

**GLUTEN-FREE DIET ADHERENCE IN
ADULT COELIAC DISEASE:
EXPLORING MULTIPLE
PERSPECTIVES**

By

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Abstract

Background: Coeliac disease is an autoimmune disease triggered by an inappropriate immune response to dietary gluten. This condition affects around 1% of the population and can lead to serious health complications, including nutrient deficiencies, infertility, osteoporosis and cancer. Symptoms, such as diarrhoea, pain, fatigue and bloating, can be debilitating. The only treatment is a life-long gluten-free diet. Up to 58% of adult patients have sub-optimal adherence to a gluten-free diet, yet the reasons for this are poorly understood. The aim of this study was to gain a better understanding of the factors affecting adherence to a gluten-free diet in adult coeliac patients.

Methods: Concept mapping is a participatory mixed method that involves generation of ideas through brainstorming. Ideas are prioritised and grouped for similarity by participants, producing visual concept maps that represent participants' perceptions about what affects adherence to a gluten-free diet.

Results: Seventy-three participants were recruited (34 adult coeliac patients; 21 adults who live with them (household members); and 18 healthcare professionals). Analysis revealed a concept map containing 13 thematic clusters: The high cost of gluten-free food was perceived to be the most important factor. Healthcare professionals perceived the availability of gluten-free sandwiches to be significantly less important than people with

coeliac disease and household members. Other factors included: knowledge and information about coeliac disease and the gluten-free diet; access to gluten-free food; motivation and support; and difficulties eating away from home. There was a high degree of consistency between the perceptions of the three stakeholder groups.

Conclusions: This study identified a complex interplay of factors associated with adherence to a gluten-free diet and their relative importance. This study provides a better understanding of how to support adherence to a gluten free diet in adults with coeliac disease. This knowledge could be used to inform interventions to improve dietary adherence.

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List of abbreviations

ACBS	Advisory Committee on Borderline Substances
BDA	British Dietetic Association
BMD	Bone mineral density
BSG	British Society of Gastroenterology
CD	Coeliac disease
CDQ	Celiac disease questionnaire
DH	Dermatitis herpetiformis
GF	Gluten-free
GFD	Gluten-free diet
GFF	Gluten-free food
GP	General Practitioner
GSRS	Gastrointestinal symptom rating scale
HLA	Human lymphocyte antigen
HRQoL	Health related quality of life
IDA	Iron-deficiency anaemia
NHS	National Health Service
NICE	National Institute for Health and Care Excellence
NNUH	Norfolk and Norwich University Hospital
NTD	Neural tube defect
QoL	Quality of life
SCT	Social cognitive model
TPB	Theory of planned behaviour

tTG	Tissue transglutaminase
UK	United Kingdom
VA	Villous atrophy
WHO	World Health Organization

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Chapter 1: Introduction and background

1.1 Introduction

Coeliac disease (CD) is a chronic disease characterised by an immune response to dietary gluten (Hall *et al.*, 2013). The wide-range of symptoms, nutrient deficiencies and serious health conditions associated with CD can only be avoided if patients adhere to a strict, life-long gluten-free diet (GFD) (Silvester & Rashid, 2007; Jacobsson *et al.*, 2012). A diet that is free of gluten (principally from wheat, rye and barley) is the only successful treatment for CD (Green & Cellier, 2007; Herman *et al.*, 2012). Gluten is ubiquitous in the modern Western diet and following a strict GFD for life places a great demand on people's capacity to adapt. Commonly consumed gluten-containing products, such as bread, pasta, pizza, cakes, beer and breakfast cereals should be avoided by people with CD.

Effective management of CD can be difficult to achieve and following a GFD can affect individuals both personally and socially (Lerner, 2010). As many as 58% of patients do not adhere to treatment, however, the reasons for suboptimal adherence are not well understood (Hall *et al.*, 2009).

The aim of this study was to gain a better understanding of the factors affecting adherence to a GFD in adults with CD. A further aim was to identify similarities and differences in the perceptions of three stakeholder groups (patients, people who live with them (household members) and healthcare professionals) in relation to adherence to a GFD. A better understanding of the factors affecting dietary adherence and the perceptions of different stakeholder groups could be helpful in the design of an intervention to improve adherence to a GFD in adults with CD.

I begin this chapter with an introduction to CD, incorporating an overview of the history of our understanding of the condition and leading up to a modern definition. The prevalence, clinical features and symptoms of CD are explored along with the associated conditions that can develop when CD is not treated with a strict GFD. Contradictory evidence exists on the impact of CD and the GFD on patient quality of life (QoL) and I discuss some of the research in this area. Recent advances in diagnostic testing for CD are presented and I explore the search for alternative treatments for CD. The complex issue of adherence to a GFD is presented and I provide a definition of the term 'adherence' in relation to the GFD. I describe some of the psychological models of behaviour that are most relevant to adherence to treatment. In this chapter I review current healthcare and other sources of support available to adults with CD in the UK. I conclude this chapter with the aims and objectives for this piece of research along with an outline of the structure of this thesis.

1.2 Coeliac disease

1.2.1 What is coeliac disease?

CD is a life-long autoimmune disease characterised by an inappropriate immune response to dietary gluten, principally from wheat (gliadin), rye (horedin) and barley (secalin) (Zarkadas *et al.*, 2006). The term 'coeliac disease' (also known as gluten-sensitive enteropathy and coeliac sprue) originates from the word *koeliakos*, meaning 'suffering in the bowels' (Cataldo & Montalto, 2007). This condition occurs in genetically predisposed individuals who carry the human lymphocyte antigen (HLA)-DQ2 or -DQ8 (Trynka *et al.*, 2010). Genetic studies have found the prevalence of CD for first-degree relatives to be around 10% (Berrill *et al.*, 2012).

Erosion of the intestinal villi (villous atrophy (VA)) in response to dietary gluten leads to a reduced capacity to absorb most nutrients (Ciclitira *et al.*, 2005; Kaukinen *et al.*, 2010). CD presents with a wide-range of symptoms and untreated patients are at a higher risk of developing a number of serious health complications, including malignant diseases (Green *et al.*, 2003), infertility (Raymond *et al.*, 2006) and osteoporosis (Garcia-Manzanares & Lucendo, 2011). Rubio-Tapia *et al.* (2009) report that mortality rates in CD are double that of the wider population. A strict GFD is the only effective treatment for CD (Lerner, 2010). Symptoms resolve when dietary intakes of

gluten are removed and a strict GFD leads to the normalisation of mortality rates (Leffler *et al.*, 2008).

1.2.2 The prevalence of coeliac disease

The prevalence of CD has been underestimated in the past because many people with CD are asymptomatic or they experience mild symptoms which are not investigated (National Institute for Health and Care Excellence (NICE), 2009a). In the past, CD was believed to be mostly confined to Northern Europe and Australasia (Kang *et al.*, 2013). Evidence from large CD screening studies shows a prevalence of between 1% and 3% in most parts of the world (World Gastroenterology Organisation, 2007). A systematic review in 2013 reported that CD is rare in sub-Saharan Africa and the Orient (Kang *et al.*, 2013). Of the 266 studies included in the systematic review by Kang *et al.* (2013) only six biopsy-proven cases in ethnic Japanese and eighteen cases among ethnic Chinese were reported. In the UK, the prevalence of CD is estimated to be around 1% (Cataldo & Montalto, 2007; Rubio-Tapia *et al.*, 2009; Aggarwal *et al.*, 2012). Fifteen studies using serological tests on adult populations showed a prevalence of 0.07% to 1.9% (National Institute for Health and Care Excellence (NICE), 2009a). Three of the 15 studies were conducted in the UK and these showed a prevalence of 0.8 – 1.9%.

The rates of CD diagnosis are increasing in many countries, including the UK (Green & Jabri 2003; Mustalhati *et al.* 2010; Violato *et al.* 2012). The

increase in diagnosis has been attributed to improvements in the accuracy of diagnostic testing and better awareness of the wide-ranging symptoms (Loftus & Murray, 2003). Despite the rise in diagnosis, CD remains undiagnosed or misdiagnosed in the majority of cases (Lohi *et al.*, 2007; National Institute for Health and Care Excellence (NICE), 2009a). This may be due to the protean nature of CD symptoms and the fact that approximately 50% of people with CD are asymptomatic (Casellas *et al.*, 2006; Tursi *et al.*, 2009).

Some researchers argue that environmental factors have led to a true rise in CD, regardless of whether or not cases are diagnosed (Lohi *et al.*, 2007; Ivarsson *et al.*, 2013). Gluten is being used more commonly by food manufacturers as an ingredient where it didn't used to be present (e.g. as a thickener and food coatings) (McCary, 2010; Zarkadas, 2006). It is possible that this increased exposure to gluten is linked with the increased prevalence of CD. Bardella *et al.* (2000) and Shale *et al.* (1982) found discordance for CD in monozygotic twins and this is suggestive of an environmental influence on the development of the disease as well as the genetic association. It has been suggested that the age of weaning a child onto gluten foods may influence the development of CD (Ivarsson *et al.*, 2013; Guandalini, 2007). It is recommended that children should be weaned no sooner than three months and no later than seven months of age (Guandalini, 2007). The evidence on the environmental influences on the development of CD is limited and more research is needed.

CD was previously thought to have the highest prevalence in people of European origin (Devlin *et al.*, 2004; Cataldo & Montalto, 2007), however, new evidence shows that CD is common across many ethnic groups (Barada *et al.*, 2010; Araya *et al.*, 2000; Cataldo & Montalto, 2007). Populations with high exposure to dietary gluten, such as the Italian population, tend to have a higher prevalence of CD (Volta *et al.*, 2001). The highest prevalence of CD (5.6%) was identified in a North African tribal population who consume a wheat-based diet (Barada *et al.*, 2010). Over-exposure to gluten could account for the high prevalence of CD identified in this tribe.

Two studies that compared frozen serum samples taken several years ago with current samples reported that CD had increased (Rubio-Tapia *et al.*, 2009; Lohi *et al.*, 2007). Rubio-Tapia *et al.* (2009) compared CD test results on 9,133 frozen serum samples collected from men in the US Air Force between 1948 and 1954 with current serological tests from 7,210 healthy young men and 5,558 older men. This study identified a 4 and 4.5-fold increase in the prevalence of CD. The difference could be explained by the fact that people with CD may have been less likely to join the Air Force due to poorer health. Lohi *et al.* (2007) compared the results of CD tests in a cohort of 8,000 adults taken between the years 1978 and 1980 with the test results for a sample of 8,028 adults taken between the years 2000 and 2001. After adjusting for age and sex, this study reported that CD prevalence had doubled over two decades. The evidence from these two studies suggests a true increase in the prevalence of CD, regardless of improvements in detection methods.

Richard Logan developed the concept of the '*coeliac disease iceberg*' in 1991 to highlight the fact that for every diagnosed case of CD, many cases remain undetected (World Gastroenterology Organisation, 2007; West *et al.*, 2007). Despite the recent increase in diagnosed cases, evidence suggests that 75-90% of the coeliac population remains undetected in Western countries and mass-screening has been advocated by some researchers (Kaukinen *et al.*, 2010). Poor detection of CD may be due to misdiagnosis (CD symptoms are often indistinguishable from irritable bowel syndrome (IBS) symptoms) or because CD is often asymptomatic (Fasano & Catassi, 2001). Although mass-screening could reduce or eliminate the '*clinical iceberg*', it could result in reduced QoL as a result of the drastic changes patients are required to make following diagnosis (Collin, 2005; Paavola *et al.*, 2012).

CD can be diagnosed at any age after an infant has been weaned onto a gluten-inclusive diet (National Institute of Health and Care Excellence (NICE), 2009a). In the past, CD was most commonly diagnosed in childhood, however, the pattern of diagnosis has changed and the average age of diagnosis in the UK is currently between 40-60 years (Rashtak and Murray, 2009).

More females than males are diagnosed with CD, however, Ageep (2012) tested 172 patients suspected to have CD and found that men and women were equally affected. One possible explanation is that women are more likely to visit their GP and CD is more likely to be detected during

pregnancy when immune function is altered and nutrient deficiencies may be identified (Gazzola, 2011). Conflicting evidence on the association between gender and adherence to a GFD exists and further research is needed (Hall *et al.*, 2009).

Regardless of whether environmental factors or improved diagnosis are responsible for the rising prevalence of CD, improving adherence to the GFD in this growing population is a significant challenge. A better understanding of the factors affecting adherence is needed. In the next section I report how our understanding of CD has developed over the years.

1.2.3 The history of coeliac disease

Because wheat, rye and barley have not always been present in the human diet, CD has not always been around. As agricultural settlements developed and crop cultivation spread across the world, dietary intolerance to gluten emerged (Gazzola, 2011). CD was unlikely to have been recognised as a new illness because the symptoms of diarrhoea, lethargy and nutrient deficiency would have been common at that time due to bacterial infections.

Recognition of CD originates from around 50AD (Cataldo & Montalto, 2007). However, it was not until the late nineteenth century when Samuel Gee recognised that patients could only be treated by diet (Richman, 2012). Paediatrician Sydney Haas claimed that all eight children in his study were cured on a banana diet, whereas, two additional children, who did not follow

the banana diet, died (van Berge-Henegouwen & Mulder, 1993). At the end of the second world war, the Dutch paediatrician, Willem-Karel Dicke observed that the health of children with CD improved when wheat and rye flour supplies were cut off by the Nazis and relapsed when the crops became available again (Van Berge-Henegouwen & Mulder, 1993; Gazzola, 2011). Dicke had suspected the role of wheat in CD and by the 1950s he identified the role of gluten as the causative antigen of CD (Gazzola, 2011).

In 1954 the British physician, John Paulley, reported that the damage to the small intestinal mucosa found in CD is caused by dietary gluten (Gazzola, 2011). Paulley also found that a GFD could restore the gut architecture and the inflammation of the intestinal lining in coeliac patients (Gazzola 2011). Intestinal biopsy was subsequently developed as a technique to identify CD and today this method of diagnosis is considered to be the 'gold standard' (Ludvigsson & Green, 2011). In the 1970s, the genetic underpinnings of CD were discovered and the discovery of tissue transglutaminase (tTG) was made in 1997 (Schuppan *et al.*, 2005). This discovery led to the development of tTG blood tests for use in the diagnosis and monitoring of CD. Much of our knowledge about CD and the GFD is relatively new and there remain many unanswered questions (Hall *et al.*, 2009; Leffler *et al.*, 2008).

1.2.4 Modern definition of coeliac disease

The first consensus definition of CD in 1970 did not take account of the genetic or immunological aspects of CD. This reflects the lack of understanding about these factors at that time (Meeuwisse, 1970). The 1970 consensus definition identified CD as:

'a permanent condition of gluten intolerance with mucosal flattening that reversed on a gluten-free diet (GFD) and then relapsed on re-introduction of gluten'.

(Meeuwisse, 1970)

Until recently, there has been a lack of consensus on a revised definition of CD. In 2011, a multidisciplinary task force consisting of 16 physicians from seven countries redefined CD following a search of the literature for terms relating to CD (Ludvigsson *et al.*, 2013). Our previous understanding of CD as a gastrointestinal disorder which mainly affects children has progressed to the modern definition of CD (known as the 'Oslo definitions'), which is:

'a chronic small intestinal immune-mediated enteropathy precipitated by exposure to dietary gluten in genetically predisposed individuals'.

(Ludvigsson *et al.*, 2013)

It has been argued that this definition is not very lucid (Marsh, 2013) and the terminology used may not be easily understood by the lay person. However, it does clear up some confusion around the terminology used to describe gluten-related disorders.

In developing their definition of CD, Ludvigsson *et al.* (2013) identified a number of terms used to describe the spectrum of disorders related to the ingestion of gluten and they developed a set of definitions. Table 1.1 lists the definitions for terms that are relevant to this study. The umbrella term 'gluten sensitivity' includes the terms listed in Table 1.1 as well as 'gluten intolerance', which is a response to gluten without significant immune reaction and with a negative blood test for CD. Refractory CD is a rare type of CD occurring in approximately 5% of patients and presents with persistent malabsorptive symptoms and VA, despite strict adherence to a GFD for 12 months or longer (Rubio-Tapia & Murray, 2010; Ludvigsson *et al.*, 2013). This thesis addresses the types of CD listed in Table 1.1 with the exception of latent, potential and subclinical CD, which would not fit the criteria for a CD diagnosis. Refractory CD cannot be treated with a GFD and, therefore, is not relevant to this study. Dermatitis herpetiformis (DH) is a skin condition that is related to CD and is also treated with a GFD. I chose not to include DH in this study because the factors affecting adherence to a GFD in DH may differ to those in CD.

Table 1.1 - Definition of terminology for disorders relating to gluten ingestion

Name of disorder	Definition
Asymptomatic coeliac disease ¹	No symptoms are exhibited. Patients with asymptomatic coeliac disease are usually detected through screening.
Atypical coeliac disease ²	Historically, this term has been used to describe patients who do not experience weight-loss but they do exhibit one or more of the following symptoms: gastrointestinal symptoms; extra intestinal manifestations; neurological problems; reproductive problems; oral/cutaneous disease; and skeletal disorders.
Classical coeliac disease	Presenting with signs and symptoms of malabsorption, which includes diarrhoea, steatorrhoea (fatty stools), weight-loss or growth failure.
Latent coeliac disease ³	Positive serological test for coeliac disease but absence of villous atrophy.
Non-classical coeliac disease	Presenting without signs of malabsorption.
Overt coeliac disease ⁴	See symptomatic coeliac disease.
Potential coeliac disease ³	People with normal intestinal mucosa but with positive serology (see also latent coeliac disease).
Silent coeliac disease ¹	See asymptomatic coeliac disease.
Subclinical coeliac disease	Below the threshold of clinical detection and symptoms are not sufficient to trigger coeliac disease testing in normal practice.
Symptomatic coeliac disease ⁴	Clinically evident symptoms (gastrointestinal or extra intestinal) that can be attributed to gluten intake.
Typical coeliac disease ²	Presenting with signs or symptoms of malabsorption (e.g. diarrhoea or malnutrition).

Notes:

1. Ludvigsson *et al.* (2013) discourage the use of the term 'silent coeliac disease' and suggest the term 'asymptomatic' should be used instead.
2. Ludvigsson *et al.* (2013) discourage the use of the terms 'atypical coeliac disease' and 'typical coeliac disease' as these terms do not reflect the modern understanding of coeliac disease as a condition that presents with a wide spectrum of symptoms.
3. Ludvigsson *et al.*(2013) discourage the use of the term 'latent coeliac disease' as it is often used interchangeably with the term 'potential coeliac disease' and causes confusion.
4. Ludvigsson *et al.* (2013) discourage the use of the term 'overt coeliac disease' and suggest the term 'symptomatic coeliac disease' should be used instead.

1.2.5 Clinical features and symptoms

Patients with CD vary in the type and severity of symptoms experienced.

Until the late 1970's CD was perceived to be a disease affecting the gastrointestinal system (Lohi *et al.*, 2007). CD is no longer seen as just an intestinal disease and it is now recognised as a systemic disease that affects the entire body, has impact on a patient's social life and can cause

psychological issues (Leffler *et al.*, 2003; Fera *et al.*, 2003; Ukkola *et al.*, 2012). Evidence from several countries, including the UK, shows that as many as 50% of newly diagnosed coeliac patients present without symptoms (Fasano & Catassi, 2001). Symptoms of CD can be broken down into gastrointestinal and extra intestinal symptoms (Lohi *et al.*, 2007):

Gastrointestinal symptoms

In CD, lymphocytes detect and respond to gluten in the diet and this inflammatory response causes damage to the small intestinal mucosa, resulting in VA. VA leads to nutrient malabsorption which causes weight-loss and nutrient-deficiency in coeliac patients. The classically recognised gastrointestinal symptoms of CD include diarrhoea, abdominal distension, steatorrhoea and abdominal pain (National Institute for Health and Care Excellence (NICE) 2009b; Ludvigsson *et al.*, 2013; Nachman *et al.*, 2009). A validated gastrointestinal symptom rating scale (GSRS) has been used to assess gastrointestinal symptoms in CD (Ilus *et al.*, 2012; Roos *et al.*, 2009). The GSRS is a questionnaire divided into five categories: diarrhoea, indigestion syndrome, constipation, abdominal pain and gastro-oesophageal reflux (Ilus *et al.*, 2012). Casellas *et al.* (2006) and Tursi *et al.* (2009) found as few as 55% and 45% of patients present with classical CD symptoms respectively and, therefore, the GSRS is no use in monitoring symptoms in around half of all patients.

Extra intestinal manifestations

Signs and symptoms of CD that occur outside the intestinal system include: fatigue; neurological conditions (including depression); reproductive diseases; oral problems; and skeletal problems (Ludvigsson, *et al.*, 2013). Untreated coeliac patients are at an increased risk of developing serious complications, including malignancies, other autoimmune conditions (e.g. insulin-dependent diabetes), iron-deficiency anaemia (IDA) and osteoporosis (Brousse & Meijer, 2005; Colleran *et al.*, 2009; Freeman, 2012). CD is associated with significant morbidity and a doubling of mortality rates compared to the wider population (Rubio-Tapia *et al.*, 2009). This increase in mortality is mainly due to a heightened risk of lymphomas (Brousse & Meijer, 2005; Silano *et al.*, 2008). Five conditions that are commonly associated with CD are presented in more detail below.

- **Iron deficiency anaemia (IDA)**

The most common condition associated with CD, which is found in two-thirds of all adult CD patients, is IDA (Griffiths, 2008). The main symptom of IDA is fatigue, which is seen in approximately 82% of CD patients (García-Manzanares & Lucendo, 2011). VA in CD leads to the malabsorption of nutrients, such as iron and the British Society of Gastroenterology (BSG) (2009) recommend that patients who have IDA should be screened for CD. IDA leads to lethargy and patients with CD should be advised to eat a GFD containing high-iron foods (World Gastroenterology Organisation, 2007).

- Reduced Bone Mineral Density and Osteoporosis

As many as 40% of patients with CD have calcium and vitamin D malabsorption, resulting in diminished bone density or osteoporosis (Leeds *et al.*, 2008). People with undiagnosed or untreated CD are likely to experience calcium and vitamin D malabsorption and low bone mineral density (BMD) is common in patients diagnosed with CD over the age of 40 (Griffiths, 2008). A study by Casella *et al.* (2012) reported a prevalence of osteoporosis of 67% in male and 70% in female patients with CD aged >65 years compared to 14% for male and 9% for female patients with CD aged 18-64 years. Studies have shown that adherence to a GFD reverses calcium malabsorption and leads to a rapid increase in BMD (Bai *et al.*, 1997; Larussa, 2012).

- Malignancy

Malignancies that occur at a higher prevalence in people with CD include intestinal lymphoma, adenocarcinoma of the small intestine, the pharynx and the oesophagus (Catassi *et al.*, 2005). The risk of malignancy in unmanaged CD is thought to be two-fold that of the wider population; for small intestinal lymphoma the risk may be as high as 50-fold (Leeds *et al.*, 2008). Holmes *et al.* (1989) found that the risk of malignancy was similar to the wider population after five years on a GFD, whereas the risk was elevated in patients who did not adhere to the GFD.

- Reproductive problems - Women with CD are prone to reproductive problems. A study involving 11,000 women with CD of fertile age (15-45 years) reported that fertility was lower in this cohort for the two years prior to diagnosis compared to an age-matched control group (Zugna *et al.*, 2010). Women with CD are likely to have a reduced period of fertility due to late menarche and earlier menopause (Zugna *et al.*, 2010). Children born to women with CD have an increased risk of being born prematurely and of having a low birth weight (Martinelli *et al.*, 2000). Following diagnosis and the uptake of a GFD, fertility in women with CD has been found to return to levels equal to that of the wider population (Sher & Mayberry, 1996).
- Psychological conditions - Coeliac disease is associated with an increase in the incidence of psychological symptoms (Addolorato *et al.*, 2004; Sainsbury *et al.*, 2013a; Addolorato *et al.*, 2008), including depression (Arigo *et al.*, 2012; Ciacci, 1998; Siniscalchi *et al.*, 2005), anxiety (Addolorato *et al.*, 2004; Fera *et al.*, 2003) and eating disorders (Leffler *et al.*, 2007a).

1.2.6 Quality of life and coeliac disease

Lee *et al.* (2012) acknowledge that eating encompasses more than just meeting the physiological need for nutrients. The act of eating is interwoven into the fabric of our lives and culture, as well as our social and emotional

needs. Several studies have reported a reduced quality of life (QoL) in people with CD (Barratt *et al.*, 2011; Lee & Newman, 2003; Hauser *et al.*, 2006; Fera *et al.*, 2003).

Over the past 20 years there has been an increased focus on the development of instruments to measure health-related QoL in patients with digestive conditions (Yacavone, 2001). The Coeliac Disease Questionnaire (CDQ) is reported to be a reliable and valid tool for measuring health-related quality of life (HRQoL) in adult CD (Hauser *et al.*, 2007). The CDQ contains four subscales: gastrointestinal symptoms; emotional well-being; social restrictions; and disease-related worries.

Females commonly report a lower QoL compared to males with CD (Jacobsson *et al.*, 2012). Feelings of deprivation in relation to following a GFD and a higher desire to control the preparation of food are reported more by females than males (Hallert *et al.*, 2003). Having an intense focus on the avoidance of gluten can help in avoiding CD symptoms, but it may take the enjoyment out of eating. This focus on food may give rise to psychological issues, including eating disorders, as well as reducing QoL (Arigo *et al.*, 2012; Leffler, 2007b).

1.2.7 Diagnosing coeliac disease

In the past, CD was most commonly diagnosed in childhood, however, the pattern of diagnosis has changed and the average age of diagnosis in the UK is now over 40 years (Ciclitira *et al.*, 2010). Patients have expressed

dissatisfaction with the length of time taken by General Practitioners (GPs) to diagnose CD (British Society of Gastroenterology 2009). Delayed diagnosis could be due to the variability of symptoms as well as the common perception that CD is not a common disease. The National Institute for Health and Care Excellence (NICE) (2009) recommend that patients should be tested for CD if they show the following symptoms:

- chronic or intermittent diarrhoea
- failure to thrive or faltering growth (in children)
- persistent or unexplained gastrointestinal symptoms including nausea and vomiting
- prolonged fatigue ('tired all the time')
- recurrent abdominal pain, cramping or distension
- sudden or unexpected weight loss
- unexplained iron-deficiency anaemia, or other unspecified anaemia

In the UK, a patient suspected of having CD will be offered a serological test. No single test can diagnose or exclude CD, however, CD blood tests have become more reliable in recent years. The most highly sensitive and specific blood tests for CD are the IgA anti-tissue transglutaminase (tTG) antibody and the immunoglobulin A (IgA) endomysial antibody (EMA) indirect immunofluorescence assays (Ciclitira *et al.*, 2010). Both tests provide sensitivities of around 95% and specificity of 100% (Hourigan 2006).

If the result of the serological test is positive, the patient will be offered an intestinal biopsy to confirm the diagnosis. The diagnostic hallmark of CD is VA in the small intestine (Lee & Green, 2005), which is confirmed by intestinal biopsy. Intestinal biopsy is considered to be the gold standard in diagnosing CD (Leeds *et al.* 2008), however, problems can occur if the sample is not taken from the correct location or if insufficient sample is taken.

In order to assure correct diagnosis, patients need to be eating a gluten-containing diet at the time of diagnosis. If a person with CD is following a strict GFD when they are tested, they are likely to have no antibodies and the intestine may have healed. This would result in a false-negative result (McCary, 2010). People with severe symptoms in response to gluten may be reluctant to go back on a GFD to get a medical diagnosis. The advantage of receiving an accurate diagnosis in the UK is that patients will be offered healthcare advice and gluten-free food (GFF) on prescription. Much of this advice, however, is freely available from coeliac support groups and other sources.

Only around 10% of people affected with CD receive an accurate diagnosis and this has led to the argument in favour of mass screening for CD (Collin, 2005; Hershcovici *et al.*, 2010). Undiagnosed CD is a concern because of the symptoms and associated conditions, such as osteoporosis, infertility and malignancy. However, the risk of serious health sequelae from CD are relatively low and the health economic argument for the value of mass

screening is dubious. If mass screening for CD was introduced in the UK, the cost to the NHS may be substantial. Not only would the NHS need to pay for the cost of the test, but money would also need to be found to provide a higher number of patients with prescribed GFF and appropriate healthcare. However, the costs and negative consequences associated with CD-related illness may be reduced through mass screening, provided that patients adhere to the GFD.

1.2.8 Treating coeliac disease

In the past, it has been incorrectly suggested that CD is a curable condition (Hopman *et al.*, 2008). CD is now recognised as a life-long condition that needs to be treated with a permanent gluten-free diet. Since gluten was identified as the causative antigen in CD in the 1950s, a gluten-free diet has been the only accepted treatment (Rubio-Tapia *et al.*, 2010). The clinical end points that justify the efficacy of a GFD as a treatment for CD are the elimination of symptoms and the correction (or prevention) of nutrient deficiencies. A reduction in the long-term complications of CD is also a justification for this treatment.

With advances in our understanding of the molecular basis of CD, research into alternative treatments has been carried out in recent years. This work has been driven by the fact that the GFD is difficult to adhere to and many patients feel dissatisfied with having to stick to such a restrictive diet (Aziz *et al.*, 2011; Bakshi *et al.*, 2012). Investigations into novel treatments for

CD have included genetic modification of grains, alterations to gut permeability and vaccinations (Bakshi *et al.*, 2012). To date, no successful alternative to the GFD has been established and the GFD is likely to remain the only accepted treatment for CD for the foreseeable future (Bakshi *et al.*, 2012). As no alternative treatment is currently available, it is important that patients are supported to adhere to a GFD.

1.3 The gluten-free diet

1.3.1 The role of gluten in food production

Gluten is an alcohol-soluble protein found in wheat, rye and barley (Fasano & Catassi, 2001). The elastic and cohesive quality of gluten binds ingredients together and provides structure to baked goods. For example, gluten forms a three-dimensional mesh-like structure that traps bubbles of carbon dioxide, helping bread to rise (Shewry *et al.*, 2002).

Baking without gluten is challenging and the main problems are achieving a good flavour, structure and texture (Moore *et al.*, 2006). GF dough is much less cohesive and more like a batter, which makes it difficult to handle compared to wheat dough (Houben *et al.*, 2012). GF baking involves the replacement of gluten-inclusive flours with GF flours, such as rice flour and corn flour, which differ in flavour and appearance to gluten-inclusive flours (Gambus *et al.* 2009). The end product of GF baking is very different in taste and texture to gluten-inclusive baked products. The GF versions tend to be

more crumbly, giving a dry feeling in the mouth and a less satisfying taste (Houben *et al.*, 2012; Gambus *et al.*, 2009). In addition to having poorer sensory qualities, GF baked goods tend to have a shorter shelf-life due to the quick drying effect of the crumb and a more rapid staling time (Gambus *et al.*, 2009).

GF biscuits are less difficult to produce than GF bread and cakes because the high sugar and fat content helps with achieving the desired crispy texture (Marti & Pagani, 2013). Producing good quality GF pasta that is firm and strong enough to withstand the cooking process is challenging (Marti & Pagani, 2013). GF pasta is often produced using either rice flour or corn flour and the sensory characteristics of GF pasta are generally perceived to be poorer than gluten-inclusive pasta (Marti & Pagani, 2013).

People who follow a GFD often report dissatisfaction with the sensory characteristics of GFF compared similar gluten-inclusive products (Diaz-Amigo & Popping, 2012). Araujo and Araujo (2011) found that only 34% of participants with CD were very satisfied with the texture of GFF. A study by Sverker *et al.* (2005) reported that tasty and varied GFF was not easy to find and that following a GFD was monotonous. Studies by Biagi *et al.* (2004), Lee *et al.* (2012) and Hopman *et al.* (2008) reported participants' perceptions of GFF as 'unpleasant', 'tasteless' and 'unpalatable'.

With the aim of improving the quality of GF products, bakers have introduced natural, synthetic and biotechnological ingredients which behave

in a similar way to gluten (Houben *et al.*, 2012). Hydrocolloids, such as xanthan gum and guar gum are often used in place of gluten to improve the viscosity, crumb structure and to slow down the staling process (Huttner & Arendt, 2010). Another method of improving the sensory qualities of GFF was the introduction of Codex wheat starch in GFF production (Coeliac UK, 2014a). Starches are derived from cereal grains and they enhance the taste and texture of food. Codex wheat starch is a specially processed wheat starch that has been washed to reduce the gluten content in accordance with Codex standards (Coeliac UK, 2014a). Despite these efforts to improve the sensory characteristics of GFF, there are still no GF crusty French loaves, filo or puff pastry of comparable quality to those containing gluten (Houben *et al.*, 2012).

1.3.2 What is a gluten-free diet?

A GFD is a diet that is free from wheat, rye and barley and their derivatives (Raymond *et al.*, 2006). The GFD usually consists of both naturally GF products (e.g. fruit and vegetables, meat and dairy products) and GF substitutes (e.g. GF bread, biscuits and pasta). The term ‘gluten-free’ implies no gluten is present, however, this is not necessarily the case (Thompson, 2001). It has been argued that an absolute GFD is unrealistic and that occasional ingestion of gluten may not cause significant risk to many people with CD. Most CD patients can tolerate small amounts of gluten, but the amount of gluten tolerated, without experiencing any

deleterious effects, varies between individuals (Bold & Rostami, 2011; Kaukinen *et al.*, 1999).

Products that are labelled as 'gluten-free' are allowed to contain up to 20ppm of gluten (Food Standards Agency, 2012). Codex wheat starch, which is processed to contain less than 20ppm of gluten, is often used as an ingredient in the production of GFF (Coeliac UK, 2014a). Products containing ≤ 20 ppm gluten are considered to be safe for people with CD and most people with CD can tolerate these products. However, some patients require a naturally gluten-free diet in order to remain symptom free (Collin *et al.*, 2004; Biagi *et al.*, 2009). People with CD may also choose to consume products that are labelled as 'low in gluten'. In the UK, these products are allowed to contain up to 200ppm of gluten and are considered to be safe for most people with CD (Food Standards Agency, 2012).

Gluten-containing crops are widely consumed in the West and adapting to a GFD can involve drastic changes to normal dietary habits (Zarkadas, 2006). Patients are required to follow a strict life-long diet that is free from many of the foods they may have enjoyed before diagnosis. For the rest of their lives they cannot eat most of the food available on restaurant menus, they cannot order an ordinary pizza, they can never taste a warm crusty French baguette, or enjoy an ordinary glass of beer in the pub with friends, or a slice of birthday cake at a birthday party. Coeliac patients often complain that the quality of GF substitute foods are not equal to that of their gluten-containing counterparts (Leffler *et al.*, 2008).

1.3.3 Oats and the gluten-free diet

The safe inclusion of oats in a GFD has been contested over the years. Oats contain a type of gluten, known as avenins, which was previously believed to be harmful to people with CD (Diaz-Amigo & Popping, 2012). In recent years, studies have found that pure, uncontaminated oats can be safely consumed by people with CD. The evidence regarding the safe inclusion of oats in a GFD was synthesised in a systematic review by Haboubi *et al.* in 2012. It is now widely accepted that pure, uncontaminated oats are not harmful to coeliac patients. However, oats are often milled in the same factory as wheat, rye or barley and it is common for cross-contamination to occur. Therefore, patients with CD are advised only to consume oats that are labelled as 'Gluten-free.' Some people with CD have other dietary sensitivities which could include avenins and these patients should be advised to avoid oats.

1.3.4 The cost of gluten-free products

GFF is a niche market and GF products tend to be expensive (Mendoza 2005; Singh & Whelan, 2011). When comparing the cost of a basket of GF products with a similar basket of gluten-inclusive products in the USA, Lee *et al.* (2007) found that every GF product was more expensive than its gluten-inclusive counterpart. GF bread and pasta were found to be around twice the price of similar gluten-containing products. Similar findings have been made in studies in the UK where all categories of GF products were

found to be more expensive than equivalent gluten-containing products (Coeliac UK, 2009; Singh and Whelan, 2011). To bring down the cost of a GFD in line with that of a gluten-inclusive diet, GFF is available on prescription for people with a confirmed CD diagnosis in the UK.

1.3.5 The availability of gluten-free products

Zarkadas *et al.* (2006) reported that 83% of CD patients had difficulties with finding GFF in shops. However, this study was conducted in Canada where GFF availability may differ to that in the UK. Heller (2009) argues that GFF has become more widely available in the UK for three main reasons: 1. CD diagnosis has increased; 2. The food industry is responding to a niche market; 3. Advances in modern technology. A recent increase in anxiety over gluten-consumption in people who do not have CD has also driven the demand for GFF (Anderson 2008). This has led to an increase in the size of the market and enhanced the availability of GFF.

GF products are now widely available for purchase on the internet, although they are often expensive. Coeliac UK's Food and Drink Directory (Coeliac UK, 2013a) lists over 11,000 manufactured GF products which are available to buy in the UK. However the availability of GFF in specific contexts e.g. when travelling and eating away from home can be problematic for people with CD (Cureton, 2006). The availability of GFF on prescription in the UK enhances access for those with a diagnosis of CD (Coeliac UK, 2013b). There has been some debate over the recent cut-backs in the variety and

quantity of GFF available on prescription, and the cost of these foods to the NHS (Coeliac UK, 2013b).

1.3.6 'Hidden' gluten and gluten contamination

Avoiding gluten is a major challenge for people with CD. If gluten is consumed, either accidentally or intentionally, it can cause severe symptoms. Gluten is often present in many products where it may not be expected, such as soups, sauces, ice cream, cornflakes, chocolate bars, medicines and lip balm (McCabe *et al.*, 2012). This is sometimes referred to as 'hidden gluten'. Gluten is a very useful and cheap product to use in food production and wheat flour is often used as a thickener or coating in many products that would otherwise be free from gluten (McCary, 2010; Zarkadas, 2006). People with CD need to be careful to avoid such products by carefully reading food labels (McCabe *et al.*, 2012).

GFF can become contaminated with gluten if prepared in the same environment as gluten-containing foods. This may occur when eating out or during food preparation at home (Schuppan *et al.*, 2005). Occasionally, products that are labelled as GF are recalled by the manufacturer because they are found to have been contaminated with gluten (Food Standards Agency, 2013). Even patients who have the best of intentions to stick to a GFD only need to have out of date information, or miss a gluten containing ingredient on manufactured foods, or eat out and have their GFF fried in oil that had previously been used to fry gluten-containing foods, in order for

them to accidentally consume gluten (Biagi *et al.*, 2009; McCabe *et al.*, 2012; Schuppan *et al.*, 2005). Hence it is recommended that gluten contamination should be the first thing to be addressed when a patient does not respond to a GFD (Dewar *et al.*, 2012).

1.3.7 Gluten-free food labelling

Most regulations of GFF labelling are based on the Codex Alimentarius Standard 118-1979 which are recommendations, rather than regulative (Diaz-Amigo & Popping, 2012). New GFF labelling legislation that was introduced in January 2012 has reduced the permitted amount of gluten present in foods labelled as 'gluten-free' from 200 parts per million (ppm) to 20 ppm (Food Standards Agency 2012). Foods containing up to 100 ppm can be labelled as 'low in gluten' under this new legislation. Some people may exceed their tolerable threshold if several products labelled as 'low in gluten' are consumed (Akobeng & Thomas 2008). Some foods that were previously labelled as GF are no longer permitted under the new legislation (Food Standards Agency 2012). Although this new legislation makes GF products safer for patients with very low tolerance to gluten, it is likely to have led to a reduction in the number of products labelled as 'gluten-free'. However, I am not aware of any research that has been carried out to investigate whether or not the new labelling legislation has affected the availability of GF products.

1.4 Adherence to treatment

The World Health Organization (WHO) (2003) has adopted the following definition of adherence to long-term therapy:

‘the extent to which a person’s behaviour – taking medication, following a diet, and/or executing lifestyle changes, corresponds with agreed recommendations from a health care provider’

(World Health Organization, 2003)

Adherence to a GFD results in symptomatic, serologic and histologic remission and the normalisation of mortality rates as well as improving psychological wellbeing (Pietzak, 2005; Leffler *et al.*, 2008; Griffiths, 2008). Non-adherence to a GFD renders this treatment ineffective. Adherence to medication for chronic conditions in developed countries, including the UK, is around 50% (World Health Organization, 2003). Self-management of dietary treatment tends to be more complicated than adherence to medication and dietary adherence tends to be worse than medication adherence (DiMatteo, 2004). Managing the effects of non-adherence can place a great strain on healthcare resources. As well as the reduction in symptoms and a reduced risk of CD-associated conditions, adherence to treatment can have a broader impact on society in the lowering of healthcare costs associated with complications and increasing workplace productivity (Behner *et al.*, 2012).

In theory, dietary treatments are straightforward (i.e. do not eat gluten), but in practice they are rather complicated and require a high degree of self-management. A study investigating dietary regimen adherence in diabetic patients found a wide-range of factors affecting adherence, including socio-demographic factors, psychosocial obstacles; and healthcare providers obstacles (Uchenna *et al.*, 2010). The wide-range of factors found to be associated with following a restricted diet highlight the complexity of dietary adherence compared to medication adherence, which may involve simply remembering to take a pill each day. By developing a better understanding of the factors affecting adherence to a GFD in adult CD, it should be possible to develop an intervention that will improve adherence.

1.4.1 Defining adherence to the gluten-free diet

Hall *et al.* (2009) highlight the variability in the way adherence to a GFD is defined and measured. In some studies, Likert scales have been used to define adherence as strict, partial or fairly strict, whereas other studies defined adherence based on the results of histopathology or serological tests (Hall *et al.*, 2009). Some studies have interpreted self-reported ‘partial-adherence’ or ‘fairly adherent’ to be adherent whereas other studies considered these to be non-adherent (Hall *et al.*, 2009). In addition, some studies regard accidental gluten consumption to be non-adherent, whereas other studies do not (Hall *et al.*, 2009). There is a lack of consistency in the ways in which adherence has been defined and measured in previous CD

studies and Leffler *et al.* (2009) highlight the need for a more standardised approach.

1.4.2 Measuring adherence to a gluten-free diet

Table 1.2 summarises different methods of measuring adherence to a GFD as used in previous research. VA is used as the diagnostic hallmark of CD and recovery of the villi is an indication of dietary adherence (Lee & Green, 2005). However, recovery of the intestinal mucosa may not be a good indicator of adherence to a GFD because the rate and extent of recovery is variable and, in some cases, the villi never fully recover (Rubio-Tapia *et al.*, 2010). However, the majority of patients do show signs of mucosal recovery over time (Hutchinson *et al.*, 2010).

The presence or absence of symptoms has been used in some studies as an indicator of adherence to a GFD (Jacobsson *et al.* 2012). However, it is argued that the elimination of symptoms is not a good indicator of adherence to a GFD. This is because CD symptoms are a poor guide to mucosal healing and they are irrelevant for around 50% of people with CD who are asymptomatic (Casellas *et al.*, 2006; Tursi *et al.*, 2009).

There is no one measure of adherence that is cost effective, patient acceptable and accurate. Research in this area is inherently biased as non-adherent patients may be less likely to participate in research into measures for adherence, and may also be less likely to attend clinics. Also patients

may tend to over-estimate their level of adherence (Leffler *et al.*, 2009; Hall *et al.*, 2009).

In their systematic review, Hall *et al.* (2009) found that adherence to a GFD in adults with CD ranged from 42% to 91%. The reason for the wide range of adherence estimates reported by Hall *et al.* (2009) could be due to the different ways in which adherence to a GFD was measured.

Table 1.2 Methods for evaluating adherence to a gluten-free diet in patients with coeliac disease

Method	Benefits	Limitations
Intestinal biopsy	Gold standard for demonstration of impact on gut mucosa.	Expensive, uncomfortable, can be painful, rare risk of gut perforation. Healing of the gut mucosa is often slow or incomplete, despite adherence to a gluten-free diet, it can take one to two years, so biopsy should not be repeated until after two years (Zanini <i>et al.</i> , 2010).
Serological testing	Simple blood test that can be repeated easily. Relatively cheap. Antibody titres drop when a patient adopts a gluten-free diet. Titre is associated with gut mucosa pathology (Ciacci <i>et al.</i> , 2002a).	Antibodies are not always present when someone with coeliac disease is occasionally non-adherent. The presence of antibodies provides little information about adherence and mucosal healing. There are substantial differences between the performance of each serological assay and between commercial kits for each assay, reducing their reproducibility (Korponay-Szabo <i>et al.</i> , 2005; Ciacci <i>et al.</i> , 2002a).
Dietitian-led Evaluation	Non-invasive. High correlation with biopsy and serology results. Relatively low cost (Da Silva Kotze <i>et al.</i> , 2009).	Limitation to number of dietitians skilled in evaluating adherence. Not all people with coeliac disease see a dietitian. Assumes patient honesty in reporting diet (Vahedi <i>et al.</i> , 2003; Leffler <i>et al.</i> , 2008).
Food Diary 24 hour, 3 or 7 day	Can be completed by the patient. Relatively low cost.	Relies on accurate recall of food eaten. Also requires record of manufacturer of pre-prepared foods so that their recipes can be checked for gluten. Can also require estimation of food amounts. Relies on patient honesty (Corrao <i>et al.</i> 2001; Leffler <i>et al.</i> , 2007b).
Adherence Questionnaire e.g. The Coeliac Dietary Adherence Test (Leffler <i>et al.</i> , 2009)	Simple short questionnaires completed by patients. Low cost. Correlates highly with dietitian assessment of adherence (Leffler <i>et al.</i> , 2007b; Leffler <i>et al.</i> , 2009).	Relies on patient honesty. Over-estimation of adherence may be common.

1.4.3 The financial cost of non-adherence to the National Health Service (NHS)

Non-adherence to a GFD in coeliac patients is associated with poorer health outcomes and this is likely to increase the financial burden on the NHS. A study in Israel reported significantly higher healthcare use for a group of 1,754 coeliac patients compared to 15,040 controls. The increase was based on hospital admission, medications, laboratory and imaging, some of which may have resulted from poor adherence (Heymann *et al.*, 2013). As far as I am aware, and according to Violato *et al.* (2012), there are currently no published figures on the financial cost of non-adherence to the National Health Service (NHS) in relation to CD.

With the increase in serious health complications associated with untreated CD, such as osteoporosis, cancer, psychological disorders and infertility, it is likely that improved adherence would result in reduced healthcare spending. In order to improve adherence, however, better healthcare provision may be needed and this could be expensive. A cost-effective strategy for improving adherence to a GFD is needed in order to reduce NHS spending on CD and improve patient wellbeing and productivity. In developing interventions to improve adherence to treatment, it is important to have a good understanding of the factors affecting adherence and to understand how such factors can be addressed.

1.5 Theoretical models of behaviour change

Upon receiving a CD diagnosis, patients are required to undergo substantial changes in behaviour if they are to adhere to a GFD. The relationship between health behaviour and adherence to treatment has received a great deal of research attention in recent years (DiMatteo, 2004). Many interventions have been designed from behaviour change theory with the aim of improving adherence to medication in chronic conditions, however, less attention has been given to developing interventions to improve dietary adherence. In this section, I describe the six most common psychological theories on the determinants of behaviour change that are applicable for adherence to treatment (Leventhal & Cameron, 1987). It is possible that these models could be used in the development of an intervention to improve long-term adherence to a GFD. The development of an intervention to change behaviour requires knowledge of the concepts (or mediators of change) that need to be targeted (Sirur *et al.*, 2009).

Health belief model (HBM)

The health belief model (HBM), which was developed by Rosenstock, Strecher and Becker, explains behaviour change as the result of a set of beliefs about a situation (Rosenstock *et al.*, 1988). In this model, behaviour change is based on a balance between the barriers to and the benefits of a particular behaviour (e.g. adherence) (Blackwell, 1992). According to this

model, there are four key beliefs that are weighed up in a cost-benefits analysis and this determines behaviour: 1. Perceived susceptibility (e.g. what is the likelihood that poor health outcomes will result from non-adherence?); 2. Perceived severity (e.g. how severe will the consequences of non-adherence be?); 3. Perceived benefit (e.g. adherence will be good for my health); and 4. Perceived barrier (e.g. GFF is expensive, difficult to find and does not taste good). Albert Bandura introduced the concept of self-efficacy, which relates to how competent an individual feels to engage in a particular behaviour (Strecher & Rosenstock, 1997). The concept of self-efficacy was recently added to the HBM. A person with a high sense of self-efficacy is likely to be more motivated to take action than a person with low self-efficacy who would be likely to feel helpless and not in control of a given situation.

According to the HBM, a person with CD must believe that they are susceptible to negative consequences of non-adherence before they will change their behaviour and start adhering to a GFD. However, an individual's beliefs may not be the only reason for non-adherence and factors, such as the presence or absence of symptoms and the availability of GFF may also play a role. The HBM fails to take account of the fact that some behaviour is based on habit, rather conscious decisions.

Theory of reasoned action and planned behaviour

Azjen and Fishbein recognised that attitudes and beliefs do not account for all behaviour (Sutton, 1997). According to this theory, the intention to act is the best predictor of behaviour and the intention to change behaviour is influenced by an individual's attitudes towards the action (Sutton, 1997). The theory of planned behaviour suggests that behaviour is influenced by three factors: 1. Attitudes (beliefs about the likely outcome of behaviour); 2. Subjective norms (perceptions of other people's expectations of them to perform the health behaviour); and 3. Perceived behavioural control (i.e. you have the resources / opportunity to engage in the behaviour). This model suggests that individuals need to believe that they are able to successfully engage in a particular behaviour (self-efficacy) before they will change their behaviour. This builds upon the idea of 'locus of control' theory which suggests that a person either views events as being controlled by their own actions (internal locus of control) or by other people (external locus of control).

Sainsbury & Mullan (2011) used the theory of planned behaviour to predict adherence to a GFD in CD and found this to be a good predictor of adherence. According to the theory of planned behaviour, most behaviour is rational and an individual's intention to behave in a particular way has a greater predictive ability than health beliefs. The theory of planned behaviour, however, does not account for unconscious or irrational behaviour that may result from emotional states or psychological problems.

Despite having an internal locus of control and strong self-efficacy a patient who does not value their health may not adhere to a GFD.

Behavioural learning theory

Behavioural learning theory relates to how people learn from their experiences and the conditioning that can take place during the early years (Lovell, 2011). This model focuses on the environment and teaching the skills and strategies required in managing adherence (World Health Organization (WHO), 2003). Behavioural learning theory explains actions in relation to internal and external antecedents (thoughts and environmental cues) and the consequences of adherence behaviour (punishment or reward). Patients with CD may have unique reasons for non-adherence to a GFD which may require a patient centred approach to care. The lack of an individual approach and the emphasis on immediate reward means that behavioural learning theory may not be appropriate for understanding behaviour or designing interventions in CD.

Social-cognitive theory (SCT)

Social cognitive theory (SCT) was developed by Bandura in 1986 and it is argued that this is the most comprehensive theory of behaviour change (Bandura, 1998; Redding *et al.*, 2000). SCT relates to the choices individuals make and components of this theory include self-efficacy, beliefs and incentives or reinforcement (Chapman-Novakofski & Karduck, 2005). In a study focusing on exercise behaviour, Wallace *et al.* (2000)

applied the SCT to examine the stage of exercise behaviour change in young adults. The study reported that self-efficacy played an essential role in determining exercise behaviour. It is possible, however, that the role of self-efficacy in dietary behaviour may differ to that in exercise behaviour.

Information motivation behaviour (IMB) skills theory

The information, motivation and behavioural (IMB) skills model suggests that there are three components of behaviour change (information, motivation and behaviour skills) (World Health Organization (WHO), 2003). Information and motivation are believed to activate behaviour. Information relates to the knowledge a person has about an illness and its treatment. The IMB model suggests that, although information is a prerequisite for adherence, it is not enough to change behaviour (World Health Organization (WHO) 2003). According to this model behaviour change requires motivation and a focus on developing behavioural skills. Motivation includes the attitudes towards the behaviour, perceived social support for such behaviour and the patient's perception of how other people with the condition might behave (subjective norm). This model acknowledges the importance of having the necessary tools and strategies required for adherence. Self-efficacy is also an important aspect of this model.

Transtheoretical (stages of change) model

The Transtheoretical (Stages of Change (SOC)) Model (TTM) describes an individual's motivational readiness to change (World Health Organization, 2003). This model incorporates some of Bandura's self-efficacy theory. According to this model, behaviour change is thought to progress through a series of five stages:

1. Precontemplation (not considering behaviour change in the next six months) (e.g. I am not seriously thinking about following a GFD).
2. Contemplation (considering changing behaviour in the next six months) (e.g. I think I should follow a GFD).
3. Preparation (planning to change behaviour in the next 30 days) (e.g. I am planning in my diary to make the changes).
4. Action (currently changing behaviour) (e.g. I am buying GFF).
5. Maintenance (successful behaviour change for at least six months) (e.g. I have been sticking to my GFD every day).

The TTM is useful in understanding and predicting intentional behaviour change, however, Bandura argues that behaviour change is multifaceted and cannot be divided into discrete stages.

All the theoretical models of behaviour described above make a number of assumptions. For example, these models assume that people will take an active role and are able to use foresight, plan and make decisions (cognitive

processes). In addition, these models also assume that people will self-regulate their behaviour and behave in goal-orientated ways. Models of health behaviour have been used to develop interventions for a number of health conditions, however, few have been used for dietary interventions. As far as I am aware the TPB is the only model of behaviour change that has been applied in relation to adherence to a GFD in adult CD (Sainsbury & Mullan 2011). It is unclear whether any of the other models could be successfully used to explain adherence to a GFD or in the development of an intervention to improve adherence to a GFD in adult CD.

1.6 Financial incentives to improve adherence

A recent development has been the exploration of how financial incentives, or reward schemes, can be implemented for eliciting behaviour change and adherence to medication (Bremner *et al.*, 2013). This idea has been used successfully in relation to adherence to a number of medical treatments, including medication in patients with mental illness, treatment attendance in substance abusers and in abstinence from smoking (Burton *et al.*, 2010; Carey & Carey 1990; Tidey *et al.* 2002).

As far as I am aware, financial incentives aimed at improving adherence to a GFD in patients with CD have not been investigated. Within the NHS, GFF

is available on prescription, with the intention of reducing the cost of the diet so that it is comparable to a non-GF diet. So, whilst this is not a financial incentive per se, it mitigates the additional cost of this diet. Furthermore, Coeliac UK has suggested that the provision of prescribed GFF does help people with CD to maintain a GFD (Coeliac UK, 2013c).

1.7 Healthcare and support for adults with coeliac disease

The NHS (2013) website states that: '*Coeliac disease is usually treated by simply excluding foods that contain gluten from your diet*'. Although simple in theory, in practice following a GFD is more complicated than this statement suggest. Patients with CD need to be supported in sticking to a life-long GFD (Holmes, 2010; Del-Colle, 2010; Addolorato *et al.*, 2004; Leggio *et al.*, 2005). In this section, I focus on the support available to adults with CD in the UK, including support from the NHS, social support and support from Coeliac UK, the largest charitable organisation for people with CD in the UK. From the time of diagnosis, patients with CD in the UK are expected to self-manage their treatment with little support from healthcare professionals (Berrill *et al.*, 2012). The sudden change to previous dietary habits can be overwhelming and eliminating favourite gluten-containing foods can be challenging (Hogberg *et al.*, 2003).

Following a GFD is complicated and patients are required to acquire a vast amount of knowledge about CD and the GFD as well as coping with the psychosocial impact of living with CD (Ford *et al.*, 2012; Hall *et al.*, 2009; Ciacci *et al.*, 2002b). Patients are required to learn which foods can and cannot be eaten, where GFF can be purchased, which restaurants serve GF meals and how to communicate their dietary needs to people who prepare food for them (Leffler *et al.*, 2008; Hall *et al.*, 2013). Support for people with CD can come from healthcare professionals, family, friends and coeliac support groups. The type and amount of support available to patients with CD is variable.

1.7.1 Current healthcare provision for patients with coeliac disease in the United Kingdom

According to Da Silva Kotze *et al.*, (2009) patients with CD should receive regular evaluation by a healthcare team, including a physician and a dietitian. It is recommended that patients are seen by a dietitian at the time of diagnosis and at least annually for review (British Society of Gastroenterology, 2009; Nelson *et al.*, 2007). However, there is a paucity of dietetic services in the UK and some patients have reported waiting six months before seeing a dietitian (James & Foley, 2011; British Society of Gastroenterology, 2009). The role of the dietitian is to provide expert dietary advice to patients on how to follow a strict GFD (British Society of Gastroenterology, 2009; Coeliac UK, 2013c).

