Volume I: Research Component

BODY IMAGE IN YOUNG PEOPLE WITH INFLAMMATORY BOWEL DISEASE

HEALTHCARE PROFESSIONALS WORKING WITH CHILDREN WITH A DUAL DIAGNOSIS OF TYPE 1 DIABETES AND COELIAC DISEASE

By

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OVERVIEW: VOLUME I

This thesis is submitted for the degree of Doctorate of Clinical Psychology at the University of Birmingham. It is presented in two volumes. Volume I includes a literature review, qualitative research paper and a public domain paper. The literature review uses a systematic review process to explore body image in children and young people with Inflammatory Bowel Disease (chronic, inflammatory intestinal conditions including Crohn's Disease, Ulcerative Colitis and Indeterminate Colitis). The original research is a qualitative paper, exploring the experiences of healthcare professionals working with children with a dual diagnosis of Type 1 Diabetes and Coeliac Disease. A public domain briefing paper is presented, which provides a summary of the literature review and qualitative paper, prepared for stakeholders in the research.

OVERVIEW: VOLUME II

Five Clinical Practice Reports are presented, which were completed during placements for the Clinical Psychology Doctorate.

Clinical Practice Report 1: The case of a 13 year old boy with a severe learning disability is presented. The difficulties with his self-injurious behaviour are formulated from behavioural and systemic theoretical models.

Clinical Practice Report 2: A service evaluation completed in a CAMHS team, auditing how the service was meeting the recommended criteria set in the NICE guidelines (2004) for self-harm.

Clinical Practice Report 3: A case study using a Cognitive Behavioural approach with a 29 year old man, experiencing depression and anxiety, during an inpatient stay on an acute psychiatric ward.

Clinical Practice Report 4: A single case experimental design based on the Newcastle Model, with a 74 year old man displaying behaviours that challenged on an older adult inpatient psychiatric ward.

Clinical Practice Report 5: A case study using Acceptance and Commitment Therapy with a 57 year old man, with low mood and adjustment difficulties in response to Chronic Kidney Disease and Chronic Pain. This case study was presented orally, an abstract is included.

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Body image in young people with Inflammatory Bowel Disease: A systematic review

Paper to be edited for submission to 'Health Psychology Review'

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ABSTRACT

Background: Inflammatory Bowel Disease (IBD) includes Crohn's Disease, Ulcerative Colitis and Indeterminate Colitis, all characterised by chronic, gastrointestinal inflammation. Symptoms can be painful and embarrassing and treatment can result in invasive procedures, side effects and surgery. Qualitative studies suggest that body image may be affected by IBD.

Aim: To review published quantitative papers with a measure of body image in children and adolescents with IBD.

Method: Electronic databases (PsycInfo, Embase, Medline, Web of Science, CINAHL) were used to systematically review the literature, identifying 19 papers. These papers were reviewed using a critical appraisal tool.

Results: The review discusses themes relating to body image including: type of IBD diagnosis; age at diagnosis; severity of symptoms; gender and treatment. Trends in body image are similar to healthy peers, particularly for those with mild or no symptoms. IBD can predispose the young person to risk factors, increasing their vulnerability to poor body image, including delayed growth, increase in severe physical symptoms and intrusive or demanding treatments.

Conclusion: Routine assessment of psychosocial factors, including body image, for young people with IBD is recommended. This review highlights methodological limitations in the current body of evidence, calling for further research to improve understanding of this area.

Keywords: *Inflammatory Bowel Disease, Crohn's Disease, Ulcerative Colitis, Indeterminate Colitis, Body Image, Children, Adolescents, Systematic Review*

INTRODUCTION

Inflammatory Bowel Disease

Inflammatory Bowel Disease (IBD) is an umbrella term for idiopathic, chronic, inflammatory intestinal conditions. These encompass: Crohn's Disease (CD), Ulcerative Colitis (UC) and Indeterminate Colitis (IC). IBD presents as an intermittent disease, with symptoms that vary in severity during periods of relapse and remission, with approximately 50% of patients experiencing relapse in any one year (IBD Standards Group, 2013). Symptoms of IBD are dependent on the phenotype of the disease and the section of the intestinal tract affected, however, symptoms typically include diarrhoea, constipation, incontinence, bowel movement urgency, abdominal pain, rectal bleeding, fever, loss of appetite and weight loss. Intestinal complications are associated with IBD, such as bleeding ulcers, bowel perforation, intra-abdominal abscesses, narrowing of the bowel, fistulas, perianal disease and an increased risk of colon cancer. IBD is additionally associated with extra-intestinal complications such as arthritis, osteoporosis and spondylitis (World Gastroenterology Organization, WGO, 2009). IBD is not to be confused with IBS (Irritable Bowel Syndrome); these are two separate diagnoses, with distinctly different aetiology, clinical presentations and treatment.

There is no known cure for IBD. The British Society of Paediatric Gastroenterology Hepatology and Nutrition (BSPGHAN, 2008) outline the treatment goals in paediatric IBD, which are to bring the disease into remission, manage relapse, maintain good nutritional status and improve wellbeing. Treatment options include enteral nutrition[1], corticosteroids, immunosuppressive agents and biological therapy. Surgery is also common, with 70-80% of

[1] A glossary of medical terms is provided in Appendix 1.

CD patients and 25-30% of UC patients requiring surgery in their lifetime (BSPGHAN, 2008; WGO, 2009). Treatment and management of IBD has improved in recent years, particularly with the introduction and licensing of biological therapies (such as Infliximab). These have been reported to significantly improve the course of treatment in IBD. Reported benefits include: inducing and maintaining remission, preventing complications, reducing corticosteroid exposure and reducing the need for surgery (Gould, 2013; Rutgeerts, Vermeire & Van Assche, 2009; McGinnis & Murray, 2008; Rungoe et al., 2013; Cassinotti, Ardizzone & Bianchi Porro, 2008; de Bie, Escher, & de Ridder, 2012).

IBD can be diagnosed at any age; however, the peak age of onset is between 15-29 years (Johnston & Logan, 2008). A prospective national survey of IBD in children under 16 years in the UK found rates of 5.2 per 100,000 individuals per year, with a mean age at diagnosis of 11.9 years in under 16 year olds. Data suggest that only a small proportion of children are diagnosed under 5 years old (Sawczenko & Sandhu, 2003). A study by Van Limbergen et al. (2008) suggests that IBD diagnosed in childhood may involve a more severe disease course with more extensive intestinal involvement compared to adults.

IBD associated complications

Complications specific for young people stemming from IBD can include growth delay and delayed puberty, associated with under-nutrition, inflammation and steroid treatment (Shamir, Phillips & Levine, 2007). Growth delay is reported amongst all forms of IBD, but is most common in CD, with a prevalence rate of 40-50% in children affected (Cezard et al., 2002). Steroid treatments have a number of other associated side effects, including: ‘cushingoid’ features (fatty deposits on the face causing roundness), reddish-purple stretch marks on the body and thinning of the skin. A ‘steroid hump’ may develop, which is a

fatty deposit in the middle-upper back. Also redness of the face, facial hair growth, rapid weight gain and acne may occur. Guidance recommends against long-term steroid use in young people with IBD, however, it remains a widely used treatment, particularly in the induction of remission (BSPGHAN, 2008).

In addition to the propensity for a more severe disease course in childhood, IBD presents young people with further challenges associated with developmental changes. Disease activity in IBD is linked to poorer psychosocial development, with a greater risk for depression, anxiety, poorer social functioning and lower self-esteem (Mackner, Crandall & Szigethy, 2006; Pinquart, 2012). Implications of the disease impact upon school life, leisure activity (Lindfred, Saalman, Nilsson, Sparud-Lundin & Lepp, 2012) and developing independence (Loonen, Grootenhuis, Last, Koopman & Derkx, 2002a). For children with chronic health conditions, the demands of their illness may also conflict with their developing social relationships, their desire to 'fit in' and their self-image (Suris, Michaud & Viner, 2004).

Self-concept

The mental perception we hold of ourselves is termed self-concept. Shavelson, Hubner and Stanton (1976) state that self-concept is influenced by our evaluations of academic, social, emotional and physical attributes. More recent research describes how the development of self-concept is influenced by expectations and judgments of others, genetic predisposition and interpersonal experiences (Bong & Skaalvik, 2003). Harter (1999) surmises that there is an abundance of terminology in the literature referring to the self, and draws attention to the distinction between global self-evaluations (such as self-concept) and domain-specific evaluations (such as physical appearance).

Childhood and adolescence is a phase of intense change, with emotional, cognitive, physical, behavioural and social transitions (American Psychological Association, 2002) all contributing to the individual's sense of self or self-concept. It is a time during which many young people strive to 'fit in', whilst also shaping their personal identity and image.

Body Image

Identity and self-concept include a domain of physical appearance or body image. Body image has been theorised by Cash (2004) as the thoughts, feelings and behaviours of one's body-related self-perceptions and self-attitudes. It is considered to be a multidimensional construct (Banfield & McCabe, 2002), informed by historical, cultural, social, individual and biological factors. Changes in bodily appearance in young people can influence body satisfaction and self-esteem, with groups of adolescents who mature early and those who mature late being at risk of poorer self-evaluation, due to them not being in 'the norm' group (Williams & Currie, 2000).

Adolescence is a time of increased self-consciousness (Sebastian, Burnett & Blakemore 2008). Therefore self-evaluation and comparison to others peaks during a period when sense of personal and social identity is rapidly forming. Festinger's Social Comparison Theory (SCT, 1954) can be used to offer insight into body image perceptions. The theory posits that individuals compare themselves to others and are also compared by others; similarities and differences are then used to evaluate their own standing. Body dissatisfaction has been reported to be higher for young people with chronic illness (Pinquart, 2013). Chronic illness and its treatments can often impact on physical appearance, and lead to restrictions on lifestyle, school attendance, social functioning (Michaud, Suris & Viner, 2007) and lower self-esteem (Pinquart, 2012). Increased body dissatisfaction in children with chronic illness can pose a high risk for engaging in unhealthy weight-loss practices (Neumark-Sztainer,

Story, Resnick, Garwick & Blum, 1995) and reduced self-care and dietary management compatible with their illness (Vlahou, Cohen, Woods, Lewis & Gold, 2008).

Qualitative research on body image and IBD

In a sample of 347 adults with IBD, 67% stated that having IBD impaired their body image. Body image was most affected in participants who were female and those that had undergone surgery (Muller, Prosser, Bampton, Mountifield & Andrews, 2010). Similarly, Brydolf & Segesten (1996) reported that young people with IBD have revealed feelings of being different (to their former self and peers), perceiving negative changes in their body image, embarrassment and diminished self-respect linked to diarrhoea and faecal incontinence. Furthermore, the authors found that dependency on being near a toilet and embarrassment of changing clothes around peers can restrict their activities. Nicholas et al. (2007) reported feelings of exclusion, vulnerability, illness uncertainty and lack of control in young people with IBD. Lack of control was specifically linked to their bodies, due to repetitive, intrusive investigations and violations of personal privacy. The authors found that adolescents with IBD have reported being more critical of their appearance than younger children, with boys having more concerns about their developing strength and height, and girls being more concerned about weight gain. Furthermore, adolescents viewed themselves in relation to perceptions they received from others, with many feeling their appearance was judged and criticised. Vlahou et al. (2008) found adolescent females with IBD were less satisfied with their bodies than males, and more positive body image was associated with better dietary self-management. A study looking at children and young adults following stoma surgery for IBD (surgically-created opening from the colon or intestine to the outside of the abdomen) revealed that despite generally positive results and good psychosocial adjustment,

patients had anxieties about leakage, noise, odour, social acceptability and sexual desirability (Lask, Jenkins, Nabarro & Booth, 1987).

Existing literature reviews

Body image of children within a range of nineteen chronic illnesses (including obesity, cystic fibrosis, scoliosis, cancer, diabetes, burns and kidney disease) has been reviewed using a meta-analysis of 330 studies (Pinquart, 2013), concluding that body image is reported as less positive than healthy children, but that the average effect size was small (g=-.30 standard deviation units). The size of differences in body image was illness-specific; the strongest differences of body image were reported in obesity, cystic fibrosis and scoliosis (g=-.79, g=-.50 and g=-.41 respectively). The authors suggest that this finding may be linked to these illnesses being visible to others; however, the moderating effect of treatments was not examined in this review. Since this review only included studies with active comparisons to healthy controls, only six studies of IBD were included. A small effect size (g=-.35) was found for the IBD sample, however a smaller sample was used compared to other illness samples (n=157) and the severity of disease activity was not reported.

Mackner at al. (2006) conducted a systematic review of psychosocial function in paediatric IBD. This review concluded that children with IBD are at increased risk of psychosocial difficulties (i.e., symptoms of depression, anxiety, poorer social functioning and self-esteem), but these problems are only clinically significant in a small number of children. Body image (assessed within quality of life measures) was cited in six articles; however, due to conflicting results no conclusions regarding body image were drawn. In a systematic review of psychosocial functioning in paediatric IBD, Ross, Strachan, Russell and Wilson (2011) summarised that health-related quality of life (HRQoL) is lower amongst this group,

with an increased incidence of self-reported symptoms of depression and anxiety. However, more specific conclusions were limited due to methodological flaws across studies and conflicting evidence for associations with psychosocial functioning. From this review, the authors recommend that body image in IBD is an area of interest, which warrants further consideration.

At the time of reporting, there has been no review of the literature specifically focussing on body image in children and adolescents with IBD. Body image and body dissatisfaction in the IBD literature is frequently found in HRQoL studies, of which there is a wealth of articles. This literature includes generic measures and disease specific measures to assess HRQoL, both of which have their strengths. Generic measures allow for comparisons with normative data, whereas disease-specific measures yield more sensitivity to aspects of health with regards to particular conditions (Wiebe, Guyatt, Weaver, Matijevic & Sidwell, 2003). The IMPACT questionnaire is an IBD-specific measure, developed for young people aged 8-18 years (Griffiths et al., 1999). It includes a body image scale regarding satisfaction with height, weight and general appearance. The most newly developed version is the IMPACT III, developed by Otley et al. (2002).

Summary and aims

In summary, the consequences of the symptoms and treatments of IBD in children and adolescents can result in severe abdominal symptoms, invasive procedures, possible surgery and unpleasant treatment side effects. In addition to these challenges, under-nutrition and poor growth and development is associated with IBD. All of these factors contribute to psychosocial risk factors in the developing child. Chronic health conditions in childhood are posited to influence poorer body image perception in children and adolescents. Considering

the theories of body image, it seems plausible to hypothesise that IBD-related issues could serve as risk factors in the establishment of poorer body image in young people. There certainly exists a small body of qualitative research on this topic, previously discussed, which provides evidence for this hypothesis (Brydolf & Segesten, 1996; Lask et al., 1987; Nicholas et al., 2007; Vlahou et al., 2008). However, at the time of reporting, no published systematic reviews exploring literature on body image in children with IBD is available. Body image has been referred to as a factor of interest within the context of wider reviews on HRQoL in paediatric IBD; however the conclusions to date remain ambiguous. These reviews have included only small numbers of papers reporting on body image, too few for robust conclusions. There has also been an extensive period over which papers have been published. Older studies may not be representative of current experiences of young people with IBD, based on current treatments. In addition, previous reviews have not specifically reported body image findings in the context of factors such as severity of illness and age.

Therefore the aims of this systematic review are to:

- Assess and summarise the current body of evidence, which reports quantitative data on body image in young people with IBD.
- Review the quality of the evidence of body image in young people with IBD.
- Derive recommendations about how body image issues can be addressed in young people with IBD.

METHODOLOGY

Search Strategy

Five electronic databases were used to search the literature; EMBASE (1974[2]-2013 December 19); MEDLINE (1946- December week 3, 2013), PsycINFO (1967 – December week 3, 2013), Web of Science (1960 – 19 December 2013) and CINAHL (1998 – 19 December 2013). Search criteria included combinations of three elements: Body Image; Inflammatory Bowel Disease; and Child. Terms were also searched using Medical Subject Headings (MeSH). Since the databases offered different MeSH terms, each database was interrogated to compile a thorough list of relevant search terms (Appendix 2). The 'Tests and Measures' function on PsycINFO was additionally used to identify measures with a body image scale. Names of measures were added to the search if these were not already covered by the existing search terms.

[2] To gather the full breadth of papers on this topic for a thorough understanding of the published literature, initial searches included a broad timeframe. The date was then manually sorted according to the dates set in the inclusion criteria.

Table 1

Inclusion and Exclusion criteria

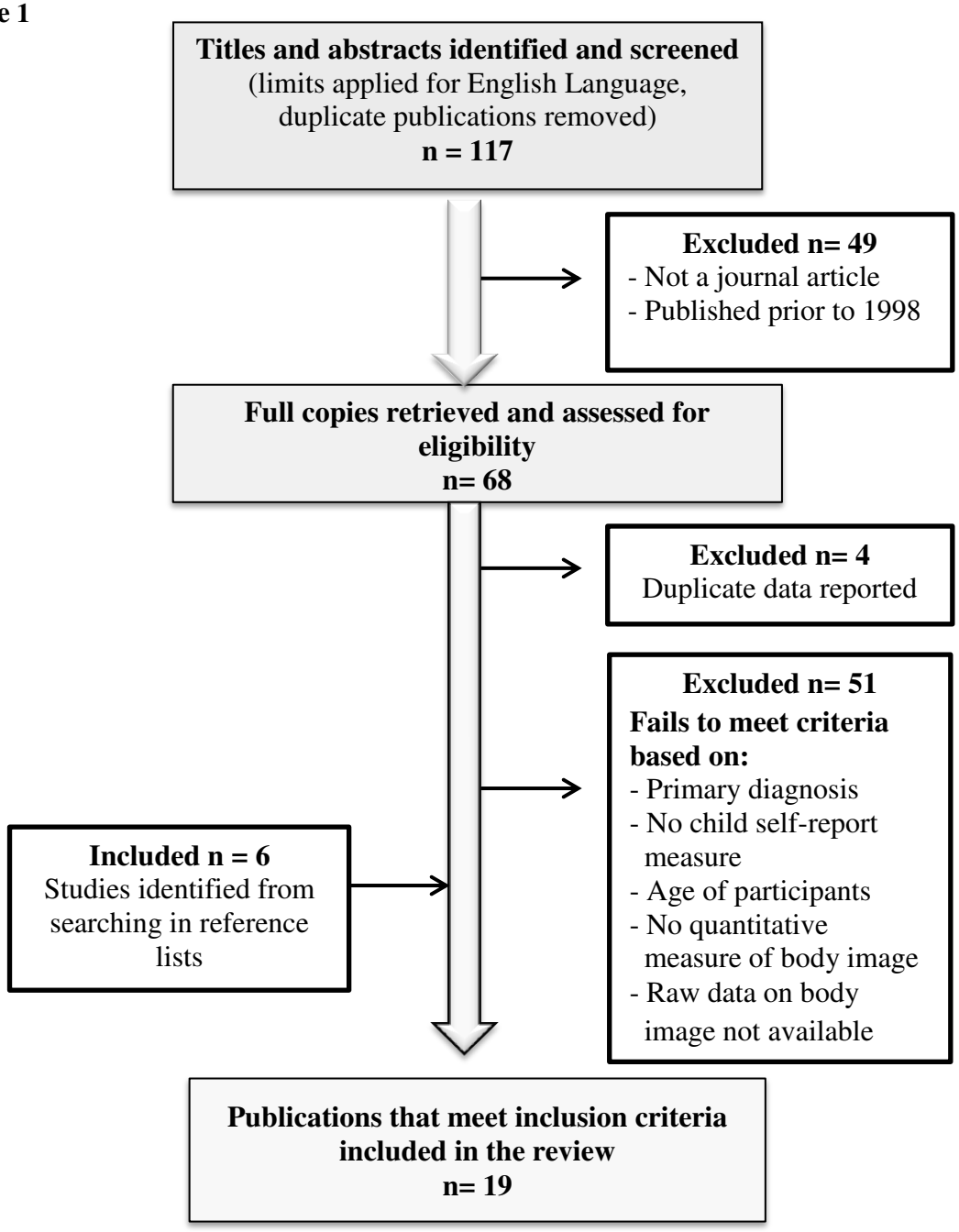
Inclusion Criteria	Exclusion Criteria
• Child self-report measure of body image. • Children aged 0-19 years. • Children with a diagnosis of Inflammatory Bowel Disease (Crohn's Disease, Ulcerative Colitis, Indeterminate Colitis). • Quantitative data provided. • Original articles. • Articles published from 1998[3] to present.	• Do not include a child self-reported measure of body image. • Age of participants is over 19 years, or separate data on those participants 19 or under is not provided. • Participants without a diagnosis of Inflammatory Bowel Disease. • Participants identified as having co-morbidities with other chronic physical health problems. • Qualitative research design. • Not published in English language. • Papers published prior to 1998. • Not an original article (i.e. review, editorials, letters, conference abstracts or case studies).

Additional papers of interest were found through the reference lists of articles obtained through the electronic search. Where papers did not report the scores of body image, the authors were contacted directly to request this information. In three cases, these data were not available; these three papers were identified as potential articles of interest, but could not be included in the review.

Figure 2 shows the search strategy employed to elicit the final set of nineteen articles. A summary of the articles included is provided in Table 2. An index of measures is provided in Appendix 3, this provides the full name and a brief description of the measures stated in Table 2, which may be used as a reference guide.

[3] This date marks the initial licensing and use of biological therapies, as detailed previously; this treatment has led to significant improvements in the management of IBD.

Figure 1



Search Strategy

SEARCH RESULTS

Table 2

Summary of Included Studies

Authors, Year	Study Type, Aims	Sample	Intervention (if applicable)	Measures Used (body image in bold)	Key Findings	Limitations
Health related quality of life (HRQoL) studies.						
Herzer, Denson, Baldassano & Hommel. 2011 *Duplicate data in Gray, Denson, Baldassano & Hommel (2011).* United States	HRQoL study. Relationship between family functioning and HRQOL.	n= 62, 13-17 years Male n=35 <u>CD n=49</u> - 92% inactive – mild symptoms - 8 % moderate – severe) <u>UC n=13</u> - 61% inactive - 31% mild and responding to treatment - 8% active disease but unresponsive to treatment)	No intervention. Treatment details provided.	-PCDAI -LCAI -FAD -IMPACT III (modified using 4 subscales with good-excellent reliability)	- Elevated family functioning difficulties associated with lower HRQoL. - Body image mean score[4]: 11.32 (2.41) - Significant associations between disease severity and all Impact scores (body image r= -0.32, p=0.01) Gray et al. add: - Internalising symptoms (social withdrawal, somatic complaints, anxiety, depression)	- Unrepresentative sample, predominance of inactive/mild status, CD, Caucasian and adolescent age, outpatient group. - Possible sample bias from volunteers/payment system; clinically elevated family functioning at baseline. - Body image was not correlated with family functioning. - LCAI used as opposed to PUCAI for UC group.

