	Mackner & Crandall (2006)	Odell, Sander, Denson, Baldassano & Hommell, (2011)	Reed-Knight, et al., 2018	Schuman, Graef, Janicke, Gray, & Hommel (2013)	Szajnberg, Krall, Davis, Treem & Hyams (1993)	Tojek, Lumley, Corlis, Ondersman, & Tolia (2002)	Werner et al. (2015).	*Wojtowicz* (2014)
Were the methods (including statistical methods) sufficiently described to enable them to be repeated?	✗	✓	✓	✓	✓	✗	✓	✗	✗	✓	✗	✓	✓	✓	✗	✓	✗	✓
Were the basic data adequately described?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Does the response rate raise concerns about non-response bias?	?	✗	✗	✗	✗	✗	✗	?	?	✗	?	✗	✗	✗	✓	✗	✗	✗
If appropriate, was information about non-responders described?	?	✗	✓	✗	✗	✗	✗	?	✗	✓	✗	✗	✗	✗	✗	✗	✓	✗
Were the results internally consistent?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Were the results presented for all the analyses described in the methods?	?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	?	✓	✓	✓

	Burke, Neigut, Kocoshis, Chandra, & Sauer (1994)	Engström (1991)	Giannakopoulos et al. (2016)	Gray, Graef, Schuman, Janicke, & Hommel (2013)	Gumidyala & Greenley, (2014)	Herzer et al (2010)	Herzer, Denson, Baldassano, & Hommel (2011)	Jelenova et al. (2015)	Jelenova et al. (2016)	Kunz, Greenley, & Howard (2011)	Mackner & Crandall (2006)	Odell, Sander, Denson, Baldassano & Hommell, (2011)	Reed-Knight, et al., 2018	Schuman, Graef, Janicke, Gray, & Hommel (2013)	Szajnberg, Krall, Davis, Treem & Hyams (1993)	Tojek, Lumley, Corlis, Ondersman, & Tolia (2002)	Werner et al. (2015).	*Wojtowicz* (2014)
Were the authors' discussions and conclusions justified by the results?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓
Were the limitations of the study discussed?	×	×	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	×	✓	✓	✓
Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?	×	×	×	✓	×	✓	✓	×	×	×	×	✓	×	✓	?	×	×	×
Was ethical approval or consent of participants attained?	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	✓	?	✓	✓	✓

✓ = Yes, × = No, ? = not stated

Synthesis

Whilst most studies looked directly at family functioning in the context of IBD (n=13) in other studies family functioning was embedded as part of a wider investigation into family life and IBD such as child or parental mental health (n=3) or quality of life (n=2). The following is a synthesis of what the studies discovered about family functioning in the context of IBD.

Family Functioning in the context of youth IBD

Some studies simply eluded to the presence of family stress in the context of youth IBD. One study reported that family stress was present in 55% of families with a young person with IBD (Werner et al., 2015). However, Herzer et al. (2011) found only 25% of families of youth with IBD reported clinically significant dysfunction.

Looking more directly at the relationship between family functioning and IBD, the findings are mixed. Three studies found a relationship between family functioning and IBD. Szajnberg, et al. (1993) found that family stress was related to higher disease severity and marital problems. Reed-Knight et al. (2018) found a positive relationship between family dysfunction and youth pain and passive coping. This relationship was mediated by parental depression and passive coping. Additionally, Tojek et al. (2002) found that family dysfunction was related to a higher frequency of adolescents' bowel movements which the authors suggest point to poorer physiological functioning. Kunz et al. (2011) found a partial relationship which suggested a possible difference in the relationship depending on the gender of the caregiver. They found that disease severity was only significantly related to family functioning as reported by fathers. For mothers, youth emotional and behavioural symptoms were better predictors of family functioning.

Four studies did not find a significant relationship between family functioning and IBD. Schuman et al. (2013) found no significant relationship between family functioning

and IBD disease severity, however family affective involvement predicted parent-reported youth depression whereas family problem solving significantly predicted youth self-reported symptoms of depression. These findings suggest that family functioning may be more important for a young person's emotional health than IBD severity. Similarly, Wojowicz (2014) found no significant relationship between family functioning and youth abdominal pain. Although not significant, Giannakopoulos et al. (2016) found a non-significant trend towards higher levels of family dysfunction for families of young people with active IBD compared to those in remission. Similarly, Odell et al. (2011) again found non-significant associations between family functioning and IBD. Instead, youth externalising behaviour was a better predictor of family dysfunction.

Although findings were mixed, four studies suggested that family functioning was not significantly worse in the context of IBD compared to other conditions or controls. Jelenova et al. (2016) found no parent-reported differences in functioning between families with and without IBD. Herzer et al. (2010) found no significant differences in family dysfunction across five paediatric conditions. Family dysfunction was better predicted by family characteristics such as children being older, than by health condition. Further, Mackner and Crandall (2006) found no difference in family functioning between families with and without IBD. Additionally, IBD severity was not significantly related to family dysfunction. However, Engström (1991) found that family dysfunction was reported higher by mothers of young people with IBD than mothers of healthy controls, children with diabetes or fathers of young people with IBD. Similarly, Jelenova, et al. (2015) found that parents of young people with IBD reported poorer functioning than parents of children without IBD.

Of course, the mixed findings could be due to methodology and study quality; many of the papers recruited participants with lower disease severity and from out-patient

clinics. Similarly, all papers relied on self-report measures of family functioning limiting the reliability of the findings. Further, the cross-sectional nature of the studies means that studies do not capture any potential changes in family functioning over time, nor can issues of causation be addressed.

Mental Health

Family functioning and IBD was examined by three studies investigating parent or child mental health. Two studies suggest a possible link between IBD, family functioning and poor mental health. Gray et al. (2013) found that parents experienced higher stress when family functioning was poor and when their child experienced higher internalising symptoms and active CD. Szjanberg et al. (1993) found prevalence of psychiatric disorders in 11 of 15 youth with IBD as well as parent reported attachment difficulties. However, interestingly, Burke et al. (1994) found that young people with IBD who were more severely depressed were also more likely to have less severe IBD but more family dysfunction.

Quality of Life

Studies looking at quality of life in the context of IBD and family functioning found that family functioning may affect quality of life. Gumidyala and Greenley (2014) found that of a range of variables including gender, age, disease severity, socioeconomic status, anxiety and depression, poorer family functioning and lower socioeconomic status were the most consistently associated with both adult and youth reported measures of quality of life explaining up to 19% of the variance in all quality of life measures. Similarly, although Herzer and colleagues (2011) found no significant relationship between family dysfunction and IBD, they did find that regardless of disease severity or type, young people with less well-functioning families reported the poorest quality of life.

Further, Kunz et al. (2011) found that higher disease severity was related to poorer parental quality of life and poorer family functioning as reported by fathers.

In summary, although some studies suggested that families of youth with IBD do report dysfunction, this was not the case for the majority of families. Taken together the studies included in the current review suggest that a strong relationship between family functioning and IBD is unlikely, with only a minority of studies finding a significant link and studies lacking high quality recruitment. However, a stronger finding was that poor family functioning was no more likely to be poor for families of youth with IBD than for families with other chronic health conditions or families of healthy children. However, when family functioning is low it seems that it does affect the quality of life for young people with IBD and their parents. Further, the presence of psychological problems may result in more family stress.

Table 3. Summary of included papers

Author (year)	***Participants***	***Comparison Group***	***Family/Parent Measures***	***Child/Adolescent Measures***	***Statistical Analyses***	***Key Findings***
Burke et al. (1994)	IBD, Children and Adolescents, n=36 (mean age 11.98 years) and caregiver	None	A-SADS-L FRI FILE	Kiddie-SADS-E Gastroenterologist rating of IBD severity.	Non-parametric tests of significance	Children with higher levels of depression were more likely to have less severe IBD, a parent with higher levels of depression and a higher level of conflict and lower cohesion within the family.
Engström (1991)	IBD children and adolescents n=20 (mean age 16.5 years)	Age matched healthy controls n=20 and children with diabetes millitus n=20	FACES	LOCS CAS	Non-parametric analysis of variance and correlation	Mothers of children with IBD reported significantly more family dysfunction than mothers of children with diabetes or healthy controls. Fathers of children with IBD showed a similar pattern of family dysfunction but this was not significant.
Giannakopoulos et al. (2016)	IBD, children and adolescents, n=85 (mean age 13.2 years) and caregiver	None	FAD CLES SCL-90-R	CDI PCDAI PUCAI RCMAS	Parametric tests of significance, non-parametric tests of significance, correlations and regressions	Parent reported life events, child reported anxiety and parent mental health were predictive of disease activity. There was a trend towards higher levels of family dysfunction in children with active disease compared to those in remission, but this was not significant.
Gray et al. (2013)	IBD, Adolescents, n=130 (mean age 15.64 years) and caregiver	None	PIP FAD	CBCL Short PCDAI LCAI	Parametric tests of significance, analysis of variance and correlations	Parenting stress was significantly higher amongst those with poorer family functioning, higher child internalising symptoms and more active Crohn's disease
Gumidyala &Greenley, (2014)	IBD, Adolescents, n=50 (mean age 15 years) and caregiver	None	PedsQL-FI FAD	RCADS PGA PedsQL 4.0	Parametric correlations	Family functioning and lower socioeconomic status most associated with poorer quality of life.
Herzer et al. (2010)	Five chronic paediatric conditions n=103 (IBD n=43, mean age 15.4 years) and caregiver	Healthy comparison group n=57	FAD	Medical history.	Parametric tests of significance, multivariate analysis of covariance and correlations. Non-parametric tests of significance	No significant group differences in family functioning. Across groups, poorer family functioning associated with children being older, lower household income and fewer children in the family.

Author (year)	***Participants***	***Comparison Group***	***Family/Parent Measures***	***Child/Adolescent Measures***	***Statistical Analyses***	***Key Findings***
Herzer et al. (2011)	IBD, adolescents, n=62 (mean age 15.5 years), caregiver	None	FAD	PDCAI LCAI IMPACT-III	Parametric tests of significance and multivariate analysis of variance	Controlling for disease severity and diagnosis, adolescents from poorer functioning families experience the poorest health related quality of life.
Jelenova et al. (2015)	IBD, adolescents n=29 (mean age 15.03 years) and caregiver	Healthy controls n=40 (mean age 14.86 years)	BDI-II BAI PedsQL-FI	CDI SAD KidScreen-10 PUCAI PCDAI	Parametric tests of significance and correlations. Non-parametric tests of significance and correlations.	Parents of children with IBD reported significantly poorer quality of life, higher levels of depression and anxiety and poorer parental functioning than parents of children without IBD.
Jelenova et al. (2016)	IBD, adolescents, n=27 (mean age 15.1 years) mothers and fathers.	Healthy adolescents n=39 (mean age 14.8)	BDI-II BAI PedsQL-FI	PUCAI PDCAI KidScreen-10 CDI SAD	Parametric tests of significance and correlations. Non-parametric tests of significance and correlations	There were no significant differences in parent functioning of parents with or without IBD. Child reported quality of life was similar between children with and without IBD. Quality of life was significantly lower for parents of children with IBD.
Kunz et al. (2011)	IBD, adolescents, n=95 (mean age 15.07 years), mothers and fathers	None	PedsQL-FI	PGA YR-PSC	Parametric tests of significance, correlations and regression	Higher disease activity was related to lower maternal and paternal reported quality of life however it was related to lower family quality of life for fathers only. Youth emotional and behavioural adjustment was a better predictor of maternal reported family functioning.
Mackner & Crandall (2006)	IBD, adolescents n=50 (mean age 14.39 years) and caregiver	Healthy comparison group, n=42 (mean age 14.39 years).	FAD	CBCL PCDAI	Parametric and non-parametric tests of significance	Illness severity was not significantly associated with family functioning, behavioural and emotional functioning, or social functioning. Family functioning was similar between adolescents with and without IBD.
Odell et al. (2011)	IBD, adolescents n=45 (mean age 15.41 years) and caregiver	None	PIP FAD	CBCL CDI PCDAI LCAI	Parametric tests of significance, correlations and regressions.	Correlations between family functioning and disease severity, parent distress, socioeconomic status and child depression were non-significant. Adolescent behavioural problems, particularly parent reported externalising, better predicted family functioning.
Reed-Knight et al. (2018)	IBD, children and adolescents n=183	None	FILE	PBCL PRI	Parametric correlations and mediation analysis	As family stress increased, children reported more pain and passive coping. This relationship was mediated by

