Volume I

Research Component

FOREVER GLUTEN-FREE: EXPLORING THE PSYCHOSOCIAL IMPACT OF LIVING WITH COELIAC DISEASE

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Summary

This thesis is presented in two main parts. Volume I is the research component comprising two papers: a literature review and empirical paper each concerned with the psychosocial effects of living with Coeliac Disease. There is also a separate executive summary of both pieces of work. Volume II is the clinical component, a compilation of five Clinical Practice Reports completed at the end of each clinical placement during the 3 years of the Birmingham Clinical Psychology Course.

Volume I

The literature review is a systematic critique of empirical research published within the last decade to investigate the effects of living with CD in respect of psychological well-being and Health Related Quality of Life (HRQoL). Twenty-one relevant studies were identified with attention being paid to the methodology, outcome measures, type of CD and sample characteristics. Difficulties when interpreting and comparing the results of the reviewed studies included differences in design and measures used, sample populations, country of origin, age group and duration of gluten-free diet. The reviewed data suggest that in addition to a reduced HRQoL, psychological distress, especially depressive symptoms is commonly found in people with CD. Although anxiety is commonly experienced, this tends to decrease on a GFD. However, depression may persist even in treated individuals.

The empirical paper describes a postal survey aimed to explore the illness perceptions and self-efficacy beliefs of adults with CD in the UK and reports their subjective levels of HRQoL and psychological well-being. Questionnaires were returned by 288 members of Coeliac UK and within this sample HRQoL and psychological well-being were found to be reduced, with levels being comparable to those found in previous

related studies. Those participants with weak beliefs in the serious consequences of CD and reduced emotional reactions to the condition were more likely to experience an enhanced HRQoL, improved psychological well-being and increased self-efficacy. The results suggest that perceived self-efficacy and illness perceptions could play an important role in informing psychological interventions for individuals with CD.

Volume II

Volume II contains the five Clinical Practice Reports (CPR's) that focus on a combination of clinical perspectives and service issues relating to each placement. CPR 5 was presented orally so that only the abstract is included.

CPR 1 presents two psychological formulations, each from a different theoretical perspective, concerning the problems of Katrina. There are four main sections. The first includes information relating to Katrina's referral, assessment (including history) and her presenting problems. In the second section, Katrina's problems are formulated from a psychodynamic perspective based upon object relations theory and constructed using Malan's triangles of conflict and person. This is followed in section three, by an alternative cognitive behavioural formulation of health anxiety. The fourth and final section is a concise critique of the two different theoretical approaches used in each formulation and includes recommendations for improvements.

CPR 2 describes a study carried out to evaluate clinical supervision groups set up for NHS nurses working in a continuing care unit for older adults. National initiatives concerning supervision for nurses within the NHS and the available evidence for the effectiveness of this are considered. The limitations of the methodology and the difficulties experienced in carrying out the evaluation are discussed and blocking factors are considered which might account for difficulties in implementing clinical group

supervision for all staff. Facilitative strategies are drawn from the relevant literature and recommendations are made for future evaluations and implementing supervision into routine clinical practice.

CPR 3 concerns Christopher, a 9 year old boy referred to a Child and Adolescent Mental Health Service by his general practitioner with a request for help with long-term sleep difficulties. Details of the referral, assessment and formulation relevant to the case are presented. These are followed by a description of the AB single-case experimental design that was employed to assess the effectiveness of a cognitive-behavioural intervention designed to increase Christopher's total number of sleep hours per night and to reduce the time taken for him to fall asleep.

CPR 4 presents the case of Marion a 58 year old woman with learning difficulties, who was reportedly displaying extreme eating behaviour by the over consumption of large quantities of food. Background information on Marion's personal and psychiatric history is provided. This is followed by a formulation of the development and maintenance of Marion's behaviour using cognitive-behavioural principles and a description of the interventions employed to manage her behaviour. Finally there is an assessment of the outcome and reflections on the work with Marion and her care staff.

CPR 5 was an orally presented report describing Tasmita a client who was referred to the CMHT where the trainee was on placement. The scope of the presentation included the reasons for her referral, the assessment methods used, the rationale and theory behind the cognitive behavioural formulation of her difficulties and the evidence for and details of the proposed treatment plan. The formulation described the development and maintenance of Tasmita's depression and obsessive compulsive behaviours and was informed by the information obtained during the assessment stage. This was followed by an evaluation of the cognitive behavioural treatment plan in terms of Tasmita's own subjective experience.

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Abstract

Coeliac Disease (CD) is an autoimmune disease in which the enzyme gluten causes inflammation and damage to the small intestine. Untreated the condition may predispose symptomatic individuals to serious diseases such as cancer, type I diabetes, osteoporosis, gynaecological problems in women, peripheral and central nervous system disorders and other autoimmune diseases. In Europe it is estimated that the condition may affect between 1 in 200 to 1 in 500 people. CD is incurable but symptoms are managed by a gluten-free diet (GFD) for life. Most research looking at CD has focused on the biological basis of the disease rather than the impact of the condition from the individual's subjective view. The few existing studies suggest that the chronicity of the condition, the limitations imposed by the need to follow a permanent restrictive diet and the risk of other associated diseases can have a negative impact on health-related quality of life (HRQoL) and psychological well-being. However, knowledge in this area remains sparse to-date.

The aim of this review was to systematically review the literature published within the last decade to investigate the effects of living with CD in respect of psychological well-being and HRQoL. Twenty-one relevant studies were identified and critically reviewed. Attention was paid to the methodology, outcome measures, type of CD and sample characteristics. Difficulties when interpreting and comparing the results of the reviewed studies included differences in: design and measures used, sample populations, country of origin, age group, CD type, duration of GFD and adherence rates. The reviewed data suggest that in addition to a reduced HRQoL, psychological distress, especially self-reported symptoms of depression are commonly found in individuals with CD. Although anxiety symptoms are commonly experienced, these tend to decrease on a GFD. However, depressive symptoms may persist even in treated individuals.

Introduction

Coeliac Disease (CD) is an autoimmune disease in which the enzyme gluten, from wheat, barley and rye, causes damage and chronic inflammation to the small intestine leading to the malabsorption of nutrients. People with the condition will have what is termed small-bowel villous atrophy or flattened villi (the tiny, finger like projections on the surface of the small intestine that help absorb nutrients). This means their ability to absorb nutrients is severely restricted (Jones, 2007). Untreated, CD may predispose symptomatic individuals to cancers such as small-bowel lymphoma (Egan, Stevens & McCarthy, 1996) non-Hodgkin's lymphoma and other gastrointestinal cancers (West, Logan, Smith, Hubbard & Card, 2004). The condition is also associated with type I diabetes, osteoporosis, gynaecological problems in women (Feighery, 2007) peripheral and central nervous system disorders (Cooke & Smith, 1966) and other autoimmune diseases (Ventura, Magazzu & Greco, 1999).

Diagnosis is achieved through serologic testing (screening blood tests using a highly sensitive immunoglobulin A assay) followed by a biopsy of the small intestine (duodenum) to detect villous atrophy (Hopper et al, 2007). CD can occur at any age, but in adults the peak incidence is in the fifth decade and females are more commonly affected than males (Jones, 2007). In Europe it is estimated that CD may affect between 1 in 200 to 1 in 500 people (Rewers, 2005; Catassi, Ratsch & Fabiani, 1994). This is comparable with prevalence rates in Canada (Cranney, Zarkadas, Graham & Switzer, 2003). However, some studies suggest a prevalence rate of 1 in 122 (Johnson, Watson, McMillan, Sloan & Love, 1997) although many cases of the disease remain undiagnosed for many years leading to chronic ill-health (Ivarsson, Persson, Juto, Peltonen, Suhr & Hernell, 1999). In the United States CD appears less prevalent, but this may be because the disease is under diagnosed

relative to Europe and the use of serologic testing is more common in Europe than the United States (Ferguson, 1997).

Coeliac Disease has been classified into four phenotypes (Rostrom, Murray & Kagnoff, 2006). In 'classic' CD, sufferers have intestinal malabsorption due to villous atrophy causing gastrointestinal symptoms including nausea, diarrhoea, bloating and abdominal pain. However, the 'atypical' (yet most common) form of the disease is asymptomatic with few or no gastrointestinal symptoms, but is characterized by other malabsorption problems including iron deficiency anaemia, osteoporosis, short stature and infertility (Goddard & Gillett, 2006). In 'silent' CD individuals have no overt symptoms, but are found to have damaged intestinal villi caused by gluten in the diet. This type of presentation may be discovered after serologic screening, endoscopy or duodenal biopsy for another reason. In 'latent disease', individuals have a previous diagnosis of coeliac disease that responded to a gluten-free diet (GFD) and have normal intestinal mucosa. Latent CD can also represent individuals with currently normal intestinal mucosa on a gluten-containing diet who will subsequently develop CD.

CD is incurable but is managed by a therapeutic GFD for life. A gluten-free diet involves the complete avoidance of all foods made from or containing wheat, rye, barley and often oats; in some cases oats may be permitted (Haboubi, Taylor & Jones, 2006). Foods containing gluten include breads, pizza bases, biscuits, cakes, pastas, flours and cereals (Coeliac UK, 2008). The GFD is very successful in managing the symptoms of CD as the removal of gluten allows the duodenal villi to re-grow, therefore leading to the normal absorption of nutrients (Mäki & Collin 1997).

However, a diagnosis of CD and the start of a GFD can cause enormous changes in the lives of sufferers and the dietary restrictions can be hard to accept (Mäki & Collin, 1997). Those with the condition can feel restricted, isolated and at times anxious about

their food intake (Sverker, Hensing & Hallert, 2005) and adults with CD often report a poor quality of life (Hallert & Lohiniemi, 1999).

Most research looking at CD has been focused on the biological basis of the disease and the autoimmune system (Cooper, Holmes & Kooke, 1978; Barone et al, 2007) and the long-term consequences of untreated CD (Dewar, Pereira & Ciclitira, 2004). However, the growing interest in patient perspectives has stimulated research on subjective outcomes, particularly those that measure health-related quality of life (HRQoL) and psychological well-being (Hallert & Lohiniemi, 1999). Consequently, the assessment of HRQoL is increasingly becoming an important outcome measure in clinical and epidemiological studies of gastroenterological disease (Hauser, Gold, Stallmach, Caspary & Stein, 2007).

HRQoL is important for measuring the impact of chronic disease. The concept refers to an individual's perceived state of health including social, emotional and physical well-being or functioning and incorporates positive and negative aspects of life (Fitzpatrick, Fletcher, Gore, Jones, Spiegelhalter & Cox, 1992). In gastrointestinal disease the most relevant aspects of HRQoL tend to comprise primarily the subjective perception of relief of abdominal symptoms and secondarily the benefits of this relief on general well-being and functional status (Hallert & Lohiniemi, 1999).

HRQoL is a multi-dimensional dynamic concept that has developed from the need to assess the psychosocial impact of disease, which includes economic welfare, psychological well-being, social environment and health status (Sajid, Tonsi & Baig, 2008). The 'health' domain in HRQoL can range from negatively valued aspects of life, including anticipating death, to the more positively valued aspects such as role function or happiness. Quality implies subjective evaluation by the individual. The boundaries of this definition usually depend on the context and purpose of any particular assessment as well as the specific concerns of patients, clinicians and researchers (Guyatt, Feeny & Patrick,

1993). The concept HRQoL is used when studying living with illness, rather than that of general QoL, because whilst widely valued aspects of life exist that are not generally considered as 'health', including income, freedom and quality of the environment, almost all of these aspects of life can become health related or affected when a person is living with illness (Guyatt et al, 1993). Health-related quality-of-life (HRQOL) is still a loose definition and its relevant aspects may vary from study to study. However, there is consensus that key dimensions of HRQoL are physical functions, sensations, self-care/dexterity, cognition, pain/discomfort and emotional/psychological well-being i.e. "all within the skin" (Sajid et al, 2008). Therefore, concepts such as health status, psychological distress and psychopathology can be viewed as specific facets amongst the many different aspects of HRQoL. The World Health Organisation has defined HRQoL as:

> *[…] an individual's perception of their position in life in the context of the culture and value systems in which they live, in relation to their goals, expectations, standards and concerns. It is a broad ranging concept affected in a complex way by the person's physical health, psychological state, level of independence, social relationships and their relationships to salient features of the environment (WHO QOL-group, 1996).*

The few studies conducted so far concerned with various aspects of HRQoL in adults with coeliac disease, have been conducted in Canada, the USA and Europe most notably Italy, Germany and Scandinavia. Studies from these countries suggest that depression (Ludvigson, Reutfors, Ösby, Ekbom & Montgomery, 2007) and lower quality of life (Addolorato et al, 2008) affect individuals with CD, and anxiety and depression have been identified as major causes of lower levels of adherence to treatment recommendations (for example, Addolorato et al, 1996; Holmes 1996).

Scandinavian studies conducted in the early 1990's cited by Hallert and Lohiniemi (1999) suggest that during the course of a therapeutic GFD, adults experience a detrimental

decline in subjective health and increased gastrointestinal symptoms and this tends to be most pronounced in females who score worse than non-coeliac women of the same age. However, knowledge of subjective health-related quality of life including self-perceived functional status of individuals diagnosed with CD remains sparse to-date and there is no literature that draws together the existing empirical studies conducted in Europe, Canada and the United States.

Aims

The main aim of this paper is to redress this situation by systematically reviewing empirical, peer reviewed studies published within the last decade that investigate the effects of living with CD in respect of various aspects of HRQoL including psychological well-being (a key dimension of this concept). The literature will be critically reviewed to establish whether having coeliac disease and living on a GFD is associated with or has a detrimental effect on aspects of HRQoL and/or psychological wellbeing. Attention will be paid to the methodology, outcome measures, type of CD and sample characteristics. A further aim is to consider the implications of the results for clinical practice.

Method

Search strategy/selection of articles

Pubmed/Medline, PsychInfo and CINAHL, were searched to identify literature on the psychosocial impact of Coeliac Disease on medically diagnosed adults published from 1997 until 2008. Search terms were 'psychosocial', 'affective disorder', 'anxiety', 'depression', 'quality of life', 'psychological adjustment', 'psychological well-being', 'gluten-free diet' and 'coeliac/celiac disease'. References in the retrieved articles were further searched for relevant citations. Prominent authors in the field were contacted by

email and asked for further relevant papers. These searches resulted in a total of 264 hits to which the following criteria were applied:

Year	Studies published during and after 1998.
Sample	Adults medically diagnosed with coeliac disease.
Study design	Studies that include a control or reference group cohort or cross-sectional studies.
Measures	Use of self-report measures of health-related quality of life (HRQoL) psychological well-being and/or distress.
Exclusion	Case studies, studies described in editorials, commentaries, or conference abstracts or other non peer reviewed studies, qualitative studies. Studies of adults focused on co-morbidities e.g. diabetes and irritable bowel syndrome.
Language	Studies published in the English Language.

Results

The next section will give a brief overview of the studies, followed by a detailed description and critique, concluded by a final section that draws the findings together.

After applying the inclusion criteria and eliminating duplicate hits, 21 relevant empirical studies published between 1998 and 2008 were available for review. A description of the studies can be found in Table 1 and a full reference list can be found in Appendix 1.

Table 1 Characteristics and Main Findings of Included Studies

Authors/ Year/ Country	Design	Target Group/ Sample Size	Outcome Measures	Gender	Main Findings
Ciacci et al, 1998; Italy	Retrospective case control: matched groups	92 adults with biopsy proven CD; 48 with chronic persistent hepatitis; 100 healthy controls	M-SDS	CDG 70 ♀; CPH 34 ♀; CG 71♀	Depression scores significantly ↑ in CD groups compared to controls; age at diagnosis and compliance with diet did not correlate with depression.
Hallert et al, 1998; Sweden	Retrospective cross-sectional: compared with normative data	89 adult biopsy-proven coeliac patients on GFD versus normative population sample: 5277	SF-36 GSRS	61% ♀ CDG; 2713 ♀ CG	After 10 years on a GFD, patients with CD had significantly lower SF-36 scores than general population. Low scoring confined to female patients.
Addolorato et al, 2001; Italy	Prospective: case-control, matched groups.	35 CD patients on GFD versus 59 healthy controls	STAI M-SDS	CDG 23 ♀; CG 32 ♀	At T0, pre-GFD, CD patients showed higher levels of state anxiety compared to controls. At T1 there was a significant drop in state anxiety in CD patients.
Green et al, 2001; USA	Retrospective cross-sectional Survey	1612 adults belonging to CD support groups across the north eastern regions in the United States. 75% biopsy proven CD.	1 QoL item non-stan-dardised	Male-to-female ratio 1:2.8	QoL after diagnosis rated as improved by 77% of sample.
Ciacci et al, 2002; Italy	Retrospective cohort:	114 adults with treated CD: 25 with untreated disease.	SAIC – developed by the authors + IBQ	87 ♀ in treated CDG.	Self-rated emotion data resulted in 3 factors: 1 (fear, anger, anxiety & sadness) 2 (reassurance & resignation) 3 (relief)
Hallert et al, 2002; Sweden	Retrospective, case-control: matched groups.	34 men and 34 aged matched women with CD on GFD; 68 matched type-2 diabetes controls	BI SF-36	Male-to-female ratio 1:1	Unlike women with diabetes, coeliac women (adhering to a GFD for several years) perceive the disease burden to be worse than men.

