

**Volume 1: Research**

**Psychosocial Factors associated with Self-Management and Well-being  
in Childhood Chronic Illness**

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## Thesis Overview

Volume I comprises a literature review and an empirical paper. The literature review explores the link between illness representations and self-management in children and young people with chronic illness. Fourteen published empirical studies were identified for the review. A risk of bias assessment was completed for each study. Consistencies and differences between papers were identified. Overall, *treatment control* beliefs were most consistently associated with self-management across a range of chronic health conditions. The literature review has been prepared for submission to the Health Psychology Review (see appendix 1).

The empirical paper details a cross-sectional study investigating associations between illness representations, self-efficacy, self-management and psychological well-being in young people with Coeliac Disease. Forty young people and 34 parents recruited from hospital outpatient clinics completed questionnaires. Results indicated *timeline-cyclical* beliefs and *treatment concerns* were associated with self-management. *Timeline-cyclical*, *identity*, *treatment control* and *coherence* were correlated with well-being. In terms of self-efficacy, young people with high levels of *self-efficacy* were more likely to have better self-management and positive well-being. Finally, dissimilarity in *timeline-cyclical* beliefs between young people and their parents was related to higher parental stress. The empirical paper has been prepared for submission to the Journal of Health Psychology. Some changes have been made related to formatting of the empirical paper in line with university guidelines for presenting a thesis (see appendix 3).

Four full length clinical practice reports and a summary of CPR 5 (oral presentation) are included in the Volume II. Firstly, a case of a 55 year old woman presenting with

depression is presented. The case is formulated from both cognitive and psychodynamic perspectives. A service evaluation of an Assertive Outreach Service is then described. Both of these clinical practice reports were completed while on an adult mental health placement. The third clinical practice report is a single case experimental design, detailing the assessment and treatment of a 15 year old boy with a mild learning disability and anxiety. This is followed by a case study of the assessment, formulation and intervention of a 15 year old boy with anger and memory difficulties. Finally, a summary of an audit of a new clinical service delivering psychosocial interventions in dementia is provided. This clinical practice report was presented orally.

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## **Literature Review**

**Illness beliefs and self-management in children and young people with chronic  
illness:**

**A review of the literature.**

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## Abstract

The present literature review explores Leventhal's common-sense model of illness representations in relation to self-management of treatment regimes in children and young people with chronic physical health conditions. Fourteen papers were identified for the review. Each paper was assessed for risk of bias and consistencies and differences between papers were identified. In terms of risk of bias, all identified studies were lacking a control group, the majority of studies were cross-sectional and small sample sizes for some studies made it difficult to generalise findings. Furthermore, the method of measuring both illness representations and outcomes was not consistent. Overall, *treatment control* beliefs were most consistently associated with self-management across a range of chronic health conditions, namely Cystic Fibrosis, Essential Hypertension and Diabetes. More specifically, short-term treatment control beliefs (i.e. effectiveness of treatment to control one's illness) were positively associated with dietary self-management in Diabetes. In addition, results indicated that illness representations were condition-and treatment-specific and therefore generalisations across conditions and particular aspects of a treatment regimen cannot be made. Together, these results suggest that children and young people who believe that their condition can be controlled by treatment are more likely to engage in more helpful self-management behaviours. The clinical implications in terms of assessment and intervention are discussed and limitations highlighted.

## *Keywords:*

*illness beliefs, illness representations, chronic illness, chronic health condition, paediatric, self-management.*

## **Introduction**

Leventhal's common-sense model of illness representation provides a theoretical framework to help understand how an individual's conceptualisation of their illness influences coping behaviour (e.g. self-management behaviours) and a range of health outcomes (e.g. well-being) (see Figure 1) (Leventhal, Brissette, & Leventhal, 2003).

The common-sense model of illness representations proposes that when individuals are faced with internal or external health threats, related to diagnosis or management of an illness, they develop their own individual beliefs (illness representations) about their condition. These illness representations (sometimes called cognitive representations, schema or personal models) are based on a person's understanding or experience of the condition, and may not be related to the objective clinical markers of the illness (Petrie & Weinman, 2006). In parallel with these cognitive illness representations, emotional representations are also being generated and processed.

Leventhal, Halm, Horowitz, Leventhal & Ozakinci (2005) suggest that representations provide a "framework for action" influencing coping strategies and action plans, for example, self-management of treatment regimens. The model indicates that these coping strategies impact on illness outcomes and emotional wellbeing. Like other self-regulatory models, the common-sense model is a dynamic process where the appraisal of the effectiveness of coping strategies, health behaviours and/or changes in the illness process results in updating of the illness representations (Leventhal et al., 2005). This appraisal mechanism and feedback loop would suggest that the model could be particularly useful in a clinical setting when developing interventions to promote self-management of chronic health conditions (McAndrew et al., 2008).





Similarly to adults, these five dimensions of illness representation have been identified in children (Goldman, Whitney-Saltiel, Granger, Rodin, 1991). However, Paterson, Moss-Morris & Butler (1999) found that more complex, abstract concepts of illness (e.g. long-term consequences) may develop more slowly in children who may focus more on current symptoms.

More recently, Horne (2003) has argued that a more comprehensive consideration of *treatment perceptions* (sometimes called *treatment beliefs*) is important when the common-sense model is applied to exploring self-management. In particular, he looked at the relationship between necessity beliefs (i.e. beliefs in personal need for treatment self-management) and concerns (i.e. belief about side effects of medication and adverse effects of treatment on daily living). Adult studies confirmed that self-management of chronic illness is positively correlated with *necessity* beliefs and negatively correlated with *concerns* beliefs (Horne & Weinman, 1999).

Clinically, the Department of Health stated that health professionals should support individuals with long-term health conditions to develop skills in self-management (Department of Health, 2007). McAndrew et al. (2008) have started to explore how the Common-Sense model could serve as a basis for developing interventions to improve chronic illness self-management. However, more research in this area is required before any firm conclusions can be drawn (e.g. Karamanidou, Weinman & Horne, 2008).

A meta-analysis of 45 studies has provided support for Leventhal's common-sense model of illness representations (Hagger & Orbell, 2003). Furthermore, the model has aided our understanding of chronic illness self-management, psychological and social outcomes (Kaptein et al., 2003). However, Kaptein et al. (2003) highlighted that available studies testing the predictive power of dimension of illness representations were limited.

Currently, a systematic review focusing specifically on the role of illness representations in children and young people with chronic physical health conditions is not available.

In summary, there is a growing body of literature supporting the application of Leventhal's common-sense model in adults with chronic health conditions. However, given the cognitive development of children and systemic issues associated with managing a chronic health condition (e.g., role of parents in managing treatment regimens), a review focusing exclusively on children and young people is necessary. Taking this into consideration, a literature review focusing on the application of the illness representation framework to explore the management of chronic physical health conditions in children and young people is now warranted.

### **Defining a chronic physical health condition**

There is no universally accepted definition of chronic illness (Kaptein et al., 2003). However, O'Halloran, Miller & Britt (2004) completed a literature review evaluating characteristics used to define chronic conditions for use in research. The evaluation resulted in the following criteria, such that for any illness to be classed as a chronic health condition, it must: a) have a duration that has lasted, or is expected to last, at least 6 months; b) have a pattern of recurrence, or deterioration; c) have a poor prognosis; and d) produce consequences that impact on the individual's quality of life.

The World Health Organization discussion paper on Chronic Health Conditions in Adolescence (Michaud, Suris & Viner, 2007) identifies a range of chronic physical health conditions namely, Asthma, Diabetes, Inflammatory Bowel Disease, HIV/AIDS, Sickle Cell Anaemia, Thalassaemia, Bone Marrow Transplant, Growth Hormone Deficiency, Hypothyroidism, Cystic Fibrosis, and Chronic Renal Disease. In addition, the World

Health Organisation for Europe also identified Chronic Obstructive Pulmonary Disease (COPD), Cardiovascular Disease & Cancer as additional chronic health conditions (Busse, Blüme, Scheller-Kreinsen & Zentner, 2010). However, the paper did not focus specifically on children and young people. In addition, Coeliac Disease is also recognised as a chronic health condition (NICE, 2009). While it is possible that these lists are not exhaustive, the above fourteen conditions, with the addition of Coeliac Disease, were included as key words within the literature search.

This literature review has identified empirical research studies investigating illness representations theory, or dimensions of it, in relation to self-management of treatment regimes in children and young people with chronic physical health conditions. Each paper was reviewed for risk of methodological and clinical biases. Consistencies and differences between papers were identified and evaluated with a view to determining to what extent illness representations relate to self-management in this population. Differences between chronic physical health conditions and types of self-management regimes were also reviewed in relation to dimensions of illness representations. This review will then seek to determine if there is stronger evidence for particular dimensions of the model that are associated with self-management. Clinically, this review will help to improve our understanding of children and young people's illness representations in relation to self-management, which could, in turn, guide interventions to promote better self-management.

## **Method**

### **Search Criteria**

Guidelines produced by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) for the identification of articles for review were used to develop search criteria (Moher, Liberati, Tetzlaff, Altman, The PRIMA Group, 2009). These guidelines are recommended by the Journal of Health Psychology (Marks, 2010).

Different terms, used interchangeably in the literature, to refer to the key concepts of illness representations, self-management, children/young people and chronic illness were used to search the key databases (between 1996 – January 2010) of Web of Science, Psychinfo, Medline and Google Scholar to identify potential papers for inclusion. Reference lists of published studies were scrutinised for additional papers not returned from electronic searches. Search criteria, including keywords and exclusion criteria are described in appendix 2.

## **Results**

### **Description of Studies**

The search criteria identified 14 papers for review (Appendix 3). Papers were organised alphabetically by author and summarised in Table 2.

#### *Country of Origin*

The majority of studies were conducted in England (9 studies). Four studies were completed in the United States of America and one study originated from Slovenia.

#### *Type of Chronic Health Condition*

Nine studies investigated the role of illness representations in Diabetes self-management (Griva, Myers, & Newman, 2000; Iannotti et al., 2006; Law, Kelly, Huey, Summerbell, 2002; Nouwen, Law, Hussain, McGovern, & Napier, 2009; Skinner et al., 2003; Skinner & Hampson, 2001; Skinner & Hampson, 1998; Skinner, Hampson, & Fife-Schaw et al., 2002; Skinner, John, & Hampson, 2000). In contrast, limited research was available for children with Asthma (Zebracki & Drotar, 2004; Yoos et al., 2007), Cystic Fibrosis (Bucks et al., 2009), Renal Disease (Radcliff & Blount, 2010), and Essential Hypertension (Zugelj et al., 2010). No published studies, focusing on illness representations and self-management, were available for Coeliac Disease, Epilepsy, HIV, AIDS, Cardiovascular Conditions, Cancer, Chronic Obstructive Pulmonary Disease (COPD), Haemophilia, Sickle Cell Anaemia, Thalassaemia or Bone Marrow Transplant.

### *Recruitment*

The main sources of recruitment across studies were hospital outpatient clinics (10 studies). Two studies recruited via specialist paediatric practices based in New York and Slovenia and two studies recruited via the British Diabetic Association.

### *Participants*

The majority of participants were young people at least 11 years old. Only two studies included younger children starting from age 5 and 10 respectively as well as young people (Iannotti et al., 2006; Yoos et al., 2007). Two studies included young people and young adults in their sample (Griva et al., 2000; Skinner et al., 2002). No other studies included participants over the age of 18 years.

Twelve studies included information related to the gender of their participants. Across these studies, 430 girls (49%) and 452 boys (51%) were included. Although, Skinner et al. (2003) did not report their gender split, they highlighted that there was a marked gender bias in their study, with girls being over-represented. In contrast, Yoos et al. (2007) did not report the gender characteristics of their sample.

### *Study Methods*

A cross sectional design was most often used (n=12) to investigate the relationship between illness representations and self-management. One study used prospective methods (Skinner & Hampson, 2001) and one included longitudinal designs in an attempt to investigate causal associations (Skinner et al., 2000).

### *Measurement of Illness Representations*

The method of measuring illness representations varied across studies. Therefore, studies were included if their questionnaires operationalized at least one of the domains of Leventhal's self-regulatory model of illness representations (Leventhal et al., 2003). Table 1 displays a summary of the questionnaires used to measure illness representations for each study. The table also includes descriptions of their subscales and key dimensions measured. A brief description of each questionnaire is then provided.

**Table 1:** Summary of questionnaires used in studies

| <b>Authors/Study</b>   | <b>Chronic Illness</b> | <b>Questionnaire</b>                             | <b>Subscales used</b>  | <b>Domain of Illness Representation Measured*</b>   |
|--|------------------------|--|--|---|
| Bucks, Hawkins, Skinner, Horn, Seddon, Horne (2009)                              | Cystic Fibrosis        | IPQ-R  | Identity, Timeline-chronicity, Timeline-Cyclical, Consequences, Personal Control, Treatment Control, Cause, Emotional Representations  | Identity, Timeline-chronicity, Treatment Control, Emotional Representation (Consequences, Personal Control, Cause, Timeline-Cyclical were not included in the analysis)                 |
| Griva, Myers & Newman (2000)   | Diabetes               | IPQ  | Identity, Timeline, Consequences, Control, Cause   | Identity, Timeline, Consequences, Personal and Treatment Control (Cause not included in the analysis).  |
| Iannotti, Schneider, Nasel, Haynie, Plotnick, Clark, Sobel, Simons-Morton (2006) | Diabetes               | Outcome Expectations of Diabetes Self-Management | Expectations for Negative Outcomes (OEDM-N)<br>Expectations for Positive Outcomes (OEDM-P)   | Consequences  |
| Law, Kelly, Huey, Summerbell (2002).   | Diabetes               | IPQ-R  | Identity, Timeline, Consequences, Cause, Timeline Cyclical, Personal Control, Treatment Control, Illness Coherence and Emotional Representations.  | Identity, Timeline, Consequences, Cause, Timeline Cyclical, Personal Control, Treatment Control, Illness Coherence and Emotional Representations. (Cause not included in the analysis). |
| Nouwen, Law, Hussain, McGovern, Napier (2009)                                    | Diabetes               | IPQ & Personal Models of Diabetes Questionnaire  | Short-term Treatment Effectiveness & Consequences  | Consequences, Treatment Control,  |
| Ratcliff, Blount (2010)  | Renal                  | Perceived Adversity Scale                        | Perceived Adversity  | Consequences, Control   |
| Skinner, Hampson (2001)  | Diabetes               | The Personal Models of Diabetes Questionnaire).  | Effectiveness of treatment to control diabetes & Effectiveness of treatment to prevent complications (i.e. control)<br>Perceived seriousness of diabetes & perceived impact of diabetes (i.e. consequences). | Consequences, Treatment Control   |
| Skinner, Hampson (1998)  | Diabetes               | The Personal Models of Diabetes Questionnaire    | Effectiveness of treatment to control diabetes & Effectiveness of treatment to prevent complications (i.e. control)<br>Perceived seriousness of diabetes & perceived impact of diabetes (i.e.                | Consequences , Treatment Control  |



|   |                        |   | consequences).   |   |
|---|------------------------|---|--|---|
| Skinner, Hampton, Fife-Schaw (2002)   | Diabetes               | IPQ & Personal Models of Diabetes Questionnaire   | Perceived Consequence Scale (10 items of the perceived consequence scale of IPQ)<br><br>Perceived Treatment Effectiveness to Control Diabetes and Prevent Complications (based on Glasgow, 1997)             | Consequences, Treatment Control   |
| Skinner, Howells, Greene, Edgart, McEvilly, Johansson (2003)                | Diabetes               | Diabetes Illness Representations Questionnaire (based on IPQ & Personal Models of Diabetes Questionnaire. | Identity, Cause, Timeline, Perceived Consequences (Impact and Threat), Treatment Effectiveness (Control and Prevent)   | Identity, Timeline, Consequences, Control, (NB: Cause only analysed for inter-correlations between illness representations)   |
| Skinner, John, Hampson (2000)   | Diabetes               | Personal Models of Diabetes Questionnaire   | Effectiveness of treatment to control diabetes & Effectiveness of treatment to prevent complications (i.e. control)<br>Perceived seriousness of diabetes & perceived impact of diabetes (i.e. consequences). | Consequences, Treatment Control   |
| Yoos, Kitzman, Henderson, McMullen, Sidora-Arcoleo, Halterman, Anson (2007) | Asthma                 | Asthma Illness Representation Scale (AIRS)  | Treatment expectations, attitudes towards medication use, facts regarding asthma, nature of asthma symptoms, emotional aspects of medication use.  | Treatment control   |
| Zebracki & Drotar (2004)  | Asthma                 | Outcome Expectancy Scale (8 items)  | Negative outcome expectancy<br>Positive outcome expectancy   | Treatment Control   |
| Zugelj, Zupancic, Komidar, Kenda, Varda, Gregoric (2010)                    | Essential Hypertension | Brief IPQ   | Consequences, Timeline<br>Personal Control, Treatment Control, Identity, Concern, Coherence, Emotions.   | Consequences, Timeline, Personal Control<br>Treatment Control, Identity, Coherence, Emotional Representations (Concern and Emotions) (Cause not included in the analysis) |

\*Conceptualisation of how subscales map onto particular aspects of Leventhal's common-sense model of illness representations.

### *Illness Representations Questionnaire (IPQ)*

The Illness Representations Questionnaire (IPQ) (Weinman et al., 1996) provides a quantitative assessment of the five dimensions of cognitive illness representations described by Leventhal's self-regulation model, namely: perceived *identity*, *cause*, *consequences*, *timeline acute/chronic*, and *control (personal and treatment)* (70 items) (Leventhal et al., 2003). More recently, a revised version (IPQ-R; Moss-Morris et al., 2002) has been published (70 items). The revised version included, on theoretical and psychometric grounds, three additional subscales (*emotional representations*, *timeline-cyclical* and *illness coherence*). Since the IPQ was developed a short version has also been developed. The Brief Illness Perception Questionnaire (BIPQ; Broadbent, Petrie, Main & Weinman, 2006) comprises of 9 items addressing each dimension listed in the IPQ-R. The IPQ was the most commonly used method of measuring illness representations by seven studies included in this review either in its complete form (Bucks et al., 2009; Griva et al., 2000; Law et al., 2002; Zugelj et al., 2010) or through the use of particular sub-scales (Nouwen et al., 2009; Skinner et al., 2002; Skinner et al., 2003).

### *Personal Models of Diabetes Questionnaires*

The Personal Models of Diabetes Questionnaire (Hampson, Glasgow & Toobert, 1990) was used in six studies. The questionnaire is a brief, eight-item self-report instrument evaluating beliefs about the efficacy of treatment regimen (to control and prevent complications of Diabetes) and consequences of diabetes (i.e. seriousness/worry and impact of Diabetes on daily life). The questionnaire maps onto key dimensions of Leventhal's model, namely *treatment control* and *consequences* dimensions (Nouwen et al., 2009) and has been validated (Glasgow et al., 1997). Three studies included in this

review used the complete questionnaire in their research (Skinner & Hampson, 2001; Skinner & Hampson, 1998; Skinner et al., 2000), whereas others included items from both the Personal Models of Diabetes Questionnaire and items from the IPQ (Nouwen et al., 2009; Skinner et al., 2002; Skinner et al., 2003). Adequate internal consistency has been reported (Nouwen et al., 2009; Skinner et al., 2003).

#### *The Perceived Adversity Scale (PA scale)*

The PA Scale is a 10-item scale based on the *consequences* component of the self-regulation model, with particular emphasis on understanding what aspects of living with a transplant are perceived to be most aversive (Radcliff & Blount, 2010). The authors developed the measure specifically for their study and reported good internal consistency associated with the measure. Other dimensions of the self-regulation model were not investigated in this study.

#### *Outcome Expectations of Diabetes Self-Management*

Bandura describes outcome expectations as “detrimental or beneficial physical effects, favourable or adverse social reactions, and positive or negative self-evaluative reactions” (Bandura, 1997). The Outcome Expectations of Diabetes Self-Management questionnaire (Iannotti et al., 2006) consists of two independent factors: expectations for negative outcomes (12 items) and expectations for positive outcomes (12 items). Good internal consistency was reported for both subscales for young people aged 10-16. Outcome expectations have been described as equivalent to the perceived *consequences* dimension of illness representations (Nouwen et al., 2009). Therefore this study has been included in the review.

### *Outcome Expectancy Scale*

Zebracki & Drotar (2007) defined outcome expectancy as the “individual’s expectations with regard to the effectiveness of the recommended treatment and relevant health-related behaviours”. The outcome expectancy scale consists of 8 items based on the Treatment Efficacy Scale (Brusch, Schwankovsky, Gilbert & Zeiger, 1999) and a section of the Caretaker Expectations Regarding the Management of Pediatric Asthma Scale (Holden, Wade, Mitchell, Ewart, & Islam, 1998). The authors describe good internal consistency. Similarly to the Outcome Expectations of Diabetes Self-Management (Iannotti et al., 2006), this questionnaire was not explicitly based on Leventhal’s self-regulatory model. However, the questionnaire measures treatment effectiveness (i.e. prevention or reduction of current or future health related difficulties). This is seen as equivalent to the *treatment-control* domain of illness representations.

### *Asthma Illness Representation Scale (AIRS)*

The final study included in this review was conducted by Yoos et al. (2007). The Asthma Illness Representation Scale (AIRS) was developed by the authors to assess parents’ beliefs related to a) facts about asthma, b) nature of symptoms, c) attitudes towards inflammatory medications, d) emotional aspects of medication use, and e) treatment expectations. In contrast, details of the child’s “symptom evaluation” were not clearly described in the study, as the main focus was the role of parental illness representations on disease management in childhood asthma. Good internal consistency has been reported (Sidora-Arcoleo, Feldman, Serebrisky, Spray, 2010; Yoos et al., 2007). Although this scale was not developed to map onto Leventhal’s illness representations framework, their

treatment expectations scale could be conceptualised as being related to the *treatment-control* domain of Leventhal's common-sense model. Details of items included in this scale were not provided, so it is unclear if other subscales operationalize other domains of the model. Overall, the authors' results related to self-management are discussed very generally in terms of the overall AIRS score, rather than specific subscales. This makes it difficult to draw conclusions from the study based on Leventhal's model.

### *Statistical Analysis*

All studies reported statistical analysis using either Regression (11 studies) or Structural Equation Modelling (3 studies).