Author (year)	***Participants***	***Comparison Group***	***Family/Parent Measures***	***Child/Adolescent Measures***	***Statistical Analyses***	***Key Findings***
	(mean age 13.75 years) and caregiver			CDI PCDAI PUCAI		parent reported passive coping and symptoms of depression.
Schuman et al. (2013)	IBD, adolescents n=122 (mean age 15.7 years) and caregiver	None	FAD	CBCL LCAI PCDAI	Parametric tests of significance, analysis of variance, correlations, regressions and moderation analysis	No significant relationship between family functioning and IBD severity. Family affective involvement predicted parent reported youth depression and family problem solving predicted youth self-reported depressive symptoms.
Szajnberg et al. (1993)	IBD, children and adolescents n=15 (mean age 11.6 years) and caregiver	None	AAI MCMI FILE IOF COBI Locke-Wallace Marital Scale	Kiddie-SADS-P CBCL Irvin Sentence Completion WISC-R	Parametric tests of significance and correlations	13 of the parents were categorised as insecurely attached to their children. Marital problems and disease severity correlated with family stress.
Tojek et al. (2002)	IBD, adolescents n=62 (mean age 15.1 years) and mothers	None	FAD PILL PANAS	CDL FDI	Parametric tests of significance, correlations and regressions. Non-parametric tests of significance	Family dysfunction was significantly related to more frequent adolescent bowel movements. Higher maternal positive affect was related to lower adolescent depression and lower functional disability
Werner et al. (2015)	IBD, children and adolescents n=125 (mean age 13.3 years) and caregiver	Age matched healthy controls	SCL-27 FILE	SDQ	Parametric tests of significance. Non-parametric tests of significance.	Significant family stress reported in 55% of families of children with IBD
Wojtowicz (2014)	IBD, children and adolescents n=76 (mean age 14.3 years) and caregiver	None	PedsQL-FI	PGA FDI	Parametric regression and mediation analysis.	Family functioning was not predictive of abdominal pain

A-SADS-L = Adult Schedule for Affective Disorders and Schizophrenia, Lifetime Version (Mannuzza, Fyer, Klein, & Endicott, 1986). FRI = Family Relationship Index Scale (Hoge, Andrews, Faulkner, & Robinson, 1989). FILE = Family Inventory of Life Events (McCubbin & Thompson, 1987). Kiddie-SADS-E = The Kiddie Schedule for Affective Disorders and Schizophrenia, Epidemiologic Version (Orvaschel, & Puig-Antich, 1987). PIP = The Pediatric Inventory for Parents (Streisand, Braniecki, Tercyak, & Kazak, 2001). FAD = The McMaster Family Assessment Device (Epstein, Baldwin, & Bishop, 1983). CBCL = Child Behavior Checklist (Achenback, 1991).

Short PCDAI = The Short Pediatric Crohn's Disease Activity Index (Hyams et al., 1991). LCAI = Lichtiger Colitis Activity Index (Lichtiger et al., 1994). PedsQL-FI = Pediatric Quality of Life Inventory Family Impact Module (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004). RCADS = Revised Children's Anxiety and Depression Scale (Chorpita, Ebesutani, & Spence, 2015). PGA = Physician Global Assessment (Hanauer et al., 1993). PedsQL 4.0 = Pediatric Quality of Life Inventory Generic Core Scale (Varni, Seid, & Kurtin, 2001). IMPACT=III = IMPACT-III Health Related Questionnaire (Otley, Xu, Yan, Olson, Liu, Griffiths, & REACH Study Group, 2006). YR-PSC = Youth Report Pediatric Symptom Checklist (Jellinek, Murphy, & Burns, 1986). CDI = Children's Depression Inventory (Kovacs, 1992). PBCL = Pain Behaviour Checklist (Kerns et al., 1991). PRI = Pain Response Inventory (Walker et al., 1997). PUCAI = Pediatric Ulcerative Colitis Activity Index (Turner et al., 2007). AAI = Adult Attachment Interview (George, Kaplan, & Main, 1996). MCMI = Millon Clinical Multi-Axial Inventory (Millon, 1997). IOF= Impact on Family Scale (Stein & Reissman, 1980). COBI = Clinician's Objective Burden Index (Stein & Jessop, 1982). Locke-Wallace Marital Scale (Locke & Wallace, 1959). Irvin Sentence Completion (Irvin, 1972). WISC-R = Weschler Intelligence Test for Children-Revised (Weschler, 1974). PILL = Pennebaker Inventory of Limbic Languidness (Pennebaker, 1982). PANAS = Positive and Negative Affect Schedule (Watson, Clark, & Tellegen, 1988). FDI = Functional Disability Index (Walker & Greene, 1991). BDI-II = Beck Depression Inventory II (Beck, Steer, & Brown, 1996). BAI = Beck Anxiety Inventory (Steer & Beck, 1997). SAD = Scale of Anxiety in Children (Müllner et al., 1983). KidScreen-10 (Ravens-Sieberer, Erhart, Rajmil, Herdman, Auquier, Bruil,... & Mazur, 2010). CLES = Coddington's Life Events Questionnaire (Coddington, 1972). RCMAS = Revised Children's Manifest Anxiety Scale (Reynolds & Richmond, 1985). SCL-90-R = Symptom Checklist 90 Revised (Derogatis, 1977). CAS = Child Assessment Schedule (Hodges et al., 1981). FACES = The Family Adaptability and Cohesion Evaluation Scale (Olsen, 1986). SCL-27 = Symptom Checklist 90 Revised Short Version (Hardt, & Gerbershagen, 2001). SDQ = Strengths and Difficulties Questionnaire (Goodman, 1997).

Discussion

The current review aimed to synthesise what is known about the functioning of families who include a young person with IBD. Eighteen papers were reviewed. Overall, the papers met most criteria using the AXIS quality appraisal tool. However, the weakness of the studies lay in their recruitment and participants. Studies generally failed to give enough detail around their recruitment procedures, failed to justify their sample sizes, relied heavily on outpatient sites at the exclusion of community or inpatient sites. Further, details about non-responders were often omitted making it unclear whether studies had non-responders and if so, whether they differed from participants. This raises questions about how applicable findings are to all young people with IBD and their families. Studies relied heavily on cross-sectional designs which in the context of a relapsing and remitting condition such as IBD may not be as helpful as longitudinal or qualitative designs. Readers may wish to be mindful of this when considering the findings.

Overall, studies failed to find a consistent and clear link between IBD and family dysfunction. Despite facing significant challenges and burdens it seems that families of young people with IBD are no more likely to experience family dysfunction than other families. However, when considering the literature, this finding may not be so surprising. Models of family functioning suggest that families can be well equipped to deal with difficulties such as chronic illness. The FAAR model (Patterson, 1988) holds that crises can be an opportunity for families to develop strengths and coping strategies. It may be that the presence of a chronic stressor can strengthen families. Indeed, Knafl et al. (2013) have argued that the majority of families supporting a young person with a health condition are able to adjust. The idea of family resilience suggests that whilst some

families do suffer in the face of a crisis such as chronic illness others emerge stronger and better able to cope (Walsh, 2003). The current studies incorporate both possibilities.

Although not a majority, there was a sizable proportion of the studies included for review that did find a relationship between family functioning and IBD. It may be that the if there been better quality recruitment, more studies would have found a relationship. Indeed, Szajnbern et al. (1993) found higher disease severity and marital problems were related to higher family stress. Two studies suggested family dysfunction was higher for families of young people with IBD than families without (Engström, 1991; Jelenova et al., 2015). Similarly, two studies suggested that family dysfunction affected IBD outcomes in the young person (Reed-Knight et al., 2018; Tojek et al., 2002). Further, two studies that did not find a significant relationship did find trends towards higher levels of dysfunction in families of young people with IBD (Giannakopoulos et al., 2016; Odell et al., 2011). It is therefore difficult to draw conclusions from the current review in either direction.

One explanation for the inconsistent findings may be due to a lack of a developmental perspective in the available studies. Most would agree that families are well equipped to deal with single stressors (Walsh, 2003) however when faced with ongoing and repeated stress, even well-functioning families may begin to struggle. Further, a family life-cycle perspective would suggest that the timing of the stresses may be important. Families experience stress during predictable or unpredictable events (Carter & McGoldrick, 1988). For example, a transition such as a child moving from primary to secondary school, whilst predictable, remains a stressor to a family just as the death of a family member is unpredictable and stressful (Carter & McGoldrick, 1988). At times of transition a family may be less able to handle the unpredictable and changing nature of a health condition such as IBD. To better understand the relationship between

family functioning and IBD it seems essential to conduct more developmentally sensitive, longitudinal research. This echoes criticism posed to family research generally, which some argue is too inflexible to fit with the complexities of family relationships (Walsh, 2003).

The studies eluded to the importance of family functioning for health-related quality of life and mental health. So perhaps whilst it remains unclear whether family functioning is important for physical health, it may be clearer that well-functioning families are necessary for good quality of life and mental health for young people with IBD. Two studies found that youth health-related quality of life was predicted by family functioning (Gumidyala & Greenley, 2014; Herzer et al., 2011). Further, Szjanberg et al. (1993) found that psychiatric disorders were prevalent in an, albeit small, sample of youth with IBD. Further there was evidence of parent reported attachment difficulties. This would fit with research which has consistently found that young people with IBD may be at risk for developing mental health problems (Lavinge & Faier-Routman, 1992). Additionally, Gray et al. (2013) found that active CD, high levels of child internalising symptoms and poor family functioning were significant predictors of high parenting stress. This would help explain the established link between parental burn out and child chronic illness (Lindström et al., 2010). It may be that if families become unable to continue defending against the onslaught of stressors in the face of chronic illness, parental or youth mental health may be at risk.

Limitations and future directions

The current review should be considered in light of several limitations. The studies included were most likely underpowered and were therefore unlikely to find a relationship should one exist. Further, the quality appraisal tool used in the current review has yet to be validated. Although it may have been better to use a validated tool, and this is certainly

recommended in the literature (Centre for Reviews and Dissemination, 2009), to the researcher's knowledge, no validated tool currently exists specifically for use on cross-sectional studies.

Further, the current review did not specify inclusion criteria that studies primarily investigate the relationships between IBD and family functioning. This was due to concerns that doing so would have been too restrictive and could have resulted in missed valuable information about family functioning and IBD that could be extracted from studies looking at these issues from a different angle. But this does mean that comparisons across studies is difficult as the studies differed in terms of their primary aim, outcomes used and the analyses they conducted. In time, perhaps this review could be repeated with enough literature available to look more directly and exclusively at the relationship between IBD and family functioning. Additionally, the studies included in the current review were looking for family weaknesses rather than looked at what made families strong and resilient. This is a criticism of paediatric health research in general which typically focuses on the negative aspects of illness (Tedeschi & Calhoun, 2004). Future reviews may wish to focus on the positive aspects of families managing childhood illness. Indeed, this may more easily translate to clinical practice where learning from what makes families resilient may be used to support those who are struggling.

Additionally, the broad definition of family functioning meant that papers that exclusively explored siblings were excluded. Although these studies would not have added to the understanding of the whole family in the context of IBD, it means that the voice of siblings may have been overlooked. Being a sibling of a child with IBD is likely to bring unique challenges that would may influence the functioning of a family system (Mamula et al., 2003). However, only two studies were found in the current searches and addressed siblings' quality of life rather than siblings' perspectives of family functioning.

Further, there was little emphasis in the studies on the differences between male and female caregivers. One study eluded to differences in how mothers and fathers perceive family functioning; Engström (1991) found that levels of family dysfunction were higher in families of young people with IBD compared to families of young people with type one diabetes for mothers only and, whilst there was a trend, this finding did not reach significance for fathers. Consequently, future reviews of family functioning may wish to attempt to explore individual family members perspectives.

Implications for Clinical Practice

The results of the current review are unable to firmly conclude whether families of young people with IBD will be at risk of family dysfunction, but equally, the findings cannot rule it out. Given that in some studies family dysfunction was related to youth IBD outcomes, it certainly suggests that the wider family context should be considered by services supporting young people with IBD to improve quality of life and possibly even physical health. If the criticism posed to family research is correct, that assessments tools such as those used in the current review only assess a snapshot of family functioning (Walsh, 2003), then clinicians may be wondering how best to assess family functioning. Indeed, the use of such tools may have contributed to the mixed findings of the current review. Whilst the answer to this question is beyond the scope of this review, Walsh (2003) advises using a family resilience perspective which is underpinned by the expectation that there will be no one model to understand families and that assessments of family functioning should include families' values, beliefs, structures and resources as well as past, present and potential future events.

Conclusion

Family functioning is a dynamic, complex concept (Walsh, 2003). When exploring families of young people with IBD it seems unlikely that a simple explanation

will arise and there are likely many other factors such as family events, mental health and quality of life that would need to be considered to make sense of any relationship. However, the first step should be to establish a consistent link; is family dysfunction more likely in families of youth with IBD and if so, does it influence the young person's physical health? Such a relationship between family functioning and youth IBD, at least according to the findings of the current review, has yet to be found in a robust way. This may be addressed in future research by designing studies with broader and more thoughtful recruitment strategies and incorporating a developmental perspective. Alternatively, it may the case that when it comes to family functioning in the context of IBD, no one size fits all.