Table 1 continued/ ...

Authors/ Year/ Country	**Design**	**Target Group/ Sample Size**	**Outcome Measures**	**Gender**	**Main Findings**
Mustalahti et al 2002; Finland	Prospective case-control**:** un-matched groups	19 adults on GFD with screen detected CD; 21 with symptom-detected CD; 105 healthy controls	GSRS PGWBI	Mostly ♀	GFD associated with improved QoL for symptom detected and screen-detected groups at 1 year.
Usai et al 2002; Italy	Retrospective case-control: matched groups	68 patients with CD on GFD versus 136 healthy controls	SF-36	CDG: 54 ♀ CG: 112 ♀	CD group obtained significantly worse scores than healthy controls on the SF-36 – GFD compliers showed better results than non-compliers
Ciacci et al 2003; Italy	Retrospective cross-sectional	581 members of 5 regional coeliac societies	SAIC	410 ♀	Coeliac disease not associated with a low level of self-perceived quality of life**.** Anxiety related to feeling different.
Fera et al, 2003; Italy	Retrospective case-control: matched groups	3 groups of 100 patients: 100 CD (biopsy proven) on GFD, 100 with diabetes and 100 healthy controls	M-SDS STAI-Y1/Y2 SF-36 IBQ	CDG: 75 ♀; DG 73 ♀ CG: 68 ♀	M-SDS & STAI-Y2 scores significantly higher in patients with CD and diabetes than in healthy controls. QoL was poorer in both CD and D patients than in controls & significantly correlated with anxiety.
De Rosa et al, 2004; Italy	Retrospective case-control: matched group	29 adult biopsy proven CD patients versus 47 healthy comparisons	IBQ EPQ PQ	CDG 25 ♀; CG 40 ♀	More than 70% of the CD group scored in the pathological range on at least one scale of the IBQ.
Johnston et al 2004; Northern Ireland, UK	Prospective case control: unmatched groups	14 adults on GFD with screen-detected CD & 23 controls; 17 clinically detected CD patients on GFD & 26	SF-36	Mostly ♀	No significant differences on QoL between screen-detected CD group and controls and at 1 year follow-up.
Siniscalchi et al, 2005; Italy	Retrospective case control: healthy comparison group	59 CD patients on GFD, 71 CD patients on ND, 80 healthy controls.	M-SDS CFS FSS	CDG 112 ♀ CG 63 ♀	All CD patients scored higher on depression and fatigue scales than healthy controls. In CD group on GFD, depression and fatigue scores did not significantly differ from CD group on ND.

Table 1 continued/...

Authors/ Year/ Country	Design	Target Group/ Sample Size	Outcome Measures	Gender	Main Findings
Viljamaa et al, 2005; Finland	Retrospective case control: unmatched groups	81 CD screen detected adults versus 44 CD symptom detected adults; 54 untreated CD patients; 110 healthy adult controls	PGWBI SF-36	Greater number of ♀ than men	In screen detected participants QoL and gastro-intestinal symptoms were similar to those in symptom detected patients or non CD controls.
Häuser et al 2006; Germany	Retrospective, cross-sectional survey	446 members of the German Coeliac Society – biopsy proven diagnosis.	SF-36 HADS	71.7% ♀	Despite a GFD, participants suffered a reduced HRQoL and a high burden of extra-intestinal symptoms.
Roos et al 2006; Sweden	Retrospective case-control: matched groups	51 medically diagnosed adults with CD on a GFD versus 182 age matched healthy adults	PGWBI	59% ♀ CDG; 57% ♀ CG	Long treated adult CD participants showed no difference in psychological well-being to controls.
Zarkadas et al 2006; Canada	Retrospective cross-sectional survey	2681 adult members of the Canadian Coeliac Association – biopsy proven diagnosis	SF-12 + CD specific questions developed by authors.	74.5% female	QoL scores similar to normative data, but significantly lower for females and newly diagnosed participants.
Addolorato et al, 2008; Italy	Retrospective case control, matched groups	40 adult-biopsy proven Classic CD patients: 25 on GFD & 15 newly diagnosed versus 50 healthy controls	LSAS M-SDS	35 female CD group: 40 female controls	Significantly higher prevalence of social phobia in CD patients compared to controls. Depression was present in a significantly higher percentage of CD patients compared to controls
Casellas et al, 2008; Spain	Prospective cohort with internal controls	Serological and symptom diagnosed adults with CD: 163 on GFD, 177 newly diagnosed not on GFD	2 HRQoL Scales: GIQLI EQ	103 newly diagnosed females; 121 GFD females	GIQLI scores significantly better in patients on GFD; EQ scores significantly better in treated versus non-treated participants

Table 1 continued/…

Authors/ Year/ Country	Design	Target Group/ Sample Size	Outcome Measures	Gender	Main Findings
Collin et al, 2008; Finland	Prospective case-control: non-matched groups	20 adult patients with biopsy proven CD on GFD versus 1199 non-CD comparisons	CCEI; TAS-20	15 ♀ CD group: 50 ♀ controls	Somatic anxiety higher in CD patients before GFD started. No alexithymia found, but scores improved at 1 year follow-up. Scores no different from comparisons.
Nachman et al, 2008; Argentina	Prospective case-control, non-matched groups	97 adults on GFD with classical CD symptoms; 25 atypical symptoms/10 silent CD; 70 healthy controls	SF-36 GSRS BDI	117 ♀ across CD groups	GFD produced substantial and rapid (3 month) improvement of most outcome measures in classical and atypical patients but not in silent cases. All subgroups had similar 1-year scores to healthy controls

Key to abbreviations:

CDG	Coeliac Disease Group
CG	Control/Comparison Group
CPH	Chronic Persistent Hepatitis
DG	Diabetes Group
GFD	Gluten Free Diet
HRQoL	Health-related Quality of Life
ND	Normal Diet
♀	Women

Key to outcome measures:

BDI	Beck Depression Inventory
BI	Burden of Illness
CCEI	Crown-Crisp Experiential Index
CFS	Chronic Fatigue Scale

Key to outcome measures cont'd:

EPQ	Eysenck Personality Questionnaire
EQ	EuroQol-5D
FSS	Fatigue Severity Scale
GIQLI	Gastrointestinal Quality of Life Index
GSRS	Gastrointestinal Symptoms Rating Scale
HADS	Hospital Anxiety and Depression Scale
IBQ	Illness Behaviour Questionnaire
LSAS	Liebowitz Social Anxiety Scale
M-SDS	Modified Zung Self-Rating Depression Scale
PGWBI	Psychological General Wellbeing Index
PQ	Pychophysiological Questionnaire
SAIC	Self-Administered Inventory for Celiacs
SF-12/36	Short Form-12/36 Health Survey
STAI	State-Trait Anxiety Inventory
TAS-20	Toronto Alexithymia Scale

Country of origin

Eight studies originated from Italy, three from Sweden and three from Finland, with the remaining seven originating from Germany, the UK, Canada, the USA, Spain, Finland and Argentina.

Gender

Apart from one study where men and women were evenly matched, women were over-represented in the majority of the studies.

Design of Studies

Table 2 below shows the methodological design of the included studies. There are thirteen case-control studies: eight of these are retrospective and five prospective. A further seven are cross-sectional and retrospective in design and the remaining two are cohort studies, one prospective and the other retrospective.

Table 2 Design of Studies Included in the Review

Sampling Method	Prospective Studies	Retrospective Studies
Case-control/comparison	Addolorato et al, 2001 Mustalahti et al, 2002 Johnston et al, 2004 Collin et al, 2008 Nachman et al, 2008	Ciacci et al, 1998 Hallert et al, 2002 Usai et al, 2002 Fera et al, 2003 De Rosa et al, 2004 Siniscalchi et al, 2005 Viljamaa et al, 2005 Addolorato et al, 2008
Cohort	Casellas et al, 2008	Ciacci et al, 2002
Cross-sectional		Hallert et al, 1998 Green et al, 2001 Ciacci et al 2003 Häuser et al 2006 Roos et al 2006 Zarkadas et al 2006

Sample Characteristics

The samples in the majority of the studies were drawn from hospital populations apart from four national surveys that recruited participants from Coeliac Support Group networks. Size was variable depending on study design (please see Table 1) although prospective studies tended to have less than adequate numbers.

Type of Coeliac Disease

All participants in the studies had either biopsy proven and/or serologically detected CD. Only one study distinguished between classical CD symptoms, atypical symptoms and Silent Disease and another referred to participants as those with classic form CD.

Outcome Measures

Table 3 overleaf, shows the different self-report outcome measures chosen in each study to assess psychological distress and/or HRQoL (please see Appendix 2 for a glossary of abbreviations and referenced list of measures). Eight studies focused on psychological symptoms and/or psychiatric problems in CD, a further nine studies were concerned exclusively with assessing HRQoL, the majority using measures specifically designed to measure physical and emotional functioning. Four studies used specific measures to explore psychological distress and generic multi-item scales to assess HRQoL.

The majority of studies that used standardised measures of HRQoL focused on the effects of living on and adhering to a gluten free diet. The most commonly used instrument for measuring HRQoL was the Short Form 36 Health Survey (SF-36) that was used in eight studies. For investigating patient self-reported symptoms the modified Zung Self-Rating Depression Scale (M-SDS) was employed in five studies. The three items of

this scale that evaluated gastro-enteric symptoms of depression (decreased appetite, weight loss & constipation) were omitted to avoid bias. The Illness Behaviour Questionnaire (IBQ) was also used in three different studies as was the Psychological General Well-being Index (PGWBI). Apart from the previous examples, in general there was little consistency in terms of outcome measures employed across the 21 reviewed studies.

Table 3 Research focus and self-report (S-R) measures used in the included studies

Studies of Psychological and/or Psychiatric Factors in CD		
Study Focus	**S-R Outcome Measures**	**Authors**
Depressive Symptoms	M-SDS	Ciacci et al, 1998
Anxiety & Depression on GFD	STAI, M-SDS	Addolorato et al, 2001
Psychological dimensions	SAIC, IBQ	Ciacci et al, 2002
Illness Behaviour & Personality	IBQ, EPQ, PQ	De Rosa et al, 2004
Fatigue & Depression	M-SDS, CFS, FSS	Siniscalchi et al, 2005
Psychological well being	PGWBI	Roos et al, 2006
Social Phobia & Depression	LSAS, M-SDS	Addolorato et al, 2008
Alexithymia & Anxiety	CCEI; TAS-20	Collin et al, 2008
Studies of Health Related Quality of Life in CD		
Study Focus	**S-R Outcome Measures**	**Authors**
Impact of GFD	SF-36, GSRS	Hallert et al, 1998
Clinical spectrum of CD & QoL	Non-standardised scale	Green et al, 2001
Impact of GFD	SF-36, BI	Hallert et al, 2002
Impact of GFD	GSRS, PGWBI	Mustalahti et al, 2002
Adherence (or not) to a GFD	SF-36	Usai et al, 2002
Impact of GFD	SAIC	Ciacci et al, 2003
Impact of GFD	SF-36	Johnston et al, 2003
Impact of GFD	SF-12, authors' own	Zarkadas et al, 2006
Impact of GFD	GIQLI, EQ	Casellas et al, 2008
Studies of Quality of Life and Psychological Distress in CD		
Study Focus	**S-R Outcome Measures**	**Authors**
Effects of GFD on QoL & mood disturbance	M-SDS, SF-36, IBQ STAI-Y1/Y2	Fera et al, 2003
Impact of dietary compliance on QoL & mental health	PGWBI, SF-36	Viljamaa et al, 2005
Clinical spectrum of CD, HRQoL & mood disorders	SF-36, HADS,	Hauser et al, 2006
Time-course impact of GFD On QoL and depression	SF-36, GSRS, BDI	Nachman et al, 2008

For key to outcome measures please see table 1

Main Review

Studies using self-report measures of psychological factors and psychiatric disorders

Ciacci, Iavarone, Mazzacca & De Rosa (1998) found depressive symptoms in young adult out-patients with CD compared to a healthy control group measured by the M-SDS. The depressive symptoms were not influenced by age at diagnosis, gender, socioeconomic variables, duration of or self-reported adherence to a GFD. The average duration of GFD was 7.9 years. The authors suggest that depressive symptoms are a feature of CD and are related to living with the condition rather than to an organic cause i.e. not a consequence of brain function disorders due to intestinal malabsorption. A disadvantage of this retrospective case-control study is that although it may prove an association between CD and depressive symptoms, it does not demonstrate causation.

In a longitudinal study Addolorato et al (2001) studied individuals with CD before and after a year on a GFD. The presence of symptoms of anxiety and depression were assessed with the STAI and M-SDS respectively and compared to healthy matched controls. At baseline, a significantly higher percentage of individuals with CD showed high levels of state anxiety compared to controls but no significant difference was found in trait anxiety. The percentage of individuals with CD who reported depressive symptoms was also significantly higher compared to controls. At 1 year follow-up a significant decrease was found in the percentage of state anxiety in individuals with CD compared to controls. There were no significant changes in trait anxiety or depression, the latter remaining significantly higher in treated (GFD) individuals with CD. The authors concluded that in people with CD, anxiety exists in a 'reactive form' (rather than as a personality trait) and decreases after starting a GFD. They suggest that the symptoms of depression that remained present in treated individuals could be attributed to the reduction in quality of life related partly to a decreased feeling of well-being and also due to dietary

restrictions leading to difficulties in daily social relationships. For example, not being able to eat the same food as one's peers can lead to a reduced social life and feelings of inadequacy and difference. A limitation to this otherwise sound study is the relatively brief follow-up period of 1 year after adherence to a GFD. A longer follow-up period would allow for the exploration of the persistence of depressive symptoms in people with CD.

In a study by Ciacci, Iavarone, Siniscalchi, Romano & De Rosa (2002) the emotional impact of a diagnosis of CD in adulthood was explored. Using a questionnaire developed by the authors (SAIC) the psychological and emotional aspects of living with CD were investigated. Scores of self-rated emotions were entered into a principal components analysis that generated 3 factors: (1) fear, anger, anxiety & sadness; (2) reassurance & resignation; (3) relief. Anger was found to represent the predominant emotion that reduced adherence to GFD and led individuals to transgress. Significant positive correlations were found between 'feeling different' and sadness, anger, fear and anxiety. The non-heterogeneous sample in this study that included twenty five untreated individuals with CD is a significant weakness. Furthermore, the items of the SAIC measure (some based on a less robust visual analogue scale) were only partially validated against similar questions of the Illness Behaviour Questionnaire (IBQ) completed by twenty seven individuals with CD.

De Rosa, Troncone, Vacca & Ciacci (2004) used the IBQ to investigate the influence of CD and its treatment on key personality components and adherence to a GFD. Other measures were the EPQ and PQ. The latter instrument explores level of stress-induced emotional activation. The IBQ scales on which the highest number of individuals had scores in the pathological range were 'affective inhibition' and 'irritability'. The EPQ scores of individuals with CD differed significantly from controls on the P scale

(psychoticism) and the L scale (lie scale). On the PQ, individuals who received a diagnosis in adulthood had significantly higher scores than the healthy controls. On the basis of these results, the authors suggest that CD is associated with a 'Coeliac Profile' composed of two main characteristics: (1) irritability with related psychophysiological reactiveness and (2) a type of conformism that reflects difficulty in expressing personal feelings and the desire to have a good self-image. They conclude that this profile characterises a lifestyle limited by chronic disease. However, 86% of their sample were women thus biasing these results.