**Table 2:** Summary of Studies

| Authors (year)   | Health Condition   | Recruitment (no. of sites)   | Participants (response rate)                              | Inclusion criteria                                   | Treatment Regime  | Measures  | Findings  |
|--|--|--|---|--|---|---|---|
| Country<br><br>Bucks, Hawkins, Skinner, Horn, Seddon, Home (2009)<br><br>England | Time since Diagnosis<br>Cystic Fibrosis<br><br>Time since diagnosis not stated | Design<br>Paediatric outpatient departments (5 hospitals)<br><br>Cross Sectional | 11-17 yr olds<br><br>20 females & 18 males (73%)          | Not receiving in-patient care during data collection | Chest Physiotherapy (CPT)<br><br>Nebulised antibiotics<br><br>Enzyme Supplements (ES) | <u>IPQ-R</u><br><br><u>BMQ</u><br>Necessity of a specific treatment & concerns about potential adverse consequences<br><br><u>MARS</u><br>Self-management | <b>Correlations</b><br>Longer <i>timeline</i> perceptions associated with better self-management of antibiotics ( $r=0.35$ ; $p<0.05$ ). Not Chest Physiotherapy (CPT) or Enzyme Supplements.<br><br>Greater belief in <i>treatment-control</i> associated with antibiotic use ( $r=0.42$ ; $p<0.01$ ).<br><br>Stronger BMQ necessity beliefs associated with all measures of CPT ( $r=0.71$ ; $p<0.001$ ) and antibiotic use ( $r=0.38$ ; $p<0.05$ ).<br><br><b>Hierarchical Linear Regression (predictor variables)</b><br>CPT and Antibiotic use predicted by IPQR <i>treatment control</i> ( $r=0.09$ ; $p<0.09$ ), <i>timeline chronicity</i> ( $r=-0.06$ ; $p<0.05$ ) & BTQ necessity ( $r=-0.35$ ; $p<0.05$ ).<br><br><b>Correlations</b><br>Dietary self-management associated with less diabetes symptoms ( <i>identity</i> ) ( $r=-0.41$ ; $p<0.001$ ).<br><br>Stronger <i>control</i> beliefs associated |
| Griva, Myers & Newman (2000)<br><br>England                                      | Diabetes<br>Diagnosis of IDDM for at least 1 year before study                 | Hospital Outpatients (diabetes care clinics).                                    | 15-25 yrs old (mean 20.6 yrs; SD =4.68)<br><br>31 males & | No severe complications or other chronic health      | Insulin, diet, blood glucose monitoring & exercise                                    | <u>IPQ</u><br>NB: No differentiation between treatment-control and personal-  |   |

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|   | Mean- 7.7 years (range = 1-17, SD=4.60)         |  | 33 females (48.4%)<br><br>Cross-sectional  | conditions<br><br>English literate  |   | control beliefs.<br><br><u>Generalised self efficacy scale &amp; self efficacy of diabetes scale.</u><br><br><u>Self-report adherence scale</u><br><br><u>Glycosylated haemoglobin:</u><br>Objective measure of self-management  | with dietary self-management (r=0.48; p<0.001); exercise (r=0.30; p<0.01); glucose monitoring (r=0.69; p<0.001) and insulin recommendations (r=0.41; p=0.001).<br><br><b>Hierarchical Multiple Regression</b><br>Only perceived control dimension (IPQ) contributed significantly to overall self-management (39% of variance of total self-management) (R=0.39, t=6.46, p<0.001). |
| Iannotti, Schneider, Nasel, Haynie, Plotnick, Clark, Sobel, Simons-Morton (2006)<br><br>USA | Diabetes<br><br>At least 1 year since diagnosis | <b>Phase 1 sample</b><br>Diabetes support network<br><br><b>Phase 2 sample</b><br>Paediatric diabetes clinics. | <b>Phase 1 sample</b><br>11 child and parent dyads<br><br>8-18 yrs old: (5 boys, 4 girls)<br><br><b>Phase 2 sample</b><br>168 yp-parent dyads (76%).<br><br>10-16 yr olds and parents (mean age= 13.6 yrs) | No major chronic illness (except well-controlled asthma or thyroid problems)<br><br>Youth or parent not English literate<br><br>Those who participated in | Diabetes self-management i.e. insulin-administration routine, self-management, adjustment, dietary routine, blood glucose testing frequency and exercise frequency. | <b>Phase 1 Item development</b><br>Semi-structured interview, literature review, develop questionnaire items and second visit complete self-report questionnaire<br><br><b>Phase 2 Scale level:</b><br><br><u>Self efficacy &amp; outcome measures</u> (young people only) | <b>Hierarchical multiple regressions</b><br>Relationship between self-efficacy and all 3 measures of self-managements was mediated by expectations for positive outcomes (i.e. positive consequences).<br><br>Parents- high expectations for positive outcomes associated with poorer dietary self-management and glycaemic control,   |







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| USA                                       | weeks post-transplant.                                   |  | 11-20 yr olds (M=15.9yrs; SD=2.5). Male =61%; Female =39%).<br><br><i>Cross-Sectional</i><br><br>Questionnaires and semi-structured interview  | cognitive deficits  | obtained as indicators of adherence  | <u>Perceived Adversity scale</u><br><br><u>A-Cope Coping</u><br><br><u>Medical Adherence Measure</u><br><br><u>Self-management: Serum Immunosuppressant Drug Assay Levels.</u>                        | <p>immunosuppressant doses.</p> <p>Significant association between PA for medical procedures and total number of immunosuppressant levels above recommended range (<math>r=0.30</math>, <math>p&lt;0.07</math>).</p>   |
| Skinner, Hampson (2001)<br><i>England</i> | Diabetes<br><br><i>At least one year since diagnosis</i> | Outpatient clinics in four regional hospitals in southern England. | <p>12-18 years old (32 girls, 42 boys)</p> <p>74 completed questionnaire (51.4% of eligible population)</p> <p>Follow-up at 12 months: 54 completed follow-up. (25 females 29 males)</p> | 12-18 yrs, diagnosis for at least one year, able to complete questionnaire unaided. | Diabetes self-management (i.e. diet, exercise, blood glucose monitoring, injecting). | <u>The Well-being Questionnaire</u><br><br><u>Summary of Diabetes Self Care Schedule</u><br><br><u>The Personal Models of Diabetes Questionnaire.</u><br><br>GHb assays (measuring glycaemic control) | <p><b>Correlations</b></p> <p>Perceived effectiveness of treatment to control diabetes (<i>short-term treatment-control</i>) was correlated significantly with changes in dietary self-management (<math>r= 0.42</math>; <math>p&lt;0.005</math>), but not with any other measures of self-management.</p> <p>Greater perceived impact of diabetes (<i>short-term consequences</i>) was predictive of greater anxiety (<math>r=0.37</math>; <math>p&lt;0.01</math>).</p> |

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| Skinner, Hampton (1998)<br>England             | Diabetes<br><i>Diagnosis for at least 6 months</i> | Outpatient clinics – 4 regional hospitals south of England                     | Prospective( 1 year follow-up)<br>74 participants (51.4% response rate).<br><br>32 girls, 42 boys.<br><br>12-18 years old<br>(M=15.18; SD = 2.01).<br><br>Cross sectional | 12-18 yrs old<br><br>Able to complete questionnaire unaided. | Diabetes self-management (i.e. diet, exercise, blood glucose monitoring, injecting).          | <u>The Well-being Questionnaire</u><br><br><u>Summary of Diabetes Self Care Schedule</u><br><br><u>Personal Models of Diabetes Questionnaire</u><br><br><u>Perceived Social Support from Family questionnaire.</u><br><br><u>Perceived Social Support from Friends questionnaire.</u><br><br><u>DFBC</u><br><br><u>DIPS</u> | <b>Correlations</b><br>Short-term and Long-term <i>treatment-control</i> beliefs were related to dietary self-management. Control (i.e. short-term beliefs) $r=0.45$ , $p<0.01$ .<br>Complications (i.e. long-term beliefs) $r=0.37$ , $p<0.01$ .<br><br>Short-term and Long-term consequences beliefs were related to depression (impact $r=0.44$ , $p<0.01$ ; seriousness $r=0.35$ , $p<0.01$ ) and anxiety (impact $r=0.57$ , $p<0.001$ ).<br><br><b>Mediator Analysis</b><br>None of the personal models variables predicted blood glucose monitoring or insulin injecting.<br><br>Perceived efficacy of the treatment regime in controlling diabetes at least partially mediates the link between social support and dietary self-management (reduced beta weight for family support from 0.40 to 0.28). |
| Skinner, Hampton, Fife-Schaw (2002)<br>England | Diabetes<br><br>At least 1 year since diagnosis    | Youth Diabetics category of membership of the British Diabetic association for | 12-30 years old.<br><br>460 completed questionnaire (38% response   | Diagnosis for at least 1 yr                                  | Diabetes self-management (i.e. diet, exercise, blood glucose monitoring, injecting-medication | <u>Summary of Diabetes Self-Care Activities</u><br><br><u>Big Five Inventory 44 (assessed personality).</u>   | Bivariate Analyses<br>Short-term treatment-control beliefs associated with self-management of diet ( $r=0.32$ , $p<0.005$ ), exercise ( $r=0.31$ , $p<0.005$ ), blood glucose monitoring ( $r=0.33$ , $p<0.005$ ), insulin administration ( $r=0.20$ , $p<0.005$ ).   |

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|  |  | adolescents and young adults with diabetes.  | rate) – final sample 338 (due to missing data, age).<br><br>Cross Sectional Design  | taking.   | Perceived consequence scale (10 items of the perceived consequence scale of IPQ and<br><br><u>Perceived treatment effectiveness to control diabetes and prevent complications</u> (based on Glasgow, 1997)<br><br>Demographic Information and medical information | Long-term treatment control beliefs associated with self-management of diet ( $r=0.17$ , $p<0.005$ ), exercise ( $r=0.18$ , $p<0.05$ ), blood glucose testing ( $r=0.15$ , $p<0.05$ ).<br><br>Perceived threat (long-term consequences) associated with self-management of diet ( $r=0.23$ , $p<0.005$ ), exercise ( $r=0.18$ , $p<0.005$ ), blood glucose testing ( $r=0.12$ , $p<0.05$ ), insulin administration ( $r=0.23$ , $p<0.005$ ).<br><br>Structural equation Modelling (SEM)<br><br>Perceived treatment effectiveness to control diabetes (short-term treatment-control) was a significant predictor of all self-management measures. |
| Skinner, Howells, Greene, Edgart, McEvilly, Johansson (2003).<br><br>England | Diabetes<br><br><i>Time since diagnosis not stated</i> | Youth Diabetics category of membership of the British Diabetic association for adolescents and young adults with diabetes. Questionnaire booklet | <b>Study 1:</b> 115 participants (12-18 yrs).<br><br><b>Study 2:</b> 79 participants (12-25 yrs)<br><br><b>Study 3:</b> 44 participants | Self-management (i.e. diet, exercise, blood glucose monitoring, insulin taking) | <b>Study 1:</b><br><br><u>2 IPQ-R scales:</u> perceived consequences of diabetes & perceived effectiveness of the treatment regimen.<br><br><i>Summary of Diabetes self-Care Activities</i>   | <b>Study 1: Scale Development</b><br><br>Treatment effectiveness to prevent complications was significantly correlated with diet ( $r=0.25$ ; $p<0.01$ ).<br><br>Treatment effectiveness to control diabetes sig correlated with diet ( $r=0.25$ ; $p<0.01$ ) and blood glucose monitoring ( $r=0.27$ ; $p<0.005$ ).<br><br><b>Study 2: Internal consistency and coherence</b>   |



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|---|---|--|---|---|--|---|---|--|
|   |   |  |   |   |  |   | <u>Well-Being questionnaire.</u><br><br><u>HbA and medical records reviewed.</u>  |  |
| Skinner, John, Hampson (2000)<br><br><i>England</i> | Diabetes<br><br><i>Diagnosis of IDDM of at least 9 months</i> | Outpatient lists at four regional hospitals in South of England. | 74 (32 girls, 42 boys) recruited at baseline.<br><br>52 (24 girls, 28 boys) completed follow-up questionnaire booklet.<br><br><i>Longitudinal study</i> | 12-18 yrs old<br><br>Ability to complete questionnaire unaided. | Self-management (i.e. diet, blood glucose testing insulin injecting. | <u>Well-Being questionnaire</u><br><br><u>Summary of Diabetes Self Care Schedule</u><br><br><u>Personal Models of Diabetes Questionnaire</u><br><br><u>Perceived Social Support from Family questionnaire.</u><br><br><u>Perceived Social Support from Friends questionnaire.</u><br><br><u>DFBC</u><br><br><u>DIPS</u> | <b>Longitudinal Mediator Analysis</b><br>Perceived control of diabetes (short-term treatment-control) predictors of dietary self-management.<br><br>Greater perceived seriousness (long-term consequences) of diabetes, the poorer dietary self-management. |  |
| Yoos, Kitzman, Henderso n, McMullen                 | Asthma<br><br><i>Not stated</i>                               | Six clinical paediatric practice sites in New York               | 228 families participated (82% response rate).  | 5-12 yrs old<br><br>Family English                              | Asthma Medication usage (i.e. anti-inflammatory,                     | <u>Demographic characteristics</u><br><br><u>Background characteristics</u>   | Parental illness representation had a significant direct effect on the medication regimen (p=0.05)<br><br>Formal asthma education had a   |  |

|  |                             |                                   |   |   |  |   |  |
|--|-----------------------------|-----------------------------------|---|---|--|---|--|
| Sidora-Arcoleo, Halterman, Anson (2007)<br>USA |                             |                                   | 5-12 yrs old.<br>Cross Sectional<br>Semi-structured home interviews with children and their parents & retrospective review of medical records         | speaking<br>Child diagnosis of asthma and at least 2 asthma related HCP visits in the prior 12 months                           | rescue, other medication)  | <u>AIRS</u><br><u>Parent-HCP relationship instrument.</u><br><u>A structured assessment of asthma severity and control.</u>   | positive effect on the medication regimen ( $p=0.02$ ), whereas informal advice-seeking was associated with a less adequate medication regimen ( $p=.0003$ ).<br>Specific illness reps not presented in article only overall association of illness representations on treatment management. |
| Zebracki & Drotar (2004)<br>USA                | Asthma<br>Minimum of 1 year | Routine pulmonology clinic visits | 77 young people and their caregivers (94% response rate).<br>11-17 years olds<br>(M=13.8yrs old, SD=1.8).<br>Child:<br>Males- 51.9%<br>Females- 48.1% | 11-17yrs old,<br>Experience mild persistent to severe symptoms<br>Asthma symptoms for longer than 3 days in previous 12 months. | Medication<br>Self-Management of episodes of asthma symptoms<br>Asthma prevention behaviours | <u>Demographic information</u><br><u>Children's Health Survey of Asthma</u><br><u>Outcome expectancy Scale</u><br><u>Child Asthma Self-Efficacy Scale</u><br><u>Family adherence to medical regimen measure</u><br><u>Self Management</u> | <b>Multiple hierarchical regression</b><br>High outcome expectancy predicted greater asthma morbidity.<br>Positive outcome expectancy for asthma management was not associated with self-management to asthma treatment.   |

|  |  |                                    |   |   |   |   |  |
|--|--|------------------------------------|---|---|---|---|--|
|  |  |                                    | Caregivers 28-67yrs old (M=42.8yrs old, SD=6.8)<br><br><i>Cross Sectional</i><br><br>Questionnaires and interviews while in clinic. | No other respiratory conditions /chronic illness, English speaking  |   | Scenarios describing hypothetical episodes of asthma symptoms & asthma prevention behaviours.<br><br><u>Asthma morbidity Items from CHSA</u>                          |  |
| Zugelj, Zupancic, Komidar, Kenda, Varda, Gregoric (2010)<br><br>Slovenia | Essential Hypertension<br><br><i>Time since diagnosis not stated</i> | Two paediatric clinics in Slovenia | 97 adolescents (75% response rate)<br><br>13-23 yrs (M=17.34; SD = 2.28)<br><br>34% female, 66% male.<br><br><i>Not stated</i>      | Diagnosis of essential hypertension<br><br>Diagnosis at least 4 months prior to participation.<br><br>No co-morbid condition. | General self-management: general tendencies to adhere to medical regimens.<br><br>Specific self-management: Low salt diet, low fat diet, medication taking, exercise, stress reduction. | <u>Demographic and Medical Data.</u><br><br><u>Brief IPQ</u><br><br><u>Inventory of Child/Adolescent Individual Differences</u><br>Assessing personality<br><br>MOSAQ | <b>Hierarchical multiple regression analyses.</b><br><br><i>Treatment-control</i> accounts for 16% of the variance of specific self-management scale.<br><br>Variation in self-management to medication-taking behaviour could well be accounted for by <i>treatment-control</i> , <i>emotional burden</i> , <i>concern</i> (43% of variance). |

Notes: IPQ-R = Illness representations questionnaire-revised version. BMQ = Beliefs about Medicines Questionnaire: specific. MARS = Medication Adherence Scale. PA Scale= Perceived Adversity Scale. MAM=Medical Adherence Scale. DFBC=Diabetes Family Behaviour Checklist. DIPS = Diabetes Inventory of Peer Support. SDQ=Strength and Difficulties Questionnaire. HbA1c = Glycosylated Haemoglobin. AIRS = Asthma Illness Representations Scale. HCP =Health Care Provider. MOSAQ= Medical outcomes study adherence questionnaire.



## **Risk of Biases**

The issue of biases has been addressed in this review by consulting key documents that provide guidance on the reporting of systematic reviews and assessment of risk of bias in research studies.

Both PRISMA (Liberati, 2009) and the Cochrane Handbook for Systematic Reviews of Interventions (Higgins & Green, 2009) used the term “risk of bias” rather than “quality”. They argue that “risk of bias” assesses the “extent to which results of included studies should be believed” (Higgins & Green, 2009). The term “risk of bias” will therefore be used when critically evaluating research studies in this review.

Risk of bias was assessed using the Transparent Reporting of Evaluations with Non-Randomized Designs (Des Jarlais, Lyes & Crepaz, & The TREND Group, 2004). This is recommended by the Journal of Health Psychology (Marks, 2010) as well as criteria for publication of experimental and quasi-experimental research in psychology (Ramos-Alvarez & Moreno-Fernandez, 2008). Both were adapted for the purposes of this review. In addition, a scoring system has been included (i.e. 2 = yes, 1=partially, 0 = no or unknown).

A summary of the risk of bias assessment is displayed in Table 3 highlighting common strengths and limitations across studies. In terms of strengths, each study included in this review had a clearly focused question and background information to justify the importance of the research. Appropriate procedures and analyses were selected to meet research aims across studies and authors highlighted limitations and biases. All, except one study, interpreted their results and statistical analysis in the context of current evidence and theory (Yoos et al., 2007).

Whilst there were strengths across research articles included in the review, the majority of studies were cross-sectional, thus meaning that conclusions based on causal associations were not possible. Moreover, small sample sizes for some studies made it difficult to generalise findings. The method of measuring both illness representations and outcomes was not consistent with varying levels of validity reported by authors depending on the measure used.

Overall, studies showing the lowest risk of bias explored illness perceptions in Cystic Fibrosis (Bucks et al., 2010), Diabetes (Iannotti et al., 2006; Nouwen et al., 2009; Skinner et al., 2002; Skinner et al., 2000; Skinner & Hampson, 1998), Asthma (Zebracki & Drotar, 2004) and Hypertension (Zugelj et al., 2010). Given that the evidence provided by these studies is stronger based on a risk of bias assessment, this review will focus more on the evidence from these studies.

**Table 3:** Risk of Bias Assessment -based on *The Transparent Reporting of Evaluations with Non-randomized Designs (Jarlais et al., 2004)* & *Criteria of the peer review process for publication of experimental and quasi-experimental research in Psychology (Ramos et al., 2008)*.

- 2= yes (i.e. low risk of bias)
- 1= partially (i.e. medium risk of bias)
- 0= no/unknown (i.e. high risk of bias, or not discussed).

| <b>Risk of Bias Criteria</b>   | Bucks et al (2009) | Griva et al (2000) | Nouwen et al (2009) | Iannotti et al (2006) | Ratcliff et al (2010) | Skinner, et al (2002) | Skinner, et al (2000) | Skinner & Hampson (1998) | Skinner & Hampson (2001) | Skinner, et al (2003) | Law et al (2002) | Yoos et al (2007) | Zebracki & Drotar (2004) | Zugelj, et al (2010) |
|--|--------------------|--------------------|---------------------|-----------------------|-----------------------|-----------------------|-----------------------|--------------------------|--------------------------|-----------------------|------------------|-------------------|--------------------------|----------------------|
| <b>Title and abstract</b>  |                    |                    |                     |                       |                       |                       |                       |                          |                          |                       |                  |                   |                          |                      |
| <i>Is there a clearly focused question including information on target population or study sample?</i>   | 2                  | 2                  | 2                   | 2                     | 2                     | 2                     | 2                     | 2                        | 2                        | 2                     | 2                | 2                 | 2                        | 2                    |
| <b>Introduction</b>  |                    |                    |                     |                       |                       |                       |                       |                          |                          |                       |                  |                   |                          |                      |
| <i>Does the background literature to justify the importance of the research problem/clear rationale?</i> | 2                  | 2                  | 2                   | 2                     | 2                     | 2                     | 2                     | 2                        | 2                        | 2                     | 2                | 1                 | 2                        | 2                    |
| <i>Is there a clear hypothesis follows from theory?</i>  | 2                  | 1                  | 2                   | 2                     | 2                     | 2                     | 2                     | 2                        | 0                        | 0                     | 1                | 1                 | 2                        | 2                    |



| Discussion  |    |    |    |    |    |    |    |    |    |    |    |    |    |    |    |    |    |   |   |   |
|---|----|----|----|----|----|----|----|----|----|----|----|----|----|----|----|----|----|---|---|---|
| <i>Is the interpretation of results and statistical analysis congruent with research problem explained in the introduction section?</i> | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2 | 2 | 2 |
| <i>Are the limitations of research and potential bias discussed?</i>  | 2  | 1  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2 | 2 | 2 |
| <i>Are the general interpretations of the results in the context of current evidence and theory?</i>                                    | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2  | 2 | 2 | 2 |
| <b>Total</b>  | 20 | 19 | 21 | 22 | 18 | 21 | 21 | 21 | 21 | 20 | 18 | 19 | 18 | 20 | 15 | 20 | 22 |   |   |   |

## **What does the evidence tell us about relationships between Illness Representations and Self-Management?**

In this review, evidence for each dimension of illness representations is discussed, with a view to ascertaining the extent to which illness representations can help in understanding self-management in children/young people with chronic health conditions. Given that illness *coherence*, *emotional representations* and *timeline-cyclical* beliefs have been integrated into the IPQ-R for theoretical and psychometric reasons, they will also be included in this review.

### **Identity**

Illness *identity* refers to symptoms a person views as part of their illness and at a more abstract level, the illness label (e.g. diabetes) (Scheier & Carver, 2003). Evidence for the association between the perceived identity of chronic health conditions in children and young people and self-management is extremely limited.

Five studies included in this review measured perceived *identity* (Bucks et al., 2009; Griva et al., 2000; Law et al., 2002; Skinner et al., 2003; Zugelj et al., 2010;). However, only one of these studies found a significant and moderate relationship ( $r = -0.41$ ,  $p < 0.001$ ) between illness *identity* and dietary self-management. Specifically, Griva et al. (2000) found that those who experienced fewer Diabetes symptoms had better dietary self-management, however, no significant findings were found for other self-management behaviours, including insulin administration, exercise or blood glucose monitoring.

Whilst Griva et al. (2000) report an association between *identity* beliefs and dietary self-management in Diabetes, other studies of Diabetes did not find this association (Law

et al., 2002; Skinner et al., 2003). Similarly, no significant findings were reported for *identity* and any aspect of self-management measured in Cystic Fibrosis (Bucks et al., 2009) and Hypertension (Zugeli et al., 2010). In terms of risk of bias, Griva et al. (2000) used the IPQ in their research whereas others used the IPQ-R or brief IPQ-R. This is an important consideration given that the identity scale in the IPQ had poorer test-retest reliability (0.06,  $p>0.05$ ) than the IPQ-R over a six-month period (0.57,  $p<0.001$ ) (Moss-Morris et al., 2002; Weinman et al., 1996). It could be argued, therefore, that Griva's finding related to *identity* is questionably stable.