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Appendices

Appendix A. AXIS Quality Appraisal Tool

Appraisal of Cross-sectional Studies

	Question	Yes	No	Don't know/ Comment
Introduction				
1	Were the aims/objectives of the study clear?			
Methods				
2	Was the study design appropriate for the stated aim(s)?			
3	Was the sample size justified?			
4	Was the target/reference population clearly defined? (Is it clear who the research was about?)			
5	Was the sample frame taken from an appropriate population base so that it closely represented the target/reference population under investigation?			
6	Was the selection process likely to select subjects/participants that were representative of the target/reference population under investigation?			
7	Were measures undertaken to address and categorise non-responders?			
8	Were the risk factor and outcome variables measured appropriate to the aims of the study?			
9	Were the risk factor and outcome variables measured correctly using instruments/measurements that had been trialled, piloted or published previously?			
10	Is it clear what was used to determined statistical significance and/or precision estimates? (e.g. p-values, confidence intervals)			
11	Were the methods (including statistical methods) sufficiently described to enable them to be repeated?			
Results				
12	Were the basic data adequately described?			
13	Does the response rate raise concerns about non-response bias?			
14	If appropriate, was information about non-responders described?			
15	Were the results internally consistent?			
16	Were the results presented for all the analyses described in the methods?			
Discussion				
17	Were the authors' discussions and conclusions justified by the results?			
18	Were the limitations of the study discussed?			
Other				
19	Were there any funding sources or conflicts of interest that may affect the authors' interpretation of the results?			
20	Was ethical approval or consent of participants attained?			

Section Two. Research Report

The Experience of Post Adversarial Growth in Young People with Inflammatory Bowel Disease

Abstract

Objectives. Post-adversarial growth (PAG) refers to a deep and personal transformation following adversity. Previous research has questioned whether young people can experience PAG. The aim of the current study was to explore whether young people with IBD experience PAG as a result of their illness and in what way they experience PAG.

Method. Fourteen young people with IBD aged between 16 and 25 were recruited through research groups, charities and social media. Participants took part in semi-structured interviews exploring their experiences of IBD and in what ways they felt they had experienced PAG. Interviews were transcribed and analysed using Template Analysis.

Results. The final template revealed several strong themes reported by participants. The main themes were *'initiation in the world of IBD'*, *'a life on hold'*, *'standing up to the illness'*, *'love surrounding pain'* and *'active acceptance'*. Results suggested that participants experiences were consistent with the concept of PAG and mirrored previous research suggesting that PAG involves a combination of social and individual factors.

Conclusions. Young people with IBD may be able to experience PAG, particularly when well supported and able to develop ways to regain control over their

lives. PAG in youths with IBD may involve increased compassion for others, new directions and positive psychological changes.

Practitioner Points.

- Openness in relationships may be particularly valuable for promoting PAG in youths with IBD and therefore positive adjustment to their illness. Practitioners working with young people with IBD may wish to look for ways to open lines of communication for their patients or support them to talk openly with trusted members of their social support networks.
- Compassion focussed interventions may help promote the development of PAG in youths with IBD, although this requires further research.
- Youths with IBD should be encouraged to make decisions and take control over their condition where possible and appropriate. Practitioners working with youths with IBD may wish to support patients to set and achieve goals to increase perceived control.

Limitations.

- The current study is limited by a small sample size, a homogenous sample, the absence of an objective measure of PAG and by not considering the influence of time since diagnosis. This limits the generalisability of the findings.

Introduction

Inflammatory Bowel Disease (IBD)

Inflammatory Bowel Disease (IBD) is a term that incorporates two main illnesses; Crohn's Disease (CD) and Ulcerative Colitis (UC; Sirois & Hirsch, 2017) and is characterised by periods of remission and periods of painful flare-ups, including symptoms such as stomach cramps, weight loss, diarrhoea, nausea and fatigue (Purc-Stephenson, Bowlby, & Qaqish, 2015). Being a lifelong condition, the severity can vary and fluctuate greatly between individuals which is hard for medical professionals to predict, meaning patients are often uncertain of their prognosis. Treatment through medication can involve severe side effects (de Rooy Toner et al., 2001). In extreme cases patients undergo surgery to create a temporary or permanent external pouch for waste. Not surprisingly, studies have documented the negative impact of living with IBD. One qualitative study found that 80% of adults with IBD reported that their condition resulted in social isolation and constraints to life (Purc-Stephenson et al., 2015).

IBD in Young People

Adolescence may be a particularly difficult time to live with the consequences of IBD. IBD can be associated with malnutrition and consequently slowed puberty and growth (Suris, Michaud, & Viner, 2004). A young person who experiences delayed puberty may experience low self-esteem and infantilization (Suris et al., 2004). Whereas some young people report enhanced coping when receiving social support, other young people experienced complications in their relationships with their parents, such as parents worrying about them, being intrusive, or not understanding the illness (McCombie, Mulder, & Gearry, 2013). Additionally, young people with chronic conditions may have

poorer educational attendance than their peers without a chronic condition through severity or treatment demands (Suris et al., 2004).

Additionally, adolescence is generally associated with greater risk of developing mental health conditions therefore the risks for a young person with IBD could be higher. Neuendorf, Harding, Stello, Hanes and Wahbeh (2016) found evidence from a systematic review that anxiety and depression are higher for patients with IBD than in healthy controls. Further, anxiety and depression were higher with active disease than in remission. Greenley et al. (2010) found that parents and youth with IBD reported lower quality of life compared to youth without IBD. This combination of biological and psychosocial factors can leave young people with chronic conditions feeling a sense of being different or feeling isolated from their peer groups (Suris et al., 2004).

Unsurprisingly, both biological and psychosocial factors have found to be important predictors of health-related quality of life (Santos, Gaspar de Matos, Marques, Simeos, Leal, & Machado, 2017). In a review of literature, Jordan, Sin, Fear and Chalder (2016) found several studies demonstrating that psychological factors such as perceived stress were associated with negative outcomes in IBD but social support may mitigate the relationship between stress and poorer mental health in IBD. However, they noted that most studies focus on factors that are associated with negative outcomes in IBD and suggest future research should also investigate the factors associated with positive outcomes in IBD. Indeed, whilst a qualitative study found that 80% of adults with IBD reported that the condition negatively affected their lives in some way, positive changes were also frequently recognised such as new life paths, personal growth and valuing life (Purc-Stephenson et al., 2015).

Post-adversarial Growth (PAG)

Clearly, highly stressful situations can and do cause distress and understandably the bulk of research on psychological consequences of stressful events focuses on the negative consequences (Tedeschi & Calhoun, 2004). However, although positive consequences of stressful events may not be experienced by all, there is evidence that stressful events offer the possibility for positive change (Tedeschi & Calhoun, 2004).

This positive change is often referred to as Post Adversarial Growth (PAG), benefit finding, posttraumatic growth, positive-growth, or stress-related growth. These terms refer to the reconceptualization of fundamental beliefs about the self, others and the future (Aspinwall & Tedeschi, 2010). PAG differs from concepts such as resilience in that it requires a significant personal transformation following adversity rather than resisting or adapting to it (Aspinwall & Tedeschi, 2010). PAG involves viewing the stressful event in a positive way because of perceived benefits (Helgeson, Reynolds, & Tomich, 2006). PAG is a concept that reaches far back in history; it has been documented following catastrophic and traumatic events such as terrorist attacks, war, cancer, bereavement and natural disasters but has only relatively recently received attention in research (Helgeson et al., 2006). Further, less is known about PAG in the context of health conditions compared to other traumatic events (Hefferon, Grealy, & Mutrie., 2009). One meta-analysis investigated the relationship between benefit finding and psychological and physical health outcomes. The results of 87 studies suggested that PAG was related to lower levels of depression and higher positive well-being. Interestingly, it was also related to more intrusive thinking and avoidant thoughts. The authors suggest that whilst initially counter-intuitive, this reflects an attempt to make sense of events rather than a marker of deteriorating mental health (Helgeson et al., 2006). Indeed, much of the literature suggests that the psychological nature of PAG can be both positive and negative simultaneously

(Soltani, Neville, Hurtubise, Hildenbrand., & Noel, 2018). Considering research that associates PAG with poorer psychological functioning, it has been argued that distress may be a prerequisite of PAG or necessary for its maintenance (Kilmer, Gil-Rivas, Griese, Hardy, Hafstad, & Alisic., 2014).

PAG in Youth

Whilst PAG has been well documented in adults, there is less research on the experiences of PAG in the young (Kilmer et al., 2014). The above description of PAG suggests the presence of fully formed cognitive schemas. Thus, there may be an assumption that young people will not experience PAG or that it is more applicable to adults as opposed to young people (Tedeschi & Calhoun, 2004). However, a small body of literature suggests PAG can occur in young people. For example, assessing PAG with the Benefit Finding Scale for Children (Phipps, Long, & Ogden, 2007), Rassart, Luyckx, Berg, Oris, and Wiebe (2017) found evidence of PAG in young people with type-one diabetes. Although PAG fluctuated over a six-month period in some, those that reported more stable PAG also reported higher diabetes self-care at a six-month follow-up. This suggests that PAG may better health protection in youths with chronic conditions. Further, a meta-analysis of PAG of young people with serious illnesses found that PAG does occur in young people as measured by the Post Traumatic Growth Inventory (PGTI; Tedeschi & Calhoun, 1996) and may include greater appreciation of life, improved relationships or recognition of new paths in life (Picoraro, Womer, Kazak, & Feutdner, 2014). Individual characteristics such as optimism, being older and more social support increased the likelihood of PAG (Picoraro et al., 2014). Further, cognitive processes such as rumination were linked to a higher likelihood of PAG (Picoraro et al., 2014). Interestingly, PAG appears more likely when the stressful event creates a certain degree of emotional distress (Picoraro et al., 2014).

However, despite the small number of studies of PAG in young people, PAG continues to be poorly defined and how PAG may evolve in young people is yet to be fully explored. Further, a large portion of the literature on PAG has utilised cross-sectional, questionnaire-based research (Jayawickreme & Blackie, 2014). Some have questioned whether such studies are genuinely finding instances of PAG, as questionnaires may be priming the participants responses (Jayawickreme & Blackie, 2014). The aim of the current study was to use qualitative methods to explore PAG in young people with IBD who self-identify with the concept of PAG. The current study aimed to explore how and in what way participants experience PAG because of their illness and to identify any common themes within this concept.

Method

Design

The concept of PAG has not yet been explored in the context of youth IBD. Further, it cannot be assumed that the processes of PAG proposed in previous quantitative or qualitative research would equally apply to IBD (Hefferon et al., 2009). Qualitative research is useful for exploring under researched topics and when using a quantitative methodology may risk missing an understanding of the whole picture (Hefferon et al., 2009).

Using a critical realist framework, Template Analysis was selected due to its flexible, structured approach to analysing data across cases and its emphasis on a hierarchical coding framework (Brooks, McClusky, Turley & King, 2015). Unlike many qualitative methods, Template Analysis allows the development of a priori themes based on existing literature provided these are tentatively used while engaging in participants' experiences and knowledge (Brooks et al., 2015).

Participants and Recruitment

Participants were sought who met the following inclusion criteria:

- Aged between 16 and 25 years inclusively. Many services define young adulthood as aged between 16 and 25 (Dovey-Pearce, Hurrell, May, Walker & Doherty, 2005). This was the definition used for the current study.
- Able to converse in English
- Diagnosis of IBD as stated by participants. No objective verification was sought.

Participants were excluded on the following criteria

- Younger than 16/older than 25
- Unable to converse in English
- Not diagnosed with IBD

An email invitation was circulated to a university held database of approximately 70 local people with IBD between the ages of 16 and 25 who had expressed interest in taking part in research. The study was also advertised nationally through several UK based IBD charities including IBD UK, Crohn's and Colitis UK, CICRA as well as IBD support groups on social media. Participants who took part in interviews were invited to send the advert to their own personal contacts provided that the individual met the inclusion criteria and were made aware that they did not have to respond. The advertisement (see appendix A) advised interested participants to contact the lead researcher via email. All interested participants were then sent a participant information sheet.

A total of 25 people contacted the lead researcher expressing interest in taking part and were sent the study information sheet (appendix B). There was no further contact from 9 participants. Another two potential participants were unable to take part due to not meeting inclusion criteria for age. The remaining 14 gave consent (appendix C) and took part in interviews. Table one below summarises the characteristics of the participants who took part in the research. Participants were largely female aged between 22 and 25, employed, White British with a diagnosis of Crohn's disease but in remission.