[4] Possible score range is between 3-15, with 15 representing more positive rating.

Herzer et al. continued		All taking anti-inflammatory medication. Recruited from 2 sites. Financial compensation provided.			strongly correlate with disease severity and predicted HRQoL. -Higher levels of externalising symptoms (behavioural problems and aggression) associated with lower HRQoL.	
Hill et al. (2010) Australia	HRQoL study. Investigate time since diagnosis and QOL in children with CD. To determine most significant predictions of QoL.	n= 41, 7-17 years CD Male n=32 Mean disease severity: 13.7 mild[5] From hospital database. Newly diagnosed and existing patients.	No intervention. Standard treatment course. Details not provided.	-PCDAI **-IMPACT III**	- Body image significantly higher in girls (t=3.09, p<0.01) - Age significantly related to body image (r=-0.26, p<0.01)). Increasing age associated with decreased body image - Height significantly related to body image (r=0.41, p<0.01)) - Disease activity significantly negatively correlated with QOL (Inc. body image p<0.01)	- Small numbers in some treatment groups (enteral nutrition n=7, this group also had significantly higher disease activity & lowest height, weight and BMI scores). - Causal relationships difficult to assess. - Some data sets discussed but not provided in article e.g. shorter children had lower body image, QoL improved with increasing height Z score.

[5] PCDAI disease severity classifications: ≤10 remission; 11-29 mild disease activity, ≥moderate-severe disease activity

Hill et al. continued					- No significant association between body image and disease duration. Body image[6] lowest for those diagnosed <6 months (10.6), improving to 11.7 after 5 years. - Poorer body image for those receiving enteral nutrition (mean 10.1 (+-2.7, p<0.05), followed by immunosuppressant's + steroids (10.8 +-2.2, p<0.05)	- Small number of girls (n=9) represented.
Kilroy, Nolan & Sarma. 2011 Ireland	HRQoL study. To obtain a profile of IBD related QOL.	n=79, 9-17 years Male n=46 CD n=41 UC n=23 IC n=12 Postal recruitment. Identified from clinic databases.	No intervention. Details of current medical treatment not provided.	-SCAS **-IMPACT III**	- Anxiety level significant predictor of QOL (p=0.001) - Qualitative measure: Feelings of anger and embarrassment. - Body image mean 3.56[7] (SD 0.76) - IMPACT-III high internal reliability (alpha=0.90)	- Qualitative feedback suggested wide variability in symptoms - Symptom severity not measured - Possible response bias (Postal measures) - Different scoring structure of IMPACT-III to other studies makes comparisons difficult.

[6] Possible score range is between 3-15, with 15 representing more positive rating.
[7] Answers transformed into a score ranging from 1-5, with higher scores denoting more positive rating and a score of 3 representing the mid-range answer.

Loonen, Grootenhuis, Last, Koopman & Derkx (2002a) *Duplicate data in Loonen et al. 2002b)* Netherlands	HRQoL study. Impact of IBD on HRQOL using a generic and disease specific measure.	<u>IBD Group</u> n= 83, 8-18 years CD n=41 UC n=40 IC n= 2 Male n=45 Disease activity: mild n=50 moderate n= 19 severe n=12 unknown n=2 Age (years): 8-11 n=18 12-15: n=41 16-18: n=24 Postal recruitment from 2 hospital databases. <u>Reference sample</u> n=1810 Without chronic condition Data taken from validation study of TNO-AZL (Vogels et al., 2000)	No intervention. Details of current medical treatment not provided.	TACQOL **-IMPACT II (NL)** -Symptom severity card	Body image mean 71[8] - CD 71 (+/- 22.8) - UC 70 (+/- 20.3) -12-18 years groups had significantly impaired HRQOL on TACQOL compared to healthy peers. - Younger children (8-11 years) had comparable QoL scores to healthy peers. - Impact-II (NL) discriminated well between patients in all age groups, with varying disease activity. - Body image significantly lower with increased disease activity ($p<0.05$) and increased disease severity ($p<0.05$). - Body image domain: cronbach's alpha 0.65 (satisfactory).	- Fewer children in the younger group, they also had lower symptom scores (11 had inactive or mild disease) - TACQOL has no body image scale; therefore this domain could not be compared. - Symptom Card is not a validated measure. - 7 week interval between test re-test. HRQoL may have changed within this time frame. - Responsiveness to change of IMPACT-II (NL) requires more vigorous testing in a longitudinal study design.

[8] Scores transformed to a 0 -100 scale, with higher scores representing more positive ratings.

Mackner & Crandall (2005a) *Duplicate data Mackner & Crandall (2005b)* United States	HRQoL study. To compare psychosocial functioning of children with IBD (in remission or mild disease activity) to healthy children	<u>IBD group</u> n=50, 11-17 years CD 76%; UC 8%; IC 16% Male 62% Disease activity: remission 62% mild 32% moderate-severe 6% Identified via Gastrointestinal clinic. <u>Control</u> n=42, 11-17 years no chronic condition Male 52% Identified via ambulatory clinics & employees	No Intervention. Current treatment details not provided, however hospitalisation and steroid use were shown.	- YSR - CDI - RCMAS **- PHSCS** - CSI - SSQ - PCDAI	- No significant difference on self-concept scale. - 20% IBD group had clinically significant behavioural/emotional symptoms (similar to control group). Physical appearance scores[9] IBD group 56.9 (9.97) Healthy 58.1 (9.63) - No significant difference between physical appearance scores in IBD and healthy group. - Mackner et al. (2005b) add: better family functioning and child coping strategies correlate with medication adherence.	Predominance of inactive mild disease status (94%), may be less representative of moderate-severe disease activity. - PCDAI used for all IBD subtypes (this measure was developed for CD).

[9] Piers Harris Self Concept Scale, T scores are provided with a mean of 50 and standard deviation of 10. T score lower than 34 is clinically significant.

Ogden et al. (2008) United Kingdom	HRQoL study. To identify changes needed to adapt the IMPACT for use in British children with IBD and assess two versions of the scale.	n=20, 8-16 years Male n=11 CD n=17 UC n=2 IC n=1 Recruitment details not provided.	No intervention. Details of current medical treatment not provided.	**IMPACT-UK** (two versions trialled, one with a VAS and one with a likert scale)	Overall body image scores[10]: Likert scale =60, VAS =65. These scores were significantly different (p=0.01) - Body image scores were higher for males than females and higher for 8-12 years age group than 13-16 years on both versions of the scale. - The Likert scale was preferred by participants.	- Further validation needed to ensure instrument is reliable and valid. Difference between scores on body image suggests unreliability of this domain. - Small sample, predominance of CD. - Details of recruitment were not provided. - Disease severity was not measured. Therefore representativeness of sample unknown.
Otley et al. (2006) United States	HRQoL study. Analysis of HRQOL from diagnosis to 12 months post-diagnosis.	n= 218, 9-17 years CD n=167 UC/IC n=51 Male n=123 Disease severity: mild n=57 moderate n=120 severe n=36.	No intervention. Details of medications, intervention or surgery provided and assessed alongside QoL scores.	**-IMPACT II** - Physician Global Assessment of Disease Severity	- Significant improvements in HRQOL during the 12 months from diagnosis (p<0.01). - Differences in body image scores[11] from baseline to 12 months not significant. Baseline 14.5 (4.6) 6 months 15.3 (4.8)	- Statistically significant changes reported, not clear if they are also clinically significant. - Some findings are discussed but the data are not reported.

[10] Scores transformed to a 0 -100 scale, with higher scores representing more positive ratings.
[11] Based on a 7 point Likert scale, scores range from 3-21, with higher scores representing more positive ratings.

Otley et al. continued		Data analysed from the 'Pediatric IBD Registry' (a large research programme in North America)			12 months 16.2 (5.1) - All other IMPACT domains showed significant change from baseline to 6 months ($p<0.01$) - Disease severity did not reach statistical significance as independent variable for body image ($p>0.05$) - Only age was a significant predictor of HRQoL 1 year post diagnosis ($p<0.01$)	- IMPACT was not consistently completed in close proximity of the disease activity assessment.
Perrin et al. (2008) United States	HRQoL study. Survey of children with IBD. Recruitment from 6 gastroenterology centres in the US. Randomly recruited using numbers generated from a database.	n=220, 8-17 years 8-11 n=39 12-15 n=94 16-18 n= 86 CD n=161 UC n=59 Male n=108 Diagnosed >6 months. Inactive/ mild symptoms Recruited from 6	No intervention. Details of current medical treatment not provided.	-**IMPACT II** -PedsQL -PCDAI - UCS	Body image mean scores[12]: overall 68.1 (SD 19.6) CD 67.9 UC 68.6. - Body image scores lower with higher disease activity. This was more significant in CD group ($p<0.01$) than UC ($p=0.02$) - Body image factor analysis revealed borderline	- Symptom severity data not consistently collected at the same time as QoL data, this may have changed during the time period. - UC symptom data collected using a measure (UCS) with no previous use or validation. - Predominance of CD

[12] Scores transformed to a 0 -100 scale, with higher scores representing more positive ratings.

Perrin et al. continued		IBD centres. Participant numbers based on statistical recommendations for factor analysis.			acceptable reliability (0.63). Authors suggest adding additional body image items may improve reliability.	and low disease activity in sample. - Participants not categorised into disease severity groups for comparison.
Van der Zaag-Loonen, Grootenhuis, Last & Derkx. (2004) Netherlands	HRQoL study. Coping styles of adolescents with IBD and healthy peers. Exploring the association between coping style and HRQOL.	<u>IBD Group</u> n= 65, 13-17 years Male n=45 52% CD, 45%UC, 3%IC Median disease severity: 8.5 (scale of 4-17, higher score=more severe) Postal recruitment. Identified from databases at 2 hospitals. <u>Reference sample</u> n= 660, 13-15 years no chronic condition 47% male Normative data from UCL-A (Bijstra et al., 1994)	No intervention. Details of current medical treatment not provided.	-UCL-A -CCSS-c **-IMPACT II (NL)** - Disease symptoms	- Adolescents with IBD use more avoidant coping styles than healthy peers ($p<0.001$). - HRQOL associated with disease related coping styles and disease activity. - Disease activity significantly associated with body image ($p<0.05$) - Coping styles not significantly associated with body image.	- Raw scores for HRQoL domains not provided, comparison of body image scores with other studies was not possible. - CCSS-c was an invalidated measure at the time of use.Validated measures of disease symptom and severity not used. - Reference sample taken in 1994, may not be comparable to youth 9 years later. - Postal recruitment with a 64% response rate. Possible response bias.

Werkstetter et al. (2012) Germany	HRQoL study. HRQoL in paediatric IBD in relation to physical activity compared to a healthy control group.	<u>IBD Group</u> n=39, 7-19 years CD n=27 UC n=12 Remission/ mild activity Male n=24 Recruited from hospital database. <u>Control</u> n=39 No chronic illness Matched for age and gender. School peers of IBD group.	No intervention. Current medical treatment details were provided.	-KINDL -**IMPACT III** -PCDAI -PUCAI - SenseWear armband QoL measured using the IMPACT for the IBD group and the KINDL for the control group.	- Median body image score[13] in IBD group: 80 (60-87). No body image data from control group. - IBD and control group did not differ on overall QoL. -Trends: reduced levels of activity (p=0.058) and reduced steps (p=0.064) in IBD group. - IBD group = lower physical activity levels (not statistically significant.) - Reduced lean body mass in IBD group (p<0.05).	- Armband suggested to overestimate low levels and underestimate high levels of activity- questioning reliability of results. - Design does not allow for conclusions regarding physical activity and QoL domains. - KINDL provides different domain scores, no body image scale, therefore IBD and control could not be compared on this measure. - Associations between lean body mass and body image were not explored.
Intervention Studies (medical interventions)						
Afzal et al. (2004) UK	Intervention study. Change in QOL,	n= 26, 12-16 years Male n=16 CD	Treatment with enteral nutrition	-**PCDAI** -IMPACT II	- In 8 weeks, significant improvements were seen in QoL scores on all domains	- Effect sizes are not provided. - No follow up to see if

[13] Scores transformed into a 0-100 scale, higher scores indicate more positive rating.

Afzal et al. continued	disease activity and intestinal status after treatment in Crohn's Disease.	All active symptoms Disease severity: Mild n=4 Moderate-severe n=21 Recruited from multiple UK sites at the point of diagnosis.			(p<0.05). - More positive rating of body image[14] was found following treatment (p<0.01) Pre-treatment 52 Post-treatment 72 -Clinical remission observed in 23 participants.	effects were maintained. - Less generalisable to younger children. - Participants were enrolled at the point of diagnosis; it is not clear if the same treatment in patients further along their course in managing the disease would demonstrate similar results.
DeBoer, Barnes, Styglels, Sutphen & Borowitz (2012) United States	Intervention study. Changes in Inflammatory, hormonal and QoL following Infliximab treatment.	<u>Symptomatic group</u> n=14, 9-22 years CD n=12, UC n=2 Male n=8 Mild symptoms only <u>Asymptomatic group</u> n=10, 9-22 years CD n=9, UC n=1 Male n=4 Body image was only measured in	Infliximab-biological therapy over 14 days.	**- IMPACT III** - PCDAI IMPACT measure was only used for the 9-17 years age group. Therefore the body image data provided are for this group.	<u>9-17 year age group:</u> No significant difference between body image scores for symptomatic and asymptomatic group at baseline or Day 14. <u>Symptomatic group:</u> Body image[15]: Baseline 70.4, Day 14: 71.7 (p=1.00) <u>Asymptomatic group:</u> Body image: Baseline 71.4, Day 14: 73.3 (p=0.67) Following treatment at day	-PCDAI calculated only for patients with CD. -Groups assigned based on researcher questions rather than validated measure of symptoms. -Symptomatic group had mild symptoms only. -Some data discussed but not reported in article. -A longer follow up may

[14] Scores transformed to a 0 -100 scale, with higher scores representing more positive ratings.
[15] Mean scores were converted in percentage scores.

DeBoer et al. continued		the 9-17 years age group, demographics for this age group not provided. Recruited from US clinic, scheduled to start Infliximab.			14, significant increases found in emotional symptoms, and social functioning ($p<0.05$)	be appropriate for QoL factors, 14 days is a limited time to show change. -Confounding variables such as serum and antibody levels of infliximab at baseline and follow up not assessed. -Unclear how many participants were in the 9-17 year old age group.
Malik et al. (2013) Canada	Intervention study. Compare HRQOL of procto-colectomy patients with those treated with conventional therapy. To determine factors that influence HRQOL in UC.	10-18 years UC Recruited from hospital database. <u>Surgical Group</u> n=33 Mean age 17.9 years (3.7) Male n=21 Disease severity: mild n= 0 moderate n=6 severe n=27	Details of current medication for each group provided.	**- IMPACT III** - IBDQ - PUCAI - HRQL - EuroQoL - EQ-5D/VAS HRQoL measured with IMPACT for ages 10-17 years (n=65) and the IBDQ only in	- No significant difference on Body image scores[16] between surgical and nonsurgical groups ($p=0.54$) Nonsurgical 11.7 (2.6) Surgical 11.3 (2.1) - No significant differences on HRQoL for surgical and nonsurgical group($p=0.09$). - Higher QoL scores in patients who are; in	- Despite IMPACT being designed for under 18 years, a separate measure for the 18 years olds made comparisons problematic. - IBDQ used in patients aged 18; however this had no body image scale. -The two groups showed significant differences between; age ($p<0.005$);

[16] Based on 5 point Likert scale. Subscale scores range from 3-15, with 15 representing a more positive rating.

Malik et al. continued		<u>Non-surgical</u> n=55 Mean age 14.6 years (4.3) Male n=25 Disease severity: mild n= 13 moderate n=22 severe n=20		patients > 18 years.	remission (p=0.05); younger (P=0.06); lower disease severity (p<0.001) and less likely to be on medication (P<0.05). - Females with higher QoL had menses onset at younger age (p<0.05), and lower menstrual irregularities (p=0.01). - Lower QoL in patients who had experienced weight loss (p<0.05).	disease duration (p<0.05) and non-surgical group had significantly more patients with moderate symptoms (P<0.005).
Smith, Field, Bingaman, Evans & Mauger (2013) United States	Intervention study. Pilot clinical trial, evaluating treatment for CD. Participants randomised to treatment or placebo, cross-over design.	n=14, CD 8-17 years Male n=5 All moderate/severe disease activity Power calculations to reach participant number required. Recruitment from single US site.	Opioid antagonist, Naltrexone for 8 weeks, with a further 8 weeks of treatment as usual. Details of other medications are provided.	-PCDAI -**IMPACT III** - Physical examination - Blood chemistry	- Significant reductions in disease activity after treatment (p<0.005). - Baseline mean body image score[17]: Treatment group 10.7 Placebo group 10.8 - No significant difference between baseline and treatment for body image.	- Despite power calculations, it is a smaller sample. - Cross over design, therefore all participants knew to expect treatment during the study, which may have influenced patients' perceived improvements. - 16 week data collection

[17] Based on a 10 point Likert scale. Scores range from 0 to 21. With higher scores reflecting more positive ratings.

Smith et al. continued					- Significant differences after treatment for systemic CD symptoms and social well-being ($p<0.05$)	period is a small time period to allow for changes in HRQoL/ body image. - Recruitment details not provided, it is difficult to judge how representative the sample is.
Intervention Studies (psychosocial intervention)						
Grootenhuis, Maurice-Stam, Derkx & Last, (2009) ***Duplicate data in DeBoer, Grootenhuis, Derkx & Last. (2005)*** **Netherlands**	Intervention study. Effectiveness of a psychoeducational group for adolescents with IBD. Outcomes measured at baseline, 0-6 weeks post intervention and 6-8 months post intervention.	<u>Intervention Group</u> n= 22, 12-18 years CD 77% UC 23% Diagnosed <6 months Male n=13 Recruited from hospital database, <u>Control Group</u> n=18, 12-18 years Male n=6 CD 61%: UC 22%; IC 17% IBD patients who	<u>Group intervention:</u> 'OK Programme', 6 sessions. <u>Group aims:</u> -Psychoeducation -Role playing for health consultations -Relaxation -CBT based positive thinking & thought challenging Details of current medical treatment	-**Dux-25** -**SPAA** -STAIC -CBCL - Cognitive Control Strategies Scale	• SPAA Physical appearance mean scores[18]: <u>Intervention group:</u> baseline 13.0, post intervention 15.5 <u>Control:</u> baseline 13.4, post intervention 13.8 • Dux-25 body image mean scores[19]: <u>Intervention group:</u> baseline 55.4, post intervention 68.9 <u>Control:</u> baseline 60.0, post intervention 59.0.	-Data 0-6 weeks post intervention not provided. -Response rate of 46%, may suggest response bias, those more proactive in receiving support took part or those faring better chose not to take part? -Relatively small sample size and dropout rate (n=8) at follow up. Participants who dropped out had better baseline

[18] SPAA physical appearance, possible score range 6- 24
[19] Scores range from 1 - 100, with 1= very bad to 100=very good

Grootenhuis et al. continued		lived too far away to participate in intervention served as the control. No significant demographic, medical or outcome variable differences at baseline.	not provided.		-6-8 months post intervention, physical appearance more positive ($p<0.01$) & body image more positive than baseline ($p<0.05$) -Physical appearance correlated with physical functioning (0.74*) and emotional functioning (0.63*). -Physical appearance correlated highly with HRQoL (0.72*) and anxiety (-0.55*). -Self-esteem correlated highly with HRQoL (0.77*) and anxiety (-0.63*). -Social functioning, physical appearance and self-esteem= high correlation with internalising, externalising and behavioural problems (all $r\geq0.3$). *$r\geq0.3$	scores for behavioural-emotional functioning. -SPAA- not an accurate measure for children with chronic illness (Aasland & Diseth, 1999) -Disease activity and severity not monitored. -Dux-25(QoL instrument) had not been validated.

Shepanski et al. (2005) United States	Intervention study. Prospective analysis of HRQoL in children and adolescents with IBD attending an IBD summer camp.	n= 61, 9-16 years CD:UC ratio 2:1 Male n=11 Participants recruited from IBD camp.	Week long IBD summer camp. No psychosocial intervention. Details of current medical treatment not provided.	-**IMPACT II** - STAIC Data collected on first and last day of camp.	- Overall body image scores[20] showed small (non-significant) improvements post camp: Pre-camp: 12.6 Post-camp: 13.2 - All age groups showed small improvements in body image scores, apart from the 15-16 year group. - Total QOL, Social functioning, bowel symptoms, treatment and interventions showed significant differences pre and post camp ($p<0.05$).	- Clinical significance of results not indicated - Not all participants returned questionnaires, possible sample bias. - Large sex differences in sample. No disease severity measure, minimal characteristics of sample provided. - Those attending the camp may be more inclined to actively seek information and support than other IBD patients.
Evaluation of Psychometric Properties						
Abdovic et al. (2013) **Croatia**	Evaluation of psychometric. IMPACT-III measure adapted for Croatian children.	n= 104, 14-18 years Male n=49 CD n=74 UC n=30 Diagnosed for >6 months Disease severity:	None	-PCDAI -PUCAI **-IMPACT III (HR)** -PedsQL	Mean Body image score[21]: 77.1 - Inactive symptoms: 78 - Mild: 76.6 - Moderate-severe: 71.9 Body image domain-	Body image subscale in this adapted version had only borderline reliability. Possibility of sample bias in those who returned the

[20] Based on a 10 point Likert scale. Scores range from 0 to 21. With higher scores reflecting more positive ratings.
[21] Scores transformed to a 0 -100 scale, with higher scores representing more positive ratings.

Abdovic et al. continued		-Inactive n=59 -Mild n=37 -Moderate-severe n=8 Identified from hospital database.			borderline acceptable internal reliability (0.64). HRQoL score highest for quiescent IBD, significantly higher than those with moderate/severe symptoms ($p<0.001$)	questionnaire.
Hutchings et al. (2007) United Kingdom	Evaluation of psychometric. The development and validation of MMQL-UK to assess HRQOL in children with chronic conditions. Including Asthma, Diabetes, Cancer, Care, IBD and healthy controls.	Total n=1238 <u>IBD Group</u> n=69 8-18 years IBD diagnosis Recruited from clinic database. <u>Healthy control</u> n=824 Recruited from schools across the UK.	None. Details of current medical treatment not provided.	-**MMQL** -PedsQL	- Physical appearance mean score[22] IBD=67.8, Control=69.8 - Physical appearance range 61.5 (cancer) to 77.1 (Diabetes). - Appearance scale score lower (not significantly) than control group for all conditions others than diabetes. - Compared to control group, none of chronic condition groups scored significantly different on physical appearance.	- Demographics not provided. - Little detail on inclusion/ exclusion criteria and recruitment. - Differences within chronic condition groups were not reported e.g. variable such as disease severity, treatment

[22] Higher score represents more positive rating.

