CHILDREN, YOUNG PEOPLE AND COELIAC DISEASE

VOLUME I: RESEARCH COMPONENT

BY

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Overview

Volume I

Volume I comprises a review of the literature about children and young people with Coeliac Disease, a common autoimmune disease characterised by an immune response to the protein gluten, found in wheat, barley and rye. It also comprises a qualitative research paper detailing interviews with young people that aimed to tap their lived experiences of the disease. The final part of Volume I comprise a public domain briefing paper summarising both the literature review and research paper.

The literature review considers the evidence for problems with adherence to treatment (a lifelong diet free from gluten) for young people with Coeliac Disease as well as psychological effects of having the disease in childhood. Studies of parental views were also included. The results suggested that there is some element of psychological distress associated with having CD in childhood, and that adherence to the treatment appears to be influenced by age and gender. Studies of parents' views about their child's Coeliac Disease suggested that parents' belief in their ability to manage the disease is important to how young children adhere to treatment.

The research project describes how 5 children and young people were recruited and interviewed about their experiences of living with Coeliac Disease. The resulting data were analysed using Interpretative Phenomenological Analysis and two themes were subsequently identified. These themes related to identity as a young person with Coeliac Disease and to perceptions about food.

Volume II

Volume II comprises five Clinical Practice Reports:

Clinical Practice Report 1 describes the “challenging behaviour” of a 42 year old man with learning disabilities. Subsequently, formulations of this behaviour from a behavioural and a systemic perspective are presented.

Clinical Practice Report 2 is a service evaluation conducted in the Heart of Birmingham that aimed to measure outcomes for adults with a learning disability, as well as considering outcomes separately for different ethnic groups.

Clinical Practice Report 3 is a case study of a 16 year old girl with anger difficulties using CBT within the Solihull Approach model.

Clinical Practice Report 4 describes cognitive-behavioural intervention with a 39-year old man suffering with anxiety as a result of residual psychotic symptoms.

Clinical Practice Report 5 was an oral presentation of a piece of clinical work completed with staff at a day hospital for older adults, conducted in order to help the staff adjust in their move to a new location.

Acknowledgements

I would like to thank all the children and young people, and their parents, who took part in my research. Their time and interest in the project was fantastic, and I wouldn't have been able to complete this thesis without them. I would also like to thank the two local Coeliac support groups for their enthusiasm and help recruiting the children, young people, and their parents to the study. My thanks also to Dr Goldstein, paediatrician at Birmingham Children's Hospital, and Ros Blackmore, dietician, for their help in the early stages of the project. I also appreciate the time and support of both research supervisors, Dr Ruth Howard and Dr Gary Law, for putting up with my questions and tantrums, and working at weekends to go through drafts! Finally, I ought to mention my fiancé, and to thank him for supporting me throughout the whole course, and especially during these last few months – I promise I won't do another degree!

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Children, young people and Coeliac Disease:

A review of the literature

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Abstract

Background: Coeliac Disease (CD) is a common autoimmune disease characterised by an immune response to the protein gluten, found in wheat, barley and rye. Currently, it can only be treated through a life-long gluten-free diet. Much literature exists in relation to adults with the disease, but little with regard to the experiences of children and young people.

Methods: Databases were systematically reviewed using search terms specific to children, young people, and their experiences of CD. All papers were assigned quality ratings in three areas: rationale and design, methods, and results.

Results: Overall, 24 papers were included in the review and fell into three categories: treatment adherence, psychological consequences of CD, and parental views. Methodological quality varied greatly across studies, and factors purportedly relating to adherence were contradictory, as were outcomes about psychological consequences. Reports about parents' attitudes were more consistent.

Conclusions: Some factors were consistently associated with adherence; being younger and female. Further, some studies reported equivalent quality of life between children with CD and those without, but the majority of studies found some element of psychological distress associated with CD. Finally, the importance of parents in helping young people to adapt to the GFD was

reported. Three main themes emerged from the data: the importance of self-efficacy, developmental stage of young person, and illness representations. However, methodological quality tended to be low, and further studies need to be completed to explore childhood reactions to having CD.

Introduction

What is Coeliac Disease?

Contrary to popular belief, Coeliac Disease (CD) is not an allergy, but a chronic autoimmune disease with an estimated UK prevalence of 1:87 (West et al., 2003). The immune system of people with CD produces antibodies that target their own body triggered by eating gluten (a protein found in wheat, barley and rye; Coeliac UK, 2007). The classic triad of symptoms found in children with CD who are consuming gluten (whether undiagnosed or failing to adhere to treatment) is failure to thrive, malabsorption and persistent diarrhoea. Other symptoms include depression, abdominal pain, anaemia, arthritis, delayed puberty, problems with dental enamel, problems with bone mineral density, and fatigue (Mearin, 2007). CD is diagnosed through biopsy of the small intestine which should show mucosal abnormality. Tests for specific antibodies, although not essential, will add weight to the diagnosis and it is expected that there should be total remission of all clinical symptoms upon commencing treatment (Walker-Smith, Guandalini, Schmitz, Shmerling, & Visakorpi, 1990). Recommendations from the British Society of Gastroenterologists (2002) suggest that a further biopsy should be carried out approximately 4 to 6 months after starting treatment in order that repair of the small intestine be observed. Although not essential, it is strongly recommended for those children where (i) diagnosis is doubted, (ii) treatment was started without an initial biopsy, or (iii) diagnosis was made before the age of 2 years.

Currently, there is no cure for CD and management is achieved through adherence to a gluten-free diet *for life* (GFD; Hill, Dirks, Liptak, Colletti & Fasano, 2005). This includes a diet free from items such as bread, pasta and pastries, as well as less obvious foods such as some fruit squashes (which contain barley), some sweets, and table sauces. Adherence to such a GFD can be compromised by a range of difficulties, including contamination of naturally gluten free foods with wheat flour, inadequate labelling of food items, and enduring gluten remaining in gluten free wheat starch used in some gluten free products (Mearin, 2007). The gluten content of food items is constantly changing, and therefore people who manage CD need to keep up-to-date with such changes (Mearin, 2007). People with CD are at a greater risk of reduced bone mass and osteoporosis, and there is an increased risk of tumours such as gut lymphoma if a GFD is not followed; the risk of the latter is increased by 25 to 120 fold in children who do not follow a GFD (Mihailidi, Paspalaki, Katakis, & Evangeliou, 2003). The risk of cancer decreases to the same as a person without CD after a GFD has been followed for 3 to 5 years (Coeliac UK, 2007).

In addition to possible adverse short- and long-term physical consequences to having CD, there are also potential psychological and social implications of managing such a chronic illness (Mearin, 2007). Managing a GFD provides a challenge to the children with CD, their families, schools, and health professionals, and this is particularly accentuated by the Western diet which is particularly heavy on foods that contain products containing wheat, rye and barley (Mearin, 2007).

Hawkes and colleagues (2000) state that CD is diagnosed in less than 1 in 2500 children in the UK. However, Bingley et al. (2004) investigated the prevalence of undiagnosed CD in 5470 children aged 7 in the UK using antibody markers for CD; prevalence was approximately 1%. Importantly, some authors posit that published prevalence figures are usually an underestimate, due to both difficulty diagnosing the disease and misdiagnosis (Hill et al., 2005). The chance of developing CD if an immediate family member has it is increased, with a prevalence of 1 in 10 (Coeliac UK, 2007). Despite this prevalence, there is a paucity of psychosocial research concerning CD.

Why focus on children and adolescents?

Research on chronic illness and childhood is more complex to conduct than for adults because there are issues of developmental stage to be taken into account (Eiser, 1990; Schmidt, Petersen & Bullinger, 2003). This not withstanding, there is evidence to suggest that children with chronic physical disorders, as well as their mothers, are more likely to have difficulties in psychosocial adjustment when compared to healthy peers (Wallander & Varni, 1998). Further, Hysing and colleagues (2009) cite various studies that confirm a range of what they term emotional and behavioural difficulties in children diagnosed with chronic illness, extending from restrictions in leisure activities to physical pain and family difficulties.

Upon diagnosis of CD, it is likely that parents begin to develop a sense of control about understanding what their child needs and how they fit in with this (Huff, 1997), and it becomes clear here that diagnosis in childhood can have

wider family and social implications than those diagnosed in adulthood (Olsson, Hornell, Ivarsson & Sydner, 2008). Younger children can be somewhat controlled in gluten ingestion by parents who take responsibility for the dietary regimen, whereas responsibility for self-managing the GFD is an issue that families will need to negotiate as their child enters adolescence and seeks to become more independent in managing their diet (Greco, Mayer, Ciccarelli, Troncone & Auricchio, 1997). Adolescence provides children with challenges, and their reactions to such stressors while they are growing up is likely to be influenced by the presence of a chronic illness. Of course, the illness itself may bring challenges of its own, which may include checkups and other procedures or therapies (Koopman, Baars & Mearin, 2003). In particular, childhood CD can affect how and what the whole family eat and this can make the family more vulnerable to life events because managing CD on a daily basis requires much thought, effort and financial output (Huff, 1997). Further, adolescence is recognised as a time when adherence with GFD may falter because young people are keen to bond with peers and to distance themselves from authority figures, including doctors (Booth, 1991). As such, they may eat gluten containing products to avoid the difficulties of others not understanding the importance of the diet, and to 'fit in' (Olsson, Hornell, Ivarsson, & Sydner, 2008).

Increasingly, there is a focus on health related quality of life for children (HRQOL) in which there is an attempt to standardise assessments of children's wellbeing in relation to chronic illness management (Grootenhuis, Koopman, Verrips, Vogels, & Last, 2007; Varni, Seid, Kurtin, 1999). Authors such as Koopman et al. (2003) acknowledge that there are both negative physical and

social effects of having a chronic illness, but also psychological effects including: uncertainty about the future, feeling scared, depression and loneliness. There may be other issues, such as pain management and possible trauma from medical procedures. Indeed, "children with any kind of chronic disease have twice as many psychosocial problems as children not affected by chronic disorders" (p. 291; Koopman et al., 2003). If children and families are helped to adapt, they are more likely to develop the self-esteem, sense of control and resilience needed to confront the typical stressors that adolescence brings (Huff, 1997). As such, measurement of HRQOL may help to establish the impact of chronic disease and its management on the child (Connolly & Johnson, 1999). However, this is not to assume that every child is adversely affected by chronic illness; indeed, there are reports about "benefit-finding", whereby positive outcomes have developed as a result of chronic illness diagnosis (e.g. Eiser, 1990).

Given the potential impact a chronic illness can have on the lives of children, young people and their families, together with dietary self-management as the cornerstone of treatment in CD, the aims of this literature review are to:

- collate papers on childhood CD and adherence to the GFD, psychosocial consequences of managing CD, and parents' views on CD and its management for their offspring.
- review and evaluate findings to establish what is currently known about childhood CD and its management and psychosocial consequences of CD.

- propose recommendations for clinical practice and future research.

Method

Search strategy

Literature was identified using two databases that comprise Web of Knowledge: Web of Science with Conference Proceedings (1900-2009) and MEDLINE (1950-2009), as well as PsycINFO (1967-2009), PsycARTICLES and EMBASE (1980-2009). Search criteria consisted of three concepts: Coeliac Disease; target population; and terms to tap psychosocial knowledge (Appendix 1). Reference sections of each retrieved paper were searched by hand for further relevant papers.

Inclusion and exclusion criteria

In order for studies to be considered for inclusion they had to meet certain criteria. Inclusion and exclusion criteria are shown in Table 1.

Table 1.

Inclusion and exclusion criteria.

Inclusion criteria	Exclusion criteria
Report an aspect of childhood CD. Include a sample of children or young people aged 0 to 25 (to reflect different education systems in different countries) OR a sample of parents reflecting on management of their child's CD.	Papers published prior to 1984. Not published in English. Reported prevalence rates of CD only. Reported links between learning disability/autism and CD only. Reported links between other physical illness (e.g. diabetes) and CD only. Reported functional outcome or screening exercises only. Not an original article (i.e. review, editorials, letters, conference abstracts).

Data extraction

In order to ease extraction of information from papers, a data extraction form was used to note important study criteria (Appendix 2).

Description of studies

The initial search yielded 716 papers. Upon removing duplicates, and sifting according to exclusion criteria, 18 papers remained (Figure 1). Searching of references yielded a further 6 papers, leaving a total of 24 for review. Of these 24, ranging in date from 1985 to 2009, papers were grouped into one of three themes: GFD adherence (10 studies), psychological aspects of CD (12 studies), and parents' views of children's CD (2 studies). The main features of each study are summarised in Table 2.

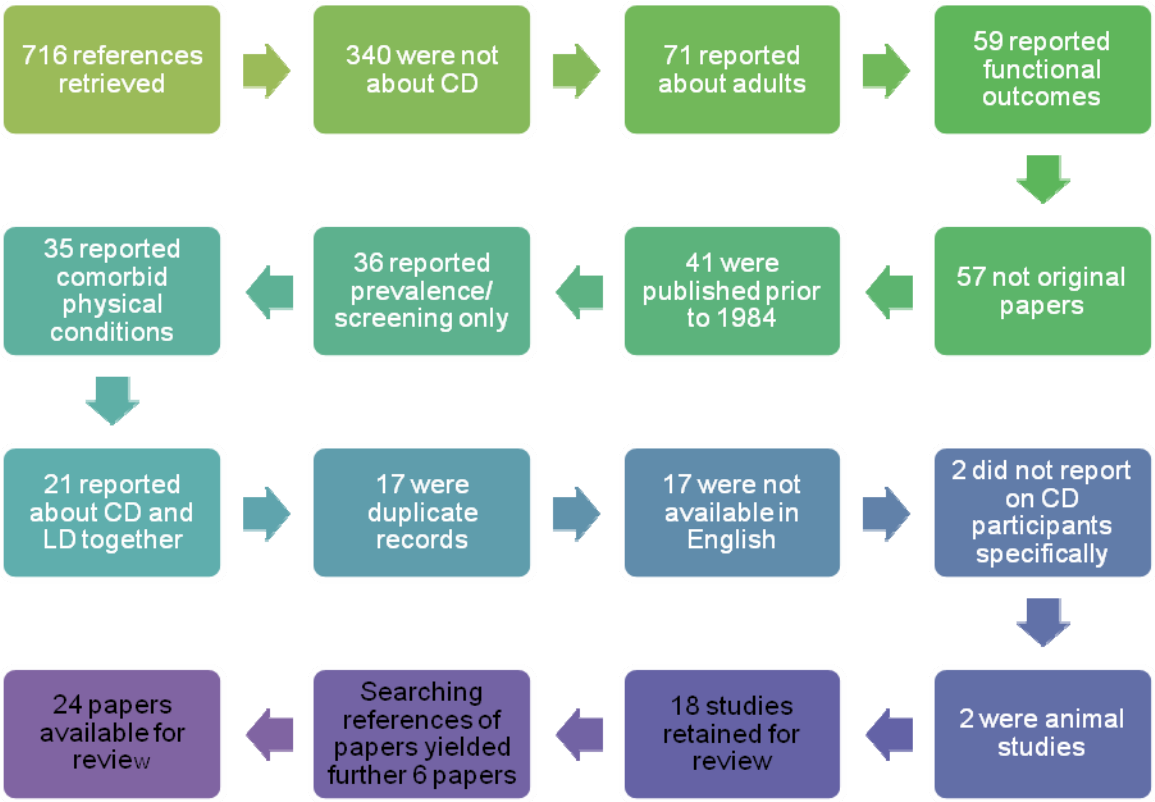

Figure 1. Flowchart of paper sift.

Table 2.

Features of each study in review.

Study *Origin*	Setting	Participant information		Timing of study in relation to diagnosis	*Type of design* Comparison group?	Outcome measures	Results
		N (% uptake) Age Gender	Inclusion criteria				
Studies reporting adherence/follow up of children diagnosed with CD							
Fabiani, Catassi, Villari, Gismondi, Pierdomenico, Ratsch, Coppa & Giorgi (1996) *Italy*	Screening detected students	23 (82.1%) Not stated 17 female, 11 male [a]	1.+ test for antigliadin antibodies aged 11-14 years when screened 1992-94	Follow up average 23 months post-diagnosis (S.D 7)	*Cross-sectional* Not reported	Clinical and anthropometric assessment. Routine blood tests for CD markers Dietary questionnaire to ascertain knowledge of disease and GFD adherence	1. Twelve children stated they were on a strict GFD (52.2%). 2. Eleven reported that they sometimes ate gluten (47.8%). 3. Weekends and friends were main indicators non-adherence. 4. Acceptance of the diet: 6 people stated it was good, 11 moderate and 6 low. 5. The majority of young people (20 of 23) reported some form of physical improvement on GFD.
Fabiani, Taccari, Ratsch, Di Guiseppe, Valentino	Screening detected students (Group A) and previous	Group A = 22 diagnosed by mass screening (81.5%)	1.Diagnosed with ESPGHAN guidelines	Not reported	*Cross-sectional with control group* Group B = 22	Clinical and anthropometric assessment Dietary interview	1. Anxiety and depression scores equitable across the two groups, regardless of sex or dietary adherence 2. Children diagnosed by mass

Coppa & Catassi (2000) *Italy*	paediatric patients (Group B)	Group A = 17.9 years (S.D 1.3), Group B = 16.1 years (S.D 3.6) 13 female and 9 male in both groups			previous paediatric patients diagnosed in childhood (91.7% response rate). Age matched, symptoms same at time of study, and no significant difference in education/social values	Institute for Personality and Ability Testing Anxiety Questionnaire, and Clinical Depression Questionnaire Routine blood tests for CD markers	screening less compliant (5/22 compliant; 22.7%) with the GFD compared to those diagnosed in childhood (15/22 compliant, 68% i.e. never ate gluten, or ate gluten ≤ once a month).
Greco, Mayer, Ciccarelli, Troncone & Auricchio (1997) *Italy*	Clinic	306 (100%) Mean 15.9 years; range 10-27, 92 people = 10 -13 years; 170 people = 13 -18 years; 44 people = 19 – 27 years 186 female, 120 male	1. Consecutively diagnosed and recruited 2. Diagnosed by biopsy (n = 284) or diagnosed on basis of clinical findings and *improvement on a GFD (n = 22)*	Not reported	*Cross-sectional* Not reported	Clinical and anthropometric assessment One day diary of diet A retrospective one month questionnaire to assess adherence	1. 223 people strictly adhered to the GFD (73%). 2. 46 people ate gluten 2 or 3 times a month (15%). 3. 37 people were either on a gluten containing diet or frequently ate gluten (12%). 4. Four factors related to better adherence: Being female, younger, having good school grades, having high self-esteem.
Hopman, le Cessie, von Blomberg & Mearin (2006)	Dutch Coeliac Society	N = 132 (33.4%; most analyses; 21 did not complete	1. Members aged 12-25 years 2. Diagnosed through	Mean 9.6 years (S.D 6) on diet	*Cross-sectional* Not reported	Clinical and anthropometric assessment Food diary, 2	1. Strict adherence in 75% of participants. 2. Occasional ingestion of gluten was reported by 23%. 3. Two participants on a full gluten

Netherlands		nutrition data, 26 no blood test: N = 85, 21.5%) Mean 16.6 years (S.D 4.4) 87 female (66%), 45 male (34%)	biopsy (ranged from 0.3-23.6 years, median = 4.3 years) 3. Understood Dutch			week days and 1 weekend day Questionnaire (34-item questionnaire developed from focus groups with 10 people with CD in the targeted age range) Blood tests	containing diet. 4. Sweets (53%), chocolates or crisps (47%), and fast food (31%) main gluten-containing products eaten, and significantly more by older participants. 5. Special occasions (60%) or at home (49%) most often places/times when gluten-containing food consumed.
Jadresin, Misak, Kolacek, Sonicki & Zizic (2008) *Croatia*	Paediatric patients	71 (66.4%) Mean 12 (S.D 5), range 5 – 30 years 37 female, 34 male	1. Diagnosed children's hospital 1972 – 1994 2. Diagnosed with either ESPGHAN criteria, revised criteria, or two biopsies	Mean 9 years post diagnosis	*Cross-sectional* Not reported	Clinical and anthropometric assessment Blood tests Adherence (strict, eating gluten less than once a week, gluten more than once a week) Attitude towards GFD (burden or not)	1. Using questionnaire, 42 (59.1%) on strict GFD, 19 (26.8%) ingesting small amounts of gluten, and 10 (14.1%) on gluten-containing diet. 2. Using immune markers, 38/71 compliant (53.5%), as opposed to 42. 3. People on GFD had experienced significantly more biopsies (in line with ESPAGAN criteria) than partial compliers and non-compliers. 4. Participants eating gluten found GFD significantly more difficult to maintain, and significantly more likely not to be followed up regularly.

Kumar, Walker-Smith, Milla, Harris, Colyer & Halliday (1988) *England*	Paediatric clinic	102 (100%) Range 12-20 years 56 female, 46 male	1. Transferred from paediatric to adult clinic 1974 - 1984	Not reported	*Cross-sectional* Not reported	Clinical and anthropometric assessment Blood tests (although some people refused as felt well) Biopsy (only 44 people agreed)	1. 57 (55.9%) said they were on a strict GFD (no gluten; self-rated). 2. 36 were partially strict (several items of gluten per week; self-rated). 3. 9 people were on a full gluten-containing diet, self-rated. 4. Followed up in adult clinic year later, adherence was: 45 (44.1%) on strict GFD, 46 partially strict, and 11 on full gluten-containing diet, self-rated. 5. Those on full gluten containing diet deemed GFD more difficult to adhere to due to practical and social reasons. 6. Children tended to eat gluten-containing food with friends, despite gluten-free food being available
Ljungman & Myrdal (1993) *Sweden*	Only paediatric clinic in county for biopsies	47 (100%) [b] Range 12-17 years Not reported, but worked out to be 35 female, 12 male	1. Born 1973-78 2. ESPGHAN criteria met in childhood 3. Diagnosed with CD for at least 10 years 4. Diagnosed <2 years old	Diagnosed for at least 10 years	*Cross-sectional with control group* Yes; 66 children (10 did not reply; response rate 86.8%) matched for age, sex, school class, geographical location (1 had DM), although not clear where sampled from	Questionnaires synthesised; questions about health and self-esteem (used in other studies but not properly validated); Knowledge of CD and adherence with the GFD (only to CD children)	1. No differences between children with CD and those without on: Perception of health, How people felt physically, Self-esteem, Fitness, Attitude to school, School meals, Sports, Classmates, Socialising with friends. 2. 81% (n = 38) deemed compliant, in that they adhered to a strict GFD and if they ate gluten, this was in error and less than once a month. 3. 11% (n = 5) had gluten more than once a month, 2% more than once a week (n = 1).