Exploration of inter-relations between illness beliefs highlighted that those with more Diabetes related symptoms were more likely to believe that they have less control over their condition and more negative consequences (Griva et al., 2000). This is consistent with Weinman et al. (1996) who highlighted that Leventhal's illness dimensions are not necessarily independent. Therefore, given that only one study found that illness identity was associated with self-management and other studies have lower risk of bias, it could be hypothesised that a combination of illness representations may be more predictive of self-management than when illness identity is considered individually.

### **Timeline (chronicity and cyclical)**

*Timeline-chronicity* refers to an individual's expectation regarding the duration of their illness (i.e. acute or chronic). Low internal consistency values led to the inclusion of new items assessing cyclical timeline beliefs in the revised IPQ (*timeline-cyclical*) (Moss-Morris et al., 2002).

Five studies measured *timeline-chronicity* beliefs (Bucks et al., 2009; Griva et al., 2000, Law et al., 2002; Skinner et al., 2003; Zugelj et al., 2010). One study reported that

young people with Cystic Fibrosis, who believed their illness would last for a long time, were better at managing their antibiotic use (Bucks et al., 2009). Similar findings were not reported for others aspects of self-management (i.e. physiotherapy or enzyme use). Bucks et al. (2009) suggested that difficulties with enzyme self-management in Cystic Fibrosis were mainly due to forgetting to take the enzymes rather than a decision to self-manage a particular aspect of treatment. Other studies did not find a link between *timeline-chronicity* beliefs and self-management.

*Timeline-cyclical* beliefs were measured using the IPQ-R in two studies (Bucks et al., 2010; Law et al., 2002). Law et al. (2002) reported that *timeline-cyclical* beliefs were not associated with self-management. Furthermore, Bucks et al. (2010) described *cyclical timeline* beliefs as psychometrically unsound and therefore did not include them in the analysis. *Timeline* perceptions (chronicity or cyclical) were not measured in any other studies.

Findings could suggest that *timeline-chronicity* beliefs are condition- and treatment-specific in children and young people. This idea would fit with the findings that timeline beliefs were associated specifically with antibiotic use in Cystic Fibrosis, but not chest physiotherapy or enzyme use in the same study (Bucks et al., 2009), nor with self-management of other chronic health conditions. However, further research exploring beliefs in Cystic Fibrosis is necessary to confirm or disprove this hypothesis.

## **Consequences**

The *consequences* domain reflects the perceived seriousness of a condition, severity of pain and the impact that the condition has on an individual's life. Twelve studies included



in this review tapped into the *consequences* domain. However, their method of measuring this dimension of illness representations varied considerably across studies (see Table 1).

Results indicated that *consequences* beliefs did not significantly affect self-reported self-management in Diabetes (Law et al., 2002; Griva et al., 2000; Nouwen et al., 2009; Skinner & Hampson, 1998; Skinner & Hampson, 2001) or self-management of diet, medication, exercise and stress reduction in Hypertension (Zugelj et al., 2010). In contrast, Renal Transplant patients with poor self-management of medication had difficulties related to not being able to do what others are doing ( $r=.36$ ,  $p<.04$ ) (Radcliff & Blount, 2010). Bucks et al. (2009) did not complete an analysis of the *consequences* scale in their studies, due to low internal-consistency levels (i.e. 0.66).

A factor analysis of perceived *consequences* scale items in the Personal Models of Diabetes Questionnaire (Hampson, Glasgow & Toobert, 1990) indicated two aspects to perceived consequences, namely the impact of Diabetes on the individual (short-term beliefs) and threat of Diabetes to health (long-term beliefs) (Skinner et al., 2002). In studies that made this differentiation, perceived impact (short-term beliefs) was unrelated to self-management (Skinner et al., 2000; Skinner et al., 2002). However, mixed findings were reported for perceived threat (long-term beliefs). For example, a cross-sectional study reported that greater perceived threat was associated with better self-management of diet, exercise, blood-glucose testing and insulin administration (Skinner et al., 2002). Contrary to this, a longitudinal study indicated the opposite effect, that young people who thought their Diabetes was serious had poorer dietary self-management (Skinner et al., 2000). Both studies had low risk of bias (Skinner et al., 2000 & Skinner et al., 2002). However, findings reported by Skinner et al. (2000) are more consistent with Hagger and Orbell's meta-analysis, where beliefs in serious consequences were positively associated with

avoidance/denial coping strategies and negatively associated with adaptive outcomes (Hagger and Orbell, 2002). According to Leventhal's model, this would suggest that young people who perceive their Diabetes to be very serious would have difficulty managing their self-care regimen. Alternatively, it could also be hypothesised that those who poorly manage their diabetes, have lots of symptoms, which results in them perceiving negative consequences associated with the condition.

In addition to the studies that specifically tapped into the consequences dimension of illness representation, a study conducted by Iannotti and colleagues (Iannotti et al., 2006) was included in this review because, within social cognitive theory, *outcome expectancies* relates to the *perceived consequences* domain of illness representations (Nouwen et al., 2009). Iannotti et al. (2006) found that when positive outcome expectations were high, self-efficacy had a greater association with Diabetes self-management. This is consistent with social cognitive theory that the more confident people are in managing their diet, the less they experience negative consequences of Diabetes (Nouwen et al., 2009).

Whilst findings for the *consequences* domain of illness representations and self-management are mixed, the studies did seem to suggest that how a person perceives the consequences of their condition was important in their overall psychological well-being. Associations between consequences and anxiety (Law et al., 2002; Skinner & Hampson, 2001; Skinner et al., 2000), positive well-being, depression, general well-being (Skinner et al., 2000) and diabetes distress (Nouwen et al., 2009) were all reported in the sample of studies selected for this review. Whilst it is not within the scope of this review to investigate the role of illness representations to psychological well-being, a literature

review exploring illness representations and psychological well-being, particularly related to childhood chronic health conditions, would be useful.

## **Causes**

Causal illness representations have been categorised according to four main components, namely psychological attributions (e.g. stress or worry), risk factors attributions (e.g. hereditary - it runs in my family), immunity attributions (e.g. a germ or a virus) and chance attributions (e.g. chance or bad luck) (Moss-Morris et al., 2002).

Of the four studies that investigated all dimensions of the model, items in the *cause* scale were not analysed (Bucks et al., 2009; Griva et al., 2000; Law et al., 2002; Skinner et al., 2003). Where inter-correlations between dimensions were completed, no significant correlations between cause and other dimensions were reported (Skinner et al., 2003). Consequently, the studies included in this review cannot confirm if causal beliefs can contribute to our understanding of self-management in children and young people with chronic health conditions.

## **Control**

The extent to which a person perceives their illness as preventable, curable or controllable is referred to as the *control* domain of illness representations (Leventhal et al., 2003). The IPQ-R distinguishes between *personal control* (including self-efficacy beliefs) and *treatment control* (i.e. belief in treatment or recommended advice).

Overall, the literature highlighted that *treatment control* beliefs were a significant predictor of self-management of antibiotics in Cystic Fibrosis (Bucks et al., 2009) as well as diet, medication, exercise, and stress reduction in Hypertension (Zugelj et al., 2010).

Furthermore, in diabetes, *treatment control* beliefs were associated with dietary self-management (Nouwen et al., 2009; Skinner et al., 2003; Skinner & Hampson., 1998; Skinner et al., 2000; Skinner et al., 2002), blood glucose monitoring (Skinner et al., 2003; Skinner & Hampson., 1998; Skinner et al., 2003) and exercise (Skinner et al., 2003) in Diabetes. In contrast, treatment-control beliefs were not associated with self-management in Asthma (Zebracki & Drotar, 2004). Therefore, those who believed their treatment regimen would be effective in controlling their illness, had better self-management of specific aspects of their treatment regimen (in Diabetes, Cystic Fibrosis and Hypertension, but not Asthma).

In contrast, *personal-control* beliefs were only investigated by three studies (Bucks et al., 2009; Law et al., 2002; Zugelj et al., 2010). Bucks et al. (2009) excluded this scale from their analysis due to low internal consistency values, and other studies indicated that personal control did not have any influence over any aspect of self-management in young people (Law et al., 2002; Zugelj et al., 2010).

These results, taken together, suggest that children and young people who believe that their condition can be controlled by treatment are more likely to engage in more helpful self-management behaviours. Moreover, believing in the effectiveness of the treatment regimen seems to be more important than one's own beliefs about personal control over the illness.

Contrary to this, Law and colleagues (2002) found neither *personal* nor *treatment control* were linked to self-management but, instead, that control beliefs were linked to emotional well-being. This is the only diabetes study that did not report a relationship between perceived control and self-management, and the authors recognised the limited generalizability of this study, due to their small sample size.

When investigating the role of long-term and short-term control beliefs in Diabetes, results were mixed. Three studies reported that beliefs related to the short-term (i.e. effectiveness of treatment to control diabetes), rather than long-term expectations (i.e. treatment to prevent complications), influenced dietary self-management (Skinner & Hampson, 2001; Skinner et al., 2000). Nouwen et al. (2009) also reported short-term treatment effectiveness beliefs to control Diabetes were associated with dietary self-management, although they did not compare this to more long-term beliefs related to preventing complications. In contrast, other studies indicated that both short-term and long-term control beliefs were associated with dietary self-management (Skinner & Hampson, 1998; Skinner et al., 2003) and blood glucose monitoring (Skinner & Hampson, 1998). More research investigating the role of short- and long-term beliefs across other chronic health conditions particularly in younger children is necessary before any conclusions can be made.

Like many of the other studies presented throughout this review, *treatment-control* illness representations appeared to be largely specific to particular aspects of a complex treatment regimen. Only one study reported that treatment effectiveness beliefs and self-management were associated with all aspects of self-management in Diabetes (Skinner et al., 2002). Whereas, the majority of studies found that perceived *treatment-control* was associated with particular aspects of self-management and not others. For example, young people who believed their treatment regime would control their Diabetes were better at self-managing diet and blood glucose monitoring in Diabetes (but not insulin injections) (Skinner & Hampson, 1998), medication in Hypertension (but not exercise and diet) (Zugelj et al., 2010) and antibiotic use in Cystic Fibrosis (but not chest physiotherapy or enzyme use) (Bucks et al., 2009). Therefore, when children are expected to manage

complex treatment regimens, generalisations cannot be assumed across treatments for the same condition. This may be because particular aspects of self-management routines are managed or prompted by parents and therefore children's beliefs do not have a causal role to play. In order to investigate this hypothesis further, studies that consider parents' beliefs and other systemic factors would be useful.

### **Emotional Representations**

*Emotional representations* relate to feelings associated with the illness (e.g. "my illness makes me feel angry"). Emotional Illness Representations were included in the revised IPQ-R because Leventhal's self-regulatory model is a parallel process including both cognitive and emotional illness representations (Moss-Morris et al., 2002). Zugelj et al. (2010) reported that lower emotional burden about having Hypertension was associated with better medication taking compared to those who were able to cope with the emotional impact of living with a chronic illness (Zugelj et al., 2010). In contrast, *emotional representations* were not associated with any of the self-management behaviours in Diabetes (Law et al., 2002) or Cystic Fibrosis (Bucks et al., 2009). Emotional representations were not measured by other studies.

### **Illness coherence**

Similarly to the *emotional representation* subscale, the perceived *coherence* subscale was included in the revised IPQ-R. Illness coherence assesses the extent to which one's illness makes sense or is puzzling. Although this is not a domain of Leventhal's self-regulatory model, Moss-Morris et al. (2002) described this as an overarching meta-cognition reflecting the way a person evaluates their understanding or helpfulness of his/her illness

representations. Results of studies indicated that *illness coherence* was not associated with self-management in Hypertension (Zugelj et al., 2010) or Diabetes (Law et al., 2002).

*Illness coherence* was not measured by the other studies included in this review.

## Discussion

This review aimed to determine the extent to which illness representations can promote understanding of self-management in childhood chronic health conditions. It highlights those dimensions of illness representations that seem to be associated with specific self-management behaviours within a condition.

The review has demonstrated that *treatment-control* beliefs have been found to be associated with self-management in Diabetes, Hypertension and Cystic Fibrosis (Bucks et al., 2009; Griva et al., 2000; Nouwen et al., 2009; Skinner & Hampson, 1998; Skinner & Hampson, 2001; Skinner et al., 2002; Skinner et al., 2003; Zugelj et al., 2010). Children and young people who believe that their condition can be controlled by treatment are more likely to engage in more helpful self-management behaviours. Moreover, *short-term treatment-control* beliefs were consistently associated with self-management. These findings are consistent with a meta-analysis reporting that perceived controllability is related to active coping and cognitive re-appraisal (Hagger & Orbell, 2003)..

The condition- and treatment-specific nature of illness representations was also highlighted by this review, particularly in relation to *control* beliefs (Skinner & Hampson, 1998; Zugelj et al., 2010), *timeline-chronicity* (Bucks et al., 2009) and *emotional representations* (Zugelj et al., 2010). The difference in findings between these elements of a treatment regimen was discussed by the authors. This indicates that generalisations across conditions or aspects of complex treatment regimens cannot be made.

Inter-correlations between illness representations dimensions were also found (e.g. Griva et al., 2000). This is supported with Hagger and Orbell's meta-analysis who reported consistent associations between *control*, *consequences*, *identity* and *timeline* dimensions (Hagger and Orbell, 2002). Whilst each dimension was discussed individually in this



review, it is conceivable that a combination of illness representations have a joint influence over self-management.

Overall, the current review has highlighted that across studies the relationship between illness beliefs and self-management varied depending on the condition and particular aspect of the treatment regimen. However, most consistently, *short-term treatment control* beliefs were most often associated with self-management in children with chronic health conditions. Other illness beliefs had more limited/mixed findings (*timeline-chronicity, identity, emotional representations, consequences*) or no/questionable evidence (*causes, illness coherence, personal control, timeline-cyclical*), but have been linked to other aspects of outcome, such as psychological well-being.

### **Other factors that may influence self-management**

It is acknowledged that self-management is influenced by many different factors. Fielding & Duff (1999) proposed a multi-factorial model that considers the influence of individual factors & resources, treatment factors, family factors, social and material resources when understanding self-management of treatment regimes.

A number of variables in addition to children/young people's illness representations were investigated in the articles identified for this review, namely social support (Skinner, et al., 2000; Skinner & Hampson, 1998), personality (Skinner et al., 2002; Zugelj et al., 2010), self-efficacy (Griva et al., 2000; Iannotti et al., 2006; Nouwen et al., 2009; Zebracki & Drotar, 2004); treatment beliefs (Bucks et al., 2009), parental illness representations (Yoos et al., 2007), and age (Bucks et al., 2009). It is also recognised that the influence of factors within the wider health care system also need to be considered when supporting

children and young people to manage complex treatment regimens (e.g. access to healthcare and patient-clinician communication; Yoos et al., 2007).

It is not within the scope of this review to explore the role these other variables have in relation to self-management. However, studies included in this review have indicated that illness representations may act as a mediating factor for other variables (i.e. self-efficacy, personality and demographic factors). Further research is needed to add to our understanding of how illness representations are associated with other variables. It is recognised that role of parental illness representations and treatment perceptions are particularly related to this review topic and therefore are discussed in more detail below as directions for future research.

#### *Parental Illness Representations*

Parental factors (Iannotti et al., 2006; Law, 2002) warrant further exploration in relation to illness representations and self-management. This seems crucial considering the influence of family at different stages of a child's development. Moreover, research has indicated that beliefs that clinicians and families have about a particular health condition can be very different, which may in turn influence self-management (Yoos et al., 2007).

#### *Treatment Perceptions*

Since the self-regulation model was initially developed, treatment perceptions have extended our understanding of illness representations and self-management of treatment regimens (Horne, 2003). One study included in this review applied Horne's necessity and concerns framework to understanding self-management in children with chronic health conditions (Bucks et al., 2009). Bucks and colleagues found that necessity beliefs were

associated with all measures of chest physiotherapy and antibiotic use in Cystic Fibrosis. Further research exploring the role of treatment perceptions in childhood chronic illness is needed.

### **Clinical Implications**

Clinically, an understanding of children and young people's beliefs about their chronic illness can, potentially, help to improve both outcomes and communication in medical consultations (Petrie & Weinman, 2006).

More specifically, assessments that focus on particular beliefs, namely *treatment control*, might be useful when young people are struggling to manage their treatment regime effectively. Prevention has also been suggested by screening young people for beliefs that put them at risk of problems related to self-management (Iannotti et al., 2006).

In terms of intervention, cognitive behavioural interventions that seek to explore individual's cognitive perceptions (related to treatment-control) may improve self-management in those who believe that their condition is not susceptible to treatment control (Bucks et al., 2009; Nouwen et al., 2009). Further intervention studies investigating the effectiveness of developmentally appropriate cognitive-behavioural therapy (CBT) for children and young people who struggle to manage their treatment regime is needed (Law et al., 2002).

### **Biases**

Research studies identified have a number of biases that warrant further discussion. Firstly, the authors acknowledged that their use of self-report methods of self-management as a primary outcome measure raised issues of reliability and validity when compared to more

objective measures (Bucks et al., 2009; Griva et al., 2000; Nouwen et al., 2009; Skinner & Hampson, 2001; Skinner, John & Hampson, 2000; Zebracki & Drotar, 2004). Other more reliable ways of measuring self-management has also been proposed namely, electronic metered dose inhaler monitors in Asthma (Zebracki & Drotar, 2004) and multiple informants e.g. parent report (Nouwen et al., 2009). Furthermore, Griva et al. (2000) found that metabolic control was associated with different aspects of self-management in diabetes. However, the authors recognised that metabolic control may also be influenced by other factors in their study e.g. direct physiological impact of emotional distress (Griva et al., 2000).

A second methodical issue relates to how illness representations are measured. Whilst the majority of studies used questionnaires that explicitly mapped onto particular dimensions of Leventhal's illness representations, three studies did not (Iannotti et al., 2006; Yoos et al., 2007; Zebracki & Drotar, 2004). This means that their validity in terms of illness representations is unclear. It is recognised that questionnaires based on Leventhal's illness representations also had some limitations, especially when adaptations were made to generic measures to take into account specific characteristics of each condition. For example, it was recognised by Skinner and Hampson in their study that the personal models questionnaire referred to self-management in very general terms rather than exploring specific aspects of self-management (Skinner and Hampson, 2001). Skinner et al. (2003) argued that a "combination of generic and disease specific scales is likely to be the most efficacious way of assessing" an individual's beliefs about their condition (Skinner et al., 2003). Although the psychometric properties of condition specific scales have been completed for certain conditions (e.g. diabetes), there is clearly a need to develop the reliability and validity of scales for other less researched conditions such as

Cystic Fibrosis (Bucks et al., 2009). The usual constraints associated with using questionnaires were also discussed namely, that key important beliefs may not be included in the questionnaire. Skinner et al. (2003) attempted to overcome this bias by including the opportunity for qualitative descriptions of individuals' beliefs.

The cross-sectional nature of the majority of studies identified for this review is also acknowledged as a limitation of the research in this area by authors (Bucks et al., 2009; Griva et al., 2000; Iannotti et al., 2006; Law et al., 2002; Nouwen et al., 2009; Zebracki & Drotar, 2004). Longitudinal research would provide an indication of causality. It could also investigate changes in illness beliefs and self-management over time in order to identify how symptom fluctuations impact on variables (Bucks et al., 2009).

Low participant numbers (Bucks et al., 2009; Law et al., 2002; Ratcliff & Blount, 2010; Skinner et al., 2000), low response rates (Nouwen et al., 2009; Skinner & Hampson, 1998), bias towards higher socio-economic groups (Skinner et al., 2000; Zebracki & Drotar, 2004) and high DNA rates at clinic (Skinner & Hampson, 1998) are re-occurring problems which have limited the generalisation of these findings. Furthermore, the condition-specific and treatment-specific nature illness representations also mean that generalisations cannot be made at this stage.

Finally the nature of the questionnaires used in these studies means that children and young people who are not English literature were excluded. Future research should seek to adapt their research include participants from diverse backgrounds (Ratcliff & Blount, 2010).

Whilst it seems that there is more evidence for the *treatment-control* dimension of the illness representations model, it is recognized that studies with larger sample sizes should seek to explore the *treatment-control* dimension of Leventhal's common-sense

model in children and young people with chronic health conditions. It is proposed that future research should also focus on the mediating role of treatment *necessity* and *concerns* beliefs (Horne, 2003) as well as distinguishing between short-term and long-term treatment effectiveness beliefs when applying this model to children and young people (Skinner et al., 2002).

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**Empirical Paper**

**Examining illness representations, treatment perceptions, and self-efficacy in relation  
to dietary self-management and psychological well-being, in young people with  
Coeliac Disease (CD) and their parents**

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## Abstract

A cross-sectional questionnaire based study is presented exploring Leventhal's illness representations, Horne's treatment perceptions, Bandura's self-efficacy, self-management and well-being in young people with Coeliac Disease (CD) and parents. Forty young people and 34 parents (34 parent-child dyads) were recruited from hospital outpatient clinics in the United Kingdom.

Bivariate correlations showed that *timeline-cyclical* beliefs and treatment *concerns* were associated with poorer self-management. In terms of well-being, *timeline-cyclical*, *identity* and *coherence* were related to negative well-being, whereas *treatment control* was associated with positive well-being. In terms of self-efficacy, young people with high levels of *self-efficacy* were more likely to have better self-management and positive well-being. Finally, incongruence in *timeline-cyclical* beliefs between young people and their parents was related to higher parental stress.

Results are described within the context of published empirical research. Limitations of the present study are recognised and suggestions for future research are provided with a view to address some of these limitations. Finally, implications for clinical practice are discussed.

*Keywords: illness representations, illness beliefs, coeliac disease, paediatric, self-management, well-being*

## **Introduction**

### **What is Coeliac Disease?**

Coeliac Disease (CD) is an autoimmune disease of the small intestine. The condition is characterised by flattened villi, associated with eating foods containing gluten (i.e. wheat, rye and barley) (Fasano & Catassi, 2005). The clinical presentation of CD in children can vary considerably. Intestinal signs and symptoms include persistent nausea, vomiting, diarrhoea, re-current abdominal pain, cramping and bloating (NICE, 2009). Non-intestinal signs and symptoms include anaemia, failure to thrive, poor growth, sudden unexpected weight loss and prolonged fatigue (NICE, 2009). Detection of the condition is further complicated because some children with CD may not experience any obvious symptoms (Fasano & Catassi, 2005; NICE, 2009). The range of clinical presentations associated with the condition means that CD is under-diagnosed (Fasano & Catassi, 2005; Jones & Sleet, 2009; NICE, 2009). More specifically, it has been estimated that around 2.5 million people with CD are still undiagnosed in Europe (Sabatino & Corazza, 2009).

Although there is currently no cure for CD, in the majority of people it can be controlled through life-long self-management of a gluten-free diet. Possible long-term complications of untreated CD include increased risk of infertility, bone fractures and malignancy (NICE, 2009). Consequently, routine monitoring of children with a diagnosis is essential to promote long-term management of the condition (Haines, Anderson & Gibson, 2008).

A recent large European population study of 29,212 participants (adults and children) reported prevalence figures of 1% across the sample, when screened for CD (Mustalahti et al., 2010). This finding is consistent with a longitudinal child UK study (Bingley et al., 2004).

## **Self-Management of a Gluten-Free Diet in Coeliac Disease**

Published epidemiological studies have suggested that 40-95% of children and young people with CD are on a strict gluten-free diet (Chauhan, Kumar, Dutta, Srikanta & Kumar, 2010; Errichiello et al., 2010; Jackson, Glasgow, & Thom 1985; Kumar, Walker-Smith, Colyer & Halliday, 1988; Mayer, Troncone, Auricchio & Marsh, 1991; Rashid et al., 2005). This is similar to a systematic review of adult CD research reporting rates for strict self-management ranging from 42% to 91% (Hall, Rubin & Charnock, 2009).