Hutchings et al. continued					- Adaptations were made to the MMQL to increase suitability for UK children. - Good internal reliability found for all components of the MMQL-UK apart from physical functioning.	
Lowe, Kenwright, Wyeth & Blair (2012) New Zealand	Evaluation of psychometric. Cross-cultural validation of IMPACT-III (via debriefing interview) with paediatric patients with CD.	n=16, 9-18 years CD Male n=8 Mean disease severity (mild) Postal recruitment. Identified from hospital database and Crohn's & Colitis Support Group.	No intervention. Details of current medical treatment not provided.	**-IMPACT III** -PCDAI -NZDep06	- Body image mean score 9.8[23] (+/-3.1) - Positive correlation: disease duration and QoL ($p<0.05$) - Negative correlation: disease activity and QoL ($p<0.05$) - No difficulties in interpretation of body image questions. - Moderate to strong negative correlation between PCDAI and HRQOL ($p<0.05$) **Qualitative feedback:** -Visual reminders of CD	- Small sample size. - Possible selection bias. - Possible bias in response: Half completed questionnaires via post, half came to clinic and met with the researcher. - Disease severity scores taken from most recent outpatient appointments. Length of time since these differed, therefore could affect validity. - Data not provided for associations between body image and: age; disease duration;

[23] Possible score range is between 3-15, with 15 representing more positive rating.

Lowe et al. continued					(e.g. 3) caused embarrassment. - Not knowing others with IBD - Treatment regimen tiring - Need for social support.	severity.

Description of Studies

The publication date of studies included in the review ranges from 2002-2013. The countries of origin include Croatia (1), United States (8), United Kingdom (4), Netherlands (3), Australia (1), New Zealand (1) and Germany (1). The age ranges of participants were 7-19 years; all included both male and female participants. The majority of the studies included a mixture of IBD diagnosis (14), however, in the remaining studies, exclusive diagnoses of CD (4) and UC (1) were investigated. All of the participants were drawn from clinical populations, recruited from the IBD database at their clinic or hospital (18), or from an IBD register (1). The question of how representative these samples were on the basis of diagnosis can be addressed in terms of the distribution across diagnostic categories. The most prevalent of the IBDs is CD, followed by UC, with IC seen in much fewer cases. This trend was reflected in 6 of the 14 studies that reported a mix of IBD diagnoses.

The details provided on the diagnosis or confirmation of IBD were varied. The inclusion of participants with a diagnosis according to the criteria provided by ESPGHAN (European Society of Paediatric Gastroenterology, Hepatology and Nutrition) was specified in one study; one study stated that diagnosis was verified by endoscopic or radiographic reports; and one study included participants from the point of their diagnosis with gastrointestinal endoscopy, ilieo-colonoscopy and barium meal. Fifteen studies stated that participants had a confirmed diagnosis of IBD, and one study assumed diagnosis from the participants being members of a Crohn's specific organisation. Two studies included participants specifically from the point of diagnosis; one study looked at those diagnosed for less than 6 months; four stated their participants had been diagnosed for more than 6 months; five reported an average disease duration between 1-3 years; three reported an average duration of over 3 years and

four did not include details. Mostly, studies measured and reported the symptom severity of their sample; six studies included inactive/mild symptoms; six studies included a mix of symptom severity; two reported moderate-severe symptoms; and four studies did not specify or did not measure this factor.

The primary aim of ten of the studies was to investigate HRQoL in children and adolescents with IBD, while four papers assessed outcomes or effectiveness of a medical intervention (enteral nutrition, biological therapy, opioid receptor antagonist and surgery), two papers measured outcomes of non-medical interventions (IBD camp, CBT group) and three were evaluations of the psychometric properties of measures.

In this review, body image was measured by the body image scale of the IMPACT questionnaire in 16 studies. The remaining studies used generic, non-illness-specific measures, including the physical appearance scale of the Piers Harris Self Perception Scale (PHSCS, Piers, 1996), the body image scale of the Minneapolis-Manchester Quality of Life Instrument (MMQL-UK, Bhatia et al., 2002), and the physical appearance scale of the Self Perception Profile (Harter, 1988). All are standardised quantitative measures.

Body Image Scores

Despite the predominant use of the IMPACT questionnaire, there are different versions of the questionnaire, which differ in scoring methods and limit the ease of comparability. The majority of authors transformed the IMPACT scores onto a scale of 0-100 (Abdovic et al., 2013; Afzal et al., 2004, Loonen et al., 2002a; Ogden et al., 2008; Perrin et al., 2008; Werksetter et al., 2012). These papers reported a range of body image scores from 52 to 80, with most scores falling in the 70's.

Four papers used the IMPACT III (Otley et al., 2006) with a five-point Likert scale, reporting the overall mean in the sample from the sum of the three body image questions (Herzer et al., 2011; Hill et al., 2010; Lowe et al., 2012; Malik et al., 2013). With a possible score range from 3-15, scores reported in these papers range from 9.8 to 11.7.

Two papers used the IMPACT II (Loonen et al., 2002a), with a seven-point scale, using the same scoring system denoted above for the IMPACT III, but with a possible score range from 3-21 (Otley et al., 2006; Shepanski et al., 2005). Scores reported in these papers range from 12.6 to 16.2.

Kilroy et al. (2011) reported a mean score of 3.6 from the five-point Likert scale, with 1 being the most problematic to 5 being the least problematic. Due to the IMPACT being a disease specific measure, no healthy control group could complete the questionnaire for comparability. Despite the measure being used with different scoring systems, all of the findings reported above reveal body image scores which fall within the top third of the possible scoring range.

The PHSCS is a generic measure including a physical appearance scale. Mackner and Crandall (2005a) employed this measure and found no significant difference between physical appearance in the IBD group and a healthy control group (56.9 and 58.1 respectively). A T-score lower than 34 is suggested as clinically significant on this measure. The Self Perception Profile for Adolescents (SPPA, Harter, 1988) used in Grootenhuis et al. (2009) study, yields a score range on the physical appearance scale of between 6 to 24. At baseline the IBD group scored a mean of 13, followed by a statistically significant increase to 15.5 following a CBT group intervention. Physical appearance for an IBD group measured by the MMQL-UK

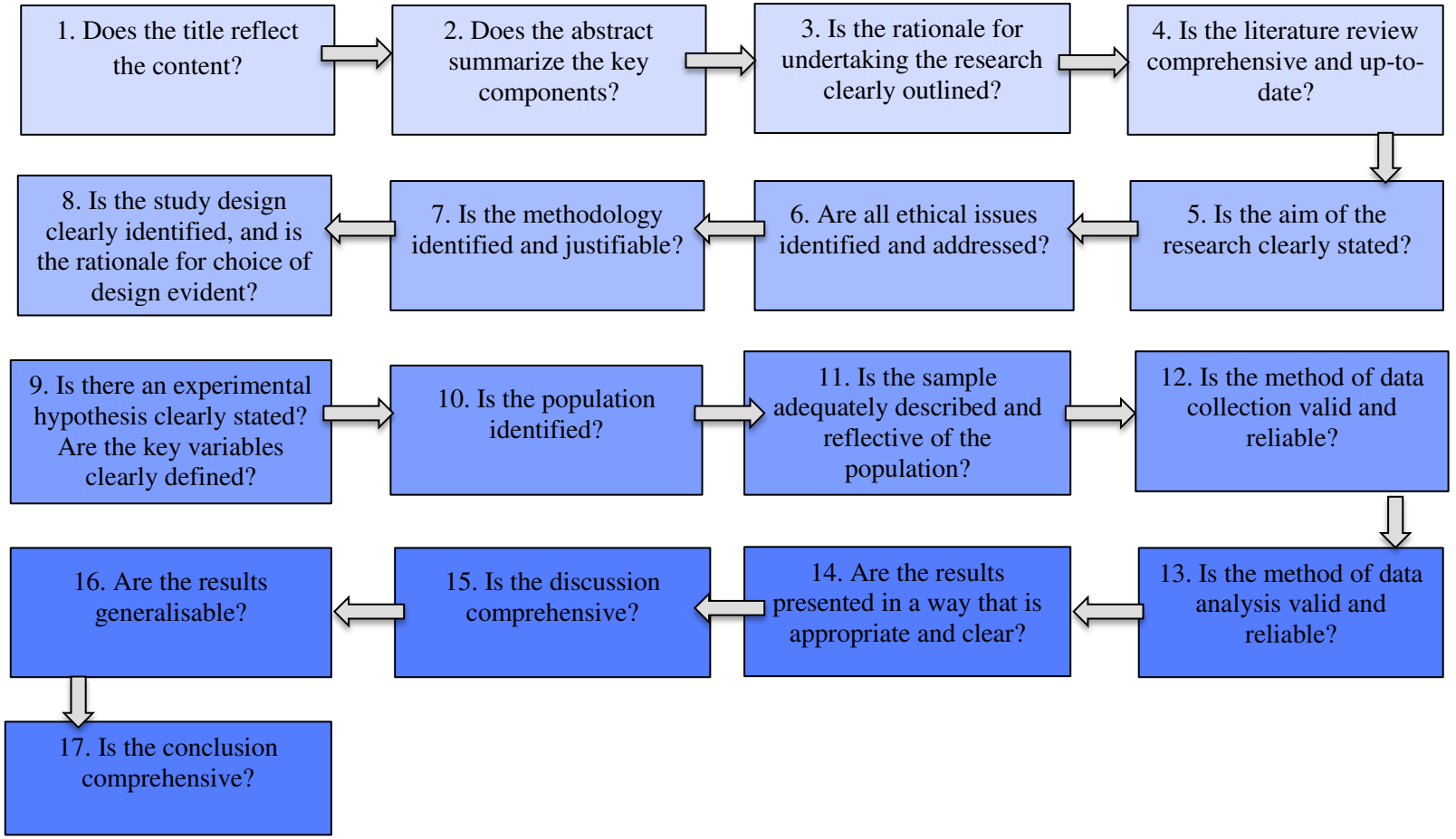
(Hutchings et al., 2007) revealed a score that was comparable to other chronic illnesses, but lower than that of a healthy control group (however this was not statistically significant).

Quality Review Framework

The framework presented by Caldwell, Henshaw and Taylor (2005) was chosen to review this body of literature (see Figure 2). This framework was designed for critiquing health-related research. The framework states the criteria for critique, however for additional clarity and detail, a numerical scale was added to the framework for this review. Each criteria of the framework was assessed according to a 0, 1 or 2 point system; where 0 conveys no evidence or that the criteria was unmet; 1 conveys partial evidence for meeting the criteria; and 2 reflects that the criteria had been sufficiently met. Detailed published guidelines were followed in applying each stage of the quality framework (Caldwell, Henshaw & Taylor, 2011). Additionally, one of the criterion 'Are the authors credible?' was excluded, since it was felt that the quality and plausibility of a piece of research should not be judged on author's credentials having been provided.

Inter-rater reliability checks on the quality review scoring process were performed to independently assess for risk of bias. The researcher's academic supervisor chose three of the final articles at random, and completed a blind quality rating using the quality review framework. A high level of agreement was found between the researchers and supervisors scores, which promotes reliability in the final scores assigned. This process was discussed in supervision following the inter-rater reliability checks, with both supervisor and researcher justifying why scores were assigned for each article. The researcher then reviewed the quality scores assigned for the remaining articles; however it was decided that scores remained appropriate.

Figure 2



Quantitative Quality Review Framework

Table 3
Article Critique[24]

Article	Title reflects content?	Abstract summary	Rationale for research	Literature review	Clear aims	Ethical issues	Methodology identified?	Design rationale	Hypothesis stated	Population identified	Reflective sample	Data collection	Data analysis	Clear results	Discussion comprehensive	Results generalisable	Conclusion comprehensive	**Total Score**
Afzal et al. (2004)	2	2	2	1	2	1	2	1	1	2	2	2	2	2	2	1	2	**29**
Otley et al. (2006)	2	2	2	2	2	1	2	2	1	1	2	1	2	1	2	2	2	**29**
Perrin et al. (2008)	1	2	2	2	2	1	2	1	2	2	2	1	2	2	2	1	2	**29**
Smith et al. (2013)	1	2	2	2	2	2	2	2	0	2	1	2	2	2	2	1	2	**29**
Loonen et al (2002a)	2	2	2	2	2	1	2	1	0	2	2	1	2	2	2	2	1	**28**
Herzer et al. (2011)	2	2	2	2	2	1	1	1	2	2	1	1	2	1	2	1	2	**27**
Mackner & Crandall (2005a)	2	2	2	1	2	1	2	2	0	2	1	1	2	2	2	1	2	**27**
Malik et al. (2013).	2	2	2	2	2	1	2	2	0	2	1	1	2	2	2	1	1	**27**
Grootenhuis et al., (2009)	2	2	2	2	2	0	2	1	1	2	1	1	2	2	2	1	2	**27**
Kilroy et al. (2011)	2	2	2	2	2	2	2	2	1	1	1	1	1	1	2	1	1	**26**

[24] Papers with the highest total score (between 24- 34) are shaded in blue. These are considered to be the papers of highest quality in this review.

Article	Title reflects content?	Abstract summary	Rationale for research	Literature review	Clear aims	Ethical issues	Methodology identified?	Design rationale	Hypothesis stated	Population identified	Reflective sample	Data collection	Data analysis	Clear results	Discussion comprehensive	Results generalisable	Conclusion comprehensive	**Total Score**
Van der Zaag-Loonen et al. (2004)	2	1	2	2	2	2	1	1	2	1	2	1	2	1	2	1	1	**26**
Abdovic et al. (2013)	2	2	2	1	2	1	1	1	0	2	2	1	2	2	2	1	2	**26**
DeBoer et al. (2012)	2	2	2	1	2	1	2	1	2	2	1	1	2	1	2	1	1	**26**
Hill et al. (2010)	1	2	2	1	2	1	2	2	0	2	1	2	2	1	2	1	1	**25**
Shepanski et al. (2005)	2	2	2	2	2	1	1	1	0	1	1	1	1	2	2	1	1	23
Werkstetter et al. (2012)	2	2	1	1	2	1	1	1	1	2	1	1	2	1	2	1	1	23
Hutchings et al. (2007)	1	2	2	1	2	1	2	1	0	1	1	1	1	1	1	1	1	20
Ogden et al. (2008)	2	1	2	1	2	1	1	1	0	1	1	1	1	2	1	1	1	20
Lowe et al. (2012)	2	2	2	1	2	1	0	0	0	1	1	1	1	1	2	1	1	19

Strengths in the articles are visually represented by the scoring and colour coded system in Table 2. The articles generally provided a good literature review, rationale, descriptions of methodology and succinct discussions. The rationale for design choice was less well discussed. All of the articles used clinical populations, which were generally representative of IBD populations; however, generalisability of samples could be influenced by sampling bias, based on those who chose to take part. Across studies, good descriptions of inclusion and exclusion criteria for the population were mostly reported. Flaws were also evident in data collection methods, since valid symptom severity measures and control over data collection variables were not consistently evident.

The critical appraisal scoring system used in Table 2 yields a maximum score of 34. For the purpose of identifying the strongest articles to base conclusions on within this review, articles scoring between 24- 34 are considered to be of higher quality. This cut-off was decided based on visual analysis of the quality scores in Table 2; those below the cut-off have lower scores, particularly in the design and methodology areas.

THEMES ASSOCIATED WITH BODY IMAGE

In addition to scores of body image, the articles reviewed also revealed a number of themes which were associated with body image. These include; symptom severity; age, gender; diagnostic categorisation; duration of illness; and treatment. These themes were derived from either the main focus of the article or from the results reported. These themes and the reported findings will now be discussed.

Symptom Severity

The most widely reported factors associated with body image in these papers were those of symptom severity and disease activity. There was a strong association between higher disease severity and poorer body image (Abdovic et al., 2013; Herzer et al., 2011; Hill et al., 2010; Loonen et al., 2002a; Perrin et al., 2008; Van der Zaag-Loonen et al., 2004). Lower QoL was also associated with increased disease severity (Afzal et al., 2004; DeBoer et al., 2012; Malik et al., 2013; Otley et al., 2006). This finding was found amongst the higher quality papers in this study, which adds weight to the conclusion that this is a strong relationship.

The association between increased disease severity and poorer body image is supported by studies that have compared participants with mild disease severity to those in remission. DeBoer et al. (2012) observed no significant difference in body image scores between those in remission and those with mild symptoms. Mackner and Crandall (2005a) reported few differences in psychosocial outcomes between those in remission and healthy controls. It can be concluded that it is not valid to combine data concerning body image of those in inactive disease status with those experiencing moderate or severe symptoms.

Age

Age was found to be significantly related to body image, with increasing age associated with decreased body image in children with IBD (Hill et al., 2010). Otley et al. (2006) purport a decrease in QoL domain scores by five points for every additional year of age and reported that this trend was also evident on the body image scale (however these data were not reported). Loonen et al. (2002a) reported data on separate age groups, observing comparable QoL scores to healthy peers in 8-11 year olds, and significantly impaired QoL scores in older children in 12-18 years age group. This is evidence of psychosocial changes with increased age; unfortunately the QoL measure used for the healthy controls did not include a measure of body image, and therefore the two groups could not be specifically compared in aspect of body image. Despite the paper receiving a lower quality score, this finding was also found in Ogden et al. (2008), who showed that body image score was higher in the 8-12 year group than the 13 -16 year group.

Additional evidence provided by Malik et al., (2013) relates the age of diagnosis to HRQoL. This study found significantly increased QoL in females who had begun menses at a younger age (ages not specified) and those who had less menstrual abnormalities. For females, if symptoms of IBD and health complications influence or delay their physical development, this may be an influential factor for psychosocial difficulties.

These age trends would also be expected in healthy children (Muris, Meesters & Fijen, 2003). The transition through puberty can intensify body image concerns, which is associated with reduced self-esteem and self-worth (Croll, 2005). However, contrary evidence in a recent longitudinal study of young people in the UK, found body satisfaction to remain stable and positive during four years of adolescence (Morin et al., 2011).

Based on the stronger papers in the review, Loonen et al. (2002a) and Hill et al. (2010) highlight that increased age (specifically the 12-18 year old group) is associated with poorer body image compared to younger age groups (8-11 year olds). The evidence suggests that children and adolescents with IBD experience similar scores in body image to their healthy peers at younger ages. Despite following similar trends to healthy peers in terms of increased body image concerns during adolescent years for those with IBD, body image concerns may be exacerbated or hindered for those with IBD related complications e.g. delayed or problematic menstrual cycles in girls.

Gender

Despite both sexes reporting body image concerns, healthy females are liable to report lower body satisfaction than healthy males (Burrowes, 2013). In this review, body image scores were higher for males than females in Ogden et al. (2008), however, levels of significance were not reported and this paper yielded a low quality review score. According to Hill et al. (2010) body image scores were significantly higher in girls. Despite a significant result, this should be considered with caution since the study had a gender split of 32:9 (male: female) and the sample featured a mean symptom severity score in the mild range. Hill et al. also found that body image was significantly related to height; this may explain the gender body image differences in the study, since research has shown that body satisfaction in adolescent males is associated with strength and height in IBD groups (Nicholas, Otley, Smith et al., 2007) and healthy peers (McCabe & Ricciardelli, 2004).

There is conflicting evidence regarding gender differences and body image. However despite the lower ratio of females in Hill's study, this paper was rated a higher quality score and had specifically defined groups (i.e. all CD, mild disease activity, specified data for ages was provided). Based on the available data, it seems likely that differences in body image

between genders follow similar trends to healthy peers, in terms of delays in growth and height influencing body image of boys.

Diagnostic categorisation

In relation to other chronic illnesses, Hutchings et al. (2007) found physical appearance scores for children and adolescents with IBD and other chronic illnesses to be lower than the control group (apart from diabetes, scoring higher). However, in none of the chronic conditions were scores significantly different from healthy peers. This article scored a low quality score, largely due to insufficient demographic and methodological details provided in the paper. However, it does appear to support findings by Pinquart (2013) and Mackner et al. (2006), reporting small differences in body image and psychosocial factors between chronic illnesses (including IBD) and healthy peers.

There are differences in the symptoms and management of the types of inflammatory bowel diseases under the umbrella term IBD. While UC leads to inflammation of the inner lining of the colon, CD can affect any section of the digestive tract from mouth to anus, and inflammation can occur in all layers of the bowel walls (Crohn's and Colitis UK, 2011). Differences can include greater propensity for rectal bleeding in UC (which is uncommon in CD), whereas strictures, fistulas and fissures (restrictions, cavities or tears) are more common in CD, along with the development of an abdominal mass. In severe cases, surgery for UC will include complete removal of the colon (colectomy), with the surgical creation of an internal pouch (ileostomy) made from healthy small intestine; in some cases this is considered a 'cure'. However, due to the wider spread of affected tissue, surgery in CD seeks to preserve as much tissue as possible. Sections of diseased tissue may be removed, connecting remaining healthy sections. In cases of colectomy, internal pouches are not an option, due to the risk of

CD occurring in the pouch, leading to a stoma and external pouch. It is possible that these differences between CD and UC could affect both physical health and psychosocial outcomes, and therefore diagnostic categorisation is a confounding variable in IBD research. Individuals with IBD have an increased risk of developing colorectal cancer, which increases the longer the duration one has the disease (Bernstein, Blanchard, Kliewer & Wajda, 2001). Mortality from IBD has declined over the past 50 years; more recent mortality trends show similarities between UC and CD (Sonnenberg, 2007), with an increased mortality rate (0.5% per year) in IBD patients compared to the general population (Card, Hubbard & Logan, 2003).

A level of significance for the association between poorer body image and increased disease severity was reported in CD than UC (Perrin et al., 2008), however, Loonen et al. (2002a) found almost the same body image scores for CD and UC patients; and Kilroy et al. (2011) did not find that type of IBD was a significant predictor of HRQoL. Five of the 19 studies specifically included only one type of IBD in their sample (Afzal et al., 2004; Hill et al. 2010; Smith et al., 2013; Malik et al. 2013, Lowe et al. 2012), however under inspection, the body image scores from these studies appear consistent with those from studies using mixed diagnoses. There was no evidence in this review to conclude that the type of inflammatory bowel disease was associated with differences in body image.