							4. 6% were on a gluten-containing diet (n = 3). 5. Girls and younger children (12-14 years) more compliant, and more knowledgeable suggested more compliant. 6. Children more likely to be compliant at home, followed by school, followed by when out with friends.
Mariani, Viti, Montouri, La Vecchia, Cipolletta, Calvani & Bonamico (1998) *Italy*	Outpatients and students	47 (100%) Mean 15.2 years (S.D 2.3) 37 female, 10 male	1. Diagnosed with ESPGHAN guidelines 2. Middle/high school students in Rome area	Not reported	*Cross-sectional with control group* Yes; 47 (13 males, 34 females, average age 15.7 years, S.D 2.3) healthy age-matched control subjects	Diary of their diet for 3 days (2 weekdays and a Sunday) Blood samples (CD group only)	1. 30 participants with CD (64%) reported strict adherence to a GFD. 2. 14 (30%) ate gluten once or twice a week. 3. 3 (6%) admitted to a full gluten-containing diet. 4. However, 5 (16%) participants stating they were on strict GFD had blood tests suggestive of gluten ingestion; adherence was 25 of 47 (53%). **5.** Being overweight and obese more common in group who followed strict GFD (72%) compared to those not on such a strict diet (51%) and the control participants (47%).
Mayer, Greco, Troncone, Auricchio & Marsh (1991) *Italy*	Outpatients	123 (100%) Mean 13.7 years, range 10.6-23	1. Diagnosed ≤ 3 years old 2. Seen at least annually for review	Diagnosed for mean 11.5 years, range 9-16 years	*Cross-sectional* Not reported	Clinical and anthropometric assessment Biopsy (36 participants	1. 65% (n = 80) were on a GFD, 11.4% (n = 14) were following the GFD with occasional gluten ingestion and 23.6% (n = 29) were on a full gluten-containing diet.

		52 male, 71 female				only) Questionnaire to assess adherence completed by dietician	2. Lapses mainly occurred in order to avoid difficulties in social situations. 3. Bread and cakes were the gluten containing foods most often consumed 4. Those diagnosed using official guidelines and thus most likely subject to follow up more likely to be following strict GFD (only 5 people diagnosed by ESPAGAN guidelines were on gluten containing diet).
Rashid, Cranney, Zarkadas, Graham, Switzer, Case, Molloy, Warren, Burrows & Butzner (2005) *Canada*	Canadian Celiac Association	168 (100%) Mean 9.1 years, range 2-15 years; 19 aged 2-3, 46 aged 4-7, 43 aged 8-11, 60 aged 12-15 97 female, 71 male	1. Member Canadian Celiac Association 2. Biopsy confirmed CD 3. Under 16 years old	Not reported	*Cross-sectional* Not reported	76 item-questionnaire developed by researchers: demographics, symptoms pre-diagnosis, diagnoses given prior to CD, with CD linked disorders, family history of CD, and questions to tap wellbeing and lifestyle choices of children	1. Most participants (95%) reported strict adherence with diet, and 89% had seen an improvement in their health. *2.* Two things that would improve their QOL: *Better labelling 63%, Better availability of foods in shops 49%, Better GF choices on menus 49%, Earlier diagnosis 34%, Better dietary advice 7%* 3. Percentage of children that responded sometimes, most of time, all of time: *feeling left out of school activities, or activities at friend's houses (61%), feeling different from others due to CD (69%), embarrassment to bring GF foods to parties (53%), feel angry about GFD (72%), thought teachers/friends did not understand (53%), being healthy*

							without following GFD (26%)
Studies reporting psychosocial characteristics							
Calsbeek, Rijken, Bekkers, Kerssens, Dekker, van Berge Henegouwen & participating centres (2002) *Netherlands*	Outpatients from 25 specialists in 11 medical centres AND patient organisation [c]	124 (not reported, but between 48.7% and 67.2%) Mean 17.5 years (S.D 3.7), range 12-25 years 80 female, 44 male	1. Certified medical specialist given diagnosis 2. Duration CD minimum 6 months 3. Aged 12-25 4. Not being hospitalised 5. Aware of diagnosis 6. No terminal illness 7. Intellectually capable 8. Fluent in Dutch 9. If recruited from patient organisation, diagnosis by at least one small bowel biopsy	Not reported; minimum 6 months	*Cross-sectional with control group* Yes; 306 people aged 12-25 (response rate 54.7%), population based control group, randomly recruited from 173 GP practices (patient files used); same criteria used as for study participants (except 1 and 9 not needed)	Questionnaire considering 24 aspects of social position sent by post: categories were education, leisure, friendship, "labour participation", finances, partnership, and sexuality.	1. Children and young people with CD do not appear to have impaired social position when compared to healthy controls; no significant differences between children and young people with CD and control group. 2. Although not significant, children and young people with CD have more time off school, fewer have a job, more need re-education to get a job, when compared to control group. 3. Those with chronic liver disease and inflammatory bowel disease are more likely to be impaired in social functioning than young people with CD.
Calsbeek, Rijken, Bekkers, Dekker, & van Berge Henegouwen	Outpatients from 25 specialists in 11 medical centres AND patient	124 (not reported, but between 48.7% and 67.2%)	1. Certified medical specialist given diagnosis 2. Duration CD	Not reported; minimum 6 months	*Cross-sectional with control group* Yes; 306 people aged 12-25	Questionnaire considering 24 aspects of social position sent by post: categories were education,	1. Children and young people with CD hospitalised significantly more often than control group (after controlling for age, gender, and economic status). 2. Children and young people with

(2006a) *Netherlands*	organisation [c]	Mean 17.5 years (S.D 3.7), range 12-25 years 80 female, 44 male	minimum 6 months 3. Aged 12-25 4. Not being hospitalised 5. Aware of diagnosis 6. No terminal illness 7. Intellectually capable 8. Fluent in Dutch 9. If recruited from patient organisation, diagnosis by at least one small bowel biopsy		(response rate 54.7%), population based control group, randomly recruited from 173 GP practices (patient files used); same criteria used as for study participants (except 1 and 9 not needed)	leisure, friendship, "labour participation", finances, partnership, and sexuality. Burden of disease assessed through questions: physical complaints, anxiety and depression (measured by HADS), "disability in endurance" (adapted from TACQOL), hospitalisation, medication, needing to adhere to diet, toilet use.	CD reporting greater burden associated with dietary adherence compared to control group (adherence "necessary"; after controlling for age, gender, and economic status). 3. Chronic liver disease, inflammatory bowel disease and food allergy sufferers reported a greater amount of burden associated with their illness than children and young people with CD.
Calsbeek, Rijken, Bekkers, van Berge Henegouwen & Dekker (2006b) *Netherlands*	Outpatients from 18 specialists in 9 medical centres AND patient organisation	61 (not reported, but between 38.6% and 58.8%) Mean 18.4 (S.D 3.7) years, range 12-25 years;	1. Certified medical specialist given diagnosis 2. Duration CD minimum 6 months 3. Aged 12-25 4. Not being	Not reported; minimum 6 months	*Cross-sectional with control group* Yes; 274 people aged 12-25 (response rate 49%), population based control	Burden of disease assessed through questions: physical complaints, anxiety and depression (measured by HADS), "disability	1. Children and young people use task-oriented, emotion oriented and avoidance as coping strategies to the same extent as those with chronic liver disease, inflammatory bowel disease, food allergy or congenital disorders, as well as a control group of healthy adolescents and young

		11 aged 12-14 years, 13 aged 15-17 years, 14 aged 18-20 years, 23 aged 21-24 years 41 female, 20 male	hospitalised 5. Aware of diagnosis 6. No terminal illness 7. Intellectually capable 8. Fluent in Dutch 9. If recruited from patient organisation, diagnosis by at least one small bowel biopsy		group, randomly recruited from 173 GP practices (patient files used); same criteria used as for study participants (except 1 and 9 not needed)	in endurance" (adapted from TACQOL), hospitalisation, medication, needing to adhere to diet, toilet use. Coping assessed by CISS-21 School/leisure activities assessed by questions: school absences, going out, educational level, friends and engaging in cultural activities	people.
Cinquetti, Trabucchi, Menegazzi, Comucci, Bressan & Zoppi (1999) *Italy*	Paediatric department	39 (75%) Mean 15.5 years, range 10-21 years 24 female, 15 male	1. At paediatric clinic 2. On GFD	Average time on GFD 13 years	*Cross-sectional* Not reported	25 item questionnaire relating to psychological implications of CD and requiring a GFD administered with psychologist in the room. Questionnaire of demographics and illness specific questions (duration of	1. Adolescents aged 10-12 didn't appear to notice any difference with friends, those aged 13-17 were "uneasy", and in older adolescents, CD was generally accepted. 2. 23 people (59%) said they may give into temptation (more likely to be older), 13 of which admitted feeling guilty afterwards. Eight endorsed concerns about physical health only, 6 said in relation to health and conscience. 3. In terms of relationship to food, a

						illness, for example), plus questions evaluating adolescents attitudes towards food, mood, and relationships with other people.	sense of hatred for 2 people, resentment for 5, dislike for 19, and the remaining 13 had relaxed attitude. 4. Desserts most commonly missed on the GFD, followed by bread and pasta.
Grootenhuis, Koopman, Verrips, Vogels & Last (2007) *Netherlands*	Dutch Celiac Patients Society (and their parents) [d]	104 (99%) Range 8 – 11 years 60 female, 45 male	1. Can read and understand Dutch 2. Aged 8 -11	Not reported	*Cross-sectional with control group* Yes; 913 healthy children aged between 8 and 11 years old.	Completed TNO-AZL/TACQOL questionnaire Considered prevalence of children at risk for HRQOL problems using 25th percentile from normal population as cut-off	1. Compared to healthy children, children with CD had significantly lower HRQOL score for social functioning. 2. In terms of prevalence of children at risk for HRQOL problems, children with CD at risk for problems in areas of motor functioning (35% at risk), cognitive functioning (35% at risk), and social functioning (41% at risk).
Karwautz, Wagner, Berger, Sinnreich, Grylli & Huber (2008) *Austria*	Outpatients and Austrian and German Coeliac Societies	283 (83%) first wave; 168 (59%; due to drop out of non-eating pathology participants) second wave Mean female 14.8 years	**1.** BMI <10th percentile & EDE-Q Restraint, Shape, or Weight Concern scale score ≥2 **OR** 2. Current bingeing vomiting,	Not reported	*Cross-sectional with control groups and longitudinal* Two control groups: First was group of adolescents with DM Type 1; second was 1080 girls, mean	Eating disorders questionnaires, Eating Disorder Inventory (EDI-2) and Eating Disorder Examination Questionnaire (EDE-Q) Clinical and anthropometric	3. 11 (4.8%) lifetime prevalence of ED; 8 (3.9%) current ED; 21 (10.2%) had lifetime history of subclinical ED, with 22 (10.7%) diagnosed with current subclinical ED, more frequent than when compared to national estimates from the USA and Europe. 4. CD diagnosis typically preceded ED diagnosis (in 85.7% of cases) between 2-17 years (mean 9.87

		(S.D 3), mean males 13.9 years (S.D 2.7), range 10-20 years 210 female, 73 male	laxative use, eating gluten foods to affect shape/weight **OR** 4. EDI-2 scores ≥ 9 for Drive for Thinness, ≥5 Bulimia, ≥15 Body dissatisfaction		age 17.7 years (S.D 2.2) and 580 boys, mean age 13.7 years (S.D 2.0) obtained from school sample.	assessment Laboratory tests for CD markers For those who were randomly selected for second stage, Eating Disorder Examination (EDE) completed to assess current and lifetime prevalence for eating disorders (ED)	years, S.D 4.7 years). 5. Those with eating pathology more often non-compliant with GFD (i.e. ate gluten ≥ twice/month). 6. Overall, female participants with CD had lower eating pathology compared to normal school controls. Lower eating pathology when compared to people with DM Type 1 and comorbid eating pathology also.
Kolsteren, Koopman, Schalekamp & Mearin (2001) *Netherlands*	Dutch Celiac Patients Society (and their parents) [d]	92 (29.0%) Range 8-16 years 8-11 year olds; 51 female, 41 male	1. Diagnosed by ESPGHAN guidelines 2. Currently "treated" CD (GFD prescribed)	Not reported	*Cross-sectional with control group* Yes; 41 12-16 year olds; 28 female, 13 male; 1183 (and 1805 parents) children aged 8-12 years (not matched) from a study previously used to validate the TACQOL/DUCATQOL measures	Generic instruments of HRQOL: TACQOL, DUCATQOL. Questions specific to GFD: management over last few weeks: any problems with GFD, disliking GF food, problems met as not allowed sweets with gluten, problems	1. No significant differences in QOL on TACQOL between adolescents in the reference sample and sample of CD patients. 2. In 8-11 year olds, complaints and positive emotions significantly greater than for reference sample. 3. Adolescent girls reported significantly greater positive complaints than boys. 4. No presentation of parents' responses, but parents reported adolescent sons to experience more positive emotions than adolescent daughters.

						due to other children eating things they couldn't, problems because have to eat something different, problems missing sweet things everyone else eats.	
Olsson, Hornell, Ivarsson & Sydner (2008) *Sweden*	Prospective incidence register (national from 1998) and paediatric departments	47 (29.6%) Range 15 – 18 years 32 female, 15 male	1. On GFD for at least 1 year 2. Diagnosis confirmed by ESPGHAN guidelines 3. Aged 15-18 years	Not reported	*Qualitative* Not reported	10 focus groups lasting 60-80 minutes analysed using Grounded Theory. Asked about experiences of living with CD/GFD in different contexts, beliefs, knowledge, attitudes, expectations, perceptions and needs and experiences relating to CD and managing the GFD, and support/attitudes of others.	1. Non-adherence with GFD compromised by: a. Significant others having poor knowledge of CD/GFD b. Eating outside the home c. Difficulties getting hold of the food, and when they do, the palatability of such food d. Lack of support socially e. How they perceive themselves if they ingest gluten

Pynnonen, Isometsa, Aronen, Verkasalo, Savilahti & Aalberg (2004) *Finland*	Hospital	29 (78.4%) Mean 14.2 years (S.D 1.9), range 12-17 years 16 female, 13 male [e]	1. Biopsy conducted between 1982-1999 indicated CD	Not reported, but ~ 6 years	*Longitudinal* Yes; 29 adolescents aged 12-18 years (average 14.4, ±1.9) symptoms suggestive of CD, but not having CD. People with IBS excluded. Matched for age at biopsy (± 6 months if < 12 years), gender, age at study (± 1.2 years).	K-SADS-PL: psychiatrists not blind to diagnosis. YSR CBCL BDI and BAI HDRS and HARS Reports from case notes to assess psychiatric symptoms and symptoms before biopsy, and for CD group, after GFD started.	1. Lifetime prevalence of depression significantly greater in adolescents with CD than without (31% v. 7%), as was "double depression" (dysthymic disorder and major depressive disorder; 21% v. 0%) and "disruptive behaviour disorders" (28% v. 3%). 2. Pre-biopsy, people with CD significantly more likely to have any depressive disorder than control group (37% v. 0%), particularly major depressive disorder (26% v. 0%). 3. CD and parental depressive disorder both predict pre-biopsy and lifetime depressive disorder (as does the interaction between them). 4. Parental depressive disorders more common in comparison group, so there is something about parental depressive disorder and CD together that gives rise to greater risk for lifetime prevalence.
van Doorn, Winkler, Zwinderman, Mearin & Koopman (2008) *Netherlands*	Dutch Celiac Patients Society	510 (67.5%) [†] Overall mean 11.3 years, S.D 2.7, range 8 – 18 years, 278 aged 8 –	1. Diagnosis by at least 1 small bowel biopsy 2. No comorbid chronic illness 3. No	Mean age at diagnosis 3.7 years (S.D 3.5), therefore, mean time since	*Cross-sectional with control group* Yes; 991 healthy children aged 8 – 15 years, and 71	Focus groups to assess impact effect of CD on everyday lives of children (Phase 1). Selection of 24	1. Questionnaire able to discriminate between severity of CD as reported by parents. 2. Low to neutral QOL reported on CDDUX by children with CD. 3. Parents scored children as significantly lower than children on QOL.

		11 years (mean 9.1, S.D 0.9); 184 aged 12 – 15 years (mean 13.1, S.D 0.9); 48 aged 16 – 18 years (mean 16.7, S.D 0.7) 8-11 years: 167 female, 111 male; 12-15 years: 116 female, 68 male; 16-18 years: 31 female, 17 male; Overall, 314 female, 196 male	intellectual impairment	diagnosis worked out to be 7.6 years.	children with asthma and 29 children with diabetes (both aged 8 – 15 years) no further information given)	items from these to generate disease specific questionnaire to be tested. This questionnaire (CDDUX) and generic DUX-25 used in remainder of study (Phase 2). Parents assessed same dimensions on a parent version of questionnaire, as well as questions about general health.	4. Those children who had an enhanced sense of their health had a higher score on the new HRQOL measure (the CDDUX). 5. Lower QOL on generic measure (DUX-25) when compared to healthy children without CD. 6. Using generic measure of QOL (DUX-25), children with CD perceived themselves as having a poorer quality of life when compared to healthy comparison group, apart from in emotional domain in 8 – 11 year olds and 12 – 15 year olds. 7. Children with CD perceived selves as having higher quality of life when compared to children with diabetes and asthma. 8. Those aged 16 – 18 years scored their QOL as lower than younger age groups on DUX-25.
van Koppen et al (2009) *Netherlands*	Outpatients	32 (100%); follow up 22 (69%) Range 12-14 years Not reported	1. Aged 2-4 yrs at diagnosis 2. Diagnosed via mass screening (19 GFD, 13 gluten-containing diet; 14 randomly allocated to	10 years	*Longitudinal comparison group (prospective)* Sample of 251 children aged 1-5 years from general Dutch population	Clinical and anthropometric assessment Serum antibodies for CD HRQOL assessed using TAPQOL at CD diagnosis, and	1. 81% (n = 26) on GFD after 10 years; 18 of 19 children who started on GFD, and 8 of 13 who started on gluten containing diet. 2. Children who had symptoms when diagnosed with CD showed improvement in QOL scores after starting GFD. 3. HRQOL for symptom free children diagnosed with CD similar to reference sample pre- and 1 year post GFD.

			groups, 18 not)			after 1 year on GFD HRQOL assessed using CDDUX (CD specific scale), 10 years post diagnosis	4. 10 years post diagnosis similar HRQOL for patients and reference sample. 5. Children with CD and on GFD reported low to neutral HRQOL.
Wagner, Berger, Sinnreich, Grylli, Schober, Huber & Karwautz (2008) *Austria*	Outpatients and Austrian and German Coeliac Societies	283 (83% originally 70.2% from Austria and 29.8% from Germany) Mean female 14.8 years (S.D 3), mean males 13.9 years (S.D 2.7) range 10-20 years 210 female, 73 male	1. Biopsy and antibody proven CD 2. Adherence to GFD at least 1 year 3. Aged 10-20 years 4. No other chronic conditions	At least 1 year	*Cross sectional with control group* 82 adolescents from Austrian schools without chronic illness, matched for age, sex, education, and social status.	Inventory of Life Quality in Children and Adolescents (ILC; Mattejat & Remschmidt, no date) Berner Subjective Wellbeing Inventory (BFW; Grob et al., 1991, as cited in Wagner et al., 2008).	1. 80% adhered strictly to GFD, 14.9% ate gluten 2-3 times/month, 4.3% ate gluten frequently. 2. Noncompliant adolescents significantly older than compliant. 3. Later diagnosis associated with greater school problems and social problems, poorer physical health and higher burden associated with CD. 4. More incidences of eating gluten lower QOL (more physical problems, felt more burdened by disease, more unwell, had more problems during leisure time, more family problems). 5. Equivocal QOL for people compliant with GFD, and those with no chronic illness.

Studies reporting parents' perspectives							
Anson, Weizman & Zeevi (1990) *Israel*	Outpatient from lone medical centre	43 sets of parents (89.6%) Age range refers to participants' children: "Compliant" = mean 11.1 years (S.D 4.4); "Non-compliant" = mean 10.0 years (S.D 3.3) Compliant = 15 female, 16 male; Non-compliant = 8 female, 4 male	1. Jewish 2. Child diagnosed > 1 years old by biopsy and also through response to GFD 3. Child ≤ 18 years at time of study	Followed up mean 6.9 years (± 3.3)	*Cross-sectional with comparison group* Yes; parents of children compliant with GFD (n = 31) v. non-compliant (n = 12), as defined by presence of clinical symptoms of disease, biopsy results, and presence of CD specific antibodies in blood samples	Demographics (completed by nurses blind to condition) Parents' knowledge of disease, GFD, and managing a menu Interviews of attitudes towards health generally, attitudes towards CD, barriers to adherence, dietary behaviour of parent and child Blood tests	1. Found parents of compliant patients were: a. better educated (related specifically to father's occupation) b. from higher social class 2. Parents of compliant children compared to parents of non-compliant children significantly more likely to deem themselves sufficiently informed (80% v. 50%), despite no difference in actual knowledge. 3. Overall, more concerned about future, better adherence. 4. The following barriers significantly more common in those parents of non-compliant children: Perceiving diet as difficult, children insisting on eating gluten containing foods, special meals at home and parties
Jackson, Glasgow & Thom (1985) *Northern Ireland*	Outpatients	50 sets of parents (100%) Age range refers to participants' children: Range 1.5 –	1. Diagnosed by biopsy and marked improvement on GFD	Not reported	*Cross-sectional* Not reported	Questionnaire developed and used at review which aimed to tap knowledge of both CD and GFD, assess attitudes and motivation	1. 30 stated GFD "strictly maintained" (60%), 18 said it was "broken regularly or sometimes" (36%), and 2 (4%) said "diet kept poorly or ignored". 2. Less than strict adherence linked to less knowledge about the disease, linked to more socially disadvantaged.

		19 years (median 9.9 years) 29 female, 21 male				towards CD and the GFD, and evaluate dietary adherence. Demographics Clinical and anthropometric assessment	3. Parents who were members of the Coeliac Society (23 of 50 parents) generally demonstrated a greater understanding of the disease, as well as having children whose dietary adherence was greater. It was also linked to social index, but direction not stated.