Table 3. Participant Characteristics (N=14)

Variable	*N (%) of Participants*	*Range*	*Mean (SD)*
Gender			
Male	2 (14.3%)	-	-
Female	11 (78.6%)	-	-
Prefer not to say	1 (7.1%)	-	-
Age			
16-21	3 (21.4%)	18-19	18.3 (0.58)
22-25	11 (78.6%)	22-25	23.6 (1.03)
Occupational Status			
Employed	9 (64.3%)	-	-
In Education	2 (14.3%)	-	-
Neither	2 (14.3%)	-	-
In education & employed	1 (7.1%)	-	
Ethnicity			
White British	10 (71.4%)	-	-
Asian British	1 (7.1%)		
White Scottish	1 (7.1%)		
White Icelandic	2 (14.3%)		
Diagnosis			
Crohn's Disease	9 (64.3%)	-	-
Ulcerative Colitis	4 (28.6%)	-	-
Crohn's & Ulcerative Colitis	1 ((7.1%)	-	-
Illness Status			
Remission	10 (71.4%)	-	-
Active	2 (14.3%)	-	-
N/A (surgical intervention)	2 (14.3%)	-	-

Measures

Demographic data were collected prior to interview. This included ethnicity, age, gender, employment/educational status, diagnosis and disease status.

The Short Inflammatory Bowel Disease Questionnaire (SIBDQ; Irvine, Zhou, & Thompson, 1996; appendix D) was used. This is a 10-item questionnaire designed to assess health related quality of life in people with IBD. Items are rated on a 7-point Likert scale with lower scores indicating lower health related quality of life. The SIBDQ has good test-retest reliability (0.65) and internal consistency (α=.75; Irvine et al, 1996). The SIBDQ can be divided into four domains; bowel, systemic, social and emotional domains. A total score can range between 10 and 70. Studies using the SIBDQ have used scores continuously and dichotomously; with those scoring under 50 reporting poor quality of life (Ulitsky et al., 2011).

Procedure

Ethical approval for the current study was granted from the University Research Ethics Committee (appendix E). In addition, feedback for the research protocol was sought from an expert by experience. Potential participants who responded to the study advertisement were provided with the participant information sheet. In preparation for the interview, consenting participants were invited to choose a piece of media that they felt represented their experience with IBD. This could include an object, photo, image, piece of art, piece of music, short piece of writing or poem. For participants having their interview over the phone, they were invited to email the lead researcher their chosen piece of media. Participants were instructed that this aspect of the interview was entirely optional, and all participants would be asked the same interview questions regardless of whether they chose anything to represent their growth. Where participants did choose a piece of media, typically photographs, this was discussed before beginning the main

interview (see appendix F). This draws on photo elicitation research which can be particularly useful when conducting research with young people (Drew, Duncan & Sawyer, 2010).

Consenting participants were then offered an interview time at their convenience. One participant was recruited from the university held database and was interviewed face to face. The other 13 were recruited through charities, social media and snowballing. These 13 interviews took place by phone.

Immediately prior to interview, demographic data were collected and the SIBDQ completed. Interviews took approximately 40 minutes and were recorded using an encrypted audio recording device. No identifiable information was asked for during the interviews, however where potentially identifying information was mentioned during the interview, this was anonymised during transcription. Interviews were deleted from the audio recorder as soon as they were uploaded to the secure University online cloud storage. The transcriber was a member of University staff who had signed a confidentiality agreement in line with University policy.

The interview comprised of two sections; the optional photo elicitation section and the main interview. The photo elicitation section asked participants to describe what they had chosen and why as well as how it relates to their IBD and PAG. The main interview asked participants to recount how they came to be diagnosed with IBD and how it impacted on their life following diagnosis. They were then asked to describe how they felt they had experience PAG and to think about why they had experienced it.

Following their interview, participants were debriefed and given the opportunity to ask any questions. They were asked for feedback and advised that they could get in touch should they have any further questions. Those that had opted to receive a copy of the research report were advised on when this might be.

Analysis

Data were analysed using Template Analysis which has been used extensively in healthcare research (Waring & Wainwright, 2008). When using Template Analysis, it is acceptable to use a priori themes if these are held tentatively and removed if there is no data to support them (Brooks, et al., 2015). Template Analysis is typically used when analysing across cases to look for common themes (Turley, Monro, & King, 2016). In the current study the following process was used:

Step One: Four main a priori themes were derived from published research (appendix G). Given the consistent finding in research that distress may be necessary to experience PAG (Picoraro et al., 2014, Kilmer et al., 2014) the subjective experience of the adverse event was chosen as a theme. Similarly, rumination and sense making have been highlighted in research (Kilmer et al., 2014; Helgeson et al., 2006) suggesting another theme; cognitive processing. Particularly for PAG in young people, the importance of social support has been proposed (Kilmer et al., 2014). This produced the third theme; social support. The fourth theme was individual characteristics based on research finding characteristics such as optimism may increase the likelihood of PAG (Picoraro et al., 2014).

Step Two: As recommended by King (2004; 2012), the first step in the analysis following data collection was to read through the transcripts for familiarity and to identify the sections relevant to the research question.

Step Three: Four transcripts chosen randomly were used to create the initial template. The relevant sections of transcripts were coded. The codes were then grouped into meaningful main themes and subthemes that represented the codes from across the subset of transcripts. The a priori themes were used as part of this process; some of these

themes were included in the initial template. What was produced at this point was an initial template (see appendix H).

Step Four: The initial template was then applied to another subset of four transcripts and amended to incorporate new codes or to reflect a new understanding of a theme or subtheme from the initial template (see appendix I).

Step Five: The revised template was then applied to all transcripts (see appendix J for example). Any further gaps or parts of the template that were not widely applicable were again amended (see appendix K).

Step Six: The final template was then applied again to the transcripts to help interpret the data and begin to make sense of it and begin writing the research report (King, 2004, 2012).

Reflexivity

In qualitative research, it is important that researchers are open about their own biases and perspectives (Elliot, Fischer, & Rennie, 1999). Being open during the research process will enable researchers to consider how their perspective will influence the interpretation of the data collected as well as allowing readers to view the report with an understanding of the relationship the researcher has had with the data (Elliot et al., 1999). Reflexivity is a constant process of critical self-evaluation and internal consideration of the researchers position in relation to the research and how that may influence the research process (Berger, 2015). A reflexive log was kept throughout the research process which included the analyst's reasoning, judgements and emotional reactions (Berger, 2015).

In addition, a data-orientated audit trail was kept throughout the template development which logged the rationale and reflections on every addition, removal and amendment to the template as is recommended in qualitative research to ensure that, as

far as reasonably possible, results are a product of the data rather than deriving from the researchers own thoughts and perceptions (Shenton, 2004).

Quality Checks

The draft interview schedule was scrutinised by the research supervisor, an expert working in the field of IBD and by an expert-by-experience. Following the development of an initial coding template, independent scrutiny from the research supervisor was also sought. As the template is developed and during the final write-up, this independent scrutiny was repeated. Research supervisor consultation was utilised to gain feedback on the reflexive log and how it might relate to the research and results.

Results

Participant Characteristics

Table two below summarises the SIBDQ scores. On average, participants total score was 50.4. If a score of 50 reflects good quality of life, participants had a borderline good quality of life. Similarly, the scores suggest participants reported a slightly lower quality of life than others with IBD; Irvine, Zhou and Thompson (1996) assessed 150 patients with CD and 45 patients with UC where SIBDQ scores ranged from 3.6 to 6.7 compared to the current 2.9-5.8. However, social quality of life was reported to be better than other aspects of living with IBD.

Table 2. SIBDQ Scores

Short Inflammatory Bowel Disease Questionnaire (SIBDQ) Scores (n=14)	***Average Raw Score***	***Mean (SD)***	***Range***
Raw Score	706	50.4 (8.44)	29-58
SIBDQ	70.5	5 (0.84)	2.9-5.8
Systemic	64.5	4.6 (1.95)	1-8.5
Social	80	5.7 (1.75)	1-7
Bowel	71.9	5.1 (0.92)	3.3-6.3
Emotional	69.2	4.9 (0.67)	4-5.7

Photo Elicitation

Five participants chose to take part in the photo-elicitation aspect of the interview. Two participants provided photos of themselves when ill and when well. The other two participants provided photos of themselves achieving goals such as running a marathon and taking a flying lesson. One participant chose the Disney (2016) movie 'Moana' and identified with its message of overcoming barriers to achieve goals. These images fitted within the themes derived from the interview data and did not add any additional themes.

Interview Data

Figure one below shows the final template (see appendix L for conceptual map). Analysis of participants transcripts produced five main themes relating to the '*initiation into the world of IBD', 'a life on hold', 'standing up to the illness', 'love surrounding pain and active acceptance*'. As can be seen in figure one below, each main theme generated second and/or third level themes. No themes emerged that were specific to diagnosis (CD or UC) nor gender. The themes were present in nearly all transcripts (see appendix M) although several themes were less common but present in over half of the transcripts. These included *'the value of openness in relationships', 'embodying genuine support'* which was divided into *'empathy for others and recognising the invisible'* and *'giving back means the pain was worth it'* as well as *'making social comparisons'*. Below each theme will be described using an example from the transcripts. Further examples can be seen in appendix N.

1. Initiation into the world of IBD
    a. Searching for answers
    b. A body out of control
    c. Making sense of life with IBD
2. A life on hold
    a. Frozen in time
    b. Acknowledging losses – physical and emotional
3. Standing up to the illness
    a. Carrying on regardless
    b. Taking back control
    c. Rolling with the punches
4. Love surrounding pain
    a. Learning the real meaning of friendship
    b. Social support = survival
    c. The value of openness in relationships
    d. Embodying genuine support
        i. Empathy for others and recognising the invisible
        ii. Giving back means the pain was worth it
5. Active acceptance
    a. Achievements and new directions
    b. Appreciating good health
    c. It's all about perspective
        i. Making social comparisons
        ii. Looking for the positives
        iii. IBD perks – gaining psychological strengths and emotional independence

Figure 1. Final Template

1. **Initiation to the world of IBD.** An important part of the participants stories was why IBD had been such a challenge. Across participants there was a shared sense that at the beginning of their lives with IBD, there was much to learn and adjust to.

1.1.*Searching for answers.* Initially, participants searched to explain troubling symptoms, often struggling with symptoms being dismissed or misdiagnosed. Many participants had considerable periods of time left with this period of ambiguity.

P9 "And my mum was researching online and she said 'I really think my daughter's got Crohn's disease', and like my bloods were only showing like slightly raised RP levels, but it wasn't enough for them to consider it to be

something major, it was ridiculous and it was 4 years of going back and forth, constant blood tests…"

1.2.*A body out of control.* Around the time of diagnosis and beyond, participants reported ongoing symptoms or treatment side effects. Many participants reported significant challenges in learning to manage their condition such as being unable to tolerate food and the ongoing struggle with relapses and remissions.

P14 "So I realise I was a pretty nasty child to my parents anyway, because – then you sort of realise you didn't really have control over how you were acting, and it was sort of the fact that I had no satiety when I was eating and I constantly craved these really high fat foods and I'd constantly – I'd be very demanding…"

1.3.*Making sense of life with IBD.* Most participants identified with not fully understanding what having IBD meant at first, sometimes only realising much later the impact it would have on their lives

P5 "...I did a bit of research online…I think that I kind of maybe felt a bit better after that because I was reading about all the treatments and things, um, that it was still quite like – at this point I think I still thought in my head that like they would put me on some tablets and I'd be normal again, I didn't realise that there wasn't ever going to be a normal, if that makes sense?"

2. A life on hold. Following their initiation into the world of IBD, the participants described disruption to their lives whilst they adjusted to their condition, often feeling that they could not grow and develop as their peers could.

2.1.*Frozen in time*. Participants described that following diagnosis they would often experience a period of stagnation; whether this was being housebound, not being able to carry on with work or education or losing independence.

P13 "…Um, and I was saying to my mum, because sometimes we'd just be sitting in silence and she'd just say, 'are you going to speak to me today' and I'd just say 'I've literally nothing new to update you on. Like you know everything that's going on, nothing new has happened, I've got nothing, literally nothing in my head to say'."

2.2.*Acknowledging losses – physical and emotional*. Participants were aware of the losses they had experienced because of IBD. Many gave up hobbies, career plans or educational goals. For others, the losses were more emotional such as feeling left behind.

P10 "It was quite crushing to be told I couldn't pursue a career in what I wanted to do…when I kind of had my dreams set on that, because I was a gymnast and I could have joined the navy, etc, but I wasn't allowed. Um, so yeah, quite a big impact, but I tried to stay positive, but I got on the course I wanted to do, so yeah."

3. **Standing up to the illness.** Participants' early experiences with IBD seemed to foster defiance towards the illness and what it had done to their lives.

3.1.Carrying on regardless. There was a strong sense that participants became determined to carry on with life despite having IBD whether this was due to being unaware of the severity of their illness or due to a determination to live as normally as possible;

P14 "...for myself it was difficult to sort of appreciate how ill I actually was, because I was always, like I said, a really outgoing sort of – I want to do everything, a really kind of active person, so I just kind of got on with it…;"

3.2.*Taking back control*. Participants regained control where they could. Learning to live with IBD meant that participants knew themselves, their bodies, what they could withstand and what treatments would work best for them.

P1 "Um, and also kind of taking responsibility for your own health because I think a lot of people 'oh well I've got Crohn's I'm poorly and that's it' whereas I thought, no, I'm going to exercise and sleep a lot, I'm going to go vegan, I'm going to change my lifestyle..."