Duration of Illness

The characteristically intermittent nature of IBD can make it an unpredictable illness to manage, with patients often passing between periods of relapse and remission during the natural course of the condition. Otley et al. (2006) found that body image scores increased from the point of diagnosis up to 12 months post-diagnosis, however, results were not statistically significant. In a five-year study, Hill et al. (2010) did not find any link between

the time since diagnosis and body image, however Otley et al's study had a much larger sample than Hill et al's and included more participants with greater disease severity.

There is uncertainty in the evidence for the influence of time since diagnosis on body image. Otley et al's study suggests that poorer body image is found at the point of diagnosis, and may improve during the following year. This hypothesis is linked to symptom severity, with children experiencing more severe symptoms at the point of diagnosis, which decrease once treatments are commenced and remission is brought about. However, due to lack of statistically significant data, no firm conclusion can be drawn. Given the intermittent nature of IBD over time, there is potential for individuals to learn more and develop their understanding of the implications of IBD and possibly experience further relapse and complications as time goes on.

Treatment

Medical trials and effectiveness studies have included psychosocial elements alongside medical evaluations. Significant improvements in body image were found after treatment with enteral nutrition for eight weeks (Afzal et al., 2004). Some improvements in body image (not reaching statistical significance) were observed after biological therapy for fourteen days (DeBoer et al., 2012) and no changes in body image were reported after treatment with an opioid antagonist over 16 weeks (Smith et al., 2013). These points raise the question of the mechanism by which body image could be being influenced and how long it would take to influence this concept. Improved quality of life and decreased symptoms may be observed in fourteen days; however a longer period of time may be required for changes to translate to concepts such as body image.

Malik et al. (2013) add an interesting element to the review by exploring the impact of surgical interventions in UC. These data suggest that body image is lower for the surgical groups (however this did not reach statistical significance, perhaps due to significant differences between the two groups, with the non-surgical group experiencing greater symptoms). It would not be surprising to find changes in body image in surgical groups, which could be interpreted in light of surgical patients having an increase in invasive procedures performed, physical changes and scarring brought about from surgery (for intestine resection, colectomy or to treat abscesses or fistulas) and possible changes in shape through weight loss or weight gain post-surgery. This study showed that quality of life is higher amongst patients not taking medication, indicating that treatment regimes may also influence psychosocial factors. This is supported by Hill et al. (2010) who found significantly poorer ratings of body image for those taking enteral nutrition, followed by immunosuppressants plus steroids compared to patients taking other IBD medications (both of these treatment groups had lower mean body image scores than the surgical group in Malik et al. 2013).

Psychosocial interventions included a group CBT intervention (Grootenhuis et al., 2005) and a one-week IBD summer camp (Shepanski et al., 2005). The CBT group boasted significant improvements in body image, which were maintained at the 6-8 month follow up. The camp did not show changes for body image, but did have changes for overall QoL and social functioning. Again, one week for the follow-up in Shepanski et al. may not have been long enough to influence change for body image, and this study has a number of methodological flaws.

This evidence suggests that the type of treatment may be influential in perceived body image. Treatments that bring about remission and alleviation of symptoms may aid

improvement in body image; however some treatments are also associated with poorer body image. The causal factor is not clear from these studies, it could be hypothesised that the long term regimes, dietary management and side effects involved in some medications may have a negative influence, however since other variables were not controlled in the studies, no firm conclusion can be drawn. Psychological support, such as the group intervention in Grootenhuis et al. (2009), has been shown to improve body image via a process of psychoeducation, relaxation and CBT based exercises.

Additional Psychological Factors

There were psychological factors of interest highlighted in the review, which may be linked to body image or self-perception. It was posited that autonomy, motor functioning and emotions are affected in adolescents with IBD (Loonen et al., 2002a); these factors could play a role in the development of the adolescent's sense of self and natural progression of gaining independence. Higher internalising complaints (social withdrawal, somatic complaints, anxiety and depression) and social problems were reported for children with IBD compared to healthy peers and a marked difference was found in boys (DeBoer et al., 2005). High correlations were found between social functioning, physical appearance, self-esteem, internalising, externalising (behavioural problems and aggression) and total behavioural problems. Furthermore, clinically elevated family dysfunction in IBD families (Herzer et al., 2011) may be a factor which influences emotional and self-development.

Differences in children and adolescents' coping styles emerged as significantly different to healthy peers in this review. Mackner and Crandall (2005b) reported a higher occurrence of 'wishful thinking', and avoidant coping styles were observed by van der Zaag Loonen et al. (2004). Avoidant style may correspond to the embarrassment expressed by

young people with IBD and attempts to conceal their illness, symptoms and treatment. Meijer, Sinnema, Bijstra, Mellenbergh and Wolters (2002) found that avoidance was not a strong predictor of psychosocial adjustment in adolescents with chronic illness and reported that active coping methods are preferable to passive coping (including avoidance and depressive coping behaviours).

It was suggested that a diagnosis of IBD prior to the onset of puberty might have fewer negative psychosocial consequences than a diagnosis during puberty, since females who had menses at a younger age had significantly higher HRQoL and fewer menstrual irregularities (Malik et al., 2013). This concurs with previous studies regarding the benefit for adolescents who develop at the same time as the majority of their peers, fitting into the developmental 'norm' (Williams & Currie, 2000) and influencing how the young person perceives their body.

The review revealed clinically relevant data, showing that young people with IBD adjust their lifestyle, doing less activity and resting more than their healthy peers even in periods of remission (Werksetter et al., 2012). The authors described the role of physical activity in building and maintaining healthy musculature, which may be lacking in children with IBD, which in turn, based on the evidence particularly for adolescent males with IBD, could influence self-esteem and body image.

In conclusion, the trends in body image reported for children and adolescents with IBD appear to follow similar patterns as their healthy peers, particularly for those in remission or with only mild symptoms. However, the nature of IBD may present the young person with a range of risk factors, which in turn has the potential to increase their vulnerability to poor

body image. These include delayed growth or height, and increase in severe physical symptoms and particular treatment regimes.

METHODOLOGICAL QUALITY AND RESEARCH FINDINGS

This is the first systematic review to explicitly assess body image in children and adolescents with IBD. There were a number of flaws within the body of evidence, which limit the strengths of conclusions on this topic, and warrant further research to address these aspects and offer more robust findings.

Firstly, no specific measures of body image were found in the literature for children or adolescents with IBD, therefore conclusions are reliant on body image and physical appearance scales based within broader quality of life measures. The IMPACT questionnaire is most commonly used to report on body image, however since this only includes 3 items, a more detailed body image questionnaire may be more informative. Despite the overall high internal reliability on the IMPACT questionnaire, the body image scale has been called into question. Abdovic et al. (2013), Loonen et al. (2002a) and Perrin et al. (2008) found borderline to satisfactory acceptability on the body image scale. Perrin et al. (2008) suggest the addition of items to this domain to increase reliability. Additionally, since this is a disease-specific measure, no comparison group or normative data are available. This is particularly pertinent, since one of the fundamental questions that remains unclear, is whether body image in children with IBD differs from the body image concerns reported in healthy young people.

Alongside a measure of body image which would allow for comparison with healthy controls, future research would also benefit from a longitudinal design to examine changes over time and the long term influences of IBD on body image, and to assess body image in IBD patients alongside developmental changes. This is currently lacking in the existing body of evidence.

It is also important that severity of symptoms is measured and reported with an appropriate measure for the disease type, such as the PCDAI (Hyams et al., 1991) for CD and PUCAI (Turner et al., 2007) for UC, since severity of symptoms is a confounding variable, which is not consistently reported. The significance of accurately measuring and recording the severity of symptoms should also be noted, with validated measures such as. The current research has demonstrated the need for interpretation of body image data based on inactive, mild, moderate-severe symptoms, medication, age and gender. Hypotheses have been suggested in the literature regarding each of these variables; however in the absence of a controlled study to address these, firm conclusions cannot be made. Small clinic-based samples of children with separate IBD's make age-specific analysis difficult (Sonnenberg, 2007). Additionally, there is a lack of information provided on effect size and clinical significance. This may be due to the subjective nature of self-report scales and lack of clear cut-off points for scales such as the IMPACT questionnaire. With the awareness of psychosocial factors in measuring disease-related outcome, clinicians may benefit from guidance on how to interpret these measures, or encouragement to set goals for raising baseline data i.e. improving HRQoL scores amongst their patients (Lowe et al., 2012).

A criticism of the quality review framework utilised is that an equal weighting was assigned to each critique item in the review process. For example, clarity of title of the paper is scored and weighted equally to the appropriateness of data analysis used. Despite this framework producing a clear overall picture of important aspects of a research study, a more robust weighting system or scoring procedure, which takes into account the more critical elements to the quality of a study may be beneficial. There were also articles of interest, with missing data, which the author could not access that may have been influential in elucidating some of the queries regarding body image in this review.

CLINICAL IMPLICATIONS

Adolescence is a crucial time of transition and development (American Psychological Association, 2002). Literature shows that future symptoms of depression and poor self-esteem can be predicted by body dissatisfaction during early and middle adolescence (Holsen, Kraft & Roysamb, 2001). Evidence in the literature examining the impact of IBD on children and adolescents highlight a range of risk factors, which may impact on the development of self-image. Research in healthy populations shows that poor self-image can have a detrimental effect on psychological wellbeing; therefore it is recommended that clinical services supporting children with IBD routinely check psychosocial factors in addition to physical health assessments, to identify all psychosocial issues, including emerging body image concerns. Group support, similar to the programme described by Grootenhuis et al. (2009), would be beneficial for children to strengthen positive coping strategies and provide an opportunity to meet other young people with IBD; this could be offered as a standardised programme for children after diagnosis or at least at the point of an identified psychosocial concern. It is important that clinicians keep psychosocial factors, such as body image, in mind when assessing children with IBD, particularly as feelings towards one's body can influence adherence to treatment regimens and diets (Vlahou et al., 2008) and may be an indicator of the need for additional support.

CONCLUSION

This systematic review examined nineteen published quantitative papers with a measure of body image in children and adolescents with IBD. The results show that, particularly for those with mild or no symptoms, trends in body image are similar to healthy peers in that body image declines with increased age during adolescence. However, IBD may predispose the young person to risk factors, increasing their vulnerability to poorer body image. There is some evidence suggesting that increase in disease severity and some treatments may negatively influence body image. In the absence of higher quality studies, these conclusions remain cautious. Methodological flaws limit the strength of conclusions which can be made from this review. There are a number of potentially confounding variables relating to body image, which were not consistently reported and controlled for. Furthermore, the current body of evidence is lacking in data that can be compared against control groups. Research with a specific focus on body image in IBD is warranted to address the issues and methodological flaws outlined in this review.

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Healthcare professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease: An Interpretative Phenomenological Analysis

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By

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ABSTRACT

Background: The psychosocial issues and challenges for young people with a dual diagnosis of type 1 diabetes and coeliac disease and their parents have been explored and uncertainties within screening and diagnostic processes are documented. However, the experiences of healthcare professionals (HCPs) working with this group has to date remained unexplored.

Method: Twelve HCPs (doctors, nurses, dieticians) working in this field from three paediatric teams were interviewed about their experiences. Transcripts were analysed using Interpretative Phenomenological Analysis.

Results: Three superordinate themes emerged; 'Connection and burden'; 'Diagnoses together, but separate and uneven'; and 'Sitting with certain uncertainty and complexity'. These represent participants' connection to the experience of patients and adding to their burden; a sense of disparity in managing the two conditions; uncertainty and complexity with the dual diagnosis, and a repertoire of coping strategies utilised. Positive aspects of the role (making a difference, improving health and reducing risk) along with coping strategies are likely to buffer the challenges of working with this client group.

Conclusion: Recommendations include a dual review clinic and further support for staff. Additional research on medical and psychosocial aspects of this dual diagnosis is needed to develop services to support both patients and HCPs.

Keywords: *Healthcare Professionals, Experiences, Coping Strategies, Children, Type 1 Diabetes, Coeliac Disease, dual diagnosis, Interpretative Phenomenological Analysis.*

INTRODUCTION

Working with Chronic Health Conditions

With increasing emphasis on extending wellness and improving health, healthcare is becoming more complex with fresh challenges for professionals (Department of Health; DH, 2008). This seems particularly pertinent to the area of chronic conditions, which are acknowledged to be complex and challenging for healthcare professionals (HCPs), (DH 2004; Royal College of Physicians, 2004).

Caring for individuals with chronic health conditions requires a range of skills, which fall outside the traditional biomedical model of prevention, diagnosis and treatment (Kane, Priester & Totten, 2005). Interventions are often rooted in patient self-management, which healthcare providers play a key role in facilitating (Schulman-Green et al., 2012), however this role can be complex (Thorne & Paterson, 2001). A study of GPs found this complexity was linked to their sense of responsibility for delivering high quality care and needing to feel in control to meet professional responsibilities (Blakeman, MacDonald, Bower, Gately & Chew-Graham, 2005).

Exposure to ongoing difficulties experienced by patients with chronic conditions may increase anxiety, feelings of professional inadequacy and emotional burden amongst clinicians (Turner & Kelly, 2000). Burnout, a syndrome of emotional exhaustion (Maslach & Jackson, 1981), has been linked to healthcare professions. Ekstedt and Fagerberg (2003) highlight the association between burnout and ongoing involvement in emotionally demanding situations. Figley (2002) asserts that the additional energy required to provide compassion and empathy to individuals with chronic illnesses can have a cost, emphasising the importance of self-care. Furthermore, to avoid burnout, Kane et al. (2005) posit the need

to draw satisfaction from supporting patients in managing long-term conditions as opposed to being able to cure them.

The Effort-Reward Imbalance (ERI) model (Siegrist, 1996) postulates that imbalance in the reciprocity between efforts and rewards at work can lead to stress. This model has been applied to the study of nurses, showing an association between high extrinsic effort, low extrinsic rewards and emotional exhaustion and depersonalisation (Bakker, Killmer, Siegrist & Schaufeli, 2000). Given the importance of the therapeutic relationship in managing chronic illness and the patient's role in self-management, interactions may not be rewarding if the professional does not feel their investment in the patient relationships leads to good outcomes, or if there is slow or no improvement in health.

A further challenge of working with chronic conditions is their nature of uncertainty. Han, Klein and Arora (2011) assert that inability to abolish uncertainty in healthcare creates difficult challenges for clinicians and patients. Mishel's model of illness uncertainty (1988) proposes that the psychological effects of uncertainty may provoke feelings of vulnerability and avoidance of decision making in HCPs. The importance of maintaining a sense of control for physicians is documented (Kearney, Weininger, Vachon, Harrison & Mount, 2009). This theme is supported by Shapiro, Shapiro, Robitshek and Shapiro (2011), who discuss how treating individuals with multiple, coexisting health conditions has been associated with feelings of loss of control.

Working with Children with Chronic Conditions

Children and adolescents' experience of chronic conditions has been associated with poorer emotional wellbeing, psychological distress (Yeo & Sawyer, 2005), longer hospital admissions, loss of school days, restrictions to activities (Newacheck & Halfon, 1998), and

may have implications for family functioning (Sawyer & Aroni, 2005). Therefore, HCPs working with children are not only responsible for providing medical care, but also have a role in being alert to psychological wellbeing and social circumstances.

Studies discussing the experiences of HCPs caring for children with chronic illness (including a range of disciplines i.e. nurses, medical specialists, physicians) highlight concerns about: sharing illness information (Lochrie, Wysocki, Burnett, Buckloh & Antal, 2009); the child and family's coping abilities (Hoey et al., 2006) and capacity for illness self-management (Lake & Staiger, 2010). In addition, providing care within the complex relationships which can emerge between families and multiple services may create further challenges (Dickinson, Smythe & Spence, 2006). Two chronic conditions that may be experienced during childhood are type 1 diabetes (T1D) and coeliac disease (CD).

Diabetes

T1D is a lifelong condition where the body does not produce the hormone insulin to regulate blood glucose levels. An estimated 22,947 children under 18 years in the UK have diabetes (this gives a prevalence of 209 per 100,000), 97% of whom have type 1 (Royal College of Paediatrics and Child Health, 2009). The UK is the fifth highest country in the world for rates of childhood T1D (Diamond Project Group, 2006), with prevalence rates rising rapidly (Hsia et al., 2008).

Diagnosis is based on prolonged elevated blood glucose concentration. Symptoms at diagnosis include excessive thirst, urination, and can escalate to diabetes ketoacidosis (DKA), a life threatening form of dehydration (Pilek & Starkman, 2014). There is no cure for T1D; treatment involves lifelong management of blood glucose levels through diet, blood glucose monitoring, insulin replacement and exercise. This is crucial to avoid acute complications

arising from low blood glucose levels (hypoglycaemia) and longer term complications associated with high blood glucose (hyperglycaemia). Consequences of hypoglycaemia range from trembling, blurred vision and difficulties concentrating, to loss of consciousness, seizure and possible death (Clarke, Jones, Rewers, Dunger & Klingensmith, 2009). Hyperglycaemia is linked to risk of renal failure, eye damage (retinopathy), nerve damage (neuropathy), and cardiovascular disease (Daneman, 2006).

Coeliac Disease

CD is a lifelong autoimmune condition where the body reacts adversely to a protein called gluten, found in wheat, barley and rye. CD is recognised as a multi-systemic disorder, with the intestine as the primary disease site (Green, 2005). The body's immune system responds to gluten, causing inflammation and damage to tissue of the small intestine, termed villous atrophy (Alaedini & Green, 2005). CD can present with a varied and wide range of symptoms including: chronic gastrointestinal symptoms, abdominal pain, weight loss, metabolic bone disease, anaemia, delayed puberty and nonspecific arthritis (Green & Jabri, 2003; Mearin, 2007). The clinical range can span from asymptomatic (villous atrophy in the absence of the classic symptoms; Alaedini & Green, 2005) to severely symptomatic. Heterogeneity in presentation of CD can lead to poor detection. A host of screening studies reveal between 0.5-1% of populations across Europe, South America, Australasia and the USA may have undiagnosed CD (van Heel & West, 2006). Prevalence rates of 1% are reported in UK children (Bingley et al., 2004).

Diagnosis is by blood test and intestinal biopsy. If results are positive, lifelong adherence to a gluten free diet (GFD) is the only form of management (Kaukinen, Lindfors &

Mäki, 2014). A review by Goddard and Gillett (2006) concludes that left untreated, CD is associated with risk of reproductive complications, osteoporosis and small bowel lymphoma.

Dual Diagnosis

The prevalence of CD in children with T1D is reported at between 5-7 times higher than the general population (Sud et al., 2010). Children with a dual diagnosis of T1D and CD (hereafter referred to as dual diagnosis) usually present with asymptomatic or 'silent' CD (Telega, Bennet & Werlin, 2008). Therefore, CD is likely to go undiagnosed unless they go through screening (Saukkonen, Vaisanen, Akerblom & Savilahti, 2002). The European Society for Paediatric Gastroenterology, Hepatology, and Nutrition (ESPGHAN) advise that children with an increased risk for CD should be screened (Husby et al., 2012). Even if asymptomatic, those testing positive are recommended to follow a GFD. The daily reality for children with a dual diagnosis include: making food choices based on only gluten free options; following a healthy diet with particular control over sugar intake; carbohydrate counting at each meal in order to calculate insulin dosage; monitoring insulin pumps or self-injecting insulin; and regular blood glucose monitoring.

Psychosocial issues, including depression, anxiety, social withdrawal and lower quality of life, have been reported for young people with T1D (Dantzer, Swendsen, Maurice-Tison & Salamon, 2003; Hanberger, Ludvigsson & Nordfeldt, 2009; Weglage et al., 2000) and emotional distress among parents (Bowes, Lowes, Warner & Gregory, 2009; Haugstvedt, Wentzel-Larsen, Rokne, Graue, 2011). Adherence difficulties, restrictions of activities and negative emotional responses (such as anger and feeling misunderstood) are documented for young people with CD (Altobelli, Paduano, Gentile et al., 2013; Roma, Roubani, Kolva et al., 2010). Adhering to the GFD is in itself associated with psychosocial difficulties (see Howard

& Law, 2014). Thus it seems plausible that the experience of managing the dual diagnosis would magnify these challenges. Love (2013) explored the challenges for young people with a dual diagnosis and their parents in a qualitative study. The nature of chronicity and variability with the diagnoses was linked to burden. A theme of protection and loss emerged, threatening future hopes and expectations for parents and adolescents, loss associated with the adolescents' relationship with food and parental protection from future health complications.

There are ambiguous reports of whether CD and the GFD have implications for blood glucose control in T1D (Mohn et al., 2001; Saadah, Zacharin, O'Callaghan, Oliver & Catto-Smith, 2004; Schwarzenberg & Brunzell, 2002). The evidence remains unsubstantiated, increasing the complexity of managing both conditions. Studies of the long term risks of untreated CD in children with T1D are also inconclusive, raising controversy for HCPs regarding optimum management (Freemark & Levitsky, 2003; Sud, Marcon, Assor, Daneman, & Mahmud, 2011). These authors conclude that unclear evidence about the long-term risks for asymptomatic CD patients, creates difficulties in developing evidence-based approaches.

The costs and benefits of screening and the additional burden of recommending a GFD (particularly to those asymptomatic for CD) has been discussed (Sud et al., 2011). Challenges associated with the diagnostic process include the ethics of screening, requirement for sound information and support for patients and families (Mimnagh & Thornton, 2006).

Aims

Available literature provides evidence of the challenges for HCPs delivering care to those with chronic conditions, notably working with uncertainty, a lack of control and the need to promote self-management for patients. The challenges facing children and their

parents associated with T1D and CD include daily, life-long self-management, which can create physical and psychosocial complications that HCPs are required to identify and support. Therefore, besides the medical complications presented by this group, psychosocial issues add to the complexity for young people, their parents and HCPs (Howard, Law & Petty, 2012). Furthermore, the dual diagnosis can bring additional complexity associated with screening practices and recommendations. To date, the experiences of HCPs working with children and young people with a dual diagnosis remain unexplored. The current study explores the experiences of HCPs, working in multi-disciplinary teams with children and young people with a dual diagnosis, with the hope of furthering the understanding of this area of healthcare, potentially enhancing staff and service support.