Practitioners should regularly evaluate patients to identify nutrient deficiency and to check that patients are adhering to a strict GFD (Hart *et al.*, 2011). Healthcare for patients with CD in the UK is variable and one study found just 62% of patients with CD were receiving active follow-up (Bebb *et al.*, 2006). Similar findings were made in the USA where only 65% of participants received follow-up that was consistent with the American Gastroenterological Association (AGA) guidelines (Herman *et al.*, 2012). However, both these studies included fairly small sample sizes which limits the reliability of the findings.

Currently there are no clear guidelines on how best to follow-up patients and monitor their adherence to a GFD (Da Silva Kotze *et al.*, 2009). The National Institute for Health and Care Excellence (NICE) provide guidelines for UK healthcare professionals on how best to manage a number of health conditions but they currently only offer guidance on the diagnosis of CD and not long-term management. NICE are in the process of developing a set of guidelines for managing CD which are due to be published in June 2015 (National Institute for Health and Care Excellence (NICE), 2014). The British Society of Gastroenterology (BSG) produced the document '*The Management of Adults with Coeliac Disease*' (Ciclitira *et al.*, 2010). In this document, the BSG recommend that patients receive annual follow-up, however, they acknowledge that many patients do not receive this level of care.

Many patients with CD have expressed dissatisfaction with healthcare in relation to the lack of expertise and continuity of care (British Society of Gastroenterology, 2009). Ukkola *et al.* (2012) reported patient dissatisfaction in relation to doctor-patient communication. Establishing a good doctor-patient relationship is believed to reduce the burden of CD for patients (Ukkola *et al.*, 2012). There is debate about what constitutes the most appropriate method of follow-up for patients with CD (Berrill *et al.*, 2012). Bebb *et al.* (2006) found that patients' preferred follow-up option was to see a dietitian and to have a doctor available and Stuckey *et al.* (2009) reported a preference for dietetic support over support from clinicians. However, patients have different needs and healthcare should be tailored to the individual (Bebb *et al.*, 2006). Patients who are already established on a GFD may prefer to self-manage their condition and only consult with healthcare professionals if they experience problems (Berrill *et al.*, 2012) whereas newly diagnosed patients are likely to need more support.

1.7.2 Prescribed gluten-free food

In the UK, patients with CD are supported by the provision of staple GF products (such as bread, flour, pasta, breakfast cereals and crackers) on prescription from the NHS (Coeliac UK; 2013d; British Specialist Nutrition Association Limited, 2013). GF products that have been approved by the Advisory Committee on Borderline Substances (ACBS) are prescribed to patients by their GP (Coeliac UK, 2013d). GPs have the responsibility of

deciding how much GFF is prescribed to patients based on national guidelines. Table 1.3 shows the monthly recommended amount of prescribed GFF for adults. Different amounts are recommended for different age and gender groups. In England patients are currently required to pay a prescription charge of £7.85 per item or they can purchase a pre-payment certificate for three or 12 months for £29.10 or £104 respectively (Coeliac UK, 2013d). Some patients in England are exempt from this charge and patients living in Scotland, Northern Ireland and Wales do not have to pay a prescription charge for their GFF (Coeliac UK, 2013d).

Table 1.3 Recommended amount of prescribed gluten-free food for adults with coeliac disease in the UK

	Number of units ¹ recommended per month
Males	
18-59 years	18
60-74 years	16
75+ years	14
Females	
18-74 years	14
75+ years	12

Note:

1. 1 Unit = 400g bread/rolls/baguette; 200g biscuits/crackers; 250g pasta; 2 pizza bases. 1.5 Units = 500g oats; 300g breakfast cereal. 2 Units = 500g mix (Agarwal, 2012).

The provision of GFF on prescription began in the 1960s at a time when the availability of substitute GFF was limited (Drug and Therapeutics Bulletin,

2013). The cost to the NHS of providing GFF on prescription was around £27 million in 2011 (Drug and Therapeutics Bulletin, 2013). It is possible that wider availability of GFF in recent years has made the provision of GFF through the NHS unnecessary. However, Coeliac UK (2013d) believe the provision of GFF on prescription is essential. Prescribed GFF is especially important for people who have difficulty obtaining GFF, such as those on a limited income who may find the price of GFF prohibitive, those who live in rural areas where GFF is not easy to find and the elderly who may experience difficulties with accessing GFF (Coeliac UK, 2013b). The provision of GFF on prescription is believed to make it easier for people with CD to follow a GFD (Coeliac UK, 2013d). Any savings made by the NHS through cancelling GF prescriptions may result in higher NHS spending on treating the consequences of non-adherence to a GFD.

1.7.3 Social support

The traditional medical model of treating disease, which is often criticised for being too paternalistic and reductionist (focusing only on the biological aspects of the disease and not the whole person), may not be appropriate for treating chronic conditions such as CD (Del-Colle, 2010). Following a GFD requires permanent and drastic behaviour change that affects an individual's entire social support system. Healthcare provision may be insufficient in helping patients adapt to a new life-long diet and the support provided by family and friends is believed to be integral in managing CD (Herman *et al.*,

2012; Del-Colle, 2010). Leffler *et al.* (2008) found that friends with and without CD were better sources of information about CD for patients compared to healthcare professionals. Socialising in environments where food is provided is greatly affected by CD and support from family and friends can help people to cope better with following a GFD during social situations (Hallert *et al.*, 2002; Zarkadas, 2006).

1.7.4 Coeliac support groups

Coeliac UK is the largest support group for people with CD in the UK and currently has over 60,000 members (Coeliac UK, 2013e). This charitable organisation was established in 1968, at a time when there was less awareness about CD and the GFD (Coeliac UK, 2013e). Coeliac UK is not funded by the government and relies on donations and membership fees, which are currently £20 per year for an individual or carer/parent, £25 for household membership or £10 for people on a low income, such as those receiving benefits or state pensions and students (Coeliac UK, 2013f).

Coeliac UK has a remit of providing members with advice and support, with a strong focus on GFD knowledge and skills. Members are supported with information about the GFD, including being provided with a copy of Coeliac UK's annual Food and Drink Directory (Coeliac UK, 2013a). The Food and Drink Directory, which is updated online monthly, lists thousands of products that are suitable for people with CD along with advice about

choosing the right food and information about the recent changes to GF labelling laws. Access to a helpline, recipe database, local support group and a quarterly publication (Crossed Grain magazine) is also provided to members. There are over 90 local Coeliac UK support groups throughout the UK which fulfil a number of roles, including GF cookery demonstrations, fundraising, hosting food fairs and providing members with the opportunity to meet other people with CD (Coeliac UK, 2013f).

1.8 Aims and objectives

The aim of this study was to gain a better understanding of the factors affecting adherence to a GFD in adults with CD. A further aim was to identify similarities and differences in the opinions of three stakeholder groups (adults with CD, adults who live with them and healthcare professionals). To achieve these aims, I set three main objectives:

1. To conduct a systematic review to identify the factors associated with adherence to a gluten-free diet from previous research and to synthesise the results with the results of a systematic review by Hall *et al.* (2009).
2. To conduct a concept mapping study to explore the factors affecting adherence to a gluten-free diet from the perspectives of three

stakeholder groups (adults with CD; adults who live with them; and healthcare professionals).

3. To develop a model of adherence to a GFD that represents the findings of this study and that can be used in the design of an intervention aimed at improving adherence to a GFD.

1.9 The structure of this thesis

The remainder of this thesis is divided into four chapters. I present the existing evidence on the factors affecting adherence to a GFD in a systematic review in Chapter 2. The method of investigation, concept mapping, is presented in Chapter 3 and I provide a justification for my decision to use this method. The results of this study are presented in Chapter 4 and, in this chapter, I pull together the results of this concept mapping study with the results of the systematic review to provide an up-to-date overview of the factors affecting adherence to a GFD. In chapter 5, I discuss the implications of the findings of this study and I present a model of adherence to a GFD that I have developed from this piece of research.

1.10 Summary

In order to achieve optimal health outcomes for the 1% of the population who have CD, strict adherence to a GFD is required. Despite research into

alternative treatments, the GFD remains the only treatment for CD. This treatment eliminates symptoms and normalises mortality rates for people with CD. The GFD is simple in theory, however, in practice following a GFD is restrictive, costly and it may affect quality of life. Research has shown that as many as 58% of people with CD do not adhere to a strict GFD, however, the reasons for this are unclear (Hall *et al.*, 2009). The aim of this thesis is to gain a better understanding of the factors affecting adherence to a GFD. In Chapter 2, I provide an update to a systematic review into the factors affecting adherence to a GFD in adults with CD by Hall *et al.* (2009).

Chapter 2: A systematic review of factors affecting adherence to a gluten-free diet in adults with coeliac disease

2.1 Introduction

In Chapter 1, I highlighted the importance of adherence to a gluten-free diet (GFD) in eliminating symptoms, improving quality of life (QoL) and normalising mortality rates in people with coeliac disease (CD). A GFD is complicated and as many as 58% of patients with CD do not adhere to this treatment (Hall *et al.*, 2009; Leffler *et al.*, 2008). Studies into the factors affecting adherence to a GFD have produced inconsistent results and a systematic review by Hall *et al.* (2009) (with the search being conducted in 2007) concluded that more rigorous research is needed. Since the systematic review was carried out by Hall *et al.* (2009), a number of new studies relating to adherence to a GFD in adults with CD have been published. In this chapter, I present an update to the systematic review by Hall *et al.* (2009).

The aim of this chapter is to systematically review the evidence of factors that affect adherence to a GFD in adults with CD published between 2007 and August 2013. To develop a broader overall picture of the evidence, I will present the results of my update along with the results of the original systematic review by Hall *et al.* (2009).

2.2 Background and rationale for this systematic review

2.2.1 Background

Adherence to a strict, life-long GFD can be challenging for patients with CD and research into the factors affecting adherence to a GFD is inconclusive (Hall *et al.*, 2009). Poor adherence to a GFD results in an increased risk of morbidity and mortality and sub-optimal adherence can affect a patient's QoL. Knowledge about the factors affecting adherence to a GFD could help to better understand how to provide appropriate care for patients with CD and improve adherence to the GFD.

2.2.2 Rationale for this systematic review

Adherence to a GFD in adults with CD is notoriously poor. It is likely that any attempt to develop a targeted intervention to improve adherence would

prove futile without a good and up-to-date understanding of the factors affecting adherence to a GFD. The evidence presented in the 38 studies included in the systematic review by Hall *et al.* (2009) was often inconsistent and the authors suggested that further research was needed. Since that time, several new studies have been carried out, however, I am not aware of any up-to-date systematic review that has pulled together the results of this new research. Conflicting evidence reported in the Hall *et al.* (2009) systematic review may be resolved through synthesis with the new research in this area.

Since the systematic review was conducted by Hall *et al.* (2009) several factors relating to living with CD and a GFD may have changed. For example, Hall *et al.* (2009) reported the availability of GFF was a problem for people with CD, however, Coeliac UK claims that the retail market for GFF has grown substantially in the last five years (Coeliac UK, 2013g)

The aim of this systematic review was to identify peer reviewed journal articles that provide evidence on the factors affecting adherence to a GFD in adults with CD. In this chapter, I report the results of my systematic review and I synthesise the evidence with the findings from the study by Hall *et al.* (2009).

2.3 Methods

This systematic review is an update to the review by Hall *et al.* (2009) and the method employed here is based on that used by Hall *et al.* (2009) with some adaptations as detailed in the sections below.

2.3.1 The systematic search

Relevant experimental and non-experimental studies were identified using a method based on that employed by Hall *et al.* (2009). In order to capture as many relevant citations as possible, I conducted an extensive search of seven medical and scientific databases: AMED (Ovid, 1985); CINAHL (EbscoH, 1982); Cochrane Library; Embase (Ovid, 1974); Medline (Ovid, 1946), Psycinfo (1806); and PubMed. The previous review by Hall *et al.* (2009) collected information up to and including 2007, therefore, I searched the databases for relevant studies published after 2007 up to and August 2013.

After searching the up-to-date literature for relevant key words, I slightly adapted the search string used by Hall *et al.* (2009) to create a comprehensive set of search terms. To ensure I identified as many relevant papers as possible, the following search string was used: (celiac or coeliac or gluten sensitiv\$ or gluten-sensitiv\$ enteropathy) and (adher\$ or comply

or complian\$ or concordan\$ or manag\$ or non-adher\$ or non-complian\$) and (gluten or gluten-free or diet\$ or treat\$ or therap\$). The search string was adapted slightly to allow for differences between the required searching criteria for each database. Where the databases provided the facility to use MeSH terms, the search strategy included the MeSH terms 'celiac disease' and 'Diet, gluten-free' and all their derivations. The full search string used for each of the seven databases is shown in Appendix 1.

To avoid duplication, all 2007 articles that were already included in Hall *et al.*'s (2009) systematic review were excluded. The reference lists from included papers were examined and any eligible articles found in this way were included in the review. The search was limited to human studies, English language studies and full papers where the databases allowed. I did not have the facilities to translate papers from non-English languages.

2.3.2 The identification and selection of relevant papers

Records were imported to Mendeley referencing manager (Mendeley, 2013) and all duplicated papers were removed. A review of the title and abstract was carried out and any studies that were not relevant were removed. I obtained and read the full text articles for the remaining studies and excluded those that did not fit my inclusion criteria (Table 2.1). Only peer

reviewed published articles were included. All types of study design were eligible for inclusion in this review.

Table 2.1 – Systematic review inclusion and exclusion criteria

Inclusion Criteria	Exclusion criteria
English language	Childhood studies or studies involving adults and children where data is combined
Human studies	If coeliac disease is not the primary disease being studied
Adult studies or studies including children where adult results are presented separately	Non-English language
Primary research	Animal studies
Studies published between 2007-August 2013	Poster abstracts and dissertations
Full paper articles only	Already included in 2007 systematic review by Hall <i>et al.</i> (2009).
Qualitative, quantitative and mixed method studies with be included	
Evidence of at least one factor affecting adherence to a gluten-free diet in coeliac disease	

2.3.3 Data extraction and quality assessment

Having identified the studies for inclusion in this review, the next step was to extract the relevant data and assess the quality of each of the included studies. To do this, it was necessary to establish the most appropriate data extraction and quality appraisal tools. The Cochrane Handbook for systematic reviews (Higgins & Green, 2011) advises that quality assessment should check for validity and sources of bias. I judged the tools used in a systematic review in adherence to medication in Parkinson’s disease by Daley *et al.*, 2012) to be the most suitable for assessing the quality of studies in this systematic review. Hall *et al.* (2009) used appraisal tools

developed by Crombie (1996) and Poppay *et al.* (1998). I chose to use the recently developed tools by Daley *et al.* (2012) because they focused more on the quality of methodological rigor and the risk of bias.

Data extraction

The full text of the included articles was reviewed and data were extracted using a slightly adapted version of the pre-determined form taken from Daley *et al.* (2012). The use of a pre-determined form can reduce the likelihood of bias and it helps to ensure the data extraction process is systematic. I adapted the data extraction sheet slightly by including the percentage of participants who were adherent/non-adherent and by including both the factors that affect adherence in both a negative and a positive way (rather than the factors affecting non-adherence only). I also decided to include the country where the study was conducted as this could be used to explain any conflicts in the evidence (for example, the availability of GFF may differ between countries).

The extracted data were tabulated and the emerging adherence factors were grouped together according to the six themes identified in the systematic review by Hall *et al.* (2009):

1. Sociodemographic factors
2. Knowledge, attitudes and beliefs
3. Illness and symptom factors
4. Treatment factors

5. Socio-cultural/Environmental factors
6. Quality of life and psychological well-being

Quality assessment for risk of bias/internal validity

The purpose of the quality assessment is to check for bias and internal validity. Systematic error and bias are terms that are interchangeable with the term ‘internal validity’. Internal validity is the extent to which the design and conduct of a research study are likely to affect the reliability of the results (Higgins & Green, 2011).

Daley *et al.* (2012) developed a quality appraisal tool to assess the risk of bias in the observational studies included in their systematic review. The assessment tool included an overall summation of the risk of bias for each study (Daley *et al.*, 2012). Although summary scores are not recommended by some reviewers (Higgins & Green, 2011) because the influence (weighting) of each quality item is not equal, I used this because it helped with descriptive clarity in appraising which study’s results were trustworthy or not. The quality checking form used by Daley *et al.* (2012) was adapted slightly for this study. For example, Daley *et al.* (2012) specified their diagnostic criterion for Parkinson’s disease, and I specified that CD should be biopsy confirmed.

The studies included in my review were assessed against five potential sources of bias using this adapted version of the quality checking criteria developed by Daley *et al.* (2012):

1) Selection bias

Diagnostic inaccuracy

Diagnosis of CD using an intestinal biopsy is considered to be the gold standard (Leeds *et al.*, 2008). I believe the accuracy of CD diagnosis is an important factor in this review because patients who have been incorrectly diagnosed with CD may behave differently in relation to following a GFD. Papers that included CD patients who were diagnosed by internal biopsy were judged to have a low risk of selection bias in relation to diagnostic accuracy. Where any other method of diagnosis was used the risk of bias in relation to diagnostic inaccuracy was deemed to be high.

Participant representativeness

Studies that included a population that is representative of the wider population of people with CD was regarded as low risk in relation to participant representativeness. Where participants were recruited from coeliac support groups (which are known to have members that are not representative of the wider population (Butterworth *et al.*,

2004)), or other non-representative groups, the risk of selection bias was judged to be high.

Sampling methods

Studies were judged on whether or not they employed an appropriate sampling method to discount selection bias, such as random sampling. Both the source and the method of sampling were considered in this assessment. Samples that were not likely to be representative of the wider population were given a lower rating than samples that provided the opportunity for a more representative sample to be selected.

2) Random variation/chance

Sample size calculation

Studies that reported a sample size calculation and the target population was reached were judged to be low risk with regards to random variation/chance.

3) Detection bias

Validity of adherence measures

In assessing the validity of adherence measures I considered the methods of measuring adherence to a GFD. Serological testing and

intestinal biopsy were considered to be valid measures, whereas self-reporting, interview or assessment by a healthcare professional (based on the patient's self-reporting) were considered not to be valid measures of adherence.

Follow-up

Was follow-up the same for cases and controls (where applicable)?

Were appropriate measures taken at follow-up?

4) Attrition bias

Loss to follow up

Participants who are lost to follow-up (where applicable) can lead to bias in the results and this can compromise the validity of the results. Participants who drop out of a study may differ in some way to those who continue to participate. Studies that showed a high loss to follow-up were rated as having a high risk of attrition bias.

5) Reporting bias

Appropriateness of analysis

Reporting bias was judged based upon whether the authors used an appropriate method of analysing the data. I also considered whether significance was likely to be a result of chance and whether missing data was dealt with appropriately.

2.3.4 Data analysis

Once data extraction was completed, I grouped the data in order of the six themes identified in the systematic review by Hall *et al.* (2009). I also included the results from the Hall *et al.* (2009) systematic review in my results. Merging the results of my updated systematic review with the results of the original systematic review by Hall *et al.* (2009), allowed me to generate an overall summary of the evidence. This summary is presented in the following section.

2.4 Results

2.4.1 Search results

The literature search, using seven databases, resulted in the identification of 1812 records. Seven hundred and twenty-two duplicated papers were removed. A hand search of relevant reference lists yielded a further 17 papers, giving a total of 1107.

Table 2.2 Results of the systematic review search of seven databases

DATABASE	NUMBER OF RECORDS
AMED	1
CINAHL	221
Cochrane Library	45
Embase	1011
Medline OVID	488
PsychInfo	7
PubMed	39
Hand search	17

2.4.2 The selection of papers for inclusion

The potential relevance of all 1107 citations was reviewed. Figure 2.1 is a PRISMA flow diagram showing the steps taken in the selection of papers for inclusion in this review. One thousand and sixteen papers were excluded as irrelevant from the title and abstract. The full papers of the remaining 91 citations were assessed to select relevant primary studies that matched the inclusion criteria. These criteria excluded 70 studies and left 21 in the review. Appendix 2 shows a list of the 70 excluded papers with the reasons for exclusion.

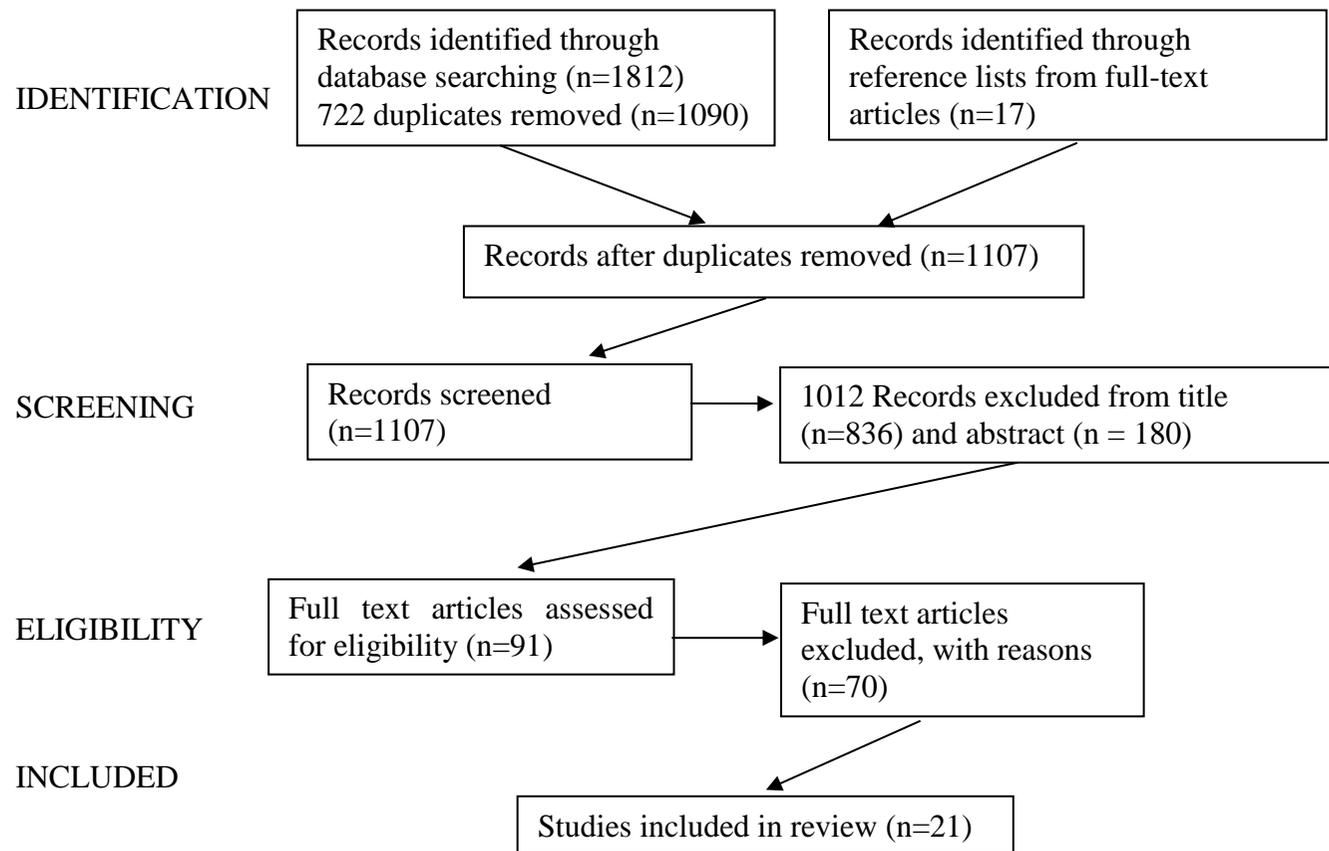


Figure 2.1 PRISMA flow diagram of the selection process of studies for inclusion in the systematic review

2.4.3 Summary of the studies included in this systematic review

I identified 21 studies for inclusion in this systematic review. The characteristics of the included papers are detailed in Appendix 3. Studies that provide figures on gender all show fewer male participants than female. Adherence ranged from 44.2% to 100%. The included studies came from nine countries (Table 2.3) and included a total of 12,222 participants.

Table 2.3 Country where the research was carried out for the 21 studies included in the systematic review

Country of study	Number of included articles
UK studies	6
USA	5
Australia	2
Finland	2
Italy	2
Argentina	1
Canada	1
Netherlands	1
Sweden	1

Table 2.4 shows the risk of bias for each of the 21 studies included in this systematic review. A low risk of bias is represented with a ‘✓’ and a high risk of bias is represented with an ‘X’. Where ‘n/a’ is shown, this measure was not relevant to that particular study’s methodology. Where ‘?’ is shown, this measure of bias could not be assessed because the author did not provide sufficient information for a judgement to be made.

I calculated the overall risk of bias and this was used to represent the quality of each study. The overall risk of bias was calculated by dividing the number of ✓ by the total number of risk of bias items (X and ?) and multiplying by 100 to produce a percentile for each study ($\checkmark / (X + ?) \times 100 = \%$). Where a source of bias was not applicable (N/A) this was not included in the calculation. Studies scoring >70% were judged to be of high quality; 40-69% were moderate quality; and <40% was low quality.

Table 2.4 shows that seven of the 21 studies were of high quality (low risk of bias); ten had a moderate risk of bias (moderate quality); and four studies were deemed to be of low quality because they had a high risk of bias. Five of the 21 studies used a measure of adherence that was deemed to be valid. Five studies employed a sample size calculation and 12 of the 21 studies recruited participants from a population that was judged to be representative of the wider population of adults with CD. The majority of studies (n=19) included coeliac patients who had been diagnosed with CD by intestinal biopsy. All papers included in this review were judged to have used appropriate methods of analysing the results.

Table 2.4 Quality assessment of the 21 included studies

Study	Risk of Bias/threat to validity									
	Diagnostic inaccuracy	Participant representativeness	Sampling methods	Sample size calculation	Validity of Adherence measures	Follow-up	Loss to follow-up	Appropriateness of analysis	Overall risk of bias	Overall quality
Barratt <i>et al.</i> , 2011	?	✓	✓	?	X	n/a	n/a	✓	$(3 \div 6) \times 100 = 50$	Moderate
Black & Orfila, 2011	✓	X	✓	?	X	n/a	n/a	✓	$(3 \div 6) \times 100 = 50$	Moderate
Casella <i>et al.</i> , 2012	✓	✓	✓	?	✓	✓	X	✓	$(6 \div 8) \times 100 = 75$	High
Edwards-George <i>et al.</i> 2009	✓	✓	✓	?	✓	✓	✓	✓	$(7 \div 8 \times 100 = 87.5)$	High
Errichello <i>et al.</i> 2010	✓	✓	✓	?	X	n/a	n/a	✓	$(4 \div 6) \times 100 = 67$	Moderate
Ford <i>et al.</i> 2012	✓	X	✓	✓	X	n/a	n/a	✓	$(4 \div 6) \times 100 = 67$	Moderate
Hall <i>et al.</i> , 2013	✓	✓	✓	?	X	n/a	n/a	✓	$(4 \div 6) \times 100 = 67$	Moderate
Hopman <i>et al.</i> , 2009	✓	✓	✓	?	X	n/a	n/a	✓	$(4 \div 6) \times 100 = 67$	Moderate
Hutchinson <i>et al.</i> , 2010	✓	✓	✓	✓	X	✓	✓	✓	$(7 \div 8) \times 100 = 87.5$	High
Kabbani <i>et al.</i> , 2012	✓	✓	✓	?	X	✓	✓	✓	$(6 \div 8) \times 100 = 75$	High
Kurppa <i>et al.</i> , 2012	✓	✓	✓	✓	✓	n/a	n/a	✓	$(6 \div 6) \times 100 = 100$	High
Lee <i>et al.</i> , 2012	✓	X	X	?	X	n/a	n/a	✓	$(2 \div 6) \times 100 = 33$	Low
Mahadev <i>et al.</i> , 2013	✓	X	X	?	X	n/a	n/a	✓	$(2 \div 6) \times 100 = 33$	Low
Nachman <i>et al.</i> , 2010	✓	✓	✓	?	✓	✓	X	✓	$(6 \div 8 \times 100 = 75)$	High
Paavola <i>et al.</i> , 2012	✓	✓	✓	?	✓	✓	✓	✓	$(7 \div 8) \times 100 = 87.5$	High
Sainsbury & Mullan 2011	✓	X	X	?	X	n/a	n/a	✓	$(2 \div 6) \times 100 = 33$	Low
Sainsbury <i>et al.</i> , 2013a	✓	X	X	✓	X	n/a	n/a	✓	$(3 \div 6) \times 100 = 50$	Moderate
Sey <i>et al.</i> , 2011	✓	X	X	?	X	✓	✓	✓	$(4 \div 8) \times 100 = 50$	Moderate
Smith and Goodfellow 2011	✓	X	X	✓	?	n/a	X	✓	$(3 \div 7) \times 100 = 43$	Moderate
Sverker <i>et al.</i> , 2009	✓	✓	✓	?	?	n/a	n/a	✓	$(4 \div 6) \times 100 = 67$	Moderate
Van Hees <i>et al.</i> , 2013	X	X	X	?	X	n/a	n/a	✓	$(1 \div 6) \times 100 = 17$	Low

Note:
X = high risk of bias; ✓= low risk of bias; ?=the author did not provide enough information for a judgement of the risk of bias to be made and, therefore, this was considered to represent a high risk of bias.

2.4.4 Factors affecting adherence to a gluten-free diet

This update to the systematic review by Hall *et al.* (2009) has identified a number of new factors associated with adherence to a GFD. The studies included in this update varied in the number of adherence factors they identified. Appendix 3 shows a list of the factors with details of the studies that identified each adherence factor. Hall *et al.* (2009) grouped the factors associated with adherence to a GFD identified in their study into six themes. In the following six sections, I present the results of my update of Hall *et al.* (2009) systematic review using the same six themes. I have also included the results of the systematic review by Hall *et al.* (2009) to provide an overall summary of the evidence.

Tables 2.5 to 2.10 show the results of my analysis along with the results of the quality assessment for each study included in this review. I have reported the quality of the papers used in Hall *et al.*'s (2009) systematic review as moderate/high which reflects the decision by the authors to exclude papers that were judged to be of poor quality from their review. I have included papers from my update to Hall *et al.*'s (2009) systematic review that I judged to be of low quality, however, I have taken into account the limitations of these studies when interpreting the results.

Sociodemographic factors

Table 2.5 shows the results of the analysis for studies that report on sociodemographic factors in relation to adherence to a GFD. All of the studies that assessed sociodemographic factors were judged to be of moderate or high quality except for one low quality study (Lee *et al* 2012). The results of this study were in line with all of the other new studies results which report that gender is not associated with adherence to a GFD, so it was judged that the result was probably trustworthy.

Hall *et al.* (2009) identified two studies that reported education as a factor that was associated with adherence to a GFD, but five where it was not. I identified a further two studies, one reported better adherence in people with a university education (Barratt *et al.* (2011) but the other study (Hall *et al.*, 2013) found no association. Therefore, education does not have a consistent relationship with adherence to a GFD.

Age was identified by Hall *et al.* (2009) as a factor that was associated with adherence in three studies, and not associated in a further seven studies. I identified three more studies which found an association with age (n=1124), and four that did not (n=3437). The three studies that found an association between age and adherence to a GFD, all reported that older age was associated with better adherence. Overall, age does not appear to have a consistent relationship with adherence.

Hall *et al.* (2009) identified just one study that associated gender with adherence and seven studies that did not. I identified a further five studies (n= 4745) that showed no association of gender with adherence. Overall, the evidence suggests that gender is not associated with adherence to a GFD.

No association was found between social class and adherence to a GFD in two studies included in the systematic review by Hall *et al.* (2009). I identified one more study that showed an association with having an affluent background. Social class does not appear to have a consistent relationship with adherence.

Hall *et al.* (2009) identified one study (n=234) that showed an association with urban living with adherence. I identified no further studies that examined this factor. Overall, there is weak evidence for an association of urban living with adherence.

Hall *et al.* (2009) identified two studies that showed no association of employment status with adherence, and I did not identify any additional studies. Therefore overall there is evidence that employment status is not associated with adherence.

One study (n=154) identified in the systematic review by Hall *et al.* (2009) showed marital status was associated with adherence. I identified one more study (n=255) that showed no association between marital status and

adherence. Therefore, marital status does not have a consistent relationship with adherence to a GFD.

Hall *et al.* (2009) identified one study (n=87) that showed ethnicity was associated with adherence, but I identified one larger study (n=679) that showed no such association. Therefore overall it is unlikely that ethnicity is associated with adherence to a GFD.

Finally I identified one study (n=204) that showed smoking status was associated with adherence. This single study provides weak evidence that non-smokers have better adherence to a GFD. Overall there is little evidence that any of the sociodemographic factors associate with adherence behaviours.

Table 2.5 Sociodemographic factors (including results from Hall *et al.*'s (2009) systematic review)

Authors	Adherence factor	Number of participants in studies that show an association with adherence	Number of participants in studies that did not show an association with adherence	Assessment of quality
Hall <i>et al.</i> , 2009	Education	2/7 ¹ (n=971)	5/7 ¹ (n=566)	Moderate/High
Barratt <i>et al.</i> , 2011	Education (university education is associated with better adherence)	n = 573		Moderate
Hall <i>et al.</i> , 2013	Education		n=287	Moderate
Hall <i>et al.</i> , 2009	Age	3/10 ¹ (n=673)	7/10 ¹ (n =1132)	Moderate/High
Barratt <i>et al.</i> , 2011	Age		n=573	Moderate
Casellas <i>et al.</i> 2012	Age		n=1898	High
Ford, 2012	Age (older age is associated with better adherence)	n=228		Moderate
Hall <i>et al.</i> , 2013	Age		n=287	Moderate
Hopman, 2009	Age (older age is associated with better adherence)	n=53		Moderate
Kabbani <i>et al.</i> , 2012	Age		n=679	High
Kurppa, 2012	Age (older age is associated with better adherence)	n=843		High
Hall <i>et al.</i> , 2009	Gender	1/7 ¹ (n=128)	6/7 ¹ (n=1806)	Moderate/High
Barratt, 2011	Gender		n=573	Moderate
Hall, 2013	Gender		n=287	Moderate
Hutchinson, 2010	Gender		n=284	High
Kabbani <i>et al.</i> , 2012	Gender		n=679	High
Lee <i>et al.</i> , 2012	Gender		n=2922)	Low
Hall <i>et al.</i> , 2009	Social Class/socioeconomic status		2/2 ¹ (n=282)	Moderate/High
Barratt <i>et al.</i> , 2011	An affluent background/wealthy achievers show better adherence	n=573		Moderate
Hall <i>et al.</i> , 2009	Urban residence	1/1 ¹ (n=234)		Moderate/High
Hall <i>et al.</i> , 2009	Employment status / Occupation		2/2 ¹ (n=544)	Moderate/High
Hall <i>et al.</i> , 2009	Marital status	1/1 ¹ n=154		Moderate/High
Barratt <i>et al.</i> , 2011	Marital status		n=225	Moderate
Hall <i>et al.</i> , 2009	Ethnicity	1/1 ¹ (n=87)		Moderate/High
Kabbani, 2012	Ethnicity		n=679	High
Errichello, 2010	Non-smokers had better adherence	n=204		Moderate

Note:

1. The figures shown here indicate the number of studies included in the systematic review by Hall *et al.* (2009) in relation to a particular adherence factor. For example, for 'Education' two out of seven (2/7) studies from the Hall *et al.* (2009) systematic review showed an association between education and adherence to a GFD and five out of seven (5/7) studies showed no association between adherence and education.

Knowledge, attitudes and beliefs

Table 2.6 shows the results of the analysis for studies that report on knowledge, attitudes and beliefs in relation to adherence to a GFD. All of the factors relating to knowledge, attitudes and belief were identified in just five out of the 21 studies included in this systematic review update. There may be issues of the same or similar factors being labelled differently by different authors. I have grouped factors together in a way that seemed logical to me.

Hall *et al.* (2009) and I both identified one study each that identified knowledge and understanding of a GFD as being associated with better adherence. One new study showed that improved awareness and understanding were believed to make adherence to a GFD easier. Therefore, it appears that knowledge and understanding is associated with better adherence to a GFD.

Hall *et al.* (2009) identified two studies that investigated the association between reading food labels and adherence to a GFD. One of these studies was associated with adherence to a GFD and the other was not. In my update, I found one further study (N=278, low quality) that found reading food labels to be associated with better adherence to a GFD.

Hall *et al.* (2009) found a study showing an association between adherence to a GFD and beliefs about the harm of exposure to gluten. Two new studies

showed evidence of association between worry about the long term impact of the disease with adherence and another new study showed no association. One new study showed an association between having a low belief in the cyclical nature of CD and better adherence to a GFD. Overall, it appears that beliefs about the seriousness of the consequences of non-adherence are associated with adherence to a GFD.

A high quality study by Edwards-George (2009) (N=154) examined personality traits and found that higher conscientiousness, values, order, self-discipline and deliberation traits were all associated with better adherence. Another study by Sainsbury (2013a), which was larger (n=390) but of moderate quality, identified that intention to adhere, task orientated coping and emotion orientated coping were all associated with improved adherence to a GFD. Overall, it appears that personality trait, intention and coping style are associated with adherence to a GFD.

Two new studies showed that self-efficacy was associated with better adherence. This may also be related to one study that showed that being prepared and organised was associated with better adherence. Additionally, a low quality study (N=278) (Sainsbury *et al.*, 2011) showed that having the confidence to ask about contamination is associated with improved adherence. Trusting other people to prepare their food was associated with poorer adherence.

Overall knowledge, attitudes and beliefs are usually associated with adherence. However, reading food labels is unlikely to be associated with adherence.

Table 2.6 Knowledge, attitudes and beliefs (including results from Hall *et al.*'s (2009) systematic review)

Authors	Adherence factor	Number of participants in studies that show an association with adherence	Number of participants in studies that did not show an association with adherence	Assessment of quality
Hall <i>et al.</i> , 2009	Understanding of gluten-free food	1/1 ¹ (n=154)		Moderate/High
Sainsbury <i>et al.</i> , 2011	Knowledge of gluten-free ingredients was associated with better adherence	n=278		Low
Hall <i>et al.</i> , 2013	Improved awareness and understanding was linked with better adherence	n=287		Moderate
Hall <i>et al.</i> , 2009	Understanding and use of food labelling	1/2 ¹ (n=87)	1/2 ¹ (n=234)	Moderate/High
Sainsbury <i>et al.</i> , 2011	Reading food labels was associated with better adherence	n=278		Low
Hall <i>et al.</i> , 2009	Beliefs about harm from exposure to gluten	1/1 ¹ (n=154)		Moderate/High
Ford, 2012	Belief in the serious consequences of non-adherence was associated with better adherence	n=288		Moderate
Hall <i>et al.</i> , 2013	Worry about the long-term impact of gluten consumption was associated with better adherence	n=287		Moderate
Sainsbury <i>et al.</i> , 2011	Worry about the long-term impact of gluten consumption		n=278	Low
Ford <i>et al.</i> , 2012	Weaker belief in the cyclical nature of coeliac disease was associated with better adherence	n=288		Moderate
Edwards-George, 2009	Higher conscientiousness was associated with better adherence	n=154		High
Edwards-George, 2009	Higher values trait was associated with better adherence	n=154		High
Edwards-George, 2009	Higher order trait was associated with better adherence	n=154		High
Edwards-George, 2009	Higher self-discipline trait was associated with better adherence	n=154		High
Edwards-George, 2009	Higher deliberation trait was associated with better adherence	n=154		High
Sainsbury <i>et al.</i> , 2013a	Intention to adhere was associated with better adherence	n=390		Moderate
Sainsbury <i>et al.</i> , 2013a	Task oriented coping (problem solving) was associated with better adherence	n=390		Moderate

Sainsbury <i>et al.</i> , 2013a	Emotion oriented coping (feeling anxious or blaming oneself) was associated with better adherence	n=390	Moderate
Ford, 2012	Higher self-efficacy was associated with better adherence	n=288	Moderate
Hall <i>et al.</i> , 2013	Higher self-efficacy was associated with better adherence	n=287	Moderate
Sainsbury <i>et al.</i> , 2011	Being prepared and organised was associated with better adherence	n=278	Low
Sainsbury <i>et al.</i> , 2011	Confidence to ask questions about contamination was associated with better adherence	n=278	Low
Sainsbury <i>et al.</i> , 2011	Having trust in others to prepare GFF was associated with poorer adherence	n=278	Low

Note:

1. Figures shown here indicate the number of studies included in the systematic review by Hall *et al.* (2009) in relation to a particular adherence factor. For example, for ‘Understanding of gluten-free food’ one study out of one (1/1) from the Hall *et al.* (2009) systematic review showed an association between understanding of gluten-free food and adherence to a gluten-free diet

Illness and symptom factors

Table 2.7 shows the results of the analysis for studies that report on illness and symptom factors in relation to adherence to a GFD.

I identified two new studies examining time since CD diagnosis in relation to adherence, however, neither of these studies found an association with adherence to a GFD. Hall *et al.* (2009) found one small study (N=76) (out of three) that reported an association between adherence to a GFD and time since CD diagnosis. Overall, it is likely that time since CD diagnosis is not associated with adherence.

Hall *et al.* (2009) reported that most studies in their systematic review found no association between age at diagnosis and adherence to a GFD. I

identified three new studies with more disparate results. Two of these studies reported that diagnosis at an older age was associated with better adherence to a GFD. There are now three studies that found an association, and six that found no association between age at diagnosis and adherence. So overall there is no strong evidence that age at diagnosis is associated with adherence to a GFD.

Two studies in the review by Hall *et al.* (2009) found the presence of symptoms at diagnosis to be associated with adherence to a GFD and one study that did not find an association. I found three new studies relating to symptoms at diagnosis which showed no association with adherence to a GFD. Overall, symptoms at diagnosis is now a factor with inconsistent evidence to support it.

Diagnostic delay was reported to be associated with adherence in one study (n=300) that was included in the review by Hall *et al.* (2009). I did not identify any further studies for this factor.

Hall *et al.* (2009) identified inconsistent evidence regarding whether having CD symptoms was associated with adherence. I identified three further studies that were also inconsistent in their results. One of these three studies showed no association with the presence of symptoms at diagnosis (n=278). One study reported poorer adherence in individuals who had experienced symptoms in the past four weeks (n=154) and one study showed better

adherence in individuals who experience severe symptoms if gluten is consumed (n=390). In total four studies identified an association between CD symptoms and adherence to a GFD and five studies did not find an association, therefore, the evidence is inconclusive.

The presence of additional food intolerances was found to be associated with better adherence in one study in Hall *et al.*'s (2009) systematic review. I identified one further study that also found this association (Edwards-George *et al.*, 2009).

I identified one large (n=1018) high quality study that reported an association between body weight and adherence to a GFD. This study showed that having a normal body weight was associated with better adherence, whilst being overweight was associated with non-adherence.

Overall, the level of symptoms, whether they led to a diagnosis, the time the person has had a diagnosis for CD, and at what age they received the diagnosis were not associated with adherence. The association between diagnostic delay and adherence to a GFD was evident in one study in the systematic review by Hall *et al.* (2009). There are two small studies that suggest that having additional food intolerances may be associated with adherence. One study that suggests that a patient's body weight is associated with adherence.

Table 2.7 Illness and symptom factors (including results from Hall *et al.*'s (2009) systematic review)

Authors	Adherence factor	Number of participants in studies that show an association with adherence	Number of participants in studies that did not show an association with adherence	Assessment of quality
Hall <i>et al.</i> , 2009	Time since diagnosis	1/3 ¹ (n=76) ²	2/3 ¹ (n=230) ²	Moderate/High
Barratt <i>et al.</i> , 2011	Time since diagnosis		n=573	Moderate
Hall <i>et al.</i> , 2013	Time since diagnosis		n=287	Moderate
Hall <i>et al.</i> , 2009	Age at diagnosis	1/6 ¹ (n=29) ²	5/6 ¹ (n=1284) ²	Moderate/High
Hall <i>et al.</i> , 2013	Age at diagnosis (those diagnosed as adults had better adherence)	n=287		Moderate
Hopman, 2009	Age at diagnosis (those diagnosed at an older age had better adherence)	n=53		Moderate
Hutchinson, 2010	Age at diagnosis		n=284	High
Hall <i>et al.</i> , 2009	Presence of symptoms at diagnosis	2/3 ¹ (n=454) ²	1/3 (n=154) ²	Moderate/High
Barratt <i>et al.</i> , 2011	Presence of symptoms at diagnosis		n=573	Moderate
Paavola, 2012	Screen detected/ symptom detected		n=576	High
Kabbani, 2012	Type of symptoms present at diagnosis (gastrointestinal or extra-gastrointestinal)		n=679	High
Hall <i>et al.</i> , 2009	Diagnostic delay	1/1 ¹ (n=300) ²		Moderate/High
Hall <i>et al.</i> (2009)	Coeliac disease symptoms experienced	2/6 ¹ (n=590) ²	4/6 ¹ (n=642) ²	Moderate/High
Edwards-George (2009)	Symptoms experienced in the 4 weeks prior to the study was associated with poorer adherence	n=154		High
Sainsbury <i>et al.</i> , 2011	Coeliac disease symptoms experienced		n=278	Low
Sainsbury <i>et al.</i> , 2013a	Higher severity of symptoms was associated with better adherence	n=390		Moderate
Hall <i>et al.</i> , 2009	Presence of additional food intolerances was associated with better adherence	1/1 ¹ (n=154) ²		Moderate/High
Edwards George <i>et al.</i> , 2009	Presence of additional food intolerances was associated with better adherence	n=154		High
Kabbani, 2012	Body weight (obesity higher in non-adherent group; normal BMI is higher in adherent group)	n=1018		High

Note:

1. The figures shown here indicate the number of studies included in the systematic review by Hall *et al.* (2009) in relation to a particular adherence factor. For example, for 'Time since diagnosis' one out of three (1/3) studies from the Hall *et al.* (2009) systematic review showed an association between time since diagnosis and adherence to a gluten-free diet and two out of three (2/3) showed no association between adherence and time since diagnosis.

Healthcare treatment factors

Table 2.8 shows the results of the analysis for studies that report on healthcare treatment factors in relation to adherence to a GFD. Hall *et al.* (2009) identified five studies that showed disparate results regarding the duration of following a GFD with adherence. Although there were four studies showing no association, the total number of participants in these studies (n=385) was not substantially larger than the one study that showed an association (n=200). I identified no further studies for this factor.

I identified two new studies that showed that people who believe that following a GFD is difficult was associated with adherence, however, the results of these two studies were conflicting. One study (n=2922) found poorer adherence in those who believed the GFD was difficult to follow, whereas the other study (n=278) found poorer adherence in individuals who believe it is not difficult to eat a nutritionally balanced GFD. Hall *et al.* (2009) found one small (n=73) study that showed no association between adherence and perception of difficulty in following the GFD. Although my two new studies were large (total N= 3200) their quality was low and the conflicting findings lead me to conclude that there appears to be no association between perceived difficulty in following a GFD being associated with adherence.

Receiving GFF on prescription was a new factor that one moderate quality study by Hall *et al.* (2013) identified as being associated with better adherence to a GFD (N=287).

In their systematic review, Hall *et al.* (2009) found inconsistent support for satisfaction with information from a healthcare provider being associated with adherence. I found no new studies in relation to this factor.

I identified one new study that showed no association between attendance at a coeliac clinic and adherence to a GFD (n=413). This finding is in contrast to Hall *et al.* (2009) who found one study that showed an association between attendance at a coeliac clinic and adherence to a GFD (n=99). Overall there is inconsistent support to show that this factor is associated with adherence to a GFD.

The regularity of follow-up was shown by Hall *et al.* (2009) to have inconsistent support for its association with adherence. I identified one more study that showed an association with adherence, and overall the balance of evidence does appear to be tending towards there being an association. In total, three studies (total N=764) reported an association between adherence and the regularity of follow-up and one study (N=207) did not find an association.

I identified one more study that showed that membership of a coeliac support group is associated with better adherence, which is in concordance with the two studies reported in the systematic review by Hall *et al.* (2009).

Overall I have identified inconsistent support for duration of GFD, difficulty of GFD, satisfaction with information from healthcare providers, attendance at coeliac clinics and regularity of follow-up. There is limited evidence that provision of GFF on prescription may enhance adherence. There is increasingly strong evidence that membership of a coeliac support group is associated with better adherence to a GFD.

Table 2.8 Healthcare Treatment factors (including results from Hall *et al.*'s (2009) systematic review)

Authors	Adherence factor	Number of participants in studies that show an association with adherence	Number of participants in studies that did not show an association with adherence	Assessment of quality
Hall <i>et al.</i> , 2009	Duration of gluten-free diet	1/5 ¹ (n=200)	4/5 ¹ (n=385)	Moderate/High
Hall <i>et al.</i> , 2009	Difficulty of gluten-free diet		1/1 ¹ (n=73)	Moderate/High
Lee, 2012	Finding the gluten-free diet difficult to follow was associated with poorer adherence	n=2922		Low
Sainsbury <i>et al.</i> , 2011	Belief that eating a nutritionally balanced gluten-free diet is not difficult was associated with poorer adherence	n=278		Low
Hall, 2013	Receiving gluten-free food on prescription was associated with better adherence	n=287		Moderate
Hall <i>et al.</i> , 2009	Satisfaction with information from health care provider	2/3 ¹ (n=321)	1/3 ¹ (n=154)	Moderate/High
Hall <i>et al.</i> , 2009	Attendance at coeliac clinic	1/1 ¹ (n=99)		Moderate/High
Mahadev, 2013	Attendance at coeliac clinic (use of dietician)		n=413	Low
Hall <i>et al.</i> , 2009	Regularity of follow-up	2/3 ¹ (n=477)	1/3 ¹ (n=207)	Moderate/High
Hall, 2013	Receiving regular follow-up was associated with better adherence	n=287		Moderate
Hall <i>et al.</i> , 2009	Coeliac support group membership	2/2 ¹ (n=241)		Moderate/High
Hall, 2013	Coeliac support group membership was associated with better adherence	n=287		Moderate

Note:

1. The figures shown here indicate the number of studies included in the systematic review by Hall *et al.* (2009) in relation to a particular adherence factor. For example, for 'Duration of gluten-free diet' one study out of five (1/5) from the Hall *et al.* (2009) systematic review showed an association between duration of gluten-free diet and adherence to a gluten-free diet and four out of five (4/5) studies showed no association between adherence and duration of gluten-free diet.

Socio-cultural and environmental factors

Table 2.9 shows the results of the analysis for studies that report on socio-cultural and environmental factors in relation to adherence to a GFD. One small study (n=146) identified a new factor, eating convenience foods, as being associated with poorer adherence.

Hall *et al.* (2009) identified inconsistent evidence that eating away from home was associated with adherence with one study showing an association with adherence and another showing no association. I identified five further studies (total n=3526) that all associated eating away from home with poorer adherence to a GFD. Overall the evidence suggests this factor is associated with adherence to a GFD.

Having supportive family and friends was not associated with adherence to a GFD in the systematic review by Hall *et al.* (2009). I identified no further research on this factor.

I identified four new studies examining the association of the availability of GFF with adherence, which added to the singular study identified by Hall *et al.* (2009). Overall, the evidence was inconsistent with three studies (n=499) showing an association with adherence and two studies (n=395) showing no association for this factor.

A new factor regarding the level of choice or restriction represented by a GFD was identified in two studies both of which found this to be associated with adherence. Hall *et al.* (2013) reported that having a better choice of GFF would improve adherence and participants in a larger study by Lee *et al.* (2013) reported a reason for non-adherence was because the GFD was too restrictive. These two studies provide evidence that having a larger choice of GFF is associated with improved adherence to a GFD.

Hall *et al.* (2009) identified inconsistent evidence that the quality (e.g. taste and texture) of GFF was associated with adherence (one study was associated with adherence, one study was not). I identified three further studies (total n=3365) that all associated the quality of GFF with adherence. Overall the evidence suggests that the poor quality of GFF is associated with non-adherence to a GFD.

The higher cost of GFF compared to non-GFF has inconsistent support in the evidence identified. Overall two studies (N=308) found no association, but four other studies (n=3599) did find an association. Overall the evidence suggests that the cost of GFF is associated with adherence to a GFD.

Overall the availability of GFF is inconsistently associated with adherence. Having supportive family and friends has not been associated with adherence to the GFD. However eating convenience foods, eating away

from home, the choice of GFF, the quality of GFF and the cost of GFF are all associated with adherence.

Table 2.9 Socio-cultural/environmental factors (including results from Hall *et al.*'s (2009) systematic review)

Authors	Adherence factor	Number of participants in studies that show an association with adherence	Number of participants in studies that did not show an association with adherence	Assessment of quality
Black <i>et al.</i> , 2011	Eating convenience foods was associated with poorer adherence	N=146		Moderate
Hall <i>et al.</i> , 2009	Eating away from home	1/2 ¹ (n=154)	1/2 ¹ (n=234)	Moderate/High
Black <i>et al.</i> , 2011	Eating away from home in restaurants/take-away was associated with poorer adherence	N=146		Moderate
Hall <i>et al.</i> , 2013	Difficulty when eating out (unclear information) was associated with difficulty adhering	N=287		Moderate
Lee <i>et al.</i> , 2012	Eating in restaurants was associated with non-adherence	N=2922		low
Sey, 2011	Eating away from home when travelling	N=15		Moderate
Smith, 2011	Eating away from home in restaurants and when travelling was reported to make adherence difficult	N=156		Moderate
Sainsbury <i>et al.</i> , 2011	Having supportive family and friends		N278	Low
Hall <i>et al.</i> , 2009	Availability of gluten-free food		1/1 ¹ n=241	Moderate/High
Black and Orfila, 2011	Poor availability of gluten-free breakfast cereals was reported to make adherence difficult	N=146		Moderate
Edwards-George <i>et al.</i> , 2009	Poor availability of gluten-free food was associated with poorer adherence		N=154	High
Sverker <i>et al.</i> , 2009	Poor availability of gluten-free food was associated with poorer adherence (hunger leads to gluten consumption)	N=66		Moderate
Hall <i>et al.</i> , 2013	Improved availability of gluten-free food was believed to make adherence easier	N=287		Moderate
Hall <i>et al.</i> , 2013	Improved choice of gluten-free food was believed to make adherence easier	N=287		Moderate
Lee <i>et al.</i> , 2013	Belief that the gluten-free diet is too restrictive was associated with non-adherence	N=2922		Low
Hall <i>et al.</i> , 2009	Quality of gluten-free food	1/2 ¹ (N=154)	1/2 ¹ (N=234)	Moderate/High
Hall <i>et al.</i> , 2013	Participants believed that better quality of gluten-free food would make adherence easier	N=287		Moderate
Lee, 2012	Poor quality (taste) of gluten-free food was associated with non-adherence	N=2922		Low
Smith, 2011	Poor quality (taste) of gluten-free food was associated with poorer adherence	N=156		Moderate
Hall <i>et al.</i> , 2009	Cost of GFF (satisfaction with the price of GFF)	1/2 ¹ (n=234)	1/2 ¹ (n=154)	Moderate/High
Edwards-	Cost of gluten-free food		N=154	High

George <i>et al.</i> , 2009			
Hall <i>et al.</i> , 2013	The higher cost of gluten-free food was reported to make adherence difficult	N=287	Moderate
Lee <i>et al.</i> (2012)	The higher cost of gluten-free food was associated with non-adherence	N=2922	Low
Smith, 2011	The higher cost of gluten-free food was associated with non-adherence	N=156	Moderate

Note:

1. The figures shown here indicate the number of studies included in the systematic review by Hall *et al.* (2009) in relation to a particular adherence factor. For example, for 'Eating away from home' one of two studies (1/2) from the Hall *et al.* (2009) systematic review showed an association between eating away from home and adherence to a gluten-free diet and one study (1/2) showed no association between adherence and eating away from home.

Quality of life and psychological well-being factors

Table 2.10 shows the results of the analysis for studies that report on Quality of life and psychological well-being factors in relation to adherence to a GFD.

Hall *et al.* (2009) identified inconsistent evidence for depression being associated with adherence. I also found inconsistent results, identifying three studies that showed an association between depression and non-adherence (total n=822) and one large low quality study (n=2265) that showed no association between depression and adherence. Overall there is inconsistent evidence for any association between depression and adherence to a GFD.