3.3.*Rolling with the punches.* Although participants expressed the desire to carry on despite the barriers IBD placed in their way, there was the need to adapt and modify things somewhat in order to work around the condition. With a chronic condition, this meant being able to adapt and compromise.

P12 "I always wanted to do the usual thing; finish uni, do a gap year, obviously that didn't happen…-and that was it, so I left, no gap year and that's how it is, just deal with it, to a point where now, work notwithstanding, I will be taking extended holidays up to fairly faraway places, just – I'll just know how I need to manage it more, I'll just, yeah I'll take more precautions. Fine, it may not be as long as I want to be, but do you know what, I'll still get to the vast majority of the places I want to get to, just in more trips."

4. Love surrounding pain. A strong theme was that social support was paramount to being able to live a fulfilled life with IBD.

4.1.*Learning the real meaning of friendship.* Many participants lost friends or partners following their diagnosis. Some felt they could no longer maintain these relationships due to a realisation that they had less in common than previously. However, many reported finding new friends where there was a deeper connection;

P2 "…and like when you're in such a state, like I had been, you see the beauty in the world, you see the beauty in people and the beauty in people's ways of how they can help you just to feel strong. Like last year, my friends would come in (to

hospital) every week, and we're all music students, so they'd all sing with me and just keep me going and that's a beautiful thing that you have to take."

4.2.*Social support = survival.* Support from others, particularly family members, was essential. Whether this was to provide emotional support, financial support or to provide the right environment under which they could get their condition under better control. Many felt that this, along with emotional support, positivity and encouragement was a crucial facilitator of experiencing PAG.

P4 "they're just great, I don't know how to describe it. They come to appointments with me, ask all the questions I'd probably forget to ask, make sure that I'm taking my medication, like remind me about it 50 times a day because I'm forgetful"

4.3.*The value of openness in relationships.* IBD is largely an invisible illness. It also causes many embarrassing symptoms. For this reason, some chose initially to keep their condition private from the outside world. However, it seems that participants found it beneficial to have some people with whom they could be open about their condition. This lead to valuing honesty and openness

P8 "I would say it has definitely strengthened my relationships as well. I think at first, maybe at first I was kind of embarrassed about it, but then my friends were very, um, kind of supportive and um, a couple of them studied medicine-type things and then they were really interested as well in kind of more about the disease and stuff, so it helped make it more of an open conversation; you didn't have to be awkward about your symptoms and what was going on."

4.4.*Embodying genuine support.* One of the ways in which many participants felt they had grown was in offering support to others. This seemed to be in two ways:

4.4.1. ***Empathy for others and recognition of the invisible.*** Participants understood that their health, whilst largely invisible to others, impacted greatly on their mood and behaviour and that this could be the case for others.

P7 "Yeah um, I think I'm a lot more – this is what my mum calls it – emotionally intelligent, so I'm like very aware of other people's emotions and how they react to stuff, so I'm always the kind of person that says like even if someone's like really angry or doing something negative, like there might be something going on behind the scenes that you don't know about, because having this condition makes you so aware that people with like invisible illnesses and you don't know what people are struggling with, so don't judge them straight away."

4.4.2. ***Giving back means the pain was worth it.*** Many participants derived a sense of purpose and meaning from being able to share their experiences with others who were diagnosed with IBD and to be able show them a positive outcome.

P2 "I think because my story has been so big, I've been able to kind of um, reach it out to everybody else, I've been able to speak about it to a lot of different people which has brought other people positive growth as well"

5. Active Acceptance. Although participants first experienced the difficulties of IBD, all participants came to view their condition positively and actively embraced life with IBD.

5.1. Achievements and new directions. Despite the barriers, participants worked hard for academic achievements, to reach goals, try new hobbies and find new careers. For some, it was the barriers that increased the value of the things they had achieved.

P8 "...for a little while I wasn't sure that I was going to get through it, just because I was quite stressed about it, how to manage the workload as well as the illness. I think going from there and then kind of calming down a little bit and just

focusing on the work and then – and then I managed to still do quite well, even though I had to have an extension and things like that, so that kind of made me more proud of it in a way, or it made it seem more worth it."

5.2.Appreciating good health. Participants all felt appreciative and made the most of their health, particularly when they felt their condition was well managed or in remission.

P2 "…I really appreciate my food, I really like – even if it's just a bowl of soup, like, it's amazing that I can eat it because I've been, I've had so many times where I'm starving to death…"

5.3.*It's all about perspective.* There was a strong theme that a positive outlook had helped participants to manage their condition, stumbling blocks and indeed feel that they had benefitted from their condition.

5.3.1. ***Making social comparisons.*** A common way that participants achieved this was to compare themselves to those less fortunate or themselves earlier in their journey with IBD.

P7 "Um, but I think also like um, I always think like there are – I kind of try and look on like the positive side of things, like there are people who are in a lot worse condition than I am, like with my condition. Like there is people who are in hospital every week and I'm lucky to not be in that."

5.3.2. ***Looking for the positives.*** Participants also generally looked for positives in their situation, which brought them comfort and strength;

P11 "That it's not always about how damaged you are…but if two people have the same illness and one was positive about it and would do everything they could to almost fight it and the other just gave up, it just changes so much"

5.3.3. ***IBD perks – gaining psychological strength and emotional independence.*** The difficulties of IBD helped participants develop resilience and feel that they could take on future challenges more successfully.

P8 "…and I think it makes you maybe more resilient if you face other set-backs, even in work or um, or academic life that you know that you can, you do have the strength to get through it somewhere, even though it seems really daunting at first."

In summary, participants described having IBD as an adverse event which disrupted their lives even before diagnosis. Living with the condition began with being unable to move forward in their lives until they had learned to manage or adapt to their condition. Participants experienced emotional losses as well as physical. However, what seemed to help participants to achieve PAG was successfully navigating and adapting life with IBD with help from friends and family. Participants came to view IBD as a positive influence in their lives reporting closer and more meaningful relationships, appreciating good health, being able help and support others as well as gaining psychological strengths. The participants described achievements whether from new paths or from achieving goals by overcoming the challenges of IBD.

Quality Assurance

A reflective log was kept during the process of recruitment, data collection and analysis of the current study with the aim of ensuring that the researcher was aware of and minimised potential biases whilst conducting the research. There were possible biases that readers may wish to consider when appraising these results. In parallel to the current research, the researcher was conducting a literature review on family functioning in the context of IBD which may have resulted in the amplification of family in the current research. Further, at the beginning of the research the lead researcher was working in a

paediatric gastroenterology psychology service as a trainee clinical psychologist. This may have elicited a therapeutic style of interviewing which may have uncovered more psychological aspects than if a more neutral stance had been taken. Further, the lead researcher had experience of working with patients with IBD who described experiences similar to PAG and was therefore positive about the possibility of identifying PAG in other young people with IBD. This may have biased the researcher towards eliciting these experiences from participants.

These issues were discussed with the research supervisor. In addition, scrutiny and feedback was sought throughout the development of the template and its use in the analysis from the research supervisor. It was noted that the first version of the template felt descriptive. This lead to a significant shift in the template themes. Discussions continued until there was total agreement between the researcher and the supervisor.

Discussion

This study aimed to explore the experiences of PAG in 14 young adults with IBD. Template analysis was used to analyse transcripts from semi-structured interviews exploring young people's experience of IBD and PAG. The analyses identified several themes relating to the concept of PAG. The adverse nature of IBD was represented by the theme *'initiation into the world of IBD'* involving a period of ambiguity (*'searching for answers'),* a body no longer in their control (*'a body out of control')* and needing to *'making sense of life with IBD'.* The adverse nature of IBD was further revealed in the theme of *'a life on hold'* and two subthemes; *'frozen in time'* and *'acknowledging losses – physical and emotional'*. However, three main themes emerged which were consistent with the concept of PAG; *'standing up the illness'*, *'love surrounding pain'* and *'active acceptance'*. Participants were defiant in the face of the IBD ('*standing up to the illness'*)

which meant *'carrying on regardless'* and increased perceived control (*'taking back control')* as well as adapting to their new lives (*'rolling with the punches').* The importance of social support was evident (*'love surrounding pain')*. Participants felt they gained a new understanding and increased depth in their relationships ('*learning the real meaning of friendship', 'the value of openness in relationships')* and that their health and wellbeing depended on those relationships (*'social support = survival'*). Participants gained from being able to provide support to others (*'embodying genuine support')*. The determination of participants and their support networks meant they were able to achieve 'active acceptance' and embrace life with IBD. This included *'achievements and new directions'* and *'appreciating good health'*. Participants described a new outlook on life *('it's all about perspective')* derived from *'making social comparisons'*, *'looking for positives'* and *'IBD perks – gaining psychological strength and emotional independence'*.

These findings mirror some previous research on the development of PAG in young people. The development of acceptance and positivity of participants fits with findings that despite the challenges of living with IBD, young people with IBD can and do develop enhanced coping strategies, particularly when well supported by family and friends (McCombie et al., 2013). Indeed, much of the literature on PAG has advocated the importance of social support (Picoraro et al., 2014) which was a strong theme in the current study. In fact, Tedeschi and Calhoun (2004) have claimed that confiding in others about the adverse event helps individuals make sense of and derive meaning from the event and therefore develop PAG. This would certainly mirror the findings in the current study about the role of social support.

Previous research has eluded to a link between rumination and PAG (Helgeson et al., 2006). Attempts to explain this link have suggested that sense-making is necessary for PAG. Here, it seems that participants made attempts to make sense of their illness. In

the literature, distress has also been highlighted as an important component of PAG (Kilmer et al., 2014) and indeed participants were clear in the ways IBD had negatively affected their lives. However, the negative aspects did not diminish the positive growth participants described. This agrees with research that suggests distress is necessary to experience PAG. Indeed, Park (2010) argued that PAG develops when individuals separate from previous beliefs and develop new ones that incorporate the difficult experience. Further, it supports the idea that PAG is both positive and negative simultaneously (Soltani et al., 2018). So, perhaps it is not that the negative aspects reduce but that they are viewed differently.

One interesting finding was participants' increased compassion for others. Few studies have found or explored the concept of increased compassion in PAG and it is argued that this has been overlooked in research on PAG (Morris, Shakespeare-Finch & Scott, 2012). However, one study reporting a similar finding was interestingly also in the context of physical health. Morris et al. (2012) found that cancer survivors reporting PAG frequently cited increased appreciation for life but also compassion for others. As found in the current study, the authors suggest that compassion towards others, particularly those in a similar position, can deliver meaning and purpose following negative experiences (Morris et al., 2012).

Similarly, whilst studies of PAG refer to the importance of individual characteristics such as optimism, few have discussed the role of perceived control in PAG. Models of psychological adjustment to health conditions however frequently highlight the importance of perceived control and it is frequently linked with positive adjustment to illness (Leventhal, Leventhal & Cameron, 2001). Feeling in control is a challenge in the context of chronic illness which can fluctuate over time despite treatment (Stanton, Revenson & Tennen., 2007). Despite initially feeling out of control, the current study

found participants developed a strong sense of defiance and took back control where possible. It may be that control is a key part of adjustment and PAG in the context of chronic illness. Indeed, Connerty and Knott (2013) found that perceived control was an important part of positive growth in cancer survival.

Limitations and Future Directions

Previous research has suggested that time since the adverse event is an important determinant of PAG and positive outcomes (Helgelson et al., 2006). The current study is limited in that it did not explore the influence of time since diagnosis. It was clear from interviews that some participants were relatively newly diagnosed compared to others who had been diagnosed at a young age and had grown up with the illness. Further, it would have been interesting to explore PAG in participants who had experienced relapses and remission. It may be that PAG would fluctuate under these circumstances. For instance, a study of PAG in paediatric diabetes found that PAG was unstable, fading over time for some participants but was stable over time for others (Rassart et al., 2017). Future studies could directly compare the testimony of those who have been diagnosed for longer, those diagnosed at a young age and the influence of changes in disease activity.

The number of participants recruited was slightly lower than anticipated; we had intended to recruit 15 participants. Further, nine participants chose not to take part after expressing interest and receiving information. This suggests that the procedure was perhaps too laborious for some. Alternatively, it may be that the current participants were more comfortable talking about their experiences or more strongly identified with the concept of PAG and may therefore represent a unique sub-group of young people with IBD. Further, it may be that those choosing not to take part had experienced PAG but did not identify with the definition of PAG or the terms used in this study. Future research

could attempt utilise a measure of PAG as a screening tool to widen and more objectively recruit.

Further, the sample of participants was fairly homogenous. Future research should make efforts to recruit more males, younger participants and participants with active disease. In doing so, more conclusions could be drawn about whether PAG is possible even in the midst of disease activity and across a broader range of population characteristics.

There were also methodological limitations. The transcripts and themes were not passed to participants for feedback prior to write-up. It is therefore possible misunderstanding may have arisen which could influence the conclusions drawn.