METHODOLOGY

Design

Interpretative Phenomenological Analysis (IPA) is a qualitative approach which explores how individuals make sense of their personal and social world (Smith & Osborn, 2008). This in-depth approach is grounded in the key areas of hermeneutics, phenomenology and idiography; with a focus on understanding an individual's lived experience through the meanings they make. It is particularly suitable for health psychology research (Brocki & Wearden, 2006). IPA was chosen for this study since the aim of the research was to gain an understanding of what it is like for the HCP's to work with this patient group and the meanings they assign to their experiences. This focus is fundamental to IPA, in contrast to other approaches such as grounded theory, which generates theory from data (Glaser & Strauss, 1967) or narrative inquiry, which explores ways that people construct stories to interpret the world (Webster & Mertova, 2007).

IPA recommends data collection methods that facilitate the collection of rich, detailed, first-person accounts of experiences (Smith, Larkin & Flowers, 2009). Semi-structured interviews lend themselves well to this form of analysis; they offer the participant the opportunity to lead the discussion about their experiences. Indicators suggested for high quality IPA studies (Smith, 2011, Appendix 1) were consulted to guide and reflect on quality throughout the research process.

Key themes from the literature on working with chronic illness were used to guide the development of initial questions, forming a list of open ended questions. A dietician (not otherwise involved in this research) from the charity Coeliac UK, with experience of the dual diagnosis field, reviewed the interview schedule to assess the relevance and suitability of

questions. The final interview schedule is shown in Appendix 2. In concordance with IPA principles, this interview schedule was used flexibly, to guide the exploration of experiences and meaning-making of participants, whilst following their concerns and ideas elicited during the interview.

Procedure

The study was granted approval by a University Research Ethics Committee (Appendix 3) and the Research and Development departments at recruitment sites. Lead clinicians from each service distributed participant information sheets (Appendix 4) to their team; this was followed up by the researcher attending a team meeting to allow potential participants to ask questions before making a decision about taking part. Participants provided written consent (Appendix 5) for data collection and inclusion of verbatim extracts in publications.

Individual, semi-structured interviews (approximately 60 minutes) were conducted in the participant's place of work. Interviews were digitally recorded and transcribed verbatim. There is a risk that discussing difficult work experiences or concerns about patients may evoke participant stress or anxiety, and therefore participants were debriefed immediately after interviews (Appendix 6) and information from their Trust staff counselling service was provided. Following interviews, participants were sent a copy of the transcript for approval. They were given a four week period to request changes to be made or to fully withdraw from the study.

Service Contexts

Participants were recruited from three services, from separate geographical locations across England and Scotland. All have paediatric diabetes teams working in close liaison with a gastroenterology team. All screen T1D patients for CD automatically in the presence of symptoms suggestive of the condition. The screening frequency for CD in asymptomatic T1D patients varies between the three services (see Table 1).

Table 1

Frequency of screening for CD in T1D

Service	Initial CD screening	On-going CD screening
Service A[25]	CD screening 12 months after T1D diagnosis.	CD screening every second year.
Service B	CD screening recommended after T1D diagnosis, patient/parents decides whether to be screened.	CD screening every second year.
Service C	CD screening at the point of T1D diagnosis.	CD screening annually.

Children and young people with a dual diagnosis are managed by the diabetes team for both conditions in all sites, with additional input provided by the gastroenterologist if there are further gastro-related complications. All three services offer quarterly appointments and an annual review. The annual review in services A and B are held with the diabetes team. Service C offers a dual annual review clinic, where the patient is seen by members of the diabetes team and a Consultant Gastroenterologist.

[25] Services labelled A, B and C to preserve confidentiality

Participants

Inclusion criteria stipulated that participants were HCPs (of all healthcare disciplines) working in multidisciplinary teams in the National Health Service (NHS) working with children with a dual diagnosis of T1D and CD. Participants were required to have worked in this area for a minimum of three months (to allow them sufficient experience of working with a dual diagnosis to allow for meaningful reflection in interviews).

Due to the risk that some findings generated by the research could be attributed to influences of the service, participants were recruited from multiple services which the University of Birmingham had established links with. A purposive sampling method was followed; this approach is used when participants are selected due to having knowledge or experience of a particular area.

Four to ten participants are recommended for IPA in doctoral level research (Smith et al., 2009). Ten participants were sought, however, a total of twelve volunteered and were included in the final analysis, representing a response rate of 40% (response rate calculation based on the total number of staff working in the clinical teams at each site meeting the inclusion criteria and the number who actually took part). Twelve may be viewed as a large sample for IPA, however in an evaluation of IPA papers, Smith (2011) gives examples of good quality IPA studies, which have used samples of twelve and guidance is given on appropriate IPA with large samples. He recommends that evidence from at least three or four participants supports each theme and the prevalence of a theme is indicated. The sample consisted of twelve HCPs (female n=11) from three services; participant demographics are shown in Table 2.

Table 2

Participant Demographics

Pseudonym	Profession	Team	Years working with dual diagnosis	Age group (years)
Fran[26]	Dietician	Diabetes	5 years	40-45
Charlotte	Dietician	Diabetes	4 years	25-30
Mandy	Dietician	Diabetes	5 years	30-35
Ann	Paediatrician	Diabetes	22 years	50-55
Sophie	Paediatrician	Diabetes	8 years	30-35
Kate	Consultant Paediatrician	Diabetes	13 years	35-40
Sam	Consultant Paediatrician	Diabetes	13 years	40-45
Ashley	Consultant Paediatrician	Diabetes	16 years	40-45
Clare	Specialist Diabetes Nurse	Diabetes	16 years	50-55
Rachel	Specialist Diabetes Nurse	Diabetes	26 years	55-60
Chris	Consultant Paediatric Gastroenterologist	Gastroenterology	19 years	46-50
Alex	Consultant Paediatric Gastroenterologist	Gastroenterology	20 years	40-45

[26] Pseudonyms have been assigned for anonymity.

Data Analysis

IPA data analysis followed the approach detailed by Smith et al. (2009, see Table 3). Credibility was boosted by cross-checking coding with two academic supervisors. This involved the independent coding of samples from three transcripts by supervisors, which was then discussed in supervision, in a process of exploring concordance and disagreement of themes and interpretations. Emerging themes were discussed in supervision and an IPA analysis group throughout the analysis process. Furthermore, a sample script was cross-checked for credibility via an IPA analysis group[27], where members of the group completed a section of coding followed by a group discussion.

Resulting super-ordinate and subthemes were sent to the lead clinicians from each site for their views and as an opportunity for them to give feedback. A sample transcript of data coding is shown in Appendix 7, this process was conducted for all transcripts. The right-hand column includes initial comments on the data; these include descriptive, linguistic and conceptual comments. The left-hand margin details the emergent themes. Appendix 8 provides an example of data supporting one of the super-ordinate themes. All quotes in the transcripts that supported each theme were copied into a table.

[27] A group of 10 Trainee Clinical Psychologists, using IPA for their doctoral thesis, facilitated by a member of the course team, with extensive research experience of using IPA.

Table 3

Summary of IPA Analysis Process

Step 1: Active engagement with the data	Reading and re-reading the transcript, listening to the interview, making note of powerful recollections and initial striking observations.
Step 2: Initial noting	Line by line analysis examining the semantic content and language on an exploratory level, noting descriptive, conceptual and linguistic comments and why these seem significant.
Step 3: Developing emergent themes	Analysing exploratory comments to identify emergent themes. Emphasising convergence, divergence, commonality and nuance. Reduction in the volume of detail of notes, mapping interrelationships, connections and patterns between exploratory notes.
Step 4: Connections between themes.	Re-evaluation of the importance of themes, mapping how the themes fit together and producing a structure which allows the most interesting and important aspects of the participants account to be identified.
Step 5: Moving to the next case.	Analysis process is repeated for the next transcript. This new transcript is explored and analysed based on its own individuality, while 'bracketing' previous analysis ideas as much as possible.
Step 6: Looking for patterns.	Assessing the connections between each case, how does a theme in one case illuminate a different case? And assessing the most potent themes. Possible reconfiguration and relabeling of themes.

Reflexivity

To engage with participants' experiences within data, it is recommended that researchers reflect on their own experiences and assumptions, and how these have the potential to influence analysis (Larkin & Thompson, 2011). This process of reflexivity was supported during regular supervision sessions.

I do not have a chronic health condition and have no special dietary requirements; however, my husband has a diagnosis of type 2 diabetes. I observed and shared the shock, difficulties and concerns surrounding his diagnosis and life with diabetes since this point. I appreciate the restrictions, frustrations and efforts involved in healthy glycaemic control and related aspects of health for individuals with diabetes. Prior to this research, my knowledge of CD was limited. Along with the development of my understanding of CD, a strong appreciation for the challenges and implications for those with this condition has grown during the research process. During interviews and data analysis I acknowledged that given my experience, it was easy for me to connect to diabetes-related worries, expressed by particpants. I have endeavoured to reflect on these and hold them aside while engaging in the participant's experiences as best as possible, making use of supervision and a peer IPA group to discuss this.

I think my experience of working alongside various HCPs in inpatient and outpatient physical healthcare has been beneficial to contextualising how professionals respond to work challenges, since some of the ideas described in interviews (particularly coping strategies) were similar to those I have observed. I had an expectation of how HCPs may respond to questions regarding their own experiences at work, possibly subjugating expression of their own work-related emotions as secondary to the emotions and experiences of the patients they care for.

RESULTS

Three superordinate themes emerged from the data. A summary is provided in Table 4. The results section will consider each theme, with quotations[28] for illustration and for participants to be heard directly. Attention will be paid to the general ideas found across participants, the nuances that were important to individuals and the connections between these. Guidance for conducting IPA with larger samples, provided by Smith, Flowers & Larkin (2009), recommends group level themes, which are illustrated with examples from individuals. Furthermore, the identification of recurrence of themes across participants is recommended for larger samples. The numeration or recurrence of themes is indicated in Table 4.

[28] For brevity within quotes, unnecessary text has been replaced with …..
Words which were highly emphasised during the interview are in bold typeface.

Table 4

Summary of Themes

Superordinate Theme	Number of participants contributing to superordinate theme	Sub-theme	Participants contributing to sub-theme
Connection and burden	All	Empathy and negative view of the diagnoses	All
		Feelings of adding to the burden (some guilt)	All
		Acting in the patients' best interests	Fran, Kate, Clare, Chris, Ann, Ashley, Alex, Mandy, Rachel, Sophie
Diagnoses are together; but separate and uneven	10	Diabetes comes first and is often prioritised	Fran, Kate, Chris, Ann, Ashley, Charlotte, Mandy, Rachel, Sophie, Alex
		Diagnoses going along in parallel but difficult to join up.	Fran, Kate, Chris, Ann, Charlotte, Mandy, Rachel,
Sitting with certain uncertainty and complexity	All	Complexity of the role	Clare, Chris, Kate, Charlotte, Rachel, Sophie
		Diagnostic grey areas can feel difficult	Chris, Ann, Ashley, Charlotte, Alex, Mandy, Sophie
		Striving for accurate control, sitting with sub-optimal control of the conditions	Fran, Kate, Clare, Chris, Ann, Ashley, Charlotte, Mandy, Sam, Rachel, Sophie

Super-ordinate Theme 1: Connection and burden

Participants described their perceptions of the difficulties associated with receiving these diagnoses, the impact on the young person's life, family, future and psychosocial factors. They described a connection to what it must feel like and a sense of responsibility for adding to the burden. A process of meaning making with regards to their role was shown.

(i) Empathy and negative view of the diagnoses

The nature of having this dual diagnosis was frequently described negatively and as burdensome; a sense of connection to the young person and family was found.

> *…it's every impact of their life, absolutely everything. So it's not just insulin and diabetes and food, it's just everything. The whole family is affected. Clare, Nurse; 29-30*

All participants talked about the challenges of having a dual diagnosis. There was a repetition of the notion "it's bad enough having T1D, but to get CD too". There were descriptions of the unfairness of the conditions, concerns about the impact they may have on the young person's quality of life and a sense that it is harder for those diagnosed with CD experiencing few or no symptoms (asymptomatic).

> *It seems very unfair in some respects to have one chronic condition that has a real problem long term, to then have to deal with another problem that's actually a dietary issue… it must be very difficult to do that. Alex, Gastroenterologist; 49-58*

> *… you do wonder what the long term impact is … they have to focus so much on their diabetes and their coeliac disease as well, you know whether that takes away some of the energy and um thought they have for other things as well. Fran, Dietician; 436-442*

Empathy was demonstrated by participants positioning themselves in the shoes of the young person or parent, questioning how they would cope with these challenges and relief that they do not have to. This depth of perspective taking may enhance the emotional connection HCP's have towards the patients and their experiences.

> *I think as a parent how hard that would be… I've always thought throughout my whole career, 'thank goodness my child doesn't have diabetes'. And then that added thought of having to deal with Coeliac disease on top is a massive challenge. Chris, Gastroenterologist; 28-30*

However, there is also recognition of the range of responses found in families, with a feeling of admiration towards the families that cope well with the challenges.

> *We have some families that are spot on you know, very diligent and avoid gluten at all costs and do all the things they need to do…… I can't imagine having to worry about things like that. Rachel, Nurse; 232-234*

(ii) Adding to the burden

In general with children with this dual diagnosis, T1D is diagnosed first. Therefore they are usually known to the diabetes team before receiving the CD diagnosis. Receiving a second lifelong diagnosis evoked compassion in participants. The language used to describe the second diagnosis ("the double whammy", "a slap in the face", "extra burden") suggests how negatively they view this outcome.

> *Massive, it's a massive challenge. Erm, you know having type 1 diabetes in itself is a massive challenge, but then getting the second double whammy of the impact of having another lifelong condition compounds all that they have to do. Clare, Nurse; 20-22*

> *I feel sorry, we all feel sorry for them, because actually we know them because they've got diabetes so we've engaged with them, so you do feel, we feel sympathetic, so 'oh, it's the last thing they need', you sit there and feel 'ahh, what a shame', Yeah you do feel that, you do. You feel 'ahh, I didn't want it for them'. Ashley, Paediatrician; 377-380*

All participants shared the view that the second diagnosis creates additional challenges and burden for young people and their families. Ashley's excerpt (above) is reflective of having developed a relationship with the patient and family, connecting with them and reacting to the news of the second diagnosis, i.e. "I didn't want it for them". Sam, illustrated by the quote below, provides an oppositional relationship within this theme, stating there is more to do practically with CD but that this is no different to managing T1D alone.

> *I think it's more challenging but I don't think it's different….. they've got two things to do. They've got to count their carbs and give insulin and they've got to make sure those carbs haven't got gluten in them. So they've got two things haven't they, rather than one. Sam, Paediatrician; 153-158*

Almost half of the participants gave an account that portrays a sense of responsibility, and in some cases guilt, of their role in 'giving' patients an extra set of issues to manage.

> *… when you throw in… Coeliac disease as well, you're, you're putting another pressure on the child…. you do feel bad kind of… having to lump something else on them as well…diabetes was bad enough but then having to now have Coeliac disease as well, so you feel bad, feel awful for the child. Fran, Dietician; 83-102*

Children with T1D who are asymptomatic for CD are picked up through the CD screening process. In these situations, the team diagnose a condition they see as burdensome and advise

a GFD. This seems to be one of the challenges of working with this group, since these are situations where HCPs are not visibly bringing about improved health for the patient in the short-term (despite potentially improving health in the long-term), and perhaps elicit less gratitude from families, reflected by Alex below.

> *....none of them hate you but don't thank you for giving them the "problem"[inverted commas action made with hands], er because I think some families do see it as that because they find it incredibly difficult. Alex, Gastroenterologist; 186-191*

(iii) Acting in their best interests

The challenges described by participants were mostly followed up with descriptions of how they manage these feelings or make meaning from them. Ashley (below) refers to having responded differently with increased experience. Perhaps less guilt is experienced and a process of rationalising develops over time.

> *I used to feel more guilty about telling them about the diagnosis, I don't think I feel that guilt so much now. I think as you get older you feel you have the rationale that it's not your fault for it, so I don't feel guilty. Ashley, Paediatrician; 386-391*

Other nuances of this process of rationalising difficulties in their role include; knowing you have done as much as you can; following protocol; and acting in the child's best interests. Fran (below) describes a view shared by all the participants; their role is to give the child the best start and to keep them as healthy as possible.

> *.... that's what it's all about, it's about keeping them healthy now and trying to keep them healthy as they get older... we've got to make a good start on it really. Fran, Dietician; 366-371*

This idea is further supported by a longer, poignant narrative from Ann:

> *This goes back a long, long way but probably the one that stays in my mind the longest is someone who is now in her twenties, but sadly registered blind. She really struggled with her diabetes as a child and this was before we routinely screened for Coeliac disease…. we thought [she] was significantly struggling with her diabetes, she was failing to thrive…. it took us longer than it ought to have taken us to screen for… Coeliac disease. She was clearly positive….it made quite a dramatic difference to her glycaemic control… I look back and think, if we had done all of that earlier, would she still have the diabetes complications that she has…. so I can see the benefits of our screening programme. If I need to question why we do it, I do always remember that girl and I suppose her missed diagnosis. Ann, Paediatrician; 171-173*

Participants describe an important element of their role is to reduce risk; drawing this meaning from their role is perhaps a coping strategy, supporting them in bringing about a burdensome diagnosis and self-management regimes. The starkness of risk is highlighted in Alex's quote (below).

> *…if you didn't pick these patients up, if you didn't screen them, what actually are the risks to the patient of gut cancer, other autoimmune conditions and bone issues or infertility… I actually don't honestly know that we truly really know what the proper risks are……. but it's not zero, and as I say, you get a gut lymphoma in Coeliac disease and it's a death, the 5 years survivals are think are about 10%, so it's not good. Alex, Gastroenterologist; 209-221*

Testing for CD may also bring some security, since the long term risks are uncertain and "not zero". This element of illness prevention was reinforced by eight of the participants. Seeing health improvements or being responsible for making a difference was a powerful aspect of job satisfaction.

> *…once they're established on the diet they generally, their symptoms can improve and that's quite nice to see that you've made a difference maybe or made them feel better, despite it posing some additional challenges for their day-to-day life. Sophie, Paediatrician; 144-148*

In summary, this superordinate theme features a strong connection from the participants to the experience of the young person and their family. The empathy and connection perhaps heightens the challenges involved in making a diagnosis and supporting the management of the conditions. The feeling of responsibility for their role in this process seems to be managed well through a process of meaning making; seeing their role as one to keep the child healthy now and in the future. Experience in the role also seems beneficial in developing coping strategies.

Superordinate Theme 2: Diagnoses are together; but separate and uneven

The co-occurring nature of the two conditions seemed to automatically lead participants to thoughts of comparison, considering which condition is the worst to have or work with. There appear to be challenges in joining up the two conditions, working with them together and equally.

(i) Diabetes comes first and is often prioritised

Narratives comparing the conditions were provided by seven participants. The dominant view described T1D as more severe and risky in the short and long-term. Ashley portrays an interesting analogy of quantifying the burden.

> *... if you look at the weighted burden of managing a gluten free diet versus the weighted burden of managing diabetes, the diabetes burden is far greater. I think if they were to be able to quantify those on a set of scales, the burden of diabetes is greater. Ashley, Paediatrician; 44-49*

Participants spoke respectfully about the challenges experienced with CD, however there was an inclination towards describing T1D as the "worse" condition to have. This was linked to the possibility of reversing the symptoms of CD as long as the GFD is followed, and their view of the complications associated with both conditions. An opposing stance is depicted by Charlotte and Clare; they describe how CD can be more arduous on a daily basis for families.

> *....quite often, parents will say to me "diabetes was bad enough, but out of the two I'd rather have the Diabetes than the Coeliac to be honest" because they feel that it impacts on so much more of their life... they find that more of a struggle. Clare, Nurse; 47-56*

Perhaps the HCP's role is an influencing factor on this perception, with dieticians and nurses perhaps seeing different challenges based on the type of contact they have with families.

Participants described young people and families prioritising T1D over CD, as shown by Chris (below). This interpretation was linked to how families view the risks of T1D as higher, perhaps lacking the energy and resources to manage another condition or in some cases a lack of CD symptoms.

> *....maybe [families] would be erm more inclined to say 'well look I'll adhere to the diabetic treatment and diet because of the risk of going blind or getting kidney disease' for example, which is really, really serious, 'but actually we'll just get by with the tiredness', and' I can't think about fertility for now because that's twenty years away' er so it might be that you do pick up families that will have that kind of er approach. Chris, Gastroenterologist; 193-201*

This trend also emerged within the participants, with six reflecting a sense of priority towards the management of T1D or a perception that it is prioritised by others in the team. However, contrary to this, Mandy was clear in her message of giving equality to both in order to treat the whole rather than the parts:

> *I don't look at it as one or the other. For me they're both equally as important, from a dietetic point of view ....we're looking at it from the whole thing, it's not just the diabetes that's more important or the Coeliac disease we're treating the whole thing for the child. Mandy, Dietician; 258-264*

It is reasonable to assume that diabetes teams would be inclined towards prioritising T1D based on it being the team focus, and considering the immediate, serious consequences of poorly controlled T1D. However, viewing the two conditions entirely separately perhaps creates barriers to working with the conditions more holistically.

(ii) Diagnoses going along in parallel but difficult to join up

Participants referred to factors that appear to create barriers to working with the conditions equally. Chris reflects a sense of responsibility to families to discuss their CD diagnosis, however, due to clinic resources, often cannot see them within a preferred time frame:

> *.....clinics are so full that they're always too full to see children in a very timely manner and this is something that I'm conscious of, that by the time I'm often seeing them too long has gone by and I'm often erm feeling er that er, that we perhaps should've met up sooner… you might have already heard the diagnosis from the diabetic team. Chris, Gastroenterologist; 138-145*

Use of phrases, such as "in the back of our mind" and being "conscious of" and the tone used was indicative of a deliberation, perhaps reflecting that the system is not ideal to fit the way Chris would like to support families. There appeared no additional time in appointments for dual diagnosis patients, which can create a challenge of covering everything, more so if there are difficulties with one of the conditions. With more information to cover, Ann questioned how much families may actually take in and remember about CD during initial discussions:

> *...we do have our diabetes handbook, which is a big A4 folder with lots and lots of pages in it. And there is a section, paragraphs on Coeliac disease in there… it would be interesting to know what they have remembered other than there's a blood test at the next clinic visit. Ann, Paediatrician; 267-274*

> *.....they don't get a longer appointment slot in their annual review so it can be difficult to cover everything you want in that appointment…you very quickly have to prioritise. Charlotte, Dietician; 276-291*

The importance of providing a consistent message to families about how to manage the conditions was indicated by half of the participants. The value of consistency from the team

was linked to gaining trust and the message it sends to families. Charlotte's narrative suggests inconsistent messages can make her role difficult; she talks about less consistency with the team's approach to CD, compared to T1D:

> *...always the first thing is getting the diabetes under control and everybody agrees and quite often it's led from the doctors, they'll be like 'don't worry about the gluten free diet' but then that's a difficult situation when later down the line you're trying to then re-engage. Definitely for the Coeliac disease there isn't as much of a team message, it tends to be more to the dietician that is constantly as the families would say 'on at them' whereas the doctors kind of leave it to us to deal with. Charlotte, Dietician; 522-537*

There is also a feeling of hierarchy here, perhaps that families pay more attention to the advice given by the consultant. Service C offers a joint annual review, which was described as sending a subtle but positive message to the families about the importance of both conditions.