Having an eating disorder was a new factor that was associated with poorer adherence in one study (n=390) (Sainsbury *et al.* (2013a).

Five studies included in the systematic review by Hall *et al.* (2009) found no association between anxiety and adherence (total n=1696). I identified a further three studies (total n=3354) that all showed an association between anxiety and poor adherence to a GFD. Therefore, there is inconsistent evidence to support any association between anxiety and adherence to a GFD

Hall *et al.* (2009) identified two studies that showed no association between mood or stress, anger, or psychological disturbance and adherence. I identified no new studies for these three factors.

In the systematic review by Hall *et al.* (2009), five studies were reported to show an association between QoL and adherence to a GFD and two studies showed no association. In my update to this systematic review, I found QoL to be inconsistently associated with adherence. In a study by Lee *et al.* (2012) (n=2922) participants who are adherent to a GFD were reported to have a lower QoL, whereas Nachman (2010) and Sainsbury *et al.* (2011) reported reduced QoL in participants who are non-adherent. In total eight studies showed an association between QoL and adherence (n= 4882) and five studies did not (n=1656). The evidence suggests that there is not a strong relationship between QoL and adherence to a GFD.

One new study by Ford *et al.* (2012) reported that wellbeing was not associated with adherence (n=288). No other studies reported on wellbeing in association to adherence to a GFD.

Overall the evidence for an association between adherence to a GFD and the factors in the area of quality of life and psychological well-being is not strong. One study found an association between eating disorders and adherence to a GFD.

Table 2.10 Quality of life and psychological well-being (including results from Hall *et al.*'s (2009) systematic review)

Authors	Adherence factor	Number of participants in studies that show an association with adherence	Number of participants in studies that did not show an association with adherence	Assessment of quality
Hall <i>et al.</i> , 2009	Depression	1/3 ¹ (n=66)	2/3 ¹ (n=540)	Moderate/High
Edwards George <i>et al.</i> , 2009	Depression was associated with poorer adherence	n=154		High
Sainsbury, 2011	Depression was associated with poorer adherence	n=278		Low
Sainsbury, 2013a	Depression was associated with poorer adherence	n=390		Moderate
Van Hees, 2013	Depression		n=2265	Low
Hall <i>et al.</i> , 2009	Presence of psychological disturbance		1/1 ¹ (n=154)	Moderate/High
Sainsbury, 2013a	Eating disorders were associated with poorer adherence	n=390		Moderate
Hall <i>et al.</i> , 2009	Anxiety		5/5 ¹ (n=1696)	Moderate/High
Edwards George, 2009	Anxiety was associated with poorer adherence	n=154		High
Lee, 2012	Anxiety (uncomfortable in social settings) was associated with poorer adherence	n=2922		Low
Sainsbury, 2011	Anxiety (worry about inconveniencing or offending other people associated with poorer adherence)	n=278		Moderate
Hall <i>et al.</i> , 2009	Anger		1/1 (n=139)	Moderate/High
Hall <i>et al.</i> , 2009	Mood or stress		1/1 (n=154)	Moderate/High
Hall <i>et al.</i> , 2009	Quality of life	5/7 ¹ (n=1559)	2/7 ¹ (n=507)	Moderate/High
Ford <i>et al.</i> , 2012	Quality of life		n=288	Moderate
Lee <i>et al.</i> , 2012	Quality of life was lower in adherent participants	n=2922		Low
Nachman, 2010	Quality of life was lower in non-adherent participants	n=123		High
Barratt <i>et al.</i> , 2009	Quality of life		n=573	Moderate
Sainsbury, 2011	Quality of life was lower in non-adherent participants	n=278		Low
Ford <i>et al.</i> , 2012	Wellbeing		n=288	Moderate

Note:

1. The figures shown here indicate the number of studies included in the systematic review by Hall *et al.* (2009) in relation to a particular adherence factor. For example, of the three studies from the Hall *et al.* (2009) systematic review that showed results for depression, one of the three studies (1/3) showed an association between depression and adherence to a gluten-free diet and the other two (2/3) showed no association between adherence and depression.

2.5 Discussion

Since the previous systematic review search was carried out in 2007 by Hall *et al.* (2009) several new studies reporting on factors affecting adherence to a GFD have been published. This update to Hall *et al.*'s (2009) systematic review identified 21 new studies. In this section, I present a brief discussion of the results of this systematic review. A fuller discussion of the results will be presented in the discussion chapter of this thesis along with the results of my concept mapping study.

In the systematic review by Hall *et al.* (2009) levels of strict adherence to a GFD was found to vary considerably (range = 42% to 91% adherence). In my update to Hall *et al.*'s (2009) review, I also found that adherence to a GFD was variable (range = 44.2% to 100% adherence). However, the one study that reported 100% adherence had an inclusion criterion that only allowed adherent participants to be recruited (Sey *et al.*, 2011). Two other studies reported high levels of adherence to a GFD at 98% (Lee *et al.* 2012) and 96% (Black & Orfila, 2011). However, Lee *et al.* (2012) highlighted the fact that participants had over-estimated their level of adherence with a 'surprising number' later admitting to dietary transgressions. Similarly, a study by Leffler *et al.* (2008), which was included in the systematic review by Hall *et al.* (2009) found that participants tended to over-report their level of adherence to a GFD. It is likely that this over-estimation of adherence also occurred in other studies included in this systematic review. Further, it

is known that members of CD advocacy groups tend to be better at adhering to a GFD (Leffler *et al.*, 2008). As several of the studies included in this and the previous systematic review recruited participants from coeliac advocacy groups, it is possible that the levels of adherence presented here are lower than those seen in the wider population of adults with CD. In addition, individuals who volunteer to take part in research may be more likely to be adherent to a GFD. Therefore, the true rates of non-adherence may be far higher than is reported in this systematic review.

This systematic review found no consistent relationship between adherence and demographic factors, which supports the findings from the previous systematic review by Hall *et al.* (2009). Education, age, gender, social class, employment status, marital status and ethnicity do not appear to be associated with adherence to a GFD. Further evidence is needed in relation to the association between smoking and adherence to a GFD.

Studies have examined a number of factors relating to knowledge and understanding in relation to adherence to a GFD. Being knowledgeable about the GFD and understanding the consequences of no-adherence appear to be associated with better adherence. However, no association was found between adherence to a GFD and understanding or reading food labels. There may be a need for better education in relation to CD and the GFD.

Personality traits and self-efficacy were associated with adherence to a GFD. Developing patients' organisational skills, teaching them the skills

needed to confidently ask for GFF when eating away from the home may help to improve adherence to a GFD.

Interestingly, having trust in others to prepare GFF was associated with poorer adherence. The authors suggest this is related to being less vigilant in relation to avoiding gluten (Sainsbury *et al.*, 2011).

Factors relating to illness and symptoms vary in their association with adherence to a GFD. No association was found between adherence and time since diagnosis, age at diagnosis, the presence of symptoms at diagnosis or the presence of symptoms when gluten is consumed. One study from the previous systematic review by Hall *et al.* (2009) reported that diagnostic delay was associated with adherence to a GFD. However, this is based on just one study and more evidence is needed.

Having an additional food intolerance may be associated with better adherence to a GFD. However, the reason for the association with adherence is not reported and further research is needed. Investigations into the association between body weight and adherence to a GFD suggest that obesity is linked with poorer adherence. This evidence is based on just one study, however, the study was judged to be of high quality and it included a large population (n=1018).

The provision of GFF on prescription in the UK is thought to make it easier for people with CD to follow a GFD (Coeliac UK 2013d). One UK study

reported that receiving GFF on prescription was associated with better adherence to a GFD. Although this evidence is based on just one study (n=287), it does support Coeliac UK's argument against the recent cutbacks in prescribed GFF (Coeliac UK 2013b).

No association was found between adherence to a GFD and the duration of the GFD or perceiving the GFD to be difficult.

Healthcare professionals are responsible for providing the patients with information and advice at the time of CD diagnosis and it is recommended that patients are regularly reviewed after this time. The regularity of follow-up with healthcare professionals was associated with better adherence in this review and it is possible that healthcare for people with CD may need to improve in order to increase adherence to a GFD. This review did not find strong evidence of an association between adherence and satisfaction with the information received from healthcare professionals or attendance at a coeliac clinic.

Coeliac support groups are available in several countries and this systematic review provides strong evidence that membership to a support group is associated with better adherence to a GFD. However, the direction of causality is not known and it is possible that those people who are better at sticking to a GFD are more likely to join a coeliac support group than those who are non-adherent.

Travelling and eating away from the home can be problematic for people with CD and the evidence from my review shows that eating away from home is linked with poorer adherence. Poor awareness of CD by staff in restaurants (Karajeh *et al.*, 2005) and poor availability of GFF when eating away from the home can make adherence to a GFD difficult. People with CD were reported to have difficulties with adhering to a GFD because it is restrictive with limited choice. These findings suggest that better provision of GFF is needed for people with CD when eating outside of the home. However, the availability of GFF was a factor that was not associated to adherence to a GFD.

Although it is claimed that the quality of GFF has improved in recent years, evidence from this systematic review highlights the ongoing dissatisfaction with the taste and texture of GFF and this is linked with poorer adherence. Olsson *et al.* (2008) found that the poor sensory qualities of GFF was linked with non-adherence in Australian adolescents. Patients are often unwilling to give up their favourite gluten-inclusive foods because they taste better than comparable GF products (Stuckey, 2008).

One of the barriers to adherence to a GFD may be the high cost of substitute gluten-free foods (Cureton 2007). The cost of speciality GFF is usually more expensive than gluten-inclusive equivalent foods (Lee *et al.*, 2007) and the high price of GFF has been found to be associated with non-adherence to a GFD. In the UK, prescribed GFF is provided to patients with a CD diagnosis and this is believed to bring the cost of a GFD in line with a

gluten-inclusive diet. In this systematic review, one study that reported the high cost of GFF as a barrier to adherence was conducted in the UK. The other new studies that associated cost with adherence were conducted in the USA where GFF is not provided through the healthcare system.

Adhering to a GFD in the face of psychological problems, may be particularly challenging, however, the link between psychological symptoms and GFD adherence is tenuous. Contradictory evidence exists in relation to the association between depression and anxiety and adherence to a GFD. Although some studies found depression and anxiety to be related to poorer GFD adherence, the direction of causality is unknown.

Although several studies have reported on the relationship between QoL and adherence to a GFD, the evidence is contradictory and more research is needed.

This systematic review has some limitations. Nine different countries were represented in the studies included in this review. It is likely that people with CD will experience different problems associated with following a GFD depending on the country in which they live. For example, some countries may provide better healthcare and resources for CD patients and this may affect the types of problems faced by people trying to follow a GFD. In the UK, GFF is provided on prescription, and this mitigates some of the costs associated with following a GFD. Furthermore, the availability

of GFF is likely to vary between countries. These differences could explain some of the conflicting findings reported in this systematic review.

Another limitation is the differences between how adherence to a GFD is assessed and defined. This makes it difficult to make comparisons between studies. The reliability of the methods of measuring adherence to a GFD was generally poor. Self-reported adherence is not regarded as a reliable measure, however, this method was used in several of the studies included in this and the previous systematic review. There is a requirement for further research to identify the true levels of adherence to a GFD using more reliable means of assessment. In addition agreement on what constitutes adherence to a GFD is needed.

Much of the research into the factors affecting adherence to a GFD in coeliac patients has produced conflicting results. In summary, this systematic review found that the factors affecting adherence to a GFD include knowledge and understanding of CD and the GFD, self-efficacy, organisational skills, the presence of additional food intolerances, body weight, the provision of GFF on prescription, membership of a coeliac advocacy group, eating away from the home and the cost and quality of GFF. I have been unable to find conclusive evidence about many of the factors reported in relation to adherence to a GFD and further studies are required. Additionally the relative importance of these factors to dietary adherence is not clear, thus making targeting of an adherence intervention

difficult, as it is impossible to currently know which factors have the greatest impact on adherence behaviours.

Chapter 3: Methods

3.1 Introduction

“The best way to have a good idea is to have a lot of ideas”

Linus Pauling.

Concept mapping is a mixed methods participatory approach that allows the views of participants from multiple stakeholder groups to be explored. In this study, participants were recruited from three stakeholder groups who were experienced in coeliac disease (CD): adults with CD; adults who live with them (household members); and healthcare professionals who work with adult coeliac patients.

In the first part of this chapter I provide a rationale for selecting concept mapping as an effective method for investigating the factors affecting adherence to a gluten-free diet (GFD). Comparisons are made between concept mapping and alternative methods that were considered for this study. In the remainder of this chapter I explain how the six-step concept mapping process was used to plan the study, gather and analyse the data and in interpreting the results.

3.2 Steering group

A steering committee was set up to guide decisions about how this project was conducted from start to end. Along with myself, the steering group consisted of: two PhD supervisors (KD and RG); two representatives from Coeliac UK (NM and LM (LM was later replaced by HU); a Gastroenterologist (IF) working at Norfolk and Norwich University Hospital (NNUH); two experts in the concept mapping method (SS and AI) and a lay advisor with CD (GN).

This expert panel included representatives from each of the three stakeholder groups that participated in this study: Two members have CD; one has a wheat intolerance; one member lives with an adult who has CD; and one is a health care professional working with adult coeliac patients. I invited our lay advisor to all steering group meetings but, unfortunately, he could only attend one.

Having a steering group helped to ensure that service-user views and expert opinion were taken into account in the design and conduct of this study. As Chief Investigator, I was responsible for chairing the meetings and ensuring steering group members were clear about the aims of the project.

3.3 Choice of method

To achieve the aims of this study it was important to select the most appropriate research method. The primary aim of this study was to gain a better understanding of the factors affecting adherence to a gluten-free diet (GFD). A further aim was to uncover differences and similarities in the views of three stakeholder groups (adults with CD; household members; and healthcare professionals).

3.3.1 Potential methods of investigation

To understand health behaviour, it is necessary to listen to the people whose behaviour we want to change. Currently, little is known about the impact of CD on daily living from patients' perspectives (Ukkola *et al.*, 2012). In order to get a better understanding of the problems encountered by patients in relation to sticking to a GFD, I endeavoured to select a method that would allow me to collect patients' views. In addition, I also wanted to gather the perspectives of other stakeholders who have close contact with people with CD.

The views of people who live with coeliac patients was deemed to be important for this study because these individuals are likely to have experience of what life is like for the person with CD, how they cope with avoiding gluten and what causes them to be non-adherent. Household

members may also be more willing to disclose information relating to non-adherence than patients.

I also wanted to collect the views of healthcare professionals who work with adult coeliac patients because they are likely to have a good insight into the patients' views about what helps them to stick to their GFD. Healthcare professionals are likely to work with a diverse range of coeliac patients and I felt this would be a useful way of collecting information about the factors affecting adherence from a broader perspective.

The steering group met to discuss ideas about the most appropriate research method for this study. From the five methods that were considered as potential approaches, concept mapping was selected as the most appropriate for achieving the study's aims. In addition, concept mapping was deemed to be a suitable method for working within the resources available for this study.

Mixed methods research has gained support in recent years and it can draw on the strengths from both qualitative and quantitative methods. Mixed methods research is useful for obtaining rich, subjective data and transferring it into a useful quantitative format. A mixed methods approach was suggested during the steering group meeting as a useful method for understanding the complexities of dietary behaviour and developing the results in a quantifiable way that can be used to develop a model of adherence for use in an intervention.

Table 3.1 shows the criteria used in selecting the most appropriate research method. Unlike some studies into adherence to a GFD, I wanted to avoid using pre-determined questionnaires as this would not allow participants to generate new ideas in relation to what helps them stick to a GFD.

Table 3.1 – Criteria for confirming the most appropriate research method for this study

	Comparisons can be easily made between stakeholder groups	Data is easily quantifiable	Identifies new adherence factors, rather than pre-determined factors
Concept Mapping	Yes	Yes	Yes
Focus Groups	No	No	Yes
Interviews	No	No	Yes
Questionnaires	Yes	Yes	No
Delphi technique	No	No	No

Concept mapping uses sophisticated multivariate analysis to produce quantitative results which are presented in the form of easily interpreted visual maps. These maps can be used to guide the development of an adherence intervention. Focus groups are a less sophisticated conceptualisation approach which, unlike concept mapping, does not generate a well-defined set of quantitative results (Kane & Trochim, 2007).

Interviews can be a useful method for gaining a deeper insight into complex human behaviour. However, interviews are time consuming, open to bias and the results are not usually quantifiable. Questionnaires can be a useful way of collecting quantifiable data. However, questionnaires relating to

adherence to a GFD mostly rely on using pre-determined adherence factors identified in previous studies, rather than allowing participants to freely determine the influences on adherence to a GFD. The questionnaires designed by (Leffler *et al.*, (2008) and Butterworth *et al.* (2007) are examples of this.

Both the Delphi technique and concept mapping involve participants who are considered to be expert in the subject being explored. The Delphi technique is useful for gaining consensus on things such as healthcare policies and guidelines. The Delphi technique was deemed to be inappropriate for this study because it does not represent a range of ideas in a framework that can be used for planning an intervention. In addition, the Delphi technique would not produce data that would allow comparisons to be made between different stakeholder groups.

Concept mapping was successfully used in a previous adherence study which investigated adherence to medication in people with schizophrenia (Kikkert *et al.*, 2006). This study used three stakeholder groups with approximately 25 participants in each group. The success of this study confirms that concept mapping is a suitable method for investigating adherence to treatment and it provides a useful framework for the design of this study.

3.4 Background to concept mapping

Concept mapping is a participatory mixed methods approach that was developed by Trochim in the 1980s (Kane & Trochim, 2007). This participatory mixed methods approach is designed to increase understanding of complex topics, such as health behaviour. The method involves participants at each stage of a six step process and seeks to yield an interpretable visual map of their ideas. Statements generated during brainstorming are sorted in order of priority and clustered into themes by participants. Data are analysed to produce interpretable concept maps. The topology of thoughts and ideas generated from this study can be used to guide the development of a novel adherence intervention for people with CD. This will be the first study to explore the factors affecting adherence to a GFD in adults with CD using concept mapping.

3.5 The six steps in concept mapping

3.5.1 Step 1: Preparing for concept mapping

Planning the project

Concept mapping commences with the development of a focus for the study and the identification of suitable stakeholder groups from which samples of participants can be recruited (Kane & Trochim 2007). This step also

involves making preparations for the remaining steps in the concept mapping process.

Deciding on the focus

Concept maps are constructed in reference to a particular focus question on a topic that is familiar to the participants (Novak & Canas, 2008). To achieve the aim of this study, the task for the steering group was to decide on a focus question, or prompt, that would generate responses from participants during brainstorming in answer to the research question. The focus statement was also used during the statement sorting exercises (prioritising and clustering) and was intended to act as an instruction or prompt for responses from participants. Ideas for the focus question were discussed during a meeting with members of the steering group and the group arrived at a decision to use the following focus prompt: *'It would be easier for adults with coeliac disease to stick to a gluten-free diet if...'*. The prompt was designed to encourage only relevant statements to be generated.

Ethical approval

An ethics application was drawn up which included a study protocol and several participant documents (Appendices 4 to 22). A positive opinion was gained from the North East – Newcastle and North Tyneside 2 Research Ethics Committee (Appendix 23). A research and development application was also approved by the NNUH and Norfolk PCT (Appendix 24).

Recruitment

Concept mapping projects are most successful when they involve a wide variety of relevant people (Kane & Trochim, 2007). The steering group identified three key stakeholder groups with expert knowledge about the topic of interest. These were: adult coeliac patients, adults who live with them (household members) and healthcare professionals who work with adult coeliac patients. Kane & Trochim (2007) suggest that there is no strict limit concerning the number of participants included in a concept mapping study. Successful concept generation has been conducted with one lone participant, and also groups of 75-80 people.

I based my recruitment strategy on that employed by Kikkert *et al.* (2006) who conducted a concept mapping study to investigate adherence to medication in schizophrenia. This study included 27 patients; 29 carers and 28 healthcare professionals. I aimed to recruit 25 adult patients with CD, 25 adults who live with an adult who has CD (household members) and 25 healthcare professionals who work with adult coeliac patients. Although it was not possible to do a power calculation for this study, I felt confident that I would reach data saturation with a population of 75 participants. Sainsbury & Mullan (2011) interviewed 13 participants on factors affecting adherence to a GFD and 30 out of the 36 items raised were endorsed by five or more people and the remaining six were endorsed by either three or four people, suggesting data saturation.

To help me collect a diverse range of opinions relating to the factors affecting adherence to a gluten-free diet, I sought to recruit patients with varying levels of adherence to a GFD. In the context of this study, the term ‘adherence’ refers to the life-long exclusion of gluten from the diet. Although foods labelled as ‘gluten-free’ or ‘low in gluten’ often contain small amounts of gluten, consumption of these products is not regarded as non-adherent. The reason for this is that the gluten content is thought to be too low to trigger a response and the use of these products is recommended by healthcare professionals (Food Standards Agency, 2012).

In this study I only regarded those people who knowingly consume gluten as being non-adherent. A problem with this definition is that some people who regard themselves to be adherent may be unaware that they are inadvertently consuming gluten (Hall *et al.*, 2013). Reasons for this could be that they are consuming ‘hidden gluten’, as described in Section 1.3.6, their food may be contaminated with gluten or they may be misinformed about which products do or do not contain gluten. Unfortunately, this limitation in my definition of adherence to a GFD cannot easily be avoided, particularly when using self-reporting to measure adherence, as I am doing in this study.

Due to financial constraints, the steering group agreed that adherence should be self-reported by participants, rather than being confirmed through clinical testing, which is costly. To establish the level of adherence to a GFD in potential participants with coeliac disease were asked to report their level of adherence on a 3-point Likert scale. This measure has been used in several

previous studies into adherence to a GFD (Hall et al., 2009). Participants were asked to select from the following statements the one that was most relevant to them:

- 1). I always stick to a strict gluten-free diet
- 2). I occasionally consume food/drinks containing gluten
- 3). I do not follow a gluten-free diet

The inclusion criteria for each stakeholder group is shown in Table 3.1.

Table 3.1 - Inclusion criteria for the recruitment of participants from three stakeholder groups

Adults with coeliac disease	Household members	Healthcare professionals
All participants must be aged ≥ 18 years		
All participants must be capable of giving informed consent		
Because the method involves agreeing the wording of statements, only English speaking participants will be enrolled		
Has been medically diagnosed with coeliac disease	Must have lived with an adult with coeliac disease for at least 1 year	Currently working with at least 1 adult patient with coeliac disease
		Has at least one year's experience of working with coeliac patients

Early in the study, the steering group agreed that recruitment should be within a 10 mile radius of Norwich. This would make it more convenient for participants attending group data collection sessions at the University of East Anglia. As we were offering to refund travel expenses, this strategy would help to keep expenses to a minimum. However, I later decided to extend the recruitment area to the whole of England and Wales when I

found recruitment of healthcare professionals and partially- or non-adherence participants with CD to be slow. All participants who lived outside 10 miles of Norwich were invited to complete the data collection exercises remotely by email or post. Participants who lived further than 10 miles away from Norwich were advised that they were welcome to join the group sessions, but that we would not be able to refund their travel expenses. I gained approval for this change to the study design from the North East – Newcastle and North Tyneside 2 Research Ethics Committee (Appendix 25).

Seven methods of recruitment were used in this study:

1. Invitation letters posted by Coeliac UK

Coeliac UK has approximately 1000 Members living within 10 miles of Norwich (Postcodes: NR1-NR10; NR12-NR16 and NR18). This includes both rural and urban areas. It would not have been appropriate for me to send invitation letters to all 1000 Coeliac UK Members as I only aimed to recruit 25 participants with CD. I agreed with the steering group and Coeliac UK that a random sample of 100 members was a sufficient number to invite. Computer randomisation was used by Coeliac UK to select 100 of their adult Members with CD in the Norwich area. Invitation letters (Appendix 6) were distributed by post to this random selection of Coeliac UK Members. To avoid the need for me to gain access to personal information, invitation letters were sent out by Coeliac UK on my behalf. A reply slip and a postage paid return envelope were enclosed.

Coeliac UK Members who responded to the invitation letter were sent a full information pack by post (Appendices 7, 12, 15, 18 and 21) along with an information pack addressed to 'Household Member' (Appendices 9, 13, 16, 19 and 22). The patient information pack consisted of an invitation letter, an information sheet, a consent form, a self-assessment of adherence and a postage paid return envelope. People with CD were asked to identify an adult in their household who might be interested in participating in the study and to pass on the household member's pack to that person.

2. Invitation packs distributed by Norfolk and Norwich University Hospital (NNUH).

Invitation packs were handed to patients with CD during outpatient consultations at NNUH. Community dietitians were given invitation packs to distribute to non-adherent patients who they came in contact with during their working activities. These invitation packs also contained a household member's pack.

3. Press release in two local newspapers.

Not all coeliac patients join Coeliac UK, and patients referred to NNUH are normally discharged after diagnosis and proof of adherence to a GFD. Therefore to pick up a more representative population of people with CD, I put an advertisement in two local newspapers (The Eastern Daily Press and the Norwich Evening News) to invite partially-adherent and non-adherent people with CD and household members to participate in this study

(Appendix 26). This was done after I had recruited a sufficient number of adherent participants.

4. Advertisement on Coeliac UK website.

An advertisement for the project was placed on the Coeliac UK website (Appendix 27). This targeted healthcare professionals, partially-adherent or non-adherent people with CD and household members. This was done after I had recruited sufficient adherent participants.

5. Advertisements on the University of East Anglia website.

An advert for the project was placed on the University of East Anglia's website (Appendix 28). This targeted healthcare professionals, partially-adherent or non-adherent people with CD and household members. This was done after I had recruited sufficient adherent participants.

6. E-mail to the British Dietetic Association (BDA) Gastroenterology Specialist Group.

An e-mail to the BDA Gastroenterology Specialist Group was sent by Coeliac UK to invite healthcare professional members who work with coeliac patients to participate in the study (Appendix 29). Interested professionals were invited to contact me to request an information pack.

7. Invitation packs posted to GP practices in Norfolk.

One hundred and twelve invitation packs were posted to GPs, GP assistants, Nurse Practitioners and Practice Nurses working in GP surgeries listed on the PCT (now CCG) website as being within 10 miles of Norwich.

Effort was made to ensure diversity within the group of participants. The information packs contained a short questionnaire which captured demographic data from participants. For the patient group, I also captured some clinical data, including self-reported level of adherence to a GFD (Appendices 15 to 17). Table 3.2 shows a list of participant characteristics captured by the questionnaires. I also asked participants to inform me about their availability for attending the group brainstorming and statement sorting sessions.

Table 3.2 Recorded Participant Characteristics

Adults with coeliac disease	Household members	Healthcare professionals
Age	Age	Gender
Gender	Gender	Profession
Ethnicity	Ethnicity	
Level of education	Level of education	
Level of adherence to a GFD		

Informed consent was obtained from all participants. This was done face-to-face for patients and household members living in the Norwich area and by telephone and post for those living outside of the Norwich area. All healthcare professionals were assumed to have a high level of competency and, therefore, consent was completed by post.

Confidentiality

To maintain participant anonymity, random 3-digit participant reference numbers were generated using a random-three-digit code selector. Any 3-digit numbers generated that were too memorable (e.g. 999) were not used. Reply slips and consent forms were stored securely in a locked filing cabinet inside a locked room at the University of East Anglia. Electronic documents were password protected and stored on a University of East Anglia computer with password security. Participants were allocated a 3-digit reference number and this was used in place of the participant's name whenever relevant.

Preparation for the next steps

During Step 1, action was taken to prepare for the following steps in the concept mapping process. The steering group members agreed that the University of East Anglia would be a suitable venue to host the group meetings with participants. The venue provided free car parking, refreshment facilities, disabled access, toilet facilities and good public transport links.

I contacted participants who had indicated that they wished to attend the group sessions to advise them of the dates when the meetings would be held. The dates were scheduled to accommodate the availability of as many participants as possible as they had indicated on their reply slips (Appendices 21 and 22). Participants were also given the option to complete brainstorming remotely by e-mail or post if they preferred to do so. To

reduce the likelihood of drop-out or disruption at the start of the group sessions, reminder letters were sent out to participants in advance (Appendix 31). This letter contained information about travel to the venue with a map, venue details and a travel expenses claim form.

To ensure that participant attending the group brainstorming sessions were given the opportunity to contribute their ideas, I included a maximum of eight participants in each group brainstorming session. This also helped me to ensure that perspectives on adherence to a GFD were explored in depth. All adults with coeliac disease and household members were invited to either attend group brainstorming sessions with other participants at UEA or to complete brainstorming remotely by post or e-mail. To encourage recruitment and prevent drop out from a group that proved difficult to recruit, all health professionals were invited to complete brainstorming remotely and no group sessions were held for this group.

To ensure consistency in the information that was provided during group sessions, a script was designed which included instructions to participants (Appendix 31). For those completing the tasks remotely, instructions were posted or e-mailed according to participants' preferences. The steering group agreed on a list of prompts that could be used during group brainstorming if the flow of ideas dried up (Appendix 31). These prompts were based on evidence taken from the literature.

In preparing for the group brainstorming and statement sorting sessions, I sent e-mails or letters to 28 gluten-free food (GFF) manufacturers to request donations of gluten-free (GF) products so that I could provide participants with snacks during the group sessions (Appendix 32). Nine companies donated products which enabled me to provide participants with snacks during the group sessions.

3.5.2 Step 2: Brainstorming

Concept mapping begins with a structured brainstorming session where participants are asked to generate statements in response to a focus prompt. The focus prompt used in this study was: *'It would be easier for adults with coeliac disease to stick to a gluten-free diet if...'*. Participants were asked to brainstorm their ideas in response to this focus prompt until all ideas were exhausted.

Participants who completed brainstorming remotely by post or e-mail were asked to record as many statements as they could think of in response to the focus prompt. Responses were returned to me by post or e-mail.

Group brainstorming sessions were held for adults with CD and household members. Where sessions were held at the same time for these two groups of stakeholders, separate rooms were used. The sessions were facilitated by one of four members of the research team (HF, KD, SS or AI) with another of the four members acting as an assistant. All facilitators were given the

same set of instructions. Written and verbal instructions were provided to all participants at the start of the session. The time allowed for all group brainstorming sessions was 2 hours. The facilitator read out instructions at the start of the session and used prompts from a list (taken from the existing literature) if the session dried up (Appendix 31). The assistant wrote the statements up on a flip-chart and pinned the statements on the wall where they could be viewed throughout the session. The duration of the brainstorming sessions was two hours with 15 minute refreshment break half way through.

In a concept mapping study by Bayer *et al.* (2010), it was recognised that participants did not participate in the first brainstorming session during research into adolescent sexuality. It was suggested that this may be due to the fact that some adolescents may have felt too embarrassed to raise certain issues in relation to their sexuality. However, in subsequent brainstorming sessions, participants were given the opportunity to write responses individually and this modification ‘catalysed’ the generation of statements. In the current study, participants were given a pen and some paper to use during brainstorming sessions so that they could contribute any statements that they didn’t wish to disclose to the group or if they thought of something while someone else was speaking and did not want to forget it. This helped to ensure that only one person spoke at a time, which helped to ensure no statements were missed or not picked up on the audio recording.

Participants were encouraged to generate as many statements as they could think of during brainstorming and criticism of other people's statements was discouraged. Participants were advised that, if they disagreed with a statement, then they could raise a counter statement.

Data were collected by e-mail or post from participants who did not take part in group sessions.

3.5.3 Step 3: Structuring the statements (prioritising and clustering)

Statement reduction

The steering group met to review the full set of statements generated by all participants. The aim of this meeting was to ensure the final set of statements was 98 or fewer. The Ariadne® software package for concept mapping (NcGv/Talcott, 1995) can only accept a maximum of 98 statements). This was achieved by synthesising statements that were the same or similar and by excluding statements that were irrelevant or too vague.

Different coloured paper was used to print the statements from each group. This helped to clarify any ambiguous statements and allowed us to ensure that the statements included in the final set included a fair representation of the statements generated by all three groups. A random number generator was used to allocate a random number to each statement in the final set. The

final set of statements was printed on small cards (8cm x 3cm) and two full sets of 91 cards were presented to each participant for the prioritising and clustering tasks. The two sets of cards were printed on different coloured card in order to prevent the cards from becoming mixed. Each set of cards was shuffled to ensure that the participants would view the statements in a different order to one another as it was felt that this would reduce any chance of bias from the order in which statements were viewed.

Preparation for prioritising and clustering

In preparation for the prioritisation and clustering tasks, each of the 91 statements was allocated a random number from 1 to 91 using a random number generator. Statements were printed on individual coloured cards with the random number displayed in the top left corner as shown in Figure 4.1 below. Appendix 34 shows the final set of 91 statements in numerical order using the number allocated by the random number allocator. Each full set of 91 statement cards was shuffled so that participants would view the statements in a random order that was different to other participants. Participants were provided with two full sets of the 91 statements, one for the prioritising task and the other for clustering. The sets of cards were printed on different coloured card to reduce the chances of the two sets of cards getting mixed up during the prioritising or clustering tasks.

2. ...if there was more availability of savoury gluten-free snacks and not just sugary cakes and biscuits.

Figure 4.1: Example of a statement card for use in the prioritising and clustering task

Structuring: Prioritisation and clustering

Prioritising and clustering were performed individually by each participant without the input from other participants or from the researchers.

Prioritising

Participants were asked to rate the importance of the 91 statements for importance on a 5-point Likert-type scale with 1 being the least important and 5 the most important. Participants were asked to place the statements into five fairly equal piles representing the priority rank assigned to them (Appendix 35). It is unlikely that any of the statements generated during brainstorming would be considered to be completely unimportant, and it was stressed that the level of importance of a statement should be judged in relation to that of the other statements and ranked accordingly.

Clustering

Participants were asked to group the statements into themes in a way that made sense to them. Participants were instructed that they should not put all the statements into 1 group or to have each individual statement as its own group. Also, participants were advised that they should not have a 'miscellaneous' or 'other' group where a group of unrelated statements were

grouped together. Any statements that were unique and could not be grouped with other statements should be put in a 'pile' on their own as a single statement. After sorting the statements into themes, participants were asked to assign a name to each of their piles and to complete a data sheet (Appendix 36) listing the random number of the clusters under each of their cluster names.

3.5.4 Step 4: Analysing the data

Data entry

Data were entered into the Ariadne® software package for concept mapping (NcGv/Talcott, 1995; Severens, 1987). To ensure anonymity, random 3-digit reference numbers were used to identify participants, rather than imputing participants' names. A coding system was used to identify which of the three stakeholder groups each participant belonged to. The full list of statements was entered into Ariadne along with the random reference number for each statement. The priority rating and clustering data for each participant was entered using the statement reference numbers. For the prioritisation data imputing, statement reference numbers were entered into five columns which represented the different levels of priority (1 = least important; 5 = most important). In the same way, the statement reference numbers were entered in columns to represent how each participant had grouped the statements during the clustering task.

Double data inputting was carried out using two separate computers to reduce the risk of data imputing errors. Ariadne allows for duplicated entries and missing entries to be identified and this further reduced the risk of imputing errors.

Data analysis

The aim of data analysis was to identify the main themes, or concepts, relating to adherence to a GFD and to establish the relative importance of each concept in relation to the others. Data analysis was also used to identify similarities and differences between the perceptions of the three stakeholder groups. Data were analysed using multivariate statistical techniques (multidimensional scaling (MDS) and cluster analysis) to produce interpretable visual concept maps.

Cluster analysis

The programme calculated how frequently statements were sorted into the same group, or theme, by participants during the clustering task. The output of this was a point map which showed each individual statement as a data point on a two-dimensional plot (point map).

The number of clusters to include on a map can be increased or decreased until an appropriate number which accurately reflects the concepts of the topic are represented. The steering group decided on the most appropriate number of clusters to include on the concept map. The steering group were asked to decide on names for each cluster. The aim was to decide on names

that represented the core or common ideas within the group of statements in the cluster. The cluster name was agreed on by all members of the steering group. In this plot (point map), the similarity between each statement is represented as a geographical distance. Statements that were judged to be similar by participants appear closer together on the two-axis matrix.

Prioritisation analysis

The mean priority rating for each statement was calculated by adding together the mean priority ratings (from 1 to 5) and dividing the result by the number of participants who completed the prioritisation task. The mean rating for each cluster was calculated from the mean score of all the statements contained within the clusters. This allowed me to identify which clusters were more or less important.

Identifying differences and similarities between stakeholder groups.

I wanted to find out whether or not there was a difference in medians between stakeholder groups in relation to the level of importance attributed to each of the statements identified from brainstorming. The Kruskal-Wallis test is appropriate for use with ranked data and this test was used to identify significant differences between the three stakeholder groups. Where significant differences were found using the Kruskal-Wallis test, pairwise comparisons among the three groups were made using the Mann-Whitney U test.

3.5.5 Step 5: Interpreting the maps

Each cluster on the concept map was given a label that best described the content of the statements clustered within it. Members of the steering group were presented with the cluster map as an image on a projector screen and on a sheet of paper on the desk. They were instructed to interpret and discuss the meaning and importance of the map in relation to the focus question and to select appropriate labels for each cluster. Members were also given a list of the statements contained in each cluster

3.5.6 Step 6: Utilisation

Step 6 involved the translation of the concept maps. The visual concept maps can be used to inform subsequent work and concept mapping is a useful method for health researchers when planning and evaluating projects, generating hypotheses and developing theories. In this study the concept map, together with the results of the systematic review, were used to develop a model of adherence to a GFD. Discussion about the model of adherence to a GFD and further utilisation of the results from this study are presented in Chapter 5.

3.6 Summary

This study endeavours to address the gaps in the existing literature by utilizing a mixed methods approach known as concept mapping to produce a conceptual model of the factors affecting adherence to a GFD in adults with CD. This method provides meaningful results that can be easily interpreted while still maintaining the richness of data associated with qualitative research. Concept mapping facilitates the involvement of multiple stakeholder groups. Alternative methods were considered, however, concept mapping was judged to be the most appropriate method for achieving the aims of this study. This study was designed to generate a conceptual framework, or model, of adherence to a GFD using concept mapping.

Chapter 4: Results

4.1 Introduction

The aim of this study was to use concept mapping to gain a better understanding of the factors affecting adherence to a gluten-free diet (GFD) in adults with coeliac disease (CD). A further aim was to compare the perspectives of three stakeholder groups: Adults with CD; adults who live with them (household members; and healthcare professionals. To achieve this aim, I collected statements from participants during brainstorming in response to the focus prompt *'It would be easier for adults with coeliac to stick to a gluten-free diet...'*. Participants then sorted the brainstormed statements in order of priority from 1 (least important) to 5 (most important) and grouped the statements for similarity (clustering). The previous chapters explained and justified the use of concept mapping for achieving the aim of this study.

In this chapter I present the results of this mixed methods study in four sections. The results of the seven methods of recruitment employed in this study are presented in Section 4.2. In Section 4.3 the characteristics of participants from three stakeholder groups are presented (adults with CD, adults who live with them (household members) and healthcare professionals). The results of data collection from brainstorming,

prioritisation and clustering activities are presented in Section 4.4 along with results of the data reduction activities carried out by the research team. The results of data analysis, using Ariadne concept mapping software (NcGv/Talcott, 1995) and SPSS™ v.19 software (SPSS Inc. Chicago Illinois), are presented in Section 4.5. This section also includes an explanation of how the most appropriate number of clusters was decided upon to represent the emerging themes. Comparisons are made between the results obtained from each of the three stakeholder groups.

4.2 Recruitment

I sought to recruit a diverse sample of participants from three stakeholder groups: adults with CD; adult household members; and healthcare professionals who work with adult coeliac patients. Seventy-three participants were recruited using seven recruitment methods (Table 4.1). Not all seven recruitment methods were employed for all three stakeholder groups.

Table 4.1 Number of participants recruited from three stakeholder groups using seven recruitment methods

Recruitment method	Adults with coeliac disease with varying levels of adherence to a gluten-free diet				Household members	Healthcare professionals	All participants
	Adherent	Partially-adherent	Non-adherent	Total			
100 invitation letters posted by Coeliac UK	12 (80%)	4 (22%)	0	16 (47%)	10 (48%)	N/A ¹	26 (36%)
13 invitation packs distributed by Norfolk and Norwich University Hospital (NNUH)	1 (7%)	1 (6%)	0	2 (6%)	0	3 (17%)	5 (7%)
Press release in local newspapers ²	0	8 (44%)	1 (100%)	9 (26%)	8 (38%)	N/A ¹	17 (23%)
Advertisements on Coeliac UK website ²	0	3 (17%)	0	3 (9%)	2 (10%)	1 (6%)	6 (8%)
Advertisement on University of East Anglia (UEA) website ²	2 (13%)	2 (11%)	0	4 (12%)	1 (5%)	N/A ¹	5 (7%)
E-mail to the British Dietetic Association (BDA) gastroenterology specialist group	N/A ¹	N/A ¹	N/A ¹	N/A ¹	N/A ¹	12 (67%)	12 (16%)
112 invitation packs posted to GP practices	N/A ¹	N/A ¹	N/A ¹	N/A ¹	N/A ¹	2 (11%)	2 (3%)
Total⁴	15 (100%)	18 (100%)	1 (100%)	34 (100%)	21 (100%)³	18 (100%)³	73 (100%)

Notes:

1. N/A indicates that recruitment did not take place for a particular stakeholder group using the method indicated (e.g. Healthcare professionals were not sent invitation letters posted by Coeliac UK).
2. Once I had recruited sufficient adherent participants through the Coeliac UK invitation letter and Norfolk and Norwich University Hospital (NNUH), I stopped inviting adherent adults with CD to participate and only invited partially-adherent and non-adherent adults with CD.
3. Percentages have been rounded up and, therefore, do not always appear to add up to 100%
4. Total shows the sum of participants and the percentage values in each column

4.2.1 Recruitment of adults with coeliac disease and adult household members

Five methods were employed for recruiting adults with CD and adult household members.

Over one third of the participants who were recruited for this study responded to invitation letters distributed by Coeliac UK. Thirty-two Coeliac UK Members returned completed reply slips and information packs were posted to them along with a household member's pack. Twenty-five adults with CD and 11 adult household members returned a reply slip.

To avoid over-recruitment of adults with CD who adhere to the GFD, rejection letters (Appendix 33) were posted to nine adults with CD. It was not necessary for me to send rejection letters to household members, partially-adherent adults with CD or non-adherent adults with CD as I did not receive an excessive number of volunteers from these groups.

I contacted all eligible volunteers to arrange face-to-face meetings to gain informed consent and to invite the recruited participants to complete a short questionnaire (Appendices 15, 16, 18 and 19). During the face-to-face meetings, some participants with CD advised me that they did not live with another adult and others told me that their household member/s had chosen not to be involved in the study. One female participant with CD requested that her daughter, who also has CD, was recruited for the study. An

invitation pack was sent to the participant's daughter and she was also recruited. A female Coeliac UK Member and her spouse were excluded because she was diagnosed with the related condition, dermatitis herpetiformis, and not CD, which did not fit with the inclusion criteria (Chapter 4).

Information packs were distributed to adults with CD and adult household members by Norfolk and Norwich University Hospital (NNUH).

Two NNUH patients with CD (one adherent and one partially-adherent) volunteered and were recruited following the distribution of 13 information packs during consultations with healthcare professionals. No adult household members responded to the invitation packs handed out at NNUH.

Advertisements were placed in two local newspapers, on the University of East Anglia website and on Coeliac UK website.

Once a sufficient number of adherent participants with CD had been recruited, I invited partially-adherent and non-adherent adults with CD and adult household members to participate in the study. Following advertisements in two local newspapers (Appendix 26), on the UEA website (Appendix 28) and Coeliac UK's website (Appendix 27), I received telephone calls and e-mails from people expressing an interest in taking part. Fifteen people were excluded at this stage: 13 adults with CD who were adherent to the GF diet; and two people who had not received a

positive CD diagnosis. Information packs were posted to 26 adults with CD and 12 adult household members who matched our inclusion criteria (Chapter 3). Ten adults with CD and one adult household member did not respond to the information pack. Despite having already recruited a sufficient number of participants who adhere to the GFD, I recruited a further two adherent participants who replied to the advertisement on the UEA website. These participants had very recently been diagnosed with CD and this increased diversity in the group of adherent participants, many of whom had been diagnosed for many years.

4.2.2 Recruitment of healthcare professionals

Four methods were used to recruit healthcare professionals.

Information packs distributed to healthcare professionals by NNUH

Following the distribution of 22 information packs to healthcare professionals at NNUH, I received three responses from healthcare professionals who were all recruited for this study.

Invitation packs posted to GPs and Practice Nurses in Norwich

The method that resulted in the least number of participants recruited (n=2) was the distribution of 112 invitation packs to GP practices. One GP and one Nurse Practitioner volunteered to take part and were recruited for the study.

Advertisement on Coeliac UK's website

One healthcare professional responded to the advertisement on Coeliac UK's website (Appendix 27) and was recruited for the study.

E-mail sent to the British Dietetic Association's gastroenterology specialist group

Fourteen healthcare professionals responded to the e-mail sent to the British Dietetic Association's (BDA) gastroenterology specialist group. Rejection e-mails were sent to two healthcare professionals because they lived and worked in Scotland, which did not fit the inclusion criteria for this study. Twelve healthcare professionals were successfully recruited following the BDA e-mail.

4.3 Participant characteristics

The participant characteristics are summarised in Table 4.2. Over two-thirds of the 73 participants recruited for this study were female. The sample of adults with CD and adult household members was almost exclusively white British. All three stakeholder groups had a higher percentage of females compared to males with the healthcare professionals' group showing the highest percentage of females (94%). Adults with CD were 6.6 years younger than adult household members. All except one participant (who was recruited from Wales) were based in England and over two-thirds of participants were recruited from Norfolk. Half of the adults with CD and

household members were educated to university degree (or equivalent) or postgraduate level.

Healthcare professionals were recruited from four occupational professions:

- 14 (78%) dietitians (including senior, specialist and research dietitians);
- 2 (11%) Practice Nurses / Nurse Specialists
- 1 (6%) Gastroenterologist
- 1 (6%) General Practitioner

Healthcare professionals had a history of working with CD patients for a mean of 10.2 years (range 1-30 years). It was not possible to calculate the mean number of CD patients that healthcare professionals work with because one participant quoted 'more than 100 patients', another 'more than 40 patients' and others provided either a weekly, monthly or annual figure.

Table 4.2 Participant characteristics

	Adults with coeliac disease (n=34)	Adult household members (n=21)	Healthcare professionals (n=18)	All participants (n=73)
Age (years) ¹				
Mean	53	59 ¹	Data not collected	55 ¹
SD	17	11 ¹		15 ¹
95% CI	47-58	54-64		50-59
Gender				
Male	13 (38%)	8 (38%)	1 (6%)	22 (30%)
Female	21	13	17	51
Ethnicity				
White British	33 (97%)	21 (100%)	Data not collected	54 (98%)
Other	1	0		1
Geographical location				
Norfolk	30 (88%)	17 (81%)	5 (28%)	52 (71%)
Outside Norfolk	4	4	13	21
Education				
No formal qualification	3 (9%) ²	3 (14%)	Data not collected	6 (11%)
GCSE/A' Level	15 (45%) ²	6 (29%)		21 (39%)
University level	15 (45%) ²	12 (57%)		27 (50%)

Notes:

1. One adult household member did not provide information about his age. The mean age and age range for adult household members shown here is the average age of the 20 participants who provided their year of birth.
2. One adult with coeliac disease did not state their level of education.

Table 4.3 shows information on level of adherence to a GFD for the adults with CD group in relation to demographic and clinical characteristics. The number of adherent (n=15) and partially adherent (n=18) participants was fairly equal. The single non-adherent participant who was recruited had been following a gluten-inclusive diet since his wife died in the 1970s. This non-adherent participant reported that he had not experienced any CD symptoms since re-introducing gluten into his diet despite having symptoms prior to diagnosis.

The mean age of participants with CD who adhere to a GFD was 2.9 years higher than the mean age of the partially-adherent participants (50.4 years).

The non-adherent participant was more than 25 years older than the mean age of the adherent and partially-adherent participants. Sixty-nine per cent (n=9) of males with CD were either partially- or non-adherent to the GFD compared to 48% (n=10) of females. Eighteen per cent more adherent participants were educated to university degree or postgraduate level (n=8 (53%)) compared to the partially-adherent participants (n=6 (35%)).

Of the 11 participants who reported severe or very severe symptoms when they consume gluten, nine (82%) were adherent to the GFD and two were partially-adherent. Participants who experienced no symptoms were either partially- or non-adherent to the GFD.

Partially-adherent participants had been diagnosed 7 years longer than the adherent participants. The non-adherent 79-year-old participant had been diagnosed for 50 years, which was the highest of all participants with CD.

Table 4.3 A comparison of the characteristics of adults with coeliac disease who are adherent, partially-adherent and non-adherent to a gluten-free diet.

	Adherent adults with CD (n=15)	Partially adherent adults with CD (n=18)	Non- adherent adults with CD (n=1)	All adults with CD (n=34)
Age (years)				
Mean	53.3	50.4	79	52.6
SD	17	15	--	17
95% CI	46-65	41-56	--	46-58
Gender				
Male	4 (27%)	8 (44%)	1	13 (38%)
Female	11	10	0	21
Ethnicity				
White British	15	17	1	33
Other	0	1	0	1
Severity of symptoms				
No symptoms	0	3 (17%)	1 (100%)	4 (12%)
Mild/very mild	2 (13%)	7 (39%)	0	9 (26%)
Moderate	4 (27%)	6 (33%)	0	10 (29%)
Severe/very severe	9 (60%)	2 (11%)	0	11 (32%)
Number of years since diagnosis				
Mean	3.8	11.2	50.0	9.1
SD	4	14	--	13
95% CI	1-6	4-19	--	4-12
Education¹				
No formal qualification	1 (7%)	2 (12%)	0	3 (9%)
GCSE/A' Level	6 (40%)	9 (53%)	0	15 (45%)
University degree/postgraduate	8 (53%)	6 (35%)	1 (100%)	15 (45%)

Note:

1. One adult with coeliac disease did not state their level of education.

Four of the 34 subjects with CD reported following another special diet in addition to the GFD. One adherent participant reported following a special diet for diabetes and another followed a low fibre diet. The two participants who followed 'other' special diets (one potassium-regulated diet and the other a nut-free diet) both reported partial-adherence to a GF diet.

In this section I showed that participants were recruited with a range of characteristics in terms of age, gender, level of education, experience of CD

symptoms and length of time since diagnosis. Only one non-adherent adult with CD and one non-White British participant with CD were recruited. Healthcare professionals from a range of professions and with different levels of experience of working with coeliac patients were recruited.

4.4 Brainstorming

Table 4.4 shows that the characteristics of participants who completed brainstorming were similar to the total sample population with regards to age, gender, ethnicity, geographical location and education. Sixty-nine of the 73 recruited participants took part in brainstorming. One partially-adherent male adult with CD, one partially-adherent female with CD and two healthcare professionals (Practice nurse and a dietitian) did not take part in brainstorming. These four participants had requested to complete brainstorming remotely by post or e-mail, but they did not return any brainstormed data. All household members who were recruited for the study participated in brainstorming.

Table 4.4 A comparison of the characteristics of the participants who completed brainstorming with those who did not complete brainstorming

	Participants who completed brainstorming (n=69)	Participants who did not complete brainstorming (n=4)	All participants (n=73)
Age (years) ¹			
Mean	55	58	55
SD	15	14	15
95% CI	51-59	39-77	51-59
Gender			
Male	21 (30%)	1 (25%)	22 (30%)
Female	48	3	51
Ethnicity ¹			
White British	68 (99%)	2 (100%)	54 (98%)
Other	1	0	1
Geographical location			
Norfolk	50 (72%)	2 (50%)	52 (71%)
Outside Norfolk	19	2	21
Education ^{1,2}			
No formal qualification	6 (12%)	0	6 (11%)
GCSE/A' level	19 (37%)	2 (100%)	21 (39%)
University level	27 (52%)	0	27 (50%)

Notes:

1. Data were not collected on age, ethnicity and education for healthcare professionals.
2. One adult with CD did not provide information on their level of education.

Fifty-three percent (n=18) of the adults with CD attended group brainstorming sessions, 41% (n=14) completed brainstorming remotely, returning their statements by post or e-mail. Forty-three per cent (n=9) of household members completed brainstorming in group sessions and 57% (n=12) brainstormed remotely. All healthcare professionals were invited to brainstorm remotely by post or e-mail. Table 4.5 shows the number of adults with CD and household members who attended each group brainstorming sessions. Five group brainstorming sessions were held for adults with CD and three group brainstorming sessions were held for household members. The group sizes were small, ranging from one to seven participants.

Table 4.5 The number of participants attending the group brainstorming session

	Adults with coeliac disease (n=18)	Household members (n=9)
Session 1	7 (39%)	4 (44%)
Session 2	5 (28%)	N/A ¹
Session 3	3 (17%)	3 (33%)
Session 4	2 (11%)	N/A ¹
Session 5	1 (6%)	2 (22%)
Total	18 (100%)	9 (100%)

Note:

1. N/A indicates that a brainstorming session was not held for adult household members at the same time as the adults with CD brainstorming sessions.

All 15 adherent participants completed brainstorming (13 attended group sessions and two brainstormed remotely). Eleven partially-adherent adults with CD brainstormed remotely and five attended group sessions. The one non-adherent participant recruited for this study completed brainstorming remotely.

Table 4.6 is a comparison of the characteristics of adults with CD and household members who attended group brainstorming and those who brainstormed remotely. All participants who attended the group brainstorming sessions lived in Norfolk. Adults with CD and household members who brainstormed remotely were on average 6 years younger than those who attended group brainstorming. The percentage of male participants was higher in the group of adults with CD and household members who attended group brainstorming sessions (44%) compared to those who brainstormed remotely (35%).

Table 4.6 Characteristics of participants with coeliac disease and household members who attended group brainstorming sessions and those who brainstormed remotely.

	Adults with coeliac disease and household members who attended group brainstorming sessions (n=27)	Adults with coeliac disease and household members who brainstormed remotely (n=26)	All adults with coeliac disease and household members who brainstormed (n=53)
Age (years) ¹			
Mean	58	52	55
SD	15	15	15
95% CI	52-64	45-58	51-59
Gender			
Male	12 (44%)	9 (35%)	21 (40%)
Female	15	17	32
Ethnicity ²			
White British	26 (96%)	26 (100%)	52 (98%)
Other	1	0	1
Geographical location			
Norfolk	27 (100%)	19 (73%)	46 (87%)
Outside Norfolk	0	7	7
Education ²			
No formal qualification	3 (11%)	3 (12%)	6 (12%)
GCSE/A' Level	9 (33%)	10 (40%)	19 (37%)
University level	15 (56%)	12 (48%)	27 (52%)

Notes:

1. One male household member who completed group brainstorming did not provide information about his age.
2. One adult with coeliac disease did not provide information on their level of education.

Group brainstorming sessions were facilitated by one of four members of the research team (HF, KD, SS, AM) with an assistant present when available to write notes. None of the sessions ran over the scheduled two hours, however, some of the sessions ended up to 30 minutes early when participants felt they had exhausted all their ideas and the facilitator had used all the prompts (Appendix 31) as necessary.

Table 4.7 shows that group and remote brainstorming yielded 903 statements. Adults with CD generated approximately half of all the statements (n=454).

Table 4.7 The number of statements generated during group and remote brainstorming sessions by each stakeholder group.

	No. of statements generated during group brainstorming	No. of statements generated during remote brainstorming	All
Adults with CD	313 (64%)	141 (34%)	454 (50%)
Household Members	176 (36%)	102 (25%)	278 (31%)
Healthcare Professionals	N/A ¹	171 (41%)	171 (19%)
All	489 (100%)	414 (100%)	903 (100%)

Note:

1. N/A indicates that healthcare professionals did not attend group brainstorming sessions.

4.5 Statement reduction and preparation for prioritisation and clustering.

The maximum number of statements that can be analysed using Ariadne concept mapping software is 98. The aim of statement reduction was for the research team to reduce the full set of 903 statements to a final set of 98 or fewer statements which represented the full range of statements contained in the original set. Through discussions held over two sessions, the research team reduced the 903 statements to a final set of 91. This was achieved by synthesising identical and similar statements and eliminating obscure or irrelevant statements. The final set of 91 statements is listed in Appendix 34.