Notes. [a] Gender split reported for whole sample prior to drop outs. [b] 2 had Diabetes Mellitus. [c] Same sample. [d] Same sample. [e] All Caucasian. [f] Phase 2 only. Phase 1 in which questionnaire developed not reported here. Abbreviations: CD = Coeliac Disease; DM = Diabetes Mellitus; ESPGHAN = European Society for Paediatric Gastroenterology Hepatology and Nutrition (Walker-Smith et al., 1990); GFD = gluten free diet; HADS = Hospital Anxiety and Depression Scale (Zigmund & Snaith, 1983). TACQOL = TNO AZL Child Quality Of Life Questionnaire (Verrips et al., 1999). CISS-21 = Coping Inventory for Stressful Situations (Endler & Parker, 1999, as cited by Calsbeek et al., 2006b). TAPQOL = TNO-AZL Preschool Children Quality of Life questionnaire (Fekkes et al., 2000). EDI-2 = Eating Disorders Inventory - 2 (German version, Rathner & Waldher, 1997, as cited in Karwautz et al., 2008). EDE-Q = Eating Disorder Examination Questionnaire (German version, Hilbert et al., 2007 as cited in Karwautz et al., 2008). EDE = Eating Disorders Examination Version 12.0D (German version, Hilbert, Tuschen-Caffier, & Ohms, 2004, as cited in Karwautz et al., 2008). DUCATQOL = Dutch-Child-AZL-TNO-Quality-Of-Life (Kolsteren et al., 2001). K-SADS-PL = Schedule for Affective Disorders and Schizophrenia for School-Age Children- Present and Lifetime Version (Kaufman, Birmaher, Brent, Rao & Ryan, 1996). YSR = Youth Self Report (Achenbach, 1991 as cited in Pynnonen et al., 2004). CBCL = Child Behavior Checklist (Achenbach, 1991, as cited in Pynnonen et al., 2004). BDI = Beck Depression Inventory (Beck, Ward, Mendelson, Mock & Erbaugh, 1961). BAI = Beck Anxiety Inventory (Beck, Epstein, Brown & Steer, 1988). HDRS = Hamilton Depression Rating Scale (Hamilton, 1960). HARS = Hamilton Anxiety Rating Scale (Hamilton, 1959). CDDUX = Coeliac Disease DUX (van Doorn et al., 2008). DUX-25 = Self-report Dutch measure of generic quality of life (Koopman et al., 1998, as cited in van Koppen et al., 2009). Where uppermost age-range is greater than 21 years, papers included where average age was less than 18 years; papers did not separate data according to age sufficiently to allow older participants to be excluded in report here.

Participants

Of 24 studies, 23 reported the age range of participants (Fabiani et al., 1996 did not report this). Ages ranged from 1.5 years to 30 years. Sixteen studies reported mean age of participants, and the average age of all participants across studies was 14.5 years. Nine studies explicitly stated time elapsed since diagnosis, two studies reported information to enable this to be worked out (Pynnonen et al., 2004 and van Doorn et al., 2008), and the remaining 13 did not provide this information. Mean time since diagnosis, across the 11 studies reporting these data, was 7.9 years. Thus, using mean age and mean time since diagnosis, average age at diagnosis was 6.5 years old.

Country of origin

Eight studies were conducted in the Netherlands, six in Italy, two in Sweden, two in Austria, and one each from Canada, Croatia, Israel, Northern Ireland, Finland, and England.

Recruitment

The majority of samples were recruited from outpatient clinical settings; 11 studies reported assessment of either previous or current paediatric patients. Five studies sampled from national Coeliac Societies exclusively, five sampled from both outpatient clinics and national Coeliac Societies, two studies sampled from both outpatient clinics and screening detected students, and one study sampled students diagnosed with CD through antibody testing after a mass screening exercise only. The percentage uptake ranged from

29.0 % (Kolsteren et al., 2001) to 100%, the latter of which was found in 8 studies. The majority of studies reported uptake at over 60%.

Inclusion criteria

Seventeen of the 24 studies used diagnosis by childhood biopsy as one of the main inclusion criterion, with 6 of these specifically using ESPGHAN criteria (Walker-Smith et al., 1990). Purposive sampling was used in all studies, although van Koppen et al. (2009) randomly allocated young people to either GFD or gluten containing diet where possible (14 cases). Calsbeek et al. (2002; 2006a; 2006b) randomly generated the control group.

Sample size

Sample size ranged from 22 (Fabiani et al., 2000) to 510 (van Doorn et al., 2008) with the median sample size calculated as 82. It is unclear, but Fabiani et al. (1996) and Fabiani et al. (2000) appear to report the same participants. Karwautz at al. (2008) and Wagner et al. (2008) report the same participants, as do Calsbeek and colleagues in their 2002 and 2006a paper. Their 2006b paper reports a variation on these studies, but with fewer participants. Grootenhuis et al. (2007) used the same experimental sample as Kolsteren et al. (2001).

Study design

The majority of studies were cross-sectional in nature, except three that were longitudinal (Karwautz et al., 2008; Pynnonen et al., 2004; van Koppen et al., 2009), and one that was a qualitative paper. Karwautz et al. (2008)

considered lifetime prevalence of eating disorders, Pynnonen and colleagues (2004) lifetime prevalence of mental health problems, and van Koppen et al. (2009) report quality of life (QOL) at three different times (pre-CD diagnosis, 1 year and 10 years post-diagnosis). Olsson et al. (2008) report the lived experiences of 47 adolescents with CD obtained through focus groups. Ten studies implementing cross-sectional design utilised a control group, and 1 used a comparison group (i.e. a group with CD, not healthy controls).

Methods

There were no standardised questionnaire measures used to assess adherence across studies. Of the 10 studies, 2 used only non-standardised questionnaires, 6 used these and a biopsy or blood test with most emphasis on the former, and a further 2 used both questionnaires and blood tests, giving equal weight to both procedures. Ten of the 12 studies utilising quantitative methods to assess quality of life used some form of standardised questionnaires, with the remaining studies (Cinquetti et al., 1999; Olsson et al., 2008) synthesising measures.

Results

Initially, an assessment of study quality was carried out. This was based upon the protocol by Mitrofan, Paul and Spencer (2008) and adapted for this review, including the introduction of a numerical numbering system to aid data analysis. As such, a score of 0 denotes no available evidence for such a feature, 1 indicates partial evidence, and 2 indicates there was definitely evidence for such a feature in the study. A further adaptation was sorting of the categories to be scored. As the studies included in this review were not typically experimental in nature, or qualitative, some categories were removed to allow for completion of categories for all studies in the review. Findings are illustrated in Table 3.

Table 3.

Quality of studies.

	Study								
Quality criteria	Fabiani et al. (1996)	Fabiani et al. (2000)	Greco et al. (1997)	Hopman et al. (2006)	Jadresin et al. (2008)	Kumar et al. (1988)	Ljungman & Myrdal (1993)	Mariani et al. (1998)	Mayer et al. (1991)
Rationale and design									
Is there a sound rationale for study?	1	1	2	2	2	1	2	1	2
Appropriate design?	1	2	1	1	1	1	1	1	1
Does the method allow for replication?	1	2	1	2	2	1	1	1	2
Is there a control group?	0	2	0	0	0	0	2	2	0
Is the design longitudinal?	0	0	0	0	0	0	0	0	0
Sub-total	**3**	**7**	**4**	**5**	**5**	**3**	**6**	**5**	**5**
Sample and measures									
Response rate satisfactory?	1	1	2	0	1	2	2	2	2
Sampling source appropriate?	2	2	1	1	1	1	1	1	1
Are participants sampled to minimise bias?	2	2	2	2	2	2	2	2	1
Use of valid and reliable paper and pencil measures [a]	0	0	0	0	0	0	0	0	0
Sub-total	**5**	**5**	**5**	**3**	**4**	**5**	**5**	**5**	**4**
Results									
Confounding variables controlled	0	1	1	0	1	0	1	1	1
Generalisable results?	1	1	1	1	1	1	1	1	1
Sub-total	**1**	**2**	**2**	**1**	**2**	**1**	**2**	**2**	**2**
Total score	***9***	***14***	***11***	***9***	***11***	***9***	***13***	***12***	***11***

	Study						
Quality criteria	Rashid et al. (2005)	Cinquetti et al. (1999)	Calsbeek et al. (2002)	Calsbeek et al. (2006a)	Calsbeek et al. (2006b)	Grootenhuis et al. (2007)	Karwautz et al. (2008)
Rationale and design							
Is there a sound rationale for study?	2	2	2	2	2	2	2
Appropriate design?	1	1	1	1	1	1	2
Does the method allow for replication?	2	2	1	1	1	1	1
Is there a control group?	0	0	2	2	2	1	1
Is the design longitudinal?	0	0	0	0	0	0	2
Sub-total	**5**	**5**	**6**	**6**	**6**	**5**	**8**
Sample and measures							
Response rate satisfactory?	2	1	0	0	0	2	1
Sampling source appropriate?	1	1	2	2	2	1	2
Are participants sampled to minimise bias?	2	1	2	2	2	2	2
Use of valid and reliable paper and pencil measures [a]	0	0	1	1	1	1	1
Sub-total	**5**	**3**	**5**	**5**	**5**	**6**	**6**
Results							
Confounding variables controlled	0	1	1	1	1	0	1
Generalisable results?	1	1	1	1	1	1	1
Sub-total	**1**	**2**	**2**	**2**	**2**	**1**	**2**
Total score	***11***	***10***	***13***	***13***	***13***	***12***	***16***

	Study							
Quality criteria	Kolsteren et al. (2001)	Olsson et al. (2008)	Pynnonen et al. (2004)	van Doorn et al. (2008)	van Koppen et al. (2009)	Wagner et al. (2008)	Anson et al. (1990)	Jackson et al. (1985)
Rationale and design								
Is there a sound rationale for study?	2	2	2	2	2	2	2	1
Appropriate design?	1	2	2	2	2	2	2	1
Does the method allow for replication?	1	1	2	1	1	1	2	1
Is there a control group?	1	0	2	1	1	2	1	0
Is the design longitudinal?	0	0	1	0	2	1	0	0
Sub-total	**5**	**5**	**9**	**6**	**8**	**8**	**7**	**3**
Sample and measures								
Response rate satisfactory?	0	0	1	1	1	1	2	2
Sampling source appropriate?	1	1	1	1	2	2	1	1
Are participants sampled to minimise bias?	1	1	2	2	2	2	2	2
Use of valid and reliable paper and pencil measures [a]	1	n/a (1)	1	1	1	1	0	0
Sub-total	**3**	**3**	**5**	**5**	**6**	**6**	**5**	**5**
Results								
Confounding variables controlled	0	1	1	0	1	1	1	0
Generalisable results?	1	1	1	1	1	1	1	1
Sub-total	**1**	**2**	**2**	**1**	**2**	**2**	**2**	**1**
Total score	***9***	***10***	***16***	***12***	***16***	***16***	***14***	***9***

Note. [a] Scored as either present (1) or absent (0).

Adherence

Adherence with the GFD varied from 22.7% (Fabiani et al., 2000) to 95% (Rashid et al., 2005) although, in the former case, only 22 participants comprised the sample. The median estimate of adherence was 64% (Mariani et al., 1998). Fabiani et al. (2000) reported two adherence rates, one for the group diagnosed in childhood, the other diagnosed as a result of a screening programme for students. Jadresin et al. (2008) and Mariani et al. (1998) reported two rates of adherence; one obtained through self-report, the other obtained from serological markers. Kumar et al. (1988) reported two rates of adherence: adherence before transfer to adult clinics, and in an adult clinic a year later.

Table 4 shows how adherence was operationalised. No two studies utilised the same testing procedures, limiting comparability. Of 10 studies (all except Greco et al., 1997 and Rashid et al., 2005), 8 rely mainly on self-report, although blood tests or biopsies are used for corroboration of adherence (biopsies: Kumar et al., 1988; Mayer et al., 1991). The reliance on self-report could be hypothesised to be biased. Only Jadresin et al. (2008) and Mariani et al. (1998) specifically report adherence using blood test results. For the latter two studies, both report lower rates of adherence using serological markers, highlighting difficulties of using self-report measures to assess adherence. Additionally, where there is a description of adherence, categories do not map onto one another (for example Fabiani et al. 2000 compared to Mariani et al. 1998).

Table 4.

Measures of adherence used.

Study	Measure of adherence	Categories of adherence
Fabiani et al. (1996)	"Precoded" questionnaire asking particularly about breakfast, lunch, snacks and supper.	Either *strict adherence*, or *partially adhere*. No further description given.
Fabiani et al. (2000)	Dietary interview completed by dietician; "food frequency" questionnaire.	Either eats gluten *never, once a month, once a week, several times a week, normal gluten-containing diet.*
Greco et al. (1997)	One day diary of diet and retrospective one month questionnaire to assess adherence.	Either *strict adherence, eats gluten 2-3 times per month, once or more a week, normal gluten-containing diet.*
Hopman et al. (2006)	3 day food record (2 weekdays and 1 weekend day) and questionnaire synthesised from focus group (no further detail given).	Either *strict adherence, occasional ingestion (153mg/d), normal gluten-containing diet.*
Jadresin et al. (2008)	Non-standardised questionnaire. AND, separately Blood tests for EMA.	Questionnaire: Either eats gluten *never, small amounts of gluten less than once a week, regular ingestion of gluten once a week or more.* Blood test: EMA negative = adherence.
Kumar et al. (1988)	Participants asked how strictly adhered to GFD. Some participants followed up by dietician also, but details not given.	Either eats gluten *never, eating several items of gluten a week, normal gluten- containing diet.*
Ljungman & Myrdal (1993)	Non-standardised questionnaire.	Either *gluten ≤ once a month, gluten ≥ once a month, gluten ≥ once a week, normal gluten-containing diet.*
Mariani et al. (1998)	3 day food record (2 weekdays and Sunday), but mainly IgA and EMA tests from blood samples.	Self-report: Either *strict adherence, eats gluten once or twice a week, normal gluten-containing diet.* Blood test: IgA or EMA absence = adherence.
Mayer et al. (1991)	Assessment by dietician of gluten eaten per day over last week, with "precoded" questionnaire.	Either *gluten-free diet, occasional gluten (average .073 g/day), normal gluten-containing diet.*
Rashid et al. (2005)	Non-standardised questionnaire.	*Strict adherence* only category stated; no further descriptions provided.

In terms of the factors that influence adherence, during the weekend and going out with friends were times when the GFD was likely to be compromised (Fabiani et al., 1996). This was corroborated by Ljungman and Myrdal (1993) who stated that considering the context of home, school and out with friends, young people were most likely to contravene the GFD when out with friends, as did Kumar et al. (1988). Mayer et al. (1991) found that young people were more likely to consume gluten-containing foods in social situations to avoid social difficulties when out. At odds with this, however, Hopman et al. (2006) report that 'special occasions' (examples not given) and being at home are associated with non-adherence.

Ljungman and Myrdal (1993) state that boys and older children (15-17 years) are more likely to occasionally eat gluten. Supporting this, Greco et al. (1997) stated that being female and younger (10-13 years, as opposed to older than 18 years) was associated with better adherence. Older participants (17-25 years) were also found to consume significantly greater amounts of fast food containing gluten than younger participants (12-16 years). However, Mariani and colleagues (1998) report no differences in demographics between groups of participants strictly adhering to GFD, or not.

Greco and colleagues (1997) stated that better school grades, as well as higher self-esteem (associated with feeling less restricted by the GFD), were related to adherence. Jadresin et al. (2008) report that those who adhered to the GFD had experienced significantly more biopsies than those who ate gluten and this is corroborated by Mayer et al. (1991). Fabiani et al. (2000)

found that those participants who were screened for CD, as opposed to having a childhood diagnosis of CD, were less likely to adhere to the GFD. One reason posited for this is a perceived lack of benefits of the GFD in the former group, perhaps linked to whether participants were asymptomatic at diagnosis (Fabiani et al., 2000).

A finding consistently reported in studies is that those young people who were deemed non-adherent found the diet hard to manage, no matter how adherence was measured. Greco et al. (1997), Jadresin et al. (2008), Kumar et al. (1988), Ljungman and Myrdal (1993), and Mayer et al. (1991) all describe difficulties young people have in adhering to the GFD. Those deemed to have poor knowledge about CD were less likely to adhere, less likely to know others with CD, skip attendance for checkups, and state unhappiness at health professionals involved in their care (Ljungman & Myrdal, 1993). The factors reported in these studies suggest the importance of the concept of self-efficacy in managing the GFD. Indeed, Nouwen and colleagues (2009) have shown the importance of self-efficacy to dietary self-management in adolescents with diabetes.

In summary, there are some key findings that have been found consistently across studies in relation to factors predicting better adherence: being female and younger, and to a certain extent, greater number of biopsies. Other findings associated with education levels and context provide variable and contradictory evidence. Most studies were poor in terms of quality but particular weight should be given to Fabiani et al. (2000), which appears to be

the most robust study here (scoring 14/21; Table 3), followed by Ljungman and Myrdal (1993; scoring 13/21; Table 3). Fabiani and colleagues (2000) maintain that screening detected young people are less likely to adhere to the GFD than those detected after experiencing symptoms in childhood. Ljungman and Myrdal (1993) report that females and young children (12-14 years) are more likely to adhere, and more likely to adhere at home.

Psychological factors

Cinquetti et al. (1999) aimed to assess what influence, if any, eating a GFD had on the psychological development of adolescents with CD. Using a questionnaire designed for the study, they showed that a great number of adolescents felt different from their friends, reported jealousy that (a) their friends could eat anything and (b) that others were more independent than them. As such, a GFD can be difficult to accept for the majority of children and adolescents, particularly marked for young people aged 12 to 17 years (Cinquetti et al., 1999). Attempts to develop an identity can be disrupted by the diagnosis of CD and difficulties tend to primarily arise when socialising with friends. It is possible that people with CD interpret the GFD as a punishment (Cinquetti et al., 1999), particularly marked when young people are out of their normal context (Cinquetti et al., 1999).

As CD can undermine wellbeing, Cinquetti et al. (1999) posit that this can result in psychological distress. However, this was not corroborated by Kolsteren et al. (2001), who used a generic measure of HRQOL developed in the Netherlands (TNO-AZL) with children diagnosed with CD. HRQOL in

children with CD is equivalent to that of children in the general population, using a reference sample of 1183 children aged 8-12 years. However, when considering the 12-16 year old group in Kolsteren's study, there is no suitable control group used for comparison (data from 11-12 year olds used). Also, there is limited description of results, which precludes thorough critique. Calsbeek et al. (2002), in a study of better quality, found that social position (education, leisure, friendships, finances, romantic partner and sexual relationships) of children and adolescents with CD was not impaired when compared to a healthy control group. However, adherence was not reported, in addition to time since diagnosis and method of diagnosis, which help to contextualise the results. In a study considering coping strategies, Calsbeek et al. (2006b) again found equivalence in children and adolescents with CD when compared to a healthy control group in terms of coping strategies typically used in daily life (either task or emotion-oriented, or avoidance). However, Calsbeek et al. (2006a) report greater illness burden associated with dietary adherence only.

Taking a slightly different approach, van Doorn et al. (2008) reported that children with CD, when using a disease specific measure of CD, report low to neutral QOL. On a generic measure, children with CD reported a lower QOL when compared to healthy controls in all areas apart from the domain of emotions in 8-11 years and 12-15 years; it is possible that these children were under the direct care and instruction of their parent more than older participants, and had not begun to develop autonomy at that time. Fitting with this hypothesis, those aged 16 – 18 years scored their QOL as lower than

younger age groups on DUX-25 suggesting that as one grows older there are more challenges to face which are compromised by having a chronic illness (Calsbeek et al., 2006a).

More recently, however, one of the more methodologically robust studies, van Koppen et al. (2009), measured HRQOL over three time points: pre-diagnosis, 1 year post-diagnosis, and 10 years post-diagnosis. They used the same measure to assess HRQOL (TNO-AZL) prior to and 1-year post diagnosis (on GFD), and the DUX-25 (generic measure) and CD-DUX (specific CD HRQOL measure) were used 10 years post diagnosis. These authors report that for those who are symptomatic pre-diagnosis there is evidence of impaired quality of life, which is improved when on a GFD. However, for those who were asymptomatic pre-diagnosis, there is no evidence of impaired quality of life and no improvement in HRQOL after starting a GFD. Ten years on, those with CD and the reference sample (N=986) reported almost equivalent HRQOL using the DUX-25, but slightly lower HRQOL was reported by CD participants when assessed using CD-DUX; van Koppen et al. (2009) put this down to using a specific measure of HRQOL, which they state is likely to yield lower HRQOL scores. This suggests there are disease specific elements to quality of life that can be masked by more generic measures of HRQOL. However, Grootenhuis et al. (2007) did find lower quality of life on a generic measure of QOL (TNO-AZL) relating only to the social functioning of young children with CD when compared to healthy children and report that children with CD are at risk of developing problems in the areas of motor and cognitive functioning also. This difference between van Koppen's and colleagues

(2009) findings and those of Grootenhuis and colleagues may be a result of the slightly older sample in the former sample, in that young people have adjusted to time on the GFD, or that the study is better quality. As Grootenhuis et al. (2007) do not report time since diagnosis it is not possible to compare these studies more closely.

So far, the research presented suggests mixed results in terms of psychological distress for children and young people with CD. One of four studies with the highest ratings for methodological quality reported in this review, Wagner et al. (2008), assessed adherence and age at diagnosis specifically as factors that may influence QOL. Later diagnosis (older than 6 years) was associated with worse self-reported QOL in the areas of schooling, physical health, burden associated with CD, and increased difficulties in managing peer contact. A lower QOL was also found for those people generally not adhering to the GFD, both within and outside the home. As such, adolescents may eat gluten containing foods and temporarily feel excited (Cinquetti et al, 1999), but Cinquetti et al. (1999) suggest that this will lead to deterioration in both physical and psychological wellbeing, supported by results presented here. Those who adhered to the GFD had comparable QOL to those participants with no chronic illness. Thus, younger age at diagnosis and increased adherence to a GFD are both factors associated with better QOL.

In another study (Kolsteren et al., 2001), younger children with CD (8-11 years) reported significantly greater positive emotions, as well as a greater number of physical complaints, than a reference sample, with girls more

happy with home life than boys. Cinquetti et al. (1999) report that mid-teenage years (13-17 years) appeared to cause “uneasiness” for young people, whereas younger children didn't appear to notice they were on a different diet to others; older adolescents appeared to accept the diet. Young people’s relationship with food was somewhat ambivalent; 2 young people stated they hated food, 5 reported resentment towards food and 19 stated they disliked food. Significantly, young people with educated fathers were less likely to feel guilty after consuming gluten. This may be due to more educated fathers recognising that minor compromises *may* not have long lasting effects, or that these fathers are more often out of the home working and mothers are the main caregivers who subsequently become more lenient with their children. However, it is equally likely this is a spurious correlation; only 39 young people completed the questionnaire.

A study with a more sound methodology, Pynnonen et al. (2004), found lifetime prevalence of depression significantly greater in adolescents with CD than without. Pre-biopsy, people subsequently diagnosed with CD were significantly more likely to have depressive disorder than the control group (those without CD, but having had a biopsy); there were no significant differences in current functioning across groups. Interestingly, CD and parental history of depression were the only variables associated with lifetime and pre-biopsy depressive disorder but parental depressive disorders were more common in the comparison group. Thus, Pynnonen and colleagues (2004) suggested that parental depressive disorder and CD *together* may increase the risk for lifetime prevalence of depressive disorder. Further, Karwautz et al. (2008), one of the studies scoring the highest on

methodological quality, reported an increased rate of eating pathology in females with CD, especially for bulimia nervosa, compared to national estimates from the USA and Europe. However, an important point made by Karwautz and colleagues (2008) is the apparent desire of such individuals to appear “supernormal” (p.404), after finding all female participants with CD demonstrated lower eating pathology overall when compared to school controls. This calls into question the use of self-report measures, and also the way in which sensitive issues are investigated.