Although reported rates of self-management are variable, it is clear that many young people struggle to maintain a gluten-free diet, despite benefits to physical health. It is likely that a combination of factors (e.g. lack of symptoms when not on a gluten-free diet, knowledge about the disease and treatment, food labelling, availability of gluten-free meals in restaurants and schools) may contribute to difficulties with self-management (Roma et al., 2010). Therefore, a better understanding of the emotional, psychological and socio-cultural factors associated with self-management is likely to enable health professionals to support young people to better manage their gluten-free diet (Mulder & Cellier, 2005).

## **Emotional and Behavioural Problems associated with Coeliac Disease in young people**

The prevalence of emotional and behavioural difficulties in children and young people with CD is unknown (Niederhofer & Pittschofer, 2006). Mixed findings across available studies make it difficult to draw conclusions. For example, anxiety and depression (measured by Hospital Anxiety and Depression Scale) were not significantly different from controls in a sample of 124 young people and young adults (age 12-25 years old)

(Calsbeek et al., 2006). Similarly, no significant difference in prevalence of anxiety, depression or “disruptive behaviour disorder” problems at school was reported in a sample of 29 young people with CD (Pynnonen et al., 2004). However, when Pynnonen et al. (2004) took into account the lifetime prevalence of difficulties, significantly higher lifetime prevalence of “major depressive disorder” and “lifetime disruptive behaviour disorder” in young people with CD compared to controls was found. Furthermore, depressive disorder and disruptive behaviour disorder before diagnostic biopsy was higher in the CD group (compared to controls) (Pynnonen et al., 2004).

Two studies compared emotional and behavioural difficulties before and after commencing a gluten-free diet (Pynnonen et al., 2005; Pynnonen et al., 2005). Results indicated an improvement in “depressive symptoms”, behavioural difficulties (Pynnonen et al., 2005) and symptoms associated with ADHD (Niederhofer & Pittschofer, 2006). Whilst the authors attribute medical explanations for improvements, it is also important to explore possible psychosocial factors that might also contribute to these changes, such as young people’s beliefs about CD, their confidence in being able to manage their condition and improvements in physical well-being associated with self-management.

### **Health related quality of life and psychological well-being of children with Coeliac disease.**

Health related quality of life (HRQoL) in children with CD is reported to be similar to the general population (Kolsteren, Koopman, Schalekamp & Mearin, 2000). In particular, research has shown that the majority of children and young people have good family integration, good social relationships and good school integration (Errichiello et al., 2010). This indicates that the majority of young people with CD are resilient and have adapted



well to living with the condition. In contrast, a disease specific quality of life (QoL) measure (Celiac Disease DUX - CDDUX) indicated that overall, young people with CD experience poor to neutral quality of life (van Doorn, Winker, Mearin, Koopman, 2008). The authors explain this discrepancy in outcomes by highlighting the different questionnaires used to assess QoL across studies (van Doorn, Winker, Mearin, Koopman, 2008). Moreover, it is recognised that the available studies have methodological problems, such as a small sample sizes, no differentiation between children and young people and no age-matched control groups (Wagner et al., 2008). Consequently, findings are not conclusive.

Previous research has explored HRQoL within the context of dietary self-management. Wagner and colleagues (2008) found that young people who did not manage their gluten-free diet reported lower QoL, more family problems, problems in their social time and lower well-being than those who were able to manage the gluten-free diet well. Likewise, a study in India identified that those who did not maintain a gluten-free diet were more likely to spend most of their time alone, were tired easily, less interested in school, afraid of new situations, felt sad and unhappy and had trouble concentrating (Chaudan et al., 2010). These findings suggest that if young people are able to self-manage their gluten-free diet, their QoL is likely to be good (Wagner et al., 2008).

Although the majority of young people do seem to adjust well to their condition, it is recognised that there are significant challenges that young people with CD face on a daily basis. Research has highlighted issues such as the stigma of having coeliac disease (Olsson, Lyon, Hornell, Ivarsson & Sydner, 2009), impact on social activities (Errichiello et al., 2010; Rashid et al., 2005), eating at restaurants and poor palatability of food (Roma et al., 2010). It is argued that social restrictions associated with managing a gluten-free

diet might have a negative impact on QoL, even though physical health has improved (Strating, 2008). Moreover, having asymptomatic CD may also be a reason for not managing the gluten-free diet well (Roma et al., 2010). In addition to these clinical reasons for variability in outcomes, the method of measuring HRQoL across studies needs to be highlighted. It is conceivable that methodical inconsistencies across studies may also be impacting on outcomes (van de Water & Mulder, 2009).

Taken together, the research indicates that some young people do not effectively manage a strict gluten-free diet, and some may experience poorer QoL and psychological well-being. Although a series of possible explanations have been proposed by authors for these issues, no clear theoretical framework has underpinned these studies. Taking this into consideration, a theoretically driven research study is now required.

### **Theoretical Underpinnings of the Research**

Leventhal's Self-regulation theory (Leventhal, Brissette & Leventhal, 2003) and Bandura's self-efficacy theory (Bandura, 1997) have provided frameworks to understand self-management and psychological well-being in young people with chronic health conditions. It is therefore proposed that these theories could be applied specifically to CD in order to understand the factors that relate to dietary self-management and well-being in young people.

### **Self-Regulation Theory**

Self-regulation theory proposes that a person's knowledge and experience of an illness influences their beliefs about their condition (Leventhal et al., 2003). Five main components to cognitive illness representations have been suggested by Leventhal,

namely: 1) *identity* (i.e. symptoms associated with the illness), 2) *timeline* (i.e. acute/chronic), 3) *causes*, 4) *consequences* (i.e. negative impact on life), and 5) *controllability* (i.e. perceived as preventable, curable or controllable) (Leventhal et al., 2003). These cognitive representations influence, coping strategies, emotional responses to the illness and illness-specific behaviours such as self-management (Dempster, McCorry, N., Brennan, Donnelly, Murray, Johnston, 2011; Petrie & Weinman, 2006). Finally, it is suggested that illness representations and/or coping strategies are amended as individuals appraise the effectiveness of specific action plans for controlling/curing their condition (Dempster et al., 2011; Leventhal et al., 2003). Therefore, it is suggested that self-regulation theory provides a useful framework to understand both treatment self-management and psychological adjustment to chronic health problems (Edgar & Skinner, 2003) (see Figure 1).

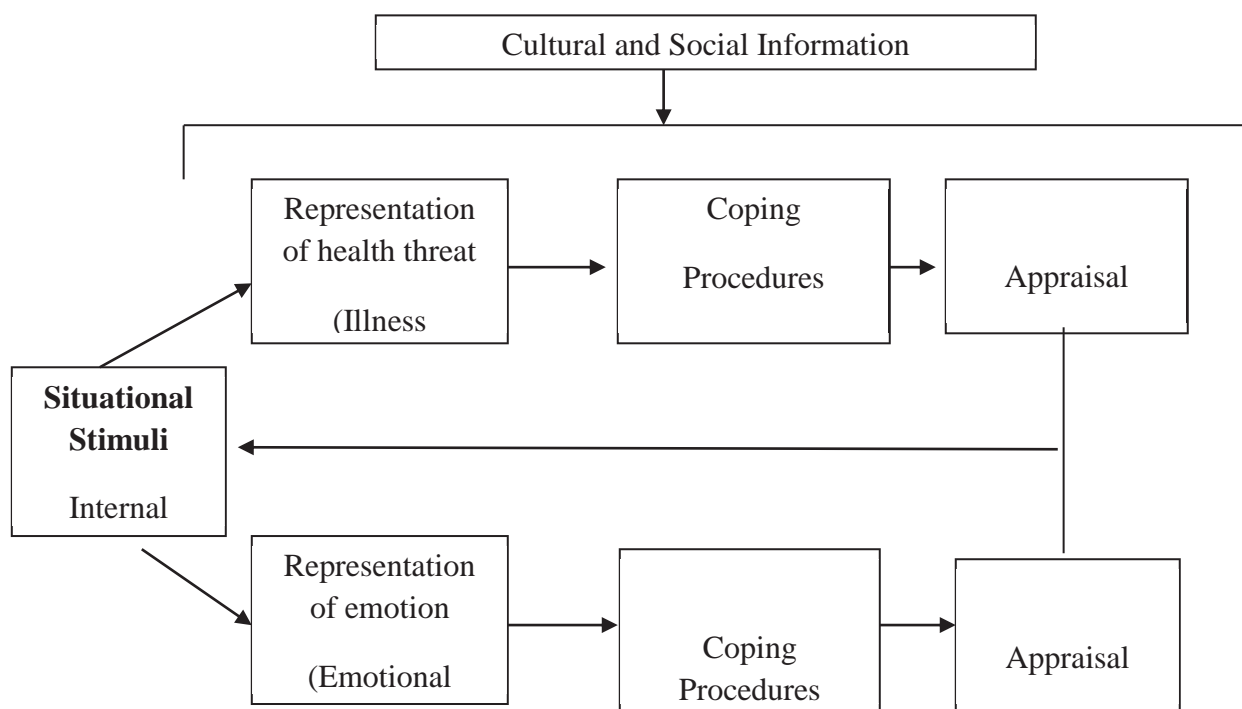


Figure 1: The common sense model (CSM) of self-regulation (based on Leventhal et al., 2003; Leventhal, Leventhal & Contrada, 1998)

The importance of theory-driven research is well recognised (Wallander, 1992). No published studies have specifically explored Leventhal's Self-regulatory theory in young people with CD. One recent study found that internal locus of control was associated with better dietary self-management in children with CD when compared to children with less internal locus of control (Bellini et al., 2011). Although internal locus of control is similar to the *personal control* domain of illness representations, the authors did not explicitly map internal locus of control onto Leventhal's illness representations.

In terms of other chronic illnesses, empirical support for self-regulation theory has been encouraging when applied to young people with diabetes, hypertension, and cystic fibrosis (Bucks et al., 2009; Griva, Myers & Newman, 2000; Nouwen, Law, Hussain, McGovern, & Napier, 2009; Skinner et al., 2003; Skinner & Hampson, 2001; Skinner, Hampson & Fife-Schaw, 2002; Skinner & Hampson, 1998; Zugelj et al., 2010). In particular, strong and consistent findings across studies have shown that *treatment control* beliefs are a significant predictor of dietary self-management in young people with diabetes (e.g. Nouwen et al., 2009; Skinner et al., 2003; Skinner & Hampson., 1998; Skinner et al, 2000; Skinner et al., 2002). In terms of well-being, links have been reported between perceived negative *consequences* of living with diabetes and anxiety (e.g. Law, Kelly, Huey, & Summerbell, 2002), positive well-being, depression, general well-being (e.g. Skinner et al., 2000) and diabetes distress (e.g. Nouwen et al., 2009).

### **Self-Efficacy**

Self-Efficacy is defined as a person's confidence in their own capabilities to carry out a particular behaviour necessary to produce a desired outcome (Bandura, 1997). Bandura (1997) proposes that this sense of personal control is important in order to manage chronic

health conditions successfully. More specifically, it is suggested that self-efficacy can influence whether individuals decide to change their health behaviours, their motivation to succeed, as well as their ability to cope with setbacks (Bandura, 1997). In addition to self-management, it is suggested that self-efficacy can impact on psychological well-being, such that those who believe in their own self-management abilities are more likely to have less emotional distress related to their condition (Bandura, 1997; Carr, 2006).

Studies have confirmed that young people with higher self-efficacy have better self-management of diabetes (e.g. Ott, Greening, Patardy, Holderby & DeBell, 2000) and asthma (e.g. Rhee, Belvea, Ciurzynski & Brasch, 2009). Similarly, a recent meta-analysis of adult studies concluded that self-management in diabetes was associated with higher self-efficacy (Gherman, Schnur, Montgomery, Sassu, Veresiu, & David, 2011).

### **Treatment Perceptions**

In addition to illness representations, a more detailed understanding of how people perceive their treatment regimens is warranted when exploring variation in self-management (Horne, 2003). Horne (2003) has explored perceptions of *necessity* (i.e. perceived personal need for treatment) and *concerns* about negative effects (i.e. belief about side effects of treatment and adverse effects of treatment on daily living). Horne (2003) makes a clear distinction between *treatment control* beliefs (measured by the IPQ-R) and *necessity* beliefs. He highlights that someone may believe that treatment is effective (*treatment control*) but yet not perceive a personal need for it (*necessity*) (Horne, 2003). Published studies have provided support for treatment perceptions. For example, self-management (i.e. chest physiotherapy and antibiotic use) was positively correlated with *necessity* beliefs in chronic health conditions (e.g. childhood cystic fibrosis; Bucks et al.,

2009), adults with haemophilia (Llewelyn, Miners, Lee, Harrington, Weinman, 2003) and adults with non-malignant chronic pain (Nicklas, Dunbar, & Wild, 2009). Similarly, *concerns* beliefs were negatively correlated with self-management (e.g. Horne & Weinman, 2002; Llewelyn, Miners, Lee, Harrington, Weinman, 2003; Nicklas, Dunbar, & Wild, 2009).

No published studies have used these theoretical frameworks to explore dietary self-management and well-being in young people with CD.

### **Parental Illness Representations**

It is recognised that parents play an important role in the development of their child's beliefs and self-management of treatment regimens (Eiser & Kopel, 2004). Researchers have found differences between adolescents' and their parent's illness representations in chronic illness (Salewski, 2003), diabetes (Law, 2002; Olsen, Berg, & Wiebe, 2008) and asthma (Yoos et al., 2007). However, the influence of this similarity or dissimilarity (in illness representations) on self-management and well-being had mixed results. Although reasons for a lack of consistency across studies are unclear, it has been suggested "dyadic perceptions" may be illness specific (Dempster et al., 2011). In terms of CD, a study in India showed that young people's self-management was better when parents had a good understanding of CD (Chauhan et al., 2010). However, comparisons between parent and child illness representations were not made. Taking this into consideration, research exploring parent and child illness representations in CD is warranted.

The current research aims to give us a better understanding of the beliefs young people and their parents have about CD. In turn, this may contribute to the development of interventions that could improve the management of CD (Leventhal et al., 2003).

The following hypotheses will be tested (see Table 1):

**Table 1:** Hypotheses

|   | <i><b>Hypothesis</b></i>  |
|---|---|
| 1 | Young People's illness representations ( <i>IPQ-R subscales</i> ) will be associated with self-reported dietary self-management ( <i>self-management: total score</i> ).  |
| 2 | Young People's illness representations ( <i>IPQ-R subscales</i> ) will be associated with well-being ( <i>KIDSCREEN: total score and SDQ: total score</i> ).  |
| 3 | Treatment perceptions ( <i>necessity and concerns</i> ) will be associated with dietary self-management ( <i>Self-Management: total score</i> ).  |
| 4 | Young People's dietary self-efficacy ( <i>Self-efficacy</i> ) will be associated with dietary self-management ( <i>SDQ: total score</i> ).  |
| 5 | Young People's dietary self-efficacy ( <i>Self-efficacy</i> ) will be associated with well-being ( <i>KIDSCREEN: total score</i> ).   |
| 6 | Incongruence of illness perceptions between young people and their parents is hypothesised to relate to the well-being of young people ( <i>KIDSCREEN: total score; SDQ: total score</i> ) and to parental well-being ( <i>DASS-21</i> ). |

## Method

### Participants

Young people aged 11-18 years old and their parents were recruited from three hospital CD out-patient clinics in England and Scotland. Age range of participants was guided by the measures used. Young people newly diagnosed with CD (in last six months), or with a co-morbid diagnosis of Diabetes, nut allergy or learning disability were excluded from the study, along with those not literate in English.

It is estimated that sixty-six young people were identified as potential participants across the three host sites. Forty (61% response rate) young people consented to take part in the study (9 boys, 31 girls) aged 11-17 years old (Mdn= 13.57, IQR=2.53). Thirty-four parents, aged 37- 65 years old (Mdn=44.38, IQR=8.88), completed questionnaires (8 men, 26 women). Of these participants, thirty-four were matched pairs (i.e. young person and their parent).

### *Age*

Young people who participated in the study were 11-17 years old (Mdn= 13.57, IQR=2.53). Parents' age range was 37-65 years old (Mdn=44.38, IQR=8.88).

### *Ethnicity*

Young people: 51.2% (n=21) described themselves as White-British, 43.9% (n=18) as Asian/British-Asian and 2.4% (n=1) as Black. Parents: 61.8% (n=21) were classified as White-British, 32.3% (n=11) as Asian or British-Asian and 2.9% (n=1) as Black.

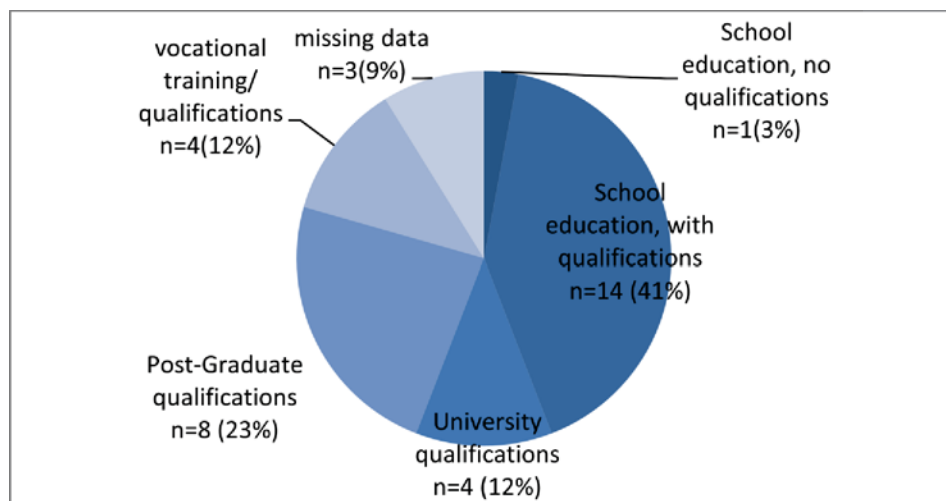


### *Parents' Marital Status*

The majority of parents were married (n=26; 78.8%). The remainder described themselves as single (n=2; 6.1%), cohabiting (n=2; 6.1%), divorced (n=2; 6.1%), or separated (n=1; 3%).

### *Parents' Education*

Parents' level of education varied across the sample (see Fig 2).



**Figure 2:** Parents' education

### *Parents' Occupation*

26.5% (n=9) of parents were in a professional occupation, 32.4% (n=11) managerial or technical, 8.8% (n=3) manual skilled, 5.9% (n=2) partly skilled and 11.8% (n=4) described themselves as home-makers.

### *Co-morbidity and Food Intolerances*

Co-morbid conditions were present in 25% of young people and included thyroid disease (n=1), asthma (n=3), heart murmur (n=1), enuresis (n=1), eczema (n=2), dyslexia (n=1) and Asperger's Syndrome (n=1). Similarly, the majority of young people did not have

other food intolerances (85.4%, n= 35), with a minority reporting intolerance to dairy (n=2), yeast (n=1), custard and eggs (n=1).

## **Design**

A cross-sectional questionnaire-based design was employed. The study was designed to explore illness representations, treatment perceptions, dietary self-efficacy, self-management and wellbeing in young people with CD. Additionally, comparisons of illness representations between young people and parents aimed to identify if dissimilarity (in illness representations) was related to well-being.

## **Ethical Approval**

The research study was approved by the National Research Ethics Service, Derbyshire Research Ethics Committee (appendix 12) and Research and Development departments from each of the three research sites (appendix 13).

## **Materials**

Young people and parents who consented to participate were asked to each complete a questionnaire pack. A summary of each questionnaire including subscales, exemplar items and guidelines for interpreting scores and psychometric properties of each questionnaire is provided below.

*The Revised Illness Perception Questionnaire (IPQ-R; Moss-Morris, Weinman, Petrie, Horne, Cameron & Buick, 2002)* was completed by young people and parents. *The young person version is a 57-item measure comprises 8 scales. The first scale (illness identity)*

asks participants to answer yes/no to experiencing 20 specific symptoms (e.g. tummy pain). Symptoms specific to CD were included in the *identity* scale. They were also asked to answer yes/no to whether they think each symptom is due to their illness. The other 7 scales ask participants to rate (on a 5-point Likert scale) the extent to which they agree or disagree with a range of statements. These scales are designed to measure the participants' illness perceptions in relation to *timeline- acute/chronic* (e.g. "My CD will last for a long-time"), *timeline-cyclical* (e.g. "Sometimes I have symptoms and sometimes they go away"), *consequences* (e.g. "My CD has a big impact on my life"), *personal-control* (e.g. "There is a lot I can do to control my symptoms"), *treatment-control* (e.g. "My gluten-free diet can control my CD"), *illness coherence* (e.g. "My CD doesn't make any sense to me") and their *emotional representations* (e.g. "My CD makes me feel angry"). The authors report good internal reliability, re-test reliability and predictive validity (Moss-Morris et al., 2002). Moreover, validation studies specifically applied to young people showed good internal consistency, and construct validity (Skinner et al., 2002). Eight additional items were added to the questionnaire to measure treatment perceptions related to a gluten-free diet, based on the Beliefs about Medicines Questionnaire (BMQ) developed by Horne et al. (1999). Horne, reports acceptable test-retest reliability (0.60-0.78) and internal consistency values in the *specific-necessity and concerns* subscales in diabetes (0.74 and 0.80).

A parental 76-item version of the IPQ-R was developed to measure parents' beliefs about their child's Coeliac Disease (9 scales). Sub-scales were designed to measure the participants' illness perceptions in relation to: *illness identity* (e.g. "Abdominal pain"), *causes* (e.g. "Hereditary – it runs in my family"), *timeline- acute/chronic* (e.g. "My child's CD will last for a long-time"), *timeline-cyclical* (e.g. "My child's CD does not make any

sense to me”), consequences (e.g. “My child’s CD has major consequences on my life”), personal control (e.g. “There is a lot which I can do to control my child’s symptoms”), treatment control (e.g. “My child’s gluten-free diet can control his or her CD”), illness coherence (e.g. “My child’s CD does not make any sense to me”), and their emotional representations (e.g. “My child’s CD makes me feel angry”). Guidance on how scores have been interpreted is provided in table 2.

**Table 2:** Interpreting IPQ-R scores

|                                  |  |
|----------------------------------|--|
| <i>Subscale</i>                  |  |
| <i>Identity</i>                  | Higher score is indicative of stronger illness identity                                    |
| <i>Timeline acute/chronic</i>    | Higher score is indicative of longer duration  |
| <i>Consequences</i>              | Higher score is indicative of more negative consequences                                   |
| <i>Personal Control</i>          | Higher score is indicative of more personal control  |
| <i>Treatment control</i>         | Higher score is indicative of more treatment control                                       |
| <i>Coherence</i>                 | Higher score is indicative of more puzzlement/confusion                                    |
| <i>Timeline Cyclical</i>         | Higher score is indicative of more the CD seems fluctuate                                  |
| <i>Causes</i>                    | Higher score (for each item) indicative of how extent each is believed to be a cause of CD |
| <i>Emotional Representations</i> | Higher score is indicative more negative emotions associated with CD                       |
| <i>Treatment Necessity</i>       | Higher score is indicative of greater perceived necessity for treatment                    |
| <i>Treatment Concerns</i>        | Higher score is indicative of more concern related to treatment                            |

*Self-efficacy for following your Gluten- Free Diet.* This dietary self-efficacy measure was based on previous literature (adapted from Senecal, Nouwen, & White, 2000). Further modifications were made following a pilot study (employing focus group methodology) to tailor the items more specifically to managing a gluten-free diet in young people with coeliac disease and their parents. The resulting measure is a 29-item questionnaire. Each

item asks participants to rate (on an 11-point Likert scale) how confident they are in their ability to manage their gluten-free diet in specific situations that are common barriers to dietary self-management (e.g. “*when I’m watching television at home*”).