Statement reduction session 1

The research team were presented with the full set of 903 statements printed on individual pieces of coloured paper. Three colours of paper were used to represent each stakeholder group. Similar statements were placed into piles and duplicates, or statements that were very similar, were synthesised. Statements that were perceived to be obscure or irrelevant were eliminated. The original words of participants were preserved whenever possible, rather than translating all statements into our own words. The occasional disagreements between research team members were resolved through discussions until a consensus was reached. By the end of the first of the two statement reduction sessions, the research team had reduced the full set of 903 statements down to a set of 161 statements.

Statement reduction session 2

The 161 statements from the first statement reduction session were printed on individual pieces of paper with the original synthesised statements attached. The process of statement synthesis and elimination was repeated by the research team and discussions took place in order for consensus to be reached on the final set of 91 statements (Appendix 34). All members of the research team agreed that further statement reduction could result in loss of meaning and loss of representation of some of the issues raised during brainstorming.

4.6 Prioritising and clustering the statements

Eleven adults with CD and five adult household members attended a group session at UEA where they completed both the prioritising and clustering tasks. All other participants who completed the prioritising and clustering tasks did so remotely.

Group prioritising and clustering sessions were facilitated by the Chief Investigator (HF). No assistant was required, however, one session was observed by another member of the research team (KD). The combined prioritising and clustering group sessions lasted no more than two hours with a 15 minute refreshment break half way through. As prioritising and clustering are individual tasks, participants were free to leave as soon as they had finished. Most participants took almost the full two hours to complete the two tasks and none ran over. Participants spent roughly an equal amount of time on each of the two tasks, completing the prioritisation task before the refreshments break.

Fifty-four participants completed the prioritising task and 52 completed the clustering task. Six male and three female participants with CD (four adherent, four partially-adherent; and one non-adherent) did not participate in prioritising the statements. Seven male and four female adults with CD (four adherent, six partially-adherent and one non-adherent) did not take

part in clustering the statements. Two male and four female household members and four female healthcare professionals did not participate in prioritising or clustering the statements. Four healthcare professionals did not complete prioritising or clustering (One practice nurse, a community dietitian, an advanced gastroenterology dietitian and a specialist dietitian).

Four female and one male household members did not complete the prioritising or clustering task. With the exception of two partially-adherent adults with CD (one male; one female), all participants who prioritised the statements went on to complete the clustering task. Tables 4.8 and 4.9 show comparisons of the characteristics of participants who prioritised and clustered the statements with those who did not complete these two tasks.

Table 4.8 A comparison of the characteristics of participants who completed the prioritisation task and those who did not.

	Participants who completed prioritising (n=54)	Participants who did not complete prioritising (n=19)	All participants (n=73)
Age (years) ^{1,2}			
Mean	52	63	55
SD	14	14	15
95% CI	47-56	55-71	51-59
Gender			
Male	14 (26%)	8 (42%)	22 (30%)
Female	40	11	51
Ethnicity ¹			
White British	53 (98%)	19 (100%)	54 (98%)
Other	1	0	1
Geographical location			
Norfolk	40 (74%)	12 (63%)	52 (71%)
Outside Norfolk	14	7	21
Education ^{2,3}			
No formal qualification	1 (2.5%)	5 (36%)	6 (11%)
GCSE/A' Level	15 (37.5%)	6 (43%)	21 (39%)
University level	24 (60%)	3 (21%)	27 (50%)

Notes:

1. One adult household member who completed the prioritising task did not state his year of birth.
2. Healthcare professionals were not asked their year of birth or level of education.
3. One male adult with coeliac disease who did not complete prioritising did not state his level of education.

Table 4.9 Characteristics of participants who completed the clustering task and those who did not.

	Participants who completed clustering(n=52)	Participants who did not complete clustering (n=21)	All (n=73)
Age (years) ^{1,2}			
Mean	52	63	55
SD	14	14	15
95%CI	47-56	55-70	51-59
Gender			
Male	13 (25%)	9 (43%)	22 (30%)
Female	39	12	51
Ethnicity ¹			
White British	51 (98%)	21 (100%)	54 (98%)
Other	1	0	1
Geographical location			
Norfolk	39 (75%)	13 (62%)	52 (71%)
Outside Norfolk	13	8	21
Education ^{2,3}			
No formal qualification	1 (3%)	5 (31%)	6 (11%)
GCSE/A' Level	13 (34%)	8 (50%)	21 (39%)
University level	24 (63%)	3 (19%)	27 (50%)

Notes:

1. One adult household member who completed the clustering task did not state his year of birth.
2. Healthcare professionals were not asked their year of birth or level of education.
3. One male adult with coeliac disease who did not complete clustering did not state his level of education.

The Ariadne manual (Talcott, 1995) advises that the maximum number of clusters that should be generated by a participant is 25. None of the participants in this study generated more than 25 clusters. The mean number of clusters generated by all participants was 10 (range 5 to 18 clusters). Table 4.10 shows the mean number of clusters generated by participants in each stakeholder group.

Table 4.10 The mean number of clusters generated by each stakeholder group.

	Adults with CD (n=23)	Household members (n=15)	Healthcare professionals (n=14)	All participants (n=52)
Number of statements generated				
Mean	10	8	12	10
Range	5-16	6-17	6-18	5-18

4.7 Data analysis

4.7.1 Data entry

Data were entered into the Ariadne software for analysis. Data inputting was carried out on two separate computers by the same data inputter and no data entry errors were identified.

The Ariadne manual states:

‘Participants are not limited in the number of piles (clusters) they form, nor in the number of statements in each pile. However, Ariadne sets an upper limit of 25 clusters per participant, each of 40 statements per cluster’.

(Talcott, 1995)

Based on the amount of overlap between the 91 statements, the research team felt it to be unlikely that the participants would generate as many as 25 clusters. The highest number of clusters generated was 18. It later became

evident that the guidance in the Ariadne manual (Talcott, 1995) was incorrect and the maximum number of statements that can be entered into the Ariadne software is 12, not 25. Ten participants generated more than 12 clusters and all of these data could not be entered into Ariadne. I sought advice from the software designer who advised me to eliminate the clusters containing the least number of statements (Appendix 35).

There were several instances where participants made errors when completing data recording sheets (Appendices 36 and 37) for the prioritising and clustering tasks. These errors, and the actions taken to resolve them, are summarised (Appendix 38). As concept mapping is used to group statements for similarity, it was necessary for me to exclude groups of unrelated statements that participants had grouped as a 'miscellaneous' cluster. Appendix 38 includes details of the removed 'miscellaneous' clusters and also the details of when more than 12 clusters were generated.

A total of 32 clusters were removed for those participants who generated more than the permitted number of 12 clusters. These 32 clusters were small and contained a total of 74 statements. The statement that appeared in the deleted clusters the most frequently was '*...if they are female*' and this statement was deleted for five participants who generated more than 12 clusters. The statement '*...if there were more resources for people from ethnic minorities*' was deleted for four participants who generated more than 12 clusters. There were six statements that appeared three times in the deleted clusters:

'...if they have supportive work colleagues'
'...if there was a wider range of Asian gluten-free products available'
'...if speciality gluten-free foods tasted nicer'
'...if speciality gluten-free food was not so high in calories and sugar'
'...if labels stated "Produced in a factory where gluten is used" rather than
"May contain gluten" so you can assess the level of risk'
'...if gluten-free biscuits were made available when gluten-inclusive biscuits
are served with tea and coffee at work'

The remainder of the removed statements appeared only one or two times each in the deleted clusters. According to the software owner, the impact of removing these small clusters should have little or no effect on the overall results.

4.7.2 Concept maps

A sequence of analyses involving multidimensional scaling and cluster analysis were used to produce the concept maps. Two types of concept maps were produced: point map and a cluster rating map. The maps represent the overall conceptual framework for the study (Kane & Trochim, 2007).

Figure 5.1 shows a point map depicting the relationships between the 91 statements sorted by participants. Each data point represents a single statement and the distance between data points on the map represents how often the statements were grouped together by participants in the clustering

task. Statements that were grouped together frequently appear close together, whereas statements that were grouped together less frequently are spatially further apart. The data points on this map are numbered and the statements that each of the numbers relates to are listed in Appendix 34.

The steering group met to decide on names for the axes on the point and cluster maps. The axes were assigned names based on the content of the statements positioned close to them. Examination of the statements in relation to the X axis indicated that statements to the right related to society's responsibility and on the left, individual responsibility. This axis could be viewed as a locus of control continuum from personal responsibility to societal responsibility. The Y axis represents psychological to practical issues relating to adherence to the gluten-free diet. The X and Y axes divide the concept map into four quadrants.

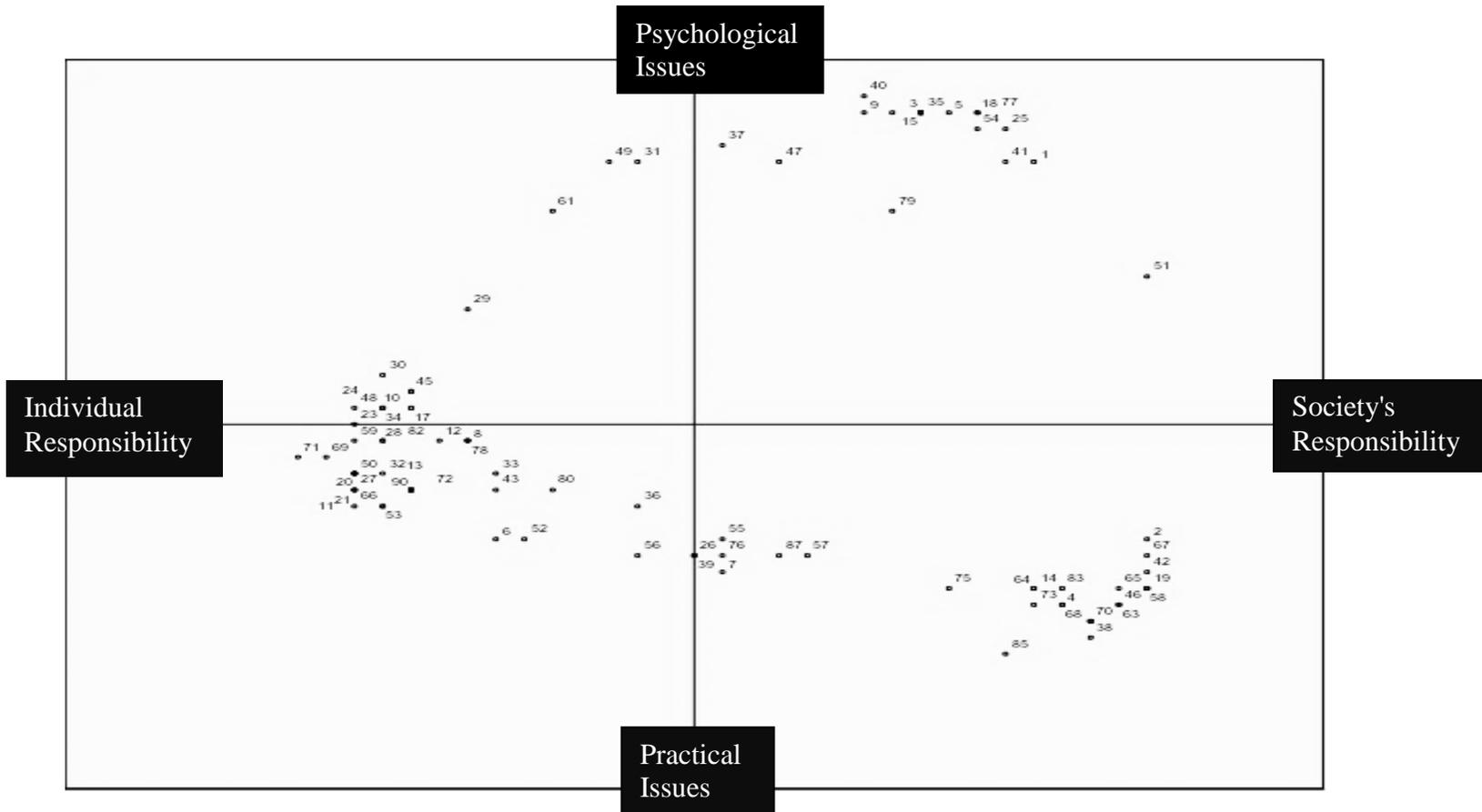


Figure 4.1 - Point map for all participants

Cluster analysis groups together the individual data points into clusters of statements that represent similar concepts. In deciding on the most appropriate number of clusters to have on the concept map, the research team tried reducing and increasing the number of clusters from the default number of eight. I mapped how the statements moved as the number of clusters was reduced or increased. Appendix 39 shows that when the cluster number was increased or decreased the amount of movement of the statements was small, often with just one statement moving to form a new single statement cluster as the number of clusters was increased. After discussion, the panel reached a consensus of a 13-cluster solution and a title for each cluster was agreed on. When looking at a 12-cluster solution, it was decided that this number of statements was not appropriate as clusters 5 and 6 from the 13-cluster solution were grouped together to form one single cluster and it was felt that the statements in the two clusters did not really belong together.

The 13-cluster concept map is shown in Figure 4.2. This cluster rating map provides an overview of the main themes of this study and the 13 clusters are ranked in order of importance from 1 (most important) to 13 (least important). The importance ranking of each cluster is based on the mean prioritisation scores of all statements contained within the cluster.

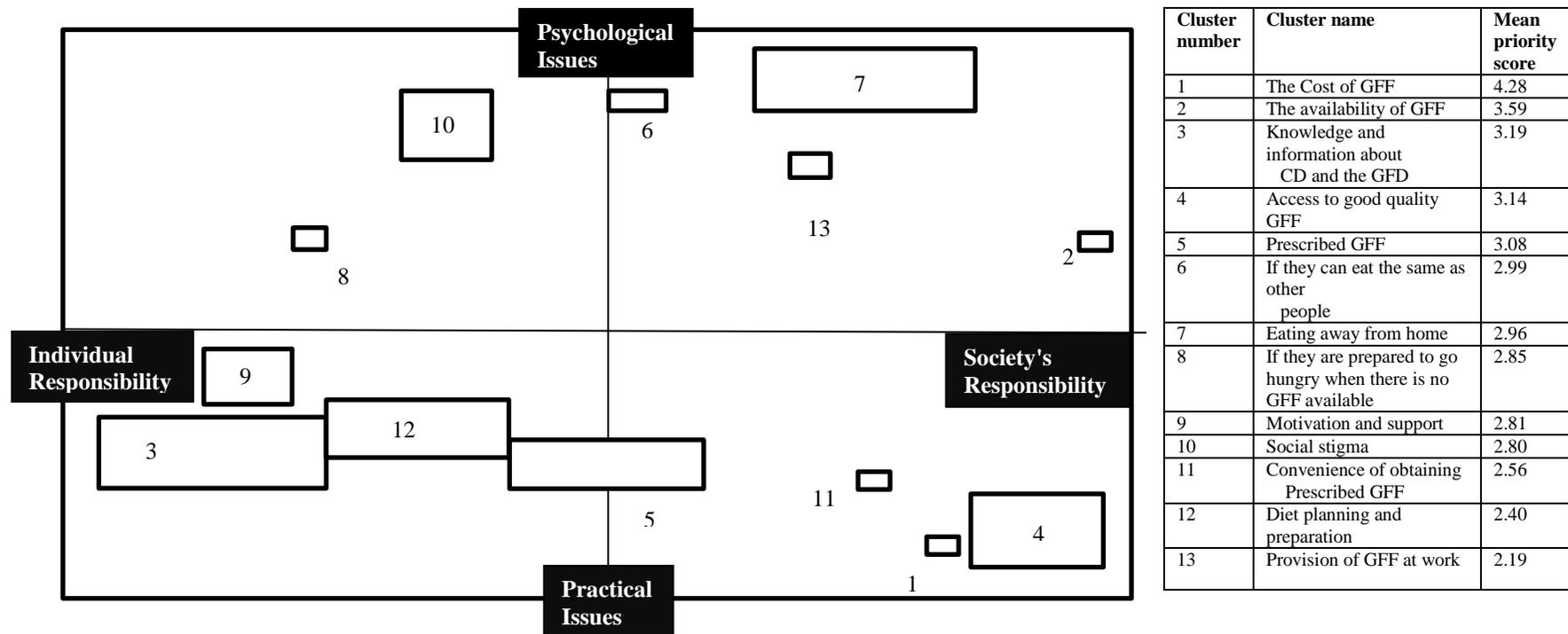


Figure 4.2 Cluster map

Note:

The clusters are numbered from 1 to 13 in order of priority (Cluster 1 = most important; Cluster 13= least

4.7.3 Analysis of the clusters

Tables 4.11 to 4.23 show the set of statements included in each cluster in order of mean preference score. Preference scores are between 1 (least important) to 5 (most important).

Cluster 1 – The cost of gluten-free food

The cost of gluten-free food (GFF) emerged as the most important theme relating to adherence to a GFD with a mean prioritisation score of 4.28 (Table 4.11). This 'cluster' contains just one single statement. The issue of the high cost of GFF was mentioned during every group brainstorming session and all except for three adults with CD, two household members and two healthcare professionals who brainstormed remotely generated a statement about the high cost of GFF. This cluster falls within the practical/society's responsibility quadrant on the concept map.

Table 4.11 Cluster 1 – The cost of gluten-free food.

Cluster 1: The cost of gluten-free food (mean preference score: 4.28)	
Statement/s:	Mean preference score
...if gluten-free food was not so expensive.	4.28

Cluster 2 – The availability of GF sandwiches

The availability of GF sandwiches was the second most important theme to emerge from this study in relation to adherence to a GFD. As with cluster 1, cluster 2 contains one single statement (Table 4.12).

Table 4.12 Cluster 2 – The availability of gluten-free sandwiches

Cluster 2: The availability of gluten-free sandwiches (mean preference score: 3.59)	
Statement/s:	Mean preference score
...if there was more availability of gluten-free sandwiches.	3.59

The statement in cluster 2 was synthesised from four of the original statements relating to GF sandwiches:

- 'More GF sandwiches everywhere!' – adult with CD
- 'If sandwich companies provided at least one choice made with GF bread' – adult with CD
- 'If there could be more variety of gluten-free foods in the fast food sections at supermarkets...snacks, sandwiches etc. In fact, there are very few outlets which provide sandwiches' – adult with CD
- 'Easy access to retail GF sandwiches and snacks' – Healthcare professional

Cluster 3 – Knowledge and information about CD and the GFD

The third highest rated theme to emerge was knowledge and information about CD and the GFD. This cluster contains 24 statements as shown in Table 4.13. The statements in the cluster relate not only to the knowledge of adults with CD but also to that of healthcare professionals.

Table 4.13 Cluster 3 - Knowledge and information about coeliac disease and the gluten-free diet

Cluster 3: Knowledge and information about coeliac disease and the gluten-free diet	
(mean preference score: 3.19)	
Statements:	Mean preference score
...if they understand the health consequences of not sticking to a gluten-free diet.	4.39
...if they received more advice about coeliac disease and the gluten-free diet around the time of diagnosis.	3.94
...if they have a better knowledge of the gluten-free diet.	3.83
...if they have appropriate follow-up care with quick and easy access to dietitians.	3.81
...if they experience symptoms when they consume gluten.	3.80
...if they have immediate access to a dietitian at the time of diagnosis.	3.74
...if they have a supportive GP.	3.67
...if healthcare professionals knew more about coeliac disease and the gluten-free diet.	3.65
...if they have access to information about coeliac disease.	3.60
...if they believe the diagnosis.	3.31
...if GPs were better informed about what patients are allowed on prescription.	3.24
...if they don't have additional special dietary requirements as well as a gluten-free diet (e.g. lactose-free, vegetarian or diabetic).	3.23
...if there was an expert point of contact for patients and healthcare professionals.	3.20
...if they join a coeliac support group (e.g. Coeliac UK).	3.11
...if they have already been following a gluten-free diet for a long time.	3.07
...if they can cook gluten-free meals from scratch.	3.00
...if they have access to another person with coeliac disease who has more experience.	2.98
...if they are given a blood test to confirm adherence.	2.93
...if separate cooking utensils are used in the preparation of gluten-free food at home.	2.56
...if they use the internet to get information about coeliac disease and the gluten-free diet.	2.39
...if TV cookery programmes included gluten-free cooking.	2.38
...if they are well educated.	2.33
...if there was more psychological support available.	2.31
...if there was a mobile phone app to advise them on where to find gluten-free food.	2.20

Cluster 4 – Access to good quality gluten-free food

The fourth most important theme related to access to good quality GFF. The statements within this cluster cover a range of issues including food labelling, palatability of GFF and availability of GFF. Table 4.14 lists the

statements in this cluster. I included the term '*access*' in the cluster label with the intention that this would cover GF labelling issues, the availability of GFF and access to free samples of GFF. The '*good quality*' part of this cluster heading covers the taste and texture of GFF, nutrient composition and the shelf-life of GFF.

Table 4.14 Cluster 4 - Access to good quality gluten-free food

Cluster 4: Access to good quality gluten-free food (mean preference score: 3.14)	
Statements:	Mean preference score
...if gluten-free bread was of the same quality in taste and texture as gluten-inclusive bread.	4.02
...if there was a universal gluten-free logo on food packaging used internationally on all suitable foods (such as the crossed grain logo).	3.94
...if there was a wider variety of gluten-free products in shops and supermarkets.	3.85
...if there was more gluten-free food available in all food shops and supermarkets.	3.78
...if gluten ingredients were not added to foods where you wouldn't expect to find gluten (such as ice cream or grated cheese).	3.72
...if speciality gluten-free foods tasted nicer.	3.67
...if there was more availability of savoury gluten-free snacks and not just sugary cakes and biscuits.	3.42
...if a wider range of non-traditional gluten-free breads were available (e.g. tortillas, chapatis, pita bread).	3.15
...if supermarket discount offers included gluten-free products.	3.13
...if gluten-free food was easier to find on the shelves when shopping in supermarkets.	3.13
...if a wider range of gluten-free ready meals were available.	3.02
...if there was more consistency between similar products regardless of brand or pack size (e.g. some brands of cornflakes are GF but others are not).	2.94
...if speciality gluten-free food was not so high in calories and sugar.	2.87
...if food manufacturers didn't change their ingredients so often.	2.79
...if they are able to try free samples of gluten-free products before buying (e.g. at roadshows).	2.78
...if labels stated "produced in a factory where gluten is used" rather than "may contain gluten" so you can assess the level of risk.	2.70
...if speciality gluten-free food had a longer shelf-life.	2.46
...if gluten-free products were kept next to similar gluten-inclusive items in supermarkets (e.g. gluten-free bread in the bread section, rather than the 'free from' section).	2.20
...if there was a wider range of Asian gluten-free products available.	2.06

Cluster 5 – Prescribed gluten-free food

Cluster 5 (Table 4.15) covers a range of issues mostly relating to prescribed GFF. During the group brainstorming sessions, there were several discussions about the recent cut-backs in prescribed GFF and inconsistencies between what GPs are willing to prescribe.

Table 4.15 Cluster 5 - Prescribed gluten-free food

Cluster 5: Prescribed gluten-free food (mean preference score: 3.08)	
Statements:	Mean preference score
...if they receive gluten-free food on prescription.	3.78
...if they get a sufficient amount of gluten-free food on prescription.	3.65
...if a wider range of gluten-free products were available on prescription (e.g. not just bread and flour).	3.54
...if gluten-free food was exempt from the prescription charge.	3.30
...if people on a low income had additional help with the cost of gluten-free food.	3.02
...if Coeliac UK's Food and Drink Directory included a wider range of products and brands.	2.81
...if they were given a personal amount of money to support the buying of gluten-free food, rather than getting it on prescription.	2.43
...if there were more resources for people from ethnic minorities.	2.11

Two additional statements also related to prescribed GFF, but these statements were grouped into different clusters:

"...if GPs were better informed about what patients are allowed on prescription" – Cluster 3: Knowledge, information and education.

"...if they didn't have to go to a pharmacy to collect prescribed gluten-free food when it should be obtainable from shops or supermarkets" – Cluster 11: Convenience of obtaining prescribed GFF.

Cluster 6 – If they can eat the same as other people

Cluster 6 (Table 4.16) contains two statements that represent eating in a social setting. The two statements in cluster 6 appear somewhat dissimilar, although they were grouped together frequently by participants. The Ariadne manual (Talcott, 1995) advises that, when choosing a name for a cluster, it is important to focus on the statements with the highest priority

rating within that cluster. Therefore, we agreed to label this cluster according to the most highly rated of the two statements *'if they can eat the same (gluten-free) food as other people'*. Both statements represent issues relating to eating in social situations.

Table 4.16 Cluster 6 - If they can eat the same as other people

Cluster 6: If they can eat the same as other people (mean preference score: 2.99)¹	
Statements:	Mean preference score
...if you can eat the same (gluten-free) food as everyone else when eating socially.	3.17
...if hospitals were better at providing gluten-free food.	2.85

Note:

The cluster preference score of 2.99 is less than the average of the two statement preference scores. The reason for this is that the number of participants who prioritised each statement differed and this was taken into account when calculating the cluster preference score. The statement *'...if hospitals were better at providing gluten-free food'* was ranked by more participants than the other statement in this cluster. As the statement that was ranked by more participants had a lower mean preference score, the overall cluster prioritisation score is lower than the average of the two statements.

The original statements that were synthesised to produce the statement *'...if you can eat the same (gluten-free) food as everyone else when eating socially'* were:

'...if you can eat identical (gluten-free) food to everyone else'

'...if people with CD are given the same types of food as non-coeliacs when eating socially'

'...if meals eaten in social situations could include gluten-free substitutes which are similar to the foods eaten by other people rather than having to eat a different type of meal'

The original statements that were synthesised to produce the statement '*if hospitals were better at providing GFF*' were:

'...if hospital restaurants could offer gluten-free meals'

'...if hospitals provided GFFF without having to inform them in advance'

'...if hospitals were better at providing GFF'

'...if hospitals were better at providing GFF for patients with CD'

Cluster 7 – Eating away from the home

The main themes arising from cluster 7 'Eating away from the home' (Table 4.17) relate to eating in restaurants and availability of GFF when travelling. This was the seventh most important theme to emerge from this study. During group brainstorming sessions, stories were told by participants about difficulties and inconsistencies experienced when travelling by air. We were told that some airlines gave passengers the option to select a GF meal, however, the airline couldn't guarantee that the passenger would get it. One participant who had ordered an on-flight GF meal was told during the flight that there were no GF options available, however, other passengers were given GF items such as fresh fruit.

Table 4.17 Cluster 7 - Eating away from the home

Cluster 7: Eating away from the home (mean preference score: 2.96)	
Statements:	Mean preference score
...if restaurants were better at labelling gluten-free options on their menus.	4.15
...if there were more gluten-free options when eating out.	4.00
...if staff working in restaurants/cafes were more knowledgeable about coeliac disease and the gluten-free diet.	3.81
...if food outlets were more careful to avoid gluten contamination.	3.48
...if they take their own gluten-free food with them when eating away from home.	3.43
...if there was legislation to ensure all food outlets provide gluten-free options.	3.33
...if they can get hold of gluten-free food when travelling abroad.	3.07
...if they are allowed to take gluten-free food with them when travelling abroad.	2.87
...if restaurants and carveries provided gluten-free gravy.	2.83
...if motorway services provided gluten-free food.	2.34
...if airlines were better at providing gluten-free food during flight.	2.30
...if there was more availability of gluten-free fish and chips.	2.26
...if pubs stocked gluten-free beers and lagers.	2.20
...if airports were better at providing gluten-free food.	2.17
...if gluten-free food was more available when travelling by train.	2.09

Cluster 8 - If they are prepared to go hungry when there is no gluten-free food available

Table 4.18 shows a single-cluster statement relating to going hungry when gluten-free food is not available.

Table 4.18 Cluster 8 - If they are prepared to go hungry when there is no gluten-free food available

Cluster 8: If they are prepared to go hungry when there is no gluten-free food available (mean preference score: 2.85)	
Statement/s:	Mean preference score
...if they are prepared to go without/go hungry rather than eat gluten when there are no gluten-free foods available.	2.85

Cluster 9 – Motivation and support

Cluster 9 (Table 4.19) relates to self-determination, social support and motivation. This cluster falls in the psychological and individual responsibility quadrant. Determination and having a positive outlook were believed to help patients stick to a GFD. In addition, the support and motivation from others was also important in relation to adherence. The statement "*if they are female*" was ranked lowest out of the 91 statements by all three participant groups. This statement was generated by a mother and daughter during a group brainstorming session. The statement arose when the mother and daughter were discussing how difficult they thought it would be for the father of their household to follow a GFD if he had CD and they believed this would be true for other males. During the follow-up group sessions with different sets of participants, it became clear that most people did not agree with this statement and several participants couldn't understand how it could be easier for females to follow a GFD compared to males.

Table 4.19 Cluster 9 - Motivation and support

Cluster 9: Motivation and support (mean preference score: 2.81)	
Statements:	Mean preference score
...if they are determined to stick to the gluten-free diet and resist temptations.	3.91
...if they have a positive outlook and focus on what they can eat, rather than what they can't.	3.81
...if they have supportive family and friends.	3.57
...if friends and family are supported/educated so they can reliably cater for them.	3.51
...if the public had a better understanding of coeliac disease and the gluten-free diet.	3.20
...if other household members eat gluten-free.	2.68
...if other people encouraged them to stick to the gluten-free diet.	2.67
...if they have supportive work colleagues.	2.38
...if they have someone to speak up for them on their behalf.	2.11
...if someone cooks for them.	1.74
...if they are female.	1.37

Cluster 10 – Social stigma

Cluster 10 is a three-statement cluster relating to issues about not being made to feel embarrassed or different when eating in public (Table 4.20). Although the cluster ranking is lower than the previous clusters, this was a theme that came up many times during group brainstorming sessions where people told stories about their experiences of embarrassment and being made to feel like they were making an unnecessary fuss by asking for GFF. The third statement in this cluster relates to the pressure felt to accept gluten-inclusive food and drink when it is offered, rather than making a fuss or feeling rude by rejecting the kindness of the person offering it. This issue arose a few times during the group sessions and participants had mixed opinions about whether they should worry about what other people think when they ask for GFF. Some participants were assertive and said they had no problem with 'making a fuss' whereas one person said she had hidden

away during a wedding because the GFF she had requested wasn't available and she didn't want the bride's mother to notice that she wasn't eating anything.

Table 4.20 Cluster 10 – Social stigma

Cluster 10: Social stigma (mean preference score: 2.8)	
Statements:	Mean preference score
...if they are confident and not embarrassed by having to ask for gluten-free food when eating out.	3.17
...if they are not made to feel different when eating socially.	3.06
...if there was less social pressure to accept any gluten-inclusive food or drink you are offered.	2.15

Note:

The cluster preference score of 2.8 is less than the average of the three statement preference scores. The reason for this is that the number of participants who prioritised each statement differed and this was taken into account when calculating the cluster preference score. The statement with the highest mean preference score was ranked by more participants than the other two statements. As the statement that was ranked by more participants had a higher mean preference score, the overall cluster prioritisation score is higher than the average of the three statements.

Cluster 11 – Convenience of obtaining prescribed gluten-free food

Cluster 11 is another single-statement cluster (Table 4.21). As discussed previously, this statement may have fit better within cluster 5 – '*Prescribed GFF*'. During group brainstorming sessions, suggestions were made for a voucher scheme whereby patients could be provided with vouchers to buy GFF in shops and supermarkets, rather than going to a pharmacy to pick up prescribed GF food. Some participants felt the prescription system was out-of-date now that GFF is more widely available in the shops.

Table 4.21 Cluster 11 Convenience of obtaining prescribed gluten-free food

Cluster 11: Convenience of obtaining prescribed gluten-free food (mean preference score: 2.56)	
Statement/s:	Mean preference score
...if they didn't have to go to a pharmacy to collect prescribed gluten-free food when it should be obtainable from shops or supermarkets.	2.56

Cluster 12 – Diet planning and preparation

We took some time to decide on the name of cluster 12. On first glance the statements listed in Table 4.22 did not appear to be clearly related, however, the four statements in this cluster are related to planning and preparing a GFD. Coeliac UK's Food and Drink Directory was discussed during brainstorming sessions and participants often use it when shopping for GFF. Members receive a copy of the Food and Drink Directory when they join Coeliac UK and they are sent a revised edition annually. Some participants told us that they did not have a copy of the Directory because they had not joined Coeliac UK. One participant cancelled her membership to Coeliac UK when the annual membership fee (currently £20) was introduced and, therefore, she no longer received the Food and Drink directory.

Table 4.22 Cluster 12 - Diet planning and preparation

Cluster 12: Diet planning and preparation (mean preference score: 2.40)	
Statements:	Mean preference score
...if they use Coeliac UK's Food and Drink Directory.	2.98
...if more gluten-free recipes were available.	2.53
...if it was free to join Coeliac UK.	2.22
...if they have plenty of freezer space.	1.87

Cluster 13 – Provision of GFF at work

The cluster perceived to be the least important related to the provision of GF food at work (Table 4.23). Cluster 13 is a single-statement cluster. **Table**

4.23 Cluster 13 - Provision of gluten-free food at work

Cluster 13: Provision of gluten-free food at work (mean preference score: 2.19)	
Statements:	Mean preference score
...if gluten-free biscuits were made available when gluten-inclusive biscuits are served with tea and coffee at work.	2.19

4.7.4 Comparison between the three stakeholder groups

Differences between the cluster preference ratings for each stakeholder group were tested for statistical significance using the Kruskal-Wallis test. No statistically significant difference was found in 12 out of the 13 clusters. Cluster 2 (the availability of GF sandwiches) shows a significant difference between the mean preference ratings for stakeholder groups (Table 4.24). A follow-up test was used to make pairwise comparisons between stakeholder groups for cluster 2 to establish which of the three groups had statistically using the Mann-Whitney U test and the results of this are shown in Table 4.25. Significant differences in mean priority scores for cluster 2 were found

between adults with CD and healthcare professionals ($P=0.02$) and also between household members and healthcare professionals ($P=0.04$). In both instances, the mean preference rating for healthcare professionals was significantly lower than the other two stakeholder groups. No other statistically significant differences were found between the stakeholder group cluster preferences.

Cluster 8 (...if they are prepared to go hungry when there is no gluten-free food available) showed a lower mean priority rating for healthcare professionals compared to the other two stakeholder groups, however, the difference was not statistically significant ($P=0.06$) (Table 4.24).

Table 4.24 Kruskal-Wallis analysis comparing the mean preference scores for the 13 clusters for three stakeholder groups.

Cluster	All participants	Adults with coeliac disease	Household members	Healthcare professionals	Kruskal-Wallis test (P-value)
1. The cost of gluten-free food					
Rank:	1	1	1	1	0.66
Mean:	4.28	4.20	4.47	4.21	
SD:	0.97	0.91	0.74	1.31	
95% CI	4.01-4.55	3.82-4.58	4.06-4.88	3.46-4.97	
2. The availability of gluten-free sandwiches					
Rank:	2	2	2	7	0.05
Mean:	3.60	3.84	3.87	2.86	
SD:	1.25	1.21	1.06	1.29	
95% CI	3.25-3.93	3.34-4.34	3.28-4.45	2.11-3.60	
3. Knowledge, information and education					
Rank:	3	5	3	2	0.21
Mean:	3.20	3.11	3.20	3.35	
SD:	0.37	0.32	0.35	0.45	
95% CI	3.10-3.30	2.98-3.24	3.01-3.40	3.09-3.61	
4. Access to good quality gluten-free food					
Rank:	4	4	6	4	0.78
Mean:	3.14	3.18	3.07	3.16	
SD:	0.44	0.40	0.58	0.35	
95% CI	3.02-3.26	3.01-3.34	2.75-3.39	2.95-3.36	
5. Prescribed gluten-free food					
Rank:	5	8	7	3	0.42
Mean:	3.08	3.00	3.05	3.26	
SD:	0.64	0.65	0.66	0.60	
95% CI	2.91-3.26	2.74-3.27	2.69-3.42	2.91-3.61	

Cluster	All participants	Adults with coeliac disease	Household members	Healthcare professionals	Kruskal-Wallis test (P-value)
6. If they can eat the same as other people					
Rank:	6	6	4	11	0.10
Mean:	2.98	3.10	3.20	2.54	
SD:	0.91	0.98	0.73	0.87	
95% CI	2.73-3.23	2.70-3.50	2.80-3.60	2.04-3.04	
7. Eating away from the home					
Rank:	7	7	8	8	0.09
Mean:	2.96	3.08	2.97	2.73	
SD:	0.50	0.57	0.35	0.43	
95% CI	2.82-3.09	2.84-3.31	2.78-3.16	2.48-2.98	
8. If they are prepared to go hungry when there is no gluten-free food available					
Rank:	8	3	5	12	0.06
Mean:	2.85	3.20	3.07	2.00	
SD:	1.56	1.53	1.67	1.24	
95% CI	2.43-3.28	2.57-3.83	2.14-3.99	1.28-2.72	
9. Motivation, support and determination/will power					
Rank:	9	9	9	9	0.48
Mean:	2.81	2.81	2.91	2.70	
SD:	0.49	0.53	0.47	0.43	
95% CI	2.69-2.95	2.60-3.03	2.66-3.17	2.45-2.95	
10. If they are not made to feel different when asking for gluten-free food					
Rank:	10	10	10	6	0.56
Mean:	2.80	2.72	2.73	3.02	
SD:	0.88	0.85	1.08	0.72	
95% CI	2.56-3.04	2.37-3.07	2.14-3.33	2.61-3.44	

Cluster	All participants	Adults with coeliac disease	Household members	Healthcare professionals	Kruskal-Wallis test (P-value)
11. Convenience of obtaining prescribed gluten-free food	11	13	11	5	0.19
Rank:	2.56	2.24	2.60	3.07	
Mean:	1.39	1.30	1.50	1.38	
SD:	2.17-2.94	1.70-2.78	1.77-3.43	2.27-3.87	
95% CI					
12. Diet planning and preparation / Coeliac UK					
Rank:	12	12	12	10	0.46
Mean:	2.40	2.34	2.35	2.57	
SD:	0.56	0.58	0.52	0.57	
95% CI	2.25-2.56	2.10-2.58	2.06-2.64	2.44-2.90	
13. Provision of gluten-free foods at work					
Rank:	13	11	13	13	0.08
Mean:	2.19	2.56	1.93	1.79	
SD:	1.27	1.29	1.16	1.25	
95% CI	1.84-2.53	2.03-3.09	1.29-2.58	1.06-2.51	

Table 4.25 Results of the Mann-Whitney U test for the cluster which showed a significant difference using the Kruskal-Wallis test.

Cluster	All participants	Adults with coeliac disease	Adult household members	Healthcare professionals	Kruskal-Wallis test (P-value)	Mann-Whitney U test (P-values) Pairwise comparisons		
						Adults with coeliac disease and adult household members	Adults with coeliac disease and healthcare professionals	Adult household members and healthcare professionals
Cluster 2 - The Availability of Gluten-free Sandwiches					0.046	0.930	0.024	0.036
Rank:	2	2	2	7				
Mean:	3.60	3.84	3.87	2.86				
SD:	1.25	1.21	1.06	1.29				
95% CI	3.25-3.93	3.34-4.34	3.28-4.45	2.11-3.60				

4.7.5 Synthesis of concept mapping results with systematic review results

In this section I will synthesise the results of my update to the systematic review by Hall *et al.* (2009) (presented in Chapter 2) with the results of this concept mapping study. Tables 4.26 to 4.31 show the results of this synthesis. I have used the same six themes that Hall *et al.* (2009) identified in their systematic review to organise the data.

Sociodemographic factors

The systematic review found very little evidence of an association between sociodemographic factors and adherence to a GFD. None of the concept mapping clusters fit into this category, which provides further evidence that sociodemographic factors are not strongly associated with adherence to a GFD. In the concept mapping study, one statement was generated regarding adherence to a GFD and gender. The statement suggested that it was easier to stick to a GFD if you are female. This statement received the lowest priority rating and several participants expressed their disagreement with this statement. Overall, sociodemographic factors have not been found to be associated with adherence to a GFD.

Table 4.26 Sociodemographic factors.

Data Source	Factor	Factors Associated with Adherence	Factors not associated with adherence	Priority ³
Review	Education	3/9 ¹ (n=1544) ²	6/9 ¹ (n=853) ²	N/A
Review	Age	6/17 ¹ (n=1797) ²	11/17 ¹ (n =4569) ²	N/A
Review	Gender	1/12 ¹ (n=128) ²	11/12 ¹ (n=6551) ²	N/A
Review	Social Class/ socioeconomic status	1/3 ¹ (n=573) ²	2/3 ¹ (n=282) ²	N/A
Review	Urban residence	1/1 ¹ (n=234) ²		N/A
Review	Employment status / Occupation		2/2 ¹ (n=544) ²	N/A
Review	Marital status	1/2 ¹ n=154 ²	1/2 ¹ n=255 ²	N/A
Review	Ethnicity (Caucasians more adherent than Asian)	1/2 ¹ (n=87) ²	1/2 ¹ (n=679) ²	N/A
Review	Smoking	1/1 ¹ N=204 ²		N/A

Notes:

1. This indicates the number of studies that found a particular factor to be associated or not associated with adherence (e.g. three out of nine (3/9) studies showed that education was a factor associated with adherence and six out of nine (6/9) studies showed no association between adherence and education).
2. n= total number of participants (including controls where relevant)
3. Priority relates to the priority rating of the concept mapping clusters. However, none of the concept mapping clusters appear in this table.

Knowledge attitudes and beliefs

The statements that were grouped into the ‘knowledge, information and education’ cluster on the concept map reflect several of the factors included in Hall *et al.*’s (2009) ‘knowledge, attitudes and beliefs’ theme. However the concept mapping cluster included the knowledge of both healthcare professionals and patients, not just patients’ knowledge, which was not highlighted in the systematic review. The closest factor identified from the systematic review in relation to the knowledge of healthcare professionals was ‘Satisfaction with information from health care provider’.

Some of the individual statements from the ‘knowledge, information and education’ concept mapping cluster can be mapped to some of the factors

included in Hall *et al.*'s (2009) 'Healthcare treatment factors', 'Socio-cultural/environmental factors' and 'Quality of life and psychological wellbeing' factors.

Table 4.27 Knowledge, attitudes and beliefs.

Data Source	Factor	Factors Associated with Adherence	Factors not associated with adherence	Priority
Review	Knowledge and understanding of GFD/ GF ingredients	3/3 ¹ (N=719) ²		N/A
Concept map	Knowledge and Information about CD and the GFD	N=73		3
Review	Understanding and use of food labels	2/3 ¹ (N=365) ²	1/3 ¹ (N=234) ²	N/A
Review	Beliefs about harm/consequences from exposure to gluten / belief in cyclical nature of coeliac disease	4/5 ¹ (N=729) ²	1/5 ¹ (n=278) ²	N/A
Review	Personality traits (higher conscientiousness/higher values trait/higher order trait/ higher self-discipline/deliberation) associated with better adherence	1/1 ¹ (N=154) ²		N/A
Review	Intention to adhere	1/1 ¹ (N=390) ²		N/A
Review	Task or emotion oriented coping associated with better adherence	1/1 ¹ (N=390) ²		
Review	Self-efficacy	2/2 ¹ (n=575) ²		N/A
Concept map	Motivation and support	n=73		9
Review	Being prepared and organised	1/1 ¹ (n=278) ²		N/A
Concept Map	Diet planning and preparation	1/1 (n=73)		12
Review	Confidence to ask questions about contamination was associated with better adherence	1/1 ¹ (N=278) ²		N/A
Review	Having trust in others to prepare GFF	1/1 ¹ (N=278) ²		N/A

Notes:

1. This indicates the number of studies that found a particular factor to be associated or not associated with adherence (e.g. four out of five (4/5) studies showed that beliefs about harm/consequences from exposure to gluten / belief in cyclical nature of coeliac disease were factors associated with adherence and one out of five (1/5) studies showed no association between adherence and beliefs about harm/consequences from exposure to gluten / belief in cyclical nature of coeliac disease).
2. n= total number of participants (including controls where relevant)
3. Priority relates to the priority rating of the concept mapping clusters. Cluster numbers 3 and 12 from the concept map appear in this table.

Illness and symptom factors

None of the concept mapping clusters appeared to fit within this theme and the systematic review identified that most factors were not supported or only had small studies to support them. The association between body weight and adherence to a GFD was not identified in the concept mapping study. Some of the factors are covered by individual statements contained within the concept mapping clusters.

Table 4.28 Illness and symptom factors.

Authors	Factors	Factors Associated with Adherence	Factors not associated with adherence	Priority ³
Review	Time since diagnosis	1/5 ¹ (N=76) ²	4/5 ¹ (N=1090) ²	N/A
Review	Age at diagnosis	3/9 ¹ (N=369) ²	6/9 ¹ (N=1568) ²	N/A
Review	Presence of symptoms at diagnosis	2/6 ¹ (N=454) ²	4/6 ¹ (N=1982) ²	N/A
Review	Diagnostic delay	1/1 ¹ (N=300) ²		N/A
Review	Coeliac disease symptoms experienced	4/9 ¹ (n=1134) ²	5/9 ¹ (n=920) ²	N/A
Review	Presence of additional food intolerances	2/2 ¹ (n=308) ²		N/A
Review	Body weight	1/1 ¹ (N=1018) ²		N/A

Notes:

1. This indicates the number of studies that found a particular factor to be associated or not associated with adherence (e.g. four out of nine (4/9) studies showed that coeliac disease symptoms was a factor associated with adherence and five out of nine (5/9) studies showed no association between adherence and coeliac disease symptoms experienced).
2. n= total number of participants (including controls where relevant)
3. Priority relates to the priority rating of the concept mapping clusters. However, none of the clusters appear in this table.

Healthcare treatment factors

The two concept mapping clusters that related to prescribed GFF fit closely within this theme. The concept map separated the content of what was prescribed and how prescribed GFF is delivered. Overall, the importance of prescribed GFF does appear to be associated with adherence to a GFD.

Several of the factors included in this theme, appeared elsewhere as statements in the concept mapping clusters. For example, the concept mapping cluster ‘motivation, support, determination/will power’ included a statement about membership of a coeliac support group, which the review identified as associated with adherence. This cluster also included support from healthcare professionals and family and friends, which is not present in this group of factors according to Hall *et al.* (2009).

Table 4.29 Healthcare treatment factors.

Data Source	Factors	Factors Associated with Adherence	Factors not associated with adherence	Priority
Review	Duration of gluten-free diet	1/5 ¹ (N=200) ²	4/5 ¹ (N=385) ²	
Review	Difficulty of gluten-free diet	2/3 ¹ (N=3200) ²	1/3 ¹ (N=73) ²	
Review	Receiving gluten-free food on prescription	1/1 ¹ (N=287) ²		
Concept Map	Prescribed gluten-free food	N=73		5
Concept Map	Convenience of obtaining prescribed gluten-free food	N=73		11
Review	Satisfaction with information from health care provider	2/3 ¹ (N=321) ²	1/3 ¹ (N=154) ²	Review
Review	Attendance at coeliac clinic	1/2 ¹ (N=99) ²	1/2 ¹ (N=413) ²	
Review	Regularity of follow-up	3/4 ¹ (N=764) ²	1/4 ¹ (N=207) ²	
Review	Coeliac support group membership	3/3 ¹ (N=528) ²		

Notes:

1. This indicates the number of studies that found a particular factor to be associated or not associated with adherence (e.g. one out of five (1/5) studies showed that duration of gluten-free diet was a factor associated with adherence and four out of five (4/5) studies showed no association between adherence and duration of gluten-free diet).
2. n= total number of participants (including controls where relevant)
3. Priority relates to the priority rating of the concept mapping clusters. However, none of the clusters appear in this table.

Socio-cultural/environmental factors

This group of factors are the ones that map most closely to the concept mapping clusters. Hall *et al.* (2009) concluded that the cost of GFF was not important from the data they had at the time. However, new data has shown that cost is a significant factor affecting adherence to a GFD in adult CD. This finding is supported by the results of this concept mapping study, which found the cost of GFF to be the most important factor affecting adherence to a GFD.

Statements relating to the availability of GFF were split into three clusters by the participants in this study. These clusters highlight not just the availability of GFF (e.g. in shops and restaurants) but the specific provision of GFF in the workplace and the decisions a person with CD may have to make if GFF is not available (i.e. go hungry). In the concept map, factors relating to eating away from home were split into two clusters: eating away from home, which includes travel and restaurants; and eating the same as other people when eating in social situations (including in the hospital).

Table 4.30 Socio-cultural/environmental factors

Data Source	Factors	Factors Associated with Adherence	Factors not associated with adherence	Priority³
Review	Eating convenience foods	1/1 ¹ (N=146) ²		
Review	Eating away from home	6/7 ¹ (N=3680) ²	1/7 ¹ (N=234) ²	
Concept map	Eating away from home	73		7
Review	Having supportive family and friends		1/1 ¹ (N=278) ²	
Concept map	If they can eat the same as other people	73		6
Review	Availability of GFF	3/5 ¹ (N=499) ²	2/3 ¹ (N=395) ²	
Concept Map	The availability of GF sandwiches	73		2
Review	Improved choice of GFF	2/2 ¹ (N=3209) ²		
Concept Map	If they are prepared to go hungry when there is no GFF available	73		8
Concept Map	Provision of GFF at work	73		13
Review	Quality of GFF	4/5 ¹ (N=3519) ²	1/5 ¹ (N=234) ²	
Concept Map	Access to good quality GFF	73		4
Review	Cost of GFF	4/6 ¹ (N=3599) ²	2/6 ¹ (N=308) ²	
Concept Map	The cost of GFF	73		1

Notes:

1. This indicates the number of studies that found a particular factor to be associated or not associated with adherence (e.g. six out of seven (6/7) studies showed that eating away from home was a factor associated with adherence and one out of seven (1/7) studies showed no association between adherence and eating away from home).
2. n= total number of participants (including controls where relevant)
3. Priority relates to the priority rating of the concept mapping clusters. However, none of the clusters appear in this table.

Only one cluster from the concept map fit within this group of factors (social stigma). Social stigma was most closely related with anxiety especially the social impact of eating different food to other people. Depression was not mentioned in any of the statements generated during brainstorming in the concept mapping study. The association between adherence to a GFD and depression is unclear from the evidence provided in the systematic review.

Table 4.31 Quality of life and psychological well-being

Data Source	Factors	Factors Associated with Adherence	Factors not associated with adherence	Priority
Review	Depression	5/7 ¹ (N=888) ²	2/7 ¹ (N=2805) ²	
Review	Presence of psychological disturbance		1/1 ¹ (N=154) ²	
Review	Eating disorders	1/1 ¹ (N=390) ²		
Review	Anxiety	3/8 ¹ (N=3354) ²	5/8 ¹ (N=1696) ²	
Concept Map	Social Stigma	73		10
Review	Anger		1/1 ¹ (N=139) ²	
Review	Mood or stress		1/1 ¹ (N=154) ²	
Review	Quality of life	8/12 ¹ (N=4882) ²	4/12 ¹ (N=1368) ²	
Review	Wellbeing		1/1 ¹ (N=28) ²	

Notes:

1. This indicates the number of studies that found a particular factor to be associated or not associated with adherence (e.g. five out of seven (5/7) studies showed that depression was a factor associated with adherence and two out of seven (2/7) studies showed no association between adherence and depression).
2. n= total number of participants (including controls where relevant)
3. Priority relates to the priority rating of the concept mapping clusters. However, none of the clusters appear in this table.

4.7.6 The construct of knowledge

All of the factors and clusters that I identified from my concept mapping study and systematic review could be mapped to the six themes in the systematic review by Hall *et al* (2009). However, my categorisation differed slightly from that used by Hall *et al.* (2009). From the results of this study, I identified the concept of knowledge as being of a broader base and more complex in nature than that identified by Hall *et al.* (2009). The results show that the construct of the concept of knowledge needed to adhere to a GFD was made up of seven components:

1. The person with CD needs significant knowledge of what foods contain gluten, how to identify gluten ingredients in manufactured foods and how to prepare GFF from scratch. They also need knowledge of the social skills required to be confident enough to ask for a restricted diet in a wide variety of social setting.
2. The family and friends of a person with CD could influence their adherence substantially by providing them with GFF at social events. This requires the family member or friend of the person with CD to understand and apply GF knowledge in their food purchasing and preparation.
3. The healthcare professionals working with CD patients need to have sufficient knowledge of the GFD. However they also needed to

promote prior organisation (e.g. taking food with you) and pragmatism (e.g. going hungry).

4. Support from other people with CD is also essential, often in the context of membership of Coeliac UK. Other people with CD have the practical knowledge of living with a restricted diet in the context of a gluten-dependent environment. They have many practical tips and advice that can be shared and applied.
5. The knowledge of restaurateurs and chefs in relation to the GFD and CD is essential if they are to providing people with CD with safe food when eating out.
6. The knowledge of people working in the food industry in relation to the GFD and CD is also essential. GFF needs to be appropriately labelled and gluten-free ingredients should be chosen over gluten— inclusive ones where possible (e.g. corn flour as a thickening agent in sauces rather than wheat flour).
7. The final group is shops and food outlets that provide food to people when on the go. The staff need to have a knowledge of the GFD and CD so that they can provide a greater choice of GFF that is acceptable to all customers (not just those with CD).

4.8 Summary

I recruited a reasonably diverse group of 73 participants from three stakeholder groups using seven recruitment routes. Participants brainstormed 903 statements relating to adherence to a GFD and data saturation was achieved. Thirteen concepts were found to be associated with adherence to a GFD. A high degree of similarity in the perceptions of the three stakeholder groups was found. The cost of GFF was the most important factor affecting adherence to the GFD. This finding, along with more recently published research, contradicts the evidence that was available at the time of a previous systematic review (Hall *et al.*, 2009) which reported no association between the cost of GFF and adherence to a GFD. The three stakeholder groups appeared to hold similar views about what would help adults with CD stick to a GFD. However, healthcare professionals perceived the second most important cluster (the availability of GF sandwiches) to be significantly less important than the other two stakeholder groups. The availability of GFF away from the home and access to GFF on prescription were issues that affect adherence to a GFD.

The results of this CM study were synthesised with my initial systematic review. All the clusters mapped to the factors identified in the review, however, the concept mapping process helped to clarify the importance of these factors to adherence behaviours and also the construction of the concepts. The concept of knowledge is broader than the construct that is

presented in Hall *et al.*'s (2009) systematic review as 'Knowledge attitudes and beliefs'. Some risk factors in the literature did not map to factors identified in this concept mapping study, in particular sociodemographic factors.

Chapter 5: Discussion

5.1 Introduction

The aim of this study was to gain a better understanding of the factors affecting adherence to a gluten-free diet (GFD) in adults with CD. A further aim was to compare the perspectives of patients, household members and healthcare professionals in relation to what helps adults with coeliac disease (CD) stick to a GFD. From the concept mapping study and the systematic review presented in this thesis, I have identified a number of factors associated with adherence to a GFD in adults with CD. In this discussion, I interpret the results of my concept mapping study and discuss my findings in the context of the existing evidence.

I begin this chapter by discussing the results of recruitment, data collection and analysis. I then discuss each of the thirteen concepts identified in this concept mapping study in order of priority rating, starting with the cost of GFF, which was perceived to be the most important factor affecting adherence to a GFD. I discuss the utility and relevance of the theoretical models of behaviour change in light of the results of this study. The model of adherence to a GFD that I have developed from the results of this study is presented in this chapter and I acknowledge the limitations of this study.

Recommendations for future research are presented at the end of this chapter.

5.2 Recruitment, data collection and data analysis

5.2.1 Recruitment

To limit the effects of selection bias, I recruited participants through seven channels. Several previous studies have recruited only through coeliac support groups, however, people who are members of coeliac support groups tend to be better at adhering to a GFD and, therefore, may not be representative of the wider population. All seven routes of recruitment have their advantages and limitations as outlined in Table 5.1.

Table 5.1 The advantages and limitations of the seven channels of recruitment used in this study.