Using a different methodology, Olsson and colleagues (2008) recruited 47 adolescents aged 15 to 18 years to 10 focus groups to talk about their experiences of living with CD and the GFD. Transcripts were analysed using Grounded Theory. Non-adherence with GFD was compromised by significant others having a poor knowledge of CD/GFD; eating outside the home; obtaining gluten-free food and, when they do, the palatability of such food; lack of support socially; and how young people perceive themselves if they ingest gluten. If young people were asymptomatic, they were more likely to eat gluten, and the desire to “fit in” suggested that young people would compromise the diet. Overall, the study appeared to be well designed, but only 47 of 159 potential participants agreed to take part, and the bias that this introduces needs to be borne in mind. Additionally, the study was not longitudinal in nature, and there is no discussion of the interview schedule being tested for utility prior to focus groups.

In summary, there is conflicting evidence about the psychological impact of experiencing CD and the GFD. Indeed, some studies offer equivocal findings

in relation to wellbeing between young people with CD and comparison groups, whereas some studies report increased depressive symptoms following diagnosis. The majority of studies report some type of psychological challenge, whether it be jealousy and ambivalence (Cinquetti et al., 1999), lower QOL (van Koppen et al., 2009; Wagner et al., 2008;), eating pathology (Karwautz et al., 2008) or depression (Pynnonen et al., 2004). The four most methodologically sound studies all report some element of psychological challenge posed by CD and its management, but it is unclear whether distress is due to the CD or the experience of the GFD, or both.

Parents' views

Anson et al. (1990) raised important points about the role of parents in helping a child with CD to adapt and manage the GFD. Forty-three sets of parents were interviewed about their knowledge of CD and the GFD, attitudes towards health generally, attitudes towards CD, barriers to adhering to the GFD, and dietary behaviour of themselves and children. Parents of children who were adhering to the diet were better educated (related specifically to fathers' occupation), and from a higher social class. Parents had equivalent knowledge of the disease in both groups, but parents of children adhering to the GFD deemed themselves to be sufficiently informed and were more able to choose gluten free meals from a menu. Overall, the more concerned the parents were about the long term impact of having CD, the better their child was at adhering to the GFD (Anson et al., 1990). Additionally, parents of children not adhering to the GFD reported more difficulties in managing the GFD and managing children who "insisted" on breaking the diet. Special

meals at home were found to be a barrier to the GFD according to parents of non-adherent children, as were parties.

Similarly, Jackson and colleagues (1985) reported that parents of children not strictly adhering to the GFD were less knowledgeable about CD, but that this was also associated with being of a lower social class. Interestingly, those parents who were members of the Coeliac Society (23 of 50 parents) generally demonstrated a greater understanding of CD, and their children had increased dietary adherence (Jackson et al., 1985). Further, at the start of the study, 8 children had biopsies due to concerns about adherence, and of these children's parents, 6 said they found the diet easy to manage. This would suggest that what parents know objectively about managing CD should be compared with their perceptions of their ability and knowledge around CD, similar to Anson and colleagues (1990). However, although laboratory tests were used to assess serological markers, there was no measure of QOL upon which to base conclusions. For example, those who were not adhering to GFD may have been asymptomatic, which may influence CD management.

Overall conclusions

There is contradictory evidence on the factors relating to adherence to the GFD, although being female and younger tends to be associated with increased adherence, and increased number of biopsies. Further, there is conflicting research about psychological consequences, if any, of having CD and maintaining the GFD. Generally, decreased adherence is associated with lower quality of life, but having CD is, in some studies, associated with

decreased wellbeing regardless of adherence. Also, managing the temptation to eat gluten-containing foods appears to be an important aspect of CD for young people, particularly when outside the home and in the company of peers. Parents appear to have an important role to play in helping children to manage the GFD and adjust to CD; parents who perceive themselves as more knowledgeable about CD are more likely to have children adhering to the GFD. However, there is limited research in this area and further studies are required to understand better the complex findings drawn from existing literature.

Discussion

Considering the literature reviewed in this paper, there appears to be some evidence that certain factors are important in relation to adherence to the GFD, as well as some evidence that psychological wellbeing can be adversely affected by either the experience of having CD, adhering to a GFD, or both. However, the methodological quality of the studies, in addition to the paucity of literature directly addressing CD in children and young people, precludes any firm conclusions.

Study findings

There are some important themes that can be drawn from the limited findings reported here. One is that of self-efficacy, and this is something that appears to be important for both young people with CD, and their parents. Bandura (1977) stated that it is not necessarily what resources a person has to perform a task that is important; it is what that person *perceives* as their resources to perform a task. Specifically, beliefs about self-efficacy are related to:

1. Previous experiences of managing a given task
2. Vicarious experiences (modelling)
3. Social persuasions
4. Arousal

Considering young people, Cinquetti et al. (1999) report feelings of jealousy and ambivalence about food, and this purported high arousal may confound a

persons' ability to manage the GFD with peers; a common finding across studies. Additionally, considering the finding that parents who perceived themselves more knowledgeable about CD (Anson et al., 1990) have children more likely to adhere to the GFD, can be seen as evidence for the importance of parents' self-efficacy beliefs in managing the GFD. Further, one mechanism that may explain why children are more likely to adhere to the GFD if their parents view themselves as more knowledgeable is that of modelling; seeing one's parents manage a situation could increase self-efficacy beliefs. The same mechanism may be at work for those young people who suffer with depression and have CD, which is more likely when their parents have depression (Pynnonen et al., 2004). However, seeing one's parents not managing as well, or having negative thoughts and ideas which most likely generalise to CD also, young people may be at increased risk of depression, particularly as CD can be difficult to manage (Calsbeek et al., 2006a) regardless of parents' mental health. Similarly, the finding that young people tend to break the GFD when outside the home may be associated with social persuasions; if peers encourage young people to break the diet, this may well have more of an impact without the support of parents who reinforce adherence. This may also link in with children's perceived expectancies as to what will happen if they do maintain the GFD, for example, that they will be ridiculed. Linked to this, Sawyer and Aroni (2003) maintain that educational professionals, for example, need an awareness of asthma treatment in order to fully support young people with asthma at school.

Additionally, there is a theme around developmental level. It is well recognised in developmental literature that as children get older they start to explore the world and develop a sense of self (e.g. Eriikson, 1982). Blos (1967, as cited in Shaw, 2001) describes the idea of separation-individuation in adolescence, whereby a young person strives to become autonomous and develop their identity. Young people will begin to assert themselves with authority figures and strive for independence from such figures. However, having a chronic illness may compromise this, in that parents may need to be involved in managing the GFD, arranging and attending hospital visits, and speaking to school staff (Eiser, 1990; Shaw, 2001). Thus, this conflict may cause issues around adherence (Shaw, 2001), but also psychological distress (Eiser, 1990).

Further, it could be hypothesised that illness representations (from the commonsense model of self-regulation; Leventhal, Meyer & Nerenz, 1980) have a role in CD management. There are five components of illness representations: Identity; Cause; Time line; Consequences; and Cure/Control. Particularly in studies that sampled screening detected students (e.g. Fabiani et al., 2000), if young people are asymptomatic there may be no perceived disease identity (i.e., symptoms attributable to CD), no perceived benefits of the GFD, and no perceived consequences of eating gluten (Olsson et al., 2008). It could be speculated that such illness representations may be related to non-adherence to a GFD. It may be of relevance to note that recent research exploring factors that are important to dietary self-management in adolescents with diabetes, has found that both self-efficacy and illness

representations (specifically perceived consequences and beliefs in the effectiveness of diet to control diabetes) are important to dietary self-care and psychological well-being (Nouwen et al., 2009). The potential utility of applying the commonsense model to explore further the factors predisposing and maintaining levels of adherence and wellbeing in children and young people with CD is within reason.

Methodological quality

Most of the studies employed a cross-sectional design without a control group, and although this can be useful, it can be difficult to draw conclusions about how a group may experience an event in relation to others without CD and to assess how the disease and its management may alter over time. Some studies reported here did utilise a control group, and only one study implemented a prospective longitudinal design (van Koppen et al., 2009), which was useful to establish how HRQOL changes over time. Studies have varied in their quality, and it is important that future studies use longitudinal designs with larger samples in order to effectively progress this area.

Further, the majority of studies were conducted in the Netherlands (where prevalence in children is 1:198; Csizmadia et al., 1999), with only one completed in the UK. This limits generalisability of results to the UK population of young people with CD, as the UK has different health, education and welfare systems which may impact on outcomes. Some studies had very few participants (Fabiani et al., 1996; Fabiani et al., 2000; Pynnonen et al., 2004), which again limits generalisability of results, as well as potentially affecting

conclusions that can be drawn. Indeed, rates of attrition were high in some studies (Hopman et al., 2006; Jadresin et al., 2008; Kolsteren et al., 2001; Olsson et al., 2008); thus a self–selection bias is more than likely present. Ideally, randomisation to condition would provide more robust results.

In only two studies (Anson et al., 1990 and Pynnonen et al., 2004) did authors describe religious background or ethnicity. Different religious and ethnic backgrounds may affect the way in which people cope and seek help, as well as how CD is construed as part of life. The lack of such information will, again, reduce the generalisability of results.

A major weakness in papers was a lack of robust paper and pencil measures to assess for dietary adherence and wellbeing, and a lack of consistency. Moreover, in the case of studies describing adherence, the definition of adherence varied. This reduces the comparability across studies and therefore different adherence rates in different groups of young people need to be interpreted with caution. De Civita and Dobkin (2005) state that the most useful way to assess treatment behaviours is through validated questionnaire measures, together with objective measures such as biological tests; in the case of CD, this could take the form of serological markers and biopsies.

In all cases generalisability was compromised by the study's country of origin, the strict selection criteria, or the lack of information presented in the study. Few studies adequately controlled for confounding variables statistically, and virtually all, apart from Pynnonen et al. (2004), failed to consider parental CD

as a possible influence on children's wellbeing. The inclusion of time since diagnosis and familial CD needs to be included in future studies, as do issues of age, gender, and socioeconomic status.

Conclusions

Given what is known about other chronic illnesses and the increased prevalence of emotional and behavioural difficulties in this group of children, early recognition of these difficulties is crucial to help increase wellbeing (Hysing et al., 2009). Although evidence presented here is contradictory, there does appear to be some psychological consequence to being diagnosed with CD and experiencing this disease in childhood. Clinically, this has implications for both the physical treatment offered to these young people as well as the need to recognise, assess and intervene to enhance psychological wellbeing. Given that CD can present with symptoms outside of the gut, for example, depression, it is imperative that health professionals are aware of the prevalence of such disorders, but also how best to manage such cases. There needs to be more focus on the theories that may explain adherence in children and young people as has been the case in other chronic illnesses, such as diabetes, to help inform interventions. Consideration of self-efficacy specifically, as well as illness representations (Hysing et al., 2009), will inform how childhood CD is managed, and how efficacy and autonomy can be supported by professionals and family alike. This will promote adherence, as well as wellbeing.

References

** denotes paper included in systematic review*

*Anson, O., Weizman, Z., & Zeevi, N. (1990). Celiac Disease: Parental knowledge and attitude of dietary compliance. *Pediatrics, 85,* 98-103.

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioural change. *Psychological Review, 84,* 191-215.

Beck, A. T., Epstein, N., Brown, G., & Steer, R. A. (1988). An inventory for measuring clinical anxiety: Psychometric properties. *Journal of Consulting and Clinical Psychology, 56,* 893–897.

Beck, A. T., Ward, C. H., Mendelson, M., Mock, J., & Erbaugh J. (1961). An inventory for measuring depression. *Archives of General Psychiatry, 4,* 561–571.

Bingley, P. J., Williams, A. J. K., Norcross, A. J., Unsworth, D. J., Lock, R. J., Ness, A. R., & Jones, R. W. (2004). Undiagnosed Coeliac Disease at age 7: Population based prospective birth cohort study. *British Medical Journal, 328,* 322-323.

Booth, I. (1991). The nutritional consequences of gastrointestinal disease in adolescence. *Acta Paediatrica Scandinavica Supplement, 373,* 91-102.

British Society of Gastroenterologists. (2002). *Guidelines for the management of patients with Coeliac Disease.* UK: British Society of Gastroenterologists.

*Calsbeek, H., Rijken, M., Bekkers, M. J. T. M., Kerssens, J. J., Dekker, J., van Berge Henegouwen, G. P., & participating centres. (2002). Social

position of adolescents with chronic digestive disorders. *European Journal of Gastroenterology and Hepatology, 14,* 543-549.

*Calsbeek, H., Rijken, M., Bekkers, M. J. T. M., Dekker, J., & van Berge Henegouwen, G. P. (2006a). School and leisure activities in adolescents and young adults with chronic digestive disorders: Impact of burden of disease. *International Journal of Behavioral Medicine, 13,* 121-130.

*Calsbeek, H., Rijken, M., Bekkers, M. J. T. M., van Berge Henegouwen, G. P., & Dekker, J. (2006b). Coping in adolescents and young adults with chronic digestive disorders: Impact on school and leisure activities. *Psychology and Health, 21,* 447-462.

*Cinquetti, M., Trabucchi, C., Menegazzi, N., Comucci, A., Bressan, F., & Zoppi, G. (1999). Psychological problems connected to the dietary restrictions in the adolescent with Coeliac Disease. *Medical and Surgical Pediatrics, 21,* 279-283.

Coeliac UK. (2007). Retrieved 23 April, 2007, from www.coeliac.org.uk.

Connolly, M. A., & Johnson, J. A. (1999). Measuring quality of life in paediatric patients. *Pharmacoeconomics, 16,* 605-625.

Csizmadia, C. G. D. S., Mearin, M. L., Von Blomberg, B. M. E., Brand, R., & Verloove-Vanhorick, S.P. (1999). An iceberg of childhood Coeliac Disease in the Netherlands. *Lancet, 353,* 813–814.

De Civita, M., & Dobkin, P. L. (2005). Pediatric adherence: Conceptual and methodological considerations. *Children's Healthcare, 34,* 19-34.

Eiser, C. (1990). *Chronic Childhood Disease: An Introduction to Psychological Theory and Research*. Cambridge, UK: Cambridge University Press.

Erikson, E. (1982). *The life cycle completed: A review.* New York: Norton.

*Fabiani, E., Catassi, C., Villari, A., Gismondi, P., Pierdomenico, R., Ratsch, I. M., Coppa, G. V., & Giorgi, P. L. (1996). Dietary compliance in screening detected Coeliac Disease adolescents. *Acta Paediatrica, 412,* 65-67.

*Fabiani, E., Taccari, L. M., Ratsch, I., Di Guiseppe, S., Valentino Coppa, G., & Catassi, C. (2000). Compliance with gluten-free diet in adolescents with screening detected Celiac Disease: A 5 year follow-up study. *Journal of Pediatrics, 136,* 841–843.

Fekkes, M., Theunissen, N. C. M., Brugman, E., Veen, S., Verrips, E. G. H., Koopman, H. M., Vogels, T., Wit, J. M. & Verloove-Vanhorick, S. P. (2000). Development and psychometric evaluation of the TAPQOL: A health-related quality of life instrument for 1–5-year-old children. *Quality of Life Research, 9,* 961-972.

*Greco, L., Mayer, M., Ciccarelli, G., Troncone, R., & Auricchio, S. (1997). Compliance to a gluten free diet in adolescents, or “what do 300 Coeliac adolescents eat every day?” *Italian Journal of Gastroenterology and Hepatology, 29,* 305-310.

*Grootenhuis, M. A., Koopman, H. m., Verrips, E. G. H., Vogels, A. G. C & Last, B. F. (2007). Health-related quality of life problems of children aged 8-11 years with a chronic disease. Developmental Neurorehabilitation, 10, 27-33.

Hamilton, M. (1959). The assessment of anxiety states by rating. *British Journal of Medical Psychology, 32,* 50–55.

Hamilton, M. (1960). A rating scale for depression. *Journal of Neurology, Neurosurgery & Psychiatry, 23,* 56–62.

Hawkes, N. D., Swift, G. L., Smith, P. M., & Jenkins, H. R. (2000). Incidence and presentation of Coeliac Disease in South Glamorgan. *European Journal of Gastroenterology & Hepatology, 12,* 345-349.

Hill, I. D., Dirks, M. H., Liptak, G. S., Colletti, R. B., & Fasano, A. (2005). Guideline for the diagnosis and treatment of Coeliac Disease in children: Recommendations of the North American Society for Pediatric gastroenterology, Hepatology and Nutrition. *Journal of Pediatric Gastroenterology and Nutrition, 40,* 1-19.

*Hopman, E. G. D., le Cessie, S., von Blomberg, B. M. E., & Mearin, M. L. (2006). Nutritional management of the gluten-free diet in young people with Celiac Disease in the Netherlands. *Journal of Pediatric Gastroenterology and Nutrition, 43,* 102-108.

Huff, C. (1997). Celiac Disease: Helping families adapt. *Gastroenterology Nursing, 20,* 79-81.

Hysing, M., Elgen, I., Gillberg, C., & Lundervold, A. J. (2009). Emotional and behavioural problems in subgroups of children with chronic illness: Results from a large-scale population study. *Child: Care, Health and Development, EPublished ahead of print, 23rd March.*

*Jackson, P. T., Glasgow, J. F. T., & Thom, R. (1985). Parents' understanding of Coeliac Disease and diet. *Archives of Disease in Childhood, 60,* 672-674.

*Jadresin, O., Misak, Z., Kolacek, S., Sonicki, Z., & Zizic, V. (2008). Compliance with gluten-free diet in children with Coeliac Disease. *Journal of Pediatric Gastroenterology and Nutrition, 47,* 344-348.

*Karwautz, A., Wagner, G., Berger, G., Sinnreich, U., Grylli, V., & Huber, W. (2008). Eating pathology in adolescents with Celiac Disease. *Psychosomatics, 49,* 399-406.

Kaufman, J., Birmaher, B., Brent, D., Rao U., & Ryan. N. (1996). Diagnostic Interview: Kiddie-Sads-Present and Lifetime Version (K-SADS-PL). Available from http://www.wpic.pitt.edu/KSADS/ksads-pl.pdf.

*Kolsteren, M. M., Koopman, H. M., Schalekamp, G., & Mearin, M. L. (2001). Health-related quality of life in children with Celiac Disease. *The Journal of Paediatrics, 138,* 593-595.

Koopman, H. M., Baars, R. M., & Mearin, L. M. (2003). Health related quality of life (HRQOL) in children with Coeliac Disease. *Coeliac Disease: Proceedings of the Xth International Symposium on CD, 10th symposium,* 291-294.

*Kumar, P. J., Walker-Smith, J., Milla, P., Harris, G., Colyer, J., & Halliday, R. (1988). The teenage Coeliac: Follow up study of 102 patients. *Archives of Disease in Childhood, 63,* 916-920.

Leventhal, H., Meyer, D., & Nerenz, D. (1980). The commonsense representations of illness danger. In S. Rachman [Ed]. *Medical Psychology* (Volume 2, pp. 7-30). New York: Pergamon.

*Ljungman, G., & Myrdal, U. (1993). Compliance in teenagers with Coeliac Disease: A Swedish follow-up study. *Acta Paediatrica, 82,* 235-238.

*Mariani, P., Viti, M. G., Montouri, M., La Vecchia, A., Cipolletta, E., Calvani, L., & Bonamico, M. (1998). The Gluten-free diet: A Nutritional risk factor for adolescents with Celiac Disease? *Journal of Pediatric Gastroenterology and Nutrition, 27,* 519-523.

Mattejat, F. & Remschmidt, H. (no date). *The inventory of life quality in children and adolescents (ILC) [German].* Available from http://www.uni-marburg.de/fb20/kjp/eval/aglq/download/ilk.fb.jugendliche.

*Mayer, M., Greco, L., Troncone, R., Auricchio, S., & Marsh, M. N. (1991). Compliance of adolescents with Coeliac Disease with a gluten free diet. *Gut, 32,* 881-885.

Mearin, L. M. (2007). Celiac Disease among children and adolescents. *Current problems in Pediatric and Adolescent Health Care, 37,* 86-105.

Mihailidi, E., Paspalaki, P., Katakis, E., & Evangeliou, A. (2003). Celiac Disease: A pediatric perspective. *International Pediatrics, 18,* 141-148.

Mitrofan, O., Paul, M., & Spencer, N. (2008). Is aggression in children with behavioural and emotional difficulties associated with television viewing and video game playing? A systematic review. *Child: Care, Health and Development, 35,* 5-15.

Nouwen, A., Law, G. Urquhart., Hussain, S., McGovern, S. & Gibbons, H. (2009). Comparison of the role of self-efficacy and illness representations in relation to dietary self-care and diabetes distress in adolescents with type 1 diabetes. *Psychology and Health, 1-14,* iFirst. DOI: 10.1080/08870440802254597.

*Olsson, C., Hornell, A., Ivarsson, A., & Sydner, Y. M. (2008). The everyday life of adolescent Coeliacs: Issues of importance for compliance with the gluten-free diet. *Journal of Human Nutrition and Dietetics, 21,* 359-367.

*Pynnonen, P. A., Isometsa, E. T., Aronen, E. T., Verkasalo, M. A., Savilahti, E., & Aalberg, V. A. (2004). Mental disorders in adolescents with Celiac Disease. *Psychosomatics, 45,* 325-335.

*Rashid, M., Cranney, A., Zarkadas, M., Graham, I. D., Switzer, C., Case, S., Molloy, M., Warren, R., Burrows, V., & Butzner, J. D. (2005). Celiac Disease: Evaluation of the diagnosis and dietary compliance in Canadian children. *Pediatrics, 116,* e754-e759.

Sawyer, S. M., & Aroni, R. A. (2003). Sticky issue of adherence. *Journal of Paediatric Child Health, 39,* 2-5.

Schmidt, S., Petersen, C., & Bullinger, M. (2003). Coping with chronic disease from the perspective of children and adolescents: A conceptual framework and its implications for participation. *Child: Care, Health and Development, 29,* 63-75.

Shaw, R. J. (2001). Treatment adherence in adolescents: Development and psychopathology. *Clinical Child Psychology and Psychiatry, 6,* 137-150.

*van Doorn, R. K., Winkler, L. M. F., Zwinderman, K. H., Mearin, M. I., & Koopman, H. M. (2008). The CDDUX: A disease specific health related quality of life questionnaire for children with Celiac Disease. *Journal of Pediatric Gastroenterology and Nutrition, 47,* 147-152.

*van Koppen, E. J., Schweizer, J. J., Csizmadia, C. G. D.S., Krom, Y., Hylkema, H. B., van Geel, A. M., Koopman, H.M., Verloove-Vanhorick, S. P., & Mearin, M. L. (2009). Long term health and quality of life consequences of mass screening for childhood Celiac Disease: A 10 year follow up study. *Pediatrics, 123,* 582-588.

Varni, J. W., Seid, M., & Kurtin, P. S. (1999). Pediatric health-related quality of life measurement technology: A guide for health care decision makers. *Journal of Clinical Outcomes Management, 6,* 33-40.