> *...it's one thing that's joint. And they know we're thinking of both conditions, it's that message to them that we're sending, it's not just your diabetes it's your Coeliac disease too that we're concerned about. Mandy, Dietician; 489-496*

This superordinate theme suggests there are varied views on prioritisation of the conditions. Despite participants acknowledging the importance of CD, the dominant stance and tone was of T1D being prioritised by young people, families and the team. Some professionals have concerns about mixed messages being given to families, which can make their role more challenging. Barriers to providing equal care for both conditions include; lack of time in clinic and lack of resources. The use of a joint clinic seems to be helpful in demonstrating an equal weighting to families.

Superordinate Theme 3: Sitting with certain uncertainty and complexity

Complexities of working with the dual-diagnosis were outlined around uncertainties of the diagnosis and management of CD in the diabetes population. The complexity of the dual diagnosis can result in less than ideal self-management.

(i) Complexity of the role

Five participants raised the issue of working with a condition that is not their area of specialty. This theme is illustrated by contributions from a diabetologist and gastroenterologist:

> *… being a diabetologist and not a gastroenterologist… sometimes we feel uncomfortable about them [diabetes patients with CD] and you're not quite sure what exactly is the right thing to do. Sophie, Paediatrician; 104-108*

> *… I do tend to hold back and not give as much information as I would do with children with just Coeliac disease. So I find that I'm probably not giving as much helpful information in an interaction… you're avoiding mixed messages and saying the wrong thing. Chris, Gastroenterologist; 124-134*

A lack of confidence was expressed by participants about working with the opposing condition to their specialist area; feelings of unhelpfulness and uncertainty were described. Charlotte spoke of wearing two hats with dual diagnosis patients:

> *… it's remembering that through the consultation that they'll ask you what about this? What about that? …. I find that quite difficult erm to remember to have both your hats on **all** the time with these patients and kind of remembering all aspects. Charlotte, Dietician; 285*

There was a tone here of needing to know or remember a lot and perhaps being asked questions outside her comfort zone. Charlotte commented on the quality of the service offered

to dual diagnosis patients; she perhaps feels responsible for offering the same specific knowledge and service for CD as the patient would have if managed by the GI team. Kate provides a different stance on complexity below; she again puts onus on the dietician to have the specialist CD skills.

> *I think you probably don't need specialist er clinical skills to look after children with Coeliac disease, they need specialist dietetic skills but not doctor skills really. Kate, Paediatrician; 100-102*

Participants described managing complexity by being honest with families about not having all of the answers and going to the 'expert' for advice (gastroenterologist or dietician). Having an expert to consult in such situations appears to offer reassurance.

> *We are so lucky to have such a good relationship with the gastroenterologist, we have a clear understanding that if there is any doubt that we have about Coeliac disease or any of our screening results, it's automatically discussed with them. So we don't ever worry about how we're going to deal with this or manage this. Ann, Paediatrician; 145-149*

The nature of responding to two conditions when you are a specialist in one appeared to add an element of complexity for some participants. There were extremes to this idea represented: for some it can result in them feeling out of their comfort zone, less helpful or feeling that they are providing a lesser service. Others did not think that specialist skills were required or acknowledged that specialist skills were held by different professionals, with whom they could consult. Experience of working with both conditions, and the relationship they have with the person deemed as the expert is perhaps key to mediating these feelings.

(ii) Diagnostic grey areas

There are uncertainties in the diagnosis, evidence base and management for the dual diagnosis and asymptomatic patients, described by half of participants. Chris talks about uncertainty for families.

> *…there always is er an element of uncertainty, because a positive blood test doesn't equate to actually having the full blown picture of Coeliac disease. So er, they will sometimes go away with a degree of uncertainty and that causes anxiety…the families are left with some anxiety related to uncertainty. Chris, Gastroenterologist; 69-81*

In circumstances where a diagnosis of CD is unclear, it seems that this brings trickiness for HCPs. Uncertainty in diagnosis tends to create anxiety in families; I wonder if this is paralleled in some of the HCPs too.

Asymptomatic patients and those testing positive on the blood test (tTG[29]) but negative on the endoscopy (frequently referred to as 'grey cases') seem the most challenging. Participants spoke about their uncertainty of not knowing what is the "right thing to do" with the grey cases. As shown in Alex's excerpt (below), it seems that clear-cut cases where HCPs can intervene and make a difference is rewarding. Circumstances where they are unable to give a definitive answer or to see that they are making a positive difference may make cases more challenging.

> *…on the one hand it's a very rewarding condition because it's a simple change of diet, albeit a big change for families, but you've made them better and you've reduced all those risks and they feel better. I think the more difficult ones are the patients that are asymptomatic. Alex, Gastroenterologist; 105-113*

[29] Tissue transglutaminase (tTG), a test used in the diagnosis for CD and to check ongoing maintenance of CD

Five participants spoke of uncertainty in the evidence base for asymptomatic patients, with reference to the lack of good quality longitudinal research into the effects of not following a GFD for asymptomatic patients. The "best" treatment options for asymptomatic patients are described by Ashley as an "unknown":

> *... what really is the best treatment, and that's an unknown….. is there a benefit to glycaemic control to go on a gluten free diet? And that is very debatable. Because* ***actually*** *there may be evidence that* ***actually****, you exclude the effect of having another diagnosis because they get exhausted by it all don't they, can't continue to carry all this burden, but actually the food you eat is different, more rapidly absorbed, less complex GI foods, so actually they may actually have a deterioration in glycaemic control following the gluten free diet, so it's complex. Ashley, Paediatrician; 69-80*

Here, the value of a GFD for asymptomatic patients is questioned, suggesting that it may even be detrimental to T1D. The doubt is emphasised in the tone and language i.e. repetition of "actually". HCPs are trained to follow evidence-based practice; when this is not clear it perhaps makes it difficult to make recommendations. Ann's quote (below) is an example that once again, empathy is shown by participants in considering "what would I do?" And again, they manage this challenging position by being honest with the families about the facts, allowing them to make their decision about how they want to manage the condition, and turning to the "experts" for guidance.

> *...we all have discussions amongst ourselves, what would we do in that situation? ... you feel if all the people in their specialist fields have a degree of uncertainty then perhaps we don't know…. So yes, there is definitely some uncertainty about what to advise. Ann, Paediatrician; 316-328*

In the grey cases, there seems to be an element of reassurance in the fact that the team has ongoing contact with the young person, and therefore they have the opportunity to detect health changes, as Ann describes:

> *... there is complete reassurance in that we know that we're still seeing the families, still seeing the children, we're able to monitor them, we're able to identify if something appears to go less well with the child ...it doesn't cause me sleepless nights. Ann, Paediatrician; 340-342*

There was also an element of acceptance from three of the consultants about the reality of medicine not always being certain. It is not ideal, but it is not something they worry about.

> *Erm, how's it feel about not knowing about what the absolute risks are? Well I don't think you can ever really know... if there's uncertainty then, it's never ideal for any of us is it, because you want to be as definitive as you can for the families. I mean does it worry me? Not really, all you can do is appraise people with the facts as you see them and hopefully those are accurate facts. Alex, Gastroenterologist; 225-234*

(iii) Striving for accurate control but accepting sub-optimal

Participants acknowledged the range of patients they see: Some cope well taking the diagnoses and recommendations "in their stride", while others struggle.

> *... I'm thinking of some very well controlled Coeliacs who have diabetes. But equally I can see not well controlled. There's a big range. Sam, Paediatrician; 32-33*

Nearly all participants referred to having families who responded very well and worked with the team; these were seen as the rewarding or easier cases. However, poor self-management was described as one of the challenges. Frustration when management of the conditions could be better was noted by eight participants. This was linked to concerns about health risk, and a

sense of putting in a lot of effort but not seeing changes was described as "disheartening" (Sophie; 222).

> *It's hard, it's frustrating….. if the parents don't take it on board then the young person isn't going to take it on board…that's why I worry, that's what we're here for, to try and reduce you know the risks to future health. Rachel, Nurse; 227-142*

Cases where participants feel families could be doing more to support the young person, and possibly to support them as HCPs were a source of frustration. The pivotal role of families in promoting effective management was highlighted by eight participants. A strong sense of empathy was shown for the challenges to families, however frustrations seemed to peak in cases where they cannot see why a family is unable to follow recommendations or "do the right thing", as shown in Charlotte's quote below. This perhaps reflects the complexity of psychosocial issues impeding adherence, which may be difficult to identify.

> *… it's very difficult…. we've got the child's best interests at heart and you are trying to support the families…there are plenty of other families that are in the same situation and they are managing to manage their diabetes and their gluten free diets. Charlotte, Dietician; 151-155.*

There was a stance from participants of needing to accept when management of the conditions is not ideal. This was in acknowledgement of T1D and CD being difficult conditions to manage day-in and day-out.

> *…if you can just keep people on board so even if they're not doing it perfectly but doing it pretty well, then actually that's great. So it's actually about facilitating that suboptimal adherence, saying you know actually it's not perfect but you know. Ashley, Paediatrician; 322-326*

The idea of sub-optimal control being 'good enough' in some situations may be challenging to HCPs, who train from a medical model to alleviate symptoms and disease. Kate suggested that she looks for improvements or outcomes beyond adherence e.g. progress with communication:

> *... you do come out and think 'well that was quite good' even though their HbA1C may be crashingly high and you know they've got Coeliac disease and they're not adhering to the diet.....once you start to have better discussions, you might actually be able to....work out what might be a way forward. Kate, Paediatrician; 264-269*

It seems that finding a balance between improving control of the diagnoses and engagement is a key aspect of the HCPs role, to improve outcomes from a clinical perspective, while having insight into the capacity of the young person and family, not "pushing them" to the point of losing them. This sense of "not losing" families was noted in five interviews, maintaining a positive relationship with families to keep them coming back to clinic, shown in the quotes below.

> *... just trying to keep that relationship going without them feeling that they are being judged um without them feeling that they have to be doing it right or else everyone will be feeling annoyed... it's not perfect but you don't want to lose them, you want them to keep coming to clinic. Fran, Dietician; 410-416*

> *...as long as it's not significantly raised you sometimes have to compromise... then they're happy to keep on with the diet. Otherwise you feel you're going to lose them completely. Charlotte, Dietician; 340-342*

There was also a theme running through the interviews of there being a limit to how much HCPs are able to do; after that it is the young people and families who choose whether or not

they follow advice. This seemed to be another coping response to managing challenges of the role.

In summary, within this superordinate theme, participants revealed the complexity that can exist within their role; however they cope with this via processes of knowing their limits, being honest with families and seeking support. The rewards from clear-cut cases seem to buffer the negative experiences of the grey cases. Another source of frustration can be poor self-management, particularly when participants do not understand the reasons for this. Despite an imbalance at times between feeling they are putting a lot of effort in and making little difference, there is recognition of the need to accept sub-optimal control and maintain engagement.

DISCUSSION

Research has begun to recognise the experiences of young people and their families with a dual diagnosis of T1D and CD (Love, 2013), and the practical challenges related to screening, diagnosis and management (Freemark & Levitsky, 2003; Sud et al., 2011). However, the phenomenology of HCPs working with this client group has not previously been explored.

Three superordinate themes emerged from this study: 'Connection and burden'; 'Diagnoses together, but separate and uneven'; and 'Sitting with certain uncertainty and complexity'. The participants' connection to experiences of the young people with a dual diagnosis and their families (particularly their role in making the diagnosis) was important, and there was a felt sense of disparity in managing the two conditions. As with other areas of healthcare in the modern NHS, participants experienced uncertainty and complexity within their roles. The use of adaptive coping strategies and ways of making meaning were evident in order to combat difficulties in the presentation of the dual diagnosis and the lack of an evidence base.

Enhanced empathy and connection to patients' experiences was revealed in this study, which may reflect a process of building therapeutic relationships. Larson and Yao (2005) summarised that engaging in clinical empathy leads to a sense of increased professional satisfaction. Connection and compassion towards patients is cited as a wellness factor associated with the absence of burnout (Eckleberry-Hunt et al., 2009), and is also important to young people with chronic illnesses and their families (Taylor, Gibson & Franck, 2008). Therefore, the findings of empathy and connection in this study are likely to be a positive influence on HCP wellbeing, the therapeutic relationship and patient care.

HCPs experience a range of presentations working with the dual diagnosis, from "clear-cut cases" (symptomatic for CD with positive biopsy results) to "grey cases" (asymptomatic or those whose CD diagnosis is not certain following biopsy). Uncertainty in the grey cases is just described in relation to CD, since T1D is described as being clear. In "clear-cut cases", facilitating a diagnosis and health improvement was reported as highly rewarding for participants. However, in the "grey cases", the sense of adding to the patient's burden by screening and possibly finding an additional life-long diagnosis was less rewarding. This led to unease in most participants and feelings of guilt in some participants. This supports findings by Wallace and Lemaire (2007), who summarised that patient interactions for clinicians are a source of both satisfaction and emotional stress; and that the feeling of making a difference is important for physician wellbeing and can buffer stressful encounters. So within the range of dual diagnosis cases, circumstances where HCPs feel able to make a positive difference are likely to buffer the effects of the more challenging "grey cases".

Despite the uncomfortable aspect of "adding to the burden", it is important to emphasise the strength of the sub-theme 'acting in the child's best interests'. Almost all accounts of challenges were followed up with narratives that reflect meaning-making, coping, and appraising of roles and position in healthcare. These included: Promoting health; reducing health risks; following guidelines; and knowing they have done as much as they can. This process of cognitive reappraisal might serve as a protective function and has been cited as a coping strategy in nurses (Lambert et al., 2007). Within the Process Model of Emotion Regulation (PMER), Gross (2002) asserts that emotional impact is altered by changing how a situation is interpreted; reappraisal can decrease the experience of negative emotions and increase positive experience. Some participants indicated that increased experience in the

field had been beneficial in creating positive re-appraisals from difficult situations, leading them to experience less guilt. Lambert et al. (2007) also found that distancing and problem-solving were coping strategies used more often by staff with more experience in the field. In this study, the HCPs' use of coping strategies to manage difficult experiences perhaps reflects professional hardiness and may serve to decrease the risk of burnout and emotional exhaustion, found within the healthcare professions (e.g. Erkstedt & Fagerberg, 2003; Maslach & Jackson, 1981).

Within the theme 'diagnoses together but separate and uneven', both narratives and interpretations gave a sense of disconnection within the dual diagnosis, with a predominant view of T1D being prioritised. This finding is not surprising since it is the diabetes team that also manages the CD in this study's sample. Interestingly, this theme was also found in a study of young people (and parents) with the dual diagnosis (Love, 2013), where T1D was viewed as more threatening; this study also reported that there was more of a focus on T1D by services and clinicians. It is possible that underlying messages of prioritising T1D could have clinical implications for present and future CD management and related physical and psychological well-being, such as not following the GFD or seeking support for self-management difficulties. This highlights the potential for service development, to perhaps move towards a more holistic approach, supporting HCPs and patients in managing the conditions together and equally.

The HCPs' understanding of how individuals with a dual diagnosis of T1D and CD understand and view these multiple conditions seems reflective of a wider issues of working with multi-morbidities, including the illness representations held by the individual. McSharry, Bishop, Moss-Morris and Kendrick (2013) reported differences in how patients viewed multi-morbidity of diabetes and depression. Some patients viewed the conditions as related, whereas

others perceived them entirely separately. The differences in illness representations was linked to; self-management behaviours; conflicts in managing the conditions; and prioritization of one condition. McSharry et al. (2013) suggest that HCP understanding of how patients perceive multi-morbidity, illness representations and the relationship between the two conditions, is likely to be important to understand within patient - physician interactions and to inform optimal patient self-management.

Service development may address the barriers to managing both conditions equally, which were cited in this study, including time in consultations, resources and inconsistent messages between professionals. Despite participants indicating good multidisciplinary relationships and support, inconsistent messages between professionals in the team may increase challenges for some clinicians, particularly those who see their role as having more responsibility for managing one or other condition (i.e. dieticians and gastroenterologists focusing on CD). Moving towards holistic management may include additional time in consultation, additional resources, and a dual review clinic (such as that run by Service C).

Increasing complexity is recognised as central to modern healthcare (DH, 2008). Participants predominantly acknowledged the uncertainty within their role; however coping responses were varied. For some, thinking they have a lack of expertise in one of the conditions seems to have led to feelings of unhelpfulness, uncertainty and lack of confidence, which was found in both diabetes and CD specialists. It seemed that those who did not experience this difficulty were those who felt buffered by a feeling of security brought about by rationalising that it is not their role to be an expert in both; those who accepted that complexity and uncertainty are realities of modern day medicine; and those who elicited support from colleagues perceived as ‘experts’. Seeking support is a coping mechanism linked to clinician wellness (Lim, Bogossian & Ahern, 2010; Mark & Smith, 2012). These findings

suggest that the position of not being an expert may contribute to feelings of stress and professional inadequacy for some HCPs.

A further source of complexity is working with families to facilitate optimal health management, and the frustrations when this is not ideal. Participants reflected how they strive for the best control for their patients; this is related to reducing risks, which is the fundamental goal of their work. Findings revealed that poor self-management for both conditions is understood when clear factors such as social issues or lack of resources (financial, practical and emotional) can be attributed to sub-optimal management. However, it seems frustrating to HCPs when families are deemed to have the resources needed to manage both conditions well, but fail to do so. Psychological assessment and shared formulations might have potential benefits in these situations.

Lack of control at work has been linked to stress and vulnerability factors in HCPs (Blackeman et al., 2005; Kearney et al., 2009; Shapiro et al., 2011). This feature of working with chronic health conditions may be salient to understanding the nature of working with the dual diagnosis. A literature review of parents of children with chronic illnesses (Fisher, 2002) described a need for parents to seek control to minimise uncertainty, a process which may be paralleled by the HCPs in this study. Siegrist's Effort-Reward Imbalance (ERI) model (1996) may also be helpful in making sense of the challenges. According to the ERI, high cost/ low gain work conditions can lead to emotional arousal. Siegrist further highlights the importance of control to cope with such demands. Using the ERI, we could postulate that the HCPs in this study may feel dissatisfied when their investments do not give rise to rewarding outcomes (i.e. good management of the conditions).

Again, coping strategies were used in response to these situations. Maintaining contact with families to monitor health, detect changes and keep them coming back to clinic were

seen as important, along with being honest about areas of uncertainty, and equipping families with the facts to make their own decisions. This response to uncertainty is similar to that described by Hayward (2006), who proposed that, to manage uncertainty in medicine, it is important for doctors to recognise processes underlying their attitudes to uncertainty and honesty regarding knowledge limitations (e.g., in the dual diagnosis cases this may relate to the uncertainty in the research / evidence base). Further responses involved an acceptance that "sub-optimal control" is sometimes a reality and a rationale that there is a limit to their capacity to bring about change.

Methodological Considerations

The use of IPA allowed for the open exploration of the understandings and experiences of HCP's working with this patient group. Since this area has not previously been researched, IPA has generated a number of themes, which can be further explored with other research methodology, such as quantitative questionnaires.

Two of the twelve participants were gastroenterologists, and ten were members of the diabetes team (occupying varying roles). The sample may be biased towards the experiences of the diabetes team; however this distribution of professional expertise is representative of the current care for young people with a dual diagnosis nationally. Three sites were included for recruitment to boost generalisability of findings. Despite this being a strength of the design, uneven numbers of participants from the sites may have biased results towards the service with the greatest representation. There was also a majority of doctors and consultants; more even representation of professionals would have strengthened conclusions and generalisability.

Despite attempts to guide participants towards discussing their feelings, there was a sense that at times, some held back in discussing their own emotions and experiences, and described an interest in the experiences of the patients. This is perhaps reflective of how HCPs view their role, selflessly supporting patients. It may also reflect a sense of discomfort or unfamiliarity with reflective practice. It is possible that further research, with specific questions based on the themes from this study or for the researcher to be a known, trusted individual to the team, would support the researcher to further explore some of the themes raised. It is important to note that the participants description and interest in their patients experience, is still linked to the phenomenology of their work and also associated to other factors that form their working experience, hence are personally salient to them. The participants' relationship to the things that matter in their lived world is the very essence of IPA. This interest in patient experience links to the HCPs own experiences of guilt, concern and complexities in their role and coping strategies they use. However, given that the participants made frequent reflections on the experience of their patients, and that this is not considered the usual focus of IPA, other qualitative methods could have been explored. One such approach which could have been used is template analysis (TA, King, 2012), which is very similar to IPA. TA is generally used with larger samples than IPA, and is useful for exploring relationships and comparing perspectives in order to find meanings. One of the strengths of TA is in exploring underlying thinking, feelings and assumptions, therefore on reflection, this approach may have been more appropriate to have used.

Checking of credibility was sought during analysis. Firstly, participants were asked to review transcripts prior to analysis to ensure the transcripts were an accurate reflection of their intended meaning, all participants reviewed and agreed to their final transcript. The process of cross-checking data analysis with academic supervisors and an IPA analysis group

adds to the confidence in the processes of arriving at the final themes. Lead clinicians from each site were sent the table of themes following analysis, and invited to contribute any feedback, unfortunately, no feedback was provided. It is acknowledged that participant checks would have been a valuable stage in assessing the credibility of the emergent themes and would have offered more insight into the trustworthiness of the results.

Clinical Implications

Since this study is the first to explore HCPs' experience in this area, findings may be of interest to other staff in the field, to help make sense of their own experiences, to highlight challenges that may have clinical implications, and to offer managers insight into service development issues.

The benefits of peer support, team meetings and increased experience to establish appropriate coping strategies were reported in this study. Mann, Gordon and MacLeod (2009) propose that reflective practice for HCPs can help to make meaning of complex situations and facilitate experiential learning. Therefore, peer group reflective practice may boost the HCPs support network in managing this group of patients. Since a range of views towards the dual diagnosis was evident, it may benefit HCPs to be mindful of their own assumptions (i.e. either thinking that the dual diagnosis is awful for the young person/ family or thinking that it is not an additional burden), to hold these to one side and be receptive to hearing the young person and family's experiences and needs.