*Strength and Difficulties Questionnaire* (SDQ; Goodman, 1997). The SDQ measures the psychological well-being of children and young people. The self-report questionnaire consists of five scales namely emotional symptoms, conduct problems, hyperactivity-inattention, peer problems and pro-social behaviour (5 items per scale). Participants indicate how much each item applies to them using a 3-point Likert scale from ‘not true’ to ‘certainly true’ (e.g., “*I worry a lot*”). In the parent version, participants indicate how much each item applies to their child using a 3-point Likert scale (e.g., “*Many worries, often seems worried*”). Higher scores, in both versions, indicate more difficulties. Reported psychometric properties of the SDQ include satisfactory reliability in a nationwide epidemiological sample, judged by internal consistency, inter-rater reliability and from test-retest stability (Goodman, 2001).

*KIDSCREEN -27: Health Questionnaire for Children and Young People – Child and Adolescent version* (The Kidscreen Group Europe, 2006). This standardized cross-national questionnaire measures health related quality of life (HRQoL) in children and young people. The 27-item measure comprises of 5 HRQoL scales: *Physical Wellbeing* (e.g., “*In general, how would you say your health is?*”), *Psychological Wellbeing* (e.g. “*Has your life been enjoyable?*”), *Autonomy & Parent Relations* (“*Have you had enough time for yourself?*”), *Social Support & Peers* (e.g. “*Have you spent enough time with your friends?*”), *School Environment* (e.g. “*Have you been happy at school*”). A higher total

score indicates better HRQoL. Internal consistency values were reported as 0.61-0.74 for different subscales of the questionnaire (Ravens-Sieberer et al., 2007).

*Depression Anxiety Stress Scale 21* (DASS 21; Lovibond & Lovibond, 1995). This is a 21-item questionnaire comprising of 3 scales measuring, *depression*, *stress*, and *anxiety*. Each item asks participants to rate (on a 4-point Likert scale) the extent to which a statement applied to them over the last week. Bieling, Cox, Murray, & Swinson (1998) report that the DASS 21 achieved good internal consistency and demonstrated good concurrent validity with the original 42 item DASS. Higher scores, across all three subscales, indicate greater levels of depression, stress and anxiety.

*About You.* Participants were asked for general demographic information together with information related to diagnosis and dietary self-management.

*About your gluten-free diet.* Young people are asked to rate (on a 5-point Likert scale) how often they have knowingly eaten foods containing gluten while at home and away from home (in the last two weeks and generally). They are also asked to rate (on a 5-point Likert scale) their concerns about accidentally eating gluten. Higher scores indicate poorer self-management and more concerns. Parents are asked the same questions about their child's gluten-free diet.

### **Internal Consistencies**

It is recognised that reliability depends on the questionnaire being used and the sample beginning tested (Streiner, 2003). Therefore, the internal consistency for each subscale was calculated (see Table 2). Results showed that Cronbach's alphas ranged between 0.52-0.97. Guidance on acceptability of alpha levels have been categorised with  $\geq 0.90$  described as excellent,  $\geq 0.80$  good,  $\geq 0.70$  acceptable,  $\geq 0.60$  questionable,  $\geq 0.50$  poor, and  $< 0.50$  unacceptable (George & Mallery, 2002). It was decided to include subscales rated as "questionable" or "poor" for exploratory purposes. However, they have been highlighted in Table 3 and are interpreted with caution.

**Table 3:** Internal Consistency Values

| <b>Young Person:<br/>IPQ-R</b>  | <b>Young Person:<br/>Cronbach's Alpha</b> | <b>Parent:<br/>IPQ-R</b>                                   | <b>Parent:<br/>Cronbach's Alpha</b> |
|---|---|--|-------------------------------------|
| Identity  | 0.84                                      | Identity   | 0.85                                |
| Timeline  | 0.79                                      | Timeline   | 0.77                                |
| Consequences  | 0.52                                      | Consequences   | 0.57                                |
| Personal Control  | 0.53                                      | Personal Control   | 0.68 (0.72 when item IP14 removed)  |
| Treatment Control   | 0.64                                      | Treatment Control  | 0.56                                |
| Illness Coherence   | 0.89                                      | Illness Coherence  | 0.80                                |
| Timeline Cyclical   | 0.78                                      | Timeline Cyclical  | 0.80                                |
| Emotional Representations   | 0.88                                      | Emotional Representations                                  | 0.40 (0.83 when item IP33 removed)  |
| Necessity   | 0.82                                      |  |                                     |
| Concerns  | 0.72                                      |  |                                     |
| <b>Young Person:<br/>Self-Efficacy</b>                                  | 0.97                                      |  |                                     |
| <b>Young Person:<br/>SDQ-total score<br/>(not including pro-social)</b> | 0.78                                      | <b>Parent: SDQ -total score (not including pro-social)</b> | 0.84                                |
| <b>Young Person:<br/>KIDSCREEN</b>                                      |   | <b>Parent: DASS</b>  |                                     |
| Physical Well-Being   | 0.86                                      | Depression   | 0.86                                |
| Psychological Well-being  | 0.79                                      | Anxiety  | 0.73                                |
| Autonomy & Parent Relations   | 0.86                                      | Stress   | 0.91                                |
| Social Support and Peers  | 0.88                                      |  |                                     |
| School Environment  | 0.77                                      |  |                                     |
| <b>Self-management</b>  | 0.89                                      | <b>Self-management</b>                                     | 0.92                                |

*Note: Highlighted values indicate 'poor' or 'questionable' internal consistency*



## **Procedure**

Methods of recruitment varied across host sites. A detailed account of the procedure for each site is provided in appendix 10. However, a general summary is given below:

A covering letter (appendix 9) and information sheets (appendix 6) were sent by the local coeliac care team to young people and parents who met inclusion criteria at least one week before their child's coeliac care review. Information sheets invited young people and their parents to take part in the research study. It also explained the rationale for the research, method, confidentiality, consent and their right to withdraw from the study at any point.

Young people and their parents were given the option to discuss the research in more detail with the chief investigator (Charlotte Tolgyesi) or principal investigators after their coeliac review appointments. Potential participants who made an informed choice to participate in the study were asked to sign a written consent form (appendix 8). If the young person was under 16 years of age, consent was also obtained from parents or from those in loco parentis.

Participants who had received a letter in the post before clinic were provided with the option to complete questionnaires either during the clinic or at home. Questionnaires took approximately 25-30 minutes to complete (appendix 5). The chief investigator and/or principal investigators were available to answer questions related to the study.

Participants who were unable to complete the questionnaires during the clinic, were able to complete the questionnaires at home and return them in a stamped addressed envelope to the University. Contact details of the researchers were provided to all participants should they have any questions related to the research.

Some young people did not attend clinic during the data collection phase of the study. These young people and parents were identified by the young person's coeliac medical care team and were sent a research pack by their Consultant Paediatric Gastroenterologist. The research pack contained a letter from their Consultant or Dietitian, participant information sheets (appendix 7), young person questionnaire booklet, parent questionnaire booklet and written consent forms. Those who decided to participate in the research were asked to post completed questionnaires and signed consent forms to the chief investigator using the envelopes provided.

## **Analysis**

Data were analysed using SPSS (PASW Statistics 18). Screening was conducted to check the data for errors. Kolmogorov-Smirnov was used to test for normality prior to the conducting of parametric or non-parametric analyses.

Descriptive statistical analysis was used to describe demographic characteristics of the sample as well as detailing frequencies, mean and standard deviations of predictor and outcome variables. Where data were not normally distributed, median and interquartile ranges are presented. Paired t-tests (or Wilcoxon tests) were used to compare scores between young people and their parent. Independent t-tests (or Mann-Whitney tests) were conducted to explore for gender differences (see appendix 11).

Bivariate correlational analysis (Pearson's  $r$  and Spearman's  $\rho$ ) explored relationships between illness representations, treatment perceptions, self-efficacy, well-being and dietary self-management in young people and their parents. Regression analyses were not conducted due to the small sample size and non-parametric data for some of the dependent variables, thus violating some of the assumptions for such analysis. Finally, where significant differences between young people and their parent's illness representations existed, "difference scores" were calculated by subtracting the parent score from the child score. Bivariate correlations were then completed between "difference scores" and variables assessing psychological well-being, in order to determine if incongruence between young people and their parent's beliefs were associated with well-being.

To avoid type 1 errors due to multiple correlations, the more conservative  $p$  value of 0.01 was used throughout.

## Results

### Hypothesis testing

Hypotheses were tested using bivariate correlations. Only results significant to 0.01 were included to reduce the likelihood of type-1 errors. In order to determine the combined effects of illness representations on self-management and well-being, a regression analysis was planned. According to Dancey and Reidy (1999) studies should have at least 15 participants per variable when using multiple regression. Taking this into consideration, the present study's sample size was not sufficient and would mean that results would not be generalizable and conclusions "invalid" (Dancey & Reidy, 1999). Therefore, multiple regression analyses were not used to test the following hypotheses.

#### *Hypothesis 1:*

*Young People's illness representations (IPQ-R subscales) will be associated with self-reported dietary self-management (Self-Management: total score).*

When self-management was correlated with illness representations, poorer *self-management (self-report)* was positively and moderately associated with young people's *timeline-cyclical* beliefs ( $r_s=0.47$ ,  $p<0.01$ ; 2-tailed). Therefore, those who believed that their condition fluctuated had poorer self-management. Dietary self-management was not associated with any other illness representations.

*Hypothesis 2:*

*Young People's illness representations (IPQ-R subscales) will be associated with well-being (KIDSCREEN: total score and SDQ: total score)*

Bivariate correlations were completed between measures of well-being in young people (i.e. KIDSCREEN total scores and SDQ total scores) and their illness representations (*IPQ-R subscales*) (see Table 4).

**Table 4:** Bivariate Correlations: Illness representations and well-being (young people)

| <b>Illness representation</b>    | <b>Psychological distress<br/>(SDQ :total score)</b> | <b>Positive Well-being<br/>(KIDSCREEN: total score)</b> |
|----------------------------------|--|---|
| <i>Identity</i>                  | <b>(.41**)</b>                                       | <b>(-.45**)</b>   |
| <i>Timeline acute-chronic</i>    | (-.12)   | (.10)   |
| <i>Consequences</i>              | .38  | -.32  |
| <i>Personal Control</i>          | .14  | -.22  |
| <i>Treatment Control</i>         | <b>(-.46**)</b>                                      | (.25)   |
| <i>Coherence</i>                 | <b>(.41**)</b>                                       | (-.32)  |
| <i>Timeline-cyclical</i>         | (.40)  | <b>(-.41**)</b>   |
| <i>Emotional Representations</i> | .31  | -.10  |

Note: \*\*p<0.01

Correlations using Spearman's rho displayed in brackets.

Stronger *illness identity* was associated with a greater psychological stress (*SDQ: total score*) and lower well-being (*KIDSCREEN: total score*). *Illness coherence* and lower perceived *treatment control* were also associated with higher levels of psychological stress (*SDQ: total score*). Finally, *timeline cyclical* beliefs were negatively correlated with well-being

(*KIDSCREEN: total score*). Other individual components of illness representations were not associated with well-being.

Overall, results provided partial support for the role of illness representations to well-being in young people. Bivariate correlations demonstrated moderate strength associations between four components of illness representations and well-being (*KIDSCREEN: total score*) and/or psychological distress (*SDQ: total score*). However, as the completion of regression analysis was not appropriate, the combined effects and unique contribution of each predictor variable could not be assessed.

### *Hypothesis 3:*

*Treatment necessity beliefs (necessity) and treatment concern beliefs (concerns) will be associated with dietary self-management (self-management: total score)*

*Necessity* beliefs were not significantly associated with *self-management* (self-report and parent-report (see Table 17). The *concerns* treatment beliefs subscale specifically assesses beliefs about the negative impact of a *gluten-free diet* on daily life. This is different to the *consequences* subscale of the IPQ-R, which assesses the perceived negative impact of CD on life. When the *concerns* subscale was correlated with dietary self-management, young people who report fewer *concerns* associated with managing a gluten-free diet reported better dietary *self-management* ( $r=0.43$ ,  $p<0.01$ ) (See Table 5).

**Table 5:** Bivariate correlations: *necessity* and *concerns* for treatment and self-management.

| Variable  |    | “Necessity” for treatment       |                       | “Concerns” for treatment        |                       |
|---|----|---------------------------------|-----------------------|---------------------------------|-----------------------|
|   |    | <i>Correlation co-efficient</i> | <i>Sig (2-tailed)</i> | <i>Correlation co-efficient</i> | <i>Sig (2-tailed)</i> |
| <b>Self-Management (self-report)</b><br><i>(Spearman’s rho)</i>   | 39 | -0.21                           | 0.209                 | <b>0.43**</b>                   | 0.006                 |
| <b>Self-Management (parent-report)</b><br><i>(Spearman’s rho)</i> | 34 | -0.21                           | 0.225                 | 0.22                            | 0.222                 |

Note: \*\*  $p < 0.01$

Overall, these results only partially support the hypothesis. Although, *necessity* beliefs were not related to *self-management*, findings indicated that young people with fewer *concerns* related to self-management were more likely to have better self-management (when measured by self-report).

#### *Hypothesis 4:*

*Young people’s dietary self-efficacy will be associated with dietary self-management (self-management: self-report).*

Bivariate correlations showed that *self-efficacy* was strongly correlated with *self-management* ( $r_s = -0.72$ ,  $p < 0.001$ ). Therefore, those with high levels of self-efficacy were more likely to have better self-management.

*Hypothesis 5:*

*Young people's dietary self-efficacy will be associated well-being (KIDSCREEN: total score & SDQ: total score).*

*Self-efficacy* was moderately associated with well-being measured by *SDQ (self-report): total score* ( $r_s = -0.54$   $p < 0.01$ ) and *KIDSCREEN: total score* ( $r_s = 0.50$ ;  $p < 0.01$ ). More specifically, young people's *self-efficacy* was associated with higher levels of *physical wellbeing* ( $r_s = -0.42$ ,  $p < 0.01$ ), *psychological well-being* ( $r_s = -0.50$ ,  $p < 0.01$ ) and *parent relations and autonomy* ( $r_s = 0.42$ ,  $p < 0.01$ ) (see Table 6).

**Table 6:** Bivariate correlations: self-efficacy and well-being.

|   | <b>Young people: Self-Efficacy</b><br>(Spearman's rho) |                                 |                       |
|---|--|---------------------------------|-----------------------|
|   | <i>N</i>   | <i>Correlation Co-efficient</i> | <i>Sig (2-tailed)</i> |
| <b>SDQ: Total Score (self report)</b>           | 38   | <b>-0.54**</b>                  | 0.002                 |
| <b>SDQ : Total score (parent report)</b>        | 33   | -.38                            | 0.31                  |
| <b>KIDSCREEN: Total Score</b>                   | 39   | <b>0.50**</b>                   | 0.001                 |
| <b>KIDSCREEN: Physical well-being</b>           | 39   | <b>-0.42**</b>                  | 0.001                 |
| <b>KIDSCREEN: Psychological well-being</b>      | 39   | <b>-0.50**</b>                  | 0.001                 |
| <b>KIDSCREEN: Parent relations and autonomy</b> | 39   | <b>0.42**</b>                   | 0.008                 |
| <b>KIDSCREEN: Social Support</b>                | 38   | 0.15                            | 0.372                 |
| <b>KIDSCREEN: School</b>                        | 37   | 0.32                            | 0.056                 |

\*\*  $p < 0.01$

Overall, results provided strong support for hypothesis 5, in that young people with high levels of self-efficacy were more likely to have positive well-being and lower levels of self-reported psychological distress.



*Hypothesis 6:*

*Incongruence of illness perceptions (between young people and their parents) is hypothesised to relate to well-being (KIDSCREEN: total score, SDQ: total score, DASS-21).*

Young people's and their parents' illness perceptions were predominately similar with only *timeline-cyclical* beliefs significantly different ( $z=3.36$ ,  $p<0.01$ ), such that young people believed more in the cyclical nature of their CD (see Table 3). Given this, hypothesis 6 was tested by calculating the difference between young people and their parent's scores for *timeline-cyclical* beliefs. Bivariate correlations were then completed between this new 'difference' variable and psychological well-being (measured by the SDQ, KIDSCREEN & IPQ-R emotional representations).

Results indicated that incongruence between young people and their parent's *timeline-cyclical* beliefs were not related well-being in young people. However, incongruence between young people and their parents *time-line cyclical* beliefs were related to higher levels of *stress* in parents (measured by DASS: stress subscale) ( $r_s=0.54$ ;  $p<0.01$ ). This indicates that when young people perceive their CD to fluctuate more than their parents, their parents are more likely to experience stress.

In summary, *timeline-cyclical beliefs* and *treatment concerns* were associated with poorer *self-management*. In terms of well-being, young people with a stronger *illness identity* and who believed their condition fluctuated (*timeline-cyclical*) reported less positive well-being (*KIDSCREEN: total score*) and higher levels of psychological distress (*SDQ: total score*).

Whereas psychological distress (*SDQ: total score*) was associated with less *treatment control*, more puzzlement (*coherence*) and a strong *illness identity*. Contrary to empirical literature, *treatment control* and *consequences* were not related to self-management and well-being respectively.

In terms of self-efficacy, young people with high levels of self-efficacy were more likely to have better self-management and positive well-being (*KIDCSREEN: total score*). Finally, incongruence in *timeline-cyclical* beliefs in young people and their parents was related to higher parental stress (*DASS: stress subscale*)

## Discussion

To our knowledge this is the first study exploring Leventhal's *illness representations*, Horne's *treatment perceptions* and Bandura's *self-efficacy* in a clinical sample of young people with CD. Furthermore, the inclusion of parental illness representations strengthens the research.

In the present study, dimensions of illness representations and self-efficacy were related to self-management and well-being. In terms of well-being, young people with a stronger *illness identity* and those who believed their condition fluctuated (*timeline-cyclical*) reported less positive well-being (*KIDSCREEN: total score*) and higher levels of psychological distress (*SDQ: total score*), whereas psychological distress (*SDQ: total score*) was associated with less *treatment control*, more puzzlement (*coherence*) and a strong *illness identity*. Poorer *self-management* was associated with *timeline-cyclical beliefs* and more *treatment concerns*. Higher levels of self-efficacy were related to better self-management and positive well-being (*KIDSCREEN: total score*). Finally, incongruence in *timeline-cyclical* beliefs in young people and their parents was related to higher parental stress (*DASS: Stress subscale*).

Findings of the present study are theoretically logical, although the extent that they are confirmed by published studies is mixed. For example, whilst *identity* and *treatment control* were related to well-being in a meta-analysis of adult studies (e.g. Hagger and Orbell, 2002), few studies have explored illness *coherence* and *timeline* cyclical beliefs (e.g. Law et al., 2002; Sawicki, Seller & Robinson, 2011). Possible reasons for this might be that these two subscales were not included in the original version of the illness perception questionnaire (Moss-Morris et al., 2002; Weinman et al., 2006) and low internal consistency values when

they have been measured (e.g. Bucks et al., 2009). Taking this into consideration, future research should seek to determine the role of *timeline-cyclical* beliefs and *coherence* in children with chronic illnesses.

Contrary to published empirical studies, *treatment control* and *consequences* dimensions were not significantly related to dietary self-management and well-being respectively in the present study (e.g. Law, Kelly, Huey, & Summerbell, 2002; Nouwen et al., 2009; Skinner et al., 2003). It is argued that *treatment control* and *consequences* beliefs should not be completely discounted in CD, but rather further research should seek to distinguish between short-term and long-term beliefs (Skinner & Hampson, 2001). This argument is supported by published research indicating that perceived effectiveness of treatment to control diabetes in the short-term (*short-term treatment control*) has been correlated with better dietary self-management in diabetes (e.g. Nouwen et al., 2009; Skinner & Hampson, 1998). In contrast, beliefs about the prevention of complications (*long-term treatment control*) were not predictive of self-management in diabetes (Skinner & Hampson, 2001). Similar distinctions have been made for short-term and long-term *consequences* beliefs (Skinner & Hampson 2001). Furthermore, psychometric issues associated with the *treatment-control* and *consequences* subscales of the IPQ-R in this study provides additional support for the development or adaptation of a questionnaire distinguishing between short-term and long-term beliefs in CD.

In the present study, illness and treatment perceptions were considered in isolation. However, it has been suggested that *timeline cyclical* beliefs and *necessity* beliefs may be inter-related (Horne and Weinman, 2002). It is therefore suggested that future research should explore the joint effects of treatment and illness perceptions on self-management.

Self-efficacy has been consistently correlated with dietary self-management (Griva, Myers & Newman, 2000; Iannotti et al., 2006; Nouwen et al., 2009) and well-being (Nouwen et al., 2009) in young people with diabetes. Similarly, *concern* beliefs have been negatively related to preventer medication in adults with asthma (Horne & Weinman, 2002). These studies provide strong support for findings related to CD in the present study.

The present study attempted to obtain an understanding of the role of parents' beliefs in the process of self-management. Previous research has reported differences between parents and young people's illness representations (i.e. perceived *consequences* and *emotional representations*) in diabetes (Law, 2002; Olsen, Berg & Wiebe, 2008). These findings were not consistent with the current study, but rather *timeline-cyclical* beliefs (i.e. belief that CD comes and goes in cycles) were significantly different in young people and parents. It is recognised that inconsistencies may be due to a series of different factors (e.g. specific characteristics of each condition, low internal consistencies of consequences scale). However, replication with a larger sample size and also across other chronic illnesses is needed before conclusions can be made. Similar to findings published by Olsen et al. (2008), the present study indicated that dissimilarity between the beliefs of young people and their parents was not related to the emotional well-being of young people. In contrast, where dissimilarity did exist in the present study (i.e. *timeline-cyclical* beliefs), parents reported higher levels of *stress*.

## Limitations & Future Research

Limitations of the study need to be highlighted. Firstly, low internal consistency values of some predictor variables (i.e. *IPQ-R: consequences, treatment control, personal control*), impacted on the reliability of results including these subscales, as previously highlighted.

Secondly, the cross-sectional nature of this research means that the directional nature of relationships between variables cannot be confirmed. It might be that *timeline-cyclical beliefs, self-efficacy* and *treatment concerns* have a direct influence on self-management. However, it is also conceivable that young people who are managing their condition well, may in turn, believe their condition is relatively stable, have fewer concerns about the treatment on daily life, and feel more confident in their ability for self-management. Moreover, illness representations, treatment perceptions, self-efficacy and self-management could influence each other. Longitudinal designs should also seek to determine whether coping mediates the relationships between beliefs and outcome (Hagger & Orbell, 2003).

The third issue relates to sample size. Despite including multiple recruitment sites, the sample size was smaller than anticipated. Issues such as high rates of co-morbidity and the presence of nut allergies in young people attending clinic impacted on the sample size. This means that generalizability of the results is limited and also more complex analyses could not be completed (e.g. multiple regression, path analysis). Finally, sample biases also exist because participants needed to be English literate to complete questionnaires and many families attending one clinic in particular did not have English as a first language.