Verrips, E. G. H., Vogels, T. G. C., Koopman, H. M., Theunissen, N. C. M., Kamphuis, R. P., Fekkes, M., Wit, J. M., & Verloove-Vanhorick, S. P. (1999). Measuring health-related quality of life in a child population. *The European Journal of Public Health, 3,* 188-193.

*Wagner, G., Berger, G., Sinnreich, U., Grylli, V., Schober, E., Huber, W., & Karwautz, A. (2008). Quality of life in adolescents with treated Coeliac Disease: Influence of compliance and age at diagnosis. *Journal of Pediatric Gastroenterology and Nutrition, 47,* 555-561.

Walker-Smith, J. A., Guandalini, S., Schmitz, J., Shmerling, D. H., & Visakorpi, J. K. (1990). Revised criteria for the diagnosis of Celiac Disease: Report of the working group of ESPGAN. *Archives of Disease in Childhood, 65,* 909-911.

Wallander, J. L., & Varni, J. W. (1998). Effects of pediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry, 39,* 29-46.

West, J., Logan, R. F. A., Hill, P. G., Lloyd, A., Lewis, S., Hubbard, R., Reader, R., Holmes, G. K. T., & Khaw, K. T. (2003). Seroprevalence,

correlates and characteristics of undetected Coeliac Disease in England. *Gut, 52,* 960-965.

Zigmund, A. S., & Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale. *Acta Psychiatrica Scandinavia, 67,* 361-370.

“Am I allowed that?” A qualitative study of young people’s experiences of living with Coeliac Disease

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Abstract

Background: In Coeliac Disease (CD) the only treatment is adherence to a strict gluten free diet. Research studies describing people’s experiences of CD are limited in the literature, particularly studies of the experience of children and adolescents with the condition. Thus, the aim of this study was to interview children with CD about their experiences of managing their illness.

Method: Five children and young people were recruited from local voluntary sector Coeliac support groups and interviewed separately.

Findings: Transcripts were analysed using Interpretative Phenomenological Analysis. Two themes are reported: *Managing identity as a young person with CD*, and *Ambivalent relationship with food.* The results illustrate how children and young people sometimes felt isolated and different from peers, but felt valued when others catered for them. Furthermore, young people talked about not necessarily enjoying food, rather, it was important to eat something and to still be able to socialise.

Conclusions: Currently, hospital annual checks attended by young people yearly could incorporate a psychological component to assess psychological wellbeing and dietary management. However, it is important to conduct additional qualitative studies and studies that involve parents to better understand the area, and longitudinal research with children to consider developmental issues. Psychological wellbeing might be useful to study, as well as young people’s coping strategies to reliably inform any interventions required.

Introduction

What is Coeliac Disease?

Coeliac Disease (CD) is not an allergy, but a chronic autoimmune disease. It is one of the most common autoimmune diseases in the UK with a childhood prevalence of 1:100 (Bingley et al., 2004). In CD, antibodies are produced and these antibodies subsequently attack the body in response to eating gluten, the protein found in wheat, rye and barley (McGough & Cummings, 2005). There are many symptoms of CD, from diarrhoea and anaemia to bone fractures and fertility problems (Troncone et al., 2008). Typically, CD was thought to be a disease of childhood, but it is now recognised that diagnosis can be made at any age (Troncone, Ivarsson, Szajewska & Mearin, 2008). Diagnosis typically involves a screening blood test followed by biopsy of the small intestine (British Society of Gastroenterologists, 2002), and in a recent review, van Heel and West (2006) suggest that the most common age of diagnosis in childhood is between 9 and 24 months old. The delay in diagnosis from onset of symptoms has been reported to be between 0 to 12 years (median 1 year; Rashid et al., 2005) in children and approximately 11 years in adults (Cranney et al., 2007; Green et al., 2001), and one possible reason for delay might include health professionals' inability to recognise and test for CD (Hill, Dirks, Liptak, Colletti & Fasano, 2005).

Currently, there is no cure for CD and management is achieved through adherence to a life-long gluten-free diet (GFD; Hill et al., 2005). This includes a diet free from food such as bread, pasta and pastries, as well other foods such

as fruit squashes that contain barley, some sweets, and table sauces. Mearin (2007) reports the difficulty in managing the GFD because industrial processes often compromise the gluten content of foods. Furthermore, adherence to the diet can be compromised by motivation to adhere to the diet, which can vary across people and stages of life (Pietzak, 2005). There are possible physical health consequences as a result of having CD, and CD has been associated with a high rate of gut lymphoma and osteoporosis, which is particularly accentuated in untreated CD (Coeliac UK, 2009). However, there are an increasing number of studies that also cite psychological and social implications of having CD (e.g. Mearin, 2007).

What is known about adults with CD?

Most research has focused on adults with CD. In a recent review, Addolorato and colleagues (2008) summarised reports of adults with CD, who also suffer with an affective disorder, including anxiety and depression, social phobia, and sexual disorders. They also cite studies of CD that report co-morbid psychiatric and developmental disorders including schizophrenia, autism, and eating disorders. Those adults non-adherent to the GFD tended to suffer with psychological distress (Addolorato et al., 2008), and this appears to be the case whether this developed prior to diagnosis (and therefore GFD not recognised as necessary, for example Ciacci et al., 1998) or through choosing not to maintain the diet once diagnosed (Hauser, Stallmach, Caspary & Stein, 2007). Additionally, women with CD report increased psychological distress (anxiety and depression) and decreased self control, vitality and general health compared with men with CD (Roos, Karner & Hallert, 2006). Interestingly,

Ciacci et al. (2005) found that for people who more often adhered to the GFD, they reported a greater level of depression, regardless of duration of GFD. This might be because the GFD is perceived to be restrictive in daily living, for example socialising (Lee & Newman, 2003).

The majority of published studies tend to be quantitative in nature, but there are reports using qualitative methods also. These aimed to develop an understanding of living with the diagnosis. Sverker, Ostlund, Hallert and Hensing (2007) interviewed close relatives of people with CD (21 partners of people with CD and 2 parents of a person with CD, age of child not reported) to explore dilemmas experienced by relatives and how these were managed. A phenomenologically informed methodology, whereby participants were asked for critical incidents that have distressed them (Critical Incident Technique, Flanagan, 1954 as cited in Sverker et al., 2007), was used. Close relatives report concerns related specifically to CD (i.e. feeling guilty they don't have CD, anxiety, and witnessing difficulties their relative has), daily activities (i.e. increased work around the house, inability to be spontaneous, and the person with CD controlling situations related to food, which also sometimes generalised to other areas of life), and their social life (i.e. others lacking information, knowledge and understanding, which meant a more restricted social life, as going out was avoided). Clearly there are some important findings here that have clinical implications for health professionals providing support to people with CD and their families.

The same authors have also interviewed people with CD aged between 20 and 40 years (Sverker, Hensing & Hallert, 2005) using the same procedure. The analysis yielded three themes: emotions, relationships and managing daily life. Specifically, participants reported feeling isolation, shame, anxiety about contamination from gluten containing products, and concerns about being a burden. In terms of relationships, participants disclosed feelings of being neglected and forgotten, but also not wanting to tell others about their CD and not wanting it to be visible. Participants also reported risks they took in managing the diet with other people, for example, eating gluten with certain family members, or because food items being eaten had previously not contained gluten, although current content had not been checked. Participants reported feeling restricted in food choice and finding gluten-free food effortful. Interestingly, those people diagnosed in childhood recalled dilemmas from adolescence, particularly dilemmas associated with eating with others outside the family home. Moreover, these authors noted how these dilemmas were often associated with intense feelings, for example, anger. These results potentially highlight that having a chronic illness in adolescence can be a major threat to a young person’s identity as they grow up (Koopman, Baars & Mearin, 2003).

So what is known about CD in relation to children and young people?

There is a dearth of research on the impact of living with CD for children and young people. What is available focuses on functional outcomes, screening exercises, and co morbid conditions (including physical conditions, autism and Down’s Syndrome), and some research reports the impact of medical

procedures children undergo during CD diagnosis and follow up (Hogberg, Nordwall & Stenhammer, 2001; Skoglosa, Falth-Magnusson & Stenhammer, 2003; Smith, 1996). There are many reports about GFD adherence, and these tend to highlight adolescence as a time when this might be compromised (Booth, 1991). Furthermore, reports about adherence tend to use non-validated measures, which are inconsistent across studies. It might be unsurprising therefore, to learn that factors linked to adherence are also contradictory, although most commonly being female and younger is associated with better adherence to the GFD (for example, Ljungman & Myrdal, 1993). There are an almost equivalent number of studies that report an aspect of psychological wellbeing, with variable findings obtained from various measures. However, what appears consistent is some element of psychological distress associated with having CD in childhood, whether it be jealousy and ambivalence (Cinquetti et al., 1999), lower quality of life (QOL; van Koppen et al., 2009; Wagner et al., 2008), eating pathology (Karwautz et al., 2008) or depression (Pynnonen et al., 2004).

Recent attempts have been made, as can be seen in the adult literature, to explore young people's ideas about CD using qualitative methods. Olsson, Hornell, Ivarsson and Sydner (2008) report focus groups results from 47 adolescents (aged 15-18 years) who were diagnosed with CD in childhood. The aim of their study was to try and elucidate reasons for compliance and non-compliance with GFD in adolescence. Using Grounded Theory, these authors report that, outside the home participants deemed it more difficult to manage the GFD, with teachers, chefs, and other family members often not

understanding what CD is and how it should be managed; these situations were reported to affect whether the young person complied with the diet. Ultimately, adolescents wanted to 'fit in' and the GFD was a barrier to this. Adolescents reported temptations to give up the GFD when other people did not understand the importance of the diet, and there were also some situations where the young person was perceived as too "self-important" (p. 362; Olsson et al., 2008) because they wanted to maintain their diet. These findings have implications for how young people are supported to manage the GFD, but these authors suggest that "for a more comprehensive understanding of what it is like to be a Coeliac adolescent, the present work could be usefully taken forward as the basis for individual interviews" (p. 366).

Aims

In summary, CD appears to be an under-researched area with regards to psychosocial outcomes in children and young people. Where reports do exist, there are contradictory outcomes, and inconsistent and non-validated measures used. Moreover, only one report was found whereby adolescents had been asked specifically about their experiences of CD. Given the paucity of data specifically obtained from children and young people, the lack of sound measures to explore wellbeing and experience of CD, the lack of understanding of childhood CD, and reports of possible psychological distress associated with CD in childhood, a qualitative methodology was selected here. This allows for a detailed exploration of a little-understood area (Krahn & Putnam, 2003), and to allow children and young people to tell others about what matters to them, in relation to their CD (Reid, Flowers & Larkin, 2005). This study aimed to explore

the lived experiences of children and young people with CD using an exclusively phenomenological qualitative methodology, to establish what is important to a group of children and young people with CD.

Method

Participants

Children and young people aged between 10 and 17 years, with a diagnosis of CD, and who were members of a local Coeliac UK support group, were invited to participate. Participants were excluded if either parent had CD, the child with CD had a known mental health diagnosis or co-morbid health condition, they did not speak English, or were older than pre-school age at diagnosis. One child was excluded because the whole family wanted to be present during the interview (possibly affecting validity of the interview), one child was excluded because they had a co-morbid health condition, and two children were excluded because they were older than pre-school age at diagnosis. A homogeneous group (as recommended by Smith & Osborn, 2003) of 5 young people were recruited, which was deemed an acceptable sample size (Brocki & Wearden, 2006). Demographic information is shown in Table 5. The children ranged in age from 10 to 16 years, with 3 girls and 2 boys interviewed. All participants were White British and subjectively rated as middle class. Age at diagnosis ranged from 12 months to 48 months (M= 28 months S.D=15.2 months), and all were diagnosed between 1993 and 2001. All young people were keen to take part in the study, and were agreeable to having the interviews recorded and results disseminated. In every case the mother was also interviewed, but parental interviews are not the focus of the study reported here and thus parental data are not included in this analysis.

Table 5.

Participant demographics.

Participant	Child gender & age in years*	Parent gender & age in years*	Parent occupation	Ethnicity	Age (year) child diagnosed	Self-reported knowledge of CD at diagnosis	Other family members with CD?
Ethan	M 14	F 51	Social worker	White British	2.5 years (1996)	No	No
Claire	F 10	F 44	Housewife	White British	4 years (2001)	No	No
Tara	F 15	F 45	Account manager	White British	14 months (1993)	No	No
Sarah	F 12	F 44**	Customer service manager	White British	3 years (1999)	No	Yes - grandparent
Tom	M 16	F 43	Teacher	White British	1 year (1993)	No	No

Note. All names have been changed to protect confidentiality. *M = male, F = female. ** Step-mother. Self-reported knowledge was assessed through asking participants whether they knew about CD at time of diagnosis.

Measures

Semi-structured interviews are suggested by Smith and Osborn (2003) as best practice for conducting research using Interpretative Phenomenological Analysis, as is the case here. Existing literature in the area was consulted, and a 10 year old child diagnosed with CD (separate from the participants) commented on the schedule once this had been done. She stated that questions should be included to reflect the impact of family in managing CD, and this was subsequently added. The resulting drafts of interview schedules were sent to a local dietician and gastroenterologist working in the field of CD for additional comment. Feedback from the gastroenterologist led to the addition of a question about "cheating" (eating gluten) and the impact this had on the young person. Comments received from the dietician resulted in inclusion of another question, to establish experience of the prescription service. Thus, final interviews involved participants describing their understanding of CD, their experiences of managing it, what it feels like to have CD, and long term implications. Susie, another young person, was interviewed using the interview schedule; her data are not presented here but instead were used as a pilot interview to test the integrity of the schedule. Subsequently, changes were made to the prompts in order that they were more open. Despite the semi-structured nature of the schedule, participants were encouraged to talk about their experiences in their own time, with prompting only as appropriate (Appendix 3).

Procedure

The study was approved by the University of Birmingham's School of Psychology Human Research Ethics Committee (Appendix 4). Participants were recruited by the researcher from support groups run by Coeliac UK (a non-NHS charitable organisation). The researcher attended two support group meetings covering different geographical areas in the West Midlands (43 children registered at one group, and 50 at the other; ages were not available but all the young people were under 18 years). At the local groups, the research was described and information leaflets made available for those families interested in participating (Appendix 5 and 6). Interested families self-referred to the research either through returning a reply slip or telephoning the researcher (see Appendix 7), and the researcher then made contact with the families to explain the research and answer queries. Subsequently, if participants wanted to take part, an appointment was made to interview the young person at home. Informed consent was obtained from each child participant and their parent (Appendix 8). It was explained to participants that they were free to withdraw from the study at any time without being removed from the prize draw, up until receipt of the transcripts, after which time, withdrawal was not possible. All interviews were digitally recorded and ranged from 15 to 75 minutes, and at the end of the interview participants were debriefed and thanked for their time. In all cases participants continued to disclose experiences after the recorder was switched off and these comments were collated in a reflective diary in order that they be included, with consent, in the analysis. Participants were sent their transcripts prior to analysis to ensure accuracy of transcription. All transcripts

were approved for use by the young people. As a reward for participating in the research, all children who took part in the interviews were entered into a prize draw to win an iPod shuffle.

Data analysis

Analysis was carried out using Interpretative Phenomenological Analysis (IPA; Smith & Osborn, 2003; for a full account, please see Appendix 9). IPA is especially useful for understanding people's experiences (rather than social processes, as in ethnography, Krahn & Putnam, 2003), and for understanding individual meaning of participants' accounts (Smith & Osborn, 2003), without imposing a theoretical structure on the findings (as in Grounded Theory; Osborne, 1994). It is frequently used in health psychology research (Brocki & Wearden, 2006), and as such has much value in allowing for thorough exploration of an under-researched area, as in this study. IPA was chosen because of its commitment to exploration of an area, rather than explanation of phenomena with theory development (Reid et al., 2005).

Reflexivity

Brocki and Wearden (2006) strongly emphasise the value of a brief reflexive account in IPA studies. Therefore, in this section, I aim to give a brief account of important factors I might bring to this research. I am a 27 year-old white British female trainee clinical psychologist. I do not have any children of my own and therefore when conducting interviews with young people I thought I could focus more on what they were saying rather than thinking about what I would do if I

was their parent. I am also a part time youth worker and therefore have contact with young people often. I think this helped me in carrying out my analysis, as I had current interactions with children and young people at work which helped to contextualise some of the issues people were describing. I do not have CD but do have close friends who have food intolerances. One friend in particular was hospitalised for a substantial amount of time before she was diagnosed, and I witnessed her anger and confusion at being unwell. I felt helpless watching my friend suffer, but at the same time tried to engage in practical tasks to reduce the visibility of her illness. I would be naïve to think that these were not in my mind during both data collection and analysis. At the start of this project I didn't know I was actually intolerant to both cow's milk and yeast, and toward the end of the project I had to omit these from my diet. I was shocked, frustrated and then felt resigned to living without these things in my diet. Consequently I went through transcripts again to see whether, as a result of this, I would have picked out any different ideas (whether phenomenological or interpretative in nature); I didn't, but I felt a stronger connection to the data and a greater desire to do justice to it. I'm particularly interested in motivation for certain behaviours and this relates in this study to adherence to the GFD, and during the study I began to question my own assumptions about the ease of maintaining a specific diet when others around you were not.

Credibility of analysis

Elliott, Fischer and Rennie (1999) report seven key guidelines that should be adhered to in order that readers can be assured of the integrity of qualitative

research. ‘Owning one’s perspective’, ‘situating the sample’ and ‘grounding in examples’ are covered in the reflexivity, participants and analysis sections, respectively. In terms of ‘credibility checks’, transcripts were sent to all participants to ensure transcript accuracy. ‘Coherence’ (Elliott et al., 1999) is an additional guideline, and this ensures that analysis makes sense and uses an appropriate narrative to inform the reader of key ideas. Reid et al. (2005) also emphasise the importance of analysis as transparent and plausible, and supervision allowed for reflection on ideas as well as discussing potential themes, and both research supervisors audited the codes generated by the researcher. Additionally, the use of a reflective diary allowed the researcher to maintain a close relationship with the data, and the researcher also attended a group of fellow IPA researchers that allowed discussion of themes and reflection of her impact on data analysis. Themes were altered through this process in order that the analysis is more interpretative and less phenomenological in the final stages, and the explanation of interviews to others helped to generate ideas and links between concepts. Elliott et al.’s (1999) remaining guidelines are covered in later sections.

Analysis[1]

Two super-ordinate themes emerged from the analysis, and Table 6 illustrates these. What follows is a narrative account, with each super-ordinate theme explored using the themes comprising it. 'Accomplishing general vs. specific research tasks' is a further guideline suggested by Elliott et al. (1999) to maintain integrity of qualitative research, and as such, the analysis that follows describes general ideas found across participants, as well as those specific to certain participants.

[1] … indicates elision where text not relevant to statement has been removed.
[text] indicates material added by author to explain a point.

Table 6.

Super-ordinate themes and themes.

Super-ordinate theme	**Participants contributing to super-ordinate theme**	**Themes**	**Participants contributing to theme**
Managing identity as a young person with CD	All	Attempts at negotiating difference	All
		Importance of others in supporting CD management	All
		CD as a mechanism to directly boost self-esteem	Claire, Tara, Sarah, Tom
		Learning to integrate CD into the self	All
Ambivalent relationship with food	All	Food as an unavoidable stressor	Claire, Tara, Sarah, Tom
		Food as potentially dangerous	All
		Food as an expected source of enjoyment	Claire, Tara, Sarah, Tom

Super-ordinate theme: Managing identity as a young person with CD

Children and young people's experiences of CD and the GFD appear to impact negatively on their perception of self and identity.

Attempts at negotiating difference

> Umm, really all I thought was my sister's eating different stuff, and my mum/I, like just take dinner, umm, I always like had, like if we have like yorkshires and stuff like that my sister's were always like huge and stuff like that and then mine are tiny. . . I wondered, I just wondered why, why they were different and so practically I knew that something was different about me, umm, like me and my sister and my family *(Claire, line 17)*.

The excerpt from Claire's interview above illustrates that there is a sense of knowing she is different when she was younger and not being quite sure why. She goes on to state that there was something different about her, but also that this affects her sister and her family. Tara also describes knowing that she was different as a result of not being able to eat the same food as others:

> Well like I was younger and I started going to like birthday parties and that, and everyone was just having like chips or burgers and stuff and I wasn't allowed nothing like that *(Tara, line 9)*.

There is a feeling of being isolated from other people her age and she "wasn't allowed" to have the same. There is something that sounds quite punitive in the

way she phrases this sentence. Sarah describes a feeling of being the “odd one out” as a result of CD, and how she left her school as a result of being bullied. Indeed, it is seems as though it is not so much having CD that is difficult, it is the perceptions of other people and their highlighting her difference that she dislikes; this might trigger feelings of shame.

> Well I got bullied quite a lot at this school. . . so I left and they were all the taking the mick out of me and two of my friends and then they were like saying things about it [CD], that’s what I hated and that *(Sarah, line 633).*

All of the children and young people, except Ethan, talked about wanting to fit in with a larger group. As Claire describes below:

> I feel like I’m the, like, I’m I feel, uhh, it’s quite sad actually 'cause I feel like I’m the odd one out. It’s like everyone else is normal and I'm different *(Claire, line 278)*.

> I just think they should like – I sometimes wish that, umm, like people like my mum and my sister and my dad, like, could try, like, like, try and swap diets but that's not able to for me. But I just want to see what it’s like on their side really so. I don't know [laughs] *(Claire, line 338)*.

> It's just when everyone else had them on the playground and I was practically the one that never had any. Like I felt like [whispers] 'gotta be the same, gotta be the same' [laughs], it's just. You just, I don't know why it just urges me, it just goes [laughs, then whispers] 'want it' [laughs] *(Claire, line 405)*.

Claire's description portrays a sense of longing to be part of the "in-group", rather than being a person with CD and therefore different. Again, there is a suggestion of feeling isolated, of injustice in Claire's accounts, and a sense of resignation that she isn't normal and never will be. Tara echoes Claire in her discussion about wanting to be part of a group, rather than standing out by having different foods that are safe to eat.

> Um, I don't know, sometimes it probably felt a bit different, when everybody else was just, could eat the food that was there and I had to go and have my like special food *(Tara, line 33).*

> You feel a bit like left out or whatever 'cos they, they don't really understand what it feels like *(Tara, line 118).*
> We all have like the same [at lunchtime]. Everyone has like potatoes. I suppose it feels quite good really. . . Oh more normal than like standing out by having like different foods all the time *(Tara, line 675).*

Again, there is something in Tara's statements about perceiving herself as abnormal, and that other people cannot understand what it is like to have CD. It does not appear that CD is a painful or distressing condition per se, rather, it is how it can result in feeling different from peers, indeed, feeling that they do not match up to peers. These young people are identifying themselves as abnormal. Sarah talks about her experiences as embarrassing:

> Um, sometimes it's a bit embarrassing. 'Cos like if my friends' new friends there I don't know them and then I've got to tell them and stuff like that (*Sarah, line 564)*.