In a study to evaluate the prevalence of fatigue, Siniscalchi et al (2005) administered the M-SDS, the CFS and the FSS to treated versus untreated out-patients with CD and healthy controls. They found that all the individuals with CD had significantly higher M-SDS depression scores and greater CFS scores compared with controls. Those individuals on a GFD had significantly higher depression scores than those in the non-treated CD group. The prevalence of clinically significant levels of depression in all individuals with CD was 17% compared to 0% in healthy controls. The authors also found a significant correlation between depression scores and fatigue scale scores in the non-treated CD group. In the GFD group, fatigue scale scores did not differ significantly from those on a normal diet and were not related to dietetic compliance. The authors state that fatigue is a common finding in people with CD and emphasise that depressive symptoms in individuals on a GFD seem to persist, supporting previous observations that a GFD alone does not significantly reduce the percentage of people with CD affected by low mood (Ciacci et al, 1998).

Roos, Kärner and Hallert (2006) used the PGWBI to assess the psychological well-being of middle aged adults with CD who had adhered to a GFD for at least ten years. Compared to controls, those with CD showed no more signs of anxiety, depressed mood or distress. However, unlike controls women with CD showed significantly lower scores on

the PGWBI than their male counterparts, most notably within the 'Vitality' sub-scale. Based on previous qualitative research the authors hypothesised that women with CD may perceive the burden of their disease as significantly worse than men in terms of inhibiting socialising with friends and having to abstain from other important activities of daily living.

The presence of social phobia (an anxiety disorder) and depressive symptoms in individuals with CD was investigated by Addolorato et al (2008) using the LSAS and M-SDS. The authors found that the percentage of participants with mild and severe social phobia was significantly higher in those with CD compared to healthy controls. There were no significant differences either for mild or severe social phobia between newly diagnosed, un-treated participants and those on a GFD. Depressive symptoms were also found in a significantly higher proportion of individuals with CD than controls and there was a direct correlation between social phobia and depressive symptoms in the CD group. The authors suggest several reasons for the onset of social phobia in individuals with CD. For instance, before diagnosis, the main symptoms of CD such as abdominal pain, diarrhoea and weight loss are often reported as the reasons for work and relationship difficulties. In addition there is a common fear of being judged as a sick person. As time progresses, the restricted lifestyle associated with a GFD might increase feelings of being different in relation to others and could result in a substantially reduced social life. The authors also suggest that the avoidance of social activities and public situations might lead to the development of depression. However, as the authors mention, the lack of a comparison group of individuals affected by another disease makes it impossible to assess whether social phobia is related to CD per se or to general symptoms.

Collin, Kaukinen, Mattila & Joukamaa (2008) evaluated whether individuals with CD suffer from psychoneurotic symptoms or alexithymia. Alexithymia is associated with

various gastrointestinal disorders and refers to a personality construct characterised by impoverished fantasy, a poor capacity for symbolic thought and an inability to experience and verbalise feelings. The CCEI which has six sub-scales was used to measure neurotic psychopathology (including depression and anxiety) and the 20-item version of the Toronto Alexithymia Scale (TAS-20) was used to measure alexithymia in a small sample of adults with CD before and after commencing a GFD. It was found that somatic anxiety was higher in individuals with CD before the introduction of the GFD than after adhering to the diet for one year. GFD had no other significant effect on CCEI scores. There was no evidence of alexithymia, but TAS-20 scores improved significantly at one year follow-up. Individuals' scores did not differ from published Finnish population estimates. The authors observe that these findings do not support studies from other countries where depression has been more commonly found in people with CD. They suggest that as their sample had only minor coeliac-related symptoms and excellent GFD adherence rates, this may have influenced the results. In addition, they state that there is much more common knowledge about CD in their country (Finland) where there is a high clinical prevalence of the disease and where gluten-free alternatives are more widely available commercially and in restaurants.

Amongst these eight studies above, the majority used validated measures of depression and/or anxiety. Apart from 2 prospective studies (Addolorato et al, 2001 and Collin et al, 2008) the remainder were all retrospective in design. In the studies using the M-SDS (including one prospective study) the presence of depressive symptoms was found in individuals with CD compared to controls. In the prospective study (Collin et al, 2008) depression and state anxiety were higher before commencement of a GFD compared to controls. At 1-year follow-up anxiety decreased but depression persisted. In a retrospective study using the PGWBI (Roos et al, 2006) individuals with CD were found to

be no more anxious or depressed than healthy controls. However, women with CD showed lower scores than their male counterparts. In the remaining Italian prospective study, compared to controls, somatic anxiety, but not depression was higher in individuals with CD before the introduction of a GFD than after 1-year follow-up on treatment.

Studies using self-report measures of Health Related Quality of Life (HRQoL)

Hallert et al (1998) used the SF-36 to assess the subjective health status of adults with CD who had been on a GFD for 10 years. Those shown to be in histological remission were also assessed on the GSRS. The SF-36 scores of female participants with CD were significantly lower than those of the general population, particularly in the General Health and Vitality sub-scales suggesting poor subjective health and excessive tiredness. Female participants also reported significantly more gastrointestinal symptoms than males as measured by the GSRS. In contrast, men with CD scored higher than their male counterparts in the general population on most of the SF-36 scales, particularly on Bodily Pain and Social Functioning domains. The reported dietary compliance rate for all participants with CD was 78% and this was corroborated by biopsy. The authors expressed surprise that after 10 years on a GFD, females with CD failed to achieve the subjective health status reported by the general Swedish population, but hypothesise that men and women may cope differently with the social inconvenience to their daily activities caused by dietary restrictions as indicated by the difference in scores on the Social Functioning domain of the SF-36. In an extension of the above study, Hallert et al (2002) investigated possible gender differences in perceived illness burden. They studied individuals with CD and matched type-2 diabetes controls treated for an average of ten years. Participants completed the 9-item Burden of Illness (BI) protocol comprising perceived worries, restrictions and subjective outcome. This measure was developed by the authors on the

basis of many years of clinical experience with adults diagnosed with CD. Cronbach's alpha reliability coefficient for the instrument was 0.73 showing good internal consistency. Construct validity was provided by a fair agreement with the PGWB index. Subjective health was also assessed with the SF-36. The results indicated that the importance of complying with a GFD was ranked high by male and female participants. However, women were less satisfied with the outcome after ten years than men and tended to express more concern about the impact on socialising with friends and the resulting abstention from important things in life. None of these concerns distinguished between males and females with diabetes. Women with CD had higher BI sum scores than men and this was inversely related to the SF-36 General Health, Mental Health and Vitality scores. The authors concluded that women with CD adhering to a GFD perceive the disease burden to be worse compared to men and the perception of restriction is a prominent feature of the disease burden. As the authors state, the weakness of the study is the lack of a control group that shares key features of CD.

In a large cross-sectional survey Green et al (2001) aimed to investigate the clinical spectrum of adults with CD in the USA. They distributed a multiple choice questionnaire in a newsletter directly to CD support groups and via the internet. The survey focused predominantly on the general characteristics of respondents rather than evaluating psychological well-being and/or HRQoL. However, to assess the effects of adhering to a GFD, the authors devised a single-item question concerning self-perceptions of QoL before and after diagnosis. Quality of life before diagnosis was rated as bad by 30%, fair 33%, good 24% and excellent 10%. After diagnosis QoL was reported to be improved by 77%, unchanged by 15% and worse by 8%. The majority of participants had a long duration of symptoms before the diagnosis was confirmed, probably due to the perception among clinicians that the disease is rare. Due to the limitations of the one-dimensional, non-

validated measure used, it is not possible to draw any firm conclusions from this survey concerning HRQoL.

Mustalahti and colleagues (2002), evaluated the effect of a GFD on the HRQoL of a small sample of individuals with screen-detected CD and those with symptom-detected disease, before and one year after initiation of a GFD. Participants completed the GSRS to assess symptoms and the PGWB to measure QoL. At baseline the authors found that participants with symptom-detected disease had poorer QoL and more gastrointestinal symptoms than those with screen detected CD. Reported compliance with the GFD was good in both groups (93% symptom detected and 95% screen-detected). HRQoL for participants in both groups was significantly improved after one year of a GFD. Similarly, GSRS scores were also improved in both groups. The authors concluded that a GFD was associated with improved HRQoL for participants with symptom-detected CD and those with screen-detected disease and that concerns about the burden of a GFD may be unfounded, at least over the short-term (Mustalahti Lohiniemi, Collin, Vuolteenaho, Laippala & Mäki, 2002). They also acknowledge that the improvements in QoL after the first year of a GFD may not persist and that a longer follow-up period would be necessary to explore this. Furthermore, they have used the PGWB to measure QoL which, as its name suggests, is probably more appropriate for assessing 'psychological well-being' than QoL especially, as it does not contain physical and social items that are accepted core dimensions of HRQoL and is loaded more towards affective items.

Usai et al (2002) carried out a study to evaluate whether HRQoL in adult CD is related to severity of illness i.e. number of symptoms at diagnosis; compliance to a GFD and the presence of associated diseases. Compared to healthy controls, participants with CD obtained worse scores on all domains of the SF-36. Compliers (59.1%) who had been gluten free for at least 2 years, showed better results than non-compliers. The lowest

scores were achieved by participants with more than six symptoms. Participants with two or more associated diseases (in particular autoimmune thyroid disorders) obtained significantly worse scores than participants with only one associated disease. On the basis of their results, the authors suggest that the role of adherence to a GFD on HRQoL appears to be more relevant in people with fewer symptoms than in more severe cases, as in these cases it is likely that the natural course of the disease is only partially modified by a GFD. In addition, they emphasise that CD in adults is frequently complicated by the presence of symptoms not necessarily related to the intestinal pathology and not responding to GFD alone. However, the authors acknowledge that the limited number of subjects and the retrospective design of the study do not allow any definitive conclusions to be made.

In a large cross-sectional survey (Ciacci et al, 2003) members of five Italian regional coeliac societies who were on a GFD for at least one year completed a partially validated questionnaire (SAIC: Self-Administered Inventory for Coeliacs) devised by the authors to investigate QoL. It was found that females adhered to a GFD more strictly than males. However, this difference was no longer significant when compliance score was corrected for age at diagnosis. Individuals diagnosed after the age of twenty years had better dietary compliance than those diagnosed earlier, and this remained after correcting for gender and age. Self-reported compliance in all participants was 74.1%. Happiness scores were higher in participants diagnosed before 20 years of age. Although levels of anxiety and depression were low, anxiety was related to feeling different and depression to an unsatisfactory sex life. The authors concluded that in general, CD was not associated with a low level of self-perceived quality of life in members of the Italian Coeliac Society. A major limitation of the study is the instrument developed by the authors for which no measure of internal consistency is reported. Using a newly developed measure also makes it difficult to compare

the results with previous studies that have used well-established standardised measures of HRQoL.

Johnston, Rodgers and Watson (2004) measured HRQoL in two groups of out-patients with different types of CD and a control group. The SF-36 was completed prospectively by all participants who agreed to have a duodenal biopsy as a follow-up to a serological screening programme. Another group of 'typical' coeliac patients also completed this measure at diagnosis. The SF-36 was repeated in both groups after one year on a GFD. The authors found no significant differences between the SF-36 scores of the screen detected coeliac group and those of healthy controls at baseline and at follow-up. However, three SF-36 sub-scales (general health, vitality and role emotional) were significantly lower in 'typical' coeliac patients compared to controls and two of these (general health and vitality) improved significantly at one year follow-up compared to the baseline data. General health significantly improved in 'typical' coeliac patients but not screen-detected patients after one year on a GFD. However, the authors acknowledge that the small numbers in their study may not have been sufficient to detect a significant difference.

In a large cross-sectional survey Zarkadas et al (2006) evaluated the impact of a GFD on members (with biopsy-proven disease) of the Canadian Coeliac Association (CCA). A postal survey was used to measure QoL using the SF-12 and coeliac specific questions devised by the authors. It was found that mean SF-12 scores were similar to normative Canadian data, but were significantly lower for females and newly diagnosed participants. Ninety percent of participants reported adherence to a GFD. The authors concluded the QoL in adults with CD could be enhanced with early diagnosis, increased availability of GF foods and improved food labelling. Limitations of the study include the lack of a matched control group and probable sample selection bias due to targeting only members of the CCA.

Casellas et al (2008) recently investigated factors impacting on HRQoL in adults with CD in a multicentre study. Two groups of participants, those controlled with a GFD and newly diagnosed individuals on a normal diet, completed two HRQoL questionnaires (the GIQLI and EQ). The results of these measures indicated that the HRQoL of untreated, recently diagnosed individuals with CD was significantly impaired on almost all dimensions and overall scores compared to the GFD group who had EQ scores that were comparable to the general Spanish population. In addition, female gender, non-adherence to a GFD and symptomatic status were significantly associated with poorer HRQoL score. HRQoL was re-assessed in recently diagnosed individuals at least six months after starting a GFD and GIQLI scores were significantly improved. The authors concluded that HRQoL improves to levels similar to those in the general population in individuals with CD controlled with a GFD for at least six months. However, the number of individuals assessed before and after commencing treatment was very small. The authors acknowledge that another limitation of the study is probable selection bias due to only including participants from a hospital population.

Of the nine studies described above, three were prospective in design. In general the results of the studies using the SF-12/36 showed a reduced HRQoL of life in individuals living on a GFD. However, in the Canadian and Swedish (retrospective) studies this impoverishment was confined only to females. In the UK prospective study, scores for 'typical' coeliac patients improved after one year of adhering to a GFD, unlike screen-detected (asymptomatic) individuals whose scores remained comparable to healthy controls before and after starting a GFD. Similarly, in the Finnish prospective study, individuals with symptom-detected disease had poorer PGWBI scores than those with screen-detected CD. However, both groups improved after one year of a GFD. In the Spanish prospective study, the authors found that compared to individuals controlled by a GFD, untreated recently

diagnosed individuals have impaired HRQoL. They also found an association between female gender and poorer HRQoL.

Studies using self-report instruments of HRQoL and psychological distress

Fera, Cascio, Angelini, Martini & Guidetti (2003) investigated the prevalence of self-reported depressive symptoms and HRQoL in adults with CD on a GFD. Three groups of participants: those with CD (on a GFD for at least a year) individuals with diabetes and healthy controls, completed the M-SDS, the STAI and SF-36. The IBQ was only completed by the CD group and patients with diabetes. Amongst those with CD the mean duration of GFD was 9.1 ± 8.7 years (range 1-32). It was found that average M-SDS scores were significantly higher in the CD group than in healthy controls but there were no differences when compared to the participants with diabetes. On the state scale of the STAI participants with CD and those with diabetes had significantly higher scores compared to healthy controls. Duration of GFD was associated with significantly higher depression scores in those with CD who had a more recent diagnosis (>1 and ≤ 3 years). HRQoL was reduced in individuals with CD and those with diabetes compared to healthy controls and significantly correlated with state anxiety scores. The IBQ showed a high psychological and somatic perception of illness in individuals with CD and those with diabetes and its sub-scales were significantly correlated with anxiety. The authors emphasise that their study found the same depression rates amongst individuals with CD and those with diabetes which may indicate that depression is a consequence of chronic disease requiring dietary restrictions. However a longitudinal study with a homogeneous sample would be needed to investigate this hypothesis further.

Using the PGWBI and GSRS, Viljamaa and colleagues (2005) investigated dietary compliance and QoL after long-term treatment (median 14 years) in those with CD identified

by serological screening. Comparisons were made between symptom detected individuals, those who were untreated and healthy controls. The SF-36 was used to assess HRQoL, but was only completed by the screen-detected individuals and then compared to a Finnish population sample. GFD adherence rates were high (96% in screen-detected and 93% in symptom detected individuals) and were not related to age at diagnosis. PGWBI total and sub-scale scores in screen-detected individuals did not differ from those of either symptom-detected individuals or non-coeliac controls. A curious finding was that SF-36, average mental health sub-scale scores were significantly better in screen-detected participants than in the general population. Total GSRS scores were lower (non-significant) in screen-detected individuals than in symptom-detected group and healthy controls, indicating fewer symptoms. The authors conclude that QoL in screen-detected individuals was comparable with that in symptom detected individuals (Viljamaa Collin, Huhtala, Sievänen, Mäki and Kaukinen 2005). However, the SF-36 a robust and commonly used measure of HRQoL was not administered to the latter group, only the PGWBI which arguably is more a measure of psychological well-being. Therefore their conclusions cannot be fully justified.

In a national survey (Hauser, Gold, Stein, Caspary & Stallmach, 2006) a set of questionnaires including the SF-36 and HADS were posted to members of the German Coeliac Society (GCS). Compared to representative samples from the German population, the participants had higher scores for anxiety but not depression on the HADS. The rate of participants with a probable psychiatric disorder was not significantly different from the general population. In all sub-scales of the SF-36 except 'physical function' participants with CD had a significantly reduced HRQoL than population comparisons. The authors acknowledge some limitations of their study. Firstly, due to their sample being recruited from the GCS they cannot rule out a possible selection bias. Secondly, there could also be a response bias of CD patients with a reduced HRQoL sending back the questionnaires.