Regular continuous professional development events are recommended, to provide opportunities for specialists in both T1D and CD to share their expertise. Support in formulating difficulties in treatment adherence may also be useful. An increased understanding of psychosocial issues (of patients struggling with self-management) could

help to reduce frustration for HCPs and to offer appropriate support for the young person and family. The benefits of integrating psychology within paediatric multidisciplinary teams include encouraging psychological thinking and advocating psychosocial factors (Jacobs, Titman, & Edwards, 2012). In this sample, psychological input is not available in all services.

Joint annual review clinics were described positively by those who use them. This system could be trialed by other diabetes teams, to evaluate the effectiveness from both a HCP and patient perspective. This may boost HCP confidence in managing the condition outside their specialism, and help them move towards a more holistic management of the conditions.

Future Research

Qualitative literature on the lived experiences of HCPs is limited and what does exist tends to be focused on staff working in trauma and end of life care; a focus on long-term conditions is scarce. Further research on caring for children with long-term conditions is warranted, to appreciate the challenges, meaning making and coping processes that arise. Comparing findings from this study to other areas of paediatrics would indicate if experiences of working with the dual diagnosis are different or reflect general findings.

With limited research and uncertainty about the dual diagnosis, further research is warranted. Increased prevalence of children diagnosed with CD (White et al., 2013) and T1D (Patterson et al., 2012), suggests a rise in the dual diagnosis can also be expected. From a medical perspective, better understanding of the relationship between glycaemic control and GFD, and the long-term consequences of not following a GFD for those with asymptomatic CD is needed. From a psychological perspective, a larger scale quantitative study of

psychosocial issues for young people, families and HCPs would strengthen the evidence base. Recommendations have been made based on the findings from this research, however, a study specifically eliciting the views of service providers and service users regarding what an ideal dual-diagnosis service would look like would support service development. With growing prevalence, expansion of this knowledge will enable services to consider specific needs in this area.

CONCLUSION

This study sought to explore the lived experience of HCPs working in the field of paediatrics, supporting young people with a dual diagnosis of T1D and CD. Findings reveal challenges in this area, centred around managing two chronic conditions (one of which may not be the HCP's area of specialty); a sense of unevenness in working with the two conditions and uncertainty around screening, diagnosis, management and the evidence base. There were also positive aspects of the role, such as being able to make a difference, improve health outcomes and reduce risk. Positive aspects of the role may serve to buffer HCPs against the more challenging aspects. An array of coping strategies were described which are likely to serve a protective function, including positive reappraisal, peer support and meaning-making. Recommendations for staff support and service development include dual review clinics, additional time in appointments, reflective practice and support for psychosocial formulations. Further research is needed to develop the evidence base and understanding of the medical and psychosocial needs of young people, families and HCPs in this area.

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Public Domain Briefing Paper

Body image in young people with Inflammatory Bowel Disease: A systematic review

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Healthcare professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease: An Interpretative Phenomenological Analysis

This paper presents a summary of a literature review of body image in children and young people with Inflammatory Bowel Disease. It also includes a summary of an original research study, exploring the experiences of healthcare professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease.

LITERATURE REVIEW

Background

Inflammatory Bowel Disease (IBD) is an umbrella term for long-term conditions of the gut including Crohn's Disease (CD), Ulcerative Colitis (UC), and Indeterminate Colitis (IC). Each cause inflammation in the intestine or digestive tract, and can lead to a range of painful, embarrassing and serious symptoms. Treatment may include a special tube-fed diet, steroids and even surgery (British Society of Paediatric Gastroenterology Hepatology and Nutrition, BSPGHAN, 2008). In some serious cases, a surgical stoma is created (opening in the abdomen) to allow waste to pass out of the body. IBD has been linked to a greater risk of depression, anxiety, poorer social functioning and self-esteem (Mackner, Crandall & Szigethy, 2006; Pinquart, 2012). Problems with body image have been found in adults with IBD (Muller, Prosser, Bampton, Mountifield & Andrews, 2010), and young people with IBD have described feeling different, embarrassed and having low self-respect linked to their bodies (Brydolf & Segesten, 1996). Long-term health conditions in childhood have been found to affect body image in children and young people; however, no published reviews of children with IBD have been completed.

Method

Five electronic databases were searched for articles published since 1998[30]. The search focussed on children and adolescents aged 0-19 years with IBD, which included a self-rating of their body image. The quality of each article was reviewed and given a quality score.

Findings

Nineteen articles were included in the review. Body image was measured within quality of life questionnaires, and there were no articles using specific body image measures. The findings of body image in young people with IBD who have mild symptoms or are free from symptoms was found to follow similar patterns to healthy young people. This includes body image being poorer with increased age from childhood to adolescence, and affected by delayed growth or being different to the norm for the young person's age group. Experiencing more severe symptoms was the strongest factor associated with lower body image. Body image was found to be lowest at the point of diagnosis; this is thought to be because of the severity of symptoms at the time before treatment begins. Treatment can reduce severity of the disease, however the side effects of some medications and treatment regimens (i.e. liquid diets and steroids) was linked to lower body image. There was no evidence in this study to show that the type of IBD (e.g. Crohn's Disease, Ulcerative Colitis, Indeterminate Colitis) significantly affects body image.

Conclusion and Recommendations

This is the first systematic review to look specifically at body image in children and adolescents with IBD. The review has brought to light the flaws in the current evidence,

[30] In 1998 a new form of treatment was licensed for IBD, which reduced the use of steroids and surgery. Therefore only research articles published after this date were included.

calling for further studies to improve the understanding of this area, which in turn may help to support those with the diagnosis. Research using a specific body image assessment, to compare with healthy children and adolescents over time would be beneficial. Recommendations include the routine assessment of psychosocial factors including body image for young people with IBD alongside their routine physical health checks.

RESEARCH STUDY

Background

Research shows there are challenges for healthcare professionals (HCPs) working with patients with chronic (long term) health conditions (Royal College of Physicians, 2004; Han and Klein, 2011; Kane, Priester & Totten, 2005; Turner & Kelly, 2000). Two chronic conditions that can occur in childhood include type 1 diabetes (T1D) and coeliac disease (CD); these involve life-long daily self-management. Young people with T1D have an increased risk of also being diagnosed with CD (Sud et al., 2010). The experiences of young people and their families with a dual diagnosis of T1D and CD has been studied (Love, 2013) and the practical challenges related to screening, diagnosis and management are reported (Freemark & Levitsky, 2003; Sud, Marcon, Assor, Daneman, & Mahmud, 2011). The aim of this study was to explore the experiences of HCPs working with children with a dual diagnosis of T1D and CD, to increase the understanding of this area of healthcare.

Method

Twelve HCPs (including doctors, dieticians and nurses) working in three NHS teams completed individual interviews. Interpretative Phenomenological Analysis (IPA) is a method used to make sense of information in order to understand a person's lived experience (Smith, Flowers & Larkin, 2009); this approach was used to understand the information from interviews. IPA is said to be particularly suitable for research in health psychology (Brocki & Wearden, 2006).

Findings

Three themes came out of the interviews; *'Connection and burden'*; *'Diagnoses together, but separate and uneven'*; and *'Sitting with certain uncertainty and complexity'*. Findings show there are both challenges and positive aspects to working with this patient group.

The HCPs showed a strong sense of connection to thinking about what the dual diagnosis must be like for the young people and families. There was also a sense of unease, and at times guilt about making the second diagnosis and advising additional health management. Managing two chronic conditions (one of which may not be the HCPs area of specialty) was linked to challenges. This was associated with a lack of expert knowledge in one of the conditions, lack of additional time and mixed messages coming from HCPs in the team. Young people with the dual diagnosis are managed by the diabetes team for both conditions and there was a sense of the T1D being prioritised over the CD. Uncertainty in areas of screening, diagnosis, and healthcare guidelines for the dual diagnosis can add to the complexity for HCPs. Due to the importance of self-management for both of these conditions,

it is at times out of the HCPs control to be able to bring about ideal management, as this relies on the young person and families.

The positive aspects of the role include being able to make a difference for the young people and their families, improving health outcomes and reducing health risks. The HCPs also use coping strategies to manage difficulties; these include making sense of their role (to keep their patients healthy and safe), knowing their limitations and seeking support from colleagues. The positive experiences and coping strategies seem to help the HCPs manage the more difficult aspects of their role.

Implications and Recommendations

It may be helpful for both HCPs and patients if services hold dual review clinics with diabetes and coeliac specialists. Additional time in appointments with dual diagnosis patients may also help the HCPs to cover all aspects of care for both conditions equally. Since HCPs use support from their colleagues to manage difficulties at work, regular reflective practice groups and opportunities to share knowledge and experiences may be beneficial. Since it seems more difficult for HCPs when the reasons for young people not following their treatment/ self-management are unclear, it is recommended that support is offered to understand and make sense of how psychological factors may lead to difficulties in self-management. Further research could continue to explore the experiences of HCPs in other areas of child healthcare, to see if there are differences depending on the patient group. There is also a need for increased understanding into both medical and psychological aspects of the dual diagnosis, and how services can best meet the needs of HCPs and the young people and families they support.

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APPENDICES

Body image in young people with Inflammatory Bowel Disease:

A systematic review

APPENDIX 1: GLOSSARY OF MEDICAL TERMS

Term	Definition
Adalimumab	Drug name. See biological therapy.
Barium meal	Procedure of radiographing the intestinal tract.
Biological therapy	Treatment aimed at various stages of the inflammatory process. They are genetically engineered medications made from living organisms that help to correct the imbalance of the gut immune system causing the diseases (Rutgeerts et al., 2009). Administered intravenously, subcutaneously or by injection. Used in the treatment of IBD.
BSPGHAN	The British Society of Paediatric Gastroenterology Hepatology and Nutrition. Professional body promoting standards of care for children with gastrointestinal, liver and nutritional diseases.
Colectomy	Surgical removal of the colon.
Corticosteroids	Also known as steroids. See steroids.
Cushingoid features	Fatty deposits on the face causing roundness. Side effect of steroid treatment.
Endoscopy	A procedure to examine the inside of the body, through a camera (endoscope) inserted through the mouth or anus.
Enteral nutrition	Delivery of a nutritionally complete feed, containing protein, carbohydrate, fat, water, minerals and vitamins, directly into the stomach or intestine.
ESPGHAN	European Society of Paediatric Gastroenterology, Hepatology and Nutrition. Professional body.
Fissure	Tear or ulcer (open sore) that develops in the lining of the anal canal (end of the bowel).
Fistula	Cavity or small channel that develops between the anal canal and the skin near the anus.
Ileocolonoscopy	Image investigation of the Ileum (small part of the bowel).
Ileostomy	The small intestine is diverted through a surgically created opening in the abdomen.
Immunosuppressive agents	Drugs that inhibit or prevent activity of the immune system, used in the treatment of IBD.
Infliximab	Biological drug. See biological therapy.
Naltrexone	Drug name. See Opioid antagonist.
Nasogastric tubes	A tube passed into the stomach via the nose. It is used for short or medium-term nutritional support.
Opioid receptor antagonist	Drugs that prevent the body from responding to opiates and endorphins.
Proctocolectomy	Surgical removal of the rectum and all or part of the colon.
Steroids	Anti-inflammatory medicine.
Steroid hump	Fatty deposit in the middle-upper back. Side effect of steroid medication.
Stoma	Surgically created opening from the colon or intestine to the outside of the abdomen.
Strictures	Restriction or narrowing of the anal canal.

APPENDIX 2: SEARCH TERMS

Search Terms:
1. exp body image/ OR self-concept/ OR self-perception/ OR body satisfaction/ OR self-image/ OR physical appearance/ OR body satisfaction.mp. OR body dissatisfaction.mp. OR body esteem.mp. OR IMPACT-I.mp. OR IMPACT-II.mp. OR IMPACT-III.mp. OR IMPACT 33.mp. OR IMPACT 35.mp. OR piers harris.mp. OR Minneapolis-Manchester Quality of Life.mp. 2. Inflammatory bowel disease.mp. OR IBD.mp. OR crohn disease/ OR ulcerative colitis/ OR colitis/ OR indeterminate colitis.mp. 3. Child* OR adolesc*OR young pe* 4. 1 AND 2 AND 3

APPENDIX 3: INDEX OF MEASURES

Abbreviation	Title of measure	Brief description
CBCL	Child Behaviour Checklist	Questionnaire for parents that measures child's competences and behavioural problems
CCSS	Cantril's Self Striving Scale	10 point global rating of quality of life. A higher score denotes a better QOL rating.
CCSS-c	Cognitive Control Strategy Scale for children	Disease related coping style instrument
CDI	Children's Depression Inventory	A brief self-report test that helps assess cognitive, affective and behavioural signs of depression in children and adolescents 7 to 17 years old.
CSI	Child Symptom Inventory	Behaviour rating scale that screens for DSM-IV emotional and behavioural disorders in children between 5 and 12 years old.
Dux-25	Dutch Children's AZL/TNO Quality of Life Questionnaire	Generic quality-of-life questionnaire
EuroQOL		Quality of life instrument, descriptive data on an individual's own rating of their quality of life.
EQ:5D	EuroQoL-5 Dimensions	Health related quality of life instrument included scales for: mobility; self-care; usual activities; pain/discomfort; sadness/anxiety/depression.
EQ-VAS	EuroQoL-Visual Analogue Scale	Thermometer like scale, provides dimensions that measure three levels of severity; no problems or some/ moderate problems or extreme problems
FAD	Family Assessment Device	60 item measure that assesses family functioning on six dimensions; problem solving; roles; affective responsiveness; affective involvement; behaviour control.
HRQL	Health Related Quality of Life	Health related quality of life instrument.

IMPACT		IBD specific HRQOL measure for 9-17 year olds. Encompassing six domains: bowel concerns, body image, functional/social impairment, emotional impairment, tests/treatments, and systemic impairment.
ITIA	I think I Am	5 dimensions of self-esteem measured (physical characteristics, skills and talents, psychological well-being, relationships with parents and relationships with others) for children aged 10-16 years.
IBDQ	Inflammatory Bowel Disease Questionnaire	A quality of life (QoL) questionnaire for patients with inflammatory bowel diseases with four dimensions: bowel, systemic, social, and emotional.
KINDL	German KINDL Questionnaire	Quality of Life scale. Likert scale items with six dimensions; physical well-being; emotional well-being; self-worth; well-being in family; well-being regarding friendships; well-being at school.
LCAI	Lichtiger Colitis Activity Index	Subjective and objective criteria to assess eight ulcerative colitis symptoms.
MMQL	Minneapolis-Manchester Quality of Life Instrument	Originally developed for child cancer survivors measuring components of; physical functioning; psychological functioning; social functioning; cognitive functioning; body image and outlook on life.
NZDep06	New Zealand Individual Deprivation Index	Index of socioeconomic deprivation
PCDAI	Paediatric Crohn's Disease Activity Index	Measure of symptoms of CD
PUCAI	Paediatric Ulcerative Colitis Activity Index	Measure of symptoms of UC
PedsQL	Pediatric Quality of Life Inventory	Measure of child and parents perspective on child HRQOL with scales for; physical, emotional, social, school related aspects of HRQOL.
PHSCS	Piers Harris Self Concept Scale	Assesses self-concept in individuals ages 7 to 18. Includes six subscales: Physical Appearance and Attributes; Freedom From

		Anxiety; Intellectual and School Status; Behavioural Adjustment; Happiness and Satisfaction; Popularity
RCMAS	Revised Children's Manifest Anxiety Scale	Measures the level and nature of anxiety, as experienced by children
Sense WearPro2 armband		Device which provides information on physical activity and energy expenditure
SPAA	Self-Perception Profile for Adolescents	7 subscales; scholastic competence; social acceptance; athletic competence; physical appearance; close friends; behavioural conduct; and general self-esteem.
SCAS	Spence Children's Anxiety Scale	44 item measure of severity of anxiety symptoms in youths.
SSQ	School Information Questionnaire	Gather information from teachers about behaviours and symptoms directly associated with Attention Deficit Hyperactivity Disorder that may displayed in a classroom setting.
SODA	Situational Obstacles to Dietary Adherence	Evaluates the ability of adolescents to cope with situational obstacles to their dietary adherence. Originally developed for adolescents with diabetes. Two scales; confidence scale and behaviour scale.
STAIC	State Trait Anxiety Inventory for Children	Self-report measure of state anxiety and trait anxiety
TACQOL	TNO-AZL Children's Quality of Life Questionnaire	A generic health-related quality of life questionnaire
UC	Ulcerative Colitis measure	Measure of symptoms in UC.
UCL-A	Utrecht Coping List for Adolescents	44 items to assess coping behaviour in day to day situations.
YSR	Youth Self-Report	Family of screening tools for behavioural and emotional problems in children and adolescents.

APPENDIX 4: INSTRUCTIONS FOR AUTHORS

This paper will be edited for submission to the journal 'Health Psychology Review'. Instructions for authors can be found on the following hyperlink.

http://www.tandfonline.com/action/authorSubmission?journalCode=rhpr20&page=instructions#.U35EZy-7n2g

APPENDICES

Healthcare professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease

APPENDIX 1: IPA QUALITY CRITERIA

What makes a good IPA paper (Smith, 2011)

1	The paper should have a clear focus
2	The paper will have strong data
3	The paper should be rigorous
4	Sufficient space must be given to the elaboration of each theme
5	The analysis should be interpretative not just descriptive
6	The analysis should be pointing to both convergence and divergence
7	The paper needs to be carefully written

APPENDIX 2: INTERVIEW SCHEDULE

(Version 1, 19.10.12)

The following interview schedule outlines the general topic areas which will be questioned during the interview and prompt areas which may be used to help facilitate the interview.

Experiences

Maybe to start, you could tell me why you chose to work with this particular patient group? How you came to work in this field?

How would you describe your role with children with T1D and CD and their families/ within the team?

What do you think about children who have a dual diagnosis?

- Do you feel you are seeing more children diagnosed with both conditions?
- If so, why do you think that is?
- Do you feel there are particular challenges for these young people and their families?
- If so, what do you think these are?
- Are there challenges to this work? Why? Why not?

Could you tell me about a patient with a dual diagnosis you've worked with that has been a good/or positive experience for you?

- What made this experience positive?
- How did you feel about it at the time?
- Was this a typical case?
- What do you find is the most rewarding aspect of your role?

Could you tell me about a patient with a dual diagnosis you've worked with that has been a difficult experience for you?

- What made this experience difficult?
- How did you feel about it at the time?
- Was this a typical case?
- What is the most challenging aspect of your role?

Are you involved in the diagnostic process or sharing information about complications (possible future complications?)

- What is your view of how this process is for the child and their family?
- How do you feel about long term complications for patients?
- Can you tell me what that's like for you?

Do you find yourself worrying about these patients?

- What are your main concerns?

- Do these concerns lead to you changing your practice, doing anything differently?
- Has your practice changed over time since working with these patients?

I understand that this area involves some focus on a patient's own self-management of both conditions. What is this process like for you?

- Do you have any particular concerns about self-management – does this apply to all of your patients or just some?
- What is the most challenging aspect of this for you as a health professional? Or what might be the most challenging aspect of this for a health professional working in this area?
- How do you respond to the challenges involved in supporting patients and families in self-management?

<u>Coping</u>

As part of your role, are you involved in dealing with situations where there are strong emotions?

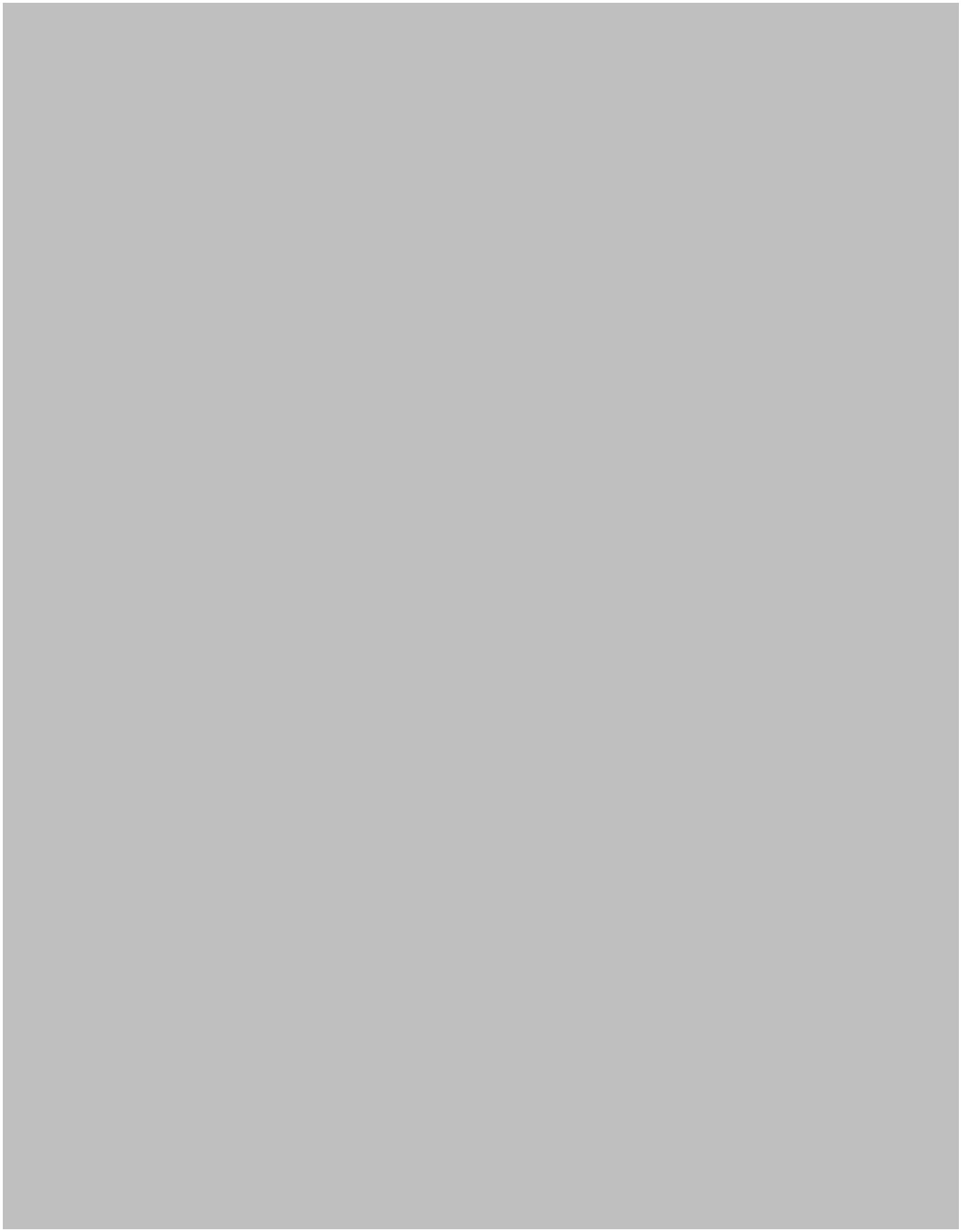
- What sort of emotions have you experienced in the children with a dual diagnosis and their parents?
- How do you feel about that/ what is it like for you?
- Do you have any particular ways of responding to these situations where patients/ families are highly emotional?
- Are there any support networks in place for you? (supervision, group supervision, formal/informal)
- Do you make use of these? How are they beneficial?
- If not, what stops you?
- Is there any form of support which is not currently available which you think would be helpful?