This is likely to make Sarah stand out from her peers and thus be perceived as different, or perhaps more negatively than that, perhaps being contagious and someone to be avoided. Indeed she goes on to say "I don't really like anyone else to know other than me and my friends". Tom talks about managing his CD, and feeling different, by disregarding the GFD:

> Like it was the 'in' chocolate bar and everyone had them and I thought I'm trying it I don't care [laughs]. That was it *(Tom, line 483)*.

The sense here is that the need to fit in and be one of the crowd is so strong that he has eaten gluten despite usually following a GFD. Other young people, for example, implore the industry to change in order that they can fit in with other young people and be afforded the same choice in food:

> Umm, they have a like a whole aisle of dog’s and cat’s food and then they have like that big of an aisle for me [demonstrates size by spreading arms]. I'm not very happy with that, that aisle ‘cos I don’t, like, it just looks like they care more for dogs and cats, animals than, umm, they do for us and it’s horrible. [Laughs] I just don't like it *(Claire, line 169)*.

> And the food's nice. . . It’s all right but I think like they should put more of a variety of like just - I know it’s hard, ‘cos they have to had/it’s/we’re not the only people in the world, they have to cater for everyone else but we like, we, we are still people so they have to still care for us *(Claire, line 599)*.

In these, Claire perceives the food industry to be reinforcing the feelings of people with CD as abnormal by not providing sufficient gluten-free food choice. She compares the available food for people with CD to that provided for household pets, which she finds upsetting; there appears to be a mental struggle for Claire in terms of trying to find her value when compared to cats and dogs, and a striving not to be forgotten. Claire is aware that there are people other than people with CD to cater for, but there is a sense of not feeling cared for by others and a sense of longing for inclusion. Perhaps this is a feeling of longing to be the same as everyone else so as to reduce embarrassment about having CD, as Tara alludes to:

> Sometimes get a bit like, like, like um, I don’t know like umm, like they [people in restaurants] don’t understand kind of thing and that they think that you can have like loads of these foods but really you can’t so sometimes it’s a bit har-- a bit hard to get through to them like, what it is *(Tara, line 109).*

> Um sometimes feel like a bit um, I don’t know really, I sometimes feel a bit like ‘oh why don’t you understand’ *(Tara, line 116).*

Thus, when other people, whether in restaurants or in the food industry do not appear to cater for children and young people with CD there is a sense of frustration. This appears to add to confusion and perhaps ambivalence about how they fit into wider social contexts. As Tom highlights when out with friends, CD can become a seemingly unwanted focus making the difference more visible:

> It’s amazing how forgettable they [friends] can be. . . They’re all like ‘why [can’t you eat that]’? I’ve got Coeliac, I told you about 8 years and last week and last month and last year. You know they always forget but when I sort of explain they go ‘oh yeah’ *(Tom, line 178)*.

> I just think ‘stupid, remember from the last time I told you’. I just say, as soon as I say Coeliac they all sort of remember me talking about the

> things at the bottom of my stomach. But, they're [friends] all right really *(Tom, line 204)*.

Claire attempts to manage feelings of difference through wishful thinking about the future:

> As I say I was like, umm, I like to think to myself. One day I'll be able to eat the stuff that they'll have, they have, but I just think to myself the day is going to come so I just better not do it [eat gluten now] just in case and I just like I, I said earlier, I'll just turn away and just don't eat it. Don't ever do it *(Claire, line 491)*.

Claire appears to know there is currently no cure for CD, but is convinced a cure will be developed that might mean she can eat gluten.

Furthermore, three young people went on to say that, actually, despite the difficulties managing feeling different, there were times when they didn't feel different or concerned about CD. Interestingly, 2 of these young people were male; perhaps males are more accepting of the disease in general:

> I just went to France in Easter, where I took my own bread and my own pizza bases and stuff, just the bloke cooked it. Give me toast for breakfast, did my dinner differently. That's it (*Ethan, line 244).*

> I felt just the/’cos I’ve been brought up with it… I used to have all this sort of, different food and I was that young and I was constantly brought up with it. It, it never, never sort of had a deep impact on me, ever *(Tom, line 682).*

> I don’t worry about it, I don’t [know] if my mum is. She never seems to, seemed to have/I don’t know really, I didn’t really think it was something to worry about it *(Tara, line 630).*

Is this a coping strategy, denial of impact of CD and GFD, or is this a reflection of the nature of managing identity as a young person with CD, and the transient nature of this? Given other statements made by young people whereby CD does cause them to be different, it is possible that actually, these young people are in the process of exploring and developing their identity, within the constraints of a chronic illness. Ultimately, children and young people want to fit in with others, but they also want to be well and free from disease. As Claire describes:

> All I know a couple of days ago my mum told me I'm, I’m more prone to osteopro/osteoporosis, stuff like that in my bones, I'm really scared now [laughs]. ‘Cos I get scared, just like I don't want to really, like that. It’s just horrible *(Claire, line 146)*.

> Umm, and I just realised when I got told that that it’s really dangerous and I could have like a threatening life when I was older like frail bones. So, I've really tried to keep to my diet, so *(Claire, line 391)*.

Importance of others in supporting CD management

As well as trying to negotiate feeling different, children and young people took value from interactions with other people who they perceived to understand what they were going through. All young people talked about having understanding friends:

> They/they don’t really say anything ‘cos they’ve all, since they’ve knew me, they’ve already, they know like what I can and I can’t eat or nothing, so they know like I couldn’t have nothing from there or anything like that *(Tara, line 285).*

> Whenever I go to their houses, they’re always like ‘oh I’ve got some stuff you can have’, ‘cos they sort of, they basically know what I can have, like chocolate, plain crisps and stuff like that. So, they always say ‘we got some of that stuff’. I just eat that (*Tom, line 347).*

One young person, Sarah, also perceived her relative with CD as someone who would be concerned for her; Sarah was the only young person with a relative with CD:

> Um, probably [family member] who’s got Coeliacs [worries most]. . . ‘Cos it’s/’cos I think she gets more ill than I do and she thinks I’m gonna get as ill as she does *(Sarah, line 512)*.

This is interesting, and is definitely something peculiar to someone with a relative or perhaps close friend, with CD. This might be reassuring to Sarah, that there is someone older, and with more responsibility and personal experience of CD, who can pave the way for Sarah in advising her in managing the GFD when older. Perhaps it relieves the burden of having CD slightly, as she is not a lone family member with CD. Similarly, given friends understood, this might make it easier for young people to worry about being young people, rather than a person with CD. Indeed, in addition to friends’ understanding, a massive boost to self-confidence generally occurred when others tried the GFD:

> My/my dad has gone on a gluten free diet. Completely, because it makes him tired or something. Wheat makes him, it makes him feel tired or something like that. . . But my dad’s now on a complete gluten free diet. It’s extra good because my dad always sort of accompanies mum pretty much going shopping and he gets to choose all the nice gluten free stuff! So I get loads more [laughs]. ‘Cos he gets all the, he gets all the/he’s got a really bad sweet tooth as well, so he always gets all the chocolate and the gluten free biscuits and like ‘oh great!’ [Laughs] *(Tom, line 616).*

> Well, um, I found it like nice ‘cause they, they never usually but sometimes occasionally when like sausages, like Hannah my sister used to really like them, so she always has them and like my chicken nuggets and stuff like that [laughs] *(Claire, line 640).*

> My stepdad’s mum, I think she went on it for a, a bit as well and he said it helped her like stomach and that and I thought it must not, the food, the food that I’m not, that I’m not eating mustn’t be that nice for people just to give it up like out of choice *(Tara, line 493) .*

> Um, well apparently all my friends have said like cakes are nicer than theirs are. Yeah, and that’s a good thing. And apparently my food tastes exactly the same as theirs [laughs]. . . Uh, good, ‘cos then I know that I’m not missing out on anything *(Sarah, line 487).*

Indeed, this validation seems important, the acceptance of other people as crucial to feeling supported. Also, the fact that people have the choice and are choosing to be the same as them feels satisfying and might help to alleviate some of the confusion that young people feel with regards having CD. But perhaps the most important person in supporting efforts to develop identity is mum. All participants reported the importance of mum as “chief checker”, “safety net”, “fountain of knowledge”, “the one who does all the shopping” and the person who will try to reduce the ‘visibility’ of the disease, in terms of eating different food:

Well, in, in the house, I’m, my mum or I ‘cos like most of the time my mum like tries to do the same so I don't feel any different but like sometimes, like Yorkshires say, I still have some but I can just tell they’re different *(Claire, line 294).*

I say can I have some of that she says no, you can't have that and then I ask her why, and she tells me ‘cos like wheat and gluten, and she lets me have a look at the packet, like stuff like that. I can just tell she really cares for me *(Claire, line 659).*

My mum buys all the food but she knows what I can have and that and like sometimes she picks up, she buys a lot of stuff from the Coeliac, um gluten-free range but most of the food I don’t eat it, I don’t really like it so my mum just buys like what I have *(Tara, line 123).*

‘Cos um, they’re always checking everything after I’ve checked it and I’m like ‘I've already checked it’ and then they double check it. . . Just after they read it they say I can have that (*Sarah, line 531).*

My mum [does it all] *(Ethan, line 36).*

My mum does [prescriptions]; bread and pizza bases and biscuits and stuff like that *(Ethan, line 65)*.

> ‘Cos she [mum] sort of, all the Coeliac books and everything are in her name even though I’ve got the CD. So yeah, she shares all the Crossed Grain and she gets the little book that to tell me what I can and can’t eat in the supermarkets and stuff like that *(Tom, line 145).*

Mum seems to be a trusted facilitator to dietary management and in doing so allows the young person to get on with being a young person; for all young people here it is seen as the norm for mum to be involved. This might be because young people are aware that others might not understand, but that mum will be a safe base and has a duty to care for them. In talking about mum and her role, Tara states:

> Feel like a, bit like a baby, asking your mum if you’re like allowed this food or whatever but got, got used to it really *(Tara, line 166)*.

Gatherings of other Coeliacs can also serve to validate one’s sense of self:

> When I was there it was really fun. The atmosphere was like ‘ah!’. . . Um, because there was loads of other Coeliacs and I wasn’t sort of, not that I usually feel singled out, but there, there was no sense of that. There was like, it was the first time I’d ever gone somewhere and I could eat everything. ‘Cos everything was Coeliac, um, gluten free and I was just like, ‘score!’ [Laughs] Biscuits and pizzas and I was like ‘yay’! Made me really chuffed [laughs] *(Tom, line 358).*

> Yes [the parties for Coeliacs were good], we used to do stuff, like Coeliac stuff that you could take home and that *(Ethan, line 120).*

Furthermore, the annual checks also received a mention, but only from Claire. This might be because she was one of the youngest interviewed, and as such, perceived this not to be a threat to her autonomy and identity as much as someone who was perhaps a little older:

> Yeah, like weighed [inaudible] and stuff like that yeah. Yeah, umm, it's helpful like, 'cause like that, umm, and they show me how I've progressed, 'cos at the moment I've gone like that [indicates upward movement with hands] I'm doing really well *(Claire, line 578)*.

CD as a mechanism to directly boost self-esteem

There also appears to be a positive connotation associated with having CD:

> And they usually cater for me always. Like at lunch, umm, our dinner lady she does special meals for me only *(Claire, line 518).*

> Say sometimes if like family members see something in like the shop that they see is gluten-free and they haven't saw that I've had it, they buy me and that for me to try. It's quite nice [laughs]. . . People are buying me food *(Tara, line 507)!*

> Sometimes you get like, special food made for you that others, like, wouldn't have or like people that buy you things that normally they wouldn't, sort of, stuff like that *(Tara, line 589).*

> Um, suppose it feels quite good really you know, like people are like buying things specially for you and that *(Tara, line 601).*

> Um, well one of my friend's mum's, um, she cooks gluten-free food whenever I go over [laughs]. [feels] good. . . Um, just the thought that they'd gone through so much stuff *(Sarah, line 265).*

> We went to this bar and they're looking at the menu and then there was sort of like loads of these meals and we're like is there wheat in this, is there wheat in this, is there wheat in? And the [inaudible] comes back and it's yes, yes, yes, there's wheat in pretty much everything. And the chef actually made a completely new meal just so I could eat it… Yeah, I feel really special. Because, 'cos you sort of, it's rare, you don't expect that to happen. You know, if you're a normal person you wouldn't say 'cook me something completely different I don't like anything on this menu'. They'd just be like, 'get out' *(Tom, line 222).*

Young people report direct benefits of having CD, and report they receive special treatment, and in some cases are treated better than if 'normal'. This feeling might be heightened as young people strive to fit in and find some

benefits of having CD, and to try and make sense of how CD fits with them and their family. However, a significant amount of what was talked about was the integration of CD into the self.

Learning to integrate CD into self

All young people report feeling special, and in some cases, being treated better than a “normal person” (Tom, above). Indeed, these mixed reports of what it’s like to grow up with CD appear to have developed over time, and although young people report feeling special, there is also a sense of resignation to having CD:

> But when I go out, I know really most of the food that I can have now and what I can’t so I just stick to that really *(Tara, line 95).*

> Then sometimes if I’m like going to a new place or whatever I just ask them and if I can’t have it then I just stick to what I’ve already eaten from that *(Tara, line 127).*

> But now I can sort of, now I sort of wised up, I know what I can have on the menu anyway *(Tom, line 218).*

There is a sense here of adjusting to managing the diet, and of having some control over doing this to some extent. Ethan, Sarah and Tara all share in the idea that there are times when they don’t feel like they are missing out, being on

the GFD, and as such have learned to rationalise their feelings so as to attempt to integrate GFD into their lives. Two excerpts from Tara succinctly illustrate this point:

> Mum bought me like these fish fingers and they were di-- they’re disgusting and she says that they taste near enough the same to what other fish fingers do so sometimes I feel like I’m not really missing out on food anyway ‘cos like with the, the gluten-free cakes and that in the shop and they make, they make me sick even though I’m allowed them ‘cos I hate the look of like shortbread, it like makes me feel sick so sometimes I don’t really feel left out or nothing because some of the food that I can have I don’t like anyway *(Tara, line 137)*.

> For him to say he was, like, leaving out [gluten] so it must not be that nice if people are choosing to not eat it *(Tara, line 490).*

And Ethan states:

> Not really [want things to be different], ‘cos I don't feel any different *(Ethan, line 86).*

Overall, there seems to be two sides to having CD, and both seem to impact on how the young person perceives himself or herself. It remains to be seen whether these two faces of CD will remain as young people get older. However,

what appears clear is that young people seem to have both positive and negative ideas about CD and its management, and its impact on them. This appears to be context dependent; when out with friends tends to be a time of high, unwanted, disease visibility. Indeed, young people’s relationship with food seems to be important, and as such, warrants a theme of its own.

Super-ordinate theme: Ambivalent relationship with food

Food as dangerous

Young people spoke about food as a potential danger to themselves, and there was a sense of feeling threatened and having to be hyper-vigilant around food. As such, young people have learned what they can tolerate before they are sick:

> Umm, some things I’m all right with where it just says traces of it or may contain; I still eat that, like Hula Hoops it says ‘may contain traces of’ whatever or produced on a line - I still eat them *(Tara, line 301).*

And Ethan reports “if I ate something different then I'd be sick” *(Ethan, line 144).* There is a sense of only eating safe foods as Tom describes:

> I just knew it made me ill at that point, ‘cos I was only like 4 or 5 so it’s/I just sort of thought you know, it just makes me ill, don’t do it *(Tom, line 101).*

He goes on to talk a lot about needing to "protect" his food in case someone else eats it, and going hungry because nothing else was safe enough to eat:

> So, in that case, in that sense, I had to sort of look after my food 'cos no-one at that party, you have/didn't think/eat whatever's there. So I had to sort of be, sort of up, conscious of other people sort of around my stuff and sort of like or hide it somewhere so I can just sort of get it out when I needed to. . . It didn't really come with like deep emotional depression [having no 'safe' food to eat at a party] or anything but I did sort of think 'ok, I'm different from all these people and I gotta look after my food otherwise I'm just going to end up hungry' [laughs] *(Tom, line 56)*.

Rather than having to protect food, Claire and Sarah completely go without certain foods if they cannot ascertain gluten content. There is a feeling of foreboding if they were to have food that possibly contained gluten, and so these young people tended to err on the side of caution:

> I know I can have it on the conveyor belt but I don't just in case [it's] contaminated 'cos she sometimes fish and stuff like that I can't have into them, so it's/she just makes stuff separately *(Claire, line 532)*.

> Um, sometimes when they like 'oh do you want some lips [unlabelled sweets]' and it's like I don't know if I'm allowed it and there's no ingredients on it. So I don't have any *(Sarah, line 378).*

Food as a potential danger is confirmed when young people eat gluten and then have to suffer the consequences. This is often followed by regrets and distress:

> Um I just hate being like; it’s like, you get a really horrible like stomach ache before and that’s worse than being sick. So, it’s not nice. It’s like um, it’s like the feeling of being sick but not actually being sick and you have like really bad pains in the stomach before I’m sick *(Tara, line 362).*

> Like sometimes like when I’m actually being sick I wish if only I was to look; like my mum says is it really worth having a couple of chips to be sick after? *(Tara, line 391)*

> I was thinking I really don’t want to be sick. I had a really bad stomach pain. Um, just worried really [about] being sick *(Sarah, line 186).*

There appears to be a marked concern around the consequences of ingesting gluten, and there is some ambivalence around wanting certain foods but not wanting to experience the consequences of eating it if they choose to. This is a great emotional demand, and given that one has a fundamental need for food, this can be incredibly taxing.

Food as an unavoidable stressor

All participants except Ethan talked about feeling tempted to eat gluten containing foods. Claire in particular talked about this:

> It’s like/I have like, it’s not like, like have you ever heard of the saying of the devil, of a devil? A devil talking in your mind, speaking to me? Uhh, I don’t know but I feel like something going ‘do it, do it, like eat, eat it’ [laughing]! It’s just like, I don't know what that it is, I think it’s, I think it’s something like the devil but it’s not the devil *(Claire, line 443)*.

Here, the use of a powerful analogy of the devil serves to illustrate how difficult Claire finds it to manage being around gluten containing food; the devil is bad and is in her head; does this mean that she feels she is a bad person? There is a sense of an intense mental battle: should I eat it or should I not? For Claire, even seeing gluten containing foods invokes strong emotion:

> But when I go shopping with my mum I look at stuff and I’m like ‘damn wheatos’, just would want some but I can’t have them *(Claire, line 161)*.

Sarah talks about having to manage situations where her friends offer her gluten-containing food, and again, there is a sense of an internal battle, that is not voiced to friends or family. Instead, young people appear to manage seemingly alone, and perhaps feel ashamed and isolated in doing so.

> Um, they like ‘oh do you want this, do you want some of my biscuit?’ One of my friends Faith did last year, and I was like ‘no I can’t finish it’, ‘oh go

> on’, ‘no I can’t be ill’ and then she went ‘oh yeah, I’d forgotten about that’ [laughs]. . . I get, I get thinking ‘stop it!’ [laughs] *(Sarah, line 682)*.

However, interactions with food are unavoidable, and as such, there is a sense of having to “make do” with any food on offer. Tom talks about a recent experience on holiday. He appears to use humour to manage having repetitive meals:

> I did want to try something else. You know, I wanted to eat something else [laughs], anything else, anything. But I couldn’t, I had chicken and chips for 2 weeks [laughs] *(Tom, line 669).*

He also reports feeling upset when he can’t eat gluten:

> ‘Cos there was like sausage rolls and you know the sausages on the sticks and stuff like that, so all the typical party food and the vast majority of that I can’t eat so when they ate my pizza I was like [sighs and makes crying noise]… *(Tom, line 36).*

Claire, having been caught eating gluten comments:

> They had these cakes on, umm, the, the stand and weren't gluten free and I was buying them and I ha/ate them and I did get real badly told off but now I know not to eat it… [on being caught cheating] And then they

> took me into a different room and just said I'm really disappointed in you, so [laughs]. Like, it was just horrible [laughs] *(Claire, line 273)*

And so there is a psychological cost for having actually consumed gluten, and Claire appears to have been very shaken by this experience. Claire reports that “they” were “disappointed” that she had eaten gluten, and her experimentation with gluten has led to a significant amount of distress, and perhaps even shame. It is not clear who exactly “they” are. However Sarah reports some element of distress from just thinking about eating gluten:

> Sometimes do [feel tempted], but then I know I shouldn’t. . . Um, like when it’s someone’s birthday and I can’t have a bit of cake [laughs] and stuff like that, but I know I shouldn’t. . . I never would *(Sarah, line 472).*

Food continues to be a stressor and a source of anxiety even when gluten containing food has been eaten, and young people seemed keen to justify why they had eaten gluten:

> I’ve ate like one packet of them once [laughs] *(Claire, line 476)*.

> It’s not like I eat the whole thing, I wouldn’t purposely go and buy a bag of chips from the chip shop, but if my friend was eating them, I’d like keep having picks out of hers. . . I wouldn’t go and buy something, it’s not like

> I'm buying it but if someone/ it was there, if someone was eating it I'd probably have a few of theirs *(Tara, line 312).*

> Like if I'm just hungry, and like say like if your friend was just eating something, you'd probably like ooh I'll have/[inaudible] on her crisps or something like that, it's just like that really *(Tara, line 328).*

> I did when I was younger. I had to admit Kinder Bueno, they were nice. The Kinder Bueno bars, the snappy off ones with the cream inside. Ohhh [laughs]. My friend used to buy them all the time and he used to give me one chunk. My treat [laughs] *(Tom, line 409).*

Food as a source of anxiety appears to be common here, and there appears to be a pre-occupation with food. This is apparent in ideas about healthy body weights for Tara, Sarah and Tom. This is not necessarily a surprising result, given the daily focus on food and what they are eating. Sarah, when asked about the annual checks comments:

> They just write it down [at annual checks] and if I was overweight they would have told me and stuff like that *(Sarah, line 448)*.

Despite not being asked about her weight, this is the first thing that Sarah comments. In this sense, Tom sees the GFD diet as advantageous to maintaining his weight:

> I don’t get fat [laughs]. I can’t think of any other positives [of being on the GFD] [laughs]. ‘Cos wheat’s really starchy and you, and it is quite fattening. And cos’ I’m not I’m just like bring on the chocolate *(Tom, line 509)*.

Food as expected source of enjoyment

One of the things that might be causing ambivalence about food is the anticipated benefits of eating it. Food can play a massive part of socialising with others, and young people talked about having to make do:

> I can’t have like baltis or nothing and I just have to have like a omelette and sometimes I end up eating food that I’m not really that keen on just because there’s nothing else to eat *(Tara, line 426)*.

Thus, rather than miss out on social events, young people felt that they had to eat food because it was the only food available. There is a sense of food being unavoidable, but also being a chore to have to manage. Both Tara and Tom want to enjoy food but feel restricted. Tara states:

> I’d like sometimes just to be able to go up and have, try everything *(Tara, line 241)*.