Nachman et al (2008) prospectively assessed differences in HRQoL and depression scores of individuals with CD to assess time-course impact of a GFD. They used the SF-36, the GSRS and BDI at diagnosis and at 3 and 12 months follow-up on treatment. Newly diagnosed individuals with classical symptoms were compared with atypical/silent cases and healthy controls. At baseline (diagnosis) both the classical symptom and atypical/silent groups had significantly lower SF-36 sub-scale scores compared with healthy controls except for 'Role Emotional' in the atypical/silent group. On the BDI compared with controls, both groups with CD had significantly higher baseline scores. On both the BDI and SF-36 atypical/silent cases faired significantly better than the classical group. In general, the GSRS scores of the two CD groups were significantly worse at baseline compared with controls. Compared with atypical/silent cases, those with classical CD revealed significantly higher baseline scores for four out of five items. Treatment (GFD) produced a substantial and rapid (3-month) improvement on most outcome measures in individuals with classical and atypical disease, but not in silent cases (n=10). After one year on treatment both CD groups achieved comparable final scores on all measures in line with normative scores. The authors state that their study shows that at diagnosis individuals with atypical/silent CD have a significantly better HQoL than those with classical symptoms. They acknowledge that a weakness of this study is the small number of cases with 'silent' disease' two of whom dropped out at final follow-up.

From the above studies, two out of the three that were retrospective in design found that scores for HRQoL were reduced compared to healthy controls/population comparisons, whilst the other found that screen-detected individuals with CD were no worse off than general population controls. The prospective study found that prior to starting a GFD, individuals with classical symptoms had lower HRQoL scores than those with atypical/silent disease – both groups having significantly reduced scores compared to healthy controls.

After one year on treatment, HRQoL scores improved in both groups in line with normative levels.

In the Italian retrospective study, depression scores were higher in the CD group compared to controls. In the Argentinean prospective study depression scores for both the classical and atypical/silent groups were higher than controls prior to starting a GFD, with the atypical group having better scores than the classical group. Both groups improved after 1 year follow-up on GFD. In the Italian study, significantly higher state anxiety scores were found in individuals with CD compared to controls. Higher scores for anxiety but not depression were found in the German survey. In the Finish study psychological well-being scores in screen-detected individuals did not differ from those in symptom detected individuals or non-coeliac controls.

Summary and Discussion

A total of 21 studies were reviewed and all except one (Hallert et al, 2002) had samples biased towards women. As emphasised in the introduction, HRQoL is a multi-dimensional concept that has developed from the need to assess the individual's subjective evaluation of the psychosocial impact of disease, including subjective health status and self-reported emotional/psychological well-being. In the reviewed literature, some of the studies aimed to assess key elements of HRQoL such as psychological well-being/distress and psychopathology in relation to CD and others used generic multi-dimensional measures that tapped various aspects of HRQoL such as physical functioning (e.g. physical pain and role limitation) and mental health (e.g. emotional vitality and social functioning).

To a small extent, the literature distinguished between chronic illness generally and coeliac disease. For example, there were 3 studies that used control groups of those with other chronic conditions namely, diabetes and chronic persistent hepatitis (CPH). Results

indicated that the perceived disease burden of individuals with diabetes and CD treated for a mean of 10 years was similar. In addition, the same rates of depressive symptoms were found amongst those with diabetes and CD (significantly higher than healthy controls) leading the authors to conclude that symptoms of depression are a consequence of chronic disease that requires dietary restrictions. Compared to individuals with CPH and 'normal' controls it was found that those with CD had significantly higher rates of self-reported depressive symptoms, thus supporting the former theory.

Overall studies tended to focus on the psychosocial aspects of living with CD itself and the specific effects of maintaining a GFD. Those that looked at the effects of a GFD distinguished between individuals who had recently been diagnosed and started a GFD and those who had been living on a GFD for a number of years having been diagnosed for some time. For example, results from 3 prospective studies revealed that asymptomatic individuals had better HRQoL scores than symptom-detected (classical) individuals at diagnosis. However, after one year on a GFD these scores were comparable for both groups. However, retrospective studies have found evidence for reduced HRQoL and poor psychological well-being in individuals treated for 10 & 14 years. Therefore, longer-term prospective studies are needed to explore persistence in improvements on a GFD.

The retrospective design of the majority of the studies makes them open to risk of selection bias and makes it difficult to ascertain cause-and-effect, because of various possible confounding factors. They also rely heavily on the accurate recall of the participants. Difficulties when interpreting and comparing the results of the reviewed literature apart from the different measures used include differences in: sample populations, country of origin, age group, CD type, duration of GFD and adherence rates. Furthermore, some studies were mostly descriptive in nature and others employed multivariate logistical techniques.

All the studies reviewed used translated, self-report instruments that may bias answers to sensitive questions such as dietary compliance. However, anonymity of the data would have minimised such bias. The most consistently used measure the SF-36 is one of the most commonly used, robust instruments for the study of HRQoL. However, it should be stressed that it is a generic tool not specifically designed for the study of CD. There is a need for further long-term prospective studies using larger samples, multivariate analysis and more specific standardised HRQoL measures. Several of the studies focussing on psychological distress implied that they measured affective/psychiatric disorder. However, these studies used self-report instruments rather than formal psychiatric interviews, so that no conclusions can be drawn from them about the prevalence of psychiatric/affective disorders in people with CD.

In three studies (one prospective) female gender was linked to poor HRQoL scores and in another reduced psychological well-being. A Swedish study later found that women with CD adhering to a GFD perceived their disease burden to be worse than men and coped less well with the social inconvenience to their daily activities.

Reduced HRQoL was consistently found in studies using the SF-36 a generic self-report instrument for assessing HRQoL however, only two were prospective and both these had small sample sizes. The presence of self-reported depressive symptoms was also invariably found using the M-SDS and one prospective study found evidence for persistence. The prevalence of anxiety is less clear cut due to different measures being used in each study. Two prospective studies found anxiety before, but not after commencement of a GFD and two retrospective studies found high anxiety scores in individuals with CD compared to controls. A further study using the PGWBI found no indications of anxiety.

Conclusions

The findings of this review suggest that in addition to reduced HRQoL, psychological distress, especially symptoms of anxiety and depression is commonly found in people with CD. Although anxiety can be experienced, the literature suggests that this tends to decrease on a GFD. However, several studies found that depressive symptoms and poor HRQoL persisted even in treated individuals. This may be due to anxiety in people with CD being mainly related to the presence of disabling symptoms such as abdominal pain, diarrhoea and weight loss that tend to dissipate on starting a GFD thus, reducing the anxiety; whereas depressive symptoms might be sustained by the reduction in quality of life (supported by several studies) due to dietary restrictions and related changes in social habits and lifestyle, perhaps more so in women.

Implications for clinical practice

In addition to reduced HRQoL, psychological distress, especially symptoms of persistent depression, is commonly found in individuals with CD. Depression can be a major reason for non-adherence to treatment in people with chronic medical diseases (DiMatteo, Lepper & Croghan, 2000). Therefore, it would seem prudent to screen men and women with CD for probable mood disorders so that appropriate psychological support can be provided which has been shown to reduce depression and improve GFD adherence (Addolorato, Lorenzi, Abenavoli, Leggio, Capristo & Gasbarrini, 2004).

A better understanding of the psychosocial effects of CD could enhance clinical management and ultimately improve the quality of life for adults with the disease. In particular, studies are needed to explore the extent to which clinically significant levels of emotional distress are accounted for by lack of psychological help to support individuals living on a gluten-free diet.

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Living with Coeliac Disease: A Cross-sectional Survey of a UK Population.

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ABSTRACT

Coeliac Disease (CD) is an incurable autoimmune condition managed by a therapeutic gluten-free diet for life. European studies suggest that the chronicity of CD, the limitations imposed by the need to follow a permanent restrictive diet and the risk of other associated serious diseases can have a negative impact on health-related quality of life (HRQoL) and psychological well-being. However, studies concerning the psychosocial effects of CD in the UK population are scarce. This postal survey explored the illness perceptions and self-efficacy beliefs of adults with CD in the UK and reports their subjective levels of HRQoL and psychological well-being. Questionnaires were returned by 288 members of Coeliac UK. In this sample, HRQoL and psychological well-being were found to be reduced with levels being comparable to those found in previous related studies. Participants with weak beliefs in the serious consequences of CD and reduced emotional reactions to the condition had a greater likelihood of having enhanced HRQoL, improved psychological well-being and increased self-efficacy. Strong beliefs in personal control over the condition and a greater perceived understanding of CD were also associated with increased self-efficacy. The results suggest that perceived self-efficacy and illness perceptions could play an important role in informing psychological interventions for individuals with CD.

Key words: Coeliac Disease; Gluten-free diet; Psychological Well-being; Illness perceptions; Self-efficacy; Health Related Quality of Life.

INTRODUCTION

Coeliac Disease (CD) is a chronic autoimmune disorder in which hypersensitivity to gluten causes damage and inflammation to the small intestine in genetically susceptible individuals (Fera, Cascio, Angelini, Martini & Guidetti, 2003). Those with an untreated condition experience intestinal malabsorption due to partial or total atrophy of the tiny finger like projections (villi) on the surface of the small intestine (Jones, 2007). The condition is also associated with osteoporosis, and fertility problems in women, type I diabetes (Feighery, 2007) and an increased risk of gastrointestinal cancer and non-Hodgkin's lymphoma (West, Logan, Smith et al, 2004). It is estimated that CD may affect up to 1 in 100 people in Western European populations, although many individuals remain undiagnosed (Hopper et al, 2007). Diagnosis is usually achieved through a screening blood test followed by a biopsy of the small intestine to detect villous atrophy. CD can occur at any age, but in adults the peak incidence is in the fifth decade and females are more commonly affected than males (Jones, 2007).

The condition is incurable but is managed by a therapeutic gluten-free-diet for life. A gluten-free diet (GFD) involves the complete avoidance of all foods made from or containing wheat, rye, barley and usually, oats such as bread, pizza, biscuits, cake, pasta and cereals. This diet is very successful in managing the symptoms of CD as the removal of gluten allows the villi to re-generate therefore leading to the normal absorption of nutrients. A GFD also protects against non-Hodgkin's lymphoma, corrects anaemia, restores normal nutritional balance, and substantially improves quality of life, particularly if unpleasant gastrointestinal symptoms have been present (Häuser, Gold, Stallmach, Caspary & Stein, 2007).

Although the literature on the immunology and physiopathology of CD is now extensive (Kagnoff, 2005; Barone et al, 2007) the impact of the condition from the individual's view is less well known. The chronicity of the condition, the limitations imposed by the need to follow a permanent restrictive diet and the risk of other associated diseases can have a negative impact on health-related quality of life (HRQoL) (Casellas et al, 2008) and psychological well-being (Addolorato et al, 2008).

It is therefore not surprising that, increasingly the focus of research is turning to the psychosocial impact of CD on those with the condition and their families (Fera et al, 2003; Hallert, Sandlund & Broqvist, 2003). Much of this research is concerned with psychological well-being and health related quality of life (HRQoL). European studies suggest that depression and lower quality of life affect individuals with CD, and anxiety and depression have been identified as major causes of lower levels of adherence to treatment recommendations (Addolorato, Stefanini, Capristo, Caputo, Gasbarrini & Gasbarrini, 1996) and poor adaptation to the disease (Ciacci, Iavarone, Mazzacca & De Rosa 1998). There is a suggestion from the existing literature that women with CD experience poorer quality of life than their male counterparts. A Swedish 10-year follow-up study (Hallert et al, 1998) for example, found that adult female coeliac patients scored significantly lower than the general population on a subjective measure of health, specifically within the domains of General Health and Vitality. A later extension of this Swedish study (Hallert et al, 2003) found that women with long-standing CD reported worse health-related quality of life than men.

However, studies on the impact of a gluten-free diet (GFD) on HRQoL have produced conflicting results. For example, US-American (Green et al, 2001), Canadian (Zarkadas et al, 2006) and Swedish studies (Roos, Karner & Hallert, 2006) report an average HRQoL for adult celiac sufferers comparable with the general population; whereas

studies conducted in Italy (Fera et al, 2003) Northern Ireland (O'Leary et al, 2002) and Germany (Häuser, Gold, Stein, Caspary & Stallmach, 2006) demonstrate a reduced HRQL compared with the general population or healthy controls.

To date, various factors have been identified as being associated with reduced HRQoL in adults with CD including younger age at diagnosis, anxiety (Ciacci, Iavarone, Siniscalchi, Romano & De Rosa, 2002) being newly diagnosed (Zarkadas et al, 2006) latency of diagnosis (Usai et al, 2002) poor adherence to a gluten-free diet (Fera et al, 2003) and somatic and psychiatric comorbidity (Häuser et al, 2007).

In a study looking at the emotional impact of CD and coping in adulthood (Ciacci, et 2002) the authors found that relief was the most intensive feeling after diagnosis. There was a positive correlation between feeling different and sadness, anger and fear. The strongest correlation was found between anger and compliance with a gluten-free diet. These authors identified a 'depressive-anxiety factor' as the main indicator of psychological disturbances in a series of long-treated young adult patients. In a study of 100 patients treated for 8 years, Fera and colleagues (2003) found a high rate of affective disorders that increased with duration of treatment and suggested a close relationship to reduced quality of life a common finding in long-treated adults with CD (Hallert et al, 1998; Lee & Newman, 2003).

In a German national survey (Hauser et al, 2006) compared to representative samples from the general population, the participants had higher scores for anxiety but not depression. The rate of participants with a probable psychiatric disorder was not significantly different from the general population. In a Swedish study (Roos et al, 2006) the authors found that compared to a healthy control group, patients with CD showed no more signs of anxiety, depressed mood or distress. However, unlike controls women with CD showed significantly reduced psychological well-being than males.

More research is needed to determine whether affective disorders and reduced quality of life are a feature of CD. At present there is a dearth of studies about the psychosocial effects of CD in the UK population. Knowledge of the prevalence of psychological distress in the UK coeliac population is important for clinical management, particularly as there is evidence from Italy that psychological counselling can improve adherence to a gluten-free diet in coeliac patients with affective disorder (Addolorato, Lorenzi, Abenavoli, Leggio, Capristo & Gasbarrini, 2004). A better understanding and greater knowledge of the psychosocial effects of CD on sufferers could enhance the clinical management of the condition and ultimately improve the quality of life for adults with the disease.

The two important concepts of illness representation (Leventhal, Nerenz & Steele, 1984; Petrie & Weinman, 1997) and self-efficacy (Bandura, 1977; 1997) feature prominently in research concerning responses to and coping with chronic illness. However, at present there has been no investigation of these concepts in relation to CD. Knowledge of these is important for informing therapeutic interventions, to help in the clinical management of the disease.

The illness representation or self-regulatory model (SRM) is the most widely used in the last decade to explain how people interpret current and potential health events or threats. It describes patients as active problem solvers whose health related behaviours are based upon, and then regulated or influenced by the representations or personal beliefs they generate about illness (Leventhal et al, 1984). The fundamental premise of the model is that people are motivated to regulate or minimise their health-related risk and act to decrease health threats in ways consistent with their perceptions of them. Sources of knowledge on which perceptions are based include the mass of cultural illness information (environmental stimulus), individual personal illness experience (perceptual symptoms) and social communication (Leventhal et al, 1984). The constant interaction of

environmental and perceptual stimuli within people's memory systems explains why different people construct different representations and devise different action plans to respond to similar medical conditions (Lau-Walker, 2006).

The SRM emphasises that individuals construct a belief about themselves as well as their condition. To be successful therefore, therapeutic interventions need to take account of and use these beliefs. In other words, not only do individuals contemplate what is happening to them and the future consequences of their condition, they also have a well established construct of themselves, based on their interpretation of their own experiences. This influences what they believe they are, or are not capable of acting upon to respond to their current health condition (Lau-Walker, 2006).

A person's 'common sense' illness representations have been shown to predict decisions to comply with medical advice and to cope successfully with chronic illness (Moss-Morris, Petrie & Weinman, 1996). In a study looking at illness representation and outcome in irritable bowel syndrome (Rutter & Rutter, 2002) the authors found that the reporting of serious perceived consequences was associated with lower quality of life and higher scores for anxiety and depression. Weaker control beliefs were also related to lower quality of life and higher depression scores.