Although it is recognised that the current study has a number of limitations, there are also strengths, such as the inclusion of parental illness representations, measuring self-efficacy and illness representations within the same study, the inclusion of treatment perceptions (i.e.

necessity and concerns) as well as obtaining multiple informants of self-management and well-being (i.e. young person and their parents).

Future research using a longitudinal research design would help to establish the causal nature of illness representations, coping and outcomes. It would also provide an opportunity to monitor how illness and treatment perceptions change over time and the impact that this change has on self-management and well-being. It is also suggested that a larger sample size (including both younger and older children and their parents) would mean that comparisons between younger and older children could be made. This distinction between younger and older children would also allow for exploration of how parent beliefs impact on younger children's self-management and well-being. Finally, a study focusing on illness representations, self-efficacy, self-management and well-being in young people with Diabetes and CD would be a logical next step in order to determine issues associated with self-management of co-morbid conditions. This is particularly crucial given the high rates of co-morbidity between Coeliac Disease and Diabetes.

### **Clinical Implications**

A collaborative approach between health professionals, young people and parents should aim to promote self-confidence in young people's own capabilities for self-management (Bandura, 1998). This should include a proactive approach involving clear planning, identifying young people's personal goals related to self-management of a gluten-free diet, monitoring, signposting young people to appropriate support and information (Health Delivery Directorate, Improvement and Support Team, 2009), developing individualized food plans

(Evert, 2004), educational-based interventions (Guevara, Wolf, Grum & Clark, 2003), as well as support to overcome temporary difficulties with self-management (Bandura, 1998).

It is important to highlight that the majority of young people with CD in the present study had good dietary self-management and well-being in the average range. However, the study has highlighted that some young people do have more difficulties. It is therefore suggested that screening in clinic for psychosocial factors that might contribute difficulties with self-management and well-being would be beneficial. In particular, young people who perceive that their CD fluctuates (*timeline-cyclical*), who have low *self-efficacy* and who perceive that the gluten-free diet has a negative impact on their life (*concerns*) may benefit from additional support with self-management. Seeking to understand young people's beliefs routinely in clinic could help to highlight potential barriers to self-management warranting further exploration and support.



## Conclusions

This cross-sectional questionnaire-based study was designed to contribute to our understanding of self-management and well-being in young people with CD. Overall, results of individual correlations indicate that illness representations (*timeline-cyclical, identity, treatment control, coherence*), *self-efficacy* and *treatment concerns* are important in relation to outcomes of young people with CD (i.e. self-management and/or well-being). It is suggested that future research should include the development of a validated questionnaire distinguishing between short-term and long-term illness representations (i.e. *treatment-control* and *consequences*) for young people CD with a larger sample size. Clinically, identifying young people who may be more likely to have difficulties with self-management and well-being would enable health care professionals to meet the needs of young people with CD and their parents.

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## **Executive Summary**

### **Psychosocial Factors associated with Self-Management and Well-being in Childhood Chronic Illness**

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School of Psychology  
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June 2011**

**Word Count: 902 (excluding references)**

## **Executive Summary**

Volume 1 details research completed, as part of a Doctorate in Clinical Psychology (ClinPsyD) at the University of Birmingham. Volume 1 is presented in two parts: 1) a review of existing literature explores how illness beliefs link to self-management in children and young people with chronic illness; 2) an empirical study investigates how illness beliefs, treatment beliefs and self-efficacy related to coping, well-being and self-management. Details of each paper are provided below:

### **1) Illness beliefs and self-management in children and young people with chronic illness:**

#### **A review of the literature.**

**Introduction:** There is a growing body of literature exploring how the beliefs young people have about their health condition impact on how they manage their treatment regimens. A Common-Sense Model of Illness Representations has provided a useful framework to describe how illness beliefs may impact on coping, self-management behaviours and well-being (Leventhal, Brissette, & Leventhal, 2003). However, no systematic reviews are available focusing exclusively on illness beliefs in children with chronic physical health conditions. The aim of the current literature review was to identify and evaluate empirical studies exploring dimensions of Leventhal's illness representations and self-management in children with a chronic health condition.

**Results:** Fourteen studies were identified for the review. Across the studies, reported findings between illness beliefs and self-management varied depending on the condition and particular aspect of a treatment regimen. However, most consistently, children and young people who believed their condition could be controlled by treatment (*treatment control*) reported better self-management. An evaluation of the studies highlighted that a lack of control groups and longitudinal research designs contributed to biases across studies.

**Discussion:** Understanding illness beliefs can potentially help health professionals to support children and young people with managing chronic health condition. In particular, seeking to understand how much children and young people believe their treatment regimen controls their condition might be useful if young people are finding it difficult to manage their gluten-free diet. Future research, with larger sample sizes across a range of health conditions would strengthen or contradict these findings. Furthermore, differentiating between short-term and long-term beliefs was highlighted as necessary, particularly in terms of consequences and controllability of chronic illness when exploring illness beliefs in children and young people.

## **2) Examining illness representations, treatment perceptions, and self-efficacy in relation to dietary self-management and well-being in young people with Coeliac Disease (CD) and their parents.**

**Introduction:** Coeliac Disease (CD) is a chronic auto-immune disease that is controlled through maintaining a lifelong gluten-free diet (Fasano & Catssi, 2005). Gluten is in food such as barley, wheat and rye (NICE, 2009). Published research studies suggest that 40-95% of

young people find it difficult to maintain a strict gluten-free diet (e.g. Kumar, Walter-Smith, Milla, Harris, Colyer, & Halliday, 1988; Rashid et al., 2005). In terms of quality of life research finding are quite mixed, with some studies reporting that young people with CD adapt well to their condition and others describing reduced quality of life and poorer psychological well-being in young people with the condition (e.g. Kolseren, Koopman, Schalekamp & Mearin, 2000, van Doorn, Winker, Mearin & Koopman, 2008). Whilst authors of studies have suggested ideas about what might impact on the self-management of treatment regimens or well-being, psychological mechanisms have not been investigated. Taking this into consideration, a research study was developed that explored two theories (Leventhal's Self-Regulation theory and Bandura's Self-Efficacy theory), previously used to understand dietary self-management and well-being in other chronic health conditions (Leventhal, Brissette & Leventhal, 2003; Bandura, 1997).

**Method:** Forty young people (11-18 years old) with CD and thirty-four parents (34 young person-parent dyads) took part in the study. Each person completed questionnaires exploring their beliefs about CD, beliefs about the gluten-free diet and their confidence to follow it, dietary self-management and well-being.

**Results:** Statistical analysis showed that young people who believed that their CD fluctuated (*timeline cyclical*) or were concerned about the negative impact that the gluten-free diet on their life (*concerns*), were less likely to report good dietary self-management.

The results also indicated that illness beliefs were related to well-being, such that young people with more positive well-being tended to feel more confident in their own ability

to stick to a gluten-free diet (*self-efficacy*) and believed that their CD was controlled by a gluten-free diet (*treatment control*). In contrast a strong *illness identity* (i.e. more symptoms associated with CD) and more puzzlement related to CD (*coherence*) were related to negative well-being.

Finally, young people and parental beliefs about CD were generally quite similar. However, young people tended to believe that CD fluctuated more than their parents thought (*timeline-cyclical*). This difference in beliefs was related to more parental stress.

**Limitations and Recommendations:** The study had a small sample size and did not include a follow-up. Therefore, it is not known the extent that particular illness and treatment beliefs contributed to differences in self-management and well-being. Moreover, it is unclear if illness and treatment beliefs impact on outcomes or vice versa. It is recommended that research completed in the future should explore the difference between short-term beliefs (i.e. impact of CD on daily life and extent that the gluten-free diet controls the condition) compared to more long-term beliefs (i.e. gluten-free diet to preventing complications and the long-term impact of CD). Published research in diabetes would suggest that this is an important distinction when seeking to understand the role of illness beliefs in children and young people (e.g. Skinner and Hampson, 2001).

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## **Appendices**

### **LITERATURE REVIEW**

#### **Appendix 1: Health Psychology Review: Instructions for Authors**

[not available in electronic copy of this thesis]

## **Appendix 2: Search criteria for literature review**

The following strategies were used:

*A:* Keyword search “illness representation\*” OR “illness perception\*” OR “illness belief\*”  
OR “personal model\*” OR “symptom representation\*” OR “treatment belief\*” OR  
“treatment perception\*” OR “self regulat\* model” OR “common sense model” OR “schema\*”

*B:* Keyword search “treatment compliance\*” OR “adherence” OR “self management” OR  
“self care” OR “health behavio\*”

*C:* Keyword search “Pediatric\*” OR “Paediatric\*” OR “Adolescen\*” OR “young people” OR  
young person” OR “youth” OR “child\*” OR “teenager\*”

*D:* Keyword Search “chronic illness\*” OR “chronic condition\*” OR coeliac OR celiac OR  
diabet\* OR asthma\* OR epilep\* OR “hypertensive” OR “renal” OR “cystic fibrosis” OR  
"HIV" OR "AIDS" OR "cardiovascular" OR cancer OR "chronic obstructive pulmonary\*" OR  
"haemophili\*" OR "sickle cell anaemia" OR thalasemia OR "bone marrow transplant".

*E:* Combine selections A **AND** B **AND** C **AND** D

Reference results = 85



### **Exclusion criteria**

Articles were excluded if they met the following criteria:

- a) Duplicates (17 excluded)
- b) Studies that do not include a sample of children or young people with a chronic physical health condition (36 excluded).
- c) Review articles i.e. not empirical studies (9)
- d) Studies that do not measure self-management (8 excluded).
- e) Articles not written in English (2 excluded)
- f) Studies that do not measure any of the dimensions of Leventhal's illness representations model (3 excluded).

**Remaining articles = 10**

Reference lists from published studies yielded 4 additional studies that met inclusion for review.

**Total articles for evaluation = 14 studies**

### **Appendix 3: Reference list of studies included in the literature review**

- Bucks, R. S., Hawkins, K., Skinner, T.C., Horn, S., Seddon, P., & Horne, R. (2009). Adherence to Treatment in Adolescents with Cystic Fibrosis: The role of illness perceptions and treatment beliefs. *Journal of Paediatric Psychology*, 34, 893-902.
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## **EMPIRICAL PAPER**

### **Appendix 4: Instructions for Authors: Journal of Health Psychology**

[not available in electronic copy of this thesis]

Date: 19/3/10  
Version 1



UNIVERSITY OF  
BIRMINGHAM

# How Does It Feel To Be Coeliac?

## Young Person Questionnaire Pack

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## The Illness Perception Questionnaire (IPQR)

### Your ideas and feelings about your Coeliac Disease (CD)

Listed below are a number of feelings or symptoms that you may or may not have had since being told you have CD. Please circle *Yes* or *No* to say whether you have had any of these symptoms and if you have, whether you think that these symptoms are caused by your CD.

|                         |  |  |  | I have experienced this symptom since my CD |    | This symptom is caused by my CD |  |
|-------------------------|--|--|--|---|----|---------------------------------|--|
|                         |  |  |  | Yes   | No |                                 |  |
| Tummy pain              |  |  |  | Yes   | No |                                 |  |
| Sore throat             |  |  |  | Yes   | No |                                 |  |
| Feeling sick            |  |  |  | Yes   | No |                                 |  |
| Weight loss             |  |  |  | Yes   | No |                                 |  |
| Feeling tired           |  |  |  | Yes   | No |                                 |  |
| Stiff joints            |  |  |  | Yes   | No |                                 |  |
| Sore eyes               |  |  |  | Yes   | No |                                 |  |
| Headaches               |  |  |  | Yes   | No |                                 |  |
| Upset tummy/ diarrhoea  |  |  |  | Yes   | No |                                 |  |
| Problems sleeping       |  |  |  | Yes   | No |                                 |  |
| Feeling dizzy           |  |  |  | Yes   | No |                                 |  |
| Feeling weak            |  |  |  | Yes   | No |                                 |  |
| Bloated tummy           |  |  |  | Yes   | No |                                 |  |
| Lots of wind (farting!) |  |  |  | Yes   | No |                                 |  |
| Feeling short of breath |  |  |  | Yes   | No |                                 |  |
| Constipation            |  |  |  | Yes   | No |                                 |  |
| Heartburn/ indigestion  |  |  |  | Yes   | No |                                 |  |
| Mouth ulcers            |  |  |  | Yes   | No |                                 |  |
| Feeling wheezy          |  |  |  | Yes   | No |                                 |  |
| Loss of hair            |  |  |  | Yes   | No |                                 |  |

## The Illness Perception Questionnaire (IPQR) continued

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

| Views about your CD                                       | Disagree<br>a lot | Disagree | Neither<br>agree nor<br>disagree | Agree | Agree<br>a lot |
|---|-------------------|----------|----------------------------------|-------|----------------|
| My CD will only last a short time.                        |                   |          |                                  |       |                |
| My CD is likely to last forever instead of going away     |                   |          |                                  |       |                |
| My CD will last for a long time.                          |                   |          |                                  |       |                |
| My CD will pass quickly.                                  |                   |          |                                  |       |                |
| I expect to have CD for the rest of my life.              |                   |          |                                  |       |                |
| My CD is a serious condition.                             |                   |          |                                  |       |                |
| My CD has a big impact on my life.                        |                   |          |                                  |       |                |
| My CD does not have much effect on my life.               |                   |          |                                  |       |                |
| My CD affects the way other people see me a lot.          |                   |          |                                  |       |                |
| My CD causes problems for my family and close friends.    |                   |          |                                  |       |                |
| There is a lot which I can do to control my symptoms.     |                   |          |                                  |       |                |
| What I do can control whether my CD gets better or worse. |                   |          |                                  |       |                |
| How CD makes me feel in the future depends on me.         |                   |          |                                  |       |                |
| Nothing I do will affect my CD.                           |                   |          |                                  |       |                |
| I have the power to change my CD.                         |                   |          |                                  |       |                |
| What I do will have no affect on the outcome of my CD.    |                   |          |                                  |       |                |
| My CD will improve in time.                               |                   |          |                                  |       |                |
| There is very little that can be done to improve my CD.   |                   |          |                                  |       |                |

## The Illness Perception Questionnaire (IPQR) continued

| Views about your CD  | Disagree<br>a lot | Disagree | Neither<br>agree nor<br>disagree | Agree | Agree<br>a lot |
|--|-------------------|----------|----------------------------------|-------|----------------|
| My gluten-free diet will be effective in curing my CD.                   |                   |          |                                  |       |                |
| The bad parts of my CD can be stopped (avoided) by my gluten-free diet.  |                   |          |                                  |       |                |
| My gluten-free diet can control my CD.                                   |                   |          |                                  |       |                |
| There is nothing that can help my CD.                                    |                   |          |                                  |       |                |
| The symptoms of CD are puzzling or confusing to me.                      |                   |          |                                  |       |                |
| My CD is a mystery to me.  |                   |          |                                  |       |                |
| I don't understand my CD.  |                   |          |                                  |       |                |
| My CD doesn't make any sense to me.                                      |                   |          |                                  |       |                |
| I have a clear picture or understanding of my CD.                        |                   |          |                                  |       |                |
| The symptoms of my CD change a lot from one day to the next.             |                   |          |                                  |       |                |
| Sometimes I have symptoms and sometimes they go away.                    |                   |          |                                  |       |                |
| I find it hard to know what is going to happen with my CD.               |                   |          |                                  |       |                |
| I go through times in which my CD gets better and then gets worse again. |                   |          |                                  |       |                |
| I feel sad when I think about my CD.                                     |                   |          |                                  |       |                |
| When I think about my CD I get upset.                                    |                   |          |                                  |       |                |
| My CD makes me feel angry.   |                   |          |                                  |       |                |
| My CD does not worry me.   |                   |          |                                  |       |                |
| Having CD makes me feel anxious or worried.                              |                   |          |                                  |       |                |
| My CD makes me feel scared or afraid.                                    |                   |          |                                  |       |                |



| Views about your gluten free diet                       | Disagree<br>a lot | Disagree | Neither<br>agree<br>nor dis-<br>agree | Agree | Agree<br>a lot |
|---|-------------------|----------|---------------------------------------|-------|----------------|
| Without my gluten free diet I would be very ill         |                   |          |                                       |       |                |
| My health, at present, depends on my gluten free diet   |                   |          |                                       |       |                |
| My life would be impossible without my gluten free diet |                   |          |                                       |       |                |
| My health in the future depends on my gluten free diet  |                   |          |                                       |       |                |
| My gluten free diet protects me from becoming worse     |                   |          |                                       |       |                |
| Having to stick to a gluten free diet worries me        |                   |          |                                       |       |                |
| My gluten free diet is a mystery to me                  |                   |          |                                       |       |                |
| My gluten-free diet disrupts my life                    |                   |          |                                       |       |                |

## Following Your Gluten-Free Diet for Coeliac Disease

Sometimes it's hard to stick to a gluten-free diet. This questionnaire lists some situations in which it might be difficult to stick to a gluten-free diet. We would like to know how sure you are that you would be able to stick to your gluten-free diet in these situations.

For each question, choose a number between 0 and 10 to show how sure you feel that you could stick to your gluten-free diet in that situation. Write the number you choose on the line next to the question. If you have never been in the situation described or you don't think that it applies to you, please write 'X' or 'N/A' instead of a number.

For example, **'Going to the cinema with friends'**

Imagine you are at the cinema with your friends. They buy lots of foods that are not gluten-free, like hotdogs and pick 'n' mix sweets. Think about how sure you are that you would not buy the same foods as your friends.

If you do not feel very sure that you would be able to stop yourself buying these foods your confidence score might be 2.

If you are very confident that you would always stick to your gluten-free diet in this situation, your confidence score would be 10.

| Confidence Scale   |   |   |   |   |                    |   |   |   |   |                    |
|--------------------|---|---|---|---|--------------------|---|---|---|---|--------------------|
| 0                  | 1 | 2 | 3 | 4 | 5                  | 6 | 7 | 8 | 9 | 10                 |
| Not at all<br>sure |   |   |   |   | Moderately<br>sure |   |   |   |   | Completely<br>sure |

Choose a number between 0 and 10 to show how sure you are that you could stick to your gluten-free diet...

|  | Confidence (0-10) |
|--|-------------------|
| When I'm watching television at home                       |                   |
| When I'm feeling tired or bored                            |                   |
| When I'm alone at home                                     |                   |
| When I'm feeling anxious, stressed or worried              |                   |
| When I see my friends eating non gluten-free foods         |                   |
| When I'm upset   |                   |
| When I'm eating out at my favourite restaurant             |                   |
| When feeling annoyed or angry                              |                   |
| When I'm out and about with my friends and get very hungry |                   |
| When I'm feeling sad                                       |                   |
| When I'm eating at school                                  |                   |

## Following A Gluten-Free Diet for Coeliac Disease continued

| Confidence Scale   |   |   |   |   |                    |   |   |   |   |                    |
|--------------------|---|---|---|---|--------------------|---|---|---|---|--------------------|
| 0                  | 1 | 2 | 3 | 4 | 5                  | 6 | 7 | 8 | 9 | 10                 |
| Not at all<br>sure |   |   |   |   | Moderately<br>sure |   |   |   |   | Completely<br>sure |

Choose a number between 0 and 10 to show how sure you are that you could stick to your gluten-free diet...

|  | Confidence (0-10) |
|--|-------------------|
| When I'm celebrating with others (e.g. at a birthday party)  |                   |
| When I'm preparing non gluten-free food for other people   |                   |
| When I'm eating out at a friend or relative's house  |                   |
| When I'm offered non gluten-free foods by my friends   |                   |
| When non gluten-free foods are available at home   |                   |
| When I'm eating out at an unknown restaurant   |                   |
| When I'm unwell  |                   |
| When I go to the tuck shop or corner shop and it doesn't have any gluten-free items available                                |                   |
| When I'm preparing my own food (such as breakfast or an after school snack)  |                   |
| When I'm faced with appealing foods that are not gluten-free in a supermarket, vending machine, or café                      |                   |
| When I'm feeling well (i.e. healthy, no symptoms)  |                   |
| When I'm on holiday abroad and eating in restaurants I don't know  |                   |
| When I'm craving foods containing gluten   |                   |
| When he/she wants more variety in their diet   |                   |
| When I'm away from home for a few days (i.e. at an activity camp or other school/college trip where your meals are provided) |                   |
| When I'm on the way to or from school, college or work   |                   |
| When I'm feeling happy   |                   |
| When he/she is not sure if something is gluten-free or not   |                   |

## 1. Physical Activities and Health

1. In general, how would you say your health is?

- ☐ excellent  
☐ very good  
☐ good  
☐ fair  
☐ poor

Thinking about the last week...

2. Have you felt fit and well?

| not at all            | slightly              | moderately            | very                  | extremely             |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

3. Have you been physically active (e. g. running, climbing, biking)?

| not at all            | slightly              | moderately            | very                  | extremely             |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

4. Have you been able to run well?

| not at all            | slightly              | moderately            | very                  | extremely             |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

Thinking about the last week...

5. Have you felt full of energy?

| never                 | seldom                | quite often           | very often            | always                |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

## 2. General Mood and Feelings about Yourself

Thinking about the last week...

1. Has your life been enjoyable?

| not at all            | slightly              | moderately            | very                  | extremely             |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

Thinking about the last week...

2. Have you been in a good mood?

| never                 | seldom                | quite often           | very often            | always                |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |

3. Have you had fun?

| never                 | seldom                | quite often           | very often            | always                |
|-----------------------|-----------------------|-----------------------|-----------------------|-----------------------|
| <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> | <input type="radio"/> |



| Thinking about the last week... |   | never                          | seldom                          | quite often                          | very often                          | always                          |
|---------------------------------|---|--------------------------------|---------------------------------|--------------------------------------|-------------------------------------|---------------------------------|
| 4.                              | Have you felt sad?  | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 5.                              | Have you felt so bad that you didn't want to do anything? | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 6.                              | Have you felt lonely?                                     | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 7.                              | Have you been happy with the way you are?                 | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |

### 3. Family and Free Time

| Thinking about the last week... |  | never                          | seldom                          | quite often                          | very often                          | always                          |
|---------------------------------|--|--------------------------------|---------------------------------|--------------------------------------|-------------------------------------|---------------------------------|
| 1.                              | Have you had enough time for yourself?                                     | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 2.                              | Have you been able to do the things that you want to do in your free time? | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 3.                              | Have your parent(s) had enough time for you?                               | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 4.                              | Have your parent(s) treated you fairly?                                    | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 5.                              | Have you been able to talk to your parent(s) when you wanted to?           | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 6.                              | Have you had enough money to do the same things as your friends?           | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 7.                              | Have you had enough money for your expenses?                               | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |

#### 4. Friends

| Thinking about the last week... |  | never                          | seldom                          | quite often                          | very often                          | always                          |
|---------------------------------|--|--------------------------------|---------------------------------|--------------------------------------|-------------------------------------|---------------------------------|
| 1.                              | Have you spent time with your friends?       | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 2.                              | Have you had fun with your friends?          | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | Always<br><input type="radio"/> |
| 3.                              | Have you and your friends helped each other? | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 4.                              | Have you been able to rely on your friends?  | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |

#### 5. School and Learning

| Thinking about the last week... |                                 | not at all                          | slightly                          | moderately                          | very                          | extremely                          |
|---------------------------------|---------------------------------|-------------------------------------|-----------------------------------|-------------------------------------|-------------------------------|------------------------------------|
| 1.                              | Have you been happy at school?  | not at all<br><input type="radio"/> | slightly<br><input type="radio"/> | moderately<br><input type="radio"/> | very<br><input type="radio"/> | extremely<br><input type="radio"/> |
| 2.                              | Have you got on well at school? | not at all<br><input type="radio"/> | slightly<br><input type="radio"/> | moderately<br><input type="radio"/> | very<br><input type="radio"/> | extremely<br><input type="radio"/> |

| Thinking about the last week... |   | never                          | seldom                          | quite often                          | very often                          | always                          |
|---------------------------------|---|--------------------------------|---------------------------------|--------------------------------------|-------------------------------------|---------------------------------|
| 3.                              | Have you been able to pay attention?        | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |
| 4.                              | Have you got along well with your teachers? | never<br><input type="radio"/> | seldom<br><input type="radio"/> | quite often<br><input type="radio"/> | very often<br><input type="radio"/> | always<br><input type="radio"/> |

[Strength and Difficulties Questionnaire not available in electronic copy of this thesis]

## About You

You're nearly finished! The last few questions are about you.