And Tom states:

> [After eating gluten] So I sort of felt as though, as if I wasn’t [Coeliac] which sort of gave a little glimmer of hope that I could go to Pizza Hut and have a massive pizza and just eat it and be fat *(Tom, line 454)*.

Claire also feels envious of “normal” food:

> It’s not, not nice being like, being able to see them, because eat them or anything, umm, because they didn't do croissants, uh, when, umm, when I had, when I tried it they didn't do them and then they came in and I bought some and then they, they didn't taste the same. They/it’s just got a real bad taste to them *(Claire, line 253).*

> It, umm, it's a very good diet but you just like [laughs], just want to, just want to do stuff you can't do [laughs] *(Claire, line 569).*

There is a sad image of a child feeling torn between having to maintain the GFD and then wanting to eat food as everyone else does. However, there is a suggestion of a developmental process, whereby young people have adhered to the GFD and then begin to push boundaries and test out the limits of their illness. This “cheating” allows the young person to experience what other young people their age will also be experiencing. In this sense, this allows young people to develop a sense of autonomy and to know what they can and can’t do

reasonably to maintain their health. Indeed, it might also lead to young people making an informed decision about how they want to manage CD and the GFD as an adult having “experimented” in adolescence.

Each of the young people, in some way, appears to want to be well and healthy but feels tempted to eat gluten. This mainly happens when outside the home. These occasions tend to be with friends, whether at school or out socialising with friends. Young people appear to have adapted to this by eating some gluten, but not generally enough to become unwell, although the memory of being ill after eating gluten remains.

Discussion

The young people in this study report conflicting feelings associated with having CD as a young person. Their ideas about how they fit in with peers appear to be restricted by CD and young people appear not to suffer major symptoms from CD, but instead feel embarrassed, ashamed, angry and isolated as a result of following the GFD, a visible manifestation of their illness. This was typically only reported when in social situations outside home and generally with friends. Indeed, parents, particularly mothers, appear to play a protective role in helping young people to manage CD and manage feelings of difference, although the success of this varied.

Like participants reported in Olsson et al. (2008), participants here report most difficulties managing the GFD when outside the home; this was likely to lead to occasional consumption of gluten containing food. Also, young people wanted to ‘fit in’ and the GFD was a barrier to this. Cinquetti and colleagues (1999) highlighted ambivalence around food for young people with CD aged 10-21 years, and this was also found here, in a small group of 10-16 year olds. What these findings potentially highlight is the importance of food in Western culture, and that many interactions occur in the presence of food. Thus, for young people that cannot eat all foods, these interactions might become problematic. Recently, DunnGalvin, Gaffney, and Hourihane (2009) sampled 62 young people aged 6-15 years to more fully understand issues associated with food allergy across early- to mid-stage childhood. They completed focus groups and,

using Grounded Theory, report six themes: what food means; autonomy, control and self-efficacy; peer relationships; risk and safety; identity; and active and interactive strategies used to manage growing up with a food allergy. Similar to DunnGalvin et al. (2009) where participants report an impact on identity, either being special or being different, participants in this study also report both difference and feeling special. Food allergy is qualitatively different from CD in the sense that food allergy can result in acute anaphylactic reactions which can be life threatening, but their common feature is the requirement to avoid certain foods. In both DunnGalvin et al.’s (2009) study and that reported here, participants reported isolation and being different from friends. A striking similarity between the two studies was the reliance on parents, namely mothers, who help to manage the condition. Similar to DunnGalvin et al. (2009), management of CD appears to be a “cumulative history of interactive processes… that are embedded in a child’s developmental organisation” (DunnGalvin et al., 2009, p. 565). Young people try to both integrate and reject the ‘CD identity’, and try to avoid gluten foods as well as engage in behaviour that could be perceived to be risky, in terms of eating some gluten containing foods; this is equivalent to the model proposed by DunnGalvin et al. (2009).

One of the key tasks for young people in this study appears to be integrating CD into their identity as they experience adolescence, which is an important time in informing future health related behaviours (Holmbeck, 2002). Lazarus and Folkman’s (1984) model of managing stress can be used to understand how young people manage with a GFD and then attempt to cope with it. This

assumes that CD (and the GFD) is a stressor, and given how young people in this study report hyper-vigilance around food, it could be seen as a demanding and emotionally-consuming task to manage a GFD. Thus, this model proposes that people will engage in primary appraisal of a situation, that is, its significance to them, and the secondary appraisal then follows; this is an appraisal of resources they perceive themselves to own that might help them to manage, as well as their perception of controllability. It could be argued that young people in this study attempted to manage CD and the GFD through what Lazarus and Folkman (1984) term emotion focused coping (reducing emotional impact), in that they perceived themselves sometimes as special because of having CD, and compared themselves to others and sometimes did not feel they were missing out. However, given the concerns that young people raised around being upset when they perceived themselves to have missed out or be different to peers, it could be hypothesised that young people felt they did not have the resources to manage this independently at the current time.

The importance of other people in helping to manage CD and, in particular, the GFD, fits well with the principles of Bandura’s (1977) concept of self-efficacy, specifically the principles of modelling and social persuasions. Indeed, young people in this study alluded to how socialising with other people with CD, and having peers that understood CD and the GFD was helpful in managing the disease burden; mothers in particular were seen as main carers in terms of CD. Using Bandura’s (1977) ideas, it could be hypothesised that having friends and family that understood the GFD and encouraged adherence increased the

child’s own self-efficacy beliefs about being able to manage the GFD. Indeed children appeared to have some level of efficacy in managing the diet, but the aim would be to develop this as they mature.

Western cultures typically eat a large proportion of wheat products (Mearin, 2007) and eating out is increasing (Coeliac UK, 2009). These factors combined might make eating out difficult for people with CD. Indeed, young people reported greatest difficulties when in the context of socializing outside home. However, the importance of food will not change and despite publication of guides for provision of food in public places (for example, The provision of allergen information for non pre-packed foods, Food Standards Agency, 2008) this remains voluntary and as such variation in foods available when out socially will remain. This is an issue that needs to be addressed given the distress this appears to cause young people. Additionally, cognitive interventions to help young people manage the GFD might be helpful; Holmbeck (2002) states that cognitive changes in adolescence might inform new thinking patterns and ideas about adherence, for example, and so result in changes in adherence and illness management.

Limitations of study and research implications

It had not been possible to receive feedback from participants at the time of writing, and as such, this useful way to triangulate information has been missed; the final guideline ‘Resonating with readers’ from Elliott et al., 1999 has not been fully operationalised. Although members of the research team and peers

were used as part of the credibility check, there was no interaction with children and young people (whether original sample or not) to validate themes, and ensure people’s stories have been plausibly explained. It is also important to note that participants interviewed in this study were of a white, middle class background. This might have resulted from a bias in selecting from local Coeliac support groups, or that other people from different ethnic and perhaps religious backgrounds do not take part in these groups, or did not want to complete the research.

The results from qualitative studies are not intended for generalising ideas, but instead provide a platform for future research and allow in depth analysis of people’s experiences (Brocki & Wearden, 2006). It would be interesting to understand how the ideas of children and young people fit with those of their parents or main caregivers. Anson et al. (1990) and Jackson et al. (1985) both report the importance of parents’ support and understanding of the GFD in helping children manage the GFD. Family coherence is important in adjustment to CD (Huff, 1997), and comparison of parent and child accounts might shed light on wider systemic issues of having CD. Also, completion of longitudinal studies focusing on early childhood to early adulthood would be key in highlighting developmental changes specific to CD. Indeed, Dovey-Pearce and colleagues (2007) highlighted that young adult development can be influenced by having diabetes, in terms of developing identity and autonomy, but that this period of growing up is, as yet, neglected in the literature. Holmbeck (2002) agrees, and states that studies of adolescent illness and health should be more

developmentally informed with focus on key developmental milestones. Qualitative studies are noticeably absent in the area of childhood CD and it is possible that this study can be replicated with other children and young people from other backgrounds and in other areas of the country, and internationally.

Greater consideration of theoretical concepts highlighted in the results here will be imperative in developing this area also, and to help researchers and professionals alike in making sense of experiences. Perhaps once more is understood about the experiences of children and young people it will be possible to usefully conduct larger scale studies of well being and adjustment using theoretical concepts.

Clinical implications

However, clinically this study might have implications for health professionals, voluntary support groups and families alike in helping young people with CD to adjust to having CD and managing the GFD in adolescence. Furthermore, it might contribute to understanding of adherence and non-adherence in young people, and contextualise such non adherence as a struggle to develop their own identity, as well as manage the daily stresses that food might bring. Annual checks young people have to attend each year could usefully incorporate a psychological component to assess psychological wellbeing and management of the diet in terms of the negative feelings reported here. Allowing young people a protected space to air some of their concerns might increase adherence and understanding of the GFD. Indeed, young people know what

food items they should and shouldn’t eat, but operationalising this seems to cause some distress. Thus, educational interventions might not be as useful as mindfulness approaches. Sawyer and Aroni (2005) call for doctors to consider psychosocial issues associated with chronic illnesses and to respect the young person and their views, and go on to say that this will facilitate effective self-management, which is important in ensuring adherence. Local Coeliac support groups could offer more age appropriate activities for young people with CD, and to encourage more discussions around managing and coping with CD in an environment where most people might not have it (e.g. at school). Similarly, Dovey-Pearce et al. (2007) suggests that the health system for managing diabetes in adolescents and young adults needs to be developed to accommodate the challenges that this group face in terms of growing up with a chronic illness; older adolescents are neither children nor adults. Given young people’s experiences reported here this could be reasonably taken forward, taking into account developmental changes this group encounter.

Reflexivity

In carrying out this study, it reminded me of some of the challenges I faced when I was growing up, and how important I also felt it was to fit in with my peer group. I feel fortunate, and also humbled, that I grew up with a relatively stress free adolescence compared to some of the experiences young people have talked about here. These young people have also helped me to cope with living without dairy and yeast, and to be pro-active in my approach to managing it. Their stories of dealing with tricky situations from such a young age have given

me hope and reassurance that things will be ok. I feel really passionate about helping young people with CD as a result of doing this study, and have decided to maintain involvement in the project after completion of the work reported here. This was somewhat unexpected for me. However, I think it is important to support children and young people through adolescence no matter what, but particularly when this time is complicated by illness.

Conclusions

The study here aimed to begin to explore the experiences of young people with CD, Indeed, findings reported here suggest there are some struggles with identity and with food relationships experienced by young people with CD, and additional studies are needed, both qualitative and quantitative in nature, so that these concepts can be explored sufficiently. Psychological wellbeing as an over-arching concept might be useful to pursue also, but more specifically young people’s methods of coping might be useful to establish, using the theoretical frameworks suggested here, and the qualitative findings reported. However, clinically, there are useful supports that professionals might considering implementation of now, including altering the content of annual checks, and providing age appropriate activities at local support groups, that help young people foster autonomy over CD management.

References

Addolorato, G., Leggio, L., D’Angelo, C., Mirijello, A., Ferrulli, A., Cardone, S., Vonghia, L., Abenavoli, L., Leso, V., Nesci, A., Piano, S., Capristo, E., & Gasbarrini, G. (2008). Affective and psychiatric disorders in Celiac Disease. *Digestive Diseases, 26,* 140-148.

Anson, O., Weizman, Z., & Zeevi, N. (1990). Celiac Disease: Parental knowledge and attitude of dietary compliance. *Pediatrics, 85,* 98-103.

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioural change. *Psychological Review, 84,* 191-215.

Bingley, P. J., Williams, A. J. K., Norcross, A. J., Unsworth, D. J., Lock, R. J., Ness, A. R., & Jones, R. W. (2004). Undiagnosed Coeliac Disease at age 7: population based prospective birth cohort study. *British Medical Journal, 328,* 322-323.

Booth, I. (1991). The nutritional consequences of gastrointestinal disease in adolescence. *Acta Paediatrica Scandinavica Supplement, 373,* 91-102.

British Society of Gastroenterologists. (2002). *Guidelines for the management of patients with Coeliac Disease.* UK: British Society of Gastroenterologists.

Brocki, J. M., & Wearden A. J. (2006). A critical evaluation of the use of interpretative phenomenological analysis (IPA) in health psychology. *Psychology and Health, 21,* 87-108.

Ciacci, C., De Rosa, A., de Michele, G., Savino, G., Squillante, A., Iovino, P., Sabbatini, F., & Mazzacca, G. (1998). Sexual behaviour in untreated and

treated Celiac patients. *European Journal of Gastroenterology & Hepatology, 10,* 649-651,

Ciacci, C., Iovino, P., Amoruso, D., Siniscalchi, M., Tortora, R., Di Gilio, A., Fusco, M., & Mazzacca, G. (2005). Grown-up Coeliac children: The effects of only a few years on a gluten-free diet in childhood. *Alimentary Pharmacology & Therapeutics, 21,* 421-429.

Cinquetti, M., Trabucchi, C., Menegazzi, N., Comucci, A., Bressan, F., & Zoppi, G. (1999). Psychological problems connected to the dietary restrictions in the adolescent with Coeliac Disease. *Medical and Surgical Pediatrics, 21,* 279-283.

Coeliac UK. (2009). Retrieved 26 May 2009, from www.coeliac.org.uk.

Cranney, A., Zarkadas, M., Graham, I. D., Butzner, J. D., Rashid, M., Warren, R., Molloy, M., Case, S., Burrows, V., & Switzer, C. (2007). The Canadian Celiac health survey. *Digestive Diseases and Sciences, 52,* 1087-1095.

Dovey-Pearce, G., Doherty, Y., & May, C. (2007). The influence of diabetes upon adolescent and young adult development: A qualitative study. *British Journal of Health Psychology, 12,* 75-91.

DunnGalvin, A., Gaffney, A., & Hourihane, J. O’B. (2009). Developmental pathways in food allergy: A new theoretical framework. *Allergy, 64,* 560-568.

Elliott, R., Fischer, C. T., & Rennie, D. L. (1999). Evolving guidelines for publication of qualitative research studies in psychology and related fields. *British Journal of Clinical Psychology, 38,* 215-229.

Food Standards Agency. (2008). *The provision of allergen information for non pre-packed foods: Voluntary best practice guidance.* UK: Food Standards Agency.

Green, P. H. R., Stavropoulos, S. N., Panagi, S. G., Goldstein, S. L., Mcmahon, D. J., Absan, H., & Neugut, A. I. (2001). Characteristics of adult Celiac Disease in the USA: Results of a national survey. *The American Journal of Gastroenterology, 96,* 126-131.

Hauser, W., Stallmach, A., Caspary, W. F., & Stein, J. (2007).Predictors of reduced health-related quality of life in adults with Coeliac Disease. *Alimentary Pharmacology & Therapeutics, 25,* 569-578.

Hill, I. D., Dirks, M. H., Liptak, G. S., Colletti, R. B., & Fasano, A. (2005). Guideline for the diagnosis and treatment of Coeliac Disease in children: Recommendations of the North American Society for Pediatric gastroenterology, Hepatology and Nutrition. *Journal of Pediatric Gastroenterology and Nutrition, 40,* 1-19.

Hogberg, L., Nordwall, M., & Stenhammer, L. (2001). Small bowel capsule biopsy in children: Parents' opinions on children's discomfort. *Acta Paediatrica, 90,* 876-878.

Holmbeck, G. N. (2002). A developmental perspective on adolescent health and illness: An introduction to the special issues. *Journal of Pediatric Psychology, 27,* 409-415.

Huff, C. (1997). Celiac Disease: Helping families adapt. *Gastroenterology Nursing, 20,* 79-81.

Jackson, P. T., Glasgow, J. F. T., & Thom, R. (1985). Parents' understanding of Coeliac Disease and diet. *Archives of Disease in Childhood, 60,* 672-674.

Karwautz, A., Wagner, G., Berger, G., Sinnreich, U., Grylli, V., & Huber, W. (2008). Eating pathology in adolescents with Celiac Disease. *Psychosomatics, 49,* 399-406.

Koopman, H. M., Baars, R. M., & Mearin, L. M. (2003). Health related quality of life (HRQOL) in children with Coeliac Disease. *Coeliac Disease: Proceedings of the Xth International Symposium on CD, 10th symposium,* 291-294.

Krahn, G. L., & Putnam, M. (2003). Qualitative methods in psychological research. In M.C. Roberts & S. S. Ilardi [Eds]. *Handbook of research methods in Clinical Psychology* (pp. 176-195). Oxford, UK: Blackwell Publishing Ltd.

Lazarus, R. S., & Folkman, S. (1984). *Stress, Appraisal and Coping.* New York: Springer.

Lee, A., & Newman, J. M. (2003). Celiac diet: Its impact on quality of life. *Journal of the American Dietetic Association, 103,* 1533-1535.

Ljungman, G., & Myrdal, U. (1993). Compliance in teenagers with Coeliac Disease: A Swedish follow-up study. *Acta Paediatrica, 82,* 235-238.

McGough, N., & Cummings, J. H. (2005). Coeliac Disease: A diverse clinical syndrome caused by an intolerance of wheat, barley and rye. *Proceedings of the Nutrition Society, 64,* 434-450.

Mearin, L. M. (2007). Celiac Disease among children and adolescents. *Current problems in Pediatric and Adolescent Health Care, 37,* 86-105.

Olsson, C., Hornell, A., Ivarsson, A., & Sydner, Y. M. (2008). The everyday life of adolescent Coeliacs: Issues of importance for compliance with the gluten-free diet. *Journal of Human Nutrition and Dietetics, 21,* 359-367.

Osborn, J. (1994). Some similarities and differences among phenomenological and other methods of psychological qualitative research. *Canadian Psychology, 35,* 167-189.

Pietzak, M. M. (2005). Follow-up patients with Celiac Disease: Achieving compliance with treatment. *Gastroenterology, 128,* 135-141.

Pynnonen, P. A., Isometsa, E. T., Aronen, E. T., Verkasalo, M. A., Savilahti, E., & Aalberg, V. A. (2004). Mental disorders in adolescents with Celiac Disease. *Psychosomatics, 45,* 325-335.

Rashid, M., Cranney, A., Zarkadas, M., Graham, I. D., Switzer, C., Case, S., Molloy, M., Warren, R., Burrows, V., & Butzner, J. D. (2005). Celiac Disease: Evaluation of the diagnosis and dietary compliance in Canadian children. *Pediatrics, 116,* e754-e759.

Reid, K., Flowers, P., & Larkin, M. (2005). Exploring lived experience. *The Psychologist, 18,* 20-23.

Roos, S., Karner, A., & Hallert, C. (2006). Psychological well-being of adult Coeliac patients treated for 10 years. *Digestive and Liver Disease, 38,* 177-182.

Sawyer, S. M. & Aroni, R. A. (2005). Self-management in adolescents with chronic illness. What does it mean and how can it be achieved? *Medical Journal of Australia, 183,* 405-409.

Skoglosa, J., Falth-Magnusson, K., & Stenhammer, L. (2003). Conscious or deep sedation: A questionnaire regarding the experience of parents, children and staff during small bowel biopsy. *Acta Paediatrica, 92,* 704-708.

Smith, J. A., & Osborn, M. (2003). Interpretative Phenomenological Analysis. In Smith J. A. [Ed]. *Qualitative psychology: A practical guide to research methods* (pp. 51-80). London: SAGE Publications.

Smith, S. (1996). Jejunal biopsy – Seeking an alternative. *Paediatric Nursing, 8,* 17-19.

Sverker, A., Hensing, G., & Hallert, C. (2005). "Controlled by food" – lived experiences of Coeliac Disease. *Journal of Human Nutrition and Diet, 18,* 171-180.

Sverker, A., Ostlund, G., Hallert, C., & Hensing, G. (2007). Sharing life with a gluten-intolerant person – the perspective of close relatives. *Journal of Human Nutrition & Diet, 20,* 412-422.

Troncone, R., Ivarsson, A., Szajewska, H., & Mearin, M. L. (2008). Review article: Future research on Coeliac Disease – a position report from the European multistakeholder platform on Coeliac Disease (CDEUSSA). *Alimentary Pharmacology & Therapeutics, 27,* 1030-1043.

van Heel, D., & West, J. (2006). Recent advances in Coeliac Disease. *Gut, 55,* 1037-1046.

van Koppen, E. J., Schweizer, J. J., Csizmadia, C. G. D.S., Krom, Y., Hylkema, H. B., van Geel, A. M., Koopman, H.M., Verloove-Vanhorick, S. P., & Mearin, M. L. (2009). Long term health and quality of life consequences of mass screening for childhood Celiac Disease: A 10 year follow up study. *Pediatrics, 123,* 582-588.

Wagner, G., Berger, G., Sinnreich, U., Grylli, V., Schober, E., Huber, W., & Karwautz, A. (2008). Quality of life in adolescents with treated Coeliac Disease: Influence of compliance and age at diagnosis. *Journal of Pediatric Gastroenterology and Nutrition, 47,* 555-561.

Public domain briefing paper

Children, young people and Coeliac Disease:

A review of the literature

and

“Am I allowed that?” A qualitative study of young people’s experiences of living with Coeliac Disease

This paper comprises a summary of a literature review detailing the experiences of children, young people and their parents on adherence to a gluten-free diet and the psychosocial consequences of having Coeliac Disease. Further, this paper also comprises a summary of an original piece of research that makes use of qualitative methods to explore children and young people's experiences of living with Coeliac Disease.

Literature review

Introduction: Coeliac Disease is a common autoimmune disease characterised by an immune response to the protein gluten, found in wheat, barley rye and oats. Currently, it can only be treated through a gluten-free diet, which has to be followed for life (Mearin, 2007). Much literature exists in relation to adults with the disease, but little with regard to the experiences of children and young people.

Method: In order to access relevant papers written about this topic, databases were systematically reviewed using search terms specific to children, young people, and their experiences of Coeliac Disease. Only papers published in the last 25 years were included, and all were assigned quality ratings in three areas: rationale and design, methods, and results.

Findings: Overall, 24 papers were included in the review and fell into three categories: treatment adherence, psychological consequences of Coeliac Disease, and parental views. Methodological quality varied greatly across studies, and factors purportedly relating to adherence were contradictory as were outcomes regarding psychological consequences. Reports about parents'

attitudes were more consistent in their content. However, what is clear is that some factors were consistently associated with adherence, namely being younger and female. Further, some studies reported equivalent quality of life between children with Coeliac Disease, and those who did not have Coeliac Disease, but the majority of authors have found that there is some element of psychological distress associated with Coeliac Disease. Finally, the importance of parents in helping young people to adapt to the treatment was reported.

Implications and recommendations: There appeared to be three main themes that could be derived from outcomes reported: the importance of self-efficacy (not necessarily what resources a person has to perform a task that is important, rather it is what that person *perceives* as their resources and/or abilities to manage a task that is important; Bandura, 1977), developmental stage of young person, and illness beliefs (how people perceive different elements of their illness which then informs management, for example, consequences of having an illness; Leventhal, Meyer & Nerenz, 1980). However, methodological quality tended to be low, and it is clear that further studies need be completed to explore childhood reactions to having Coeliac Disease.