Self-efficacy refers to the same theme as the illness representation model i.e. the belief that an individual has a well-established construct of themselves, based on their own interpretation of their experiences. Perceived self-efficacy is concerned with people's beliefs in their capabilities to perform a specific action required to achieve a desired outcome. The original theory was developed by Albert Bandura (1977) in the context of cognitive behaviour modification. He asserts that perceived self-efficacy is not a measure of the skills one has, but a belief about what one can do under different sets of conditions with whatever skills one possesses. Bandura (1982) distinguished between two types of

expectations: outcome expectation and self-efficacy. Outcome expectation is the belief that certain behaviours will lead to a particular outcome, and self-efficacy reflects the belief that one can successfully perform these behaviours to produce the outcome. Self-efficacy beliefs determine the initial decision to perform a behaviour, the effort to be expended and persistence in the face of adversity. For example, an individual may believe that regular exercise will improve his/her future health (high outcome expectancy) but may reject this strategy as they have a low efficacy expectancy (never having been a regular exerciser they will not see themselves as able to start regular exercise and will not believe they have the ability to sustain it). Therefore, generic educational material on diet that focuses only on improving health outcomes rather than addressing individuals' confidence in being able to sustain the diet is unlikely to effectively strengthen their self-management abilities.

It has been found that a strong sense of personal efficacy is related to better health, higher achievement and more social integration (Schwarzer & Fuchs, 1995). In order to promote individuals' self-efficacy for managing a long-term health condition, it is important that clear, precise and specific knowledge and competence in relevant skills are provided to them to support their own management of their particular condition (Lau-Walker & Thompson, 2009). In general, people's self-efficacy beliefs influence the health-related choices they make, the health related goals they set for themselves and the amount of effort they use to reach these goals (Wallston, Rothman & Cherrington, 2007). For example, patients with diabetes who adhere to dietary advice and other self-management tasks are more likely to report feeling competent to self-manage their diabetes (e.g. Talbot, Nouwen, Gingras, Gosselin & Audet, 1997). Based on this, it was hypothesised that adherence to a gluten free diet by individuals with CD would be associated with a strong sense of self-efficacy. It was also hypothesised that those participants with favourable

illness perceptions would have higher levels of perceived self-efficacy to manage their condition. Although illness perceptions and self-efficacy have been independently constructed they have a common theme at their core. Each posits that individuals' personal constructs of their condition and of their ability to cope with that condition are at the basis of effective self-management. Furthermore, both concepts argue that it is through an individual's accumulated experience, rather than personality, that their actions and perceptions are informed. Both theories acknowledge that individuals interpret the events that affect them and construct responses and future outcomes from a rational base that is unique to each individual (Lau-Walker, 2004). In view of the fact that there is considerable overlap within the two theoretical concepts it seems likely that there will be a relationship between the components of the two models and more specifically that illness representations will be predictive of self-efficacy.

This study has three main aims as follows:-

1. To investigate gender differences in quality of life and sense of well-being in adults with CD in the UK
2. To explore the illness perceptions and self-efficacy beliefs of adults with CD in the UK
3. To explore the relationship between individuals' personal sense of control or self-efficacy and perceptions of illness, and their well-being and quality of life.

METHOD

Design and Procedure

The study design was a cross-sectional postal questionnaire. Adult members (aged 18 years and over) of Coeliac UK (the national UK charity supporting people with CD) were invited to participate in a questionnaire survey designed to investigate the psychological and social effects of living with CD. The survey was advertised in the quarterly Coeliac UK magazine. The advertisement provided a contact telephone number for potential interested participants to ring and leave an address so that the survey pack could be sent to them. Each pack contained an information sheet, consent form, five questionnaires and a pre-paid envelope for postal return (a copy of the forms in the pack can be found in Appendix 6). Some questionnaire packs were also distributed at local Coeliac UK support meetings. Members of Coeliac UK were chosen as potential participants because all members of the Society have been medically diagnosed with CD. This was an attempt to ensure that those experiencing symptoms of CD, but who had no formal diagnosis were excluded from the study.

The information sheet detailed what the study was about including: who was taking part; the benefits and any risks; the right to withdraw; what would happen to the information; support networks if some questions were distressing and what was expected of participants. After reading this, participants were asked for their written consent to take part in the study before completing the five questionnaires. These included a social demographic questionnaire, the Perceived Medical Condition Self-Management Scale (PMCSMS) (Wallston et al, 2007) the Revised Illness Perception Questionnaire (IPQ-R) (Moss-Morris, Weinman, Petrie, Horne, Camerson & Buick, 2002) the General Well-Being Index (British adaptation of the Psychological General Well-being Schedule) (Hunt & McKenna, 1992) and the Celiac Disease Questionnaire (CDQ) – a Health Related

Quality of Life index (Häuser et al, 2007). Participants were asked to post back all the forms in the pre-paid envelop including the signed consent form. It was stressed in the information sheet that the questionnaires would be treated with complete confidentiality.

Ethical approval was obtained from the University of Birmingham, School of Psychology Research Ethics Committee (see Appendices 4 & 5).

Participants

The majority of participants were women (80%) and of White British origin (95%). Men were significantly older than women (mean difference 8.61; Z= -4.08; P= <0.001). Adherence to a gluten free diet (GFD) was high with only 13% of participants reporting that they did not adhere all the time. In general, the number of years since diagnosis corresponded with the duration of membership in Coeliac UK. Over half the sample (57%) had received their diagnosis in their forties and fifties. The majority of participants were well educated and had professional occupational status. Full sample characteristics are presented in Table 1.

Exclusion Criteria

Adults experiencing symptoms of CD, but who had no formal diagnosis were excluded. Symptoms of CD can be similar to other gastrointestinal diseases; therefore to ensure that the sample contained only those suffering from CD, they were not included unless they had a medical diagnosis. The study focused on adult sufferers; therefore, those aged below 18 years were excluded.

TABLE 1 Sociodemographic Characteristics of Study Sample (n=284)

Variable	**Number (%)**	**Mean (SD)**	Range	Median	**25th – 75th Percentile**
Sex (female)	227 (80.0)				
Age All		54.0 (14.6)	19-85	56	44-65
Male		61.0 (13.6)	23-85	63	53-70
Female		52.3 (14.4)	19-84	54	43-63
Duration of membership in Coeliac UK					
<1-5 yrs	133 (46.8)				
6-20 yrs	103 (36.3)				
>20 yrs	48 (16.9)				
Years since diagnosis					
<1-5 yrs	127 (44.7)				
6-20 yrs	103 (36.3)				
21-40+ yrs	53 (18.6)				
Age at diagnosis					
<1-20 yrs	34 (12.1)				
21-40 yrs	85 (30.1)				
41-50+ yrs	163 (57.8)				
Adherence to a GFD					
All of the time	246 (86.6)				
Most/some of the time	37 (13.4)				
Marital status					
Married/co-habiting	210 (74.0)				
Separated/divorced/widowed	36 (12.8)				
Single (never married)	36 (12.8)				
Highest educational level					
No qualifications	36 (12.8)				
Secondary School	63 (22.3)				
Vocational training	67 (23.6)				
University degree	116 (40.8)				
Occupational status (previous or current)					
Professional	137 (48.9)				
Managerial/technical	71 (25.4)				
Non-manual skilled	21 (7.4)				
Manual skilled/partly skilled	24 (9.0)				
Non-skilled/home-maker	27 (9.7)				

Measures

The social demographic questionnaire contained items related to participants' age, sex, ethnicity, educational level, occupational and marital status, age at diagnosis and adherence to a gluten-free diet (see Appendix 6).

Perceived Medical Condition Self-Management Scale (PMCSMS)

A Coeliac Disease-specific adaptation of the Perceived Medical Condition Self-Management Scale (PMCSMS) was used to assess the degree to which the participants felt competent or self-efficacious in managing their CD. The PMCSMS is an 8-item measure based upon the Perceived Health Competence Scale (Smith, Wallston, & Smith, 1995). It was developed as a template that could be made disease-specific and used with any medical condition requiring self-management. It has been successfully adapted for use with patients with diabetes (type 1 and type 2) and was found to be a reliable and valid measure (Wallston et al, 2007). Responses are rated on a 5-point Likert scale from 'strongly disagree' to 'strongly agree'. Higher scores indicate stronger perceptions of self-efficacy.

The Revised Illness Perception Questionnaire (IPQ-R)

The original Illness Perception Questionnaire (Weinman, Petrie, Moss-Morris & Horne, 1996) was developed to provide a quantitative assessment of the five components (identity, consequences, timeline, control/cure and cause) of illness representation in Leventhal's Self-Regulatory Model (Leventhal et al, 1984; 1997). Central to this model are the representations (or illness cognitions) that patients have about their illness. The illness representation gives personal meaning to symptoms and it is argued acts as a framework for guiding coping efforts. The revised version (IPQ-R) includes a new subscale relating to emotional representations and divides control beliefs into personal

attempts to control illness and control of illness by treatment. It has demonstrated sound reliability, discriminant and predictive validity (Moss-Morris et al, 2002). The illness identity subscale measures the number of commonly experienced symptoms such as upset stomach that individuals associate with their illness. The consequences subscale measures individuals' beliefs about the seriousness of their condition. The timeline scale is divided into an acute/chronic subscale and a cyclical dimension that measures whether individuals view their illness as variable over time. The control subscale is divided into personal control, that refers to beliefs about one's own ability to control symptoms and treatment control that refers to beliefs regarding treatment (or diet) as an effective way of controlling the condition. The illness coherence dimension assesses the degree to which patients feel they have a coherent understanding of their illness. The final causal items can be divided into four main subscales: psychological causes such as stress and overwork, risk attributions such as diet and heredity, causes related to immunity such as a virus and chance attributions such as an accident or bad luck.

General Well-being Index (GWBI)

The Psychological General Well-Being Index originally developed in the US by Harold Dupuy (1984) was adapted for use in Britain by Hunt & McKenna (1992) and renamed the General Well-Being Index (GWBI). This instrument assesses emotional well-being in individuals whose illnesses are not so much physically incapacitating as psychologically distressing such as epilepsy, diabetes and it is argued CD. It provides a self-report of intrapersonal affective states that reflect subjective well-being or distress. The index consists of questions that cover six affective states: anxiety, depressed mood, feelings of positive well-being, self-control, general health and vitality. The adapted measure has been shown to have good psychometric properties while being short, easy to

use and acceptable to participants (Hunt & McKenna, 1992). Responses are rated on a 5-point Likert scale from 'strongly disagree' to 'strongly agree'. The approach adopted in this study was to score the items so that higher scores indicated better psychological well-being.

The Coeliac Disease Questionnaire (CDQ)

The Coeliac Disease Questionnaire (CDQ) is a reliable and valid disease specific instrument for measuring health-related quality of life in adult patients with CD (Hauser et al, 2007). Recently developed in Germany the index has been translated into English. The CDQ comprises four subscales: gastrointestinal symptoms, emotional well-being, social restrictions and disease-related worries. Responses are rated on a 7-point scale from 'strongly disagree' to 'strongly agree'. High scores indicate a good HRQoL.

Data Analysis

The data were analysed using the Statistical Package for the Social Sciences (SPSS 15 for Windows).

Reliability analyses were carried out on the four questionnaires using Cronbach's alpha (see Table 2 overleaf). These indicated a high level of internal consistency for all the measures except four subscales belonging to the IPQ-R, namely: Timeline (acute/chronic); Treatment Control; Risk and Immunity. These items were removed from subsequent analyses.

The Kolmogorov-Smirnov test showed that most variables were not normally distributed, therefore non-parametric analyses were chosen (see Appendix 7). However, to allow comparisons with the results of other studies both medians and means are presented. Preliminary descriptive and univariate procedures were employed before bivariate tests of

association were carried out. Comparisons between variables were performed using the Mann-Whitney U or Fisher tests as appropriate. Binary logistic regression methods were employed to investigate the predictive strength of illness perceptions and other factors.

TABLE 2 Reliability coefficients for subscales of all measures

Measure and Sub-scale	Number of items	Cronbach's alpha
PMCSMS		
Total Scale	8	0.92
IPQ-R		
Identity	14	0.80
Timeline (acute/chronic)	6	0.51*
Consequences	6	0.79
Personal control	6	0.81
Treatment control	5	0.51*
Illness coherence	5	0.90
Timeline (cyclical)	4	0.92
Emotional representations	6	0.88
Psychological causes	6	0.87
Risk	6	0.67*
Immunity	3	0.42*
GWBI		
Positive well-being	4	0.86
General health	3	0.86
Depressed mood	3	0.91
Anxiety	5	0.85
Self-control	3	0.88
Vitality	4	0.87
CDQ		
Gastrointestinal symptoms	7	0.82
Emotional well-being	7	0.91
Social restrictions	7	0.85
Disease related worries	7	0.81

The dependent variables psychological well-being, HRQoL and self-efficacy were split at the median to create two equal groups of high scorers and lower scorers. Up to 25% of missing items on the PMCSMS, IPQ-R, GWBI and CDQ were replaced by the median of the items of the respective sub-scale. If more than 25% of the items of a subscale were missing the respective measure was excluded from further analysis.

RESULTS

Two hundred and eighty eight out of 433 (66%) questionnaires were received back from participating members of Coeliac UK. Four datasets had to be excluded as they were not accompanied by consent forms. A number of questionnaires were excluded as there were more than 25% missing items as follows:- GWBI 10, CDQ 8, IPQ-R 6 and PMCSM 4. In total, 14 participants were excluded from the main analyses.

Self-Efficacy

Table 3 shows descriptive data for the PMCSMS for adherence group and sex and the differences between each group. The mean scores for the total sample show a relatively high level of perceived self-efficacy. There was a significant difference in scores between the adherence groups. As predicted, those in the lower group had weaker perceptions of their own self-efficacy to manage their CD (Z= -2.0; P=0.04). Conversely, those in the higher adherence group had stronger beliefs in their ability to manage their condition. There was no difference in the level of perceived self-efficacy to manage their Coeliac Disease between men (mean= 32.2) and women (mean= 31.6).

TABLE 3 Perceived Self-Efficacy between Adherence Group and Men and Women

PMCSMS	**Total Sample** (n=274)		**Lower Adherence** (n=37)		**High Adherence** (n=237)	
	Mean (SD)	Median (Range)	Mean (SD)	Median (Range)	Mean (SD)	Median (Range)
	31.7 (5.9)	32 (14-40)	29.2 (7.2)	32 (15-39)	32.1*(5.6)	32 (14-40)
			Females (n=224)		**Males** (n=56)	
PMCSMS			Mean (SD)	Median (Range)	Mean (SD)	Median (Range)
			31.6 (5.9)	32 (14-40)	32.2 (5.7)	32 (14-40)

*****Z= -2.0; P=0.04

Psychological Well-being

Table 4 (below) shows descriptive data for the General Well-being Index (GWBI) for males and females and the differences between the two groups. The distribution of scores for the whole sample (not shown) ranged from 29 to 110 with a mean of 79.0 (s.d. 15.4) out of a possible top score of 110. Men tended to score slightly higher than women indicating better psychological well-being. These differences were significant for the total score and the following subscales: Anxiety, Depressed Mood and Self-Control. There was no significance in GWBI scores between adherence groups (Z= -0.30; P=0.76).

TABLE 4 Levels of Psychological Well-being between Men and Women

GWBI Subscale	Means (standard deviations) and Medians (ranges) **Females** (n=219)		**Males** (n=55)		**Difference test**
Anxiety	17.0 (4.0)	17 (6-25)	19.0 (3.9)	19 (10-25)	Z= -3.1; P=0.002*
Depressed Mood	11.0 (2.7)	12 (5-15)	12.2 (2.5)	12 (5-15)	Z= -3.0; P=0.002*
Positive well-being	14.4 (3.1)	15 (5-20)	15.3 (2.7)	16 (8-20)	Z= -1.7; P=0.08
Self-control	12.0 (2.3)	12 (5-15)	13.0 (2.0)	13 (8-15)	Z= -2.9; P=0003*
General Health	10.7 (2.8)	11 (3-15)	11.2 (2.9)	12 (4-15)	Z= -1.3; P=0.19
Vitality	12.5 (3.3)	12 (4-20)	13.0 (3.4)	13 (5-19)	Z= -0.9; P=0.36
Total Score	76.9 (15.4)	78 (29-110)	83.0 (14.7)	84 (48-108)	Z= -2.5; P=0.01*

*= significant difference

Health Related Quality of Life (HRQoL)

Table 5 (below) shows the mean and median scores for the Coeliac Disease Questionnaire (CDQ) for men and women and the differences between them. The distribution of scores for the whole sample ranged from 61 to 194 with a mean of 152.2 (s.d. 26.4) out of a possible top score of 196 (not shown) reflecting reduced HRQoL. Reduced HRQoL was defined by scores ≤ 10% percentile of the total CDQ score which was 10% of the sample. Men tended to score slightly higher on the CDQ than women, but the differences were only significant for the total scores and two sub-scales: Emotion and Social. There was no significant difference in CDQ total score and adherence group (Z= -1.25; P=0.20).