Most participants chose not to take part in the photo-elicitation aspect of the interview. Although photo-elicitation has been recommended in qualitative research with young people (Meo, 2010), participants in the current study were typically at the older end of the age range. In fact, those that did take part in the photo-elicitation were the four youngest participants. Possibly due to the lack of uptake, the addition of photo-elicitation did not expand the findings. Researchers have noted that the technique lacks a clear procedure (Meo, 2010). Indeed, the questions in the interview schedule were general rather than arising from the media presented by participants perhaps limiting its depth.

Theoretical and Clinical Implications

The findings of the current study suggest that it is possible for youth with IBD to identify with describe experiences consistent with the concept of PAG. Research on PAG in young people is scarce, possibly due to assumptions that young people are not old enough to experience PAG (Tedeschi & Calhoun, 2004). The current findings should be encouraging to researchers wishing to explore PAG in younger populations.

Although doubts remain about the extent to which the factors involved in the development of PAG are modifiable (Connerty & Knott, 2013) the current findings suggest several ways this could be explored. Previous research and the current findings suggests that social support, particularly being open with others, is an important part of PAG. Clinicians in medical or mental health settings should certainly encourage their patients with IBD to be open about their condition whether this is with a professional or within support networks. Future research could also explore whether talking therapies, support groups or interventions encouraging young people with IBD to openly discuss their experiences promotes the development of PAG.

Further, results here suggest that some young people with IBD could benefit from feeling more in control of their condition, which has implications for medical and psychological clinical practice. Young people with IBD should be listened to and given options where possible in their treatment and beyond. For example, the findings suggest that some young people with IBD could benefit from support in setting and meeting life goals. Indeed, Connerty and Knott (2013) have argued that encouraging use of social support, physical activity and lifestyle changes promote positive growth in cancer survivors and suggest that this is through an increased perception of control.

Additionally, compassion for others appears to have been underestimated by quantitative measures of PAG (Morris et al., 2012). The findings here that increased compassion for others supported participants to derive meaning from their illness could implicate the clinical utility of compassion focused interventions for youth with IBD. Whilst such therapies typically focus on compassion for the self, they also strengthen the skill of compassion which includes compassion for others (Gilbert, 2009). The benefit of such an approach would also promote the acceptance of being soothed and cared for by others (Gilbert, 2009) which the current findings suggest may be an important part of

PAG in youth with IBD. Future research could explore the effectiveness of compassion focused interventions for youth with IBD

Conclusions

The current study suggests that it is possible for youth with IBD to experience PAG despite the challenges of living with a chronic illness. What seems important is a blend of acknowledging the difficulties that IBD imposes but not being hindered by them. Participants regained a sense of control and carried on in the face of IBD related barriers. They valued the support they were given and developed deeper relationships as well as compassion for others. Participants derived meaning from many aspects of their illness and embraced life with IBD by viewing it positively and feeling mentally stronger because of it. They appreciated their health and made the most of it by achieving goals and moving forward in life. For anyone experiencing living with IBD these findings offer hope that despite living with a challenging condition, the reality need not be entirely negative. However, in order to apply these findings and promote PAG in the general population of young people with IBD, further research is needed.

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Appendices

Appendix A. Study Advertisement

Email: 'Living with IBD can be painful and distressing. Despite this, some people experience positive changes to their outlook on life, their beliefs or values as a result of experiencing something difficult. They might find that things like family, friends, hobbies, school work or jobs are more important and viewed more positively than they were before the diagnosis. This is called post-adversarial growth.

If this is something you feel you have experienced as a result of having IBD we would like to hear from you. We are looking for participants aged between 16 and 25 to take part in a study exploring the experiences of young people who have experienced post-adversarial growth as a result of having IBD. If you consent to taking part you would be interviewed over the phone (or in person if you live in the Sheffield area and would prefer this). Interviews last around 40 minutes. Interviews are recorded, anonymised and transcribed for analysis and discussion in a research report. You would not be identified in any way.

If this is something you might be interested in please contact Oonagh O'Hare on oohare1@sheffield.ac.uk for further information.'

Twitter 'We are seeking participants aged 16-25 for a study on experiences of positive personal growth as a result of having Inflammatory Bowel Disease. For further information please contact: oohare1@sheffield.ac.uk'

Appendix B. Information Sheet

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

Telephone:
Fax:
Email:oohare1@sheffield.ac.uk

Version 3 12/07/2018

PARTICPANT INFORMATION SHEET

What is the purpose of the study?

Although living with a chronic health condition can and does cause distress, some people experience positive changes in their outlook, values and beliefs because of the adversity they have experienced. This is called post-adversarial growth. The aim of the current study is to explore how and under what circumstances young people with Inflammatory Bowel Disease (IBD) experience post-adversarial growth.

Why have I been invited?

You are being invited because you are a young person who has been diagnosed with Inflammatory Bowel Disease and have identified with the idea of post-adversarial growth.

Do I have to take part?

No – it is entirely up to you whether you take part of not. If you choose to take part you are free to stop taking part at any time if you should change your mind.

What will happen if I take part?

You will be invited to take part in an interview. This will take place at Cathedral Court, 1 Vicar Lane or over the phone. You will be given more details on this before your interview date.

Before the interview, you are encouraged, but not required, to find or create something that represents the growth you have experienced as a result of your illness. This might be an image, a

photo taken by you or someone else, a piece of music, a piece of art work by you or someone else, or an object. If you are unsure about what to bring, but you would like to bring something, you can contact the lead researcher for advice. You will be invited to bring this item to the interview where it will be discussed with you as part of the interview.

If you'd rather not bring an item with you or you can't find something suitable – this is not a problem – we would still like to you to come to the interview!

At the interview, you will be asked to complete a short questionnaire about your illness. You will then be asked to talk one-to-one with the interviewer about your illness and how things have changed since you were diagnosed. The interviewer will have some questions they will ask you, but the intention is that it will feel more like a conversation. Although it may vary, the interviews are expected to last between 20 and 40 minutes.

What are the benefits of taking part?

Taking part in the interview will be an opportunity for you to reflect on the positive changes you have experienced. We hope that you find the experience meaningful and enjoyable.

What if there is a problem?

We don't expect that taking part will involve any risks. However, in the course of the interview and in completing the questionnaire, you will be asked about your illness. It is possible that this may be distressing. If this occurs you should tell the researcher.

If you notice significant distress following the interview, it may be helpful to speak to your General Practitioner (GP) about support. If you become concerned about your welfare and need immediate emotional support, we would suggest that you call The Samaritans on 116 123 from any phone, 24 hours a day.

We have a duty of care to keep participants safe. Although it does not occur very often, if you were to say anything in the interview indicating that you, or someone around you, is at risk of harm we will pass this information on to the appropriate services e.g. police. Where possible, we will tell you prior to doing so.

Will all the information be kept confidential?

Yes. The interview you take part in will be recorded using an audio recording device. The audio file will then be uploaded to a secure online storage facility on The University of Sheffield's server and deleted from the audio recording device.

The audio files will be anonymous as you will not be asked to identify yourself on the recording. You will be asked during the interview to avoid saying people's names or anything that might identify you but the lead researcher will delete any identifiable information from the audio recording before they are typed up. Your audio recording will only be identified by the time and date of the interview.

The interviews will be typed up by a typist who works for the University and is bound by the University's confidentiality policy.

The audio recordings of your interview made during this research will be used only for analysis and for anonymised illustration in publications, conference presentations and lectures. No other use will be made of them without your written permission, and no one outside the project will be allowed access to the original recordings.

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

We are able to offer a maximum of £5 to pay for any travel costs you may incur by attending the interview. You will need to provide tram/bus/train tickets or receipts.

What will happen to the results of the study?

The interviews from every participant will be analysed together by the lead researcher once they have been typed up. The researcher will be looking for similarities and themes across the interviews in order to understand how people with IBD experience growth. This will be written up as a research report. Anything that could possibly identify a participant will be excluded from the report. 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. You will not be able to be identified in any reports or publications unless you have given your explicit consent for this.
Following completion of the study, the data will be kept securely for a period of five years in line with University policy. During this time it will only be accessible to approved members of staff. After this time, it will be securely destroyed.

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.

Who has approved this research?

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

What if I wish to complain about the way the study has been carried out?

If you would like to make a complaint about this project, in the first instance you should contact the lead researcher. If you do not feel satisfied that your complaint has been dealt with appropriately you can contact the lead researcher's supervisor, Prof. Gillian Hardy, Professor of Clinical Psychology or Dr. Andrew Thompson, Director of Research Training.

If you feel that your complaint has not been handled to your satisfaction following this, you can contact Dr. Thomas Webb, chair of the Department Ethics Subcommittee on t.webb@sheffield.ac.uk or Prof. Glenn Waller, Head of Department at g.waller@sheffield.ac.uk.

Can I withdraw at any time?

Yes – at any time during the process you may tell the lead researcher you no longer wish to be involved.

Contact Information

This research is being conducted by **Oonagh O'Hare,** Trainee Clinical Psychologist. This research will be used to write a thesis which fulfils part of their doctoral training. If you have any questions about the research you can contact her on oohare1@sheffield.ac.uk

Alternatively, you can leave a telephone message with Amrit Sinha, Research Support Officer on: 0114 222 6650 and he will ask Oonagh to contact you.

Thank you for taking the time to read this information.

Appendix C. Consent Form

The University Of Sheffield.

Participant Consent Form

The experience of post adversarial growth in young people with inflammatory bowel disease.

Please tick the appropriate boxes	Yes	No
Taking Part in the Project		
I have read and understood the project information sheet dated 12/07/2018 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.)	☐	☐
I have been given the opportunity to ask questions about the project.	☐	☐
I agree to take part in the project. I understand that taking part in the project will include taking part in an interview which will be recorded, anonymised and transcribed for analysis.	☐	☐
I understand that my taking part is voluntary and that I can withdraw from the study at any time. 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.	☐	☐
How my information will be used during and after the project		
I understand my personal details such as my name and email address will not be revealed to people outside the project.	☐	☐
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.	☐	☐
Once written, I would like a copy of the research report	☐	☐

Name of participant [printed]	Signature	Date
Name of Researcher [printed]	Signature	Date

Project contact details for further information:
Oonagh O'Hare (lead researcher): oohare1@sheffield.ac.uk or Prof. Gillian Hardy (supervisor): g.hardy@sheffield.ac.uk. Should you wish to contact someone outside of the project you can contact

Dr. Tom Webb (Chair of the Department Ethics Committee) on t.webb@sheffield.ac.uk or Prof. Glenn Waller, Head of Department on g.waller@sheffield.ac.uk.

Save 2 copies of the consent form: 1 paper copy for the participant, 1 copy for the research data file

The template of this consent form has been approved by the University of Sheffield Research Ethics Committee and is available to view here: https://www.sheffield.ac.uk/rs/ethicsandintegrity/ethicspolicy/further-guidance/homepage

Appendix D. SIBDQ

Short Quality of Life in Inflammatory Bowel Disease Questionnaire (SIBDQ)

This questionnaire is designed to find out how you have been feeling during the last 2 weeks. You will be asked about symptoms, the way you have been feeling in general and how your mood has been as a result of your inflammatory bowel disease

1. How often has the feeling of fatigue or of being tired and worn out has been a problem for you during the last 2 weeks? Please choose from one of the following options
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

2. How often during the last 2 weeks have you had to delay or cancel a social engagement because of your bowel problem? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

3. How much difficulty have you had, as a result of your bowel problems, doing leisure or sports activities you would have liked to have done during the last 2 weeks? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

4. How often during the last 2 weeks have you been trouble by pain in the abdomen? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

5. How often during the last 2 weeks have felt depressed or discouraged? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

6. Overall, in the last 2 weeks, how much of a problem have you had with passing large amounts of gas? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

7. Overall, in the last 2 weeks, how much of a problem have you had maintaining or getting to, the weight you like to be at? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

8. How often during the last 2 weeks have you felt relaxed and free of tension? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

9. How much of the time during the last 2 weeks have you been troubled by a feeling of having to go to the bathroom even though your bowels were empty? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

10. How much of the time during the last 2 weeks have you felt angry as a result of your bowel problem? Please choose one option.
    1. All of the time
    2. Most of the time
    3. A good bit of the time
    4. Some of the time
    5. A little bit of the time
    6. Hardly any of the time
    7. None of the time

Please calculate the score by adding the number for each question. Minimum score = 10 and maximum score = 70.

Appendix E. Ethical Approval

Downloaded: 10/06/2018
Approved: 08/05/2018

Oonagh OHare
Registration number: 150123668
Psychology
Programme: Doctorate in Clinical Psychology

Dear Oonagh

PROJECT TITLE: The experience of post adversarial growth in young people with inflammatory bowel disease.
APPLICATION: Reference Number 019115

On behalf of the University ethics reviewers who reviewed your project, I am pleased to inform you that on 08/05/2018 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 019115 (dated 26/04/2018).
- Participant information sheet 1042878 version 1 (24/04/2018).
- Participant consent form 1042879 version 1 (24/04/2018).