	Advantages	Limitations
Invitation letters posted by Coeliac UK	Reaches a group of people who have expressed an interest in getting involved in research.	People with coeliac disease who are members of Coeliac UK may not be representative of all people with coeliac disease. Members of Coeliac UK are more likely to adhere to a GF diet, so recruiting partially-adherent or non-adherent participants is difficult.
Advertisement in local newspapers	Reaches those who are not currently being treated for CD and those who are not members of Coeliac UK.	Only those people who read the local paper would see the advertisement. May be inundated with responses when only a small sample is required.
E-mail to healthcare professional members of the British Dietetic Association	Reaches a range of healthcare professionals.	Members may not work with Coeliac patients. E-mails are easily ignored.
Advertisements on Coeliac UK website	Reaches a wide audience of people with CD, healthcare professionals and household members who use the website.	Members may have dermatitis herpetiformis, not coeliac disease. May be inundated with responses when only a small sample is required.
Invitation packs distributed by hand by Norfolk and Norwich University Hospital (NNUH)	Reach partially-adherent and non-adherent as well as healthcare professionals who work with adult coeliac patients.	Only a small number of healthcare professionals work with adult coeliac patients at NNUH. There is no patient database, so it was not possible to distribute patient invitation packs by mail. Invitation packs could only be handed to patients during outpatient appointment, which was a slow process.
Advertisement on the University of East Anglia (UEA) website	Reaches a wide audience of UEA staff and students.	The advertisement may not be viewed widely. May be inundated with responses when only a small sample is required.
Invitation letters posted to General Practitioners and Practice Nurses	Reaches a group of people who may have experience of the diagnosis of coeliac disease and prescribing gluten-free food.	Low response rate is common in this population. General Practitioners and Practice Nurses may have little contact with coeliac patients and, therefore, may not feel motivated to participate.

When designing this study, I considered the factors that could deter people from volunteering to take part. Providing participants with the option of completing the brainstorming and statement sorting tasks remotely may have encouraged participation in individuals who would find it difficult to attend the group sessions. Initially the recruitment of healthcare professionals and non-adherent participants was slow. To speed up the

recruitment process, I adapted my recruitment strategy to address this and was successful in recruiting a satisfactory number of participants (n=73).

The sample of participants with CD and household members represented a wide range of age groups and both males and females were represented in all three stakeholder groups. However, with the exception of one male Gastroenterologist, all other healthcare professionals were female. Dietetics and nursing are predominantly female professions and, therefore, the sample of participants could be considered to be representative of these professions (Sturrock & Lennie, 2009; Nursing and Midwifery Council, 2009).

The group of participants with CD and household members was not ethnically diverse. From these two stakeholder groups I recruited just one male participant with CD who was non-white British. I recruited the majority of participants from the Norfolk region, which has a relatively small proportion of ethnic minorities at just 7.6% of the population in 2011 compared to 19.5% in England and Wales as a whole (Norfolk Insight, 2011; Office for National Statistics, 2012).

I aimed to recruit a mix of adherent, partially-adherent and non-adherent participants for this study. Adherence was measured through self-reporting, which is a notoriously inaccurate measure. However, my aim was not to measure adherence, but to recruit a diverse group of participants in order to collect a varied range of perspectives. The cost of measuring adherence using a more accurate test would be expensive and I did not have the budget to cover this.

Only one non-adherent participant volunteered to take part in this study. This participant had switched back to eating a normal gluten-inclusive diet when his wife died approximately 30 years ago and he had experienced no CD symptoms since then. Including more non-adherent participants may not have added value to this study because the focus was on the barriers faced by people when attempting to follow a GFD. The non-adherent participant included in this study was not attempting to follow a GFD. Fifteen adherent and eighteen partially-adherent participants were recruited and, with the inclusion of the one non-adherent participant, this sample allowed for the collection of data from people with CD who do and those who do not stick to a GFD.

GPs have contact with patients at the time of CD diagnosis and when prescribing GFF. Therefore, GPs could be a valuable source of information in relation to the factors affecting adherence to a GFD. Overall, the recruitment of GPs was difficult and I only managed to recruit one GP for this study. An alternative recruitment strategy may have been more successful. Williamson *et al.* (2007) report that GPs are less likely to participate in research if they believe it will impinge too greatly on their time. On top of the time required to read and complete the participant documents (Appendices 6-22), concept mapping requires approximately three hours of participants' time. This requirement may have deterred GPs (and others) from taking part in this study. By making it clearer to participants that they could complete just one or two of the three concept

mapping activities (brainstorming, prioritising and clustering), it is possible that more people would have volunteered to take part. However, such an approach could have resulted in higher attrition rates.

Having an interest in the topic being studied is also linked with higher GP participation in research (Williamson *et al.*, 2007). In this concept mapping study, a lack of knowledge and support in relation to CD and the GFD amongst healthcare professionals was indicated by participants (Appendix 34, statements 20, 32 and 72). This could reflect a general lack of interest in CD and the GFD in this stakeholder group. However, by targeting healthcare professionals who are members of Coeliac UK and the BDA, I believe that my recruitment strategy was appropriate in targeting healthcare professionals who are likely to have an interest in CD. Although, this strategy was helpful for recruitment, this sample of healthcare professionals is unlikely to be representative of the 'average' healthcare professional who works with CD patients. Participants from these two groups are likely to be more interested and better informed about CD and the GFD.

In summary, my recruitment strategy was successful in that I recruited a mix of participants from the three stakeholder groups using seven recruitment methods. However, the sample of participants included in this study may not be representative of the wider population. In particular, the sample of patients and household members was not ethnically diverse, patients were mostly members of Coeliac UK and only one fully non-adherent patient was recruited. However, I believe that by recruiting participants from three

stakeholder groups and using seven different recruitment strategies, I was able to recruit a sample of participants that represented a broad range of opinions in relation to the factors associated with adherence to a GFD.

5.2.2 Data collection

Brainstorming

Data generation through brainstorming gave participants the opportunity to freely generate statements in relation to the focus prompt '*It would be easier for adults with coeliac disease to stick to a gluten-free diet if...*'. Nine hundred and three statements were generated through brainstorming and the high level of duplication towards the end of brainstorming indicated that data saturation had been reached. By anticipating the type of responses that could be generated, the steering group believed that the wording of the prompt would serve to elicit a wide range of ideas relating to adherence to the GFD. However, the wording of the focus prompt could have been presented in a number of ways and it is possible that different wording may have generated different responses (Kane & Trochim, 2007).

Group brainstorming facilitates the generation of a higher number of ideas compared to remote, individual brainstorming (Kane & Trochim, 2007). Whether participants brainstormed their ideas remotely or in group brainstorming sessions may have affected the type and number of statements generated. During group brainstorming, participants generated ideas through lively discussions. Participants who brainstormed remotely

would not have experienced the same level of interactivity and exchange of ideas (Kane and Trochim, 2007). However, healthcare professionals who completed brainstorming remotely in the work environment may have had their ideas triggered by patients, colleagues and other resources that would not have been available to them during a group brainstorming sessions at the UEA. Similarly, patients and household members who completed brainstorming remotely may also have had their ideas triggered by factors in their environment that they may not have considered in a group setting.

Participants who brainstormed remotely may have felt more willing to disclose sensitive information that they would have felt uncomfortable to disclose in a group setting. In addition, remote brainstorming avoids the problems associated with having outspoken participants in the group, who dominate the discussion and prevent others from voicing their ideas (Kane & Trochim, 2007). To minimise the chance of these issues occurring during group brainstorming sessions, the facilitators encouraged participants to generate ideas anonymously on pieces of paper and we ensured all participants were given equal opportunities to speak, preventing individuals from dominating the discussion. By including both remote and group brainstorming sessions in this concept mapping study, the limitations associated with each method were to some degree mitigated.

Statement reduction

Including large numbers of statements in a concept mapping study can impose practical constraints for participants when completing the

subsequent statement sorting activities. We reduced the number of statements generated during brainstorming to a final set of 91. As there was a high degree of duplication in the statements generated during brainstorming, the statement reduction task was less difficult than it might otherwise have been. Although subjective, the decisions made by the steering group when reducing the statements were reached through discussion and detailed analysis of the original data. We were careful to ensure that all ideas were represented in the final set of statements and we kept editing to a minimum in order to retain the original meaning of the statements where possible. It is possible that in changing the wording of some statements to make them clearer, or in merging two or more statements together, the original meaning may have been altered or lost (Kane & Trochim, 2007). However, by involving representatives from the three stakeholder groups in the statement reduction process and in agreeing on appropriate wording of the statements, I hoped to reduce the likelihood of this occurring.

Prioritising and clustering

Prioritising and clustering are individual activities and, therefore it should make little difference whether these tasks are completed in a group setting or remotely. Although it was possible to monitor the participants who attended the group sessions to ensure their responses were not influenced by the people around them, I cannot be certain that the participants who completed the tasks remotely were not influenced by the opinions of others when sorting the statements.

The order in which the two tasks are completed could influence the results. Participants may be likely to perceive the statements that they view at the start of the task differently to the ones they view when reaching the end. Further, participants will be familiar with the statements by the time they complete the second task and this familiarity may influence their decisions in some way (Kane & Trochim, 2007). To limit the impact of this potential source of bias, I shuffled the piles of cards so that all participants would view them in a different order.

Participants in concept mapping studies are often reluctant to rank statements with a low priority score (Kane and Trochim, 2007). The reason for this is believed to be the fact that all statements that were brainstormed are likely to have *some* importance (Kane & Trochim, 2007). Having fairly equal piles of statements in the prioritising task allows us to get a better idea of the relative values of the statements. Despite this, many concept mapping projects have allowed participants to prioritise as many statements as they like into each of the five piles on the Likert scale (1=least important, 5=most important) (Kane and Trochim, 2007). To avoid the likelihood of participants placing fewer statements in the pile with the lowest priority score, I asked them to place a fairly equal number of statements in each pile. However, this is likely to have made the task more challenging for those participants who felt inclined to give all statements higher priority ratings.

Sorting the statements into piles for similarity (clustering) helps us to identify stakeholders' views of the relationship between ideas. It is possible that some statements in this study may have been deemed to 'belong' in more than one pile. Participants were instructed not to place individual statements in more than one pile because that would make it difficult to identify the interrelationships between ideas. Although none of the participants reported this restriction to cause them problems, it is possible that it would make the deliberation process more complicated. Participants may perceive more than one way of sorting the cards and the criteria used for sorting the statements are likely to differ between participants. In this study, participants were instructed to sort the statements in a way that made sense to them. What makes sense to one person does not necessarily make sense to somebody else and it is possible that participants differed in the criteria they used to sort the statements. However, a synthesis of the results for all participants should provide a general overview of how the group as a whole perceives the relatedness of the ideas.

Not all of the participants completed brainstorming, prioritising and clustering and it is possible that the result may have differed if all participants had completed all three tasks. However, Tables 4.4, 4.8 and 4.9 in Chapter 4 showed that there were not large differences between the characteristics of the participants who completed the tasks compared to those who did not.

5.2.3 Data analysis

Data were input onto separate computers to reduce the chance of data inputting errors occurring. Some of the data could not be entered for participants who had grouped the statements into more than 12 clusters. To reduce the impact that this may have had on the results, I removed the clusters with the least number of statements in them in line with the advice provided by the software developer (Appendix 35). Some of these ‘clusters’ contained just one statement and, therefore, would not have been useful in providing information about how the statements relate to each other anyway. Therefore, it is unlikely that the results would have been substantially different whether or not these small clusters were included in the analysis. The owner of the software reassured me that the impact of removing these clusters should have little effect on the results (Appendix 35).

Some errors were identified in the data provided by participants for the prioritising and clustering tasks. Where the error could not be corrected, these data were not input into Ariadne for analysis. Although it is unlikely that this would have had much of an impact on the results, there is a chance that it could have altered the findings slightly.

Having participants enter their data directly into a computer programme, rather than manually writing their responses on paper, may have reduced the likelihood of error. However, Ariadne does not provide such a facility and

the resources available for this study were not sufficient to cover the costs of using an alternative programme, such as the Concept Systems concept mapping software (Concept Systems Incorporated 2013) which is available in the United States.

One of the key stages in the concept mapping study is deciding on the number of clusters to include in the concept map (Kane & Trochim, 2007). The steering group decided on the number of clusters that we felt was most appropriate to represent the emerging themes from the data. Decisions about how many clusters to include in a concept map are rather subjective. However, Appendix 39 shows that there were generally very few changes to the statements contained within the clusters when the number of clusters was increased or decreased.

5.3 The thirteen factors affecting adherence to a gluten-free diet

5.3.1 Cluster 1 - The cost of gluten-free food

A systematic review conducted in 2007 by Hall *et al.* (2009) found no clear association between the cost of GFF and adherence to the GFD at that time. However, a study by the same author in 2013 reported a relationship between the cost of GFF and adherence to a GFD (Hall *et al.*, 2013). My update to the systematic review by Hall *et al.* (2009) and the results of my

concept mapping study support the finding that cost plays an important role in relation to adherence to a GFD. Two studies with a total of 308 participants showed no association between the cost of GFF and adherence to a GFD, whereas five studies (including my concept mapping study) with 3672 participants reported an association between the cost of GFF and adherence to a GFD. In addition, a study by Barratt *et al.* (2011) reported that better adherence was linked with having an affluent background and being a wealthy achiever. Both these factors could be associated with the affordability of GFF. No statistically significant difference was observed between the perceptions of the three stakeholder groups in relation to the impact of the cost of GFF on adherence to a GFD. All three groups perceived the cost of GFF to be the most important factor affecting adherence to a GFD.

Studies in the USA and the UK have reported that, on average, all speciality GF products were more expensive than their gluten-inclusive counterparts (Lee *et al.*, 2007; Coeliac UK, 2009; Singh and Whelan, 2011). Helping patients to manage the cost of GFF has been argued to be equally as important as helping them to understand the GFD (Cureton, 2007).

To gain a better understanding of the cost burden of the GFD in the UK, Coeliac UK conducted the Cost Project in 2009. The Cost Project involved over 100 volunteers who visited their local supermarkets to compare the cost of GF products with the cost of similar gluten-inclusive products (Coeliac UK, 2009). The results showed that all GF items included in the

analysis were more expensive than gluten containing equivalent products, with GF bread costing over five times the amount of similar gluten-inclusive bread. No significant difference was found in the cost of GFF from supermarkets in different regions of the UK.

The results showed that supermarket own-brand GFF tended to be more expensive than branded GFF across six supermarkets (Asda, Co-op, Morrisons, Sainsbury's, Tesco and Waitrose). Coeliac UK questioned the reason for this price difference and they also questioned the reasons why three similar products (pitta bread, bread and pizza bases) should have such wide variation in price, with pitta bread costing 6.2 times the price of its gluten-containing equivalent, bread 5.1 times and pizza bases 2.4 times the price of their gluten-containing counterparts. As far as I can find, there has been no justification made for the discrepancy in the pricing of these products.

According to dietitians, the high cost of GFF is problematic for coeliac patients on low incomes (O'Donnell & Edelstein, 2009). In the UK patients are assisted with the increased cost of the GFD by the provision of a limited, basic range of prescribed GFFs. For many people this provision is not exempt from the prescription charge. Despite the availability of GFF on prescription for people living with CD in the UK, this concept mapping study found the cost of GFF to be the most important factor affecting adherence.

Recent cut-backs in the amount and type of GFF available on prescription may have exacerbated the problem of cost for many patients. 'Luxury' items such as cakes and biscuits are no longer available on prescription. If people with CD choose to eat these 'luxury' products they are likely to have to pay more for ones that are labelled as GF. Coeliac UK has campaigned against the reduction in prescribed GFF and they highlight the importance of this provision in helping patients stick to their GFD (Coeliac UK, 2013d). It could be argued that people with a wheat allergy face the same difficulties as coeliac patients when purchasing expensive speciality food. Despite this, prescribed food is not made available by the NHS for people with a wheat allergy. The reason for this could be that the long-term health problems associated with CD are not seen in wheat allergy. By assisting patients with CD to stick to a GFD, the NHS can reduce the amount of money required for treating the health problems associated with non-adherence to a GFD.

Rather than obtaining expensive speciality GF products, patients can be educated to avoid specialist GFF and to cook naturally GFF where possible. For example, rice is cheap and could be used as an alternative to expensive GF products, such as bread and pasta. However, one of the problems identified from my systematic review is that the GFD is viewed as restrictive and this is a factor that reduces adherence. Therefore, having the choice to eat specialist GFF can reduce the perception of restriction and enhance adherence.

This study has identified the cost of GFF as the most important factor affecting adherence. People with CD are being asked to pay more for food of lesser quality. However, the higher cost of GFF could act as 'salt on the wound', rather than being a factor that affects adherence. The cost of GFF is likely to be an issue for people on a limited budget but whether the issue of cost actually stops most people with CD from adhering to a GFD is unclear.

5.3.2 Cluster 2 - The availability of gluten-free sandwiches

The availability of GF sandwiches was found to be the second most important factor affecting adherence to a GFD. This finding, which had not been identified in my systematic review, was somewhat surprising to me. This 'cluster' contained just one single statement that represented a synthesis of four statements from the original brainstorming statements (see Section 4.7.3).

In Britain sandwiches are eaten commonly at lunchtime, however, this finding may not be applicable outside of British diets. The pervasiveness of the sandwich as the usual choice for lunch means that GF alternatives can be hard to find and restricted in choice. Although some shops now do provide GF sandwiches they are still not widely available and they tend to be more expensive than regular sandwiches (Coeliac UK, 2013b). Some participants stated that they had to rely on bringing packed lunches when away from home, otherwise they would go hungry. I have not been able to find any

literature relating to the role of the GF sandwich in patients with CD and this topic requires further exploration.

Healthcare professionals prioritised the availability of GF sandwiches lower than adults with CD and household members. It may well be that they do not perceive the day-to-day impact of this limited choice of lunchtime food. Healthcare professionals need to be more aware of this problem and help patients to overcome this, possibly by encouraging them to prepare sandwiches at home to take with them.

5.3.3 Cluster 3 - Knowledge and information about coeliac disease and the gluten-free diet

Cluster 3 contains 24 statements relating to knowledge and information about CD and the GFD (Table 4.13, Chapter 4). This cluster related to the knowledge held by patients, healthcare professionals and others, such as friends, family members and those working in the food industry. In Section 4.7.6 I introduced the ‘concept of knowledge’ that was developed from the findings of this study and this included many of the issues highlighted in this cluster. The concept of knowledge will be discussed later in this chapter.

In the UK, Coeliac UK is the major charity providing information about CD and supporting people to follow a GFD. It is recognised that patients have a lot to learn at the time of diagnosis, and having multiple sources of valid

information may be helpful. Coeliac UK also provides information about CD and the GFD for healthcare professionals and people working in the food industry and this charity is working hard to educate people and raise awareness about CD and the GFD (Coeliac UK, 2013e).

As well as getting information from Coeliac UK, patients also gather information about CD and the GFD from several sources. Online forums provide the opportunity for patients to chat and exchange knowledge and ideas as well as providing a source of support and encouragement (Veen *et al.* 2010). Patients also look to health professionals to provide them with information about CD and the GFD (British Society of Gastroenterology, 2009). This may include being made aware of the possible health consequences relating to non-adherence of a GFD as well as developing their knowledge about how to follow a strict GFD.

In the systematic review by Hall *et al.* (2009) knowledge is grouped together with attitudes and beliefs. These factors are all related to the patient and not healthcare professionals. Health promotion has for many years been dominated by a health education model which emphasises lifestyle and behaviour change of patients through education. This model places responsibility for health behaviour solely with the patient. An example of this approach is emphasised in the statement:

‘The empowered and knowledgeable patient is fundamental to the successful management of coeliac disease’

(British Specialist Nutrition Association Ltd., 2011).

The health education model approach has been led by healthcare professionals and it is recognised that educating patients as the only method of improving adherence is not a strategy that is likely to succeed (Willey *et al.*, 2000). A broader approach that takes into account the knowledge of healthcare professionals and other sources of support for people with CD may be more useful.

The statements contained within cluster 3 (shown in Table 4.13) reveal a lot of dissatisfaction in relation to healthcare professionals' knowledge about CD and GFD. A statement from the patient's perspective shown on the British Society of Gastroenterology website emphasises this point:

*“Patients are frustrated...by a lack of expertise and continuity
in both primary and secondary care”*

(British Society of Gastroenterology, 2009)

Several of the statements contained within this cluster suggest that healthcare professionals need to increase their level of knowledge about CD and the GFD. A study by Nelson *et al.* (2007) found one-quarter of dietitians working with coeliac patients had received no CD training in the previous two years. Coeliac UK provides a useful resource for increasing the knowledge of patients and healthcare professionals (Coeliac UK, 2013e). Nelson *et al.* (2007) reported that almost all dietetic departments included in their study referred patients to Coeliac UK. Although the British Society of Gastroenterology (2009) advises healthcare professionals to refer

patients to Coeliac UK, it is possible that healthcare professionals are relying too heavily on the supportive role of Coeliac UK rather than keeping their knowledge of CD and the GFD up-to-date.

In contrast, the healthcare professionals included in this concept mapping study were found to have similar views to patients and household members in relation to the factors that would help patients stick to a GFD. This similarity suggests that healthcare professionals have a good insight into what it is like to live with CD. However, the sample of healthcare professionals involved in this study were self-selected and, therefore, are unlikely to be representative of the majority of healthcare professionals who work with CD patients.

The people who prepare food for people with CD, such as family, friends and chefs in the food industry also need to have a good knowledge of CD and the GFD. A study by Karajeh *et al.* (2005) reported that chef's knowledge of CD was poorer than that of the general public. Restaurant chefs were more knowledgeable than take-away chefs but overall there was a clear deficit in knowledge about CD and the GFD. This study was conducted almost ten years ago and knowledge about CD and the GFD has improved since then. Despite this general increase in awareness, the statements generated from this concept mapping study and the results from the systematic review suggest that the knowledge of people working in the food industry is still less than adequate. This issue is discussed further under Cluster 7 later in this chapter.

5.3.4 Cluster 4 - Access to good quality gluten-free food

As mentioned in Cluster 1, the quality and accessibility of GFF is relatively poor compared to gluten-containing foods. Although the availability and quality of GFF in the UK has improved in recent years, poor access to good quality and affordable GFF still represents a significant barrier to adherence (Singh & Whelan, 2011). Specific problems associated with access to GFF are highlighted in the statements included in Cluster 4 (Table 4.14) and in the systematic review in chapter 2. In addition, the statements contained in Cluster 7 (Eating away from the home) shown in Table 4.17 also highlight problems relating to poor accessibility. Cluster 7 is discussed later in this chapter.

With regards to the quality of GFF, the statements contained in Cluster 4 suggest that efforts made to improve the quality of GFF have not been entirely successful and patients are still dissatisfied with the taste and texture of GFF. This is particularly evident in relation to the quality of GF bread (Table 4.14). In addition, poor access to non-traditional types of GF bread, such as pita bread, chapatis and tortillas has also been highlighted. Despite this, Coeliac UK's Food and Drink Directory (Coeliac UK, 2013b) lists several non-traditional GF breads that are available from several supermarkets. Whether these items are regularly stocked in these supermarkets is not clear and in their Cost Project, Coeliac UK (2009) found that only one UK supermarket chain had pita bread available in store.

It can be difficult for people with CD to easily identify GFF when shopping because there has not been universal adoption of the cross-grain logo in manufactured products. In the UK, new GFF labelling legislation has made it more difficult for manufacturers to label their foods as ‘gluten-free’ (Food Standards Agency, 2012). Patients are often required to have a good understanding of gluten-containing ingredients if they are to avoid them (Coeliac UK 2013a). Many food products have gluten added as a thickener, binding or bulking agent and identifying these ‘hidden’ sources of gluten can be difficult (Singh & Whelan, 2011). Manufacturers can change the ingredients used in their products without highlighting it on the packaging. Food which had previously been ‘safe’ can change to contain gluten with little or no notification to consumers and this can result in accidental gluten consumption.

Overall, the association between access to GFF and adherence to a GFD is not clear and the evidence in the systematic review shown in Table 2.9 is contradictory. The link between the quality of GFF and adherence to a GFD is clearer. The results of my systematic review and concept mapping study show that having access to GFF that tastes good and has a good texture is associated with better adherence to a GFD.

5.3.5 Cluster 5 - Prescribed gluten-free food

Clusters 5 and 11 both relate to prescribed GFF. The main theme of this cluster relates to obtaining an adequate amount and range of GF products on

prescription. Cluster 11 focuses on the practicalities of obtaining GFF on prescription. The evidence base for this issue is small because it only applies to the UK.

My systematic review identified just one study relating to the role of prescribed GFF on adherence to a GFD. This study reported that the 86.1% of participants who were receiving GFF on prescription had better adherence to a GFD compared to participants who were not receiving prescribed GFF (Hall *et al.*, 2013).

Participants in this concept mapping study reported discrepancies in the amount and type of GFF made available on prescription by GPs (Table 4.13). There is a need for more consistency in what GPs prescribe and Coeliac UK has campaigned against the cut-backs in the variety and quantity of GFF offered on prescription and the “post-code lottery” in its provision (Coeliac UK, 2013b). Recent reductions in NHS spending on prescribed GFF may prove futile if adherence to the GFD worsens as a result.

5.3.6 Cluster 6 - Eating the same as other people

There were two statements in this cluster which largely appear to be unrelated (Table 4.16). The first statement (...if you can eat the same gluten-free food as everyone else when eating socially) highlighted the social role of food, where not sharing in communal meals (e.g. at weddings)

can be perceived as being ungrateful or disrespectful. Sverker *et al.* (2005) report on the emotional aspect of not being able to eat the same as other people. People with CD report feeling isolated and ashamed when they cannot eat the same food as other people (Sverker *et al.*, 2005).

The second statement in this cluster relates to the provision of GFF in hospital. A study by Zarkadas *et al.* (2006) also identified the hospital as being a difficult setting in relation to following a GFD. The relationship between these two statements could be that they both focus on difficulties when eating in a public setting. Not only did the participants not wish to 'cause problems' for the hospital staff when asking for a specialist diet, but they were often disappointed by the lack of knowledge and GFF provision. This indicates that patients were not being supported in their adherence to a GFD in hospital. Patients in hospital are vulnerable and the consequences of non-adherence to a GFD at this time may lead to further deterioration in their health. Some patients may choose to go hungry, rather than consume gluten-containing food in hospital and this is also likely to have a negative impact on their recovery. The hospital setting is one which should be set up to promote good health and this dissonance could be regarded as unacceptable.

Similar problems can occur when eating socially and issues around not wanting to draw attention to oneself or 'make a fuss' when asking for GFF when eating socially have been evidenced in this study and in previous research (Sverker *et al.*, 2005; Zarkadas, 2006).

5.3.7 Cluster 7 - Eating away from home

At home, people with CD have more control over what GFF is available and in ensuring safety from contamination. The results of the concept mapping study and systematic review show a strong correlation between eating away from home and poorer adherence to a GFD. When eating away from the home, it can be difficult to find GFF (Zarkadas, 2006). In particular this can be problematic when travelling (Zarkadas, 2006). The statements in Cluster 7 (Table 4.17) highlight a number of problems experienced by people with CD when eating away from the home.

The results of this study suggest that patients may find it easier to stick with their GFD if they take their own GFF with them when eating out, or if they provide GF ingredients to the restaurant (e.g. GF gravy granules) Airlines need to improve their provision of GFF, particularly during long flights when passengers with CD may have to endure several hours with nothing to eat. The statements in this cluster show a great desire for restaurants and their staff to be better informed about CD and GFF so that patients' needs can be more discretely and easily accommodated. This is another component of the complex construct of knowledge that is needed to support people with CD to adhere to their diet.

5.3.8 Cluster 8 - If they are prepared to go hungry when there is no gluten-free food available

It was interesting that people with CD and those that lived with them were more pragmatic about the fact that in order to remain adherent they may at times have to go hungry. This finding had not been reported in any of the studies included in my systematic review. It was implied in the statements that this would generally mean missing just one meal or snack. It was also highlighted (in the previous cluster) that this could be mitigated by the person with CD being organised enough to take GFF with them when in locations where GFF could be hard to find.

Healthcare professionals did not rate this issue as highly as the other two stakeholder groups. This could be because they would be uncomfortable giving such advice to patients or they may believe that with the correct preparation, patients should not need to go hungry. However participants in my study highlighted that being prepared to go hungry is sometimes necessary. This reinforces the findings of my systematic review that people who have an overall tendency to plan and be organised were more likely to be adherent to a GFD (Edwards-George *et al.*, 2009).

5.3.9 Cluster 9 - Motivation and support

This cluster represents self-motivation and the social support provided by family, friends and work colleagues. People who interact with people with CD can be very influential and practically supportive in their adherence behaviours, including speaking up for them (e.g. in restaurants) and cooking GFF for them. Online conversations between people with CD can help to motivate them to stick to a GFD (Veen *et al.*, 2010).

Self-efficacy, conscientiousness and self-discipline, being prepared and organised were factors related to adherence that were reported in the systematic review (Table 2.6). Self-motivation is a similar trait that was also associated with adherence to a GFD in this concept mapping study. As well as educating patients about the GFD, it is also important that they are motivated to change their behaviour. Some of the models of behaviour change discussed in Chapter 1 may provide a useful tool for changing patient behaviour and this is discussed in more detail later in this chapter.

The sole statement relating to sociodemographic factors was located in this cluster. “*If they are female*” was a statement generated by a mother and daughter, both with CD, in relation to the father of their household who they believed would experience greater difficulty following a GFD due to his lack of kitchen skills. However, this statement was rated the lowest of all statements generated in this study. The link between gender and food preparation is highly variable within the UK and my systematic review

identified no sociodemographic factors that were consistently associated with adherence. Therefore, I do not consider this to be an important finding. Hall *et al.* (2009) also reported no consistent relationship between gender and adherence to a GFD.

5.3.10 Cluster 10 - Social stigma

It has to be recognised that food is not just nutritive, it plays an important social function. Many celebrations, ceremonies and gatherings are marked with the provision of food; often specialist to that event (e.g. Christmas pudding and birthday cake) (Gibney *et al.*, 2006). Additionally, food is a way of expressing love and care for other people. Therefore, it is unsurprising that people on a restricted diet can face negative social consequences when they refuse the food that is offered to them.

Sverker *et al.* (2005) report on some of the dilemmas faced by people living with CD including one man who was ridiculed by his work colleagues for eating GF crisp bread. People with CD need to have considerable communication skills and tact in order for them to communicate their dietary needs. However, it is also helpful if other people have the knowledge that this request is genuine and not 'faddy'. Again this highlights the complexity of the knowledge construct needed for GFD adherence.

5.3.11 Cluster 11 - Convenience of obtaining prescribed gluten-free food

Hippocrates famously said "*Let food be thy medicine and let thy medicine be food*". Thinking of food as medicine is quite common in ordinary life e.g. superfoods, nutrient supplements. Despite this, the statements in cluster 11 suggest that participants did not feel that it is appropriate to collect their food from a pharmacy when it is available in shops. Most people with CD who are adherent will have no symptoms and may not perceive themselves as being ill. It may feel odd to them to collect food from a venue where they would normally only collect medication.

To make the process of obtaining GFF on prescription less burdensome for GPs and patients, some areas of the UK have introduced a pharmacy-led supply scheme (Coeliac UK 2014b). The scheme removes the need for patients to get a prescription from a GP for their GFF (Coeliac UK, 2014b). Although this is a step in the right direction, this study has found that participants do not want to collect their prescribed GFF from a pharmacy.

The current system of prescribing GFF is politically defensible but not pragmatic. GFFs are now more widely available in the shops and having to collect prescribed GFF from a pharmacy may no longer be necessary for most people. Patients may be inconvenienced when collecting prescribed GFF in bulk from a pharmacy. This system may be inconvenient for patients who have little freezer or storage space and for those who do not own a car.

During one brainstorming session, participants discussed the benefit of having a voucher system to spend in shops and supermarkets rather than collecting large quantities of GFF from a pharmacy.

Additionally the NHS is set up to distribute medications, not foods and inevitably this leads to large mark-ups on the cost of provision of GFF compared to the cost of these foods in the supermarket. Recent media attention has highlighted the unnecessary over-spending by the NHS on prescribed GFF and a more pragmatic and cost-effective system is overdue (Coeliac UK, 2012). Perhaps it is time to review the system in which prescribed GFF is made available to patients and introduce a system that is more convenient and appropriate for patients.

5.3.12 Cluster 12 - Diet planning and preparation

The utility of membership of Coeliac UK for practical advice on GFF and recipes was recognised by participants in this study. This reinforces the finding of the systematic review which reported that membership of a support group is linked with adherent behaviours. One source of dissatisfaction that was noted from one of the statements generated in this study related to the cost of membership of Coeliac UK, which previously was free of charge.

Having the skills to identify GF products when out shopping and the skills to cook GF meals are also important in relation to following a GFD. Additionally the practicalities of storing GFF in bulk required that people considered having a large freezer. This may be particularly useful when collecting prescribed GFF in bulk, such as bread, which may not have a long shelf-life.

5.3.13 Cluster 13 - Provision of gluten-free food at work

This was a single statement cluster that focuses on the issue of biscuits being provided with coffee in a work context. Again this highlights the day-to-day impact of lack of availability of GFF, which is similar to the issue of poor availability of lunchtime sandwiches (Cluster 2). This cluster also links with issues of being prepared (taking your own GFF with you), being prepared to go hungry, feeling stigmatised by your dietary choices, and being unsupported by your work colleagues in your health.

Although this was the least important cluster in this study, its importance to the CD population in general may be higher than is indicated here. The reason for this is that many of the participants with CD in this study were of retirement age, therefore, work-related issues were likely to be of lesser importance to them.

5.4 The concept of knowledge

In Chapter 4 the results of this concept mapping study were synthesised with the results of the systematic review by Hall *et al.* (2009) and my updated systematic review. From this I identified the ‘concept of knowledge’ in relation to the GFD.

The concept of knowledge acknowledges that it is simplistic and unachievable to expect a person with CD to be the sole repository for knowledge of how to adhere to a GFD. It is interesting that my systematic review identified trusting other people to prepare their food as being associated with poorer adherence. It is impractical in modern life to expect people with CD to never eat food unless they themselves have prepared it. Therefore the knowledge base of the importance of a GFD and the health consequences that non-adherence causes, must be more generally known.

The individual with CD is required to develop a good understanding of CD and GFF if they are to stick to a GFD. They are also required to develop coping strategies for living in a gluten-dependent environment where other people may not understand their dietary requirements. Developing the social skills and resilience necessary to be able to request GFF in any situation and to always avoid gluten-containing products is not easy for the person with CD. Improved education about CD and the GFD for friends, family, people in the catering and food manufacturing industry, healthcare professionals, colleagues at work etc. is required in order to reduce some of the challenges

faced by people with CD. However, improving knowledge about the GFD is not an easy task.

In the following section, I discuss some of the models of behaviour change in relation to changing the adherence behaviour of adults with CD. These models were previously introduced in Chapter 1. I follow that by presenting a model of adherence to the GFD that I have developed from the results of this study.

5.5 Changing behaviour in adults with coeliac disease

In the previous section I acknowledged that it is not just the person with CD who needs to change in order for adherence to a GFD to be made easier. There are many societal and environmental factors that make behaviour change difficult to achieve (Ni Mhurchu *et al.*, 1997). On an individual level, knowledge, attitudes, beliefs, and intentions also play a role in determining behaviour (Ni Mhurchu *et al.*, 1997). Theoretical models of behaviour have been designed to operate at an individual, rather than a societal level (Gibney *et al.*, 2007). I will discuss how individuals with CD can be helped to adhere to a GFD in an environment where gluten is difficult to avoid.

As acknowledged in chapter 1, patients are required to make substantial changes to their behaviour when they are diagnosed with CD. Theoretical models can be used to develop interventions to target behaviour change and it is useful to know which is the most suitable model or models to use (Gibney *et al.*, 2006). In Chapter 1 I introduced six theoretical models of behaviour change that could be used to target adherence to a GFD. These include: The health belief model (HBM); the theory of reasoned action (TRA) and planned behaviour; behavioural learning theory; social cognitive theory (SCT); information motivation behaviour (IMB) skills theory; and the transtheoretical (stages of change) model. Each model has its strengths and limitations and these were discussed in Chapter 1.

The results of this concept mapping study and systematic review provide us with a better understanding of the factors that influence adherent behaviour in adults with CD. The results of this study have also provided a clearer understanding of how these factors operate at both an individual and a societal level. In addition, this study has identified how these factors interact to make it more or less difficult for a person with CD to stick to a GFD. This study has shown that adherence to a GFD is affected by a wide range of individual and societal factors, including both the psychological and practical issues associated with following a restricted diet. Therefore, a holistic approach to behaviour change would be most appropriate for addressing these issues. Rather than focusing on one single theoretical model, I will take a blended approach, drawing together several themes

from a number of models that could be useful in improving adherence to a GFD.

The transtheoretical (stages of change) model acknowledges that people have different levels of readiness to change and individuals are motivated in different ways (Gibney *et al.*, 2006). Although this model does not take account of social influences, it could be useful in addressing intentional non-adherence in people with CD. This model includes Bandura's self-efficacy theory, which could be used in targeting individual's beliefs about their ability to stick to the GFD. According to this model, helping people to believe they are able to stick to a GFD may improve adherence.

The HBM could be used to address attitudes and beliefs relating to the seriousness of non-adherence to a GFD in coeliac patients. By educating patients about the possible consequences of not following a GFD, they may begin to understand the risks associated with their non-adherent behaviour and this could trigger behaviour change. Similarly, the TRA could also be used to address attitudes and beliefs about the consequences of non-adherence to the GFD. According to this model, patients who believe that following a GFD is beneficial to their health are more likely to adhere to a GFD. However, whether an individual takes action to change their behaviour may depend on their level of self-efficacy, motivation and locus of control. Ford *et al.* (2012) found that patients with CD who have a strong sense of personal control and a better understanding of CD had higher self-efficacy. The patients with higher self-efficacy were found to be better at

adhering to a GFD (Ford *et al.*, 2012). As discussed in Chapter 1, people who feel that they are in control of their own actions are more likely to engage in behaviour change.

Feeling motivated is an important factor in behaviour change (Gibney *et al.*, 2007). In Chapter 1 I introduced the idea of financial incentives for improving adherence to a GFD. The cost of GFF was found to be the most important factor affecting adherence in this study and, therefore, it is possible that financial incentives could be a powerful motivator for adherence. As recognised in the transtheoretical (stages of change) model, individuals are motivated in different ways. Although financial incentives may work in motivating some people to change their behaviour, this will not work for everyone.

Feeling motivated and believing that you can stick to a GFD may not be sufficient to change dietary behaviour in CD. Following a GFD is complicated and this study has shown that patients also require the knowledge, skills and resources necessary for adherence. Issues such as the high cost and poor availability of GFF and a lack of the skills required to read food labels and cook GF meals cannot be resolved by changing an individual's attitudes and beliefs alone. In addition, Herman & Mack (1975) recognised the impact of imposing restraint on eating in dieters and they found that paradoxically trying to eat less resulted in overeating. Trying to cut gluten out of the diet could have the same cognitive effect, with gluten-

containing foods becoming more desirable because the person with CD is told they cannot have them.

Social cognition models, such as the TPB highlight the role of behavioural intention, attitudes and motivation (Ajzen, 1991). Research has shown that behavioural intention is linked with behaviour change in a number of health behaviours, including condom use, intention to exercise and intention to attend cervical screening appointments (Ogden *et al.*, 2007). Whether or not a patient with CD intends to stick to their GFD could be a good indicator of behaviour. In a study by Hall *et al.* (2013) 40.1% of patients reported intentionally consuming gluten in the previous six months. Assisting patients in changing their intentions towards sticking to a GFD could be helpful in an intervention to improve adherence to a GFD. Sainsbury *et al.* (2013a) reported an intention-behaviour gap in relation to adherence to a GFD in patients with CD. This was believed to be due to psychological symptoms in patients with CD, including depression (Sainsbury *et al.*, 2013a). Psychological disorders are more commonly seen in patients with CD and this may make behaviour change more problematic. However, Hauser *et al.* (2010) found no connection between adherence to a GFD and anxiety/depression.

As previously stated, I believe that an intervention to improve adherence to a GFD needs to take account of the societal as well as individual influences on behaviour. Such an intervention would also need to address both the psychological and practical aspects of following a GFD. In the following

section I introduce my model of adherence to a GFD which takes account of both the individual and societal influences on adherence as well as the practical and psychological influences.

5.6 A model of adherence to the gluten-free diet

To understand the complexity of adherence to a GFD in CD, I developed a model of adherence to a GFD based on the findings from this study. The model provides a framework for developing an intervention to improve adherence in CD. This model was developed from the concept maps and it represents the domain of ideas collected from three stakeholder groups. The concept map was divided into four quadrants which represent four overriding themes that influence an individual's ability to follow a GFD (psychological, practical, individual and societal influences). The model of adherence is based around these four interrelated themes (Figure 5.1) and it highlights the resilience that is required by patients in light of the pressures from society (e.g. high cost of GFF, poor availability of GFF). The factors associated with adherence to a GFD identified in the systematic review by Hall *et al.* (2009) and my updated review overlapped with the findings from my concept mapping study and all factors can be represented within this model. I will discuss the content of these four quadrants in more detail in the following four sections.

As with the concept map in Figure 4.2, the model of adherence is divided into four quadrants. The thirteen clusters from the concept mapping study are represented in the same quadrant on the model as they appeared on the concept map (Figure 4.2). Four of the six themes identified in the systematic review will be discussed in relation to the quadrants on the model. The other two themes (sociodemographic and sociocultural/environmental factors) span across all four quadrants and will not be discussed separately for each quadrant. For example, a person's age may have both a practical and psychological influence on adherence to a GFD and the influence of a person's age can operate at either an individual or societal level. Sociodemographic factors were not found to be associated strongly with adherence to a GFD in either the systematic review or the concept mapping study (Table 4.26).

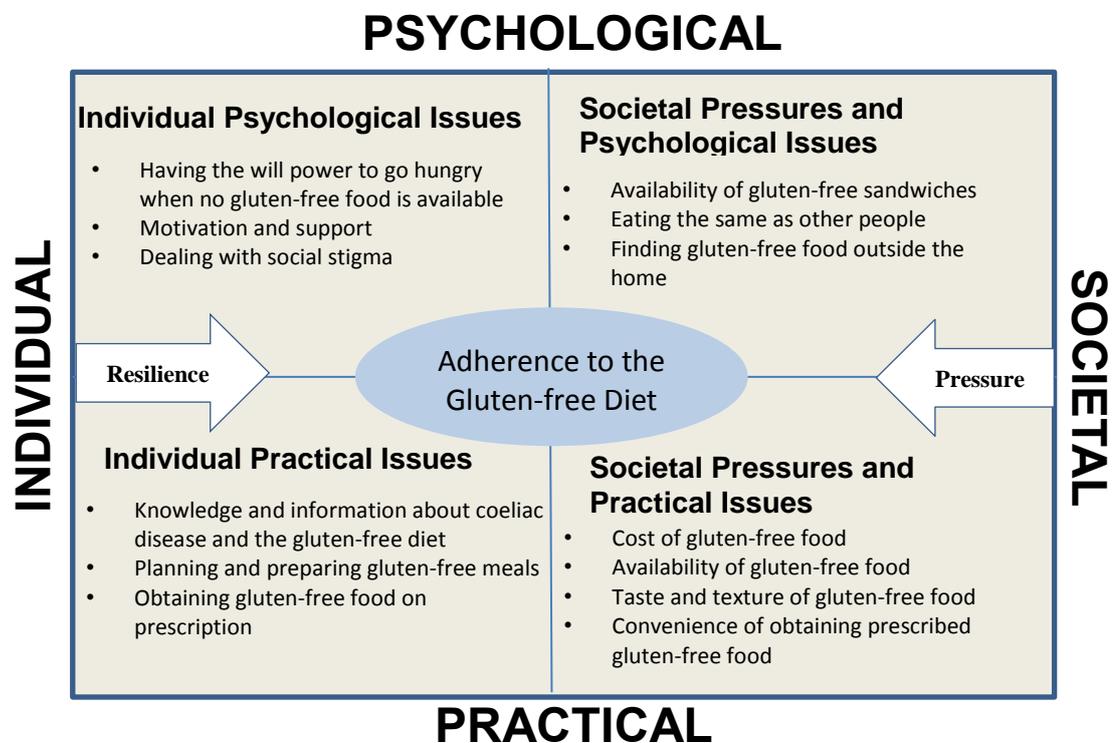


Figure 5.1 A model of adherence to a GFD in adults with CD

5.6.1 Society's responsibility/Psychological issues quadrant (top right)

Four of the 13 concepts (clusters) from the concept map are represented in this quadrant:

- Cluster 2 - The availability of GF sandwiches
- Cluster 6 - If they can eat the same as other people
- Cluster 7 - Eating away from home
- Cluster 13 - Provision of GFF at work

The theme of this quadrant relates to the availability of GFF and the difficulties associated with following a GFD in a society where gluten is regularly consumed. By being located on the right-hand side of the model, this demonstrates that the concepts in this quadrant are mostly the responsibility of society, rather than the individual. Poor availability of GFF has a psychological impact on patients, often leaving them feeling deprived and embarrassed for 'making a fuss' (Sverker *et al.*, 2005; Zarkadas, 2006).

With the exception of the sociodemographic and sociocultural/environmental factors, which spanned all four quadrants, none of the other themes from the systematic review (Hall *et al.* 2009) were closely linked with this quadrant.

5.6.2 Society's responsibility/practical issues quadrant (bottom right)

Four of the 13 concepts (clusters) from the concept map and one of the themes from the systematic review are represented in this quadrant:

- Cluster 1 - The cost of GFF
- Cluster 4 - Access to good quality GFF
- Cluster 5 - Prescribed GFF) also in next quadrant
- Cluster 11 - Convenience of obtaining prescribed GFF
- Systematic review theme - Healthcare treatment factors

This quadrant represents the interplay of factors that cause practical difficulties for patients when trying to stick to a GFD and the issues are mostly influenced by societal factors. Issues relating to the availability of GFF are covered in this quadrant as well as the previous one, however, in this quadrant the impact of poor availability is related to the practical effect this has on patients. Access to prescribed GFF is also included in this cluster along with the difficulties associated with obtaining GFF from a pharmacy. The cost of GFF is also a practical issue related to following a GFD and this was shown to be the most important factor affecting adherence to a GFD from the perspectives of all three stakeholder groups.

5.6.3 Individual responsibility/Practical issues quadrant (bottom left)

Four of the 13 concepts (clusters) from the concept map and two of the themes from the systematic review are represented in this quadrant:

- Cluster 3 - Knowledge and information about CD and the GFD
- Cluster 5 - Prescribed GFF) also in previous quadrant
- Cluster 9 - Motivation and support
- Cluster 12 - Diet planning and preparation
- Systematic review theme - Knowledge, attitudes and beliefs
- Systematic review theme - Illness and symptom factors (also under individual / practical issues)

The themes in this quadrant relate to the knowledge, skills and motivation patients require if they are to stick to a GFD. These themes also relate to the knowledge and skills that healthcare professionals, friends, family, work colleagues etc. are required to possess in order to support the person with CD. The quadrant includes the importance of motivation for patients and this could include being motivated by other people as well as self-motivation.

5.6.4 Individual responsibility / Psychological issues quadrant (top left)

Two of the 13 concepts (clusters) from the concept map and two of the themes from the systematic review are represented in this quadrant:

- Cluster 8 - If they are prepared to go hungry when there is no GFF available
- Cluster 10 - Social stigma
- Systematic review theme - Illness and symptom factors (also under individual / practical issues)
- Systematic review theme - Quality of life and psychological well-being

The themes in this final quadrant relate to the psychological issues associated with adherence to a GFD. Issues such as will power, determination and intention as well as the psychological effects of having CD are presented in this quadrant. Issues in this quadrant could be tackled to help patients stick to a GFD with the use of the models of behaviour as discussed in Section 5.5.

Overall, the model of adherence represents the wide range of factors associated with adherence to a GFD. This model demonstrates that adherence is not the sole responsibility of the patient. CD impacts patients at both a psychological and practical level and an intervention to improve adherence would address these wide-ranging factors.

In the following section I discuss how this model could be utilised in practice along with recommendations for future work and I end this chapter by discussing the limitations of this study.

5.7 Future work

To improve non-adherence it is important to understand why patients with CD do not stick to a GFD. This study has identified a wide-range of factors affecting adherence to a GFD. More research is needed to understand how these factors influence adherence and how they can be addressed. Conflicting evidence exists with regards to the relationship between demographic factors and adherence to a GFD. This was highlighted by the fact that the one statement relating to gender was given the lowest priority rating of all 91 statements. In addition, the systematic review found conflicting evidence in relation to gender and adherence to a GFD. More research is needed into the non-sociodemographic influences on adherence.

An intervention to improve adherence to a GFD would need to take into account the societal and individual influences on adherence as well as the practical and psychological aspects of following a GFD. The model of adherence developed from the results of this study would provide a useful framework for developing an intervention.

An online intervention could be the next logical step to help improve adherence in patients with CD. An online intervention should be relatively cheap to deliver, however not all homes in the UK have online access, so other delivery methods would also be required (e.g. face-to-face or telephone delivery). Sainsbury *et al.* (2013b) conducted a randomised controlled (RCT) trial in Australia which was an online intervention of six training modules that addressed many of the issues identified in this study. This RCT showed improvements in adherence to a GFD, however, only 50% of participants completed the modules. A similar intervention could be developed for a UK audience that is designed around the model of adherence presented in this chapter.

In order to help and support patients with CD, healthcare professionals are required to have sufficient knowledge and understanding of the difficulties faced when following a GFD. Therefore, a similar training package would be a useful and cost-effective method of training for healthcare professionals. This would have the potential of improving healthcare for patients.

5.8 Limitations of this study

This study possibly suffers from selection bias. Participants were self-selected and the 100 Coeliac UK members who were invited came from a population that is predominantly female, white and of a higher educational

level (Ford *et al.*, 2012). Participants were mostly recruited within approximately a 10 mile radius of Norwich which may not be representative of the wider UK population. For example, this area is not as ethnically diverse as many other parts of the UK. All but one of the 73 participants were white British. Previous studies suggest that participants from Asian ethnic groups are less adherent to a GFD and the factors affecting adherence for them may differ from my findings (Butterworth *et al.*,2004).

I only included one hospital in the recruitment of participants and this university hospital may not be representative of an average hospital in the UK. Due to this the results may not be generalisable to the wider population. By recruiting through several channels, many of the limitations identified for each individual recruitment method was reduced. For example, one limitation of recruiting through Coeliac UK is that the sample of members may not be representative of the wider population of adults with CD. By also recruiting through the local press and NNUH, the sample population could be said to be more representative.

As I had limited funds available for this study, it was not possible to obtain a clinical assessment of adherence to a GFD and I relied on self-reporting by patients. However, accurate measurement of adherence was unnecessary as the aim of this study was to recruit a group of participants with a range of levels of adherence, not to measure adherence per se. Only one fully non-adherent participant with CD volunteered to take part in the study and this participant took part in brainstorming, but not prioritising or clustering.

Although this study has identified several factors associated with adherence to a GFD, further studies with larger sample sizes are needed.

Some of the data from the prioritising and clustering tasks was either missing or could not be used. This may have affected the results of the study. However, the software designer advised me that the impact of removing the clusters containing a small number of statements should have little impact on the results.

Overall, the participants included in this study provided a broad range of opinions in relation to the factors affecting adherence to a GFD. However, the sample included only one non-adherent patient with CD, one patient from a non-white British ethnic group, one male healthcare professional, one GP and one gastroenterologist. The majority of participants were recruited from Norfolk and the results of this study may not be generalisable to the wider population.

5.9 Summary

This is the first study to explore the factors affecting adherence to a GFD in adults with CD using concept mapping. CD is a common and debilitating condition affecting approximately 1% of the population. There is no cure for CD, however, it is effectively treated with a strict GFD. Although it is clear that lifelong adherence to a GFD is difficult, we don't fully understand the

reasons for non-adherence. Up to 58% of people with CD do not adhere to this treatment and this increases their risk of developing serious health problems. The aim of this study was to gain a better understanding of the factors that affect adherence to a GFD from the perspectives of adults with CD, household members and healthcare professionals using concept mapping. Concept mapping was a useful method for studying the range of interconnected factors associated with adherence to a GFD and achieving the aims of this study.

Overall, I identified 13 concepts relating to adherence to a GFD from the perspectives of patients, household members and healthcare professionals. Concept mapping allowed me to identify the complex interplay of factors associated with adherence to a GFD and their relative importance. The high cost of gluten-free food was identified as the most important factor. The results of this study suggest that reducing the cost of GFF may be a primary target for an adherence intervention. However, it is possible that the high cost of GFF is a 'salt on the wound' issue that people are dissatisfied with, but it may not actually prevent adherence. The high cost of gluten-free food can be mitigated by providing gluten-free food on prescription. However, the effectiveness and practicality of the current method of prescribing GFF to reduce patient costs needs to be reviewed.

Knowledge about coeliac disease and the gluten-free diet has been shown to be a complex construct that does not solely reside in the person with coeliac disease. The knowledge of healthcare professionals, family, friends and staff

in restaurants is important in supporting people with coeliac disease in sticking to a gluten-free diet.

There was little difference between perceptions of the three stakeholder groups around the factors affecting adherence to a GFD. Healthcare professionals need to understand the pervasive provision of lunchtime sandwiches can be very impactful day to day, and that patients with coeliac disease may have to decide to go hungry rather than become non-adherent if they cannot find any gluten free foods when away from home. Following a GFD for life requires a good deal of planning and preparation.

Together, the results of my concept mapping study and systematic review were used to develop a comprehensive model of adherence to a GFD which can be used in designing an intervention to improve adherence to a GFD. It is anticipated that improvements in adherence to a GFD will improve the lives of people with CD and reduce NHS costs. This model of adherence to a GFD takes into account the wide range of factors that impact on adherence.

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Appendices

Appendix 1 - Systematic review search strings

Seven database searches were carried out on 16 August 2013. The full search string used for each database is shown below along with the number of articles found (hits). The search included articles published between 2007 and August 2013. Where the database allowed, I restricted the search to full articles, English language and human studies only.

1. AMED (Ovid) (1806-2013) - systematic review search string

Number of hits: 1

Search string:

Celiac OR Coeliac OR Gluten sensitiv\$ OR Gluten-sensitiv\$ enteropath\$
AND
Adher\$ OR Comply OR Complian\$ OR Concordan\$ OR Manag\$ OR Non-
adher\$ OR Non-complian\$
AND
Gluten OR Gluten-free OR Diet\$ OR Treat\$ OR Therap\$

2. CINAHL Plus (EbscoH) - systematic review search string

Number of hits: 221

Search string:

Celiac OR Coeliac OR Gluten sensitive* OR Gluten-sensitiv* enteropath*
AND
Adher* OR Comply OR Complian* OR Concordan* OR Manag* OR Non-
adher* OR Non-complian*
AND
Gluten OR Gluten-free OR Diet* OR Treat* OR Therap*

3. Cochrane Library - systematic review search string

Number of hits: 45

Search string:

Celiac disease/ (MESH term) OR Coeliac OR Gluten sensitiv* OR Gluten-
sensitive enteropath*
AND
Adher* OR Comply OR Complian* OR Concordan* OR Manag* OR Non-
adher* OR Non-complian*
AND
Diet, Gluten-free (MESH term) OR Gluten OR Gluten-free OR Diet* OR
Treat* OR Therap*

4 EMBASE (Ovid) - systematic review search string

Number of hits: 1011

Search string:

Celiac disease/ (MESH term) OR Coeliac OR Gluten sensitiv\$ OR Gluten-sensitiv\$ enteropath\$
AND
Adher\$ OR Comply OR Complian\$ OR Concordan\$ OR Manag\$ OR Non-adher\$ OR Non-complian\$
AND
Diet, Gluten-free/(MESH term) OR Gluten OR Glutens/ (MESH term) OR Gluten-free OR Diet/ (MESH term) OR Treat\$ OR Therap\$

5. Medline Ovid (1948-2012) - systematic review search string

Number of hits: 488

Search string:

Celiac disease/ (MESH term) OR Coeliac OR Gluten sensitiv\$ OR Gluten-sensitive enteropath\$
AND
Adher\$ OR Comply OR Complian\$ OR Concordan\$ OR Manag\$ OR Non-adher\$ OR Non-complian\$
AND
Diet, gluten-free/(MESH term) OR Glutens/ (MESH term) OR Gluten OR Gluten-free OR Treat\$ OR Therap\$

6. PsychINFO (1806-2013) - systematic review search string

Number of hits: 7

Search string:

Celiac OR Coeliac OR Gluten sensitiv\$ OR Gluten-sensitive enteropath\$
AND
Adher\$ OR Comply OR Complian\$ OR Concordan\$ OR Manag\$ OR Non-adher\$ OR Non-complian\$
AND
Gluten OR Gluten-free OR Diet\$ OR Treat\$ OR Therap\$

7. PubMed Central - systematic review search string

Number of hits: 39

Search string:

Celiac Disease/ (MESH term) OR Coeliac OR Gluten sensitive* OR
Gluten-sensitive enteropath\$

AND

Adher* OR Comply OR Complian* OR Concordan* OR Manag* OR
Non-adher* OR Non-complian*

AND

Gluten OR Gluten-free OR Diet* OR Nutrition* OR Treat* OR Therap*

Appendix 2 - Systematic review excluded papers

The table below shows a list of the 71 studies that were excluded from the systematic review after reading the full paper.