TABLE 5 Levels of HRQoL between Men and Women

CDQ	Means (standard deviations) and Medians (ranges)				
Subscale	**Females** (n=219)		**Males** (n=55)		**Difference test**
Emotion	33.4 (8.2)	37 (12-49)	36.0 (8.1)	37 (12-49)	Z= -1.9; P=0.04*
Social	40.2 (8.5)	46 (11-49)	42.0 (9.6)	46 (11-49)	Z= -2.0; P=0.04*
Worries	39.3 (8.1)	41(12-49)	40.6 (8.6)	43 (9-49)	Z= -1.5; P=0.12
Gastrointestinal	38.0 (7.4)	34 (7-49)	39.4 (7.0)	41 (24-49)	Z= -1.1; P=0.26
Total Score	150.9 (26.0)	156 (64-194)	157.7 (27.9)	164 (61-194)	Z= -2.1: P=0.03*

*= significant difference

Illness Perceptions

Mean scores for consequences (3.5, s.d. 0.82), personal control (4.3, s.d. 0.67) and illness coherence (4.2, s.d. 0.82) were high, reflecting a coherent understanding of CD, strong perceptions of personal ability to control it and strong beliefs about the serious consequences of CD. Participants did not attribute many symptoms to their CD, reflecting a low disease identity (mean 3.3, s.d.2.94). The most important cause identified by participants was genetic risk with over half (52.2%) attributing this to the development of their condition. Table 6 shows the mean and median scores for each of the included IPQ-R

subscales for men and women and differences between them. The results indicate that women had a significantly higher emotional response to their condition than men. Women also had significantly stronger beliefs that their CD was caused by psychological factors such as stress and mental attitude; however, there were no gender differences on the other dimensions.

TABLE 6 Mean and Median Scores for Illness Perceptions

IPQ-R	Means (standard deviations) and Medians (ranges)				
Subscale	**Females** (n=219)		**Males** (n=55)		**Difference test**
Identity	3.5 (3.02)	3.0 (1-12)	2.9 (2.58)	3.0 (1-12)	Z= -0.8; P=0.40
Consequences	3.5 (0.82)	3.6 (1.3-4.8)	3.5 (0.80)	3.6 (1.6-5.0)	Z= -0.2; P=0.83
Personal control	4.3 (0.62)	4.5 (2.1-5.0)	4.2 (0.84)	4.3 (1.0-5.0)	Z= -0.4; P=0.68
Illness coherence	4.2 (0.79)	4.4 (1.8-5.0)	4.2 (0.95)	5.6 (1.0-5.0)	Z= -0.1; P=0.90
Timeline cyclical	2.3 (1.10)	2.0 (1.0-5.0)	2.0 (0.99)	2.0 (1.0-4.5)	Z= -1.4; P=0.14
Emotional responses	2.6 (0.96)	2.5 (1.0-5.0)	2.2 (0.98)	2.0 (1.0-5.0)	Z= -2.7; P=0.005*
Psychological causes	2.0 (0.88)	2.0 (1.0-5.0)	1.7 (0.74)	1.5 (1.0-3.6)	Z= -2.0; P=0.04*

*= significant difference

Associations Between Illness Perceptions and Distress

Table 7 shows Spearman's R correlations between illness perceptions, age and self efficacy, measures of well-being and HRQoL for the whole sample. The majority of the coefficients are modest, lying between 0.40 - 0.65. Those lying between 0.19 – 0.39 are considered low (Cohen & Holliday, 1982). The lower the disease identity of participants the higher their self-efficacy, HRQoL and general well-being scores were. The weaker the beliefs of participants in the severity of their CD, the higher their self-efficacy, HRQoL and psychological well-being scores were. Stronger perceptions of personal control over the condition and a clearer understanding of CD were also associated with increased self-efficacy and improved HRQoL. The weaker participants' beliefs that CD was variable over time and the lower their emotional responses were to CD the higher their self-

efficacy, HRQoL and psychological well-being scores were. Total scores for the CDQ and GWBI were also strongly correlated (Spearman's Rho 0.76, P=0.01).

TABLE 7 Correlations (Spearman's R) between Illness Perceptions and Outcome Measures

Total Scores n= 274	**PMCSMS**	**CDQ**	**GWBI**
Age	0.19**	0.24**	0.24**
Identity	-0.37**	-0.57**	-0.44**
Consequences	-0.43**	-0.55**	-0.32**
Personal Control	0.46**	0.22**	0.14*
Illness Coherence	0.56**	0.42**	0.32**
Timeline cyclical	-0.49**	-0.54**	-0.49**
Emotional representations	-0.60**	-0.65**	-0.53**

*p = 0.01, **$p \leq 0.001$ *(based on the Bonferroni correction test, P values ≤ 0.002 are significant)*

Predicting General Well-being, HRQoL and Self-Efficacy

The results of binary logistic regression analyses to investigate predictors of general well-being, HRQoL and self-efficacy are presented in tables 8-10. Reduced scores on Timeline cyclical, Emotional representations, Consequences and older age led to a correct classification of an enhanced general well-being in 74% of the sample. This means that the lower the scores for Timeline cyclical, Emotional representations and Consequences the more likely it was that participants had higher general well-being scores. In addition the older participants were, the more likely they were to have a higher general well-being score. Low scores on Consequences, Illness coherence, Emotional representations and Identity led to a correct classification of better HRQoL in 79% of the sample. This means that the lower the scores for Consequences, Illness coherence, Emotional representations and Identity the more likely for participants to have a higher HRQoL. In the self-efficacy analysis sex was not entered as no univariate difference had been found (stated previously). Lower scores for Consequences and Emotional representations were associated with higher self-efficacy scores measured by the PMCSMS as were strong beliefs in Personal control

and Illness Coherence (clarity of understanding of CD). The correct classification rate for this model was 78%. The independent variables Consequences and Emotional representations were both predictive of all three outcome measures, general well-being, HRQoL and self-efficacy.

TABLE 8
Logistic regression analysis of Illness Perceptions predicting increased general well-being (GWBI)

Independent Variables	Odds Ratio	95% CI	B	P-value
Age	**1.03**	**1.00-1.05**	**0.31**	**0.007**
Sex (Male)	1.16	0.53-2.54	0.15	0.69
Identity	0.91	0.80-1.03	-0.08	0.16
Consequences	**0.94**	**0.87-1.00**	**-0.06**	**0.05**
Personal control	1.01	0.93-1.09	0.01	0.78
Illness coherence	1.02	0.93-1.12	0.02	0.63
Timeline cyclical	**0.82**	**0.74-0.90**	**-0.19**	**0.000**
Emotional representations	**0.89**	**0.83-0.96**	**-0.11**	**0.002**

TABLE 9
Logistic regression analysis of Illness Perceptions predicting increased HRQoL (CDQ)

Independent Variables	Odds Ratio	95% CI	B	P-value
Age	1.01	0.99-1.03	0.01	0.29
Sex (Male)	1.31	0.56-3.07	0.27	0.52
Identity	**0.82**	**0.71-0.94**	**-0.19**	**0.005**
Consequences	**0.86**	**0.79-0.94**	**-0.14**	**0.001**
Personal control	1.05	0.95-1.15	0.05	0.28
Illness coherence	**1.12**	**1.00-1.24**	**0.11**	**0.03**
Timeline cyclical	0.92	0.83-1.02	-0.07	0.14
Emotional representations	**0.87**	**0.81-0.94**	**-0.13**	**0.001**

TABLE 10
Logistic regression analysis of Illness Perceptions predicting increased Self-efficacy (PMCSMS)

Independent Variables	Odds Ratio	95% CI	B	P-value
Age	1.02	0.99-1.04	0.02	0.06
Identity	1.02	0.89-1.16	0.02	0.**77**
Consequences	**0.90**	**0.83-0.98**	**-0.10**	**0.01**
Personal control	**1.14**	**1.04-1.26**	**0.13**	**0.005**
Illness coherence	**1.31**	**1.17-1.48**	**0.27**	**0.000**
Timeline cyclical	0.91	0.82-1.00	-0.09	0.06
Emotional representations	**0.90**	**0.83-0.96**	**-0.10**	**0.005**

The internal validity of the models was good except for self-efficacy. In the omnibus test the coefficients for all three models were significant (P= <0.0001). The levels of significance in the Hosmer-Lemeshov (Goodness of fit) test of the models was 0.97 for well-being, 0.42 for HRQoL and 0.05 for self-efficacy - the latter being equal to the predefined P-value of 0.05 and therefore indicating a less reliable model.

Predictors of GFD adherence

Table 11 shows the results of a binary logistic regression analysis to predict high adherence to a gluten-free diet (GFD). Sex was not entered as there was no evidence of a univariate effect (χ^2=1.162, P=0.38 Fisher's Exact). The most predictive independent variables were older age, strong beliefs in the serious consequences and weak beliefs in the cyclical nature of CD (or conversely beliefs in the chronicity of the condition). This means that the older participants were and the stronger their beliefs in the seriousness of CD the more likely they were to stick to a GFD. Furthermore, the weaker participants' beliefs in the cyclical nature of CD the more likely they were to adhere to a GFD. The correct classification rate for the model was 86%. None of the outcome measures, self-efficacy, general well-being and HRQoL were strong predictors of high adherence.

The internal validity of this model was good. In the omnibus test the coefficients were significant (P= <0.0001) and the significance level in the Hosmer-Lemeshov (Goodness of fit) test was 0.86, above the predefined P-value of 0.05, thus confirming goodness of fit.

TABLE 11 Logistic regression analysis of factors predicting high adherence to a GFD

Independent Variables	Odds Ratio	95% CI	B	P-value
Age	**1.04**	**1.01-1.07**	**0.04**	**0.002**
PMCSMS	1.07	0.97-1.17	0.07	0.14
GBWI	0.97	0.93-1.02	-0.03	0.17
CDQ	0.99	0.96-1.02	-0.01	0.54
Identity	0.96	0.81-1.13	-0.03	0.65
Consequences	**1.15**	**1.03-1.28**	**0.14**	**0.009**
Personal control	1.08	0.96-1.20	0.07	0.16
Illness coherence	1.01	0.90-1.13	0.01	0.83
Timeline cyclical	**0.87**	**0.77-0.98**	**-0.14**	**0.02**
Emotional representations	0.94	0.85-1.05	-0.05	0.31

DISCUSSION

This study investigated the psychosocial impact of CD on a UK adult population in terms of health-related quality of life and psychological well-being. It is also the first to investigate the illness perceptions of individuals with CD. Previous European studies have indicated that depression and lower quality of life affect individuals with CD, and anxiety and depression are major causes of lower levels of adherence to treatment recommendations (Addolorato et al, 1996; Ciacci et al, 1998). There is also a suggestion from some studies that women with CD experience poorer quality of life than their male counterparts (Hallert et al, 2003).

HRQoL, Psychological Well-being and Self-Efficacy

The results for HRQoL are in line with previous research which indicates that lower quality of life affects individuals with CD. The mean and total distribution of scores for the CDQ were comparable with those found in the German Coeliac Society population by the authors of the instrument (Häuser et al, 2007). In this German study to validate the CDQ the mean score for participants (n=516) who belonged to the German Coeliac Society was 151.1 (s.d. 25.2). Reduced HRQoL was defined by scores ≤ 10% percentile of the total CDQ score which was 11% of the sample. There were significant differences between men and women for all sub-scales, reflecting better health related quality of life for men. In the current UK population, the univariate analyses showed that men had higher scores than women on the total scale and two subscales, Emotion and Social. This indicates that women were more emotionally affected by their CD than men and found the condition more socially restrictive. However, it could not be demonstrated by multivariate analysis that there was an association between male gender and increased HRQoL.

For psychological well-being the results were similar in that GWBI scores indicated a reduced overall level of psychological well-being. The mean GWBI of this CD population was slightly lower compared to individuals with long-term health problems drawn from a UK primary care population and considerably lower when compared to a healthy sub-set drawn from the same sample. In this UK sample drawn from a general practice population, the distribution of GWBI scores ranged from 29 to 109 with a mean of 82.2 (s.d. 14.6). Forty five percent of patients had a limiting long-term illness, health problem or handicap (Hopton, Hunt, Shiels & Smith, 1995). In a healthy sub-group of this sample i.e. those with no long-standing illness and absence of anxiety and depression, GWBI scores ranged from 54 to 109 with a mean of 94.0 (s.d. 10.9). In the CD sample, there were also slight differences between the mean scores of men and women on this measure for total score,

and the subscales of anxiety, depressed mood and self-control, with men having a better outcome. Once again however, gender difference was not demonstrated in the multivariate analysis.

The mean PMCSMS scores showed a relatively high level of perceived self-efficacy in this CD population meaning that individuals generally felt confident with managing their condition. Those in the lower adherence group had significantly reduced self-efficacy compared with those in the high adherence group. This is in line with early diabetes research that found patients adhering to dietary advice were more likely to report feeling competent to self-manage their diabetes (Talbot et al, 1997). However, the PMCSMS was not found to predict adherence in the multivariate analysis. Furthermore, no significant difference was found between the scores of men and women unlike the findings in a recent study of people with Diabetes where men scored higher than women (Wallston et al, 2007). In this study 398 participants with diabetes completed the PMCSM adapted for Diabetes namely, the Perceived Diabetes Self-Management Scale (PDSMS). It was found that males scored significantly higher (mean 29.7) than women (mean 28.3). These self-efficacy scores for men and women with Diabetes are slightly lower compared to those of the CD participants, indicating reduced perceived self-efficacy in the former group (Wallston et al, 2007).

Illness Perceptions

In general the participants reported a coherent understanding of their condition with strong perceptions of their personal ability to control it and strong beliefs about the serious consequences of CD. Few differences in illness perceptions were identified between men and women. The finding that women were more likely to respond more emotionally to their CD than men may be a reflection of different ways of coping between men and

women (Hallert et al, 2002). Women also believed more strongly than men that psychological causes such as stress had some bearing on the development of their condition. Again, this is perhaps a reflection of differences in western society at large in which women tend to report more psychological symptoms than men (Wittchen, 2002).

There were associations between weak identity perceptions and increased HRQoL and enhanced general well-being. Weak beliefs in the serious consequences of CD increased the likelihood of increased self-efficacy and HRQoL. Strong perceived personal control increased the probability of a higher self-efficacy score. Strong perceived illness coherence tended to increase the likelihood of better self-efficacy and HRQoL. A reduced emotional response to CD and weak beliefs that the condition was cyclical in nature increased the probability of better self-efficacy, good HRQoL and enhanced psychological well-being. At present there exist no similar studies investigating the illness perceptions of individuals with CD so that comparisons cannot be made. However, in a study focusing on illness representations and outcomes in irritable bowel syndrome (Rutter & Rutter, 2002) the authors found similarly that the reporting of serious perceived consequences was associated with reduced quality of life and poorer scores for anxiety and depression. The high correlation between CDQ and GWBI scores suggested a close relationship between psychological well-being and HRQoL.

Predictors of Psychological Well-being, HRQoL, Self-efficacy and Adherence

In the binary logistic analyses, the most consistent predictors of all three outcome measures were consequences and emotional responses. Weaker beliefs in the serious consequences of CD and reduced emotional responses were more likely to be associated with better scores. The levels of reliability for each model were satisfactory with the psychological well-being model showing particularly high reliability. Older age and

weaker beliefs in the cyclical nature of CD were more likely to predict enhanced psychological well-being. However, men were significantly older than women in this sample meaning that the finding for older age could be caused by older men who had higher total scores on the GWBI. This needs further investigation in subsequent studies. A weaker CD identity was more likely to be associated with an improved HRQoL. Greater perceived illness coherence also increased the likelihood of a better HRQoL and higher self-efficacy. Stronger beliefs in personal control were associated with increased self-efficacy.