End of the interview

We've now covered all of the questions I wanted to ask. Is there anything you've not had the opportunity to say which you'd like to add to the interview?

(Version 1, 19.10.12)

APPENDIX 3: SPONSORSHIP LETTER, UNIVERSITY ETHICS



APPENDIX 4: PARTICIPANT INFORMATION SHEET

UNIVERSITY OF BIRMINGHAM

Exploring the experiences of health professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease.

My name is Victoria Hobday, I am a Trainee Clinical Psychologist at The University of Birmingham, completing a Doctorate in Clinical Psychology. I would like to invite you take part in a research study. Details of the study are outlined below, which explains the purpose of the study and what it would involve for you. Please read this information before you decide whether or not you would like to participate. If you have any further questions please feel free to contact me.

- **What is the purpose of this research?**

There has been a growing research interest in children and young people with a dual diagnosis of type 1 diabetes and coeliac disease; current research being undertaken at The University of Birmingham is focusing on the experiences of children with this dual diagnosis and their families. This study aims to explore the experiences of health professionals working with children and young people with a dual diagnosis of type 1 diabetes and coeliac disease, to develop individual and shared accounts of the experiences of working in this field.

- **Why have I been invited to take part?**

  Your team has been identified as a multidisciplinary team working with children and young people with a dual diagnosis of type 1 diabetes and coeliac disease. Health professionals who have worked with this patient group for a minimum of three months are invited to take part.

- **What will happen to me if I agree to take part?**

  If you agree to take part in this study you will be asked to participate in an individual interview with me which will be audio-recorded and transcribed verbatim. The interview will last up to an hour and will be arranged at a time and place convenient to you. If you are interested in taking part in this study, please contact me using the details below. You will have the opportunity to ask any further questions before arranging the individual interview. Prior to the interview you will be asked to sign a consent form for your participation. For the purpose of confidentiality, you will be assigned a pseudonym. Your consent form will be kept separate from your data and only your interview data will be associated with your pseudonym.

The interview will ask questions about your experiences of working with children with a dual diagnosis of type 1 diabetes and coeliac disease. The interview will then be transcribed verbatim; any identifiable information will be omitted for the confidentiality of you and your team.

- **What will happen if I do not want to carry on with the study?**
  Your participation is voluntary and you are free to change your mind and withdraw at any time during the research interview, without giving a reason. You may withdraw your interview entirely or in part, without giving any reason and without judgement up to two weeks following the interview. Should you choose to withdraw from the research, you can contact me on the details below and your data will be deleted. The date by which you can withdraw from the study will be provided to you on the day of your interview.

- **Expenses and payments**
  Interviews will be arranged at a time convenient to you and will take place at your usual place of work. No expenses or payments will be provided to you.

- **What will happen to the results of the research study?**
  The recordings from the interview will be heard only by me, and the transcripts will be looked at by myself and my research supervisors at The University of Birmingham. Transcripts may be checked by external examiners as part of the Doctorate qualification but no identifiable information will be included in these circumstances. Data will be stored securely on a password protected device and your transcript will be associated with your pseudonym only.

  Quotes from the interview may be used in the write up of the study for my Doctorate in Clinical Psychology, and in any subsequent publications. However any identifiable information will be omitted or changed from quotes to ensure you will not be identifiable by comments used.

  An executive summary of the research and findings will be made available to you and your team. The details of any subsequent publications made as a result of this study will also be made available.

  Audio-recordings and interview transcripts will be kept by my academic supervisors at The University of Birmingham until the qualification has been awarded (not usually more than 5 years). Anonymised electronic files will be kept for 10 years from publication.

- **What happens if I have any further concerns?**
  Should you have any questions or concerns at any point during or following the research process, you can contact the researcher by telephone or email.

Thank you for reading this information and for considering the research. If you would like to discuss any aspect of this research or to indicate your interest in taking part please contact me on the details below.

(Version 1, 19.10.12)

APPENDIX 5: CONSENT FORM

UNIVERSITY OF BIRMINGHAM

Research site: .....

Study number:.....

Participant Identification Number:.....

Title of Project: Exploring the experiences of health professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease.

Researcher: Victoria Hobday

Please initial box

1. I confirm that I have understood the information sheet dated 19.10.12 (version 1) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily. ☐

2. I understand that my participation is voluntary and that I am free to withdraw at any time during the research interview, without giving any reason. ☐

3. I understand that the research interview will be audio-recorded ☐

4. I understand that following the research interview, the researcher will send me a copy of my interview transcript. I will be given a period of time for reflection and allocated a date, by which I can request changes to the transcript or to withdraw my data. The researcher will then contact me at which point I may withdraw my interview entirely or in part, without giving any reason. ☐

5. I understand that the data collected during this study will be looked at by the researcher and relevant others at the University of Birmingham to ensure that the analysis is a fair and reasonable representation of the data. ☐

6. I understand that direct quotes from my interview may be published in any write-up of the data, but that my name will not be attributed to any such quotes and that neither I nor my team will be identifiable by my comments. ☐

7. I agree to take part in the above study.

.....	.....	.....
Name of participant	Date	Signature
.....	.....	.....
Name of researcher	Date	Signature

(Version 1, 19.10.12)

APPENDIX 6: DEBRIEF SHEET

UNIVERSITY OF BIRMINGHAM

Exploring the experiences of health professionals working with children with a dual diagnosis of type 1 diabetes and coeliac disease

Thank you for taking part in this study.

If you have any worries or concerns after talking about your experiences at work, staff support is available within your Trust, the attached leaflet provides information on this service and how you can access this support.

I will be in contact to give you a copy of the transcript from your interview. If you have any questions in the meantime, please feel free to contact me.

Thanks again,

Victoria Hobday

(Version 1, 19.10.12)

APPENDIX 7: EXAMPLE EXTRACT OF DATA ANALYSIS

How does that feel for you in those situations?

Erm, it's very difficult. Erm because at the end of the day we've got the child's best interests at heart and you are trying to support the families. But then at the other end of the spectrum, you know there are plenty of other families that are in the same situation and they are managing to manage their diabetes and their gluten free diets, so just trying to work with them as best as possible.

And you said if they tend to be following the diabetes diet quite well they can often manage the coeliac diet well too. Can you tell me more about that?

Yeah definitely, I would say my experience would be that if they're well controlled with their diabetes they tend to be quite well controlled with the coeliac and it's more so the kids that aren't very well controlled with their diabetes that aren't quite as strict with their gluten free diet as well.

The other thing you said was the difficulties in keeping children and families motivated. Whose role were you referring to in keeping the child motivated?

I think it's me and the parents, a bit of both really. I think initially a lot of the families are very motivated and go about reading labels and being super, super strict And then as time goes by we do an annual review appointment and you kind of go back over the basics and you see that they say 'oh there was that takeaway and weren't quite sure if it was gluten free or not' and you can see that there's little things that are starting to slip in, so you do have to give them a reminder. And a lot of families will be like 'it's been good to meet and you know it reminds us to be a little more cautious again'. And I guess the same approach with their diabetes, we meet them annually and every so often… a lot of families will be very strict with the carb counting initially and then sometimes you'll meet them a few years down the line and things have slipped a little bit and they haven't appreciated that their child has grown and their portion sizes have changes and they're still guessing the same numbers as they did when they were smaller. So it's all about getting the weighing scales out again. So it's a very similar approach we'll take to both conditions, with a little reminder and going back to basics and the importance of being strict with both conditions.

Wanting to do the right thing, support, non-judgmentally Frustration vs empathy

Taking responsibility for motivating the child, rescuing the situation when it deteriorates

Need for accuracy, to be strict managing conditions. Control feels safer/ reassuring for her.

"We've" =collective. Have responsibility for others. Tone: Irritable/ frustrated. Trying to do the best by everyone. Sense of frustration? Why can't they just do it? Others do! Discrepancy between feelings of frustration with the families & empathy.

Copers vs non-copers. Managing is about "control/strict"
Joint responsibility. This seems different to tone of other sections where she takes more responsibility. Motivation can drop over time. Responsible for 'bringing them back' Having thanks/ reward for re-motivating them. This is valued. The task of managing both over time, they can slip. Having to go over it again. Sense of ever making progress? Control going up and down. Need to be strict, be in control, maintaining order.

The position of being responsible for doing something she wasn't trained for

Some families can learn and self-manage in a sophisticated way. Others can't and need simpler information or more support.

Aiming for good enough.

And, erm, you also said that one of the things for parents is to think about counting the carbs and their own numeracy ability comes into that. Can you tell me what's that's like for you to work with?

Er, well I think it's well known that numeracy levels in the UK are much lower than the rest of Europe, and we definitely encounter that with our families in that from diagnosis, erm it's sometimes very easy to see if families are struggling with numbers. Because diabetes is <u>all numbers,</u> your dose of insulin is a number and your meter is a number, every food label is a number. So some families, some parents have been dyslexic with numbers, others have obviously lacked school very early and didn't attend much when they were there, so haven't really encountered or understand decimal points before. So it's really going <u>back to basics</u>. I think as a dietician, you haven't been taught how to be a math's teacher at university, but it's definitely skills that we've all had to take in our approach and I kind of take a bronze, silver and gold approach to my teaching with families. So you start with very basics you know and erm with carb counting and if they're managing that then you kind of progress and progress until you get to the point when they are dose adjusting their insulin and you know they've got a very flexible way of eating. But some families don't do that, they would never cope with that, So with start with what they call 'consistent carbs' and we give them a very set amount to stick to at meal times and we work with them a lot with pictures with that and erm weighing scales so they don't have to rely on actually working things out as much themselves. So they have the benefit on the ward, they have a few days of seeing what the meals look like and a lot of families erm kind of go on that and try and give their children a very similar looking portion to what they had on the ward. And then we use the 'carbs and cals' book a lot, because then they have a picture and underneath they've got the weight of the actual portion size, so we give them free weighing scales when they first get diagnosed and a 'carbs and cals' book. So at least then all they need to do is weigh the portion and that can usually manage that. Erm and then you <u>know that the carbs will be accurate enough</u>. And a lot of families, we encourage them to bring in labels or foods they would usually eat frequently.

Additional challenge to work with. Families overwhelmed with numbers, she's asking them to do things they find hard.
Maths- not trained for, had to take on additional skills. I'm a dietician not a maths teacher! Teacher role, take them one step at a time (education language used "bronze, silver, gold")
Flexible, adaptive.
Interesting- no control discourse on this page.
Adapting style to get it right, pitch it right for their abilities.
Doing it together.
(She commented about benefits of the ward previously)The ward is beneficial-is there something about this feeling safer for her? More staff input? Try to minimise pressure on the families- practical things. A sense of reassurance that there is some accuracy.
"Accurate enough"-change in tone from being strict.

APPENDIX 8: EXAMPLE OF SUPPORTING QUOTES

Superordinate theme 2: Diabetes comes first, but is separate and uneven	
Diabetes comes first	**Going along in parallel but difficult to join up**
Erm, I think the complications with coeliac disease probably feel less severe to me than the complications of diabetes, erm, so er it doesn't feel too bad actually. I think always comparing the two then diabetes is the one that's much more of a challenge because it, the complications are much more real… erm and with coeliac disease I think you can if you manage it well than you can say everything going to be ok. Kate; 401-405	I often feel there's not quite enough time in a clinic setting. I'm thinking particularly of the individuals with a diagnosis of type 1 diabetes and coeliac disease, I probably spend a significant portion of the consultation talking about the diabetes and then ask any symptoms and how are things going with the gluten free diet, you know it's not the first thing we talk about when they come in….the focus is not massive by any manner of means on the coeliac disease. Ashley; 46-58
My feeling again, without looking at a whole clinic, is that some of our teenagers are completely ignoring their coeliac disease. Some of them are also ignoring their diabetes, but not all of them, so some of them will prioritise their diabetes over their coeliac disease and some of them won't prioritise either. Um, it's unusual in my experience to find someone who's prioritising their Coeliac over their type 1 diabetes Chris; 53-57	within the confines of our diabetic clinic appointment, most of the plans were related to her diabetes, with leaving the onus on the mother to continue with contact with the diabetes nurse specialists and to let us know how things were going from the gluten free diet point of view. Kate; 73-79
there's a huge emotional upset when you get a diagnosis of something like type 1 diabetes in particular. Er, I don't have either condition so it's difficult for me to say in terms of which one I would personally prefer to have if you were going to give me the two… a choice, then I'd prefer to have coeliac disease because it's… you change your diet, I'm going to say 'just' change your diet, it is a quite significant change to your diet, but you change your diet and then get on with your life. Whereas diabetes is much more invasive in terms of your day-to-day living, you have to count carbohydrates, do your blood tests, Alex; 62-69	you've got a lot to try and discuss in that short clinic appointment. And you know, as I'm reflecting on this now, the coeliac disease was not first and foremost, it was mentioned all of the way through the consultation, but it wasn't a 'we need to focus completely on a gluten free diet at this point'. Ann; 84-88
I think those complications mainly relate to diabetes, because the complications are much more significant and er generally are more life limiting medically, erm because the chances of something life threatening happening with coeliac disease is very very slim. Erm even if they don't stick to the diet, and that's the truth because there's many people with	I feel we do mention is, but we're also talking about a lot of other things. I know we have a plan to have something in writing to hand to them; we do have our diabetes handbook, which is a big A4 folder with lots and lots of pages in it. And there is a section, paragraphs on coeliac disease in there, so if you're saying are people informed and told, we'd say 'oh yes', but if you were to question them a week after coming to visit at the new patient clinic, again it would be interesting to know what they have remembered other than there's a blood test at the next clinic visit. Ann; 267-274
	The diabetes is what they struggle with. And maybe it's my bias as well because I see the diabetes. But in the coeliac clinic, because our patients don't go to the coeliac clinic just come

untreated coeliac disease so the risks for example of cancer are not zero but they are much smaller than the risks to the child's health from poorly controlled diabetes. Sophie; 181-186 I suppose I don't see it as the most important role in clinic to be persuading someone who has had discussions with other people about whether they do or do not go on a gluten free diet. They tell us their child is asymptomatic their child is thriving, their diabetes control may not be excellent but it's not something I push. Erm, obviously I would take guidance from the gastroenterologist and other people in the team if they strongly feel we need to make a huge effort Ann; 213-220 I think personally I would feel that the coeliac takes less and less or a priority for them, they're following it and maybe it is easier for them because of all of the products that are available, I don't know. And yeah it doesn't seem a big issue for these families. I saw a girl yesterday, I hadn't thought of her until now, who's on a coeliac gluten free diet and you know in the consultation we must've talked about that and she had some abdominal issues, we must've talked about that for 2 or 3 minutes in a 30-40 minutes consultation. Ashley; 270-276. I think it is harder because they don't always see the symptoms with coeliac disease, erm so I think a lot of families will take more risks with their coeliac disease and maybe go somewhere and think 'this is probably not fully gluten free' or 'there could be cross-contamination' but they take the risk with coeliac disease rather than…we obviously have kids with allergies and things, so if the child had a nut allergy they just wouldn't take that chance. Erm so I guess they often put a lesser significance on the coeliac diagnosis than they do erm with their diabetes, I definitely think coeliac disease comes down the list. Charlotte; 515-522 Yeah, I think I probably, I don't look at it as	to the diabetes clinic, we want to do all the things that I think we should do in the diabetes clinic. Ashley; 171-177 it's a shame for the child because I feel they need to be supported really and like I said everyone needs to be giving the same advice to them and they need to know what it is they're supposed to be doing, not er… not having one person telling them one thing and another saying different Fran; 469-472 often the dieticians will take over afterwards for the on-going management, and the diabetic dietician will primarily lead on that. Er, so it's a little bit disjointed in that it's not my gastro-dietician that provides the er main advice and follow up…that's why we have a joint clinic with the endocrinology team on occasions to try and provide coordinated advice with the dietician and the gastroenterologist and the endocrinologist in the same room. Chris; 15-20 the clinics are so full that they're always too full to see children in a very timely manner and this is something that I'm conscious of, that by the time I'm often seeing them too long has gone by and I'm often erm feeling er that er, that we perhaps should've met up sooner and so we do spend a lot of time telling them, actually you are important to us and the fact that you've waited quite a long time for a gastro appointment. And you might have already heard the diagnosis from the diabetic team, erm and I hope they're alright with that because erm, it's, they've heard it sometimes second hand. But in fact the diabetic team, I think, handle it quite sensitively. Chris; 138-145 that's something that's also in the back of our mind is that we like to see children in a really timely manner and ideally we'd see them as soon as the results come back, but I don't get their results sometimes for four weeks after their tests, which is actually too long. So we're also dealing with families who, where the system doesn't work as well as we think it should do, so we also have that sort of dynamic in our consultation Chris; 147-151

one or the other. For me they're both equally as important, from a dietetic point of view I need to make sure that they're eating enough calories, they're eating enough carbs, they're diet is nutritionally balanced, they're growing, they're not having any symptoms, they're taking their insulin, they're HbA1c is coming down or is good, we're looking at it from the whole thing, it's not just the diabetes that's more important or the coeliac disease we're treating the whole thing for the child. Mandy; 258-264 I've got one girl that's really struggling and she says it's not the diabetes it's the coeliac disease. Whereas usually they'll say it's the other way round. But she says 'I could deal with it if it was just one, I could deal with just the diabetes, I can do the insulin' but she just wants to eat the same foods as her friends. It's all about fitting in with the children Fran; 347-351 Yes, yes, yes, I think because you know the diabetes has been around for a long time and you know its been at the forefront and going back to the fact that they may not have had signs and it is difficult to avoid gluten in stuff. All of those reasons make it a second best sometimes I think. Rachel; 229-231 you know you don't want to think of them as having poor health outcomes because their health has been…their diabetes has been poorly controlled in younger life but unfortunately you do have a lot of patients who do have poor control despite our best efforts to engage them so I think that can be a very challenging erm area. I suppose in terms of the coeliac, although its that kind of dual burden and dual diagnosis, I think diabetes is often er has more of a significant impact on their day to day life and risks for long term health implications. So I think that would probably be the dominant one that would cause problems with day-to-day life. Rachel; 163-169	So you've given the information up to a point, but you can see from their point of view, if she can't see that her daughter has any abdominal symptoms, she couldn't see the link potentially between her incomplete gluten free diet and her poor diabetes control. Clare; 79-82 I would hope that I always give the opportunity for people to bring them up, but often you're aware that you've maybe spent more time on one aspect of diabetes care, Ann; 121-123 if you have team members giving different messages to families, they don't feel that they have someone that they can trust. I think the consistency in the message and honestly in the message is something that we have an obligation to give to the families. Ann; 224-230 Definitely the diabetes is easier to hold, I do that all the time, but I don't do the coeliac disease as much ….. its kind of just remembering not to suggest anything that isn't erm, suitable… it can be hard to remember about the coeliac disease as well….So it's a lot of detailed information with that, so it's trying to remember then also the coeliac disease in there can be a bit difficult. Charlotte; 360-378 And even in terms of how the team approach it, erm if you have a very poorly controlled erm child who's family are struggling already, always the first thing is getting the diabetes under control and everybody agrees and quite often it's led from the doctors they'll be like 'don't worry about the gluten free diet' but then that's a difficult situation when later down the line you're trying to then re-engage with the family and obviously they're well aware that the doctor kind of said 'don't think about the gluten free diet' and they've just accepted that point that they don't have to follow the gluten free diet, its vey hard to then re-engage because they've got that message that it's not as strict. While with diabetes, we would always be 'better control, better control' and the whole team give a much more consistent team message. And there's a lot of research in diabetes that if the team give a consistent

	message you tend to have better compliance and control, and I think it's definitely for the coeliac disease if there isn't as much of a team message, it tends to be more to the dietician that is constantly as the families would say 'on at them' whereas the doctors kind of leave it to us to deal with that issue. Charlotte; 522-537 But with coeliac disease, I don't think the doctors have as much awareness and will probably think 'oh the dietician can look at that for you' and so there isn't as much sort of team support and team consistency with the message with coeliac disease. *Why do you think that is?* Erm I think probably because we are the diabetes team and that is our main focus and a lot of our patients are picked up through screening so I guess we meet the family and the focus is on diabetes and then when they get the diagnosis of coeliac disease it is the dietician that tends to take the lead on that advice. So I think the doctors kind of always bounce it back to us because they know we're the experts and they're scared of saying something wrong. Erm so I don't think they say anything… they wouldn't say anything wrong, its just more that they don't feel confident with the coeliac disease to kind of give the answers, so they always bounce it back to us. Charlotte; 539-559 We do a joint coeliac diabetic clinic and we have the gastroenterologist that comes, and the diebetologist. We do a joint clinic and there's a dietician as well. Erm, my experience from the clinic, I don't feel that gastro-wise that there's much that they do. I think it's the dietetics that's the most important, because we're the ones that can really help them to change and we'll probably have the answers of how we can help them with the bits they are struggling. Whereas the medics are probably, if the tTGS are raised they'll say we need a diet review, so it will come back to us Mandy; 165-172 we still kept it going because we felt it was really important for them to have that joint review. And also it means they don't need to see the gastroenterologist separately and the

	diabetes team separate, its one thing that's joint. And they know we're thinking of both conditions, its that message to them that we're sending, its not just your diabetes its your coeliac disease too that we're concerned about. Because from the diabetes team we kept that going, we didn't decide to not continue it. Mandy; 489-496 If we can work together as a team and we all say the same thing, then hopefully at some point the families going to think 'well they're all saying the same thing so there must be something you know important about this. Rachel; 237-239

APPENDIX 9: INSTRUCTIONS FOR AUTHORS

This paper will be edited for submission to 'British Journal of Health Psychology'. Instructions for authors can be found on the following hyperlink.

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