The following optional amendments were suggested:

Info sheet: Suggest deleting "Where possible, we will endeavour to meet you a time that is convenient." or rewording - obviously, the interviews should take place at a time that is convenient for the participants - the "where possible/endeavour" implies otherwise! Suggest reword of "If you choose to take part you are free to stop taking part at any time if you should change your mind." Perhaps to "If you choose to take part you are free to withdraw from the study at any time if you change your mind. " Other: 1. Will participants understand what the term "post-adversarial growth" means? Consider including a lay definition in the information sheet. 2. How will participants self-identify as having experienced post-adversarial? Will this be checked in any way? For example, a brief measure of post-traumatic growth could be completed in addition to the QoL measure. Please let us know if you include further measures All the best with your research

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

Thomas Webb
Ethics Administrator
Psychology

Appendix F. Interview Schedule

Part One. Photo-elicitation.

Tell me about the item you have brought today. What made you decide to bring this item? How do you think it relates to your IBD? Would you be happy for us to use the image/image of the item you have brought with you today as part of our research report?

Part Two. Interview.

1. Intro/warm up question: Tell me about how you were diagnosed with IBD.
Prompts: How long ago were you diagnosed? How long did it take for you to receive your diagnosis? How did you receive the diagnosis?

2. What was life like in the weeks after you were diagnosed?
Prompts: How did life change in the months/years following diagnosis (if applicable)? Were there any expected changes following diagnosis? What has managing your condition been like? What are some of the negative things that have happened as a result of having IBD? Can you give me an example?

3. Some people find that going through something difficult, like having IBD, can result in positive changes in the way they view the world or themselves. They might find that some things, like friends, family, hobbies, school work or a job are more important and more positively viewed following diagnosis than they were before. You've been invited to this interview because you feel this has happened to you. Can you describe your experience of positive changes?
Prompts: Have your relationships with friends or family changed? Has anything changed in terms of hobbies or interests? Has anything changed in your work/school? In what way? What was positive about these changes? Can you give me an example? Have you learnt anything as a result of having IBD?

4. Why do you think that you experienced positive-growth following your diagnosis? Do you think everyone who is diagnosed experiences positive-growth? – why/why not?

Appendix G. A priori Themes

Main Themes
Social support
Individual characteristics e.g. optimism
Subjective experience of adverse event
Cognitive processing e.g. rumination

Appendix H. Initial Template

Main Theme	Second Level	Third Level
Subjective Experience of Adverse Event	Illness taking its toll on family	
	Invisibility of illness/lack of understanding from others	
	Interrupted life/not being able to do things	
	Treatment	Positive and Negative
	Diagnosis	Severity of Illness
Social Support	Family relationships	Relatability
	Friendships	
	Supporting others	
Individual Factors	Determination/pushing self	
	Resilience	
	Developing Maturity	
	Social comparisons	
	Taking Control/Assertiveness	
	Adaptation/Adjustment/Acceptance	
Moving Forward/Getting on with Life	Achieving things/enjoying life	
	Knowing own body and self	

Appendix I. First Revision

Main Theme	Second Level	Third Level
Subjective Experience of Adverse Event	Invisibility of illness/lack of understanding from others	
	Interrupted life/not being able to do things	
	Treatment Experiences	Positive
		Negative
	Difficulties with diagnosis	
	Individual Experience of Illness	
Social Support	Family relationships	General
		Relatability
		Impact on Family
	Friendships	
		Relatability
	Supporting others	
	Support and acceptance from others	
Individual Characteristics	Adjusting and trying to continue with life	
	Mental Strength	
	Making comparisons	With self
		With others
Defining Positive Growth	Achieving things/enjoying life	
	Taking ownership of health	
	New & helpful ways of being	
	Acceptance	

Appendix J. Example Transcript/Codes

Section of Transcript (P14)	Main Theme	Second Level	Third Level
P Ok, so I first started getting symptoms when I was 6 years old. R Wow, ok. P Yeah, I was quite young. Um, and it was basically just noticing I was going to the toilet a lot, sort of up to maybe 15 times a day it got to, and there was sort of a lot of blood in stools and that was sort of filling the blood of the toilet. So me and my mum took a lot of journey's to the GP, got told it was a fissure, got told it was just like a tummy bug, given some creams and things like that, and obviously by then I was just getting more and more fatigued, because I was losing so much blood and my mum was getting more insistent with the GP to refer me to secondary care. Um, so about - after about 11 months or so, finally got referred for some tests and without doing any sort of major tests the surgeon just said 'oh, we think it's a polyp in your large bowel, so we'll do a colonoscopy and remove it' and like 'oh, ok, well that's not a problem, it can go away.' Um, and that was a couple of days before Christmas and they did this colonoscopy and then found that my whole bowel was ulcerated and enflamed and like 'sorry, it's not a polyp, you've actually got IBD, we'll refer you to a paediatric gastroenterologist and that's sort of when it started, so by then I was 7, um, so it took about a year to get diagnosed. R Right, wow, ok. So what was it – I suppose you were very young, so you might not remember, but what – can you remember how you were feeling at the time when you were getting all these sort of answers from your doctor that didn't quite explain the issues? Yeah, what was it like? P I mean, I think, for me – it was more worrying for my mum, for me I was just 'I don't know what's happening.' Obviously it's really scary when you're bleeding that much and constantly having to run to the toilet, but my mum was the one that was like 'something definitely isn't right' and for her I think it was really tough. Um, and I mean, that also meant – I've got an older brother and an older sister and it meant this attention	Initiation into the world of IBD	Searching for answers Searching for answers	

got off them and was all sort of focused on me when I was getting diagnosed, because it was like 'this is something we haven't come across before. So for me it was just feeling that tired, I just wanted to stop feeling tired, because I'd always been a really energetic kid, I always just wanted to play out in the park and it was stopping me doing those things, because I was just absolutely shattered all the time.	A life on hold	Frozen in time	
R No, so what was life like then in the weeks and months after that? So how did you cope with the new diagnosis? P I mean for me, I was still a very determined little kid, I was like, I'm going to try and play as much as I can and whatever and obviously it did limit me but I was still trying, and pretty soon I was put on prednisone steroids. Um, which made a really big difference, and so I did get my energy levels back to sort of a baseline at that point, so I could still do the activities I wanted and things like that. R Good, that sounds good. How did you – were there any negative effects of the medication you were on? P A lot, ha. R OK. P Yeah, I mean sort of then going on steroids, and the side effects, um, did get to me a lot. Um, so I started experiencing the really bad moon face, so the big swollen cheeks, excessive hair growth on my face, which, being Indian, you're already sort of predisposed to that, so that was quite difficult, and then the swollen tummy and those kind of things. And something I only sort of reflect on now is how bad my mood swings actually were. Um, so I realise I was a pretty nasty child to my parents, anyway, because – then you sort of realise you didn't really have control over how you were acting, and it was sort of the fact that I had no satiety when I was eating and I constantly craved these really high fat foods and I'd constantly – I'd be very demanding, like 'ok, I want scampi and chips for dinner, I want all of these kind of really rich foods, which obviously just made me gain a lot of weight, on top of being on the steroids, which was quite difficult.	Standing up to the illness Initiation into the world of IBD	Carrying on regardless A body out of control	

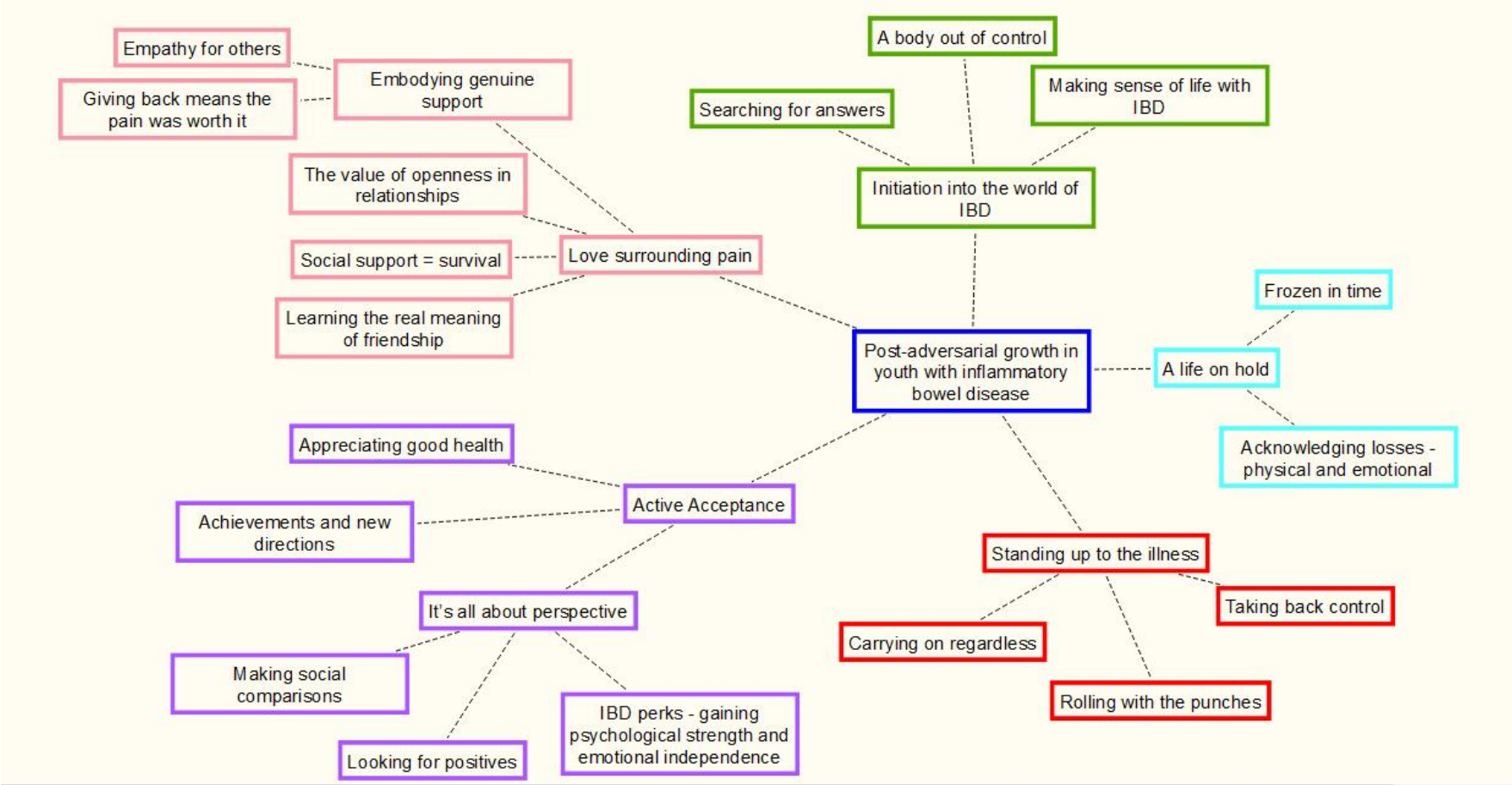
R Yeah, so it sounds like it was quite a difficult time then, particularly at such a young age. What can you remember much about what school was like after you were diagnosed? P In terms of sort of primary school, for myself it was difficult to sort of appreciate how ill I actually was, because I was always, like I said, a really outgoing sort of – I want to do everything, a really kind of active person, so I just kind of got on with it and I had my few close friends which – you know they're close friends when they don't care what you look like and they don't ask questions. Um, so it wasn't really – other than that one comment, in primary school, even though my appearance was very strange, it didn't really affect me that much in primary school, other than that one comment, and – until I went to secondary school and that's when it – I think it hit me how ill I was and other people were noticing. R Right, ok. So what makes you say that then, about high school? Tell me a bit about high school then. P So when I started year 7, um, I was quite ill, I'd lost a lot of weight, so I looked very skinny and very frail and it led to a hospital admission. Basically I ended up taking a week off of school just out of pure fatigue, um, and sort of that was really out of my character, because I was a bit of a nerd at school; I loved learning. So it's sort of when your mum picks up 'oh, something's not quite right' because she can't go to school. Um, and I basically went to have a shower and I collapsed in the shower. So I think I was unconscious for about 5 minutes and then um, got admitted to hospital with severe anaemia and dehydration, sort of seated in hospital for a week having blood transfusions and those kind of things - intravenous steroids and sort of a talk about changing other medication. Um, and I was on cyclosporine which is quite a strong immunosuppressant but it hadn't kept my flares at bay, but obviously starting school looking very frail and then re-joining school after I'd been in hospital on high-dose steroids and getting this moon face back and swollen tummy, um, people start to look at you and go 'oh, that's not what you looked like a couple of months ago, what's happened here?' And obviously going through puberty	Standing up to the illness Initiation into the world of IBD	Carrying on regardless A body out of control	

anyway is a difficult time for anyone, but add being on medications with horrible side effects just makes it that bit more difficult. Um, so it's the point where people would sort of make those odd little looks and comments and it might not be anything particular but if people look at you a certain way when you're self-conscious anyway, I think it hits you that much harder. And because I had no control at that point over the way I looked, I was very much 'I'm going to control my personality as much as I can, so I think I became quite – quite a pushover in school, because I was like 'I just want people to like me, because I can't be that girl who fits in because I'm pretty' and I fit in because I'm not stereotype girl, because I didn't fit a certain mould at that point.	Standing up to the illness	Taking back control	