The adherence rate was high with 87% of participants reporting that they stuck to a GFD all of the time, the remaining 13% reported that they adhered most or some of the time. The small numbers of low compliers make the analysis of adherence less robust. The most significant likely predictors of higher adherence were: older age, stronger beliefs in the serious consequences of CD and weaker beliefs in the cyclical nature of the condition. Eighty eight percent of the sample had been over the age of 20 when diagnosed and it can be speculated from the data that the majority of these participants had been adhering to a GFD for some time. However, it was not proven that adherence was related to time since diagnosis. Nevertheless, the study is in line with Italian research (Fera et al, 2003) that showed that adherence to a GFD was related to length of diagnosis with individuals diagnosed after the age of 20 years having a better adherence rate than those diagnosed earlier. This is an area for further investigation. Although strong beliefs in serious consequences was a likely predictor of adherence, results reported in the paragraph above indicated that weaker beliefs in serious consequences increased the likelihood of enhanced psychological well-being, HRQoL and Self-efficacy. However, there was no evidence in this study to suggest that those in the high adherence group had poorer scores than the low

adherence group on any of these outcome measures. This interaction effect needs further investigation using multivariate techniques.

Limitations of the Study

Some limitations of the study should be born in mind. For example, the participants were recruited from adult members of Coeliac UK leading to a possible selection bias. In a German study of patients with inflammatory bowel disease, membership of a self-help organization was predictive of reduced life satisfaction (Janke, Klump, Gregor & Häuser, 2005). However, there are no comparative data available between individuals with CD with and without membership of Coeliac UK. It is possible that there is a further response bias of individuals with reduced psychological well-being and HRQoL returning the questionnaires. Therefore, it seems unlikely that the findings of the study are representative of the UK general population of people with coeliac disease. Conversely this group of individuals is the only available large UK sample studied to date. The sample was predominately white Caucasian, of high educational level and biased towards women (although CD tends to affect more women than men). Individuals with a higher level of education are more likely to hold beliefs that are compatible with scientific and medical approaches (Bowling, 1989). Therefore, the results of this study may not generalize to people with CD of lower educational level or ethnic groups whose beliefs about CD and illness perceptions may differ.

The cross-sectional nature of this study should also be considered, since this means that the results show only associations between variables and prohibit conclusions being drawn about causality. The inclusion of a control group or healthy non-CD group would have facilitated the interpretation of scores for HRQoL and psychological well-being by providing normative data. It should also be mentioned that the data are self-reported which

may bias the answers to sensitive questions such as dietary compliance. However, anonymity of the data was maintained to help minimize this potential bias.

CONCLUSIONS

Amongst adult members of Coeliac UK there was evidence of reduced HRQoL and decreased psychological well-being. The gender differences in quality of life found in previous research were not repeated in the multivariate analyses used in this study. More research is needed in the UK Coeliac Disease population using robust methodologies such as case control or longitudinal studies to investigate this potential difference further.

Further investigation is also required into possible differences in quality of life and well-being between those who adhere to a GFD and those who do not. Self-efficacy and illness perceptions appeared to be influential factors in this study and could play a role in informing psycho-education for individuals who might benefit from therapeutic intervention to improve GFD adherence and enhance psychological well-being. More information is needed on the link between self-efficacy, illness perceptions and adherence to a GFD. Further knowledge of these factors is important for informing therapeutic interventions, to help in the clinical management of Coeliac Disease.

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Living with Coeliac Disease: A Research Study Conducted in Partial Fulfilment of the Requirements for the Doctorate in Clinical Psychology at the University of Birmingham

Executive Summary

Coeliac Disease (CD) is an autoimmune disease in which the enzyme gluten causes damage and chronic inflammation to the small intestine. Untreated the condition may predispose an individual to serious diseases such as cancer, type I diabetes, osteoporosis, gynaecological problems in women, central nervous system disorders and other autoimmune diseases. Across Europe it is thought that the condition may affect between 1 in 200 to 1 in 500 people (Rewers, 2005; Catassi, Ratsch, Fabiani, 1994). CD is cannot be cured but symptoms are managed by a gluten-free diet (GFD) for life.

Most research looking at CD has been focused on the biological basis of the disease rather than the impact of the condition from the individual's own point of view. The few existing studies conducted mostly in Europe and Canada suggest that the chronic nature of the condition, the limitations imposed by the need to follow a permanent restrictive diet and the risk of other associated diseases can have a negative impact on health-related quality of life (HRQoL) and psychological well-being (Aldorrato, Leggio, D'Angelo et al, 2008). There is also a suggestion from the existing literature that women with CD experience poorer quality of life than men (Hallert, Sandlund & Broqvist 2003). However, knowledge in this area remains sparse to-date particularly in the UK.

The aim of this research was two-fold: first to systematically review the literature published within the last decade to investigate the impact of living with CD on

psychological well-being and Health Related Quality of Life (HRQoL) and secondly to conduct a questionnaire survey on the Illness Perceptions and Effects of Coeliac Disease on Psychological Well-being and HRQoL in a UK Population. The perceptions that a person has about their illness or condition (illness perceptions) give personal meaning to symptoms and it is believed they act as a framework for guiding coping strategies. The concept of HRQoL refers to an individual's perceived state of health including social, emotional and physical well-being or functioning. Psychological well-being is concerned with the emotional well-being in individuals whose illnesses or conditions are not so much physically incapacitating as psychologically distressing. The term relates to internal emotional states that reflect subjective well-being or distress.

Literature Review

Twenty-one relevant studies were identified. Eight of these originated from Italy, three from Sweden and three from Finland, with the remaining seven coming from Germany, the UK, Canada, the USA, Spain, Finland and Argentina. In general, the results of these studies suggest that in addition to a reduced Health Related Quality of Life, psychological distress is commonly found in patients with CD, particularly depression. The studies showed that anxiety is also commonly experienced but this tends to decrease when individuals start a regular gluten-free diet. However, it was found that depression may persist even in people whose diets have been gluten-free for many years.

Questionnaire Survey

The postal survey investigated self-reported levels of Health Related Quality of Life (HRQoL) and psychological (or emotional) well-being in adults belonging to Coeliac UK, the main charity supporting people with CD in the UK. It also explored their beliefs and

perceptions concerning CD (illness perceptions) and how well they thought they coped with the condition (self-efficacy).

The survey was ethically approved by the University of Birmingham, School of Psychology, Research Ethics Committee and advertised in the quarterly Coeliac UK Magazine, 'Crossed Grain'. Questionnaires were received back from 288 adult members out of a total of 433 who asked to be sent a survey pack.

The results of the survey in this UK population found that Health Related Quality of Life (HRQoL) and psychological well-being were lower than they should be with levels being comparable to those found in similar European studies of CD. Unlike these previous studies, in the UK sample there was no evidence to suggest that women have poorer levels of HRQoL than men.

It was also found that those who were not worried by the possible serious consequences of CD and who did not react emotionally to their condition tended to have a higher level of HRQoL, a healthier level of psychological well-being and increased self-efficacy (felt they managed their CD well). Those who strongly believed that they had control of their condition and felt they understood their CD well also tended to have higher levels of self-efficacy.

Those participants most likely to stick to a gluten-free diet tended to be older and had stronger beliefs in the serious consequences of CD. The results suggest that knowledge of illness perceptions could play an important role in developing psycho-educational programmes aimed at helping people stay on gluten-free diets and enhancing psychological well-being and HRQoL.

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Appendices

Appendix 1

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Appendix 2

Glossary and reference list of outcome measures

BDI	Beck Depression Inventory: Beck A.T., Ward C., Mendelson M. (1961). Beck Depression Inventory (BDI). Archives of General Psychiatry 4: 561-571.
BI	Burden of Illness: Hallert C, Grännö C, Hultén S, Midhagen G, Ström M, Svensson H, Valdimarsson T. (2002). Living with coeliac disease: controlled study of the burden of illness. Scandinavian Journal of Gastroenterology, 37: 39-42.
CCEI	Crown-Crisp Experiential Index: Crown, S. & Crisp, A.H. (1966). A short clinical diagnostic self-rating scale for psychoneurotic patients: The Middlesex Hospital Questionnaire. British Journal of Psychiatry, 112: 917-23.
CFS	Chronic Fatigue Scale: Wessely, S. & Powell (1989). Fatigue syndromes; a comparison of chronic 'postviral' fatigue with neuromuscular and affective disorders. Journal of Neurology and Neurosurgical Psychiatry, 52: 940-48.
EPQ	Eysenck Personality Questionnaire: Hans Jürgen Eysenck, H.J. & Eysenck, S.B.G. (1975). Manual of the Eysenck Personality Questionnaire. London: Hodder and Stoughton.
EQ	EuroQol-5D: Brooks R, Rabin R, de Charro F, (eds): The Measurement and Valuation of Health Status Using EQ-5D: A European Perspective: Evidence from the EuroQol BIO MED Research Programme. Rotterdam: Kluwer Academic Publishers; 2003.
FSS	Fatigue Severity Scale: Krupp, L.B., La Rocca, N.G., Muir-Nash, J., Steinberg, A.D. (1989). The fatigue severity scale: application to patients with multiple sclerosis and systemic lupus erythematous. Archives of Neurology, 46: 1121-23.
GIQLI	Gastrointestinal Quality of Life Index: Eypasch, E., Williams, J.I., Wood-Dauphinee, S., Ure, B.M., Schmulling, C., Neugebauer, E., Troidl, H. (1995) Gastrointestinal Quality of Life Index: Development, validation and application of a new instrument. British Journal of Surgery, 82: 216-222.
GSRS	Gastrointestinal Symptoms Rating Scale: Revicki, D.A., Wood, M., Wiklund, I., Crawley, J. (1998). Reliability and validity of the Gastrointestinal Symptom Rating Scale in patients with gastroesophageal reflux disease. Quality of Life Research, 7: 75-83.
HADS	Hospital Anxiety and Depression Scale: Zigmond, A.S. & Snaith R.P. (1983). The hospital anxiety and depression scale. Acta Psychiatrica Scandinavica, 67: 361-370.

IBQ	Illness Behaviour Questionnaire: Pilowsky, I., & Spence, N. (1994). Manual for the Illness Behaviour Questionnaire (3rd ed.). Department of Psychiatry, University of Adelaide.
LSAS	Liebowitz Social Anxiety Scale: Liebowitz, M.R. (1987). Social phobia. Modern Problems of Pharmacopsychiatry, 22: 141-73.
M-SDS	Modified Zung Self-Rating Depression Scale: Zung W. (1973) From art to science. Archives of General Psychiatry, 29: 328-37.
PGWBI	Psychological General Wellbeing Index: Dupuy, H. (1984). The Psychological General Well-Being Index. In Wenger N., Mattson M., Furberg C. & Elinson J. (Eds). Assessment of Quality of Life in Clinical trials of Cardiovascular Therapies. pp 170-183. New York, Le Jacq.
PQ	Pychophysiological Questionnaire: Sanavio, E., Bertolotti, G., Michielin, P., Vidotto, G., Zotti, A.M. (1997). Cognitive Behavioural Assessment 2.0: Primary Scale [Italian]. Firenze, Organizzazioni Speciali.
SAIC	Self-Administered Inventory for Coeliacs: Ciacci C., Iavarone A., Siniscalchi M., Romano R., De Rosa A. (2002). Psychological dimensions of celiac disease: toward an integrated approach. Digestive Diseases & Sciences, 47: 2082-7.
SF-36	Short Form-36 Health Survey: McHorney, C.A., Ware, J.E.J., Lu J.F., Sherbourne, C.D. (1994). The MOS 36-item Short-Form Health Survey (SF-36): III. Tests of data quality, scaling assumptions, and reliability across diverse patient groups. Medical Care, 32: 40-66.
SF-12	Short Form-12 Health Survey: Riddle, D.L., Lee, M.S. & Stratford, P.W. (2001). Use of SF-36 and SF-12 health status measures a quantitative comparison for groups versus individual patients. Medical Care, 30: 867-878.
STAI	State-Trait Anxiety Inventory: Spielberg, C.D., Gorsuch, R.L., Lushene, R.E.(1983). Manual for the State and Trait Anxiety Inventory. Paolo Alto, CA, USA: Consulting Psychologist Press.
TAS-20	Toronto Alexithymia Scale: Bagby, R.M., Parker, J.D.A., Taylor, G.J. (1994). The 20-item Toronto Alexithymia Scale. I. Item selection and cross-validation of the factor structure. Journal of Psychosomatic Research, 38: 23-32.

Appendix 3

Social Science & Medicine

Guide for Authors

(Removed for copyright reasons)

Appendix 4
Letter of Ethical Approval
(Removed for reasons of Confidentiality)

Appendix 5
Response to Ethics Committee
(Removed for reasons of Confidentiality)

Appendix 4
Questionnaire Pack

Participant Information Sheet

Study of Living with Coeliac Disease
University of Birmingham

Thank you for your interest in this study. My name is Sarah Ford and I am a Clinical Psychologist in Training at the University of Birmingham, UK. As part of my doctoral degree I am conducting a study exploring the experiences of individuals who have Coeliac Disease (CD). I would like to invite you to take part in this questionnaire survey. Please read the information below before deciding whether or not you would like to take part in this survey.

.

What is the purpose of the study? Research from other countries suggests that a diagnosis of CD and staying on a gluten-free-diet leads to enormous changes in the lives of sufferers and the dietary restrictions can be hard to accept. It has been found that some people with CD can feel restricted, isolated and at times anxious about what they eat and this reduces their quality of life. So far, no studies have been conducted in the UK to explore the effects on adults of living with CD. Understanding the emotional and social effects of having CD may help in the development of effective psychological treatments for those who struggle to cope with their disease.

Who is taking part? All those aged 18 and above who belong to the Coeliac Society are being invited to participate. This is because to be a member you need to have been medically diagnosed with CD.

Do I have to take part? You are under no obligation to participate and you have the right to withdraw at any time. If after completing the survey you decide that you would like to withdraw please contact me by email: stf615@bham.ac.uk or telephone: 0121 414 7576 and I will destroy your data.

What will happen to me if I take part? You will be asked to fill in 5 different questionnaires. These may take between 20 – 40 minutes to complete.

What do I have to do? You will be asked to complete 5 questionnaires and return them in the envelope provided. These questionnaires are confidential and anonymous.

What are the possible disadvantages and risks of taking part? The survey is completely voluntary and there are no physical risks as I am simply gathering information. Some questions may be a little personal or embarrassing, but you do not have to answer any questions you do not want to.

What are the possible benefits of taking part? Participating in this survey will not benefit you personally. However, the findings may help our understanding of the effects of CD and help other sufferers in the future.

What happens when the research study stops? When you send back your questionnaires they will be entered into a database accessible only to the researchers conducting this study. It is hoped that this study will be published in an academic journal; details will also appear in the Coeliac Society Newsletter 'Crossed Grain'.

- **What if I find some of the questions distressing?** If after completing this survey you feel in need of some additional support, please speak to someone at one of the organisations below:

  - ❑ Your GP/practice nurse
  - ❑ Coeliac Society helpline: 0870 444 8804
  - ❑ NHS Direct Tel: 0845 46 47

Will my taking part in the study be kept confidential? All of the information that you send will be confidential and anonymous. When written up all participants will be considered as a group so there will be no way of knowing who participated in the study.

Contact Details: Further information can be obtained from Sarah Ford, Trainee Clinical Psychologist (email: XXXXXXXX) who is carrying out this research under the supervision of Dr Ruth Howard and Dr Jan Oyebode. They can both be contacted at the School of Psychology, University of Birmingham, Edgbaston, Birmingham B15 2TT Tel: XXXXXXX

What happens if I decide to take part? Please read and sign the attached consent form before completing the five questionnaires.

CONSENT FORM

Title of Project: Living with Coeliac Disease Study

Name of Researcher: Sarah Ford, Trainee Clinical Psychologist

I confirm that I have read and understand the information sheet for the above study. I have had the opportunity to consider the information, and receive more information (if needed) via the contact details given.

I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason.

I agree to take part in the above study.

Name _____

Signature_____ Date _____

Thank you for agreeing to take part in this survey

Now please complete the five questionnaires beginning with form 1

Form 1: Living with Coeliac Disease Study

We are interested in how well you cope with your Coeliac Disease (CD). Please indicate how much you agree or disagree with the following statements by circling one answer per statement.

It is difficult for me to find effective solutions for problems that occur with managing my CD.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

I find efforts to change things I don't like about my Coeliac Disease are ineffective.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

I handle myself well with respect to my Coeliac Disease.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

I am able to manage things related to my Coeliac Disease as well as most other people.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

I succeed in the projects I undertake to manage my Coeliac Disease.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

Typically, my plans for managing my Coeliac Disease don't work out well.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

No matter how hard I try, managing my Coeliac Disease doesn't turn out the way I would like.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

I'm generally able to accomplish my goals with respect to managing my Coeliac Disease.