Are you male or female?

What is your date of birth?

|                       |        |  |
|-----------------------|--------|--|
| <input type="radio"/> | male   |  |
| <input type="radio"/> | female |  |

**Do you live with your parent/carer or independently?**

- ☐ Parent/carer
- ☐ Independently

Which ethnic group do you belong to?

|                       |               |  |  |                       |                        |
|-----------------------|---------------|--|--|-----------------------|------------------------|
| <input type="radio"/> | White British |  |  | <input type="radio"/> | Chinese                |
| <input type="radio"/> | White other   |  |  | <input type="radio"/> | Mixed—White & Asian    |
| <input type="radio"/> | Asian         |  |  | <input type="radio"/> | Mixed—White & Black    |
| <input type="radio"/> | Black         |  |  | <input type="radio"/> | Other mixed background |

Other (please specify)

|  |
|--|
|  |
|--|

### When were you diagnosed with Coeliac Disease?

|  |
|--|
|  |
|--|

### How were you diagnosed with Coeliac Disease?

|                       |   |  |  |  |  |  |  |
|-----------------------|---|--|--|--|--|--|--|
| <input type="radio"/> | I had an intestinal biopsy and blood test |  |  |  |  |  |  |
| <input type="radio"/> | I had just an intestinal biopsy           |  |  |  |  |  |  |
| <input type="radio"/> | I had just a blood test                   |  |  |  |  |  |  |
| <input type="radio"/> | I had no tests                            |  |  |  |  |  |  |

Other (please specify)

|  |
|--|
|  |
|--|



## About You continued

**Do you have any other long term health conditions? (please tick all that apply)**

|  |  |
|--|--|
| <input type="radio"/> Type 1 diabetes mellitus | <input type="radio"/> Type 2 diabetes mellitus |
| <input type="radio"/> Thyroid disease          | <input type="radio"/> Asthma                   |
| <input type="radio"/> Heart disease            | <input type="radio"/> No other condition       |

Other (please specify)

**Do you have any other food intolerances? (please tick all that apply)**

|                                       |  |
|---------------------------------------|--|
| <input type="radio"/> Dairy (lactose) | <input type="radio"/> Fructose             |
| <input type="radio"/> Alcohol         | <input type="radio"/> Yeast                |
| <input type="radio"/> Caffeine        | <input type="radio"/> No other intolerance |

Other (please specify)

**When did you last see a dietician about your Coeliac Disease?**

**When was your last antibody blood test?**

**What was the result of the test?**

|                                |  |
|--------------------------------|--|
| <input type="radio"/> Normal   | <input type="radio"/> I can't remember           |
| <input type="radio"/> Abnormal | <input type="radio"/> I haven't had a blood test |

## About You continued

| In the last two weeks...  | Never | Once or twice | A few times | Daily | All the time |
|---|-------|---------------|-------------|-------|--------------|
| How often have you knowingly eaten foods containing gluten while at home?       |       |               |             |       |              |
| How often have you knowingly eaten foods containing gluten when away from home? |       |               |             |       |              |

| In general...   | Extremely well | Well | Quite well | Not very well | Not at all |
|---|----------------|------|------------|---------------|------------|
| How well do you stick to your gluten-free diet when you are at home?        |                |      |            |               |            |
| How well do you stick to your gluten-free diet when you are away from home? |                |      |            |               |            |

| In general...   | Extremely worried | Very worried | Quite worried | A little worried | Not worried at all |
|---|-------------------|--------------|---------------|------------------|--------------------|
| How worried are you about accidentally eating gluten? |                   |              |               |                  |                    |

| In general...  | Extremely harmful | Very harmful | Quite harmful | A little harmful | Not at all harmful |
|--|-------------------|--------------|---------------|------------------|--------------------|
| How harmful to your health do you think accidentally eating gluten is? |                   |              |               |                  |                    |

**Thank You!**

Date: 19/3/10  
Version 1



UNIVERSITY OF  
BIRMINGHAM

# Psycho-Social Factors in Coeliac Disease

## Parent Questionnaire Pack

### Research Team:

- Dr Ruth Howard Clinical Psychologist
- Dr Gary Law Clinical Psychologist
- Charlotte Tolgyesi Trainee Clinical Psychologist

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## My child's CD and me

### Your views about your child's Coeliac Disease (CD)

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

|                          |  |  |  | My child has experienced this symptom since diagnosis |    | This symptom is related to their CD |  |
|--------------------------|--|--|--|---|----|-------------------------------------|--|
|                          |  |  |  | Yes   | No |                                     |  |
| Abdominal pain           |  |  |  | Yes   | No |                                     |  |
| Sore throat              |  |  |  | Yes   | No |                                     |  |
| Nausea                   |  |  |  | Yes   | No |                                     |  |
| Weight loss              |  |  |  | Yes   | No |                                     |  |
| Fatigue                  |  |  |  | Yes   | No |                                     |  |
| Stiff joints             |  |  |  | Yes   | No |                                     |  |
| Sore eyes                |  |  |  | Yes   | No |                                     |  |
| Headaches                |  |  |  | Yes   | No |                                     |  |
| Upset stomach/ diarrhoea |  |  |  | Yes   | No |                                     |  |
| Sleep difficulties       |  |  |  | Yes   | No |                                     |  |
| Dizziness                |  |  |  | Yes   | No |                                     |  |
| Loss of strength         |  |  |  | Yes   | No |                                     |  |
| Bloating                 |  |  |  | Yes   | No |                                     |  |
| Excessive wind           |  |  |  | Yes   | No |                                     |  |
| Breathlessness           |  |  |  | Yes   | No |                                     |  |
| Constipation             |  |  |  | Yes   | No |                                     |  |
| Heartburn/ indigestion   |  |  |  | Yes   | No |                                     |  |
| Mouth ulcers             |  |  |  | Yes   | No |                                     |  |
| Wheeziness               |  |  |  | Yes   | No |                                     |  |
| Hair loss                |  |  |  | Yes   | No |                                     |  |

## My child's CD and me

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

| My child's CD and me  | Disagree<br>a lot | Disagree | Neither<br>agree nor<br>disagree | Agree | Agree<br>a lot |
|---|-------------------|----------|----------------------------------|-------|----------------|
| My child's CD will last a short time.                               |                   |          |                                  |       |                |
| My child's CD is likely to be permanent rather than temporary.      |                   |          |                                  |       |                |
| My child's CD will last for a long time.                            |                   |          |                                  |       |                |
| My child's CD will pass quickly.                                    |                   |          |                                  |       |                |
| I expect my child to have CD for the rest of his or her life.       |                   |          |                                  |       |                |
| My child's CD is a serious condition.                               |                   |          |                                  |       |                |
| My child's CD has major consequences on my life.                    |                   |          |                                  |       |                |
| My child's CD does not have much effect on my life.                 |                   |          |                                  |       |                |
| My child's CD strongly affects the way others see me.               |                   |          |                                  |       |                |
| My child's CD has serious financial consequences.                   |                   |          |                                  |       |                |
| My child's CD causes difficulties for those who are close to me.    |                   |          |                                  |       |                |
| There is a lot which I can do to control my child's symptoms.       |                   |          |                                  |       |                |
| What I do can determine whether my child's CD gets better or worse. |                   |          |                                  |       |                |
| The course of my child's CD depends on me.                          |                   |          |                                  |       |                |
| Nothing I do will affect my child's CD.                             |                   |          |                                  |       |                |
| I have the power to influence my child's CD.                        |                   |          |                                  |       |                |
| My actions will have no effect on the outcome of my child's CD.     |                   |          |                                  |       |                |
| My child's CD will improve in time.                                 |                   |          |                                  |       |                |

## My child's CD and me

| My child's CD and me  | Disagree<br>a lot | Disagree | Neither<br>agree nor<br>disagree | Agree | Agree<br>a lot |
|---|-------------------|----------|----------------------------------|-------|----------------|
| There is very little that can be done to improve my child's CD.               |                   |          |                                  |       |                |
| My child's gluten-free diet will be effective in curing his or her CD.        |                   |          |                                  |       |                |
| The negative effects of my child's CD can be prevented (avoided) by the diet. |                   |          |                                  |       |                |
| My child's gluten-free diet can control his or her CD.                        |                   |          |                                  |       |                |
| There is nothing that can help my child's CD.                                 |                   |          |                                  |       |                |
| The symptoms of my child's CD are puzzling to me.                             |                   |          |                                  |       |                |
| My child's CD is a mystery to me.   |                   |          |                                  |       |                |
| I don't understand my child's CD.   |                   |          |                                  |       |                |
| My child's CD doesn't make any sense to me.                                   |                   |          |                                  |       |                |
| I have a clear picture or understanding of my child's CD.                     |                   |          |                                  |       |                |
| The symptoms of my child's CD change a great deal from day to day.            |                   |          |                                  |       |                |
| My child's symptoms come and go in cycles.                                    |                   |          |                                  |       |                |
| My child's CD is very unpredictable.  |                   |          |                                  |       |                |
| We go through cycles in which my child's CD gets better and worse.            |                   |          |                                  |       |                |
| I get depressed when I think about my child's CD.                             |                   |          |                                  |       |                |
| When I think about my child's CD I get upset.                                 |                   |          |                                  |       |                |
| My child's CD makes me feel angry.  |                   |          |                                  |       |                |
| My child's CD does not worry me.  |                   |          |                                  |       |                |
| My child having CD makes me feel anxious.                                     |                   |          |                                  |       |                |
| My child's CD makes me feel afraid.   |                   |          |                                  |       |                |



## My child's CD and me

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

| Possible Causes  | Strongly disagree | Disagree | Neither agree nor disagree | Agree | Strongly agree |
|--|-------------------|----------|----------------------------|-------|----------------|
| Stress or worry  |                   |          |                            |       |                |
| Hereditary— it runs in my family                             |                   |          |                            |       |                |
| A germ or virus  |                   |          |                            |       |                |
| Diet or eating habits  |                   |          |                            |       |                |
| Chance or bad luck   |                   |          |                            |       |                |
| Poor medical care  |                   |          |                            |       |                |
| Pollution in the environment                                 |                   |          |                            |       |                |
| My behaviour   |                   |          |                            |       |                |
| My mental attitude e.g. thinking about life negatively       |                   |          |                            |       |                |
| Family problems or worries                                   |                   |          |                            |       |                |
| Overwork   |                   |          |                            |       |                |
| My emotional state e.g. feeling down, lonely, anxious, empty |                   |          |                            |       |                |
| Ageing   |                   |          |                            |       |                |
| Alcohol  |                   |          |                            |       |                |
| Smoking  |                   |          |                            |       |                |
| Accident or injury   |                   |          |                            |       |                |
| My personality   |                   |          |                            |       |                |
| Altered immunity   |                   |          |                            |       |                |

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

The most important causes for me:

1. \_\_\_\_\_
2. \_\_\_\_\_
3. \_\_\_\_\_

[Strength and Difficulties Questionnaire not available in electronic copy of this thesis]



## DASS 21

Please read each statement and circle a number 0, 1, 2 or 3 which indicates how much the statement applied to you *over the past week*. There are no right or wrong answers. Do not spend too much time on any statement.

*The rating scale is as follows:*

- 0 Did not apply to me at all
- 1 Applied to me to some degree, or some of the time
- 2 Applied to me to a considerable degree, or a good part of the time
- 3 Applied to me very much, or most of the time

|  |   |   |   |   |
|--|---|---|---|---|
| I found it hard to wind down   | 0 | 1 | 2 | 3 |
| I was aware of dryness in my mouth   | 0 | 1 | 2 | 3 |
| I couldn't seem to experience any positive feeling at all  | 0 | 1 | 2 | 3 |
| I experienced breathing difficulty (eg, excessively rapid breathing, breathlessness in the absence of physical exertion)           | 0 | 1 | 2 | 3 |
| I found it difficult to work up the initiative to do things  | 0 | 1 | 2 | 3 |
| I tended to over-react to situations   | 0 | 1 | 2 | 3 |
| I experienced trembling (eg, in the hands)   | 0 | 1 | 2 | 3 |
| I felt that I was using a lot of nervous energy  | 0 | 1 | 2 | 3 |
| I was worried about situations in which I might panic and make a fool of myself  | 0 | 1 | 2 | 3 |
| I felt that I had nothing to look forward to   | 0 | 1 | 2 | 3 |
| I found myself getting agitated  | 0 | 1 | 2 | 3 |
| I found it difficult to relax  | 0 | 1 | 2 | 3 |
| I felt down-hearted and blue   | 0 | 1 | 2 | 3 |
| I was intolerant of anything that kept me from getting on with what I was doing  | 0 | 1 | 2 | 3 |
| I felt I was close to panic  | 0 | 1 | 2 | 3 |
| I was unable to become enthusiastic about anything   | 0 | 1 | 2 | 3 |
| I felt I wasn't worth much as a person   | 0 | 1 | 2 | 3 |
| I felt that I was rather touchy  | 0 | 1 | 2 | 3 |
| I was aware of the action of my heart in the absence of physical exertion (eg, sense of heart rate increase, heart missing a beat) | 0 | 1 | 2 | 3 |
| I felt that life was meaningless   | 0 | 1 | 2 | 3 |

## About You

Are you male or female?

|                       |        |  |  |
|-----------------------|--------|--|--|
| <input type="radio"/> | male   |  |  |
| <input type="radio"/> | female |  |  |

What is your date of birth?

|  |  |  |  |  |  |
|--|--|--|--|--|--|
|  |  |  |  |  |  |
|--|--|--|--|--|--|

Are you a member of Coeliac UK?

|                       |     |  |  |
|-----------------------|-----|--|--|
| <input type="radio"/> | yes |  |  |
| <input type="radio"/> | no  |  |  |

If yes, how long have you been a member?

|  |  |  |  |  |  |
|--|--|--|--|--|--|
|  |  |  |  |  |  |
|--|--|--|--|--|--|

What is your marital status?

|                       |            |  |  |
|-----------------------|------------|--|--|
| <input type="radio"/> | single     |  |  |
| <input type="radio"/> | cohabiting |  |  |
| <input type="radio"/> | married    |  |  |
| <input type="radio"/> | divorced   |  |  |
| <input type="radio"/> | separated  |  |  |
| <input type="radio"/> | widowed    |  |  |

What is your highest level of education?

|                       |                                      |
|-----------------------|--------------------------------------|
| <input type="radio"/> | School education, no qualifications  |
| <input type="radio"/> | School education with qualifications |
| <input type="radio"/> | University qualifications            |
| <input type="radio"/> | Postgraduate qualifications          |
| <input type="radio"/> | Vocational training/qualifications   |

What is or was your highest level of occupation?

|                       |                         |                       |                      |
|-----------------------|-------------------------|-----------------------|----------------------|
| <input type="radio"/> | Professional occupation | <input type="radio"/> | Partly skilled       |
| <input type="radio"/> | Managerial or technical | <input type="radio"/> | Unskilled occupation |
| <input type="radio"/> | Non-manual skilled      | <input type="radio"/> | Home-maker           |
| <input type="radio"/> | Manual skilled          |                       |                      |

Which ethnic group do you belong to?

|                       |               |                       |                        |
|-----------------------|---------------|-----------------------|------------------------|
| <input type="radio"/> | White British | <input type="radio"/> | Chinese                |
| <input type="radio"/> | White other   | <input type="radio"/> | Mixed—White & Asian    |
| <input type="radio"/> | Asian         | <input type="radio"/> | Mixed—White & Black    |
| <input type="radio"/> | Black         | <input type="radio"/> | Other mixed background |

Other (please specify)

|  |
|--|
|  |
|--|

## About Your Child

You're nearly there! The last few questions are about your child with CD.

Is your child male or female?

What is their date of birth?

|                       |        |  |
|-----------------------|--------|--|
| <input type="radio"/> | male   |  |
| <input type="radio"/> | female |  |

**Which ethnic group does your child belong to?**

|                       |               |                       |                        |
|-----------------------|---------------|-----------------------|------------------------|
| <input type="radio"/> | White British | <input type="radio"/> | Chinese                |
| <input type="radio"/> | White other   | <input type="radio"/> | Mixed—White & Asian    |
| <input type="radio"/> | Asian         | <input type="radio"/> | Mixed—White & Black    |
| <input type="radio"/> | Black         | <input type="radio"/> | Other mixed background |

Other (please specify)

|  |
|--|
|  |
|--|

**When were they diagnosed with Coeliac Disease?**

|  |
|--|
|  |
|--|

### How was your child diagnosed with Coeliac Disease?

|                       |  |  |  |  |  |  |  |
|-----------------------|--|--|--|--|--|--|--|
| <input type="radio"/> | Through an intestinal biopsy and blood test  |  |  |  |  |  |  |
| <input type="radio"/> | Through an intestinal biopsy alone   |  |  |  |  |  |  |
| <input type="radio"/> | Through a blood test alone   |  |  |  |  |  |  |
| <input type="radio"/> | I diagnosed them myself based on their symptoms and/or reaction to dietary changes |  |  |  |  |  |  |

Other (please specify)

|  |
|--|
|  |
|--|

**When did your child last see a dietitian about their Coeliac Disease?**

\_\_\_\_\_

## About Your Child continued

How often does your child see a dietician about their Coeliac Disease?

|   |  |
|---|--|
| <input type="radio"/> Every 3 months or more frequently | <input type="radio"/> Every 6 months                                       |
| <input type="radio"/> Every 12 months                   | <input type="radio"/> Every 2 years  |
| <input type="radio"/> Every 3 years or less frequently  | <input type="radio"/> I've never seen a dietician about my Coeliac Disease |

When was your child's last antibody blood test?

What was the result of the test?

|                                |  |
|--------------------------------|--|
| <input type="radio"/> Normal   | <input type="radio"/> Can't remember           |
| <input type="radio"/> Abnormal | <input type="radio"/> Haven't had a blood test |

Where you given a score (number) for the test? If so, can you remember what it was?

Does your child have any other long term health conditions? (select all that apply)

|  |  |
|--|--|
| <input type="radio"/> Type 1 diabetes mellitus | <input type="radio"/> Type 2 diabetes mellitus |
| <input type="radio"/> Thyroid disease          | <input type="radio"/> Asthma                   |
| <input type="radio"/> Heart disease            | <input type="radio"/> No other condition       |

Other (please specify)

Does your child have any other food intolerances? (select all that apply)

|                                       |  |
|---------------------------------------|--|
| <input type="radio"/> Dairy (lactose) | <input type="radio"/> Fructose             |
| <input type="radio"/> Alcohol         | <input type="radio"/> Yeast                |
| <input type="radio"/> Caffeine        | <input type="radio"/> No other intolerance |

Other (please specify)

## About Your Child's Gluten-Free Diet

| In the last two weeks...  | Never | Once or twice | A few times | Daily | All the time |
|---|-------|---------------|-------------|-------|--------------|
| How often has your child knowingly eaten foods containing gluten while at home?       |       |               |             |       |              |
| How often has your child knowingly eaten foods containing gluten when away from home? |       |               |             |       |              |

| In general...  | Extremely well | Well | Quite well | Not very well | Not at all |
|--|----------------|------|------------|---------------|------------|
| How well does your child stick to their gluten-free diet when they are at home?        |                |      |            |               |            |
| How well does your child stick to their gluten-free diet when they are away from home? |                |      |            |               |            |

| In general...  | Extremely concerned | Very concerned | Quite concerned | A little concerned | Not concerned at all |
|--|---------------------|----------------|-----------------|--------------------|----------------------|
| How concerned are you about accidental gluten-ingestion? |                     |                |                 |                    |                      |

| In general...   | Extremely harmful | Very harmful | Quite harmful | A little harmful | Not at all harmful |
|---|-------------------|--------------|---------------|------------------|--------------------|
| How harmful do you feel accidental gluten-exposure is to your child's health? |                   |              |               |                  |                    |

## And Finally...

I give my permission for the research team to contact me about this and future research projects. I understand that this does not obligate me to take part in any further research.

[illegible]

# Thank You !



**Appendix 6: Participant Information Sheets (Clinic)**

**Psychological Impact of Coeliac Disease in Young People**

**INFORMATION SHEET  
FOR YOUNG PEOPLE  
11-15 YEARS OLD**

We are asking if you would join in a research project to find out what it is like for young people who have Coeliac Disease and their parents. Before you decide if you would like to take part, it is important that you understand why the research is being done and what you will be asked to do. Please read the information in this leaflet carefully. You can talk to other people about the research before you decide what to do. People you could talk to might be your family, school teacher, doctor or friends.



You or your parents/carers can also telephone us if there is anything that is not clear or if you would like to have some more information.

**WHY ARE WE DOING THIS RESEARCH?**

This research study is being completed for a university doctorate (3 year course) at the University of Birmingham. Charlotte Tolgyesi is carrying out the research and she is supervised by Dr Ruth Howard and Dr Gary Law. They are both Clinical Psychologists who work at the University of Birmingham. This research study has also been reviewed by the Derbyshire Research Ethics Committee.



The aim of this research study is to find out what it is like for young people who have Coeliac Disease. We would like to find out what makes it hard to stick to a gluten free diet, what helps young people to cope with Coeliac Disease and what makes it more difficult. This is done by completing questionnaires.

## WHO CAN TAKE PART IN THIS RESEARCH?

Young people (aged 11-18 years old) with a diagnosis of Coeliac Disease who attend review appointments at hospital will be invited to take part in the study. We will also be asking parents/carers to take part in this research if they have a child with Coeliac Disease. We hope that about 150 young people and their parents will be able to take part in the study.

## WHAT HAPPENS IF I DECIDE TO TAKE PART?



If you and your parent/carer agree to take part, then you will both be asked to sign a consent (agreement) form at your next review appointment.

You will be asked to complete some questionnaires. The booklet of questionnaires will take 25-30 minutes to complete. The questionnaires will ask about your thoughts and feelings about Coeliac Disease, and how it affects you. You can either complete the questionnaires at your next review appointment or complete them at home. We will give you a stamped and addressed envelope so you can send them back to us. After you have completed the questionnaires we will only contact your parent/carer if we think that extra support might be helpful for you or if we have concerns about you.

If you would like a summary of the results of this study, then you can contact the research team or ask your coeliac care team at the hospital. The results will be available from October 2011.

## DO I HAVE TO TAKE PART?

No. It's you and your parents' choice if you want to take part in this study. If you decide to take part you will be free to stop taking part at any time during the research without giving a reason. If you do not want to carry on with the study, just tell a researcher or your doctor. You can also withdraw from the study after you have completed the questionnaires. If you decide to stop, this will not affect your medical care.