Research study

Introduction: Research studies describing people's lived experiences of Coeliac Disease are limited in the literature and moreover, studies of the experience of children and young people with the condition is further limited. However, there are consistent reports that adolescence is a challenging time to manage Coeliac Disease (Greco, Mayer, Ciccarelli, Troncone & Auricchio, 1997), and also the

time that people are most likely to compromise the gluten-free diet (for example, Fabiani et al., 1996), which is the only treatment available. Given this, the aim of this study was to interview children with Coeliac Disease about their experiences of managing their illness, and to analyse these in order to establish what things are of importance to children and young people with the disease.

Methods: Five children and young people were recruited from local voluntary sector Coeliac support groups. Analysis was carried out using Interpretative Phenomenological Analysis (IPA; Smith & Osborn, 2003), which is a type of analysis that can be carried out on interview data. IPA is especially useful for understanding people's experiences (Krahn & Putnam, 2003), and understanding individual meaning of participants' accounts (Smith & Osborn, 2003), without imposing a structure on findings (Osborne, 1994).

Findings: Two themes emerged from the data: *Managing identity as a young person with Coeliac Disease*, and *Ambivalent relationship with food.* The results illustrate how children and young people appreciate parents as a "safe base" in terms of managing the gluten free diet, and also how they felt valued when others catered for them outside the home. Children and young people reported that they sometimes felt isolated and different from peers, and sometimes felt like they were "missing out". Further, young people talked about not necessarily enjoying food, rather, it was important to eat something and to still be able to socialise with others.

Implications and recommendations: For the group of children and young people interviewed here, it appears that managing Coeliac Disease as a young person

can be a stressful and time-consuming issue. It is important to use the results here to inform further research studies that survey a large number of children and young people. It may also be useful if further interview studies are carried out, in order to more fully understand this area. Parents also need to be involved in research interviews to get understand their experiences as well (Huff, 1997), and children also need to be interviewed over time so that issues of development can be studied and its influence accounted for (Holmbeck, 2002). Psychological wellbeing as an over-arching concept may be useful to research, but more specifically young people's methods of coping may be useful to establish. Such studies would allow for suitably informed support to be put in place for young people, should they require it. However, findings reported here suggest that, currently, the annual checks young people have to attend each year could usefully incorporate a psychological component in order to assess psychological wellbeing and management of the diet. Allowing young people a protected space to air some of their concerns may increase adherence and understanding of the gluten free diet.

Acknowledgements

I would like to thank all the children and young people, and their parents, who took part in my research. Their time and interest in the project was fantastic, and I wouldn't have been able to complete this thesis without them. I would also like to thank the two local Coeliac support groups for their enthusiasm and help recruiting the children, young people, and their parents to the study. My thanks also to Dr Goldstein, paediatrician at Birmingham Children's Hospital, and Ros Blackmore, dietician, for their help in the early stages of the project. Finally, I also appreciate the time and support of both research supervisors, Dr Ruth Howard and Dr Gary Law, for helping me throughout the entire project.

References

Bandura, A. (1977). Self-efficacy: Toward a unifying theory of behavioural change. *Psychological Review, 84,* 191-215.

Fabiani, E., Catassi, C., Villari, A., Gismondi, P., Pierdomenico, R., Ratsch, I. M., Coppa, G. V., & Giorgi, P. L. (1996). Dietary compliance in screening detected Coeliac Disease adolescents. *Acta Paediatrica, 412,* 65-67.

Greco, L., Mayer, M., Ciccarelli, G., Troncone, R., & Auricchio, S. (1997). Compliance to a gluten free diet in adolescents, or "what do 300 Coeliac adolescents eat every day?" *Italian Journal of Gastroenterology and Hepatology, 29,* 305-310.

Holmbeck, G. N. (2002). A developmental perspective on adolescent health and illness: An introduction to the special issues. *Journal of Pediatric Psychology, 27,* 409-415.

Huff, C. (1997). Celiac Disease: Helping families adapt. *Gastroenterology Nursing, 20,* 79-81.

Krahn, G. L., & Putnam, M. (2003). Qualitative methods in psychological research. In M.C. Roberts & S. S. Ilardi [Eds]. *Handbook of research methods in Clinical Psychology* (pp. 176-195). Oxford, UK: Blackwell Publishing Ltd.

Leventhal, H., Meyer, D., & Nerenz, D. (1980). The commonsense representations of illness danger. In S. Rachman [Ed]. Medical Psychology (Volume 2, pp. 7-30). New York: Pergamon.

Mearin, L. M. (2007). Celiac Disease among children and adolescents. *Current problems in Pediatric and Adolescent Health Care, 37,* 86-105.

Osborn, J. (1994). Some similarities and differences among phenomenological and other methods of psychological qualitative research. *Canadian Psychology, 35,* 167-189.

Smith, J. A., & Osborn, M. (2003). Interpretative Phenomenological Analysis. In Smith J. A. [Ed]. *Qualitative psychology: A practical guide to research methods* (pp. 51-80). London: SAGE Publications.

Appendix 1

Search terms used in systematic review

- Psycho* AND (child* OR adolescen* OR young*) AND (“Coeliac Disease” OR “Celiac Disease”)

- “Quality of life” AND (child* OR adolescen* OR young*) AND (“Coeliac Disease” OR “Celiac Disease”)

- Emot* AND (child* OR adolescen* OR young*) AND (“Coeliac Disease” OR “Celiac Disease”)

- Behavio* AND (child* OR adolescen* OR young*) AND (“Coeliac Disease” OR “Celiac Disease”)

Appendix 2

Data extraction form

Study	
Country of origin	
Setting	
Number participants	
% uptake	
Inclusion criteria	
Age	
Gender	
Ethnicity	
Social class	
Timing of study in relation to diagnosis	
Design	
Control group	

Assessments *If adherence, how measured? Categories?* *If QOL, questionnaires used?*	
Outcomes	

Conclusions	
Strengths and limitations	
References to follow up	
Other points of note	

Appendix 3

Child topic guide

- Why did you think there was something wrong?

- When did you think you might have Coeliac Disease (CD)?
    - What did you think?
    - What did you feel?
    - What did you want to have happen?

- Can you describe to me what CD is?
    - How would you describe it to someone at school or in a restaurant?

- Tell me about when you were told of the diagnosis.
    - Who told you? (Parent or doctor)
    - What happened to you; what was it like?
    - Who was with you?
    - What did you think?
    - How did you feel?
    - How do you feel about it *now*?
    - (Following on from who was with you) what did they think?
    - How did they react?

- Who does most when it comes to the CD?
    - For example, food shopping and checking, food preparation, telling other people, prescriptions?
    - What is it like for you when that's done for you? How does it feel?
    - How would you like things to be different?

- What/who helps you to manage your CD?
    - Family/friends?
    - Support groups?
    - Health professionals? (e.g. doctors, nurses, dietician, pharmacist?)
    - Tell me about annual checks?

- How often do you cheat on the diet?
    - When was the last time?
    - What makes you want to cheat?

- What is good about having CD?
    - Favourable food? Allowed to pick food items?
    - What do you think about that?

  - What does that feel like?
  - What's your favourite G-F food?
- Who do you think worries the most about the CD?
  - Who worries most, mum/dad/sister/brother/you etc? What tells you that they worry most/least?
  - Do *you* have any concerns about the CD?
  - If yes, what concerns you most? (Socialising/feeling left out/being different?)
  - How does CD affect school etc (school trips)?
  - How have things changed since you were diagnosed, if changed at all? e.g. the food you eat, going out, going on holiday, going to parties, shopping in the supermarket , how you feel (physically)etc.

Appendix 4

Copy of ethical approval

[not available in the web copy of this thesis]

Appendix 5

Information leaflet for children and young people

Information sheet for young people

Project title:

Reactions to diagnosis and what life is like now: A qualitative study of children with Coeliac disease

You are being invited to take part in some research. Before you decide that you would like to take part, it is important for you to understand why the research is being done and what you will need to do if you take part. This leaflet will tell you more!

Thank you for reading this!

Who are we?

We are researchers at the School of Psychology, University of Birmingham.

Eleni

My name is Eleni Theodosi, and I'm a student at the university. As part of my training I need to complete some research, and this is why I'm doing this project. I'm supervised by Dr Ruth Howard and Dr Gary Law, who make sure I am doing the project in the right way.

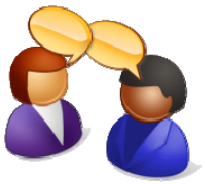
What is our research about?

We are hoping to find out about the experiences of young people with Coeliac disease. We are interested in the time when you were diagnosed, and how activities such as school and friendships are for you now. We'd also like to speak to your parents to see how they felt when you were told about Coeliac disease.

Will you tell anyone what I say?

No. All the answers you give will be kept private. I WILL NOT tell your parents what you say. I will not tell you what your parents say. However, if you tell me that you or someone else in your family is in danger, I will have to tell my supervisor, who will decide what to do with that information so that you remain safe.

Why are we doing this?



To see if we can improve the way you are told about having Coeliac disease. We also hope that after the research we will have a better idea at ways health professionals can support you to be happy after you've been diagnosed.

Who are we inviting to take part?

We are hoping to interview about 6 to 8 young people and their parents. All the young people will have Coeliac disease.

What will I have to do?

You will be interviewed by me at your home or the University of Birmingham. The interview will be very general, and you will have the chance to talk about when you were told about the disease, and what kind of things you do everyday. I will only ask short,

general questions; you can say as much as you want. The interview should take about an hour to complete. I will tape the sessions so that I can write down exactly what you say, and exactly what I say. I'm looking for patterns in what people say, and once I've done this, I will send you the information so that you can see whether you agree with it.

Are there any advantages to taking part in the study?

We are not offering any kind of treatment. However, if you choose to take part you will be entered into a prize draw where the prize is an *IPod shuffle*. It is hoped that this research will be published and make a difference to how people are diagnosed in future, and what support there is for people just diagnosed.

What are the possible disadvantages?

We do not expect you to talk about times that were very upsetting for you, unless you want to. So, we don't expect there to be any disadvantages.

What will happen to the results of this study?

I will write what I have done as a project. This will also hopefully be published in a journal interested in this topic, and I may use some sentences from my interviews with you. I will change names and places so that no-one should be able to work out that it was you that said a certain sentence.

Do I have to take part?

No. It is up to you to decide if you want to take part. If you do decide to take part you will be given this booklet to keep and be asked to sign a consent form. If you decide to take part you can stop at any time, without giving a reason. This will not affect any care that you receive.

Any questions?

We would be really pleased if you could take part in this research. If you would like further information please call the University of Birmingham Clinical office and ask for Eleni, Ruth, or Gary:

[phone number] **Thank you!**

Appendix 6

Information leaflet for parents

Information sheet for parents/guardians

Project title:

Reactions to diagnosis and what life is like now: A qualitative study of children with Coeliac disease

You and your child are being invited to take part in a research project. Before you decide that you and your child would like to take part, it is important for you to understand why the research is being done and what you will need to do if you take part. This leaflet will tell you more!

Thank you for reading this!

Who are we?

We are researchers at the School of Psychology, University of Birmingham. My name is Eleni Theodosi, and I am training to become a clinical psychologist. As part of my training, I am required to complete a doctoral thesis of which this research will comprise part of. I am being supervised by Dr Ruth Howard and Dr Gary Law, both at the University of Birmingham.

What is our research about?

We are hoping to explore the experiences of young people with Coeliac disease. We are interested in the time when your child was diagnosed, and how they get on with activities such as school and friendships now. We'd also like to speak to you, the parent or guardian, to explore how you felt when you were told about your child's Coeliac disease, and how you manage on a day to day basis. It is possible to interview more than one parent/guardian if you want me to.

Will information be confidential?

All information collected will be anonymised. I will not disclose any information about you to anyone else, including your child (unless you give permission). I will also not tell you what your child has said during the research. Information will *not* remain confidential if your child, or you, tells me that someone in your family is being hurt, or your child is at risk of any kind of harm. If this is the case I am obliged to disclose this to my supervisor, who will decide what we should do with the information.

Why are we doing this?

To see if we can improve the way you are told your child has Coeliac disease. We also hope that after the research we will have a better idea at ways health professionals can support you and your family after the diagnosis has been given. There is currently limited information available in this area.

Why has my child been chosen?

Your child has been selected because they have Coeliac disease and are aged between 10 and 17. Unfortunately, we are unable to include children who also have diabetes, or a mental health problem. At this stage, we will also be unable to interview you and your child if you also have a diagnosis of Coeliac disease, or if you have difficulty speaking in English. In total, we hope to interview about 6 to 8 young people and their parents.

What will I have to do?

You will be interviewed by me at your home or the University of Birmingham, and you and your child will be interviewed separately. The interview will be

very general, and you will have the chance to talk about when you were told that your child had the disease, and what kind of things you do on a daily basis as a result of your child's diagnosis. I will only ask short, general questions; you can say as much or as little as you want, and the interview should take about an hour to complete. I will tape the sessions so that I can transcribe verbatim. I'm looking for patterns in what people say, and once I've done this, I will send you the information so that you can see whether you agree with it.

Are there any advantages to taking part in the study?

We are not offering any kind of treatment. However, if you choose to take part, your child will be entered into a prize draw where the prize is an *IPod shuffle*. In addition, it is hoped that this research will be published and make a difference to how people are diagnosed in future, and what support is provided for families with a child who has Coeliac disease.

What are the possible disadvantages?

We do not expect you or your child to talk about times that were very upsetting for you, unless you want to. So, we don't expect there to be any disadvantages.

What will happen to the results of this study?

I will write up what have done as part of my doctoral thesis. This will also hopefully be published in a journal interested in Coeliac disease, and I may use quotes from my interviews with you and your child. I will change all identifiable information, such as names and places.

Do I have to take part?

No. It is up to you to decide if you want to take part. If you do decide to take part you will be given this booklet to keep and be asked to sign a consent form. Please note in addition to obtaining consent from you and your child separately, you will also be required to consent on your child's behalf. If you and your child do decide to take part you can stop at any time, without giving a reason. Please also note that this will not affect any support that you and your family receive.

Any questions?

We would be really pleased if you could take part in this research. If you would like further information please call the University of Birmingham Clinical office and ask for Eleni, Ruth, or Gary:

[Phone number] Thank you!

Appendix 7

Contact sheet

Are you interested in taking part in exciting new research about Coeliac Disease?

A cutting edge project has started!

We are interested in young people's experience of being diagnosed with Coeliac Disease and what life is like now. We are also keen to speak to parents about how they felt at the time of diagnosis, and how they feel now.

Interviews would take approximately 1 hour, and young people (aged between 10 and 17) and parents would be interviewed separately. Unfortunately we cannot interview young people if parents also have Coeliac Disease.

Please discuss this opportunity with your son or daughter. If you are interested in taking part, or would like more information, please call or e mail us:

[phone number]

[email addresses] (Eleni Theodosi)

Alternatively, please tear off the slip below and post to us using the stamped addressed envelope attached. Any young person that takes part will automatically be entered into a prize draw to win an **IPod shuffle**!

After we have heard from you we will contact you to explain the project in more detail and answer any questions.

✂----------

I _____ (name) have discussed this with my son/daughter _____ (their name) and we are interested in taking part.

The telephone number I would prefer to be contacted on is _____ . The best time to contact me is mornings/afternoons/ evenings (please delete as appropriate).

Appendix 8

Consent form

This part should be completed by the participant.

	Please circle the answer you agree with
I have read about the project	Yes/No
The researcher has also explained the project to me	Yes/No
I understand what the project is about	Yes/No
I have asked all the questions I wanted to	Yes/No
I have had my questions answered in a way I understand	Yes/No
I understand that I can stop taking part:	
At any time	Yes/No
Without having to give a reason	Yes/No
I understand that some of what I say (e.g. a sentence) may be written in the final report	Yes/No
I am happy to take part in the study	Yes/No

If any answers are ‘no’ or you don’t want to take part, don’t sign your name!

If you do want to take part, you can write your name below.

Your name: _____

Date: _____

Your parent or guardian must write their name here as well if they are happy for you to do the project:

I am happy for my son/daughter _____ [name] to complete this research.

Name: _____

Sign: _____

Date: _____

The researcher who explained the project to you also needs to sign:

I have explained the study to the participant and given them an information sheet. He has indicated his willingness to take part.

Your name: _____

Date: _____

Appendix 9

Summary of IPA analysis

Smith and Osborn (2003) state there is not one particular way in which IPA should be carried out, and it is at the discretion of the researcher how super-ordinate themes are achieved. However, predominantly in this study, their framework was utilised. Where there were deviations from this, they are clearly marked:

1. Read the first transcript (chosen at random) several times. The aim at this point is to become familiar with the content of the interview and any striking features that become apparent.
2. Once familiar with the first transcript, make annotations in the left hand margin that indicate significant comments, interesting use of language, instinctive ideas about meaning, similarities, contradictions, and any other remarkable features. Smith and Osborn (2003) state there is no correct way in which to do this initial analysis, but it was used in this study mainly to note preliminary ideas about what participants might be trying to convey, and to highlight interesting comments.
3. The above procedure can be carried out as many times as needed. In this study, this was completed at least twice for each transcript.
4. The next stage involves noting “emerging theme titles” (p. 68) in the right hand margin. These themes should sufficiently report the “essential quality” (p. 68) of what participants said. These should be more abstract than the initial coding, which is used to help develop

themes, and perhaps theoretical in nature, and might include psychological language.

5. At all times, it should be clear how themes are related to what participants have said. The particular “richness” (p. 71) of data will typically influence how many themes are generated, but there is no set rule about this. At this stage, all text in the transcript is data, and no data is omitted.
6. Emergent themes are then clustered into super-ordinate themes, whereby the aim is to make sense of the themes according to theory or specific concepts.
7. During this iterative process, it is important to check that the clustering process remains transparent, in that what participants actually said fits with other themes in that cluster.
8. Name the clusters. These names reflect super-ordinate themes. Some themes here may be omitted, either due to lack of evidence or lack of fit with remaining themes.
9. Tabulate clusters of themes, with cluster name (the super ordinate theme), followed by theme, and followed by key excerpts from transcripts highlighting these.
10. Continue analysis of remaining participants’ data. In this study, the emergent themes for all remaining participants were created first, and then these were subsequently clustered, with the analysis of the first participant’s transcript as a guide. Clustering was conducted using paper copies of the themes, and physically moving them around the table to consider different clusters and allow for easier manipulation.

11. Care was taken to consider similarities between participants' accounts, as well as instances where there were differences.
12. In this study, a further analysis of transcripts (step 4 onwards) was conducted after the researcher was diagnosed with food intolerances.
13. Once all transcripts have been analysed and themes clustered into interpretative super-ordinate themes, draw up a table of super-ordinate themes (Table 6 in text).
14. Check that all super-ordinate themes are sufficient conceptualisations of the transcripts; check all transcripts against super-ordinate categories.

Taken from Smith and Osborn (2003)

In order to contextualise this methodology, a short sample of analysis is included below. This displays step 9, in that it contains super-ordinate theme name, participants contributing to super-ordinate theme, theme name, participants contributing to theme, key cross references and indicative quotes. Additionally, there is a column named 'Phenomenology', and this typically represented the initial coding that took place. Thus, this format allowed for constant checking of the transparency of analysis as it progressed.

Super-ordinate theme	Participants contributing to theme	Themes	Phenomenology	Participants contributing to theme	Key cross references	Indicative quotes
Managing identity as a young person with CD	All	ATTEMPTS AT NEGOTIATING DIFFERENCE	Noticing different because eating different food to others	CLAIRE; *TARA;* SARAH; *TOM*	17; 294 *9* 10 *55; 63*	Umm, really all I thought was my sister's eating different stuff, and my mum/I, like just take dinner, umm, I always like had, like if we have like yorkshires and stuff like that my sister's were always like huge and stuff like that and then mine are tiny... I wondered, I just wondered why, why they were different and so practically I knew that something was different about me, umm, like me and my sister and my family. Most of the time my mum like tries to do the same so I don't feel any different but like sometimes, like Yorkshires say, I still have some but I can just tell they're different. Umm, um, er, so bread, I can't have bread and if you look at the size of their bread, they're, they're quite big; my bread is ...like that big [indicates with hands – smaller]. *Well like I was younger and I started going to like birthday parties and that, and everyone was just having like chips or burgers and stuff and I wasn't allowed nothing like that.* Uh, couldn't eat the same as other people. *Umm, at that point I did sort of realise*

						that I had different sort of eating habits, you could say as it were. *I did sort of think 'ok, I'm different from all these people and I gotta look after my food otherwise I'm just going to end up hungry' [laughs].*
		ATTEMPTS AT NEGOTIATING DIFFERENCE	Being the odd one out	SARAH	633; 652; 663	Well I got bullied quite a lot at this school... so I left and they were all the taking the mick out of me and two of my friends and then they were like saying things about I [CD], that's what I hated and that. Well, I got really upset about it. Um, well they were saying stuff like 'oh she can't eat this, at least we can eat what we want to eat', stuff like that... Um, 'cos they were say/just like 'ha, she can't eat this, she can't that, and we can'.
		IMPORTANCE OF OTHERS IN SUPPORTING CD BEHAVIOURS	Enjoying gatherings of Coeliacs because could enjoy food and feel like everyone's the same	CLAIRE; TARA; SARAH; ETHAN; TOM	219; 559 530 346; 398 120 354; 358; 627	It's my friend's Nan and umm, sometimes every time we go down there, just to see how she is, umm, she sometimes gives me some Coeliac food for, umm, me and then I bring some stuff down to her so we get on really well with her, and that's the only Coeliac person I know. Uhh, like it's nice just like seeing people who've, what had, what umm, that I, the people that have got the disease that

						I've got. So, umm, we're not afraid to go up to them and say hello. It's like [laughs] 'cos they/we like we know what's the matter with them and like play, umm. I used to when I was younger like the discos and that and they used to like have like really nice food. Um, they do a lot and uh, my nan [not changed] who's got Coeliacs does a lot as well, 'cos whenever we go over to her's, you can have quite a lot of good stuff there. I've been to um, like two Coeliacs discos where everyone goes. Yeah, it was good 'cos I could eat everything there. First time ever. Yes [the parties for Coeliacs were good], we used to do stuff, like Coeliac stuff that you could take home and that. I went to probably, a meeting of Coeliac something or other. And I suppose it was amazing for a 9 or 10 year old to go. When I was there it was really fun. The atmosphere was like 'ah!'... Um, because there was loads of other Coeliacs and I wasn't sort of, not that I usually feel singled out, but there, there was no sense of that. There was like, it

						was the first time I'd ever gone somewhere and I could eat everything. 'Cos everything was Coeliac, um, gluten free and I was just like, 'score!' [laughs] Biscuits and pizzas and I was like 'yay'! Made me really chuffed [laughs]. 'Cos he [dad on GFD also] gets all the, he gets all the/he's got a really bad sweet tooth as well, so he always gets all the chocolate and the gluten free biscuits and like 'oh great!' [laughs]

Appendix 10

Notes for authors

[Not available in the web copy of the thesis]