Strongly Disagree	Disagree	Neither agree nor disagree	Agree	Strongly Agree

Form 2: Living with Coeliac Disease Study - Illness Perceptions Questionnaire

YOUR VIEWS ABOUT YOUR COELIAC DISEASE (CD)

Listed below are a number of symptoms that you may or may not have experienced since being diagnosed with CD. Please indicate by circling *Yes* or *No*, whether you have experienced any of these symptoms and whether you believe that these symptoms are related to your CD.

	I have experienced this symptom *since my CD*		This symptom is *related to my CD*	
Pain	**Yes**	**No**	**Yes**	**No**
Sore Throat	**Yes**	**No**	**Yes**	**No**
Nausea	**Yes**	**No**	**Yes**	**No**
Breathlessness	**Yes**	**No**	**Yes**	**No**
Weight Loss	**Yes**	**No**	**Yes**	**No**
Fatigue	**Yes**	**No**	**Yes**	**No**
Stiff Joints	**Yes**	**No**	**Yes**	**No**
Sore Eyes	**Yes**	**No**	**Yes**	**No**
Wheeziness	**Yes**	**No**	**Yes**	**No**
Headaches	**Yes**	**No**	**Yes**	**No**
Upset Stomach	**Yes**	**No**	**Yes**	**No**
Sleep Difficulties	**Yes**	**No**	**Yes**	**No**
Dizziness	**Yes**	**No**	**Yes**	**No**
Loss of Strength	**Yes**	**No**	**Yes**	**No**

We are interested in your own personal views of how you now see your Coeliac Disease (CD). Please indicate how much you agree or disagree with the following statements about your CD by ticking the appropriate box.

VIEWS ABOUT YOUR CD	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
My CD will last a short time.					
My CD is likely to be permanent rather than temporary.					
My CD will last for a long time.					
My CD will pass quickly.					
I expect to have this CD for the rest of my life.					
My CD is a serious condition.					
My CD has major consequences on my life.					
My CD does not have much effect on my life.					
My CD strongly affects the way others see me.					

FORM 2 Continued/…

VIEWS ABOUT YOUR CD	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
My CD has serious financial consequences.					
My CD causes difficulties for those who are close to me.					
There is a lot which I can do to control my symptoms.					
What I do can determine whether my CD gets better or worse.					
The course of my CD depends on me.					
Nothing I do will affect my CD.					
I have the power to influence my CD.					
My actions will have no affect on the outcome of my CD.					
My CD will improve in time.					
There is very little that can be done to improve my CD.					
My diet will be effective in curing my CD.					
The negative effects of my CD can be prevented (avoided) by my diet.					
My diet can control my CD.					
There is nothing that can help my condition.					
The symptoms of my condition are puzzling to me.					
My CD is a mystery to me.					
I don't understand my CD.					
My CD doesn't make any sense to me.					
I have a clear picture or understanding of my condition.					
The symptoms of my CD change a great deal from day to day.					
My symptoms come and go in cycles.					
My CD is very unpredictable.					
I go through cycles in which my CD gets better and worse.					

VIEWS ABOUT YOUR CD	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY DISAGREE
I get depressed when I think about my CD.					
When I think about my CD I get upset.					
My CD makes me feel angry.					
My CD does not worry me.					
Having this CD makes me feel anxious.					
My CD makes me feel afraid.					

CAUSES OF MY COELIAC DISEASE

We are interested in what you consider may have been the cause of your CD. As people are very different, there is no correct answer for this question. We are most interested in your own views about the factors that caused your CD rather than what others including doctors or family may have suggested to you. Below is a list of possible causes for your CD. Please indicate how much you agree or disagree that they were causes for you by ticking the appropriate box.

POSSIBLE CAUSES	STRONGLY DISAGREE	DISAGREE	NEITHER AGREE NOR DISAGREE	AGREE	STRONGLY AGREE
Stress or worry.					
Hereditary - it runs in my family.					
A Germ or virus.					
Diet or eating habits.					
Chance or bad luck.					
Poor medical care in my past.					
Pollution in the environment.					
My own behaviour.					
My mental attitude e.g. thinking about life negatively.					
Family problems or worries.					
Overwork.					
My emotional state e.g. feeling down, lonely, anxious, empty.					
Ageing.					

POSSIBLE CAUSES	**STRONGLY DISAGREE**	**DISAGREE**	**NEITHER AGREE NOR DISAGREE**	**AGREE**	**STRONGLY AGREE**
Alcohol.					
Smoking.					
Accident or injury.					
My personality.					
Altered immunity.					

Below, please list in rank-order the three most important factors that you now believe caused YOUR CD. You may use any of the items from the box above, or you may have additional ideas of your own.

The most important causes for me:

1. _____

2. _____

3. _____

Thank you
Please continue by completing form 3
on the back of this page →

Form 3: The General Well-Being Index (British version)

Please tick the column which best applies to you (one answer per row)

How have you been feeling in general during the past month?	In very good spirits	In good spirits mostly	I've been up & down a lot	In low spirits mostly	In very low spirits
During the past month have you been bothered by any Illness, pains or fears about your health?	All the time	A lot of the time	Some of the time	A little bit	Not at all
Did you feel depressed during the past month?	Yes, very much so	Yes, quite a bit	Sometimes enough to bother me	A little depressed now and then	No, not at all
During the past month have you felt in firm control of your actions, thoughts or feelings?	Yes, definitely	Yes, mostly	Not too well	No, hardly at all	Not at all
Have you been bothered by your nerves during the past month?	Very much so	Quite a bit	Sometimes	A little	Not at all
During the past month how much energy or vitality did you have?	Lots of energy	Fairly energetic most of the time	Energy varied quite a bit	Low in energy mostly	No energy at all
Have you felt disheartened and sad over the past month?	All of the time	Most of the time	From time to time	Very occasionally	Not at all
During the past month how tense have you been?	Extremely tense all of the time	Very tense most of the time	A little tense sometimes	Rarely tense	Not tense at all

How happy or satisfied have you been with your personal life during the past month?	Very satisfied	Fairly satisfied	Satisfied on the whole	Rather dissatisfied	Very dissatisfied
Over the past month did you feel well enough to do the things you like to do or had to?	Yes, definitely	Yes, for the most part	About half the time	No, not often	No, not at all
Have you felt so sad, disheartened or had so many problems that you wondered if anything was worthwhile over the past month?	All the time	Most of the time	From time to time	Very Occasionally	Not at all
During the past month have you been waking up feeling fresh and rested?	Every day	Most days	Less than half the time	Not often	Not at all
Have you had any worries or fears about your health during the past month?	Yes, all the time	Most of the time	From time to time	Not a lot	Not at all
During the past month have you wondered if you were losing control over your actions, thoughts, feelings or memory?	All the time	Most of the time	From time to time	No, hardly at all	Not at all
Has your daily life been filled with things that interest you during the past month?	All of the time	Most of the time	Some of the time	A little	Not at all
During the past month how active and vigorous have you felt?	Very active every day	Mostly active	Fairly active	Seldom active	Not at all active
Have you been anxious, worried, or upset over the past month?	Very much so	Quite a lot	Sometimes enough to bother me	A little bit	Not at all

During the past month have you felt emotionally stable and sure of yourself?	All of the time	Most of the time	Some of the time	Now and then	Not at all
How relaxed have you felt over the past month?	Very relaxed all the time	Mostly relaxed	Relaxed about half the time	Rarely felt relaxed	Not at all relaxed
During the past month how cheerful have you been?	Not cheerful at all	A little cheerful now and then	Cheerful about half the time	Mostly quite cheerful	Very cheerful all the time
Have you felt tired, worn out or exhausted during the past month?	All of the time	Most of the time	Some of the time	Now and then	Not at all
Over the past month have you been under any stress or pressure?	Yes, almost more than I could bear	Yes, more than usual	About the same as usual	Yes, a little	No, not at all

Thank you
Please continue by completing form 4

FORM 4

The Coeliac Disease Questionnaire CDQ – Health related quality of life index

This questionnaire has been developed to find out how you have been feeling during the last two weeks. You will be asked about symptoms related to your coeliac disease, your general well-being and your mood. The questionnaire contains 28 questions. Each question offers seven possible answers ranked (1) to (7). Please read each question carefully and circle the answer that best describes how you felt during the past two weeks.

How many times during the past two weeks was your life affected by a sudden urge to visit a bathroom for a bowel movement?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How often during the last two weeks did you feel physically exhausted or fatigued?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How often during the last two weeks have you felt frustrated, impatient or restless?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you refuse or avoid an invitation for dinner with friends or relatives due to your coeliac disease?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How often during the last two weeks have your bowel movements been loose?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How much intellectual energy did you have during the last two weeks?

No energy At all	Very little energy	Little energy	Some energy	A moderate amount of energy	Lots of energy	I was full of energy

How many times during the last two weeks were you concerned that your children could inherit or may have inherited your coeliac disease?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks have you been troubled by cramps in your abdomen?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

Did you encounter any difficulties with recreational activities or sports due to your coeliac disease during the last two weeks? Please tick one answer below.

- ❑ Extreme difficulties, no activities possible
- ❑ Very considerable difficulties
- ❑ Considerable difficulties
- ❑ Some difficulties
- ❑ Minor difficulties
- ❑ Hardly any difficulties
- ❑ No difficulties, coeliac disease did not affect my recreational activities or sports

How often during the last two weeks did you feel depressed or discouraged?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you suffer from bloating or flatulence?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

People with coeliac disease often have worries and fears related to their disease. How many times during the last two weeks did you worry about or were afraid of getting cancer as a result of your CD?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks were you affected by a feeling of incomplete bowel evacuation?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How often during the last two weeks have you felt relaxed and free of tension?

None of the time	Hardly any of the time	A little of the time	Some of the time	A good bit of the time	Most of the time	All of the time

How many times during the last two weeks did you feel isolated from or excluded by others due to your coeliac disease?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How much of the time during the last two weeks have you felt tearful or upset?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you suffer from repeated belching?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

To what extent did your CD restrict your sexual activity during the last two weeks?

- ❑ No sex due to coeliac disease
- ❑ Considerable restraint due to coeliac disease
- ❑ Moderate restraint due to coeliac disease
- ❑ Some restraint due to coeliac disease
- ❑ Little restraint due to coeliac disease
- ❑ Almost no restraint due to coeliac disease
- ❑ No restraint due to coeliac disease

How many times during the last two weeks did you suffer from nausea or retching?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you feel that important people such as members of your family or friends showed a lack of understanding for your CD?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How satisfied, happy or pleased have you been with your personal life you during the last two weeks?

- ❑ Very unsatisfied, mostly unhappy
- ❑ Generally unsatisfied, unhappy
- ❑ Somewhat unsatisfied, unhappy
- ❑ Generally satisfied, pleased
- ❑ Most of the time satisfied, happy
- ❑ Most of the time very satisfied, happy
- ❑ Very satisfied, could not be happier or more pleased

How many times during the last two weeks did you feel that colleagues or superiors showed a lack of understanding for your coeliac disease?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you feel limited in your professional training or career by your coeliac disease?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you feel burdened by the expenses and time required obtaining gluten-free food?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you feel burdened by problems with meeting the costs of gluten-free food or other coeliac therapies?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you experience lack of expertise regarding CD from your doctors?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you worry that your CD was diagnosed too late?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

How many times during the last two weeks did you suffer from fear of medical examinations in relation to your coeliac disease, e.g. blood test or endoscopy?

All of the Time	Most of the time	A good bit of the time	Some of the time	A little of the time	Hardly any of the time	None of the time

Thank you
Please continue by completing form 5

Form 5: Living with Coeliac Disease Study – Information About You

Please state your date of birth: Day……. Month…….. Year ……..

Are you: **Female** ☐ **Male** ☐ (please tick)

Marital Status (please tick as applicable)

- ❑ Single (never married)
- ❑ Married
- ❑ Co-habiting
- ❑ Separated
- ❑ Divorced
- ❑ Widowed

Ethnicity:

- ❑ White British
- ❑ Asian
- ❑ Black
- ❑ Chinese
- ❑ Mixed – White & Asian
- ❑ Mixed – White & Black
- ❑ Other mixed background
- ❑ Any other ethnic background

Age at diagnosis of Coeliac Disease

- ❑ Childhood 5 – 10 years old
- ❑ Adolescence 11 – 20 years old
- ❑ Adulthood 21 – 30 years old
- ❑ Adulthood 31 – 40 years old
- ❑ Adulthood 41 – 50 years
- ❑ Adulthood 50 +

Education Level:

- ❑ School education, no qualifications
- ❑ School education with qualifications
- ❑ University qualifications
- ❑ Vocational training/qualifications

Highest Occupation:

- ❑ Professional occupation
- ❑ Managerial or technical
- ❑ Non-manual skilled
- ❑ Manual skilled
- ❑ Partly skilled
- ❑ Unskilled occupation
- ❑ Home-maker

How many years since your CD diagnosis?

- ❑ Less than a year
- ❑ 1 year - 5 years
- ❑ 6 years – 10 years
- ❑ 11 years – 20 years
- ❑ 21 years – 30 years
- ❑ 31 years – 40 years
- ❑ More than 40 years

In general, how strictly do you maintain a gluten free diet?	**How long have you been a member of Coeliac UK?**
❑ All of the time	❑ Less than a year
❑ Most of the time	❑ 1 year - 5 years
❑ Some of the time	❑ 6 years – 10 years
❑ Now and then	❑ 11 years – 20 years
❑ Not at all	❑ More than 20 years

Thank you for taking the time to complete this survey. Your participation is much appreciated.

Please post all the forms (apart from the information sheet) back to us using the envelope provided. Please check that you have filled in <u>both sides</u> of each form and remember to include the consent form, as we cannot process your forms without this.

Appendix 7

One-Sample Kolmogorov-Smirnov Test

		PMCSMS Total Score	IPQ-R Identity	IPQ-R Timeline	IPQ-R Consequences
N		280	278	278	278
Normal Parameters(a,b)	Mean	31.76	3.37	28.19	21.04
	Std. Deviation	5.948	2.940	2.400	4.927
Most Extreme Diffs	Absolute	.105	.148	.225	.113
	Positive	.083	.148	.225	.057
	Negative	-.105	-.126	-.200	-.113
Kolmogorov-Smirnov Z		1.763	2.496	3.758	1.883
Asymp. Sig. (2-tailed)		.004	.000	.000	.002

a Test distribution is Normal.
b Calculated from data.

One-Sample Kolmogorov-Smirnov Test

		IPQ-R Personal Control	IPQ-R Treatment Control	IPQ-R Illness Coherence	IPQ-R Timeline Cyclical
N		278	278	278	278
Normal Parameters(a,b)	Mean	26.03	19.74	21.31	9.12
	Std. Deviation	4.060	3.327	4.128	4.358
Most Extreme Diffs	Absolute	.164	.113	.196	.170
	Positive	.164	.072	.185	.170
	Negative	-.152	-.113	-.196	-.120
Kolmogorov-Smirnov Z		2.738	1.889	3.272	2.834
Asymp. Sig. (2-tailed)		.000	.002	.000	.000

a Test distribution is Normal.
b Calculated from data.

One-Sample Kolmogorov-Smirnov Test

		IPQ-R Emotional Responses	IPQ-R Psychological Factors	IPQ-R Risk Factors	IPQ-R Immunity
N		278	278	278	278
Normal Parameters(a,b)	Mean	15.35	12.40	15.27	7.49
	Std. Deviation	5.858	9.141	8.502	8.587
Most Extreme Diffs	Absolute	.105	.231	.206	.319
	Positive	.105	.144	.206	.319
	Negative	-.067	-.231	-.189	-.297
Kolmogorov-Smirnov Z		1.750	3.795	3.374	5.023
Asymp. Sig. (2-tailed)		.004	.000	.000	.000

a Test distribution is Normal.
b Calculated from data.

One-Sample Kolmogorov-Smirnov Test

		IPQ-R Accident/ Chance	GWBI Total Score	CDQ Total Score	AGE
N		278	274	276	279
Normal Parameters(a,b)	Mean	4.83	78.17	152.28	54.0758
	Std. Deviation	8.140	15.468	26.493	14.68605
Most Extreme Diffs	Absolute	.369	.065	.114	.073
	Positive	.369	.034	.065	.045
	Negative	-.350	-.065	-.114	-.073
Kolmogorov-Smirnov Z		6.218	1.071	1.902	1.227
Asymp. Sig. (2-tailed)		.000	.202	.001	.098

a Test distribution is Normal.
b Calculated from data.

Psychology & Health

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