Authors	Title	Year	Reason for exclusion
Angeli et al.	An epidemiologic survey of celiac disease in the Terni area (Umbria Italy) in 2002-2010	2012	Not factors affecting adherence
Araujo et al.	Coeliac disease. Following the diet and eating habits of participating individuals in the Federal District, Brazil.	2011	Age range of participants is not specified – the results may include patients aged <18 years
Arigo	Psychiatric comorbidities in women with celiac disease	2011	This paper makes the association between adherence and various symptoms, but not the factors affecting adherence.
Autodore	Celiac Disease and its treatment	2012	Not factors affecting adherence
Aziz et al.	Are patients with coeliac disease seeking alternative therapies to a gluten-free diet?	2011	Not factors affecting adherence
Bakshi et al.	Emerging therapeutic options for celiac disease: Potential alternatives to a gluten-free diet.	2012	Not factors affecting adherence
Barada et al.	Celiac disease in the developing world.	2012	Not primary research.
Bellini et al.	Compliance with the gluten-free diet: The role of locus of control in celiac disease	2011	Includes children and adolescents.
Biagi et al.	A gluten-free diet score to evaluate dietary compliance in patients with coeliac disease.	2009	Measuring compliance – not factors affecting adherence/compliance.
Biagi et al.	A score that verifies adherence to a gluten-free diet: A cross-sectional, multicentre validation in real clinical life.	2012	Measuring compliance – not factors affecting adherence/compliance.
Blazina et al.	Bone mineral density and importance of strict gluten-free diet in children and adolescents with celiac disease.	2010	No separate data for 18 year olds
Bold & Rostami	Gluten tolerance; potential challenges in treatment strategies	2011	Not factors affecting adherence

Cammarota et al.	A highly accurate method for monitoring histological recovery in patients with celiac disease on a gluten-free diet using an endoscopic approach that avoids the need for biopsy: a double-center study.	2007	Not factors affecting adherence.
Cassellas et al.	Factors that impact health-related quality of life in adults with celiac disease: A multicenter study	2008	Includes 15 to 17 year olds.
Chawla	Diagnosis and management of celiac disease	2010	Paper unavailable (interlending too expensive)
Comino et al.	Monitoring of gluten-free diet compliance in celiac patients by assessment of gliadin 33-mer equivalent epitopes in feces.	2012	Not factors affecting adherence.
Cosnes et al.	Incidence of autoimmune disease in celiac disease: protective effect of gluten-free diet	2008	Not factors affecting adherence
Cureton	The gluten-free diet: Can your patient afford it?	2007	Not primary research
Da Silva Kotze	A Brazilian experience of the self transglutaminase-based test for celiac disease case finding and diet monitoring.	2009	Not factors affecting adherence
Dipper et al.	Anti-tissue transglutaminase antibodies in the follow-up of adult coeliac disease.	2009	Not factors affecting adherence
Gabrovska	Monitoring of daily gliadin intake in patients on gluten-free diets.	2011	Not factors affecting adherence
Garcia-Manzanares et al.	Nutritional and dietary aspects of celiac disease	2011	Not primary research
Hauser et al.	Anxiety and depression in adult patients with celiac disease on a gluten-free diet.	2010	Measures anxiety and depression, but not in relation to adherence
Hauser et al.	Predictors of irritable bowel-type symptoms and healthcare-seeking behaviour among adults with celiac disease.	2007	Only included adherent participants. Not factors affecting adherence
Hauser et al.	Predictors of reduced health-related quality of life in adults with coeliac disease.	2007	Not factors affecting adherence
Herman et al	Patients with celiac disease are not followed up adequately.	2012	Not factors affecting adherence

Holmes & Moor	Coeliac disease in Asians in a single centre in southern Derbyshire.	2012	
Hopman et al.	Gluten tolerance in adult patients with celiac disease 20 years after diagnosis?	2008	Not factors affecting adherence
Hwang et al.	Duodenal histology vs. celiac disease-specific serology: which is the best tool for assessing compliance with the gluten-free diet at one year after diagnosis?	2013	Not factors affecting adherence
Jacobsson et al.	Impact of an active patient education program on gastrointestinal symptoms in women with celiac disease following a gluten-free diet: a randomized controlled trial.	2012	Only included women who were adherent to GFD. Not factors affecting adherence
Kaukinen et al.	Coeliac disease – A diagnostic and therapeutic challenge.	2010	Not factors affecting adherence
Kempainen et al.	Unkilned and large amounts of oats in the coeliac disease diet: A randomized, controlled study.	2008	Not factors affecting adherence
Kinsey et al.	A dietary survey to determine if patients with coeliac disease are meeting current healthy eating guidelines and how their diet compares to that of the British general population	2008	Not factors affecting adherence
Kurpa et al.	Factors associated with dietary adherence to gluten-free diet.	2012	Includes adults and children
Lanzini et al.	Complete recovery of intestinal mucosa occurs very rarely in adult coeliac patients despite adherence to gluten-free diet.	2009	Not factors affecting adherence
Lee et al.	Economic burden of a gluten-free diet.	2007	Assessment of the cost and availability of gluten-free products, but not in relation to adherence
Leffler et al.	A simple validated gluten-free diet adherence survey for adults with celiac disease.	2009	Measuring adherence, not factors affecting adherence.

Lerner	New therapeutic strategies for celiac disease.	2010	Not primary research
Lovik et al.	Oats in a strictly gluten-free diet is associated with decreased gluten intake and increased serum bilirubin.	2010	This study evaluated the effects of oat consumption on adherent adults with CD, not related to adherence or factors affecting adherence to GFD
Mancini et al.	Celiac disease and the athlete.	2011	Not primary research
Mounajjed et al.	The liver in celiac disease: Clinical manifestations, histologic features, and response to gluten-free diet in 30 patients.	2011	Not factors affecting adherence
Nachman et al.	Quality of life in celiac disease patients. Prospective analysis on the importance of clinical severity at diagnosis and the impact of treatment.	2009	Not factors affecting adherence
O'Donnell	Dietitians' perceptions of adherence to a gluten-free diet among low-income individuals with celiac disease	2009	The patient age range is not specified, but the results mention missing days off school, which suggests children were included in the Dietitians' reports.
Olen et al.	Coeliac disease characteristics, compliance to a gluten free diet and risk of lymphoma by subtype.	2011	Not factors affecting adherence
Olsson et al.	The everyday life of adolescent coeliacs: issues of importance for compliance with the gluten-free diet.	2008	<18 years of age
Olsson et al.	Food that makes you different: the stigma experienced by adolescents with celiac disease.	2009	Includes participants aged 18 years but the data is combined with children aged <18 years
Paarlahti et al.	Predictors of persistent symptoms and reduced quality of life in treated coeliac disease patients: A large cross-sectional study.	2013	Factors associated with symptoms, not adherence
Purnak et al.	Mean platelet volume could be a promising biomarker to monitor dietary compliance in celiac disease.	2011	Not factors affecting adherence
Roos et al.	Gastrointestinal symptoms and well-being of adults living on a gluten-free diet: a case for nursing in celiac disease.	2009	Gluten-free diet and wellbeing, not factors affecting adherence

Rubio-Tapia et al.	Mucosal recovery and mortality in adults with celiac disease after treatment with a gluten-free diet.	2010	Not factors affecting adherence
Ryan & Grossman	Celiac disease: implications for patient management.	2011	Not factors affecting adherence
Sainsbury et al.	Reduced quality of life in coeliac disease is more strongly associated with depression than gastrointestinal symptoms.	2013	This is the same as an included study (Sainsbury et al. 2013)
Sainsbury et al.	A randomized controlled trial of an online intervention to improve gluten-free diet adherence in celiac disease	2013	The inclusion criteria was age >16 years. Data were not presented for participants aged \geq 18 years
Shamir	Population screening for celiac disease: Follow up of patients identified by positive serology.	2007	Age of participants not reported. Researcher based at a children's hospital.
Shepherd & Gibson	Nutritional inadequacies of the gluten-free diet in both recently-diagnosed and long-term patients with coeliac disease.	2013	Nutritional intake, not factors affecting adherence
Silano et al.	Effect of a gluten-free diet on the risk of enteropathy-associated T-cell lymphoma in celiac disease	2008	Consequence of non-adherence, not factors affecting adherence.
Sverker et al.	Sharing life with a gluten-intolerant person—the perspective of close relatives	2007	How coeliac disease affects close relatives, not factors affecting adherence.
Terasaki & Ajam	Case report: Revisiting celiac disease.	2009	Not factors affecting adherence
Tursi et al.	Complications in celiac disease under gluten-free diet.	2009	Adherence is measured, but not factors affecting adherence.
Ukkola	Patients' experiences and perceptions of living with coeliac disease - implications for optimizing care	2012	Includes 16 to 17 year olds, but not separate data for this age group.
Usai et al.	Effect of gluten-free diet and co-morbidity of irritable bowel syndrome-type symptoms on health-related quality of life in adult coeliac patients.	2007	Quality of life, not factors affecting adherence

Veen	Quitting is not an option: An analysis of online diet talk between celiac disease patients.	2010	Age of participants not known
Vilppula et al.	Clinical benefit of gluten-free diet in screen-detected older celiac disease patients.	2011	Not factors affecting adherence
Violato et al.	Resource use and cost associated with coeliac disease before and after diagnosis in 3,646 cases: Results of a UK primary care database analysis.	2012	Not factors affecting adherence
Volta et al.	Usefulness of antibodies to deamidated gliadin peptides in celiac disease diagnosis and follow-up.	2008	Measure of adherence, but not factors affecting adherence
Wagner et al.	Quality of life in adolescents with treated coeliac disease: influence of compliance and age at diagnosis	2008	Children and adolescents - No separate data for 18 year olds
Whitaker	Patient perceptions of the burden of coeliac disease and its treatment in the UK	2009	Assessing the burden of coeliac disease, not factors affecting adherence
Whitehead	Obesity and coeliac disease: possible effects of the gluten-free diet.	2013	Not primary research.
Wild et al.	Evidence of high sugar intake, and low fibre and mineral intake, in the gluten-free diet.	2010	Not factors affecting adherence
Zanchi et al.	Rapid anti-transglutaminase assay and patient interview for monitoring dietary compliance in celiac disease	2013	Not factors affecting adherence
Zanini	Five year time course of celiac disease serology during gluten-free diet: Results of a community based "CD-watch" program.	2010	Measure of adherence, not factors affecting adherence.

Appendix 3 Characteristics of the studies included in systematic review

Article	Country	Study Design	Study Aims	Source of Participants	Participant Characteristics			Adherence Assessment	% non-adherence	Factors Affecting Adherence
					Intervention /study group	Control or comparison group/s	Total			
Barratt <i>et al.</i> , 2011	UK	Cross-sectional	To assess which factors impact on quality of life in CD	Recruited during outpatient appointments	n=225 26% male Mean duration on GFD = 8 years	n=348	n=573	Reported as 'fully-adherent' = every day for past 28 days; partially adherent = at least half of these days; non-adherent = none of these days.	65% adherent; 31% partially-adherent; 4% non-adherent.	Quality of life; Age, Marriage, Affluence; Education; Gender; Disease duration; Screening detection.
Black <i>et al.</i> , 2011	UK	Cross-sectional	To investigate the effect of CD and GFD on dietary habits and QoL	Postal invitation to Coeliac UK Members	N=146 18% male Biopsy-confirmed	n/a	n=146	Food frequency questionnaire	96% adherent	Availability of breakfast cereals; eating out/socialising; eating convenience foods.
Casella <i>et al.</i> , 2012	Italy	Retrospective cohort	To compare CD in older and younger adults and to assess the effects of a GFD	Identified from records on a database	N = 59 Older patients ≥ 65 years	n=1166 Younger patients 18-64 years	n=1225	Serology and histological characteristics tested before and during GFD. Assessment during interview by physician.	90% adherent.	Age

Edwards-George 2009	USA	Cross-sectional	To determine the relationship between personality traits, psychological symptoms and GFD adherence	Outpatients at clinic	n=157 Following GFD for >3 months Mean age 50.3 years	n/a	n=157	Assessment by expert Dietitian	44.2% Excellent adherence 34.4% Good adherence 21% Non-adherent	Mood (depression); Anxiety; Low values trait; low conscientiousness; low order trait; low deliberation trait; low self-discipline trait; other food intolerance; presence of symptoms when gluten is consumed; Availability of gluten-free food; Cost of gluten-free food.
Errichello <i>et al.</i> , 2010	Italy	Cross-sectional	To identify risk and protective factors in relation to GFD adherence in a group of young people	Outpatient clinic	n=204 38% male Aged 13-30 years.	n/a	n=204	Interview	73.5% no dietary transgressions; 26.5% occasional or frequent transgressions.	Non-smoker

Ford <i>et al.</i> , 2012	UK	Cross-sectional	To explore the illness perceptions and self-efficacy beliefs of adults with CD and to report their subjective levels of HRQoL	Coeliac UK	n=228 20% male 95% White British Mean age of males = 61 years Mean age for females = 52 years	n/a	n=228	Self-reported on a 5-point Likert scale	87% adherent 13% partially-adherent	Belief in cyclical nature/ chronicity of coeliac disease; Consequences of non-adherence are serious; Self-efficacy; Age; Psychological wellbeing; Quality of life.
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Hall <i>et al.</i> , 2013	UK	Cross-sectional	To determine the rates of intentional and inadvertent non-adherence and to examine the factors associated with both	Patients registered with family practices	n=287	n/a	n=287	Self-reported	40.1% reported intentional gluten consumption in the last 6 months.	Membership of coeliac support group; Better quality of gluten-free food; Cost of gluten-free food; Receiving gluten-free food on prescription; Regular follow-up; Better choice of gluten-free products; Clearer information when eating out; Availability of gluten-free food; Perceived tolerance to gluten; Clearer and universal labelling; Age at diagnosis; self-efficacy; gender
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Hopman <i>et al.</i> , 2009	UK	Cross-sectional	To investigate whether dietary (non-) compliance is associated with health-related quality of life (HRQoL) of celiac patients	Hospital patients	n=33 following a GFD	n=8 gluten-transgressions (partially adherent) n= 12 normal gluten-containing diet (non-adherent)	n=53	Self-reported adherence. Food frequency questionnaire	38% partially- or non-adherent	Duration of gluten-free diet; Osteoporosis; Osteopenia; Symptoms if gluten is consumed; Age; Age at diagnosis
Hutchinson <i>et al.</i> , 2010	UK	Retrospective cohort	To examine the time to histopathological recovery and to assess correlations between histopathological disease score, gender, age and compliance with a gluten-free diet	Hospital patients	n=284	n/a	n=284	Self-reported. Biopsy	12% poor adherence (88% good adherence)	Age at diagnosis; Gender
Kabbani <i>et al.</i> , 2012	USA	Retrospective cohort	To assess changes in BMI after diagnosis in a large coeliac population	Coeliac clinic (electronic records)	1018 24% male Biopsy proved CD	n/a	n=1018	Assessment by expert Dietitian. Gastroenterologists' notes and serology results.		Age; Gender; BMI; Ethnicity (White)

Kurpa <i>et al.</i> , 2012	Finland	Cross-sectional	To establish whether the shift of diagnostics and follow-up of coeliac disease from tertiary centres to secondary has affected the success of treatment, and to identify predictors for dietary non-adherence	Hospital patients	n=843 n=94 <18 years n=749 ≥18 years	n/a	n=843 (of which 749 were adults)	Structured dietary interview	10% partially adherent (no non-adherent).	Age
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Lee <i>et al.</i> , 2012	USA	Cross-sectional	To investigate the impact of both coeliac disease and the gluten-free diet on quality of life	Coeliac support groups	n=1743 23% male 26% age 18-45 44% age 46-65 30% over 65	n=1179 61% male 30% age 18-45 43% age 45-65 28% over 65	n=2922	Self-reported	Males and females reported 98% compliance However, a surprising number admitted to dietary indiscretion	Eating away from home (with friends and in restaurants); The poor taste of gluten-free food; Cost of gluten-free food; Restrictiveness of the diet; A gluten-free diet is difficult to follow; Anxiety (the gluten-free diet is difficult in social settings); Quality of life; Gender.
Mahadev <i>et al.</i> , 2013	USA	Cross-sectional	To determine if Dietitian use was associated with quality of life, symptom severity or gluten-free diet adherence	Celiac Disease Center of Colombia University	n=413 23% male 24% aged >60 years	n/a	n=413	CD adherence test (CDAT)	Not stated	Dietitian use

Nachman <i>et al.</i> , 2010	Argentina	Prospective cohort	To assess quality of life in coeliac disease and to determine the influence of gluten-free diet adherence on quality of life	Patients at a small bowel disease clinic	n=53 Newly diagnosed, biopsy-confirmed Mean age 38 years 9% male	n=70	n=123	Assessment by lead physician, interview with Dietitians and self-report.	Alimentary assessments did not identify any patients as non-compliant at all, possibly because those on unrestricted diet voluntarily missed the scheduled visits. At the 4-year visit, strictly compliant (n=27) and partially compliant patients (n=26)	Quality of life
Paavola <i>et al.</i> , 2012	Finland	Cross-sectional	To investigate the effect of long-term gluten-free dietary treatment on gastrointestinal symptoms and psychological well-being	Newspaper advertisements and coeliac support group newsletter	n= 96 screen detected n= 370 non-screen detected	n= 110 Non-coeliacs	n=576	Structured interview	88% adherent	Screen detection/non-screen detection (presence of symptoms at diagnosis).

Sainsbury <i>et al.</i> , 2011	Australia	Cross-sectional	To predict the level of gluten-free diet adherence in adults with coeliac disease	Coeliac support group	Phase 1 (elicitation interviews n=13 (7 male) Phase 2 (online survey) n=265 16.6% male	n/a	n=278	Self-reported (CDAT)	47.5% excellent or very good adherence. 35.8 moderate adherence 7.9% fair to poor adherence. 8.8% missing data.	Anxiety (social stigma/not wanting to cause offence or inconvenience others); Depression; Quality of life; Belief in long-term consequences of non-adherence; Knowledge of gluten-free ingredients; Trusting others to prepare gluten-free food.
Sainsbury <i>et al.</i> , 2013	Australia	Cross-sectional	To examine the potential role of psychological symptoms in limiting the translation of positive intention into strict gluten free diet adherence	Coeliac support group	n=390 17.2% male Mean age 44.2 years	n/a	n=390	Self-reported (CDAT)	56.7% excellent/very good; 37.2% moderate; 6.2% fair/poor.	Limited options; Severity of symptoms; Depression; Task-oriented coping; Emotion-oriented coping; Intention to adhere; Risk of eating disorders.

Sey <i>et al.</i> , 2011	Canada	Prospective cohort	To test the safety of oats	Patients at London Health Sciences Centre	n=15 adults with biopsy confirmed CD for ≥ 1 year Asymptomatic on GFD for 1 year and normal tTG level.	n/a	n=15	Random food diaries (self-report)	All participants were adherent at the start of the study (this was an inclusion criterion)	Travelling (Eating away from home).
Smith and Goodfellow 2011	USA	Cross-sectional	To examine factors and perceived causes that interfere with adherence to a gluten-free diet, identify coping strategies and examine the relationship between coping strategies and quality of life	Gluten intolerance group (GIG) website.	n=156 Mean age 51.5 years	n/a	n=156	Inclusion criterion was that participants were on a GFD for 6 months (adherence was not measured in any other way)	n/a	Eating with family or friends; Following a gluten-free diet in social settings is uncomfortable; Eating at a restaurant; Travelling; Dislike the taste of gluten-free food; Cost of gluten-free products

Sverker <i>et al.</i> , 2009	Sweden	Cross-sectional	To explore consequences of dilemmas in everyday lives for women and men, as personally affected by coeliac disease or as close relatives to someone affected and to put these experiences into context regarding household activities	Hospital patients (identified from Dietitians' patient registers)	n=43 adults with CD (32 female; 11 male) n= 23 close relatives (17 male; 6 female) 28 male 38 female	n/a	n=66	None	n/a	Hunger leads to gluten consumption if nothing gluten-free is available
Van Hees <i>et al.</i> , 2013	The Netherlands	Cross-sectional	To investigate whether long-term adherence to a gluten-free diet is related to depressive symptoms in coeliac disease patients	Coeliac support group	n=2265 Average age 52 years (range 18-93 years)	n/a	n=2265	Self-reported	50.2%=strict adherence 46.3% sufficient 3.6% insufficient.	Mood (depression)

Appendix 4 Ethics application



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8 April 2011

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Dear Leigh,

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

I wish to submit the above research project proposal for proportionate review by the Newcastle North Tyneside 2 Research Ethics Committee on 21 April 2011. This is a PhD student research project which is being funded entirely by the University of East Anglia.

The attached paperwork includes separate documents for the three stakeholder groups who will participate in this study. I have inserted a reference in the top right corner of each document to identify which stakeholder group the document relates to:

AWCD = Adults with coeliac disease

HM = Household members

HP = Healthcare professionals

I look forward to hearing from you in due course.

Yours faithfully

Helen Flaherty
Chief Investigator

Welcome to the Integrated Research Application System

IRAS Project Filter

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please enter a short title for this project (maximum 70 characters)

What helps adults with coeliac disease stick to a gluten-free diet? V1

1. Is your project research?

Yes No

2. Select one category from the list below:

- Clinical trial of an investigational medicinal product
- Clinical investigation or other study of a medical device
- Combined trial of an investigational medicinal product and an investigational medical device
- Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- Basic science study involving procedures with human participants
- Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- Study involving qualitative methods only
- Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- Study limited to working with data (specific project only)
- Research tissue bank
- Research database

If your work does not fit any of these categories, select the option below:

Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? Yes No
- b) Will you be taking new human tissue samples (or other human biological samples)? Yes No
- c) Will you be using existing human tissue samples (or other human biological samples)? Yes No

3. In which countries of the UK will the research sites be located? (Tick all that apply)

- England
- Scotland
- Wales
- Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

- England
 Scotland
 Wales
 Northern Ireland
 This study does not involve the NHS

4. Which review bodies are you applying to?

- NHS/HSC Research and Development offices
 Social Care Research Ethics Committee
 Research Ethics Committee
 National Information Governance Board for Health and Social Care (NIGB)
 National Offender Management Service (NOMS) (Prisons & Probation)

For NHS/HSC R&D offices, the CI must create Site-Specific Information Forms for each site, in addition to the study-wide forms, and transfer them to the PIs or local collaborators.

5. Will any research sites in this study be NHS organisations?

- Yes No

6. Do you plan to include any participants who are children?

- Yes No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

- Yes No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the NIGB Ethics and Confidentiality Committee to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

- Yes No

9. Is the study or any part of it being undertaken as an educational project?

- Yes No

Please describe briefly the involvement of the student(s):

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

- Yes No

10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

Yes No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

Yes No

Integrated Research Application System**Application Form for Research administering questionnaires/interviews for quantitative analysis or mixed methodology study**

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting [Help](#).

Please define any terms or acronyms that might not be familiar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
What helps adults with coeliac disease stick to a gluten-free diet? V1

PART A: Core study information**1. ADMINISTRATIVE DETAILS****A1. Full title of the research:****A2-1. Educational projects**

Name and contact details of student(s):

Name and contact details of academic supervisor(s):

Please state which academic supervisor(s) has responsibility for which student(s).
Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

A2-2. Who will act as Chief Investigator for this study?

- Student
 Academic supervisor
 Other

A3-1. Chief Investigator:

Title Forename/Initials Surname

Post
Qualifications
Employer
Work Address

Post Code
 Work E-mail
 * Personal E-mail
 Work Telephone
 * Personal Telephone/Mobile
 Fax

** This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.
 A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.*

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project?
This contact will receive copies of all correspondence from REC and R&D reviewers that is sent to the CI.

Title Forename/Initials Surname

Address

Post Code
 E-mail
 Telephone
 Fax

A5-1. Research reference numbers. *Please give any relevant references for your study:*

Applicant's/organisation's own reference number, e.g. R & D (if available):

Sponsor's/protocol number:

Protocol Version:

Protocol Date:

Funder's reference number:

Project website:

Additional reference number(s):

Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

Yes No

Please give brief details and reference numbers.

A5-3. US DHHS grant application.

PHS grant application number:
 Name of Program Director:

2. OVERVIEW OF THE RESEARCH

To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.

A6-1. Summary of the study. Please provide a brief summary of the research (maximum 300 words) using language easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, this summary will be published on the website of the National Research Ethics Service following the ethical review.

A6-2. Summary of main issues. Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.

Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, R&D office or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.

A6-3. Proportionate review of REC application The initial project filter has identified that your study may be suitable for proportionate review by a REC sub-committee. Please consult the current guidance notes from NRES and indicate whether you wish to apply through the proportionate review service or, taking into account your answer to A6-2, you consider there are ethical issues that require consideration at a full REC meeting.

Yes - proportionate review No - review by full REC meeting

Further comments (optional):

Note: This question only applies to the REC application.

3. PURPOSE AND DESIGN OF THE RESEARCH

A7. Select the appropriate methodology description for this research. Please tick all that apply:

- Case series/ case note review
- Case control
- Cohort observation
- Controlled trial without randomisation
- Cross-sectional study
- Database analysis
- Epidemiology
- Feasibility/ pilot study
- Laboratory study
- Metanalysis
- Qualitative research
- Questionnaire, interview or observation study
- Randomised controlled trial
- Other (please specify)

A10. What is the principal research question/objective? Please put this in language comprehensible to a lay person.

A11. What are the secondary research questions/objectives if applicable? *Please put this in language comprehensible to a lay person.*

A12. What is the scientific justification for the research? *Please put this in language comprehensible to a lay person.*

A13. Please summarise your design and methodology. *It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.*

A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?

- Design of the research
- Management of the research
- Undertaking the research
- Analysis of results
- Dissemination of findings
- None of the above

Give details of involvement, or if none please justify the absence of involvement.

A14-2. Have you tested the acceptability of using patient identifiable data in this study without consent?

Please give details.

4. RISKS AND ETHICAL ISSUES

RESEARCH PARTICIPANTS

A15. What is the sample group or cohort to be studied in this research?

Select all that apply:

- Blood
- Cancer
- Cardiovascular
- Congenital Disorders
- Dementias and Neurodegenerative Diseases
- Diabetes
- Ear
- Eye
- Generic Health Relevance
- Infection
- Inflammatory and Immune System
- Injuries and Accidents

- Mental Health
 Metabolic and Endocrine
 Musculoskeletal
 Neurological
 Oral and Gastrointestinal
 Paediatrics
 Renal and Urogenital
 Reproductive Health and Childbirth
 Respiratory
 Skin
 Stroke

Gender: Male and female participants

Lower age limit: Years

Upper age limit: Years

A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

RESEARCH PROCEDURES, RISKS AND BENEFITS

A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. These include seeking consent, interviews, non-clinical observations and use of questionnaires.

Please complete the columns for each intervention/procedure as follows:

1. Total number of interventions/procedures to be received by each participant as part of the research protocol.
2. If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
3. Average time taken per intervention/procedure (minutes, hours or days)
4. Details of who will conduct the intervention/procedure, and where it will take place.

A21. How long do you expect each participant to be in the study in total?

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

Yes No

A24. What is the potential for benefit to research participants?

A26. What are the potential risks for the researchers themselves? (if any)

RECRUITMENT AND INFORMED CONSENT

In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? For example, identification may involve a disease register, computerised search of social care or GP records, or review of medical records. Indicate whether this will be done by the direct care team or by researchers acting under arrangements with the responsible care organisation(s).

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

Yes No

Please give details below:

A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?

Yes No

A29. How and by whom will potential participants first be approached?

A30-1. Will you obtain informed consent from or on behalf of research participants?

Yes No

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

If you are not obtaining consent, please explain why not.

Please enclose a copy of the information sheet(s) and consent form(s).

A30-3. Why is it not practicable for either the researcher's organisation, or the current holder of the information required by the researcher, to seek or obtain patient consent for proposed use of patient identifiable information?

A31. How long will you allow potential participants to decide whether or not to take part?

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs?(e.g. translation, use of interpreters)

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the

study? *Tick one option only.*

- The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.
- The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.
- The participant would continue to be included in the study.
- Not applicable – informed consent will not be sought from any participants in this research.
- Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

CONFIDENTIALITY

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

Storage and use of personal data during the study

A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)? *(Tick as appropriate)*

- Access to medical records by those outside the direct healthcare team
- Access to social care records by those outside the direct social care team
- Electronic transfer by magnetic or optical media, email or computer networks
- Sharing of personal data with other organisations
- Export of personal data outside the EEA
- Use of personal addresses, postcodes, faxes, emails or telephone numbers
- Publication of direct quotations from respondents
- Publication of data that might allow identification of individuals
- Use of audio/visual recording devices
- Storage of personal data on any of the following:
- Manual files (includes paper or film)
 - NHS computers
 - Social Care Service computers
 - Home or other personal computers
 - University computers
 - Private company computers
 - Laptop computers

Further details:

A37. Please describe the physical security arrangements for storage of personal data during the study?

A38. How will you ensure the confidentiality of personal data? *Please provide a general statement of the policy and*

procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.

A39. Please specify whether identifiers will be held in the same database as the clinical data, or in a separate database and linked through a unique study or case number. If held separately, please specify how and at what point the separation will occur. If held in the same database, will the identifiers be encrypted? If so, specify what will be encrypted and who will continue to have access.

A40. Who will have access to participants' personal data during the study? Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.

Storage and use of data after the end of the study

A41. Where will the data generated by the study be analysed and by whom?

A42. Who will have control of and act as the custodian for the data generated by the study?

Title Forename/Initials Surname

Post
Qualifications
Work Address

Post Code
Work Email
Work Telephone
Fax

A43. How long will personal data be stored or accessed after the study has ended?

- Less than 3 months
 3 – 6 months
 6 – 12 months
 12 months – 3 years
 Over 3 years

A44. For how long will you store research data generated by the study?

Years:
Months:

A45. Please give details of the long term arrangements for storage of research data after the study has ended. Say where data will be stored, who will have access and the arrangements to ensure security.

INCENTIVES AND PAYMENTS

A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?

Yes No

A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?

Yes No

A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?

Yes No

NOTIFICATION OF OTHER PROFESSIONALS

A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?

Yes No

If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.

PUBLICATION AND DISSEMINATION

A50-1. Will the research be registered on a public database?

Yes No

Please give details, or justify if not registering the research.

Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

A50-2. Will the research be registered on a public database such as the Research Register for Social Care?

Yes No

Please give details, or justify if not registering the research.

A51. How do you intend to report and disseminate the results of the study? Tick as appropriate:

- Peer reviewed scientific journals
- Internal report
- Conference presentation
- Publication on website

- Other publication
- Submission to regulatory authorities
- Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- No plans to report or disseminate the results
- Other (please specify)

A52. If you will be using identifiable personal data, how will you ensure that anonymity will be maintained when publishing the results?

A53. Will you inform participants of the results?

Yes No

Please give details of how you will inform participants or justify if not doing so.

5. Scientific and Statistical Review

A56. How have the statistical aspects of the research been reviewed? Tick as appropriate:

- Review by independent statistician commissioned by funder or sponsor
- Other review by independent statistician
- Review by company statistician
- Review by a statistician within the Chief Investigator's institution
- Review by a statistician within the research team or multi-centre group
- Review by educational supervisor
- Other review by individual with relevant statistical expertise

In all cases please give details below of the individual responsible for reviewing the statistical aspects. If advice has been provided in confidence, give details of the department and institution concerned.

Title Forename/Initials Surname

Department
Institution
Work Address

Post Code
Telephone
Fax
Mobile
E-mail

Please enclose a copy of any available comments or reports from a statistician.

A57. What is the primary outcome measure for the study?

A58. What are the secondary outcome measures? (if any)

A59. What is the sample size for the research? How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.

Total UK sample size:
Total international sample size (including UK):
Total in European Economic Area:

Further details:

A60. How was the sample size decided upon? If a formal sample size calculation was used, indicate how this was done, giving sufficient information to justify and reproduce the calculation.

A61-1. Will participants be allocated to groups at random?

Yes No

A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.

6. MANAGEMENT OF THE RESEARCH

A63. Other key investigators/collaborators. Please include all grant co-applicants, protocol co-authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.

A64. Details of research sponsor(s)

A64-1. Sponsor

A64-2. Please explain how the responsibilities of sponsorship will be assigned between the co-sponsors listed in A64-1

A65. Has external funding for the research been secured?

- Funding secured from one or more funders
 External funding application to one or more funders in progress
 No application for external funding will be made

What type of research project is this?

- Standalone project
 Project that is part of a programme grant
 Project that is part of a Centre grant

- Project that is part of a fellowship/ personal award/ research training award
 Other
 Other – please state:

A66. Has responsibility for any specific research activities or procedures been delegated to a subcontractor (other than a co-sponsor listed in A64-1)? Please give details of subcontractors if applicable.

- Yes No

A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?

- Yes No

Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

A69-1. How long do you expect the study to last in the UK?

Planned start date:
 Planned end date:
 Total duration:
 Years: Months: Days:

A69-2. How long do you expect the study to last in all countries?

Planned start date:
 Planned end date:
 Planned end date
 (clinical interventions):
 Planned end date
 (all trial procedures):
 Total duration:
 Years: Months: Days:

A71-1. Is this study?

- Single centre
 Multicentre

A71-2. Where will the research take place? (Tick as appropriate)

- England
 Scotland
 Wales
 Northern Ireland
 Other countries in European Economic Area

Does this trial involve countries outside the EU?

- Yes No

A72. Which organisations in the UK will host the research? Please indicate the type of organisation by ticking the box and give approximate numbers if known:

- NHS organisations in England
 NHS organisations in Wales
 NHS organisations in Scotland
 HSC organisations in Northern Ireland
 GP practices in England
 GP practices in Wales
 GP practices in Scotland
 GP practices in Northern Ireland
 Joint health and social care agencies (eg community mental health teams)
 Local authorities
 Phase 1 trial units
 Prison establishments
 Probation areas
 Independent (private or voluntary sector) organisations
 Educational establishments
 Independent research units
 Other (give details)

Total UK sites in study:

A73-1. Will potential participants be identified through any organisations other than the research sites listed above?

- Yes No

A74. What arrangements are in place for monitoring and auditing the conduct of the research?

A76. Insurance/ indemnity to meet potential legal liabilities

Note: in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland

A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? Please tick box(es) as applicable.

Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (NHS sponsors only)
 Other insurance or indemnity arrangements will apply (give details below)

Please enclose a copy of relevant documents.

A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the

sponsor(s) or employer(s) for harm to participants arising from the design of the research? Please tick box(es) as applicable.

Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.

- NHS indemnity scheme will apply (protocol authors with NHS contracts only)
 Other insurance or indemnity arrangements will apply (give details below)

Please enclose a copy of relevant documents.

A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?

Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.

- NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)
 Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

Please enclose a copy of relevant documents.

A78. Could the research lead to the development of a new product/process or the generation of intellectual property?

- Yes No Not sure

Part B Section 10: Additional information for applications to National Offender Management Service Please consult the guidance notes before completing this section. For general advice on offender health research see the Offender Health Research Network: <http://www.ohrn.nhs.uk/>

A. Information for the National Offender Management System

1. The following categories are the NOMS Research Strategic Priorities.

Please tick the one that best applies to the research you are requesting:

- Decency
- Diversity & Equality
- Organisational Effectiveness
- Public Protection
- Offender Management and Reducing Re-offending
- Security
- Maintaining Order and Control
- Physical Health
- Mental Health

2. Broadly speaking, what type of methodology do you intend to use in order to deliver this research:

- Literature review
- Rapid evidence assessment / systematic review
- Secondary data analysis
- Primary quantitative approach
- Primary qualitative approach
- Experimental / quasi-experimental
- Economic analysis
- Other

3. Are you targeting specific groups?

- Yes No

4. Does your research cover:

- Prisons
- Probation
- Both Prisons & Probation

5. Please select each region and then select the establishments / offices within those regions where you wish to conduct the research:

- North West
- North East
- East Midlands
- East of England
- Greater London

- Kent & Sussex
- South Central
- Yorkshire & Humberside
- South West
- West Midlands
- High Security
- Wales
- Establishment(s) / Office(s) not known at this stage

6. Please advise when the outcomes are required by (and whether there are any critical deadlines when information from this research is required):

7. What are the potential benefits of the research?

To NOMS?

To academic knowledge in the field of study?

**8. Will the research include a reconviction study?
(If yes please state how this will be conducted)**

Yes No

NB. The body reviewing an application, which includes a reconviction element, should forward it to the Re-offending and Criminal Career Statistics team in OMS Analytical Services in the Ministry of Justice.

9. Please state how the results will be made available for NOMS:

10. Details of REC application

Name of REC

Address

E-mail

REC Reference number

Copy of REC opinion Enclosed
 To follow

11. Please state your reasons for choosing the selected establishment/s:

12. If you wish to conduct your research in more than four prisons, please provide further details on why this number of prisons is required:

13. Have any establishments been approached separately about this research? If so, provide details:

Yes No

14. How long will the researcher need to be inside each prison establishment?
(Please state the number of days and the number of hours per day)

15. Please list any equipment which you are intending to take into the prison establishment, if applicable
(Please note that only in exceptional circumstances would any electronic equipment be allowed into prisons due to the security risks incurred).

16. How will you identify the offenders to be involved in the research?

17. Please state how long the researcher will need to be in contact with the offender/s:

18. Please state how many staff would be involved (sampling of staff is required):

19. How long will the researcher need to be in contact with staff?

20. Are there any resource implications for NOMS?
E.g. anticipated demands on staff time, office requirements, information etc...

PART C: Overview of research sites

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. For NHS sites, the host organisation is the Trust or Health Board. Where the research site is a primary care site, e.g. GP practice, please insert the host organisation (PCT or Health Board) in the Institution row and insert the research site (e.g. GP practice) in the Department row.

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. For NHS sites, the host organisation is the Trust or Health Board. Where the research site is a primary care site, e.g. GP practice, please insert the host organisation (PCT or Health Board) in the Institution row and insert the research site (e.g. GP practice) in the Department row.

PART D: Declarations

D1. Declaration by Chief Investigator

1. The information in this form is accurate to the best of my knowledge and belief and I take full responsibility for it.
2. I undertake to abide by the ethical principles underlying the Declaration of Helsinki and good practice guidelines on the proper conduct of research.
3. If the research is approved I undertake to adhere to the study protocol, the terms of the full application as approved and any conditions set out by review bodies in giving approval.
4. I undertake to notify review bodies of substantial amendments to the protocol or the terms of the approved application, and to seek a favourable opinion from the main REC before implementing the amendment.
5. I undertake to submit annual progress reports setting out the progress of the research, as required by review bodies.
6. I am aware of my responsibility to be up to date and comply with the requirements of the law and relevant guidelines relating to security and confidentiality of patient or other personal data, including the need to register when necessary with the appropriate Data Protection Officer. I understand that I am not permitted to disclose identifiable data to third parties unless the disclosure has the consent of the data subject or, in the case of patient data in England and Wales, the disclosure is covered by the terms of an approval under Section 251 of the NHS Act 2006.
7. I understand that research records/data may be subject to inspection by review bodies for audit purposes if required.
8. I understand that any personal data in this application will be held by review bodies and their operational managers and that this will be managed according to the principles established in the Data Protection Act 1998.
9. I understand that the information contained in this application, any supporting documentation and all correspondence with review bodies or their operational managers relating to the application:
 - Will be held by the REC (where applicable) until at least 3 years after the end of the study; and by NHS R&D offices (where the research requires NHS management permission) in accordance with the NHS Code of Practice on Records Management.
 - May be disclosed to the operational managers of review bodies, or the appointing authority for the REC (where applicable), in order to check that the application has been processed correctly or to investigate any complaint.
 - May be seen by auditors appointed to undertake accreditation of RECs (where applicable).
 - Will be subject to the provisions of the Freedom of Information Acts and may be disclosed in response to requests made under the Acts except where statutory exemptions apply.
 - May be sent by email to REC members.
10. I understand that information relating to this research, including the contact details on this application, may be held on national research information systems, and that this will be managed according to the principles established in the Data Protection Act 1998.
11. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named below. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Contact point for publication(Not applicable for R&D Forms)

NRES would like to include a contact point with the published summary of the study for those wishing to seek further information. We would be grateful if you would indicate one of the contact points below.

- Chief Investigator
 Sponsor

- Study co-ordinator
- Student
- Other – please give details
- None

Access to application for training purposes (Not applicable for R&D Forms)

Optional – please tick as appropriate:

I would be content for members of other RECs to have access to the information in the application in confidence for training purposes. All personal identifiers and references to sponsors, funders and research units would be removed.

Signature:

Print Name:

Date: (dd/mm/yyyy)

D2. Declaration by the sponsor's representative

If there is more than one sponsor, this declaration should be signed on behalf of the co-sponsors by a representative of the lead sponsor named at A64-1.

I confirm that:

1. This research proposal has been discussed with the Chief Investigator and agreement in principle to sponsor the research is in place.
2. An appropriate process of scientific critique has demonstrated that this research proposal is worthwhile and of high scientific quality.
3. Any necessary indemnity or insurance arrangements, as described in question A76, will be in place before this research starts. Insurance or indemnity policies will be renewed for the duration of the study where necessary.
4. Arrangements will be in place before the study starts for the research team to access resources and support to deliver the research as proposed.
5. Arrangements to allocate responsibilities for the management, monitoring and reporting of the research will be in place before the research starts.
6. Where the research is reviewed by a REC within the UK Health Departments Research Ethics Service, I understand that the summary of this study will be published on the website of the National Research Ethics Service (NRES), together with the contact point for enquiries named in this application. Publication will take place no earlier than 3 months after issue of the ethics committee's final opinion or the withdrawal of the application.

Signature:

Print Name:

Post:

Organisation:

Date: (dd/mm/yyyy)

Declaration by applicant of the request to the Competent Authority.

D5. I hereby confirm that /confirm on behalf of the sponsor (tick which is applicable) that:

- The information provided is complete;
- The attached documents contain an accurate account of the information available;
- the clinical trial will be conducted in accordance with the protocol;
- The clinical trial will be conducted, and SUSARs and result-related information will be reported, in accordance with the applicable legislation.

Applicant of the request for the competent authority (as stated in section C.1):

Date

Signature

Print name

Appendix 5 Research protocol

Research Protocol

General Information

Protocol version and date: Version 1. 20 March 2011

Project Title: Gluten-free diet adherence in coeliac disease: Exploring multiple perspectives.

Researchers:

Helen Flaherty - Chief Investigator and PhD Student

Dr Katherine Deane - Primary Academic Supervisor

Prof Richard Gray - Secondary Academic Supervisor

Edith Cavell Building
Faculty of Health
University of East Anglia
Norwich
NR4 7TJ

Funding:

This study is a PhD research project which is funded by the University of East Anglia.

Other Organisations Involved in this Study:

Coeliac UK

3rd Floor, Apollo Centre, Desborough Road, High Wycombe, Buckinghamshire, HP11 2QW.

Norfolk and Norwich University Hospital (NNUH)

Colney Lane, Norwich, Norfolk, NR4 7UY.

Norfolk PCT

Lakeside 400, Old Chapel Way, Broadland Business Park, Thorpe St Andrew, Norwich, NR7 0WG.

Project Summary

Coeliac disease (CD) is an autoimmune disease characterised by a permanent, inappropriate immune response to ingested gluten. This condition is believed to affect around 1% of the UK population (Coeliac UK, 2010). Life-long adherence to a strict gluten-free diet (GFD) is currently the only effective treatment for CD and non-

adherence is associated with a variety of health problems and increased healthcare costs. Studies have shown that non-adherence may be as high as 58% for people with CD (Coeliac UK, 2010b; Leffler *et al.*, 2008). The aim of this study is to develop a theoretical model to understand adherence to the GFD using concept mapping.

Concept mapping is a mixed methodology designed to increase understanding of complex topics and to identify which aspects are of greater or lesser importance (Kane and Trochim, 2007). Seventy-five participants will be purposefully selected to represent the views of three stakeholder groups: 25 adults with CD; 25 adult household members; and 25 healthcare professionals. Participants will formulate statements relating to adherence to a GFD during brainstorming sessions. Next, the statements will be rated for importance and clustered into themes by the stakeholders. The results will be aggregated using 'Ariadne' concept mapping software to produce visual 'concept maps'.

The topology of thoughts and ideas generated from this study will be used to construct a model of adherence based on the common views of stakeholders. This model can be used to design an intervention, or 'adherence package' to guide healthcare professionals in providing appropriate care for people with CD. We anticipate that improvements in adherence to a GFD will reduce NHS costs as well as improve the health and quality of life of people with CD.

Abbreviations

CD	Coeliac disease
GF	Gluten-free
GFD	Gluten-free diet
GFF	Gluten-free food
IBS	Irritable bowel syndrome
NNUH	Norfolk and Norwich University Hospital
NHS	National Health Service
VA	Villous Atrophy
UK	United Kingdom

1. BACKGROUND INFORMATION AND RATIONALE

1.1 What is Coeliac Disease?

CD is an autoimmune disease found in genetically susceptible people and it is characterised by a permanent intolerance to dietary gluten. Gluten is a prolamin found in wheat, rye and barley and is present in many commonly consumed products, such as bread, breakfast cereals, pasta, beer, cakes and biscuits. The only effective

treatment for CD is the lifelong elimination of gluten from the diet (Leffler *et al.*, 2008). Poor adherence to a GFD is associated with higher rates of morbidity and mortality, often due to an increased risk of the development of enteropathy-associated T-cell lymphoma (Silano *et al.*, 2007). CD has a broad clinical spectrum ranging from asymptomatic (silent coeliac disease) to more severe symptoms including chronic diarrhoea, severe lethargy, gastric distension and pain (Dewar and Ciclitira, 2005). Inflamed and flattened small intestinal mucosa (villous atrophy (VA)) is common in CD and can result in impaired absorptive function which commonly leads to nutrient deficiency diseases, such as iron-deficiency anaemia and osteoporosis (Patient UK, 2010).

1.2 The Epidemiology of Coeliac Disease

1.2.1 Prevalence

The accuracy of estimates of the prevalence of CD has improved in recent years through the development of more reliable serological tests (Rostom *et al.*, 2005). Serological screening studies have shown that CD prevalence in the UK population is around 1%, however, as few as 1 in 10 cases are accurately diagnosed (Dewar & Ciclitira, 2005). This may be due to misdiagnosis (often CD symptoms are indistinguishable from irritable bowel syndrome (IBS) symptoms) or because the person is asymptomatic (silent CD). Improvements in the detection of CD and the possibility of mass screening may lead to a higher prevalence of CD in future years (Dewar & Ciclitira, 2005; Whittaker *et al.*, 2009). In a study by Rubio-Tapia *et al.* (2009), 9,133 frozen serum samples from healthy adults collected between the years of 1948 and 1954 were tested for CD. The results were compared with serological tests on 7,210 healthy young men and 5,558 older men. This study indicated that the prevalence of CD is increasing, regardless of whether cases are diagnosed. A Finnish study demonstrated similar results, showing that CD prevalence had doubled in the last two decades (Lohi *et al.*, 2007). Improving adherence to the GFD for an increasing number of people will present a significant challenge for healthcare professionals and a better understanding of the factors affecting adherence is required.

Genetic studies have shown that the prevalence of CD for first-degree relatives is between 10-15% (Martucci *et al.*, 2003). Ethnic-specific data for adults with CD in the UK are scarce, however, studies show that CD is common across many ethnic groups (Green, 2005). CD diagnosis tends to be more common in females than males, although the reasons for this remain unclear (Green, 2005). In the past, CD was most commonly diagnosed in childhood, however, the pattern of CD diagnosis is changing and the average age of diagnosis in the UK is now between 40-60 years (Dewar and Ciclitira, 2005; Coeliac UK, 2010d). Adherence to a GFD may be particularly

difficult for people diagnosed with CD in older age when their gluten-inclusive dietary habits have become long-established.

1.2.2 Disease Progression

CD is associated with a doubling of mortality rates compared to the normal population (Rubio-Tapia *et al.*, 2009). This condition is also linked with substantial morbidity and the clinical manifestations of CD can be devastating for patients (Rubio-Tapia *et al.*, 2009). The modes of presentation of CD vary from person to person and can be either the classic diarrhoea-predominant form, the asymptomatic 'silent' form or patients can present with atypical symptoms (Green, 2005; Griffiths, 2008). The classic clinical manifestations of CD include weight-loss, diarrhoea, steatorrhoea (fatty stools) and malabsorption of nutrients (Griffiths, 2008). The most common feature is iron-deficiency anaemia and this is found in two-thirds of adults with CD (Griffiths, 2008). Osteoporosis is also high among people with CD and this is linked with impaired calcium and vitamin D absorption. However, CD is a systemic disease, not just an ailment of the alimentary tract and a host of atypical disorders have now been attributed to the presence of CD (Griffiths, 2008; Rewers 2005). These include lethargy, infertility and neurological disorders (Griffiths, 2005). Malignancies related to CD include T-cell lymphoma, gastric, oesophageal, bladder, breast and brain cancer (Hourigan, 2006). The risk of malignancy in unmanaged CD is thought to be two-fold that of the wider population; for small intestinal lymphoma the risk may be as high as 50-fold (Leeds *et al.*, 2008). Holmes *et al.*, (1989) found that the risk of malignancy was similar to the wider population after 5 years on a GFD whereas the risk was elevated in patients who did not adhere to the GFD.

Adherence to a strict, lifelong GFD results in symptomatic, serologic and histologic remission and the normalisation of mortality rates (Pietzak, 2005; Leffler *et al.* 2008). In addition, the complete removal of gluten from the diet is associated with improved psychological wellbeing and quality of life for people with CD (Leffler *et al.*, 2008). Non-adherence to a GFD is often the reason for the persistence of symptoms after diagnosis and is associated with higher rates of morbidity and mortality (Leffler *et al.*, 2008; Hall *et al.*, 2009).

1.3 Treatment: The gluten-free Diet

The GFD was established over 50 years ago as a treatment for CD and, so far, the efficacy of this treatment has not been disproven (Ciccocioppo and Corazza, 2005). Central to this treatment is the evidence that mortality is normalised and symptoms are eliminated with the introduction of a lifelong GFD (Leffler *et al.*, 2008). The removal of gluten from the diet leads to mucosal recovery and

reduces the risks associated with gluten exposure, such as small bowel cancer (Griffiths, 2008).

Gluten is naturally present in wheat, rye and barley and some people with CD may also react to oats, although the evidence for this is inconclusive (Garsed and Scott, 2007). Gluten-containing crops are widely consumed in the UK and adopting a GFD can involve drastic changes to normal dietary habits (Zarkadas *et al.*, 2006). Some gluten-free food (GFF) is available on prescription through the NHS, although this is not exempt from prescription charges (Coeliac UK, 2010d). The provision of GFF on prescription is thought to make it easier for people with CD to follow a GFD (Coeliac UK, 2010d).

A GFD can include foods which are naturally gluten-free (GF), such as fruit and vegetables, meat and dairy products as well as gluten-free substitutes, such as GF bread, GF biscuits and GF pasta. Although the term 'gluten-free' implies that no gluten is present, this is not necessarily true (Thompson, 2001). Most people with CD can tolerate small amounts of gluten, however, the amount of gluten people with CD can consume without experiencing any deleterious effects varies between individuals (Akobeng and Thomas, 2008).

New GFF labelling legislation is currently being introduced in the UK and this will limit the permitted amount of gluten present in foods labelled as 'gluten-free' to 20 parts per million (ppm) (Food Standards Agency, 2010). Foods containing up to 100 ppm can be labelled as 'low in gluten' under this new legislation. If several of these products are eaten together, some people may exceed their tolerable threshold and experience CD symptoms (Akobeng & Thomas, 2008). Some foods that are currently labelled as GF will no longer be permitted under the new legislation (Food Standards Agency, 2010). This may reduce the number of GF products available to consumers, possibly making adherence to a GFD more difficult.

1.4 Adherence to a Gluten-free Diet

1.4.1 Definition of Adherence

The term 'adherence' is defined as the extent to which a patient's behaviour coincides with the advice of healthcare professionals (Haynes *et al.*, 2008). 'Adherence' suggests that the patient has control over their choice to follow the advice of healthcare professionals, whereas the term 'compliance' has authoritarian connotations linked with obedience (Vermeire, 2003). In the context of this study, adherence refers to a lifelong exclusion of wheat rye and barley from the diet (Leffler *et al.*, 2008). Foods labelled as 'gluten-free' or 'low in gluten' may contain small amounts of gluten, however, consumption of these products will not be regarded as non-

adherent. The reason for this is that the amount of gluten present in these products should not be sufficient to trigger a response (Food Standards Agency, 2010) and the use of these products in the CD diet is recommended by healthcare professionals.

1.4.2 Measuring Adherence to the Gluten-free Diet

A systematic review by Hall *et al.* (2009) found that adherence to a GFD in adults with CD ranged from 42% to 91%. Adherence to a GFD can be measured in several ways. In a study by Ciacci *et al.* (2002) serological test results were found to be a valid test of adherence when measured against intestinal biopsy results. Leffler *et al.* (2007) and Butterworth (2004) used self-reported measures of adherence to the GFD using Likert scales. Leffler *et al.* (2007) also applied three additional methods to confirm the validity of self-reported adherence using the same subjects. This included: analysis by a nutritionist; a 3-day food diary or 24-hour recall; and serological testing. This study found self-reported levels of adherence to be congruent with the other measures with a slight tendency for over-estimation. This inconsistency could have been due to accidental gluten contamination. To establish the level of adherence to a GFD in potential participants with coeliac disease, we will ask individuals to rate their level of adherence from the following three options:

- 1). I always stick to a strict gluten-free diet
- 2). I occasionally consume food/drinks containing gluten
- 3). I do not follow a gluten-free diet

1.4.3 Factors Affecting Adherence to a GFD in Adults with CD.

Although CD is easily and effectively treated with a GFD, the participants in a study by Whitaker *et al.* (2009) reported that following a strict GFD was a substantial burden. A systematic review investigating the factors affecting adherence to a GFD in adults with CD by Hall *et al.* (2009) found that adherence was most strongly associated with cognitive, emotional and socio-cultural factors, membership of a CD advocacy group and regular dietetic follow-up. Hall *et al.* (2009) reported that the evidence presented in the 38 studies included in this systematic review was of variable quality and further research is needed. The study by Leffler *et al.* (2008), which examined a sample of 154 adults with CD in the United States, found that adherence to a strict GFD was significantly associated with 13 factors including: membership to a CD advocacy group; understanding of the GFD; ability to follow a GFD when away from the home; and ability to follow a GFD despite changes in mood or stress levels. Low sensory acceptance and the cost of GFF have also been associated with poor adherence (Olsson *et al.*, 2008; Mendoza, 2005). Travelling and eating out can be problematic for

people with CD and a study by Karajeh *et al.* (2004) reported that less than one-fifth of UK chefs had heard of CD. Kong *et al.* (2003) found that poor availability of GFF in the workplace was a barrier to adherence.

1.5 Study Rationale

It is estimated that 1% of the UK population has CD. Following a strict GFD for life is currently the only successful treatment for this disease. The GFD is simple in theory, however, as many as 58% of people with CD do not adhere to a strict GFD (Leffler *et al.*, 2007). Poor adherence is associated with increased morbidity and mortality as well as increased healthcare costs. While the factors affecting adherence to a GFD for people with CD have been explored in previous studies, this body of research has suffered from a lack of strong theoretical and conceptual guidance. This study will develop a conceptual model of adherence to a GFD which can be used to design an adherence intervention. In a systematic review of adherence to a GFD in adult patients with CD, Hall *et al.* (2009) found little evidence of the use of interventions to improve adherence.

Concept mapping focuses on user involvement and it is a useful tool for understanding complex behaviour, such as adherence to a gluten-free diet. This methodology also allows for the generation of multiple points of view and it uncovers similarities and differences in the perspectives of different stakeholder groups.