Appendix. K Second Revision

Main Theme	Second Level	Third Level
Initiation into the world of IBD	Struggling with Ambiguity/Searching for answers	
	A body out of control	
	Making sense of life with IBD	
Putting Life on Hold	Frozen in Time	
	Acknowledging Losses – physical and emotional	
Standing up to the illness	Carrying on regardless/Denial	
	Taking back control/Taking Ownership of body	
	Rolling with the Punches	
You Can't Do It Alone	Learning the real value of friendship	
	Social support = survival	
	Openess in Relationships	
	Embodying Genuine Support	Empathy for others/recognition of the invisible
		Giving back means the pain was worth it
Active Acceptance	Finding New Paths	
	Making the Most of Your Health/Don't take Health for Granted	
	It's all about Perspective	Making social comparisons
		IBD Perks – gaining emotional independence/mental strength

Appendix L. Final Template Conceptual Map



Appendix M. Spread of Themes

Main Theme	Subtheme	3rd Level Theme	P1	P2	P3	P4	P5	P6	P7	P8	P9	P10	P11	P12	P13	P14
Initiation into the world of IBD	Searching for answers		x	x	x	x	x	x	x	x	x	x	x	x	x	x
	A body out of control		x	x	x	x	x		x	x	x	x	x	x	x	x
	Making sense of life with IBD		x	x	x	x	x		x	x	x	x	x	x	x	x
Putting Life on Hold	Frozen in Time		x	x	x	x	x	x	x		x	x	x		x	x
	Acknowledging Losses – physical and emotional		x	x	x	x	x		x	x	x	x	x	x	x	x
Standing up to the illness	Carrying on regardless		x	x	x	x	x	x	x	x	x	x	x	x	x	x
	Taking back control		x	x	x	x	x		x	x	x	x	x		x	x
	Rolling with the Punches		x	x	x		x		x	x	x	x	x	x	x	x
Love Surrounding Pain	Learning the real meaning of friendship		x	x	x	x	x	x	x	x	x	x	x	x	x	x
	Social support = survival		x	x	x	x	x	x	x	x	x	x	x	x	x	x
	The value of openness in Relationships		x		x	x	x		x	x	x	x		x	x	x
	Embodying Genuine Support	Empathy for others & recognition of the invisible	x	x	x				x				x		x	x

		Giving back means the pain was worth it		x	x	x					x	x			x	x
Active Acceptance	**Achievements and new directions**		x	x	x	x	x	x	x	x	x	x	x	x	x	x
	Appreciating good health		x	x		x	x	x	x	x	x	x	x		x	x
	It's all about Perspective	**Making social comparisons**	x	x	x	x			x		x	x	x			x
		Looking for the positives	x	x	x	x	x		x	x	x	x	x	x	x	x
		IBD Perks – gaining psychological strength & emotional independence	x	x	x	x	x	x	x	x	x	x	x	x	x	x

Appendix N. Supplementary Table of Quotes

Main Theme	2nd Level	3rd Level Theme	Quotes
Initiation into the world of IBD	Searching for answers		P8 "Yeah, I was quite worried and especially because I didn't really think that it was – it could be IBD so I just didn't really know what it was and then I didn't understand why I was feeling worse all the time and just the stomach pain mainly was um, got quite bad at some stages, so yeah, it was quite frustrating."
	A body out of control		P2 "I was on the toilet 20, 30 times a day and I couldn't make it out the door without needing the loo" P11 "I often never quite knew how my stomach would react. Sometimes I could eat things and be fine, sometimes I could eat things and be in an awful lot of pain afterwards – there was really no rhyme or reason to it." P14. "…because I had lost a stupid amount of weight, but for me to try and say – for me to try and say that I would never do that to myself, I almost wish I was doing this to myself because this is just my body, this is just what's happening…"
	Making sense of life with IBD		P1 "…I thought this isn't feeling better, I thought I was going to feel better. And then why – is this going to be the rest of my life? Is this the new healthy?"
Putting Life on Hold	Frozen in Time		P5 "I'd be cancelling – like I'd get invited to family events and I wouldn't go. Cancel like – if they say, if they're going to a restaurant, because I was getting pain after I'd eat and things like that, so that was a no-no. So there's missing out on like birthday meals and things like that." P7 "…but like I – I was super super keen to get back to school because I missed all my friends, I missed playing sports and everything, like I was so active before. Um and like when – them telling me that I couldn't do sport for three months after the operation was almost like a – worse news than actually having to have the operation itself…yeah, I missed quite a lot in years 9 and 10, um, so in year 9 I must have missed about 3 months and then in year 10 I would have missed about 4

			months…I loved doing all the like sports and drama and musicals and everything there and I missed my friends, I got like major FOMO, fear of missing out!" P9. "And um to start with the college tutors were like 'well do you want to study here, like where are you spending your time?' and I'm 'I've literally been on the toilet all this time'. And I remember like, oh, there was a few days when I had to go home because I just couldn't hack it, I just had to go home to bed." P14 "…I just remember my cousin and my sister sat on the stairs giggling, because I was 8, 9 years old and my mum had to come – come to the toilet with me and eventually and it – it sounds really silly reflecting on it now; at the time you don't realise how much it actually affects you"
	Acknowledging Losses – physical and emotional		P1 "…and it was really hard because all my friends went to uni, every single one and I had to stay at home, watch them go do all these exciting things and I couldn't" P12 "…I think I went – probably went through like a 6-month phase where I think I probably cried once a day, sometimes multiple times a day, it was just – just an outlet of like everything that had been going on…" P13 "…there's definitely days when I mope around or I think that like this is just so unfair and seeing like my friends graduate this time last year, I was definitely in a foul mood that entire day. I've already missed out on so much and now I'm missing on this big reunion for them…"
Standing up to the illness	**Carrying on regardless**		P11 "But once I had got a bit stronger and once I thought, ok, I'm probably at a level now I can play at a low competitive level, it was almost me, proving, almost telling the illness that actually, no, you're not going to take that away from me, I can still do it, ok not as well as I did, but I can still do it and, yeah, I've still got that, it was just kind of proving a point that I'm not going to let it win" P9."…I can't even say what did get me through because I don't even know myself, I just did…I think because I was brought up by a single mum, you know, I was brought up to be strong and

			independent, that's just how I am. Like, if I had something wrong then I will just literally try and get on with it as best I can, like, you know what I mean?"
	Taking back control		P1 "Um, and also kind of taking responsibility for your own health because I think a lot of people 'oh well I've got Crohn's I'm poorly and that's it' whereas I thought, no, I'm going to exercise and sleep a lot, I'm going to go vegan, I'm going to change my lifestyle..." P13. "I don't really like it when other people tell me I'll get there in the end, because I think…I just think I'd rather sort of know it myself and tell myself that I'll get there in the end." P14 "…because I had no control at that point over the way I looked, I was very much 'I'm going to control my personality as much as I can…"
	Rolling with the Punches		P11 "I had to pretty much give those two up (football and pool). Um, I also play table tennis at least once or twice a week and that was the thing I thought, ok, probably aren't going to be able to do this as well as I could before but it's something that it still doable. So my focus was, uh, from – uh, from almost a sense of achievement, something to be able to get back to some degree of where I used to be, um, so I focused on actually getting back to playing that." P13 "And I think especially with chronic illnesses like this it that it's never going to end, if that makes sense? It's not something like cancer or anything as serious as that where if you're cured you can think about life before and life after cancer, with something that's chronic, you have to think 'I need to sort of' – you have to roll with the punches, you have to take everything in your stride, or try to take it in your stride…"
Love Surrounding Pain	**Learning the real meaning of friendship**		P8. "…some people I knew less well, they would kind of shy away from addressing it or asking me about it and then I wouldn't really want to bring it up with somebody unless they, you know, say if I wasn't sure if they were wanting to ask about it. So in a way, yeah, but I think I just learned that some people, like some of my closest friends maybe understand it better than some of my more distant friends…"

			P9 "…finding my partner now, I'm like 'oh, my, is this something – something is going to happen – something bad is going to happen, he's not going to stick around. He's going to get fed up of me being at hospital all the time', but it's just not been like that at all. And even through the pregnancy, like during the surgery and that he sat with my mum the whole time and didn't leave my side and the same with when I've been admitted like on a few nights, he just sits by my bedside, even though he's been at work from 5 o'clock that morning and say we don't get home until 5 o'clock the following morning."
	Social support = survival		P9. "The days that I felt really low or really, really ill, then he would take me to the beach, um, try and get me some ice-cream, you know, like my dad would literally do anything he could to try and make me feel better." P11 "…but I was lucky as well, because a lot of my financial stuff – like I was getting credit card statements and stuff like that still coming through for things and subscriptions and stuff I was still signed on for, and my parents paid for that at the time and then I paid them back, when I was actually well and out of hospital."
	The value of openness in Relationships		P3 "Um I think its made me be very open with people. Um…yeah, I mean, I – probably because of the age I was diagnosed I sort of just sort of would speak about it with my friends and things because otherwise it would become like an elephant in the room" P10 "He understands that I have to cancel things at the last minute because I don't feel well, um, and I feel like I can be more open, now he understands, whereas before I couldn't, I'd hide more things which would then cause more anxiety, getting stressed and then be ill again, it's a vicious circle." P10 "…I think have my Crohn's has made me be even more honest, you can't hide it. You can't pretend that you're okay, so yeah, since that's happened, yeah, I think I've been more honest and open with people. You know, you've annoyed me, and I'll tell you why, because there's no point hiding it. My Crohn's is invisible, you know, my opinion is not."

	Embodying Genuine Support	**Empathy for others & recognition of the invisible**	P11 "if other people did things to almost – not take my control away, but to make things difficult I'd be getting sort of annoyed at them and now I just, again, just don't do that so much more because I realise that other people could have stuff going on that could be affecting things and not necessarily stuff that they want to discuss, but they could have things going on in the background."
		Giving back means the pain was worth it	P9 "And the next thing is I started a blog…I'd say recently in the past kind of year, it's gotten really good reviews and people have been sharing it and you know, I'll get a few messages being like 'oh I read your blog and it's so good and I didn't know that this was what' – because I don't just share about my journey, I share about other people's stories and be like 'oh my god, I thought I had it bad!' Like it's so good and you'll see them like in a few months on their facebook and they're like back to school or getting back to work and you're like 'oh my god that is so good.' Because I've been able to get them to the point where they are now, like being able to support them." P13 (about working in the medical profession and sharing own experiences) "I think it would just show that everything that I'd been through was worth it and like that there was a finish line that – at the moment it keeps getting pulled further and further away from me, and when I eventually get to it, and I'm hopefully sort of steady in my own health, I'd like to think that it was sort of worth it and I can help other people."
Active Acceptance	**Achievements and new directions**		P1 "I've done all sorts, I've done a skydive for Crohn's to raise money…my dream was to travel and help animals…because of that I went to T and went to a dog sanctuary and I'm moving over there in November to live"
	Appreciating good health		P1 "I'm going back in November to live because I don't know when I'm going to get ill again, I don't want to wait to get ill again and then not be able to go"
	It's all about Perspective	**Making social comparisons**	P14 "I went to SA for a part of it (travelling) and obviously you see everything from um, quite well to do people to the polar opposite and you think 'actually, on the grand scheme of things, what does it matter how big you are or how much hair you have on your face, ha.'"

		IBD Perks – gaining emotional independence/mental strength	P2 “At least I’m here, at least I’m sat outside in the sunshine, at least I’m not hooked up to things and that’s really positive…then you know, you’ve got to turn the positives – turn the negatives into positives”
		Looking for the positives	P2 “I know I’ll succeed and I know that I’ll get through it because I got through that life and death moment and that’s bigger than anything on this earth” P7. “And I think I am quite a naturally happy person. Everyone’s like ‘oh, why are you so happy all of the time?’ And I’m like it could be so much worse, you’ve just got to think of the positives.” P8. “…it’s a little bit almost, um, not comical but a little bit funny in a way because there’s 3 siblings and now we’ve all been diagnosed with it, so we kind of make jokes about it. But I would say in general it makes family life probably better or closer because everybody is kind of asking each other how they’re doing.” P11 “I think that’s something I learned through being in hospital. I saw lots of different attitudes of people in hospital – of people – people would actually whatever they were told would just be convinced, right, we’re going to get on with this and we’re going to get out of hospital and they generally did; and other people would get told something and would kind of sink into depression and in a few days they didn’t ever get out of hospital and I was just determined that was not going to be me.”