Much of the research into adherence to a GFD for people with CD has been conducted outside the UK. The factors affecting adherence to a GFD in the UK may differ to those found in other countries for several reasons, including: differences in the cost and availability of GFF; the provision of GFF on prescription in the UK; and differences in the type and amount of help and support available for people with CD. The method of investigation used in this study (Concept mapping) will allow for the identification of factors affecting adherence to a GFD for adults living in the UK, rather than focusing on pre-determined adherence factors identified in previous studies which may not be relevant in the UK. Concept mapping is ideally suited for investigating topics which do not have an objective, absolute truth. Three stakeholder groups will be involved in this study. Differences and similarities in opinion can be identified and a 'common denominator' can be established.

2. STUDY AIMS AND OBJECTIVES

Aim:

The aim of this project is to develop a theoretical model, or conceptual framework, of adherence to a GFD using concept mapping. This model can be used to guide the development of a novel adherence intervention for people with CD. To achieve this aim, we will work with adults who have been medically diagnosed with coeliac disease, spouses or other adults who live in the same household as a person with CD (household member) and healthcare professionals who work with people with CD to gain an understanding of adherence modifying factors and their relative importance.

Objectives:

Primary objective: To use concept mapping to develop a common framework that can be used to design an adherence intervention.

Secondary objectives: To produce visual maps depicting how the adherence statements generated during the brainstorming sessions relate to one another and how important the statements are perceived to be by the three stakeholder groups.

3. STUDY DESIGN AND METHOD OF INVESTIGATION

3.1 Steering Group and Research Team

3.1.1 Steering Group

A steering group has been set up and all members will be involved in making decisions relating to each stage of the research process. The steering group includes: The Chief Investigator (Helen Flaherty); three adults with coeliac disease; a Gastroenterology Consultant from Norfolk and Norwich University Hospital (NNUH) (Dr Ian Fellows); Coeliac UK's Head of Diet and Health (Norma McGough); and the academic supervisory team (Dr Katherine Deane and Professor Richard Gray).

3.1.2 Research Team

The research team will consist of Helen Flaherty, Katherine Deane and Richard Gray (as above). It will also have Ada Mackovova who will act as a research assistant to the study. Ada Mackovova is also a PhD student at the University of East Anglia and will be trained by Helen Flaherty and Katherine Deane in appropriate consent procedures and how to assist in the concept mapping process. All of the research team have been trained in Good Clinical Practice (GCP).

3.2 Concept Mapping

3.2.1 The Concept Mapping Process

This study is designed to generate a conceptual framework, or model, of adherence to a GFD using concept mapping methods. Concept mapping is a qualitative and quantitative methodology which will be used to explore the factors affecting adherence to a GFD from the perspectives of a diverse group of stakeholders. Participants' thoughts and ideas are articulated during group brainstorming sessions and recorded as statements. Statements are clustered and rated by participants and this data will be analysed and represented in an objective form as visual maps using the Ariadne software package. These 'maps' will allow us to see which adherence factors are of greater or lesser importance according to the group of participants. A conceptual model of adherence will be generated from the results of the data analysis. This model can then be used to inform the development of a subsequent adherence intervention aimed at assisting healthcare professionals to provide effective support and resources to help improve adherence to a GFD.

Concept mapping is a phased methodology which will be conducted as follows:

Step 1: Preparation

- Development of focus for the conceptualization: The focus is the question that will be put to the participants in the brainstorming session in order to generate as many responses as possible. The focus question for the brainstorming sessions in this study will be 'what helps adults with coeliac disease stick to a gluten-free diet?'
- Selection of participants: Three groups of stakeholders will be selected for inclusion in this study: 25 adults with CD; 25 adult household members; and 25 healthcare professionals.
- Setting a schedule: The location, dates and times will be set for the group brainstorming sessions and the clustering and rating sessions. These will be held separately for each stakeholder group.

Step 2: Generation of statements

- Brainstorming - Separate brainstorming sessions will be held for each of the three stakeholder groups. Participants will be asked to use the focus question as a starting point for free association and to generate as many statements as possible in response to the focus question "what helps adults with coeliac disease stick to

a gluten-free diet?”. An example of a possible statement is ‘*If gluten-free foods were cheaper*”.

- Statement reduction – In this step, members of the steering group will eliminate duplicated statements, statements that are not understandable and statements which are regarded to be too specific. The aim of this activity is to create a rationalised set of up to approximately 80 statements.

Step 3: Structuring of statements – clustering and prioritising

Both the clustering and the prioritising tasks are performed individually by each participant. Participants will be given a full set of up to 80 cards and each card will have a single statement printed on it.

- Clustering – for this activity, participants will be asked to sort the statements into groups or piles in a way that makes sense to them. Participants will be required to give a name to each of their piles.
- Prioritisation (ranking) – participants will be asked to assign a value from 1 to 5 for each statement with 1 representing the lowest and 5 representing the highest priority. All statements will be divided into five equal piles representing the priority assigned to them.

Step 4: Concept mapping analysis

- The data will be analysed using specialised concept mapping software (Ariadne). A series of maps will be produced to represent the data generated from the structuring tasks.

Step 5: Interpreting the maps

- At this stage, the relationships between ideas and clusters of ideas can be seen.
- Different stakeholder groups’ perspectives and the thoughts and ideas of the group as a whole can be compared.

Step 6: Utilization of maps

- At this stage, decisions are made about how the results will be used in practice.

3.2.2 The Concept Mapping Sessions

Participants will be invited to attend two group sessions in a local community centre in Norwich or at the University of East Anglia. The Chief Investigator will ensure that there is comfortable seating, adequate lighting and facilities for any participants with disabilities.

Session 1: Brainstorming

The first session will be a brainstorming session, lasting approximately two hours. This will include an introduction and a short break where gluten-free refreshments will be provided. The Chief Investigator and Research Assistant will facilitate the brainstorming sessions. Separate brainstorming sessions will be held for each of the three stakeholder groups and around eight participants will be assembled for each session.

The quality of the brainstorming session is dependent on the involvement of participants. Detailed instructions will be provided in the participant information sheet and in the introduction at the start of the brainstorming session. The date and time of the session will be set well enough in advance to fit it into participants' schedules. In addition a telephone call, e-mail or postal reminder will be provided approximately one week prior to the session.

The brainstorming sessions will involve working as a group to generate statements around the focus question 'what helps adults with coeliac disease stick to a gluten-free diet?'. The focus statement will be posted and clearly visible for the duration of the session. The essence of the brainstorming session is that participants freely associate ideas and all participants have an equal influence on the outcome. The end result of the brainstorming session will be a set of statements that describe the conceptual domains of interest.

During the session, statements will be recorded on a flipchart and displayed clearly for the participants to see. An audio recording of the session will also be made using a Dictaphone. The Chief Investigator will be responsible for the quality of statements and for ensuring that they meet certain criteria in order to be useable in the following sessions.

Remote generation of ideas: If any of the participants are unable to attend the group brainstorming sessions, it will be possible for them to generate statements remotely by the use of electronic mail, by telephone or by post. This situation is not ideal as there is less interactivity and exchange of ideas among the stakeholders. The main benefit of offering the facility of remote statement generation is that it will help to keep participant drop-out rates low.

After the brainstorming sessions, the research team will refine and reduce the statements to eliminate duplicates and any statements that are too specific. The aim of this activity is to have a final set of approximately 80 statements (the software can analyse a maximum of 98 statements).

Session 2: Structuring the Statements

The second session will also last around two hours with an introduction and a short break with gluten-free refreshments. This

stage requires the participants to complete two separate tasks; these tasks are carried out individually by each participant. Participants will be given a full set of numbered statements printed on individual cards. Both tasks will be performed in relation to the focus question which will be displayed for participants to see throughout the session.

Although these are individual tasks, we feel it is appropriate to run this as a group session because some participants may require assistance from the Chief Investigator or the assistant. For healthcare professionals and participants who cannot attend a group meeting, this will be offered as a remote activity, either electronically (e-mail) or by post.

Task 1 – Prioritisation: Participants will be asked to sort their cards into five equal piles according to how important they perceive each statement to be using the following ratings:

- 1 = Relatively Unimportant
- 2 = Somewhat Important
- 3 = Moderately Important
- 4 = Very Important
- 5 = Extremely Important

Task 2 – Clustering: Participants will be asked to group the statements into piles in a way that makes sense to them according to content and they will be asked to label each cluster. Participants may not group all the statements into one single pile or have every statement as its own pile (however, some piles may contain just one statement). Each statement can only be used once and, therefore, cannot be sorted into more than one pile.

3.2.3 Data analysis

The results of the structuring stage are entered into the Ariadne software programme for analysis.

In stage 4 of the concept mapping process, the data generated from stage 2 is entered into the computer programme, Ariadne. Ariadne will combine the ideas of the three stakeholder groups and present them as a group product. Analysis begins by counting how often participants have clustered statements into a single category. The more often statements are clustered together, the closer they will appear on the 'map'. This closeness represents a strong link between the statements. Statements that appear close together on the map are combined to form clusters of related statements and a box is drawn around them. Next, Ariadne calculates the average rating for each individual statement and for each cluster from the prioritisation task. The relative importance of the different aspects is

reflected by the height of the points on the concept map. Analysis can be carried out to compare the results from different stakeholder groups or individual participants.

In stage 5, the clusters on the concept map will be given a label which best describes the content or theme of the cluster. This task will be performed as a group discussion involving the steering group.

Stage 6 involves the translation of the concept map and agreement on how the results can be used in practice as agreed by the steering group. The concept map sheds light on the views and priorities of the group as a whole and this information can be used as the basis for decision making.

3.3 Study Population

3.3.1 Recruitment of participants and selection criteria

In order to explore a diverse range of perspectives about the factors affecting adherence to a GFD, seventy-five participants will be recruited for this study from three stakeholder groups:

- 25 adults diagnosed with CD
- 25 adults living in the same household as an adult diagnosed with CD (household members)
- 25 healthcare professionals

Participants will be selected based on the inclusion criteria listed below. Only adults will be recruited for this study as we want qualified healthcare professionals and people (either with CD or those living with adults with CD) who are able to be fully in charge of their dietary choices and food buying decisions.

INCLUSION CRITERIA: ADULTS WITH COELIAC DISEASE
Adults aged 18+
Has been medically diagnosed with coeliac disease
Living within approximately a 10 mile radius of Norwich
Subjects will be a member of Coeliac UK and/or registered as a patient with coeliac disease at Norfolk and Norwich University Hospital
Must be capable of giving informed consent
Because the method involves agreeing the wording of statements, only English speaking participants will be enrolled

Adults with celiac disease will be recruited by letter of invitation which will be sent on our behalf by Coeliac UK and Norfolk and Norwich University Hospital. Coeliac UK will include a reply slip and pre-paid return envelope with the letter. Coeliac UK members who return a

reply slip will then be sent an invitation letter, a participant information sheet, a reply slip to indicate their wish to participate in the study, a pre-paid return envelope and a household member's pack. Patients with CD attending NNUH will be sent the invitation letter along with a participant information sheet, a reply slip, a pre-paid return envelope and a household member's pack in one single mailing. Although all potential participants will receive the same documents, Coeliac UK requested that their members are sent an invitation letter and reply slip separate to the other documents, whereas NNUH would like all the documents to be sent out together.

As we are interested in hearing about factors that both help and prevent people with CD sticking to a gluten-free diet, we will endeavour to recruit participants with a range of GFD adherence behaviours. In order to do this, we will ask the person with CD to indicate their level of adherence to a GFD, along with their contact details on the reply slip. We will select a reasonably equal proportion of the three levels of adherence:

1. I always stick to a gluten-free diet
2. I partially stick to a gluten-free diet
3. I do not follow a gluten-free diet

Return of this information to the research team indicates consent for us to have information on their level of adherence to a GFD.

INCLUSION CRITERIA: SPOUSES OR OTHER ADULTS LIVING WITH AN ADULT WHO HAS COELIAC DISEASE (HOUSEHOLD MEMBERS)
Adults aged 18+ years
Has been living in the same household as a person who has been medically diagnosed with coeliac disease for at least one year
Living within approximately a 10 mile radius of Norwich
Must be capable of giving informed consent
Because the method involves agreeing the wording of statements, only English speaking participants will be enrolled

Adults with CD will also be sent a pack marked 'Adult household member's pack' enclosed with their invitation documents. We will ask adults with CD to identify their spouse or other adult who lives with them (household member) and to pass on the adult household member's pack to that person should they wish to do so. This pack will contain an invitation letter, a participant information sheet, a reply slip and a pre-paid return envelope. We intend to recruit household members for this study even if the person with CD who they live with does not also volunteer. A sample of 25 participants will be selected from the responses.

The Chief Investigator will visit adults with CD and household members at their home or other appropriate venue to take consent. During this meeting, the Chief Investigator will collect baseline characteristics through a short questionnaire.

INCLUSION CRITERIA: HEALTHCARE PROFESSIONALS
Currently working with at least one adult patient with coeliac disease
Has at least one year's experience of working with adult patients with coeliac disease
Working or living within approximately a 10 mile radius of Norwich
Must be capable of giving informed consent
Because the method involves agreeing the wording of statements, only English speaking participants will be enrolled

Healthcare professionals will be recruited on the basis that they have treated people with CD for at least one year and are currently treating at least one adult patient with CD. An invitation letter, participant information sheet, a short questionnaire, a consent form and pre-paid return envelope will be sent to all healthcare professionals who work with adults with CD at NNUH. We will also recruit by sending the invitation documents to GP practices in Norwich. Efforts will be made to recruit a diverse range of healthcare professionals including General Practitioners, Nurses, Gastroenterologists and Dietitians. We do not intend to meet healthcare professionals face-to-face to gain informed consent, they will complete the consent form and return it to us by post.

All individuals who return a reply slip indicating that they wish to take part in the study will be contacted and informed whether or not they have been selected for the study. We will notify participants of when and where the group meetings will be held and we will send reminders by post, e-mail or telephone approximately seven days prior to the meetings.

If we do not receive sufficient responses through this process, we will recruit additional participants by sending reminder letters to individuals we have previously contacted, invitation letters to individuals not previously contacted, by delivering presentations and/or publications via Coeliac UK or NNUH.

Should any suitable participants arrive at either of the two meetings having not been consented, then consent will be taken face-to-face before the session commences.

Collection of baseline characteristics

All participants will be asked to complete a short questionnaire which will collect baseline characteristics. For adults with CD and household members, questionnaires will be completed during face-

to-face meetings with the Chief Investigator at the time when informed consent is obtained. For healthcare professionals, the questionnaire will be sent out with the invitation letter and this should be completed and returned with the consent form. The following data will be collected in the questionnaires:

For people with CD:

- Name
- Address
- Telephone number/s
- E-mail address
- Gender
- Length of time since CD diagnosis
- Any other special diets followed
- Severity of symptoms when gluten is consumed
- Ethnicity
- Age
- Level of education

For adult household members:

- Name
- Address
- Telephone number/s
- E-mail address
- Relationship to the person with CD
- Gender
- Gender of the person with CD
- Ethnicity
- Age
- Level of Education

For healthcare professionals working with patients with CD:

- Name
- Address
- Telephone number/s
- E-mail address
- Job title
- Professional qualifications
- Length of time working with coeliac patients
- Number of people with CD on current patient list

We will ask all interested participants to indicate the best time of the week for them to attend the group sessions by putting ticks in the appropriate boxes on a table of possible days/times (see below). This information will be used when we plan the group sessions:

	MON	TUES	WED	THURS	FRI	SAT	SUN
Morning (9am-12pm)							
Afternoon (12pm-4pm)							
Evening (6pm-9pm)							

3.4 Duration of the Project

This study is being carried out as a three year PhD research project, ending in April 2013. It is anticipated that the group brainstorming sessions and the statement sorting sessions will be conducted within approximately an eight month period.

4. DISSEMINATION AND OUTCOME

The research findings will be disseminated through academic journals and the results will be presented at conferences. We will work closely with Coeliac UK and relevant NHS and health organisations in disseminating the research findings. A report of the results will be sent to participants at the end of this study.

The findings of this study will be used to guide the development and subsequent testing of a novel adherence intervention to enable healthcare professionals to assist patients to be better at sticking to a GFD.

5. PROBLEMS ANTICIPATED

Hall *et al.* (2009) highlight the fact that people who adhere to the GFD may be more likely to respond to research documentation, such as questionnaires, compared to people who do not adhere to the GFD. It would be unethical for us to force anybody to participate in this study, so this problem is unavoidable. However, we will seek to specifically recruit participants with a variety of levels of adherence to a GFD. We will also recruit healthcare professionals who are likely to work with patients with varying degrees of adherence and adults who live in the same household as someone with CD who may have varying levels of adherence to a GFD.

Some potential participants will spend time reading the participant information sheet and completing and returning the reply slip/questionnaire, yet they may not be selected to participate in the study. We will send out letters to those people who are not selected to thank them for their interest in the study and to explain why they

have not been selected. The findings of our research will be published in a lay report for Coeliac UK which should allow most of these potential participants to read the findings of the study even though they were unable to participate.

As this study is based in Norwich, the results may not reflect the wider population of adults with CD. In particular, we do not expect to be able to recruit an ethnically diverse population of people with CD from Norwich, as Norwich does not have substantial numbers of people from ethnic minorities in its population. In addition, the study will not include people who are not members of Coeliac UK or do not receive NHS healthcare for CD.

There is the potential for researcher bias in this study. In order to limit researcher bias, the Chief Investigator and Research Assistant will not provide prompts to generate responses during the brainstorming sessions. The focus question will be stated at the start of the session and participants will be allowed to express their ideas freely without discussion or criticism from other participants or members of the research team.

For health professionals participating, there is a risk that the Chief Investigator could be made aware through discussion of unprofessional practice. Any practice that is potentially detrimental to a vulnerable person may have to be reported to an appropriate authority. Health professionals will be advised of this risk in the information sheet provided to them.

6. STUDY ADMINISTRATION AND ETHICAL ISSUES

6.1 Informed Consent

Invitation letters and participant information sheets will be sent to potential participants belonging to the three stakeholder groups. These documents will explain the purpose and nature of the study along with details of what will be required of individuals if they decide to take part.

One of the inclusion criteria is that participants are capable of giving informed consent. To ensure this, visits will be made to the selected participants who have CD and household members prior to the study to gain informed consent face-to-face. This will allow the research team to identify and eliminate participants who are not capable of giving informed consent. It is expected that this meeting will take place in the participant's home. An alternative meeting place will be arranged if the participant requests this (e.g. a room at the university). For health professionals, professional competency is assumed and, therefore, the Chief Investigator will not request a

meeting to obtain informed consent. For this group of participants, the signed consent form can be returned by post. Should any suitable participants arrive at either of the two meetings having not consented, then consent will be taken face-to-face before the session commences.

Participation in this research will involve attending two group sessions. At the beginning of each session, the Chief Investigator will remind all group members that participation in the research is optional and individuals are free to withdraw from the study at any time without giving a reason. Participants who complete the concept mapping tasks remotely will also be reminded that participation is optional and that they are free to withdraw at any time.

We will conduct process consent throughout the research study. Should anyone indicate that they are unwilling to continue they will be able to withdraw without being required to provide a reason. This will have no influence on the clinical care provided to them.

6.2 Safety considerations

Possible risks:

- Visiting participants in their own home in order to gain informed consent carries an element of risk for members of the research team. The researchers will follow the University's Lone Worker Policy. In practice this will mean that prior to every meeting, the visiting researcher will e-mail the address and time of the meeting to the academic supervisor. In addition, the visiting researcher will carry a fully charged mobile phone in case of an emergency and will telephone the academic supervisor immediately before and after every meeting. Should the researcher feel unsafe during a meeting with a participant, she will leave the property immediately and telephone the academic supervisor (and the appropriate emergency services if necessary).
- Health and Safety precautions and insurance cover for the meeting room will be covered by the venue owners.
- Although the risk of harm is considered to be low, a potential issue that may arise is that some participants may become upset when discussing issues related to CD and the GFD. The Chief Investigator and Research Assistant will monitor for signs of distress and will withdraw a participant from the group if they show signs of becoming distressed. Participants will be reminded at the start of the group sessions that they can withdraw from the study at any time without giving a reason.
- Although it seems unlikely, some of the issues raised during group meetings may cause anger in some participants. The Chief Investigator will be accompanied by a Research Assistant for all group meetings. The risk of harm to the Chief

Investigator, Research Assistant and participants is believed to be low.

- The two group sessions will include a break with refreshments. To avoid causing harm through gluten-consumption, all the food and drinks provided will be gluten-free. The food packaging will be made available so that people with any other food intolerance or allergy can check whether the items are suitable for them. This action will be taken with all three groups.

6.3 Confidentiality

Care will be taken to avoid disclosing participants' identities. Anonymity in all publications relating to this research will be guaranteed. Invitation letters will be distributed to adults with CD on behalf of the research team by NNUH and Coeliac UK. This eliminates the need for us to gain access to personal information when recruiting subjects for this study. NNUH will distribute invitation letters and participant information sheets to healthcare professionals who work with adult patients with coeliac disease. We will recruit healthcare professionals from General Practices by sending invitation letters and participant information sheets directly to them.

Completed questionnaires and consent forms will be locked in a secure filing cabinet within a locked room at the University of East Anglia. In order to protect participants' identities, each participant will be allocated a reference number which will be printed on all their related documentation. Documents received from individuals who are not selected for participation in this study will be shredded within six months. Data will also be stored as a password protected document on a university computer with password security. Paper documents relating to participants will be shredded six months after the study ends. Electronic files will be deleted two years after the study ends.

The data collection method employed in concept mapping is a collective activity and this makes it difficult to identify which participant provided which data. In addition, the research team will screen all statements generated for personally identifying statements (e.g. I trust the advice I receive from Dr Smith) and anonymise them (e.g. I trust the advice I receive from my GP). At the start of the two group sessions, individuals will be advised that they are free to withdraw from the study at any time, however, the data gathered up until that point will still be used in the study. All information gathered during this study will be anonymised and cannot be traced back to the dissenting individual. An audio recording device will be used during the two group meetings. All audio recordings of the brainstorming sessions will be deleted after transcription.

6.4 Resource Requirements

- A Research Assistant to join the Chief Investigator for the group meetings
- Concept mapping software (Ariadne) for analysing the data
- A suitable meeting room for brainstorming sessions
- Pens, paper, flip chart, statements printed on cards and a Dictaphone
- Gluten-free (GF) Refreshments for all group meetings
- Transport costs for the participants, the Chief Investigator and Research Assistant

6.5 Study Plan

6.5.1 Flow chart

Approximately May – June 2011 - Recruitment of subjects

Distribute invitation letters and participant information sheets
Notify those who have and have not been selected for the study
Gain informed consent
Collect baseline characteristics using questionnaires
Send confirmation of the dates for the group meetings



Approximately June – July 2011

Send a reminder one week prior to the group session
Group session 1: Brainstorming



Approximately July – September 2011

Statement reduction task



Approximately October - December 2011

Send a reminder one week prior to the group session
Group session 2: Clustering and prioritising the statements



Approximately January – March 2012

Input the data using Ariadne software



Approximately April 2012 – June 2012

Develop a Model of Adherence using the concept maps



Approximately July –September 2012

Make recommendations for how the concept map could be used in
planning and assessing health care for people with CD

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Appendix 6 - Invitation letter to adults with coeliac disease from Coeliac UK

Coeliac UK logo to be added by Coeliac UK

Dear

Re: Invitation to participate in a research study: What helps adults with coeliac disease stick to a gluten-free diet?

Coeliac UK is working with the University of East Anglia (UEA) on a study to explore what helps adults with coeliac disease stick to a gluten-free diet. This study has gained ethical approval and it meets all of the necessary research standards.

As you have already expressed an interest in taking part in research, we would like to know if you would be willing to take part in this study. If you are interested and you would like to receive more information, please complete the enclosed reply slip and return it in the envelope provided by **(insert date (allow approx 2 weeks))**.

If you choose not to take part, then you do not need to do anything further. Please be assured that we have not passed any of your details to the UEA. They will only have your information if you return the consent form to them. All information collected about you during the study will be strictly confidential.

The study will involve attending two group meetings in Norwich which will last approximately two hours each. During these meetings you will be asked to share your ideas about what helps adults with coeliac disease stick to a gluten-free diet.

You can participate in this study regardless of whether or not you stick to a gluten-free diet. You will need to have been medically diagnosed with coeliac disease and aged eighteen years or over.

The UEA are recruiting for this study via a number of routes, such as GP surgeries or dietetics clinics. It is possible that you may receive more than one invitation to this study but you only need to respond to one.

Coeliac UK is dedicated to supporting research to help improve the lives of people with coeliac disease and dermatitis herpetiformis. As research also remains a high priority for our Members we do hope that you will be able to take part.

Yours sincerely

Coeliac UK

Appendix 7 – Letter sent to people who responded to Coeliac UK’s invitation letter



University of East Anglia
1.33 Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ

Phone: 01603 593665
E-mail: h.flaherty@uea.ac.uk

Dear

Re: Invitation to participate in a research study: What helps adults with coeliac disease stick to a gluten-free diet?

Coeliac UK recently wrote and invited you to participate in the above study. Thank you for contacting us to request more information about this study.

It is up to you to decide whether or not to take part. Before you decide, please read the attached information sheet and discuss it with others if you wish. If you would like to take part, please complete the reply slip and return it to us in the envelope provided as soon as possible.

We are also looking to recruit spouses or other adults who live with a person who has coeliac disease for this study. If there is someone living with you who you think might be interested in joining this study, please give them the enclosed envelope addressed to 'Adult household member'.

Participation in this study is optional for both you and your spouse or other adult household member.

Because recruitment for this study is via a number of routes it is possible that you will receive more than one invite to this study. Please ignore the second invitation.

Yours Sincerely

Helen Flaherty
Chief Investigator

Appendix 8 - Letter sent to patients with coeliac disease by Norfolk and Norwich University Hospital



Norfolk and Norwich University Hospital
Colney Lane
Norwich
NR4 7UY

Phone: 01603 288356
E-mail: ian.fellows@nnuh.nhs.uk

Dear Sir/Madam,

Re: Invitation to participate in a research study: What helps adults with coeliac disease stick to a gluten-free diet?

You are being invited to take part in a research study as a person with coeliac disease who is attending, or has attended, a clinic at Norfolk and Norwich University Hospital. It is up to you to decide whether or not to take part. Before you decide, please read the attached information sheet and discuss it with others if you wish.

If you decide that you would like to take part, please complete the enclosed reply slip and return it in the envelope provided as soon as possible.

We are also looking to recruit spouses or other adults who live with a person who has coeliac disease for this study. If there is someone living with you who you think might be interested in joining this study, please give them the enclosed envelope addressed to 'Adult household member'.

Participation in this study is optional for both you and your spouse or other adult household member.

Because recruitment for this study is via a number of routes it is possible that you will receive more than one invitation to this study. Please ignore the second invitation.

Yours faithfully

Dr Ian Fellows
Consultant, Gastroenterology and General Medicine

Appendix 9 Invitation letter sent to household members



1.33 Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 593665
E-mail: h.flaherty@uea.ac.uk

Dear Sir/Madam,

Re: Invitation to participate in a research study: What helps adults with coeliac disease stick to a gluten-free diet?

You are being invited to take part in a research study because a person who you live with has coeliac disease.

It is up to you to decide whether or not to take part. Before you decide, please read the attached information sheet and discuss it with others if you wish. Participation in this study is optional for both you and the person with coeliac disease.

If you decide that you would like to take part, please complete the enclosed reply slip and return it in the envelope provided as soon as possible.

Yours faithfully

Helen Flaherty
Chief Investigator

Appendix 10 Invitation letter sent to healthcare professionals by Norfolk and Norwich University Hospital



Norfolk and Norwich University Hospital
Colney Lane
Norwich
NR4 7UY

Phone: 01603 288356
E-mail: ian.fellows@nnuh.nhs.uk

Dear Sir/Madam,

Re: Invitation to participate in a research study: What helps adults with coeliac disease stick to a gluten-free diet?

You are being invited to take part in a research study because you are a healthcare professional who works at Norfolk and Norwich University Hospital. We are looking to recruit healthcare professionals who currently work with at least one adult patient who has coeliac disease.

It is up to you to decide whether or not to take part. Before you decide, please read the attached information sheet and discuss it with others if you wish.

If you decide that you would like to take part, please complete the enclosed consent form and short questionnaire and return them in the envelope provided as soon as possible.

Yours faithfully

Dr Ian Fellows
Consultant, Gastroenterology and General Medicine

Appendix 11 Invitation letter sent to General Practices



University of East Anglia
1.33 Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ
Phone: 01603 593665
E-mail: h.flaherty@uea.ac.uk

Dear Sir or Madam,

Re: Invitation to participate in a research study: What helps adults with coeliac disease stick to a gluten-free diet?

You are being invited to take part in a research study because you are a healthcare professional working at a General Practice in Norwich. We are looking to recruit healthcare professionals who currently work with at least one adult patient who has coeliac disease.

It is up to you to decide whether or not to take part. Before you decide, please read the attached information sheet and discuss it with others if you wish.

If you decide that you would like to take part, please complete the enclosed consent form and short questionnaire and return them in the envelope provided as soon as possible.

Yours faithfully

Helen Flaherty
Chief Investigator

Appendix 12 Participant information sheet for adults with coeliac disease



1.33 Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 593665
E-mail: h.flaherty@uea.ac.uk

Information sheet for people with coeliac disease

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

Dear Sir/Madam

We would like to invite you, as a person with coeliac disease, to take part in a research study which aims to explore how people with coeliac disease manage sticking with a gluten-free diet.

The study is being conducted by PhD student, Helen Flaherty, who is the Chief Investigator in this study. Helen is working under the supervision of two academic supervisors, Dr Katherine Deane and Prof Richard Gray at the University of East Anglia. Before you give your full consent, please take time to read the following information carefully and discuss it with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Participation in this study is optional for both you and your spouse or adult household member and you are free to withdraw at any time without giving a reason.

Part 1 tells you the purpose of this study and what will happen if you participate. Part 2 gives you more detailed information about the conduct of the study.

If you would like to take part in this study, please complete the enclosed reply slip and return it to us in the envelope provided as soon as possible. Helen Flaherty will contact you within three weeks of receiving your reply slip to arrange to visit you in your home (or elsewhere if you prefer). During this meeting Helen will discuss the study with you, answer your questions and ask you to sign a consent form. You will also be asked to fill in a short questionnaire which should take no longer than five minutes to complete.

Thank you for taking your time to read this.

Yours Sincerely

Miss Helen Flaherty
Chief Investigator

Part 1

What is the purpose of the study?

The only effective treatment for coeliac disease is a diet that is free from gluten. Gluten is found in wheat, rye and barley. Some people may also have an adverse reaction to oats. Removing food and drinks from the diet which contain gluten, such as bread, breakfast cereals, pasta, beer and cakes, can be difficult. It is known that many people with coeliac disease do not stick to a strict gluten-free diet.

In this study we will explore the factors influencing whether or not adults with coeliac disease stick to a gluten-free diet. To do this, we will collect information from 25 adults with coeliac disease, 25 spouses or other adults who live with a person with coeliac disease and 25 healthcare professionals. The study will determine whether these three groups of people have the same or different ideas about why people with coeliac disease do or do not stick to a gluten-free diet. The results of this study can be used to design an intervention aimed at improving healthcare for people with coeliac disease.

This study is being undertaken for educational purposes, as part of my PhD.

Why have I been invited?

You have been invited to participate in this study because you have coeliac disease and you are attending, or have attended, a clinic at Norfolk and Norwich University Hospital.

Are there any inclusion or exclusion criteria?

We are seeking to recruit 25 adults who have been medically diagnosed with coeliac disease to take part in this study. These people should be aged 18 years or over and live within approximately a 10 mile radius of Norwich. Participants will be registered with Coeliac UK and/or Norfolk and Norwich University Hospital. As we are interested to hear about the things that both increase and decrease a person's ability to stick to a strict gluten-free diet, we are looking to recruit people who fit in to any of the following categories:

- I follow a strict gluten-free diet at all times
- I partially follow a gluten-free diet
- I do not follow a gluten-free diet at all

Do I have to take part?

Participation in this study is voluntary and it is up to you to decide. If you agree to take part, we will ask you to sign a consent form. You are free to withdraw at any time without giving a reason. This would not affect the standard of care you receive.

What will happen to me if I take part?

Before we can include you in this research study, the Chief Investigator, Helen Flaherty, will meet with you to discuss the study and to ask you to sign a consent form. This meeting should last no longer than 30 minutes. Helen can meet with you in your own home or, if you prefer, the meeting can take place elsewhere. You will also be required to complete a short questionnaire, which should take you no longer than five minutes.

You will then be invited to participate in two group meetings with other people who have coeliac disease. These meetings will take place between November 2011

and March 2012 at the University of East Anglia. The meetings are really informal discussions and each is expected to last approximately two hours. There will be a break during both meetings and refreshments will be provided (gluten-free of course!). The group meetings will be run by the Chief Investigator, Helen Flaherty, and there will be at least one research assistant present at both meetings.

In the first meeting, you and the other members of your group will be asked to produce statements that answer the question “What helps a person with coeliac disease stick to a gluten-free diet?”. The discussion will be tape-recorded and statements will be written on flip charts. You will also have a note pad on which you can individually write your statements if you wish. There is no pressure on any individual to make statements and any information you provide during the meeting will be kept anonymous.

The second meeting involves looking through the statements that were generated by all three participant groups during the first meetings. There will be around 80 statements. You will be asked to sort the statements into five equal piles indicating their relative importance and then you will be asked to group the statements into themes.

Expenses and payments:

Although we do not have funding to compensate you for your time, we will refund your travel expenses.

What are the possible disadvantages and risks of taking part?

The risk of you being harmed as a result of this study is very low. If you feel distressed at any time during this study, you are free to withdraw without having to provide a reason. If the Chief Investigator and/or research assistant become concerned about your health at any stage, we will suggest that you withdraw from the study and we may advise you to speak to a healthcare professional.

A possible disadvantage for participants is that you will be asked to give up four hours of your time to attend two two-hour group meetings. Although you will not be compensated for your time, we will reimburse your travel expenses and we will provide gluten-free refreshments during both meetings.

What are the possible benefits of taking part?

We cannot promise that the study will help you but it is our intention to use the information we get from this study to inform a lay report of this research for Coeliac UK and a professional report for a peer reviewed journal which will, hopefully, help improve the treatment of people with coeliac disease.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my taking part in this study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

This completes part 1. If the information in part 1 interests you and you would like to participate in this study, please read the additional information in part 2 before making any decision.

Part 2

Once I take part, can I change my mind?

Yes! After you have read this information and asked us any questions, we will ask you to complete an Informed Consent Form. However, if at any time, before, during or after the study you wish to withdraw, please just contact the Chief Investigator, Helen Flaherty. You can withdraw at any time and you do not have to explain the reason why you have decided to withdraw.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. The Chief Investigator, Helen Flaherty, can be contacted by telephone: 01603593665 (office) / 07920406094 (mobile) or email: h.flaherty@uea.ac.uk. If you remain unhappy and wish to complain formally, you can do this by contacting Helen's academic supervisor, Dr Katherine Deane, by telephone: 01603 597047 or e-mail: K.Deane@uea.ac.uk. Alternatively, you can write to us at:

Dr Katherine Deane / Helen Flaherty
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against the University of East Anglia but you may have to pay your legal costs.

Will my taking part in this study be kept confidential?

Yes. All information collected about you during the course of this study will be kept strictly confidential. Your personal details, such as your name and address, will be recorded and stored anonymously according to the Data Protection Act (1998). Your name will be substituted by a coded reference number so that your information cannot be traced back to you. Paper documents which contain your data will be stored in a locked cabinet in a locked room at the University of East Anglia. Paper documents will be shredded six months after the end of this study. Electronic data will be stored securely on a password protected computer at the University of East Anglia for two years. After this, all data will be deleted. Data will only be viewed by the Chief Investigator and members of the research team.

If the data collected in this study is used in a written report, such as a thesis or published paper, we will not use your name or personal details. Any information you provide will be kept anonymous, however, you are not obliged to disclose any information that you would prefer to keep private.

What will happen to the results of the study?

The results of this study can be used to develop an intervention to help improve adherence to a gluten-free diet for people with coeliac disease. This may improve the quality of life of many people with coeliac disease as well as reducing the cost of healthcare.

We aim to publish the results of this study in a lay report for Coeliac UK, in scientific journals and the study will be used as part of a written PhD thesis. You will not be personally identifiable in any report or publication relating to this study.

The results of this study will be made available to you upon your request. Please use the contact details shown at the top of this information sheet when requesting the results.

Who is organising and funding the research?

This study is part of a three year student PhD research project funded by the University of East Anglia. The Chief Investigator is PhD student, Helen Flaherty. Helen will work under the supervision of Dr Katherine Deane and Professor Richard Gray.

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by the Newcastle North Tyneside 2 Research Ethics Proportionate Review Sub-Committee. In addition, the research study has been reviewed by staff at Coeliac UK and Norfolk and Norwich University Hospital.

Further information and contact details:

Please do not hesitate to contact us if you require further information about any of the following issues:

Specific information about this research project.

Helen Flaherty
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 597130 / 07920406094
E-mail: h.flaherty@uea.ac.uk

Advice as to whether you should participate.

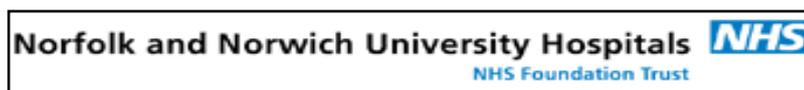
Please do not hesitate to contact your General Practitioner or other healthcare professional if you are unsure of whether to take part. You can also discuss your participation in this study with your friends and family.

Who you should approach if you are unhappy with the study.

Dr Katherine Deane
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 597047
E-mail: K.Deane@uea.ac.uk

Thank you for taking your time to read this information sheet. Please complete and return the enclosed reply slip in the envelope provided if you would like to participate in this study.

Appendix 13 Participant information sheet for household members



Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 593665
E-mail: h.flaherty@uea.ac.uk

Information sheet for spouses (or other adults) who live with a person with coeliac disease

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

Dear Sir/Madam,

We would like to invite you to take part in a research study which aims to explore how people with coeliac disease manage sticking with a gluten-free diet.

The study is being conducted by PhD student, Helen Flaherty, who is the Chief Investigator in this study. Helen is working under the supervision of two academic supervisors, Dr Katherine Deane and Prof Richard Gray at the University of East Anglia. Before you give your full consent, please take time to read the following information carefully and discuss it with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Participation in this study is optional and you are free to withdraw at any time without giving a reason.

Part 1 tells you the purpose of this study and what will happen if you participate. Part 2 gives you more detailed information about the conduct of the study.

If you would like to take part in this study, please complete the enclosed reply slip and return it to us in the envelope provided as soon as possible. Helen Flaherty will contact you within three weeks of receiving your reply slip to arrange to visit you in your home (or elsewhere if you prefer). During this meeting Helen will discuss the study with you, answer your questions and ask you to sign a consent form. You will also be asked to fill in a short questionnaire which should take no longer than five minutes to complete.

Thank you for taking your time to read this.

Yours faithfully

Miss Helen Flaherty
Chief Investigator

Part 1

What is the purpose of the study?

The only effective treatment for coeliac disease is a diet that is free from gluten. Gluten is found in wheat, rye and barley. Some people may also have an adverse reaction to oats. Removing food and drinks from the diet which contain gluten, such as bread, breakfast cereals, pasta, beer and cakes, can be difficult. It is known that many people with coeliac disease do not stick to a strict gluten-free diet.

In this study we will explore the factors influencing whether or not adults with coeliac disease stick to a gluten-free diet. To do this, we will collect information from 25 adults with coeliac disease, 25 spouses or other adults who live with someone with coeliac disease and 25 healthcare professionals. The study will determine whether these three groups of people have the same or different ideas about why people with coeliac disease do or do not stick to a gluten-free diet. The results of this study can be used to design an intervention aimed at improving healthcare for people with coeliac disease.

This study is being undertaken for educational purposes, as part of my PhD.

Why have I been Invited?

You have been invited to participate in this study because you are a spouse or other adult who lives with a person who has coeliac disease.

Are there any inclusion or exclusion criteria?

We are seeking to recruit 25 participants who are either a spouse or other adult who lives with an adult who has been diagnosed with coeliac disease. These people should be aged 18 years or over and live within approximately a 10 mile radius of Norwich.

Do I have to take part?

Participation in this study is voluntary and it is up to you to decide. If you agree to take part, we will ask you to sign a consent form. You are free to withdraw at any time without giving a reason. This would not affect the standard of care you receive.

What will happen to me if I take part?

Before we can include you in this research study, the Chief Investigator, Helen Flaherty, will meet with you to discuss the study and to ask you to sign a consent form. This meeting should last no longer than 30 minutes. Helen can meet with you in your own home or, if you prefer, the meeting can take place elsewhere. You will also be required to complete a short questionnaire, which should take you no longer than five minutes.

You will then be invited to participate in two group meetings with other spouses or adults who live with a person with coeliac disease. These meetings will take place between November 2011 and March 2012 at the University of East Anglia. The meetings are really informal discussions and each is expected to last approximately two hours. There will be a break during both meetings and refreshments will be provided. The group meetings will be run by the Chief Investigator, Helen Flaherty and there will be at least one research assistant present at both meetings.

In the first meeting, you and the other members of your group will be asked to produce statements that answer the question "What helps a person with coeliac

disease stick to a gluten-free diet?”. The discussion will be tape-recorded and statements will be written on flip charts. You will also have a note pad on which you can individually write your statements if you wish. There is no pressure on any individual to make statements and any information you provide during the meeting will be kept anonymous.

The second meeting involves looking through the statements that were generated by all three participant groups during the first meetings. There will be around 80 statements. You will be asked to sort the statements into five equal piles indicating their relative importance and then you will be asked to group the statements into themes.

Expenses and payments

Although we do not have funding to compensate you for your time, we will refund your travel expenses.

What are the possible disadvantages and risks of taking part?

The risk of you being harmed as a result of this study is very low. If you feel distressed at any time during this study, you are free to withdraw without having to provide a reason. If the Chief Investigator and/or research assistant become concerned about your health at any stage, we will suggest that you withdraw from the study and we may advise you to speak to a healthcare professional.

A possible disadvantage for participants is that you will be asked to give up four hours of your time to attend two two-hour group meetings. Although you will not be compensated for your time, we will reimburse your travel expenses and we will provide refreshments during both meetings.

What are the possible benefits of taking part?

We cannot promise that the study will help your spouse or the person you live with but it is our intention to use the information we get from this study to inform a lay report of this research for Coeliac UK and a professional report for a peer reviewed journal which will, hopefully, help improve the treatment of people with coeliac disease.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my taking part in this study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

This completes part 1. If the information in part 1 interests you and you would like to participate in this study, please read the additional information in part 2 before making any decision.

Part 2

Once I take part, can I change my mind?

Yes! After you have read this information and asked us any questions, we will ask you to complete an Informed Consent Form. However, if at any time, before, during or after the study you wish to withdraw, please just contact the Chief Investigator, Helen Flaherty. You can withdraw at any time and you do not have to explain the reason why you have decided to withdraw.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. The Chief Investigator, Helen Flaherty, can be contacted by telephone: 01603593665 (office) / 07920406094 (mobile) or email: h.flaherty@uea.ac.uk. If you remain unhappy and wish to complain formally, you can do this by contacting Helen's academic supervisor, Dr Katherine Deane, by telephone: 01603 597047 or e-mail: K.Deane@uea.ac.uk. Alternatively, you can write to us at:

Dr Katherine Deane / Helen Flaherty
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against the University of East Anglia but you may have to pay your legal costs.

Will my taking part in this study be kept confidential?

Yes. All information collected about you during the course of this study will be kept strictly confidential. Your personal details, such as your name and address, will be recorded and stored anonymously according to the Data Protection Act (1998). Your name will be substituted by a coded reference number so that your information cannot be traced back to you. Paper documents which contain your data will be stored in a locked cabinet in a locked room at the University of East Anglia. Paper documents will be shredded six months after the end of this study. Electronic data will be stored securely on a password protected computer at the University of East Anglia for two years. After this, all data will be deleted. Data will only be viewed by the Chief Investigator and members of the research team.

If the data collected in this study is used in a written report, such as a thesis or published paper, we will not use your name or personal details. Any information you provide will be kept anonymous, however, you are not obliged to disclose any information that you would prefer to keep private.

What will happen to the results of the study?

The results of this study can be used to develop an intervention to help improve adherence to a gluten-free diet for people with coeliac disease. This may improve the quality of life of many people with coeliac disease as well as reducing the cost of healthcare.

We aim to publish the results of this study in a lay report for Coeliac UK, in scientific journals and the study will be used as part of a written PhD thesis. You will not be personally identifiable in any report or publication relating to this study.

The results of this study will be made available to you upon your request. Please use the contact details shown at the top of this information sheet when requesting the results.

Who is organising and funding the research?

This study is part of a three year student PhD research project funded by the University of East Anglia. The Chief Investigator is PhD student, Helen Flaherty. Helen will work under the supervision of Dr Katherine Deane and Professor Richard Gray.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by the Newcastle North Tyneside 2 Research Ethics Proportionate Review Sub-Committee. In addition, the research study has been reviewed by staff at Coeliac UK and Norfolk and Norwich University Hospital.

Further information and contact details:

Please do not hesitate to contact us if you require further information about any of the following issues:

Specific information about this research project.

Helen Flaherty
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 597130 / 07920406094
E-mail: h.flaherty@uea.ac.uk

Advice as to whether you should participate.

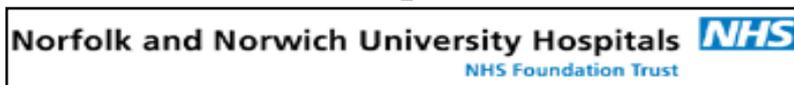
Please do not hesitate to contact your General Practitioner or other healthcare professional if you are unsure of whether to take part. You can also discuss your participation in this study with your friends and family.

Who you should approach if you are unhappy with the study.

Dr Katherine Deane
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 597047
E-mail: K.Deane@uea.ac.uk

Thank you for taking your time to read this information sheet. Please complete and return the enclosed reply slip in the envelope provided if you would like to participate in this study.

Appendix 14 Participant information sheet for healthcare professionals



1.33 Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ

Tel: 01603 593665
E-mail: h.flaherty@uea.ac.uk

Information sheet for healthcare professionals

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

Dear Sir or Madam,

We would like to invite you to take part in a research study which aims to explore how people with coeliac disease manage sticking with a gluten-free diet.

The study is being conducted by PhD student, Helen Flaherty, who is the Chief Investigator in this study. Helen is working under the supervision of two academic supervisors, Dr Katherine Deane and Prof Richard Gray at the University of East Anglia. Before you give your full consent, please take time to read the following information carefully and discuss it with others if you wish. Please ask us if there is anything that is not clear or if you would like more information.

Participation in this study is optional and you are free to withdraw at any time without giving a reason.

Part 1 tells you the purpose of this study and what will happen if you participate. Part 2 gives you more detailed information about the conduct of the study.

If you would like to take part in this study, please complete the enclosed consent form and short questionnaire and return them in the envelope provided as soon as possible. It should take you no longer than five minutes to complete these forms. After receiving your completed consent form and questionnaire, Helen Flaherty will contact you with further information about the study.

Thank you for taking your time to read this.

Yours faithfully

Miss Helen Flaherty
Chief Investigator

Part 1

What is the purpose of the study?

The only effective treatment for coeliac disease is a diet that is free from gluten. Gluten is found in wheat, rye and barley. Some people may also have an adverse reaction to oats. Removing food and drinks from the diet which contain gluten, such as bread, breakfast cereals, pasta, beer and cakes, can be difficult. It is known that many people with coeliac disease do not stick to a strict gluten-free diet.

In this study we will explore the factors influencing whether or not adults with coeliac disease stick to a gluten-free diet. To do this, we will collect information from 25 adults with coeliac disease, 25 spouses or other adults who live with someone with coeliac disease and 25 healthcare professionals. The study will determine whether these three groups of people have the same or different ideas about why people with coeliac disease do or do not stick to a gluten-free diet. The results of this study can be used to design an intervention aimed at improving healthcare for people with coeliac disease.

This study is being undertaken for educational purposes, as part of my PhD.

Why have I been Invited?

You have been invited to participate in this study because you are a healthcare professional working in Norwich who treats people with coeliac disease.

Are there any inclusion or exclusion criteria?

We are seeking to recruit 25 healthcare professionals who have a current caseload of at least one adult coeliac patient. Participants should have at least one year's experience of working with coeliac patients and should live or work within approximately a 10 mile radius of Norwich.

Do I have to take part?

Participation in this study is voluntary and it is up to you to decide. You are free to withdraw at any time, without giving a reason. If you wish to take part in this study, please return your completed consent form and questionnaire in the envelope provided.

What will happen to me if I take part?

Before we can include you in this study, you will need to complete the enclosed consent form and questionnaire and return them to us in the envelope provided. You will then be invited to participate in a group meeting with other healthcare professionals who work with adult coeliac patients. This meeting will take place between November 2011 and March 2012 in the Edith Cavell Building, opposite Norfolk and Norwich University Hospital. The meeting is really an informal discussion and is expected to last no more than two hours. There will be a break during the meeting and refreshments will be provided. The group meeting will be run by the Chief Investigator, Helen Flaherty and there will be at least one research assistant present.

In the meeting, you and the other members of your group will be asked to produce statements that answer the question "What helps a person with coeliac disease stick to a gluten-free diet?". The discussion will be tape-recorded and statements will be written on flip charts. You will also have a note pad on which you can individually write your statements if you wish. There is no pressure on any individual to make statements and any information you provide during the meeting will be kept

anonymous. If you are unable to attend the group meeting we will be able to record your statements by e-mail or post.

There will be a second activity which we will ask you to complete either electronically (e-mail) or by post. This will take place between 1 to 4 months after the group meeting. This activity involves looking through the statements that were generated by all three participant groups during the first meetings. There will be around 80 statements. You will be asked to sort the statements into five categories indicating their relative importance and then you will be asked to group the statements into themes.

Expenses and payments:

Although we do not have funding to compensate you for your time, we will refund your travel expenses.

What are the possible disadvantages and risks of taking part?

The risk of you being harmed as a result of this study is very low. If you feel distressed at any time during this study, you are free to withdraw without having to provide a reason.

For health professionals participating in this study, there is a risk that the chief investigator could be made aware of unprofessional practice during discussions and this would have to be reported to an appropriate authority.

A possible disadvantage for participants is that you will be asked to give up two hours of your time to attend the two-hour brainstorming group meeting. Although you will not be compensated for your time, we will reimburse your travel expenses and we will provide refreshments during the meeting.

What are the possible benefits of taking part?

We cannot promise that the study will help you or your patients directly but it is our intention to use the information we get from this study to inform a lay report of this research for Coeliac UK and a professional report for a peer reviewed journal which will, hopefully, help improve the treatment of people with coeliac disease.

What if there is a problem?

Any complaint about the way you have been dealt with during the study or any possible harm you might suffer will be addressed. The detailed information on this is given in Part 2.

Will my taking part in this study be kept confidential?

Yes. We will follow ethical and legal practice and all information about you will be handled in confidence. The details are included in Part 2.

This completes part 1. If the information in part 1 interests you and you would like to participate in this study, please read the additional information in part 2 before making any decision.

Part 2

Once I take part, can I change my mind?

Yes! After you have read this information and asked us any questions, you can complete the enclosed Informed Consent Form. However, if at any time, before, during or after the study you wish to withdraw, please just contact the Chief Investigator, Helen Flaherty. You can withdraw at any time and you do not have to explain the reason why you have decided to withdraw.

What if there is a problem?

If you have a concern about any aspect of this study, you should ask to speak to the researchers who will do their best to answer your questions. The Chief Investigator, Helen Flaherty, can be contacted by telephone: 01603593665 (office) / 07920406094 (mobile) or email: h.flaherty@uea.ac.uk. If you remain unhappy and wish to complain formally, you can do this by contacting Helen's academic supervisor, Dr Katherine Deane, by telephone: 01603 597047 or e-mail: K.Deane@uea.ac.uk. Alternatively, you can write to us at:

Dr Katherine Deane / Helen Flaherty
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ

In the event that something does go wrong and you are harmed during the research and this is due to someone's negligence then you may have grounds for a legal action for compensation against the University of East Anglia but you may have to pay your legal costs.

Will my taking part in this study be kept confidential?

Yes. All information collected about you during the course of this study will be kept strictly confidential. Your personal details, such as your name and address, will be recorded and stored anonymously according to the Data Protection Act (1998). Your name will be substituted by a coded reference number so that your information cannot be traced back to you. Paper documents which contain your data will be stored in a locked cabinet in a locked room at the University of East Anglia. Paper documents will be shredded six months after the end of this study. Electronic data will be stored securely on a password protected computer at the University of East Anglia for two years. After this, all data will be deleted. Data will only be viewed by the Chief Investigator and members of the research team.

If the data collected in this study is used in a written report, such as a thesis or published paper, we will not use your name or personal details. Any information you provide will be kept anonymous, however, you are not obliged to disclose any information that you would prefer to keep private.

What will happen to the results of the study?

The results of this study can be used to develop an intervention to help improve adherence to a gluten-free diet for people with coeliac disease. This may improve the quality of life of many people with coeliac disease as well as reducing the cost of healthcare.

We aim to publish the results of this study in a lay report for Coeliac UK, in scientific journals and the study will be used as part of a written PhD thesis. You will not be personally identifiable in any report or publication relating to this study.

The results of this study will be made available to you upon your request. Please use the contact details shown at the top of this information sheet when requesting the results.

Who is organising and funding the research?

This study is part of a three year student PhD research project funded by the University of East Anglia. The Chief Investigator is PhD student, Helen Flaherty. Helen will work under the supervision of Dr Katherine Deane and Professor Richard Gray.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by the Newcastle North Tyneside 2 Research Ethics Proportionate Review Sub-Committee. In addition, the research study has been reviewed by staff at Coeliac UK and Norfolk and Norwich University Hospital.

Further information and contact details:

Please do not hesitate to contact us if you require further information about any of the following issues:

Specific information about this research project.

Helen Flaherty
1.33 Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 593665 / 07920406094
E-mail: h.flaherty@uea.ac.uk

Advice as to whether you should participate.

If you are unsure of whether to take part, you can discuss your participation in this study with your friends and family or another healthcare professional.

Who you should approach if you are unhappy with the study.

Dr Katherine Deane
Edith Cavell Building
University of East Anglia
Norwich
NR4 7TJ
Tel: 01603 597047
E-mail: K.Deane@uea.ac.uk

Thank you for taking your time to read this information sheet. Please complete and return the enclosed consent form and questionnaire in the envelope provided if you would like to participate in this study.

Appendix 15 Questionnaire for adults with coeliac disease



Questionnaire

Research Study: What helps adults with coeliac disease stick to a gluten-free diet?

Please complete this questionnaire if you have been medically diagnosed with coeliac disease and you are aged 18 years or over.

1) **Your gender:** (*Please tick*) Male Female

2) **How many years ago were you diagnosed with coeliac disease?**

.....

3) **Do you follow any other special diet/s:** (*Please select all that apply*)

Vegetarian/vegan Nut-free Lactose-free Other (Please state).....

4) **How severe are your symptoms when you eat gluten?** (*Please tick*)

Very severe symptoms	Severe	Moderate	Mild	Very mild	No
<input type="checkbox"/>					

6) **Your ethnic group:** (*Please tick*)

White: British Irish Other white background

Mixed: White and Asian White and Black African

White and Black Caribbean Other mixed background

Asian or Asian British: Bangladeshi Chinese Indian

Pakistani Other Asian background

Black or Black British: Caribbean African Other black background

Other ethnic background (*Please state*)

7) **In what year were you born?** _ _ _ _

8) What is your highest educational qualification? (Please tick)

- No formal qualification
- GCSE/O Level or equivalent
- A' Level or equivalent
- University Degree or equivalent
- Postgraduate Degree/Diploma or equivalent

**Thank you for taking the time to complete this questionnaire.
Please return it to us in the envelope provided.**

Appendix 16 Questionnaire for household members



Questionnaire

Research Study: What helps adults with coeliac disease stick to a gluten-free diet?

Please complete this questionnaire if you are aged 18 years or over and you have lived in the same household as an adult who has coeliac disease for at least one year.

1) What is your relationship to the person with coeliac disease?
(e.g. husband / girlfriend / friend etc.)

2) Your gender: (*Please tick*) Male Female

3) The gender of the person with coeliac disease: (*Please tick*)
 Male Female

4) Your ethnic group: (*Please tick*)

White: British Irish Other white background

Mixed: White and Asian White and Black African

White and Black Caribbean Other mixed background

Asian or Asian British: Bangladeshi Chinese
 Indian Pakistani Other Asian background

Black or Black British: Caribbean African
 Other black background

Other ethnic background (*Please state*).....

5) In what year were you born? _ _ _ _

6) What is your highest educational qualification? (Please tick)

- No formal qualification
- GCSE/O Level or equivalent
- A' Level or equivalent
- University Degree or equivalent
- Postgraduate Degree/Diploma or equivalent

**Thank you for taking the time to complete this questionnaire.
Please hand your completed questionnaire to a member of the
research team or return it in the envelope provided.**

Appendix 17 Questionnaire for healthcare professionals



Questionnaire

Research Study: What helps adults with coeliac disease stick to a gluten-free diet?

Please complete this questionnaire if you are a healthcare professional who has worked with adult patients with coeliac disease for at least one year and if you currently work with at least one adult coeliac patient.

1) Please state your job title:

.....

2) For how many years have you worked with patients with coeliac disease?years

3) How many patients with coeliac disease do you currently work with?

4) Please provide details of your professional qualification/s:

.....

5) Please tick the appropriate box/es to show when you are most likely to be available to attend the group meeting/s:

	MON	TUES	WED	THURS	FRI	SAT	SUN
Morning (9am-12pm)							
Afternoon (12pm-4pm)							
Evening (6pm-9pm)							

Name: Title: (please circle) Dr / Mr / Mrs / Miss / Ms /

Other:.....

First name/s: Surname:

Preferred contact address:

House number/name:Street:

Town/City: Postcode:

Preferred contact tel number:

.....(Home/work/mobile)

Preferred contact e-mail address:

.....
How would you prefer to be contacted (please tick):

Phone E-mail Post

Thank you for taking the time to complete this questionnaire.
Please return your completed questionnaire in the envelope provided

along with your signed consent form. We will be in touch within 3

weeks of receiving these documents.

Appendix 18 Consent form for adults with coeliac disease



Consent form for adults with coeliac disease

Research Project: What helps adults with coeliac disease stick to a gluten-free diet?

Chief Investigator: Helen Flaherty

Participant Identification Number for this study: Please initial box

1. I confirm that I have read and understand the information sheet dated 16/05/2011 (version 3) for the research project called "What helps adults with coeliac disease stick to a gluten-free diet?".

2. I have had a chance to think about it and ask any questions. I confirm that my questions have been answered satisfactorily.

3. I know that I don't have to do this, it is my own choice. If I start joining in the research, I know that I can stop if I want to at any time. I will still be cared for in the same way, whether I join in or not and I do not have to give a reason for stopping.

4. I agree to have the group meeting audio-recorded. I understand that the audio-recordings will be listened to by the research team at the University of East Anglia and they will be deleted within 3 months.

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

Name of Participant (please print):

Signature: Date:

Name of Person taking consent (please print):

Signature: Date:

When completed: 1 copy for participant; 1 copy (original) for researcher site file.

Appendix 19 Participant consent form for household members



Consent form for spouses (or other adults) who live with a person with coeliac disease

Research Project: What helps adults with coeliac disease stick to a gluten-free diet?

Chief Investigator: Helen Flaherty

Participant Identification Number for this study:

Please initial box

1. I confirm that I have read and understand the information sheet dated 16/05/2011 (version 3) for the research project called "What helps adults with coeliac disease stick to a gluten-free diet?".

2. I have had a chance to think about it and ask any questions. I confirm that my questions have been answered satisfactorily

3. I know that I don't have to do this, it is my own choice. If I start joining in the research, I know that I can stop if I want to at any time. I will still be cared for in the same way, whether I join in or not and I do not have to give a reason for stopping.

4. I agree to have the group meeting audio-recorded. I understand that the audio-recordings will be listened to by the research team at the University of East Anglia and they will be deleted within 3 months.

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

Name of Participant (please print):

Signature: Date:

Name of Person taking consent (please print):

Signature: Date:

When completed: 1 copy for participant; 1 copy (original) for researcher site file.

Appendix 20 Participant consent form for healthcare professionals



Consent form for healthcare professionals

Research Project: What helps adults with coeliac disease stick to a gluten-free diet?

Chief Investigator: Helen Flaherty

Participant Identification Number for this study: Please initial box

1. I confirm that I have read and understand the information sheet dated 09/12/2011 (version 4) for the research project called "What helps adults with coeliac disease stick to a gluten-free diet?".

2. I have had a chance to think about it and ask any questions. I confirm that my questions have been answered satisfactorily.

3. I know that I don't have to do this, it is my own choice. If I start joining in the research, I know that I can stop if I want to at any time. I do not have to give a reason for stopping.

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

Name of Participant (please print):.....

Signature: Date:

Please keep one copy of this consent form and return the other copy to us in the envelope provided along with your completed questionnaire.

Appendix 21 Reply slip for adults with coeliac disease



REPLY SLIP

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

If you are an adult who has been diagnosed with coeliac disease and you are interested in participating in this study, please complete this reply slip and return it to us in the envelope provided. We will not give your details to anybody outside of our research team here at the University of East Anglia. Any information you provide will be kept strictly confidential.

Name:

Title: (please circle) Dr / Mr / Mrs / Miss / Ms / Other:

.....

First name/s: Surname:

Address:

House number/name:.....

Street:

Town/City:Postcode:.....

Phone:

Home: Mobile: Work:

E-mail:

As we are interested to hear about the things that both increase and decrease a person's ability to stick to a strict gluten-free diet, we are looking to recruit people who do and people who do not stick to a gluten-free diet. Please state your current level of adherence to a gluten-free diet by ticking the appropriate box below:

I stick to a gluten-free diet all of the time

I partially stick to a gluten-free diet

I do not follow a gluten-free diet

If you are returning this reply slip to us, you should be willing to attend two two-hour meetings in Norwich. Please tick the appropriate box/es to show which times during the week you are most likely to be available to attend these meeting/s:

	MON	TUES	WED	THURS	FRI	SAT	SUN
Morning (9am-12pm)							
Afternoon (12pm-4pm)							
Evening (6pm-9pm)							

Signature: Date:

Thank you for completing this form. Please return it to us in the envelope provided as soon as possible.

Appendix 22 Reply slip for household members



REPLY

SLIP

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

If you live with an adult who has been diagnosed with coeliac disease and you are interested in participating in this study, please complete this reply slip and return it to us in the envelope provided. We will not give your details to anybody outside of our research team here at the University of East Anglia. Any information you provide will be kept strictly confidential.

Name:

Title: (please circle) Dr / Mr / Mrs / Miss / Ms / Other:
First name/s: Surname:

Address:

House number/name:.....
Street:
Town/City: Postcode:

Phone:

Home: Mobile: Work:

E-mail:

If you are returning this reply slip to us, you should be willing to attend two two-hour meetings in Norwich. Please tick the appropriate box/es to show which times during the week you are most likely to be available to attend these meeting/s:

	MON	TUES	WED	THURS	FRI	SAT	SUN
Morning (9am-12pm)							
Afternoon (12pm-4pm)							
Evening (6pm-9pm)							

Signature: Date:

Thank you for completing this form. Please return it to us in the envelope provided as soon as possible.

Appendix 23 Ethical approval letter



National Research Ethics Service

NRES Committee North East - Newcastle & North Tyneside 2

Room 002
TEDCO Business Centre
Rolling Mill Road
Jarrow
NE32 4BW

Telephone: 0191 428 3565
Facsimile: 0191 428 3432

21 April 2011 Re-issued

Miss Helen Flaherty
PhD Student
University of East Anglia
Faculty of Health
1.33 Elizabeth Fry Building
Norwich
NR4 7TJ

Dear Miss Flaherty

Study title: Adherence to a gluten-free diet in adults with coeliac disease: Exploring multiple perspectives.
REC reference: 11/NE/0111

The Proportionate Review Sub-committee of the NRES Committee North East - Newcastle & North Tyneside 2 Research Ethics Committee reviewed the above application on 21 April 2011.

Ethical opinion

The sub-committee considered this to be a worthwhile study.

The sub-committee raised the following issues and as chief investigator you responded accordingly as follows -

1. It was queried if there is a possibility that recruitment for this study may be low and potential participants may have already received duplicate invitations. If so, the committee requested that you make it quite clear to potential participants in the documentation why they are being continually asked to participate.

There is indeed a possibility that the participant response rate may be low, however, this is considered unlikely. Previous studies have shown high response rates from adults with coeliac disease. In addition, Coeliac UK will only send the invitation letter to members who have previously expressed an interest in participating in research. In the invitation letters a statement has been included advising adults with coeliac disease that they may receive more than one invitation (one from Coeliac UK and one from NNUH if they are registered with both organisations). Reminders may be sent to individuals who return the reply slip only if they do not respond to further correspondence - this will only be done once if it is necessary. Individuals will not be asked continually to participate in this project. Anybody who is not interested in participating will receive a maximum of two invitation letters (one from Coeliac UK and one from NNUH if they are registered with both). If they do not respond to the invitation, they will not be contacted again.

2. It was queried if there a specific reason why recruitment is being restricted to a 10 mile radius of Norwich.

It was decided to restrict recruitment to within 10 miles of Norwich because participants will be asked to travel to a venue in Norwich for two group meetings. Funding for this study is fairly low and as participants' travel costs will be reimbursed, it would be too expensive if they were to travel from a longer distance. In addition, you intend to travel to the participants' homes to gain consent prior to the group sessions and as you do not have your own transport, this would be too problematic and expensive to travel further than 10 miles each time. Consultations have been held with Coeliac UK in relation to the number of potential members living within 10 miles of Norwich. Coeliac UK has over 1000 members who are interested in participating in research, living within 10 miles of Norwich and this is far more than is required for this study (a random sample from these members will be selected).

The sub-committee was satisfied with the responses given to the issues raised.

On behalf of the Committee, the sub-committee gave a **Favourable** ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

For NHS research sites only, management permission for research ("R&D approval") should be obtained from the relevant care organisation(s) in accordance with NHS research governance arrangements. Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at <http://www.rforum.nhs.uk>.

Where the only involvement of the NHS organisation is as a Participant Identification Centre (PIC), management permission for research is not required but the R&D office should be notified of the study and agree to the organisation's involvement. Guidance on procedures for PICs is available in IRAS. Further advice should be sought from the R&D office where necessary.

Sponsors are not required to notify the Committee of approvals from host organisations.

Additional condition specified by the REC

- The participant information sheet should note that the results of the study will be available to participants if they request this – a revised copy is requested.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Approved documents

The documents reviewed and approved were:

<i>Document</i>	<i>Version</i>	<i>Date</i>
REC application	IRAS v 3.1	12 April 2011
Protocol	v 1	20 March 2011
Letter of invitation to participant	v 1 (from UEA to Coeliac UK members)	29 March 2011
Letter of invitation to participant	v 1 (from UEA to Household members)	11 March 2011
Questionnaire: Non-Validated - Household Members	v 1	11 March 2011
Questionnaire: Non-Validated - Healthcare Professionals	v 1	11 March 2011
CV for Academic Supervisor	K Deane	11 March 2011
Questionnaire: Non-Validated - Adults with Coeliac Disease	v 1	11 March 2011
Evidence of insurance or indemnity	Zurich Municipal	16 June 2010
Investigator CV	H Flaherty	07 April 2011
Covering Letter	Helen Flaherty	08 April 2011
Reply slip Coeliac UK members	v 1	05 April 2011
Letter of Invitation - From NNUH to Healthcare Professionals	v 1	11 March 2011
Reply slip - Adults with Coeliac Disease	v 1	11 March 2011
Letter of Invitation to participants - From UEA to Coeliac UK members	v 1	29 March 2011
Letter of Invitation to participants - From UEA to General Practices	v 1	11 March 2011
Letter of Invitation to participants - From Coeliac UK to Adults with Coeliac Disease	v 1	11 March 2011
Reply slip - Household members	v 1	11 March 2011
Letter from sponsor	UEA	12 April 2011
Email Notification	H Flaherty	11 March 2011
Participant Information Sheet: Household members	v 1	11 March 2011
Participant Information Sheet: Healthcare professionals	v 1	11 March 2011
Participant Information Sheet: Adults with coeliac disease	v 1	11 March 2011
Participant Consent Form: Adults with Coeliac disease	v 1	11 March 2011
Participant Consent Form: Household members	v 1	11 March 2011
Participant Consent Form: Healthcare Professionals	v 1	11 March 2011

Membership of the Proportionate Review Sub-Committee

The members of the Sub-Committee who took part in the review are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Now that you have completed the application process please visit the National Research Ethics Service website > After Review

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

11/NE/0111**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project

Yours sincerely



**Professor Philip M Preshaw
Chair**

Email: gillian.mayer@sotw.nhs.uk

Enclosures: List of names and professions of members who took part in the review

"After ethical review – guidance for researchers" SL-AR2

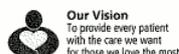
Copy to: Ms Tracy Moulton – Research Contracts Manager, University of East Anglia, Norwich NR4 7TJ

NRES Committee North East - Newcastle & North Tyneside 2**Attendance at PRS Sub-Committee of the REC meeting on 21 April 2011****Committee Members:**

<i>Name</i>	<i>Profession</i>	<i>Present</i>	<i>Notes</i>
Mr Andrew Brenikov	Historian	Yes	
Professor Philip M Preshaw (<i>Chair</i>)	Professor of Periodontology and Consultant in Restorative Dentistry	Yes	
Mr Michael Wyatt (<i>Vice Chair</i>)	Consultant General and Vascular Surgeon	Yes	

Appendix 24 Research and development

application and approvals



Norfolk and Norwich University Hospitals **NHS**
NHS Foundation Trust

Miss Helen Flaherty
University of East Anglia
Faculty of Health
1.33 Elizabeth Fry Building
Norwich
Norfolk
NR4 7TJ
United Kingdom

Research & Development Office
Level 3 East
Norfolk & Norwich University Hospitals NHS Foundation Trust
Colney Lane
Norwich
NR4 7UY

direct dial: 01603 287806
direct fax: 01603 289800
e-mail: rdoffice@nnuh.nhs.uk
website: www.nnuh.nhs.uk

22 September 2011

Dear Miss Helen Flaherty

Re: R&D Reference Number: 2011GAST04S (58-05-11)

Project Title: Adherence to a gluten-free diet in adults with coeliac disease: Exploring multiple perspectives

I am pleased to inform you that the above project has been given full NHS permission for research at Norfolk & Norwich University Hospitals NHS Foundation Trust.

This NHS permission for research has been granted on the basis described in the application form, protocol and supporting documentation. The documents reviewed were:

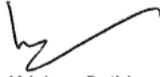
<u>Document</u>	<u>Version</u>	<u>Date</u>
Protocol	1	20/03/2011
Invitation letter - to Coeliac UK members	2	16/05/2011
Invitation letter - to household members	2	16/05/2011
Invitation letter - to GP Practices	2	16/05/2011
Invitation letter - to healthcare professionals	2	16/05/2011
Invitation letter - adults with coeliac disease	2	16/05/2011
Consent form - household members	3	16/05/2011
Consent form - healthcare professionals	3	16/05/2011
Consent form - adults with coeliac disease	3	16/05/2011
PIS - household members	3	16/05/2011
PIS - healthcare professionals	3	16/05/2011
PIS - adults with coeliac disease	3	16/05/2011
Reply slip - household members	2	16/05/2011
Reply slip - adults with coeliac disease	2	16/05/2011
Questionnaire - household members	1	11/03/2011
Questionnaire - healthcare professionals	2	12/05/2011
Questionnaire - adults with coeliac disease	1	11/03/2011

I have enclosed two copies of the Standard Terms and Conditions of Approval. Please sign both copies returning one copy to the Research Governance office at the above address and keeping the other in your study file. Failure to return the standard terms and conditions may affect the conditions of approval.

Please note, under the agreed Standard Terms and Conditions of Approval you must inform the R&D department of any proposed changes to this study and submit annual progress reports to the R&D department.

If you have any queries regarding this or any other project please contact Dr. Seema Gopinath, Research Governance Administrator, at the above address. Please note, the reference number for this study is **2011GAST04S (58-05-11)** and this should be quoted on all correspondence.

Yours sincerely



Professor Krishna Sethia
Medical Director

Enc

Carbon Copy: (POC) Dr Ian Fellows

Appendix 25 Ethics substantial amendment approval letter

NHS
Health Research Authority

NRES Committee North East - Newcastle & North Tyneside 2

Room 002
TEDCO Business Centre
Rolling Mill Road
Jarrow
NE32 4BW
Tel: 0191 428 3565
Fax: 0191 428 3432

13 February 2012

Miss Helen Flaherty
PhD Student
University of East Anglia
Faculty of Health
1.33 Elizabeth Fry Building
University of East Anglia, Norwich
NR4 7TJ

Dear Miss Flaherty

Study title: Adherence to a gluten-free diet in adults with coeliac disease: Exploring multiple perspectives
REC reference: 11/NE/0111
Amendment number: Amendment 1 (9.12.11)
Amendment date: 14 December 2011

The above amendment was reviewed at the meeting of the Sub-Committee held on 18 January 2012.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
Notice of Substantial Amendment (non-CTIMPs)	Amendment 1 (9.12.11)	14 December 2011
Covering Letter	H Flaherty	14 December 2011
Email from sponsor	v 1	13 December 2011
Questionnaire: Healthcare Professionals	v 3	09 December 2011
Letter of invitation to participant	v 1 (Adults with CD)	09 December 2011
Letter of invitation to participant	v 1 (Healthcare Professionals)	09 December 2011
Participant Consent Form: Adults with CD	v 4	09 December 2011
Participant Consent Form: Household Members	v 4	09 December 2011
Participant Consent Form: Healthcare Professionals	v 4	09 December 2011
Participant Information Sheet: Adults with CD	v 4	09 December 2011

A Research Ethics Committee established by the Health Research Authority

Participant Information Sheet: Household Members	v 4	06 December 2011
Participant Information Sheet: Healthcare Professionals	v 4	06 December 2011
Protocol	v 2	06 December 2011
Reply slip - Household Members	v 3	06 December 2011
Reply slip - Adults with Coeliac Disease	v 3	06 December 2011
Advertisement	v 2 (Advert for newspaper, BDA & UEA website)	12 December 2011

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

R&D approval

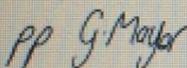
All investigators and research collaborators in the NHS should notify the R&D office for the relevant NHS care organisation of this amendment and check whether it affects R&D approval of the research.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

11/NE/0111:	Please quote this number on all correspondence
-------------	--

Yours sincerely



Professor Philip M Preshaw
Chair

E-mail: gillian.mayer@scw.nhs.uk

Enclosures: *List of names and professions of members who took part in the review*

Copy to: *Mrs Tracy Moulton – Research Contracts Manager, University of East Anglia, Norwich NR4 7TJ*

NRSS Committee North East - Newcastle & North Tyneside 3

Attendance at Sub-Committee of the REC meeting on 18 January 2012

Name	Profession	Capacity
Professor Philip M Preshaw (Chair)	Professor of Periodontology and Consultant in Restorative Dentistry	Expert
Mr Chris Barron	NCRN Team Lead	Expert
Dr Catherine F M Bartley	Consultant in Anaesthesia and Intensive Care	Expert
Mr Andrew Branikov	Historian	Lay Plus
Dr Robert Jefferson	Consultant in Environmental Medicine	Expert
Mr Peter Morton (Alternate Vice Chair)	Voluntary Worker/Former Contracts-Procurement Manager	Lay

Also in attendance:

Name	Position (or reason for attending)
Ms Gillian Mayer	Committee Co-ordinator

Appendix 26 Press release

The following press release was placed in two local newspapers (The Eastern Daily Press and the Norwich Evening News) to invite partially-adherent and non-adherent people with coeliac disease and household members to participate in this study. This was done after we had recruited a sufficient number of adherent participants.

Press Release

Researchers are looking to recruit people with coeliac disease, who do not follow a strict gluten-free diet, for a new study at the University of East Anglia.

The researchers want to find out what could help adults with coeliac disease to be better at sticking to a gluten-free diet for life.

“Following a gluten-free diet for life can be very challenging for people with coeliac disease. We are interested to hear from adults with coeliac disease who do not currently follow a strict gluten-free diet” said Dr Katherine Deane.

“Their views are very important for us in identifying the things that could help people with coeliac disease to be better at sticking to a gluten-free diet,” she added.

As well as recruiting people with coeliac disease, the researchers are also recruiting spouses or other adults who live in the same household as the person with coeliac disease to take part in the study.

Volunteers will be asked to complete two tasks which can be done in their own time and returned to the researchers by e-mail or post. The first task will take approximately 30 minutes, and the second approximately 60 minutes.

Alternatively, participants can complete the tasks during group sessions held at the University of East Anglia (with approximately six other volunteers in each group). These group sessions will take between one to two hours each and refreshments will be provided.

The researchers are looking for 15 adults with coeliac disease and 15 spouses or other adult household members to volunteer for this study. All volunteers need to be aged 18 years or over.

To find out more about the research or to volunteer, please contact the researcher, Helen Flaherty, by emailing h.flaherty@uea.ac.uk, or call 01603 593665 or 07920 406094.

Appendix 27 – Advertisement on Coeliac UK’s website for healthcare professionals

Invitation to participate in research study

Coeliac UK is working with the University of East Anglia (UEA) on a study to explore what helps adults with coeliac disease stick to a gluten-free diet. We are seeking the opinions of healthcare professionals who have worked with an adult with coeliac disease in the last year. We would like to know if you would be willing to take part in this study. The study will involve two tasks which can be completed in your own time and submitted online or by post. Each task should take you no longer than one hour to complete.

If you are interested in taking part, please contact Helen Flaherty at the University of East Anglia who will provide you with further information about the study before you decide. This study has gained ethical approval and it meets all of the necessary research standards.

E-mail address: h.flaherty@uea.ac.uk

Tel: 07920406094

Postal address: Helen Flaherty
1.33 Elizabeth Fry Building
Faculty of Medicine and Health Sciences
University of East Anglia
Norwich Research Park
Norwich
NR4 7TJ

Appendix 28 Advert on the University of East Anglia website

The following advertisement was placed on the University of East Anglia website:

Researchers at the University of East Anglia are looking to recruit people with coeliac disease who are aged 18 years or over and who do not currently follow a strict gluten-free diet.

The researchers want to find out what could help adults with coeliac disease to be better at sticking to a gluten-free diet for life.

“Following a gluten-free diet for life can be very challenging for people with coeliac disease. We are interested to hear from adults with coeliac disease who do not currently follow a strict gluten-free diet” said Dr Katherine Deane.

“Their views are very important for us in identifying the things that could help people with coeliac disease to be better at sticking to a gluten-free diet” she added.

As well as recruiting people with coeliac disease for this study, the researchers are also recruiting spouses or other adults who live in the same household as the person with coeliac disease.

Volunteers will be asked to complete two tasks which can be done in your own time and returned to the researchers by e-mail or post. The first task will take approximately 30 minutes and the second will take approximately 60 minutes. Alternatively, participants can complete the tasks during group sessions held at the University of East Anglia (with approximately 6 other volunteers in each group). These group sessions will take between 1 to 2 hours each and refreshments will be provided (gluten-free, of course!).

The researchers are looking for 15 adults with coeliac disease and 15 spouses or other adult household members to volunteer for this study.

To find out more about the research or to volunteer, please contact the researcher, Helen Flaherty, by emailing h.flaherty@uea.ac.uk or call 01603 593665 / 07920406094.

Appendix 29 Advertisement sent by Coeliac UK to the British Dietetic Association (BDA) Gastroenterology Specialist Group

Invitation to participate in research study

Coeliac UK is working with the University of East Anglia (UEA) on a study to explore what helps adults with coeliac disease stick to a gluten-free diet. We are seeking the opinions of healthcare professionals who have worked with an adult with coeliac disease in the last year. We would like to know if you would be willing to take part in this study. The study will involve two tasks which can be completed in your own time and submitted online or by post. Each task should take you no longer than one hour to complete.

If you are interested in taking part, please contact Helen Flaherty at the University of East Anglia who will provide you with further information about the study before you decide. This study has gained ethical approval and it meets all of the necessary research standards.

E-mail address: h.flaherty@uea.ac.uk

Tel: 07920406094

Postal address: Helen Flaherty
1.33 Elizabeth Fry Building
Faculty of Medicine and Health Sciences
University of East Anglia
Norwich Research Park
Norwich
NR4 7TJ

Appendix 30 Letter of confirmation for group meeting

Room 1.33



Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ

Phone: 01603 593665
Mobile: 07920 406094
E-mail: h.flaherty@uea.ac.uk

Dear

Research study: What helps adults with coeliac disease stick to a gluten-free diet?

Brainstorming session - Friday 16th September 2011 7:00pm–9:00pm

Thank you for agreeing to take part in the above brainstorming session on Friday 16th September at 7:00pm. This meeting will last up to two hours and there will be a break with refreshments.

The meeting will be held in the Edith Cavell Building, which is opposite the Norfolk and Norwich University Hospital on Colney Lane (see enclosed maps). As the main entrance to the Edith Cavell Building will be locked on Friday evening, my colleagues and I will meet you at the main entrance when you arrive.

The Edith Cavell Building has its own car park which is accessed from Colney Lane. Parking is free and you can use the disabled parking spaces if you wish. There are regular buses direct to the Hospital from Norwich centre (bus numbers 12, 21, 22 and 24).

We would like to reimburse any travel expenses you may incur. Please complete the enclosed expenses claim form, attaching your travel tickets/receipts or stating the number of miles travelled by car. You can hand the form to me on Friday or at the second meeting which you will be invited to attend later this year. Alternatively, you can post the form to me at the above address.

If you have any questions or need further information, please feel free to contact me using the details shown at the top of this letter. I will be available on my mobile phone on the evening of the meeting.

Yours Sincerely

Helen Flaherty
PhD student, University of East Anglia

Appendix 31 Brainstorming script and prompts to be used during brainstorming

Brainstorming Session Script and prompts

Script

At the start:

Thank you all for coming along tonight, it is nice to see you all again and we really appreciate you giving up your time to participate in this study.

I'd like to introduce you to my supervisor, Dr Katherine Deane and two of my fellow PhD students who have kindly volunteered to help out, Steve Smith and Ada Mackovova (we should wear name badges). (do you want to say something about yourselves or should I say more about you?). To make it easier for us to remember all your names, we would be grateful if you would wear name badges (have stickers and marker pens to hand).

Tonight's brainstorming session will last up to two hours and we'll take a break at 8pm for refreshments.

Following any restricted diet can be very difficult and studies have shown that up to 58% of adults with CD do not stick to a gluten-free diet. There are many reasons why this may be and through this research, we hope to identify what would help adults with coeliac disease to be better at sticking to a gluten-free diet.

One definition of brainstorming is: "A means of getting a large number of ideas from a group of people in a short time". The aim of tonight's brainstorming session is to generate as many of your ideas as possible in response to the focus question "what helps adults with coeliac disease stick to a gluten-free diet". To do this, we will ask you to complete the following sentence: "It would be easier for adults with coeliac disease to stick to a gluten-free diet if...". For example you could say, "It would be easier for adults with coeliac disease to stick to a gluten-free diet if gluten-free food was cheaper".

You or your household member may be very good at sticking to the GFD and you may have strategies in place to help you manage the diet which we would really like to hear about. For example, if your local shops don't stock much GFF, you may shop online as a strategy to help you stick to the diet. Two statements could be generated in relation to this:

1. "It would be easier for adults with coeliac disease to stick to a gluten-free diet if more gluten-free food was available in local shops".
2. "It would be easier for adults with coeliac disease to stick to a gluten-free diet if they shop for gluten-free food online".

You will not be required to analyse or discuss the statements. We simply want to collect as many statements as possible. There is a need to suspend judgement and to accept ideas that you might think are wild or silly. If you have an idea that you would rather not share with the group, you can write it down and hand it to one of us at the end.

Tonight's session will be tape recorded so that we can ensure we don't miss anything. The recording will be listened to again by the research team and then it will be deleted. To help with the clarity of the recording, we would be grateful if just one person speaks at a time. If you have an idea while someone else is talking and you don't want to forget it, then please write it down. Everyone should have an equal opportunity to contribute their ideas.

I also ask that you respect each other's views even if you do not agree with them. If you disagree with someone's statement, you can generate a conflicting statement.

I would also like to remind you that you are free to withdraw at any time without having to explain your reason.

Does anybody have any questions before we begin?

Fire evacuation procedures - THE EARLHAM, COLNEY AND BLUEBELL ROOMS (FIRST FLOOR)

In the event of discovering a fire, please raise the alarm.

- Leave via the entrance/exit door
- Turn left and exit via the solid "push bar" fire exit door
- Take the internal staircase down and exit through the glass "push bar" door at the bottom.
- Carefully cross the road and wait on the pavement opposite the Sportspark.

[Do not re-enter the building unless authorised to do so by a member of Sportspark Staff](#)

At the break:

Please help yourself to drinks and the GF snacks which have been donated by gluten-free food companies. In return for their generosity,

I agreed to distribute their leaflets, so please take these if you wish. I have some additional gluten-free food donations for the people with CD to take away and try at home (put together a bag for each person with leaflets in). For household members/spouses, we will give the person who you live with a pack when they attend their brainstorming session.

At the end:

Thank you all for coming along and giving up your time. I hope you have enjoyed taking part. You have provided us with some valuable data and we really appreciate your contributions. If you do have any more ideas that you think of later or if there is anything that you didn't wish to raise during the meeting, you can contact me later with the details.

I will be in touch with you again later in the year to invite you to the second and final group meeting where you will be asked to prioritise the statements generated during the brainstorming sessions and to group them into themes.

I hope you have a safe journey home. For those of you who have parked in the Sportspark car park, please don't forget to get your ticket validated as you leave. Anyone travelling by public transport or taxi, don't forget to keep your ticket or to get a receipt so that we can reimburse your costs. Does anyone want to hand in a travel expenses claim form tonight?

Prompts to be used by the group facilitator if necessary

If the session dries up, the facilitator may want to prompt on the following points...

And what about...

- The cost of gluten-free food?
- The availability of gluten-free food:
 - When Travelling
 - When eating out
 - In the workplace
 - When shopping
 - On prescription
- Social eating/drinking
- Social support from friends and family
- The role of
 - Coeliac support groups
 - Healthcare professionals
- Gluten-free food labelling
- The taste and texture of gluten-free food
- Your knowledge about the gluten-free diet
- Information available about the gluten-free diet

- Symptoms experienced if you do eat gluten (duration, severity, type of symptoms)
- Length of time on the gluten-free diet
- Additional special dietary requirements
- Age at diagnosis / age now
- Cooking skills and domestic arrangements
- Household structure

Appendix 32 Letter sent to gluten-free food companies

Room 1.33
Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ

Phone: 01603 593665
E-mail: h.flaherty@uea.ac.uk

29 February 2012

Dear Sir/Madam,

I am a PhD student at the University of East Anglia and I am conducting a study into adherence to a gluten-free diet in adults with coeliac disease. I will collect data from seventy-five participants during two group meetings.

I would like to provide my participants with gluten-free snacks and samples and, as the budget for my research is very small, I would be extremely grateful if Nature's Path Organic would be willing to donate some samples of your gluten-free products for this purpose. In return for your generosity, I will make the participants aware of your donation and I would be very happy to display any promotional leaflets you may wish to send.

Thank you for considering my request. If you have any questions or need further information, please feel free to contact me.

Yours faithfully

Helen Flaherty
PhD student, University of East Anglia

Appendix 33 Rejection letters



Room 1.33
Elizabeth Fry Building
University of East Anglia
Norwich
NR4 7TJ

Phone: 01603 593665
E-mail: h.flaherty@uea.ac.uk

07 July 2011

Dear xxxx,

Re: Research study: What helps adults with coeliac disease stick to a gluten-free diet?

Thank you for responding to the invitation to participate in the above research project. Although we appreciate you taking the time to respond to the invitation letter, we have now received sufficient numbers of replies and, regrettably, we are unable to include you in this study.

Thank you for taking the time to reply to our invitation and for your interest in our research.

Yours Sincerely

Helen Flaherty
PhD student, University of East Anglia

Appendix 34 Final set of 91 statements

This table shows a list of the final set of 91 brainstormed statements arranged in a random order and numbered from 1 to 91.

No.	Statement (in response to the focus statement: It would be easier for adults with coeliac disease to stick to a gluten-free diet...)
1	...if there was more availability of gluten-free fish and chips.
2	...if there was more availability of savoury gluten-free snacks and not just sugary cakes and biscuits.
3	...if restaurants were better at labelling gluten-free options on their menus.
4	...if gluten ingredients were not added to foods where you wouldn't expect to find gluten (such as ice cream or grated cheese).
5	...if motorway services provided gluten-free food.
6	...if they use Coeliac UK's Food and Drink Directory.
7	...if people on a low income had additional help with the cost of gluten-free food.
8	...if separate cooking utensils are used in the preparation of gluten-free food at home.
9	...if staff working in restaurants/cafes were more knowledgeable about coeliac disease and the gluten-free diet.
10	...if other household members eat gluten-free.
11	...if they received more advice about coeliac disease and the gluten-free diet around the time of diagnosis.
12	...if they can cook gluten-free meals from scratch.
13	...if they are given a blood test to confirm adherence.
14	...if there was a universal gluten-free logo on food packaging used internationally on all suitable foods (such as the crossed grain logo).
15	...if airlines were better at providing gluten-free food during flight.
16	...if airports were better at providing gluten-free food.
17	...if other people encouraged them to stick to the gluten-free diet.
18	...if restaurants and carveries provided gluten-free gravy.
19	...if a wider range of gluten-free ready meals were available.
20	...if healthcare professionals knew more about coeliac disease and the gluten-free diet.
21	...if there was an expert point of contact for patients and healthcare professionals.
22	...if someone cooks for them.
23	...if they believe the diagnosis.
24	...if they have supportive work colleagues.
25	...if pubs stocked gluten-free beers and lagers.
26	...if gluten-free food was exempt from the prescription charge.
27	...if they use the internet to get information about coeliac disease and the gluten-free diet.
28	...if they have already been following a gluten-free diet for a long time.
29	...if they are prepared to go without/go hungry rather than eat gluten when there are no gluten-free foods available.
30	...if the public had a better understanding of coeliac disease and the gluten-free diet.
31	...if they are not made to feel different when eating socially.
32	...if they have a supportive GP.
33	...if there was a mobile phone app to advise them on where to find gluten-free food.
34	...if friends and family are supported/educated so they can reliably cater for them.
35	...if gluten-free food was more available when travelling by train.

36	...if more gluten-free recipes were available.
37	...if you can eat the same (gluten-free) food as everyone else when eating socially.
38	...if supermarket discount offers included gluten-free products.
39	...if they get a sufficient amount of gluten-free food on prescription.
40	...if they take their own gluten-free food with them when eating away from home.
41	...if there was legislation to ensure all food outlets provide gluten-free options.
42	...if there was a wider range of Asian gluten-free products available.
43	...if TV cookery programmes included gluten-free cooking.
44	...if they are allowed to take gluten-free food with them when travelling abroad.
45	...if they have a positive outlook and focus on what they can eat, rather than what they can't.
46	...if speciality gluten-free food had a longer shelf-life.
47	...if hospitals were better at providing gluten-free food.
48	...if they have someone to speak up for them on their behalf.
49	...if they are confident and not embarrassed by having to ask for gluten-free food when eating out.
50	...if they had a better knowledge of the gluten-free diet.
51	...if there was more availability of gluten-free sandwiches.
52	...if it was free to join Coeliac UK.
53	...if they have immediate access to a Dietitian at the time of diagnosis.
54	...if food outlets were more careful to avoid gluten contamination.
55	...if they receive gluten-free food on prescription.
56	...if Coeliac UK's Food and Drink Directory included a wider range of products and brands.
57	...if a wider range of gluten-free products were available on prescription (e.g. not just bread and flour).
58	...if there was a wider variety of gluten-free products in shops and supermarkets.
59	...if they understand the health consequences of not sticking to a gluten-free diet.
60	...if they join a coeliac support group (e.g. Coeliac UK).
61	...if there was less social pressure to accept any gluten-inclusive food or drink you are offered.
62	...if there was more psychological support available.
63	...if gluten-free products were kept next to similar gluten-inclusive items in supermarkets (e.g. gluten-free bread in the bread section, rather than the 'free from' section).
64	...if speciality gluten-free foods tasted nicer.
65	...if gluten-free food was easier to find on the shelves when shopping in supermarkets.
66	...if they have access to information about coeliac disease.
67	...if a wider range of non-traditional gluten-free breads were available (e.g. tortillas, chapatis pita bread).
68	...if speciality gluten-free food was not so high in calories and sugar.
69	...if they are well educated.
70	...if there was more consistency between similar products regardless of brand or pack size (e.g. some brands of cornflakes are GF but others are not).
71	...if they have access to another person with coeliac disease who has more experience.
72	...if GPs were better informed about what patients are allowed on prescription.
73	...if labels stated "produced in a factory where gluten is used" rather than "may contain gluten" so you can assess the level of risk.
74	...if they have supportive family and friends.
75	...if they didn't have to go to a pharmacy to collect prescribed gluten-free food when it should be obtainable from shops or supermarkets.
76	...if they were given a personal amount of money to support the buying of gluten-free food, rather than getting it on prescription.
77	...if there were more gluten-free options when eating out.
78	...if they don't have additional special dietary requirements as well as a gluten-free

	diet (e.g. lactose-free, vegetarian or diabetic).
79	...if gluten-free biscuits were made available when gluten-inclusive biscuits are served with tea and coffee at work.
80	...if they have plenty of freezer space.
81	...if they are female.
82	...if they experience symptoms when they consume gluten.
83	...if gluten-free bread was of the same quality in taste and texture as gluten-inclusive bread.
84	...if they are able to try free samples of gluten-free products before buying (e.g. at roadshows).
85	...if gluten-free food was not so expensive.
86	...if there was more gluten-free food available in all food shops and supermarkets.
87	...if there were more resources for people from ethnic minorities.
88	...if food manufacturers didn't change their ingredients so often.
89	...if they can get hold of gluten-free food when travelling abroad.
90	...if they have appropriate follow-up care with quick and easy access to dietitians.
91	...if they are determined to stick to the gluten-free diet and resist temptations.

Appendix 35 E-mail communication with the owner of the Ariadne concept mapping software.

From: Talcott [peter.severens@talcott.nl]
Sent: 18 June 2012 10:15
To: Helen Flaherty (NSC)
Subject: RE: Ariadne question

Helen,

Sorry I missed the email. My Talcott account is from my holding company. I do not look very regular at it.

I cannot check from here what is in the manual but 12 clusters is indeed what the software handles. What I normally did is leave out the very small clusters. It is no problem for the statistical procedure (clusters with only 1 already had no effect, clusters with 2 items have a very small effect). Another (but not very scientific option because is not following a non-ambient rule) is to put together clusters that resemble each other (this at least works in my business but we use Ariadne only to generate good ideas for marketing). The first option is to prefer from your perspective.

I cannot help you further with that. At least I will check the manual and change it.

Peter

From: Helen Flaherty (NSC) [<mailto:H.Flaherty@uea.ac.uk>]
Sent: Monday, June 18, 2012 10:42 AM
To: 'peter.severens@talcott.nl'; P.W.M.Severens@inter.NL.net; peter.severens@acxiom.com
Cc: 'directie@sbodevonder.nl'
Subject: RE: Ariadne question
Importance: High

Hi Peter,

I e-mailed you on 28th May with a question about Ariadne. I have just found some other e-mail addresses for you and I wondered whether the one I sent my previous e-mail to may no longer be the one you use. Please see below my question about the Ariadne software. I hope you are able to help.

Many thanks,
Helen

Helen Flaherty

PhD student
1.33 Elizabeth Fry Building
Faculty of Medicine and Health Sciences
University of East Anglia
Norwich Research Park
Norwich
NR4 7TJ

Tel: 01603 593665
Mobile: 07920406094

From: Helen Flaherty (NSC)
Sent: Monday, May 28, 2012 12:08 PM
To: peter.severens@talcott.nl
Subject: FW: Ariadne question

Dear Peter,

I am a PhD student at the University of East Anglia, England and I am using the Ariadne software that you kindly provided last year. I met Barbara when she came over to deliver some training for us about a year ago and this was really helpful.

I hope you don't mind me contacting you but I have a question about the software and I would be very grateful if you or Barbara could help. I sent the e-mail below to Barbara, but I'm not completely sure if I have the correct e-mail address for her.

When sorting the data, a few of the participants in my study have generated more than 12 clusters. Page 14 of the Ariadne handbook says that participants can generate up to a maximum of 25 clusters, however, the software only seems to allow me to input data for a maximum of 12 clusters per participant. Do you know whether it is possible to input data for more than 12 clusters and, if so, could you please advise me on how to do this.

Many thanks,
Helen

Helen Flaherty

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1.33 Elizabeth Fry Building
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University of East Anglia
Norwich Research Park
Norwich
NR4 7TJ

Tel: 01603 593665
Mobile: 07920406094

From: Helen Flaherty (NSC)
Sent: Friday, May 18, 2012 9:24 AM
To: 'directie@sbodevonder.nl'
Subject: Ariadne question

Hi Barbara,

I hope you are well. You may remember me from the Ariadne training session that you ran for us last June in Norwich. I have a quick question about Ariadne and I wondered whether you may be able to help.

I have reached the stage of asking my participants to rate and cluster the statements and I have started inputting the data into Ariadne. As per the guidance in the Ariadne handbook, I have told participants that they can produce as many clusters/groups as they like (max of 25). However, I

cannot fathom out how to increase the number of groups from 12 in Ariadne. I would be very grateful if you are able to advise.

Many thanks,
Helen

Helen Flaherty

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Appendix 36 Prioritisation data collection sheets

Task 1

Ranking the statements in order of importance from 1 to 5

You have a set of 91 statements. Each statement completes the sentence “*It would be easier for adults with coeliac disease to stick to a gluten-free diet if...*”.

There are two steps involved in task 1. Please read the instructions below before starting this task.

Step 1

Please divide the statements into five groups in order of how important you think each one is in relation to the focus statement: “*It would be easier for adults with coeliac disease to stick to a gluten-free diet if...*”. You can use the boxes numbered 1 to 5 on the attached sheet (page 2) to put your statements in order.

Score 1 refers to statements which you think are **least important**.

Score 5 should be the statements you consider to be **most important**.

It is important that you divide the statements into five groups of roughly the same size. Each group should contain no less than 16 statements and no more than 20 statements.

Every statement must be put in a group and none of the statements should be left out.

Step 2

Once you have completed step 1, please fill in the grid on page 3. Each statement has a random number printed on it. Please write the numbers shown on the statements in each of your five piles in the appropriate boxes on the grid.

Once you have completed the grid on page 3, please return it to us in the envelope provided (along with your completed grid for Task 2 (page 3 of Task 2)).

Task 1 - Ranking the statements from 1 to 5

Step 1 - Please rank the statements in order of importance from 1 to 5 in relation to the focus statement: *“It would be easier for adults with coeliac disease to stick to a gluten-free diet if...”*

When you have finished, please complete the grid on page 3 and return it to us in the envelope provided.

Least Important  Most Important	1	Place your statements here	No fewer than 16 statements and no more than 20 statements in each pile.
	2	Place your statements here	
	3	Place your statements here	
	4	Place your statements here	
	5	Place your statements here	

Appendix 37 Clustering data collection sheets

Task 2

Grouping the statements into themes

You have a set of 91 statements. Each statement completes the sentence “*It would be easier for adults with coeliac disease to stick to a gluten-free diet if...*”.

There are two steps involved in task 2. Please read the instructions below before starting this task.

Step 1

Please group together the statements which you consider to be related, or which have something in common. The importance of the statements is not relevant here.

You can use the attached sheets (all numbered as ‘page 2’) to help put your statements into groups.

You may form as many groups as you like.

Please make sure that:

- Not all statements are placed into one single group.
- Statements are not all put into their own individual group.
- Each statement is only used once.
- You do not have a group of ‘miscellaneous’ statements or a group containing statements that you think are not related to one another.
- Every statement must be put in a group and none of the statements should be left out.

Step 2

Once you have completed step 1, please choose a name for each of your groups and write the names in the grid on the attached sheet (page 3). You can use the additional sheet (also labelled as page 3) if necessary.

Each statement has a random number printed on it. Please write the numbers shown on the statements in each of your groups in the boxes next to the relevant group name on the grid (page/s 3).

Once you have completed the grid, please return it to us in the envelope provided (along with your completed grid for Task 1 (page 3 of Task 1)).

Task 2 - Grouping the statements into themes

Step 2

Please write the name of each of your groups in the left-hand column in the grid below. The order in which you list the groups is not important. Each statement has a random number printed on it. Please write the numbers shown on the statements next to the appropriate group name on the grid. The order in which you write the statement numbers is not important.

You can write your list of statement numbers over more than one line if there is not sufficient space in a single line. Please use the additional sheet if necessary.

Name of group																				

Please return this sheet to us in the envelope provided along with your results for Task 1 (Page 3 of Task 1)

Appendix 38 Data inputting errors and actions taken to resolve them

Errors with the data generated from the prioritising and clustering exercises:

Participant ref no.	TASK 1 - Prioritisation	TASK 2 – Clustering
258	No errors	I removed a 15-statement cluster labelled ‘trivial’ because the statements contained within it are not necessarily related to each other. The prioritisation of statements shows whether or not statements are perceived to be trivial or not.
680	No errors	More than 12 clusters generated (2 clusters removed: Food Labelling; and taste)
482	1 Statement missing	No errors
615	No errors	3 statements missing and more than 12 clusters generated (3 clusters removed: Recipes; ethnic; and IT Help).
455	No errors	1 statement missing.
879	No errors	1 statement missing and 1 statement was duplicated. Both entries of the duplicated statement were removed.
126	1 statement missing and 1 statement was duplicated. Both entries of the duplicated statement were removed.	No errors.
366	No errors	More than 12 clusters generated (2 clusters removed: Gender; and different ethnic backgrounds).
970	No errors	1 statement missing and more than 12 clusters generated (5 clusters removed: Product information; habit; other dietary issues; self-determination; and packaging information).
954	1 statement was duplicated. Both entries of the duplicated statement were removed. 18 statements were missing (task not completed).	Task not completed (no data).
512	No errors	More than 12 clusters generated (4 clusters removed: Ethic origin; financial; diet; and no coeliac relevance – this last cluster would have been removed anyway as the statements are not necessarily related).
598	No errors	1 statement missing and more than 12 clusters generated (6 clusters removed:

		Media support; work environment; public understanding; gender; ethnic food choices; and healthy eating).
116	1 statement missing and 1 statement was duplicated. Both entries of the duplicated statement were removed.	1 statement was duplicated. Both entries of the duplicated statement were removed.
904	No errors	2 statements missing. I have removed five clusters as they were not themes/concepts and the statements contained within them were not necessarily related. The clusters were named: Nice touch; things I consider adequate; not necessary; not likely; does this make a difference?
162	No errors	More than 12 clusters generated (1 cluster removed: Eating out).
600	No errors	Task not completed (no data).
540	No errors	1 statement missing.
710	2 statements missing	1 statement was duplicated. Both entries of the duplicated statement were removed. 4 statements were missing
647	No errors	1 statement missing and more than 12 clusters generated (3 clusters removed: Storage; labelling; and work).
178	No errors	1 statement missing and more than 12 clusters generated (3 clusters removed: Dietetic services; ethnic minorities; and financial).
909	No errors	More than 12 clusters generated (5 clusters removed: Psychological support; ingenuity; financial help; being normal; and taste and texture).
679	1 statement was duplicated in the same group (removed one entry) 1 statement missing	1 statement missing.
475	No errors	More than 12 clusters generated (3 clusters removed: Gender; at work; and believing the diagnosis).
685	1 statement missing and 1 statement was duplicated. Both entries of the duplicated statement were removed.	Had a 'miscellaneous group' – removed.
269	No errors	Had an "Irrelevant" group – removed.
637	No errors	1 statement missing.
715	No errors	Had a "Miscellaneous" group – removed.

Appendix 39 Movement of statements between clusters as the number of clusters was increased or decreased

Statements within the clusters are shown in order of mean preference score.

Comparison of 4 and 5 clusters for all participants:

	4 Clusters	5 clusters
Cluster 1	Statements: 3, 77, 9, 54, 40, 41, 37, 49, 89, 31, 44, 47, 18, 5, 15, 1, 25, 79, 16, 61, 35.	NO CHANGE
Cluster 2	Statements: 85, 83, 14, 58, 86, 4, 64, 2, 67, 38, 65, 19, 70, 68, 88, 84, 73, 75, 46, 63, 42.	NO CHANGE
Cluster 3	Statements 59, 11, 91, 50, 45, 90, 82, 55, 53, 32, 20, 39, 66, 74, 57, 34, 23, 26, 72, 78, 21, 30, 60, 28, 7, 12, 6, 71, 13, 29, 56, 10, 17, 8, 36, 76, 27, 24, 43, 69, 62, 52, 33, 48, 87, 80, 22, 81.	Statements: 55, 39, 57, 26, 7, 6, 56, 36, 76, 52, 87, 80
Cluster 4	Statement: 51	59, 11, 91, 50, 45, 90, 82, 53, 32, 20, 66, 74, 34, 23, 72, 78, 21, 30, 60, 28, 12, 71, 13, 29, 10, 17, 8, 27, 24, 43, 69, 62, 33, 48, 22, 81.
Cluster 5		NO CHANGE

Cluster 3 in the 4-cluster table splits to create clusters 3 and 4 in the 5-cluster table.

Comparison of 5 and 6 clusters for all participants:

	5 Clusters	6 clusters
Cluster 1	SPLIT	3, 77, 9, 54, 40, 41, 89, 44, 18, 5, 15, 1, 25, 79, 16, 35
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	37, 49, 31, 47, 61
Cluster 6		NO CHANGE

Cluster 1 in the 5-cluster table has split to form 2 clusters (clusters 1 and 5) in the 6-cluster table.

Comparison of 6 and 7 clusters for all participants:

	6 Clusters	7 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	SPLIT	59, 11, 91, 50, 45, 90, 82, 53, 32, 20, 66, 74, 34, 23, 72, 78, 21, 30, 60, 28, 12, 71, 13, 10, 17, 8, 27, 24, 43, 69, 62, 33, 48, 22, 81.
Cluster 5	NO CHANGE	29
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7		NO CHANGE

The only difference is that statement 29 moves from cluster 4 in the 6-cluster table to form its own cluster (cluster 5) in the 7-cluster table.

Comparison of 7 and 8 clusters for all participants:

	7 Clusters	8 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	SPLIT	6, 36, 52 & 80
Cluster 4	NO CHANGE	55, 39, 57, 26, 7, 56, 76 & 87
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8		NO CHANGE

The only difference between the cluster-7 and the cluster 8-table is that cluster 3 from the 7-cluster table is divided into 2 clusters (cluster 3 and 4) in the 8-cluster table.

Comparison of 8 and 9 clusters for all participants:

	8 Clusters	9 clusters
Cluster 1	SPLIT	3, 77, 9, 54, 40, 41, 89, 44, 18, 5, 15, 1, 25, 16, 35.
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9		79

The only difference between these two is that cluster 1 in the 8-cluster table has split to form 2 clusters (cluster 1 and cluster 9) in the 9-cluster table.

Comparison of 9 and 10 clusters for all participants:

	9 Clusters	10 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	SPLIT	49, 31, 61
Cluster 8	NO CHANGE	37, 47
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10		NO CHANGE

The only difference between the 9-cluster and 10-cluster tables is that cluster 7 in the 9-cluster table has split in 2 to form clusters 7 and 8 in the 10-cluster table.

Comparison of 10 and 11 clusters for all participants:

	10 Clusters	11 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	SPLIT	83, 14, 58, 86, 4, 64, 2, 67, 38, 65, 19, 70, 68, 88, 84, 73, 46, 63, 42.
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	NO CHANGE	85, 75
Cluster 11		NO CHANGE

The extra cluster is formed in the 11-cluster table from 2 statements taken from cluster 2 in the 10-cluster table.

Comparison of 11 and 12 clusters for all participants:

	11 Clusters	12 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	SPLIT	75
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12		85

Cluster 10 in the 11- cluster table is split to form cluster 10 and cluster 11 in the 12-cluster table.

Comparison of 12 and 13 clusters for all participants:

	12 Clusters	13 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	SPLIT	59, 11, 50, 90, 82, 53, 32, 20, 66, 23, 72, 78, 21, 60, 28, 12, 71, 13, 8, 27, 43, 69, 62, 33.
Cluster 6	NO CHANGE	91, 45, 74, 34, 30, 10, 17, 24, 48, 22, 81.
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	NO CHANGE	NO CHANGE
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12	NO CHANGE	NO CHANGE
Cluster 13		NO CHANGE

The only difference was that cluster 5 split to form a new clusters 5 and 6 in the 13-cluster table.

Comparison of 13 and 14 clusters for all participants:

	13 Clusters	14 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	SPLIT	6, 52
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9	NO CHANGE	36, 80
Cluster 10	NO CHANGE	NO CHANGE
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12	NO CHANGE	NO CHANGE
Cluster 13	NO CHANGE	NO CHANGE
Cluster 14		NO CHANGE

The 4 statements in cluster 3 were split into 2 clusters (cluster 3 and cluster 9) in the 14-cluster table.

Comparison of 14 and 15 clusters for all participants:

	14 Clusters	15 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	SPLIT	49, 31
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	NO CHANGE	NO CHANGE
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12	NO CHANGE	61
Cluster 13	NO CHANGE	NO CHANGE
Cluster 14	NO CHANGE	NO CHANGE
Cluster 15		NO CHANGE

Cluster 8 split to form clusters 8 and 12 in the 15-cluster table.

Comparison of 15 and 16 clusters for all participants:

	15 Clusters	16 clusters
Cluster 1	SPLIT	41, 1
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	3, 77, 9, 54, 40, 89, 44, 18, 5, 15, 25, 16, 35
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	NO CHANGE	NO CHANGE
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12	NO CHANGE	NO CHANGE
Cluster 13	NO CHANGE	NO CHANGE
Cluster 14	NO CHANGE	NO CHANGE
Cluster 15	NO CHANGE	NO CHANGE
Cluster 16		NO CHANGE

Cluster 1 was split to form cluster 1 and cluster 3 in the 16-cluster table.

Comparison of 16 and 17 clusters for all participants:

	16 Clusters	17 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	SPLIT	55, 39, 26, 7, 56, 76.
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	NO CHANGE	NO CHANGE
Cluster 8	NO CHANGE	NO CHANGE
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	NO CHANGE	NO CHANGE
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12	SPLIT	NO CHANGE
Cluster 13	NO CHANGE	57, 87
Cluster 14	NO CHANGE	NO CHANGE
Cluster 15	NO CHANGE	NO CHANGE
Cluster 16	NO CHANGE	NO CHANGE
Cluster 17		NO CHANGE

Cluster 5 in the 16-cluster table has split to form clusters 5 and 13 in the 17-cluster table.

Comparison of 17 and 18 clusters for all participants:

	17 Clusters	18 clusters
Cluster 1	NO CHANGE	NO CHANGE
Cluster 2	NO CHANGE	NO CHANGE
Cluster 3	NO CHANGE	NO CHANGE
Cluster 4	NO CHANGE	NO CHANGE
Cluster 5	NO CHANGE	NO CHANGE
Cluster 6	NO CHANGE	NO CHANGE
Cluster 7	SPLIT	78, 12, 8, 43, 33.
Cluster 8	NO CHANGE	59, 11, 50, 90, 82, 53, 32, 20, 66, 23, 72, 21, 60, 28, 71, 13, 27, 69, 62.
Cluster 9	NO CHANGE	NO CHANGE
Cluster 10	NO CHANGE	NO CHANGE
Cluster 11	NO CHANGE	NO CHANGE
Cluster 12	NO CHANGE	NO CHANGE
Cluster 13	NO CHANGE	NO CHANGE
Cluster 14	NO CHANGE	NO CHANGE
Cluster 15	NO CHANGE	NO CHANGE
Cluster 16	NO CHANGE	NO CHANGE
Cluster 17	NO CHANGE	NO CHANGE
Cluster 18		NO CHANGE

Cluster 7 split to create clusters 7 and 8 in the 18 cluster table.