## WHO CAN I CONTACT IF I HAVE ANY WORRIES OR CONCERNS AFTER COMPLETING THE QUESTIONNAIRES?

You should speak to a member of your coeliac care team at the hospital if you have any worries or concerns. You can also contact your General Practitioner (GP) or Coeliac UK (tel: 0845 305 2060) who will be able to provide advice about where to access further information and/or support.

If you would like to make a complaint about the research study then you can contact the Patient and Advice Liaison Service (PALS) on *{insert number for local trust}*

### **WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?**

Information that could reveal your identity will be kept confidential (private) to the researchers, medical team and your GP. However, if there were worries about you or someone else then we would need to follow the appropriate guidelines and procedures

### **EXPENSES AND PAYMENTS**

You will not be given any money for taking part in the research. However, young people who complete the questionnaires will be entered into a free prize draw to win an IPOD shuffle. We will also provide a stamped addressed envelope for you to return completed questionnaires.

### **WHO CAN I CONTACT IF I HAVE ANY QUESTIONS ABOUT THIS STUDY?**

You may ask the study staff any questions you may have about this study at any time. If you would like to discuss any part of this research please contact:



**Name:** Charlotte Tolgyesi

**Email:** [REDACTED]



**Telephone:** [REDACTED]



**Post:** Charlotte Tolgyesi  
Trainee Clinical Psychologist  
School of Psychology  
University of Birmingham  
Edgbaston  
Birmingham  
B15 2TT

**OR**



**Name:** Dr Ruth Howard

**Email:** [REDACTED]



**Telephone:** [REDACTED]



**Post:** Dr Ruth Howard  
Clinical Director, ClinPsyD  
School of Psychology  
University of Birmingham  
Edgbaston  
Birmingham  
B15 2TT

Thank you for reading about this study. This is your copy of the information sheet.



## Psychological Impact of Coeliac Disease in Young People

### INFORMATION SHEET FOR YOUNG PEOPLE 16-18 YEARS OLD

We are asking if you would join in a research project to find out what it is like for young people who have Coeliac Disease and their parents. Before you decide if you would like to take part, it is important that you understand why the research is being done and what you will be asked to do. Please read the information in this leaflet carefully. You can talk to other people about the research before you decide what to do. People you could talk to might be your family, school teacher, doctor or friends.



You or your parents/carers can also telephone us if there is anything that is not clear or if you would like to have some more information.

#### WHY ARE WE DOING THIS RESEARCH?

This research study is being completed for a university doctorate (3 year course) at the University of Birmingham. Charlotte Tolgyesi is carrying out the research and she is supervised by Dr Ruth Howard and Dr Gary Law. They are both Clinical Psychologists who work at the University of Birmingham. This research study has also been reviewed by the Derbyshire Research Ethics Committee.



The aim of this research study is to find out what it is like for young people who have Coeliac Disease. We would like to find out what makes it hard to stick to a gluten free diet, what helps young people to cope with Coeliac Disease and what makes it more difficult. This is done by completing questionnaires.

#### WHO CAN TAKE PART IN THIS RESEARCH?

Young people (aged 11-18 years old) with a diagnosis of Coeliac Disease who attend review appointments at hospital will be invited to take part in the study. We will also be asking parents/carers to take part in this research if they have a child with Coeliac Disease. We hope that about 150 young people and their parents will be able to take part in the study.

## WHAT HAPPENS IF I DECIDE TO TAKE PART?



If you and your parent/carer agree to take part, then you will both be asked to sign a consent (agreement) form at your next review appointment.

You will be asked to complete some questionnaires. The booklet of questionnaires will take 25-30 minutes to complete. The questionnaires will ask about your thoughts and feelings about Coeliac Disease, and how it affects you. You can either complete the questionnaires at your next review appointment or complete them at home. We will give you a stamped and addressed envelope so you can send them back to us. After you have completed the questionnaires we will only contact you if we think that extra support might be helpful for you.

If you would like a summary of the results of this study, then you can contact the research team or ask your coeliac care team at the hospital. The results will be available from October 2011.

## DO I HAVE TO TAKE PART?

No. It's you and your parents' choice if you want to take part in this study. If you decide to take part you will be free to stop taking part at any time during the research without giving a reason. If you do not want to carry on with the study, just tell a researcher or your doctor. You can also withdraw from the study after you have completed the questionnaires. If you decide to stop, this will not affect your medical care.

## WHO CAN I CONTACT IF I HAVE ANY WORRIES OR CONCERNS AFTER COMPLETING THE QUESTIONNAIRES?

You should speak to a member of your coeliac care team at the hospital if you have any worries or concerns. You can also contact your General Practitioner (GP) or Coeliac UK (tel: 0845 305 2060) who will be able to provide advice about where to access further information and/or support.

If you would like to make a complaint about the research study then you can contact the Patient and Advice Liaison Service (PALS) on *{insert number for local trust}*

## WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?





Information that could reveal your identity will be kept confidential (private) to the researchers, medical team and your GP. However, if there were worries about you or someone else then we would need to follow the appropriate guidelines and procedures.

## EXPENSES AND PAYMENTS

You will not be given any money for taking part in the research. However, young people who complete the questionnaires will be entered into a free prize draw to win an IPOD shuffle. We will also provide a stamped addressed envelope for you to return completed questionnaires.

## WHO CAN I CONTACT IF I HAVE ANY QUESTIONS ABOUT THIS STUDY?

You may ask the study staff any questions you may have about this study at any time. If you would like to discuss any part of this research please contact:

 **Name:** Charlotte Tolgyesi  
 **Email:** [REDACTED]  
 **Telephone:** [REDACTED]  
 **Post:** Charlotte Tolgyesi  
Trainee Clinical Psychologist  
School of Psychology  
University of Birmingham  
[REDACTED]

OR

 **Name:** Dr Ruth Howard  
 **Email:** [REDACTED]  
 **Telephone:** [REDACTED]  
 **Post:** Dr Ruth Howard  
Clinical Director, ClinPsyD  
School of Psychology  
University of Birmingham  
[REDACTED]

Thank you for reading about this study. This is your copy of the information sheet.

## Psychological Impact of Coeliac Disease in Young People

### INFORMATION SHEET FOR PARENTS

We are asking if you and your child would join in a research project to find out what it is like for young people who have Coeliac Disease and their parents. Before you decide if you would like to take part, it is important that you understand why the research is being done and what you and your child will be asked to do. Please read the information in this leaflet carefully. You can talk to other people about the research before you decide what to do. You can also telephone us if there is anything that is not clear or if you would like to have some more information.

#### WHY ARE WE DOING THIS RESEARCH?

This research study is being completed for a university doctorate (3 year course) at the University of Birmingham. Charlotte Tolgyesi is carrying out the research and she is supervised by Dr Ruth Howard (Clinical Director), Dr Gary Law (Child Clinical Psychologist and Senior Academic Lecturer). They both work at the University of Birmingham. This research study has also been reviewed by the Derbyshire Research Ethics Committee

The aim of this research study is to find out what it is like for young people who have Coeliac Disease and their parents. We would like to find out what makes it hard to stick to a gluten free diet, what helps young people to cope with Coeliac Disease and what makes it more difficult. This is done by completing questionnaires.

#### WHO CAN TAKE PART IN THIS RESEARCH?

Young people (aged 11-18 years old) with a diagnosis of Coeliac Disease who attend review appointments at hospital will be invited to take part in the study. We will also be asking parents/carers to take part in this research if they have a child with Coeliac Disease. We hope that about 150 young people and their parents will be able to take part in the study.

#### WHAT HAPPENS IF I DECIDE TO TAKE PART?

If you and your child agree to take part then you will both be asked to sign a consent form (at your child's next medical review appointment). You will then be asked to complete a booklet of questionnaires. The questionnaires will take approximately 25-30 minutes to complete. The questionnaires will ask you about your thoughts and feelings about your child's condition and how it affects you both. Questionnaires will also ask your son/daughter about their own thoughts and feelings about being coeliac. You can either complete the questionnaires after

your child's next review appointment with his/her doctor or complete them at home. We will give you a stamped and addressed envelope so you can send them back to us. After you have completed the questionnaires we will only contact you if we think that extra support might be helpful for your child or if there are concerns about your child's safety. If your child is 16-18 years old then we will contact them directly.

### **DO I HAVE TO TAKE PART?**

No. It's your choice if you want to take part in this study. If you decide to take part you and your child will be free to stop taking part at any time during the research without giving a reason. If you do not want to carry on with the study, just tell the research staff or your child's doctor. You can also withdraw from the study after you have completed the questionnaires (until April 2011). If you decide that you don't want your questionnaires to be used in the research then you can contact us at the telephone number or address on this information sheet. We will destroy any completed questionnaires or other information that you have given by shredding them. If you decide to stop, this will not affect the medical care your son/daughter receives.

If completing the questionnaires causes any upset or concern for you or your child, then you should speak to a member of your child's coeliac care team at the hospital. You can also contact your General Practitioner (GP), who will be able to provide advice about where to access further support.

### **WHO CAN I CONTACT IF I HAVE ANY WORRIES OR CONCERNS AFTER COMPLETING THE QUESTIONNAIRES?**

You should speak to a member of your coeliac care team at the hospital if you have any worries or concerns. You can also contact your General Practitioner (GP) or Coeliac UK (tel: 0845 305 2060) who will be able to provide advice about where to access further information and/or support.

If you would like to make a complaint about the research study then you can contact the Patient and Advice Liaison Service (PALS) on *{insert number for local trust}*

### **WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?**

Information that could reveal your identity will be kept confidential to the researchers, medical team and GP. However, if there were worries about your child's safety or someone else's safety then we would need to follow local child protection procedures.

### **EXPENSES AND PAYMENTS**

You will not be given any money for taking part in the research. However, young people who complete the questionnaires will be entered into a free prize draw to win an IPOD shuffle. We will also provide a stamped addressed envelope for you to return completed questionnaires.

### WHAT WILL HAPPEN TO THE RESULTS OF THE RESEARCH STUDY?

Results of the research study will be written in a research thesis. Results will be completely anonymous. A hard bound copy of the research will be held in University of Birmingham library. Results of the research might also be published in an academic journal. However, no personal information will be published.

If you would like a summary of the results of this study, then you can contact the research team or ask your coeliac care team at the hospital. The results will be available from October 2011.

### WHO CAN I CONTACT IF I HAVE ANY QUESTIONS ABOUT THIS STUDY?

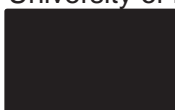
You may ask the study staff any questions you may have about this study at any time. If you would like to discuss any part of this research please contact:

**Name:** Charlotte Tolgyesi

**Email:** [REDACTED]

**Telephone:** [REDACTED]

**Post:** Charlotte Tolgyesi  
Trainee Clinical Psychologist  
School of Psychology  
University of Birmingham



**OR**

**Name:** Dr Ruth Howard

**Email:** [REDACTED]

**Telephone:** [REDACTED]

**Post:** Dr Ruth Howard  
Clinical Director, ClinPsyD  
School of Psychology



Thank you for reading about this study. This is your copy of the information sheet.

**Appendix 7: Participant Information Sheets (Home)**

**Psychological Impact of Coeliac Disease in Young People**

**INFORMATION SHEET  
FOR YOUNG PEOPLE  
11-15 YEARS**

We are asking if you would join in a research project to find out what it is like for young people who have Coeliac Disease. Before you decide if you would like to take part, it is important that you understand why the research is being done and what you will be asked to do. Please read the information in this leaflet carefully. You can talk to other people about the research before you decide what to do. People you could talk to might be your family, school teacher, doctor or friends.



You or your parents/carers can also telephone us if there is anything that is not clear or if you would like to have some more information.

**WHY ARE WE DOING THIS RESEARCH?**

This research study is being completed for a university doctorate (3 year course) at the University of Birmingham. Charlotte Tolgyesi is carrying out the research and she is supervised by Dr Ruth Howard and Dr Gary Law. They are both Clinical Psychologists who work at the University of Birmingham. This research study has also been reviewed by the Derbyshire Research Ethics Committee



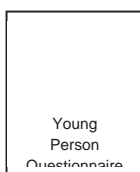
The aim of this research study is to find out what it is like for young people who have Coeliac Disease. We would like to find out what makes it hard to stick to a gluten free diet, what helps young people to cope with Coeliac Disease and what makes it more difficult. This is done by completing questionnaires.

**WHO CAN TAKE PART IN THIS RESEARCH?**

Young people (aged 11-18 years old) with a diagnosis of Coeliac Disease who attend review appointments at hospital will be invited to take part in the study. We will also be asking parents/carers to take part in this research if they have a child with Coeliac Disease. We hope that about 150 young people and their parents will be able to take part in the study.

### WHAT HAPPENS IF I DECIDE TO TAKE PART?

If you and your parent/carer agree to take part then please can you:



- Sign the consent (agreement) forms enclosed with your parent/carer.
- Complete the “Young Person Questionnaire Pack”. It will take 25-30 minutes to complete. The questionnaires will ask about your thoughts and feelings about Coeliac Disease, and how it affects you.
- Your parent/carer will be asked to complete the “Parent Questionnaire Pack”.
- Please can you and your parent post to us the signed consent forms with completed questionnaires in the stamped addressed envelope provided. You can keep this information sheet.
- After you have completed the questionnaires we will only contact your parent/carer if we think that extra support might be helpful for you or if we have concerns about you.

If you would like a summary of the results of this study, then you can contact the research team or ask your coeliac care team at the hospital. The results will be available from October 2011.

### DO I HAVE TO TAKE PART?

No. It's you and your parents' choice if you want to take part in this study. If you decide to take part, you will be free to stop taking part at any time during the research, without giving a reason. If you do not want to carry on with the study, just tell your parent, the researcher or your doctor. You can also withdraw from the study after you have completed the questionnaires. If you decide to stop, this will not affect your medical care.

If you have any worries or concerns after completing the questionnaire, then you should speak to a member of your coeliac care team at the hospital. You can also contact your General Practitioner (GP), who will be able to provide advice about where to access further support.

### WHO CAN I CONTACT IF I HAVE ANY WORRIES OR CONCERNS AFTER COMPLETING THE QUESTIONNAIRES?



You should speak to a member of your coeliac care team at the hospital if you have any worries or concerns. You can also contact your General Practitioner (GP) or Coeliac UK (tel: 0845 305 2060) who will be able to provide advice about where to access further information and/or support.

If you would like to make a complaint about the research study then you can contact the Patient and Advice Liaison Service (PALS) on *{insert number for local trust}*

### **WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?**

Information that could reveal your identity will be kept confidential (private) to the researchers, medical team and your GP. However, if there were worries about you or someone else then we would need to follow the appropriate guidelines and procedures.

### **EXPENSES AND PAYMENTS**

You will not be given any money for taking part in the research. However, young people who complete the questionnaires will be entered into a free prize draw to win an IPOD shuffle. We will also provide a stamped addressed envelope for you to return completed questionnaires.

### **WHO CAN I CONTACT IF I HAVE ANY QUESTIONS ABOUT THIS STUDY?**

You may ask the study staff any questions you may have about this study at any time. If you would like to discuss any part of this research please contact:

**Name:** Charlotte Tolgyesi



**Email:**



**Telephone:**



**Post:**

Charlotte Tolgyesi  
Trainee Clinical Psychologist  
School of Psychology  
University of Birmingham



**Name:** Dr Ruth Howard



**Email:** [REDACTED]



**Telephone:** [REDACTED]



**Post:** Dr Ruth Howard  
Clinical Director, ClinPsyD  
School of Psychology  
University of Birmingham



Thank you for reading about this study. This is your copy of the information sheet.

## Psychological Impact of Coeliac Disease in Young People

### INFORMATION SHEET FOR YOUNG PEOPLE 16-18 YEARS

We are asking if you would join in a research project to find out what it is like for young people who have Coeliac Disease. Before you decide if you would like to take part, it is important that you understand why the research is being done and what you will be asked to do. Please read the information in this leaflet carefully. You can talk to other people about the research before you decide what to do. People you could talk to might be your family, school teacher, doctor or friends.



You or your parents/carers can also telephone us if there is anything that is not clear or if you would like to have some more information.

### WHY ARE WE DOING THIS RESEARCH?

This research study is being completed for a university doctorate (3 year course) at the University of Birmingham. Charlotte Tolgyesi is carrying out the research and she is supervised by Dr Ruth Howard and Dr Gary Law. They are both Clinical Psychologists who work at the University of Birmingham. This research study has also been reviewed by the Derbyshire Research Ethics Committee.



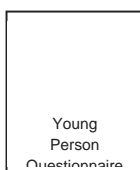
The aim of this research study is to find out what it is like for young people who have Coeliac Disease. We would like to find out what makes it hard to stick to a gluten free diet, what helps young people to cope with Coeliac Disease and what makes it more difficult. This is done by completing questionnaires.

### WHO CAN TAKE PART IN THIS RESEARCH?

Young people (aged 11-18 years old) with a diagnosis of Coeliac Disease who attend review appointments at hospital will be invited to take part in the study. We will also be asking parents/carers to take part in this research if they have a child with Coeliac Disease. We hope that about 150 young people and their parents will be able to take part in the study.

### WHAT HAPPENS IF I DECIDE TO TAKE PART?

If you and your parent/carer agree to take part then please can you:



- Sign the consent (agreement) forms enclosed with your parent/carer.
- Complete the “Young Person Questionnaire Pack”. It will take 25-30 minutes to complete. The questionnaires will ask about your thoughts and feelings about Coeliac Disease, and how it affects you.
- Your parent/carer will be asked to complete the “Parent Questionnaire Pack”.
- Please can you and your parent post to us the signed consent forms with completed questionnaires in the stamped addressed envelope provided. You can keep this information sheet.
- After you have completed the questionnaires we will only contact you if we think that extra support might be helpful for you or if we have concerns about you.

If you would like a summary of the results of this study, then you can contact the research team or ask your coeliac care team at the hospital. The results will be available from October 2011.

### DO I HAVE TO TAKE PART?

No. It's you and your parents' choice if you want to take part in this study. If you decide to take part, you will be free to stop taking part at any time during the research, without giving a reason. If you do not want to carry on with the study, just tell your parent, the researcher or your doctor. You can also withdraw from the study after you have completed the questionnaires. If you decide to stop, this will not affect your medical care.

If you have any worries or concerns after completing the questionnaire, then you should speak to a member of your coeliac care team at the hospital. You can also contact your General Practitioner (GP), who will be able to provide advice about where to access further support.

## WHO CAN I CONTACT IF I HAVE ANY WORRIES OR CONCERNS AFTER COMPLETING THE QUESTIONNAIRES?

You should speak to a member of your coeliac care team at the hospital if you have any worries or concerns. You can also contact your General Practitioner (GP) or Coeliac UK (tel: 0845 305 2060) who will be able to provide advice about where to access further information and/or support.

If you would like to make a complaint about the research study then you can contact the Patient and Advice Liaison Service (PALS) on *{insert number for local trust}*

## WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?





Information that could reveal your identity will be kept confidential (private) to the researchers, medical team and your GP. However, if there were worries about you or someone else then we would need to follow the appropriate guidelines and procedures.

## EXPENSES AND PAYMENTS

You will not be given any money for taking part in the research. However, young people who complete the questionnaires will be entered into a free prize draw to win an IPOD shuffle. We will also provide a stamped addressed envelope for you to return completed questionnaires.

## WHO CAN I CONTACT IF I HAVE ANY QUESTIONS ABOUT THIS STUDY?

You may ask the study staff any questions you may have about this study at any time. If you would like to discuss any part of this research please contact:

 **Name:** Charlotte Tolgyesi  
 **Email:** [REDACTED]  
 **Telephone:** [REDACTED]  
 **Post:** Charlotte Tolgyesi  
Trainee Clinical Psychologist  
School of Psychology  
University of Birmingham  
[REDACTED]

 **Name:** Dr Ruth Howard  
 **Email:** [REDACTED]



**Telephone:** [REDACTED]



**Post:** Dr Ruth Howard  
Clinical Director, ClinPsyD  
School of Psychology  
University of Birmingham



Thank you for reading about this study. This is your copy of the information sheet.

## **Psychological Impact of Coeliac Disease in Young People**

### **INFORMATION SHEET FOR PARENTS**

We are asking if you and your child would join in a research project to find out what it is like for young people who have Coeliac Disease and their parents. Before you decide if you would like to take part, it is important that you understand why the research is being done and what you will be asked to do. Please read the information in this leaflet carefully. You can talk to other people about the research before you decide what to do. You can also telephone us if there is anything that is not clear or if you would like to have some more information.

#### **WHY ARE WE DOING THIS RESEARCH?**

This research study is being completed for a university doctorate (3 year course) at the University of Birmingham. Charlotte Tolgyesi is carrying out the research and she is supervised by Dr Ruth Howard (Clinical Director), Dr Gary Law (Child Clinical Psychologist and Senior Academic Lecturer). They both work at the University of Birmingham. This research study has also been reviewed by the Derbyshire Research Ethics Committee.

The aim of this research study is to find out what it is like for young people who have Coeliac Disease and their parents. We would like to find out what makes it hard to stick to a gluten free diet, what helps young people to cope with Coeliac Disease and what makes it more difficult. This is done by completing questionnaires.

#### **WHO CAN TAKE PART IN THIS RESEARCH?**

Young people (aged 11-18 years old) with a diagnosis of Coeliac Disease who attend review appointments at hospital will be invited to take part in the study. We will also be asking parents/carers to take part in this research if they have a child with Coeliac Disease. We hope that about 150 young people and their parents will be able to take part in the study.

## **WHAT HAPPENS IF I DECIDE TO TAKE PART?**

If you and your child agree to take part then please can you both:

- Sign the consent (agreement) forms enclosed
- Complete the “Parent Questionnaire Pack”. It will take 25-30 minutes to complete. The questionnaires will ask you about your thoughts and feelings about your child’s condition and how it affects you both.
- Your child will be asked to complete the “Young Person Questionnaire Pack”. Questionnaires will ask your son/daughter about their own thoughts and feelings about being coeliac.
- Please can you post to us the signed consent forms with completed questionnaires in the stamped addressed envelope provided. You can keep this information sheet.
- After you have completed the questionnaires we will only contact you if we think that extra support might be helpful for your child or if there are concerns about your child’s safety. If you child is 16-18 years old then we will contact them directly.

If you would like a summary of the results of this study, then you can contact the research team or ask your coeliac care team at the hospital. The results will be available from October 2011.

## **DO I HAVE TO TAKE PART?**

No. It’s your choice if you want to take part in this study. If you decide to take part, you will be free to stop taking part at any time during the research, without giving a reason. If you do not want to carry on with the study, just tell the research staff or your child’s doctor. You can also withdraw from the study after you have completed the questionnaires (until April 2011). If you decide that don’t want your questionnaires to be used in the research then you can contact us at the telephone number or address on this information sheet. We will destroy any completed questionnaires or other information that you have given by shredding them. If you decide to stop, this will not affect the medical care you receive.

If completing the questionnaires causes any upset or concern for you or your child, then you should speak to a member of your child’s coeliac care team at the hospital. You can also contact your General Practitioner (GP), who will be able to provide advice about where to access further support.

## **WHO CAN I CONTACT IF I HAVE ANY WORRIES OR CONCERNS AFTER COMPLETING THE QUESTIONNAIRES?**



You should speak to a member of your coeliac care team at the hospital if you have any worries or concerns. You can also contact your General Practitioner (GP) or Coeliac UK (tel: 0845 305 2060) who will be able to provide advice about where to access further information and/or support.

If you would like to make a complaint about the research study then you can contact the Patient and Advice Liaison Service (PALS) on *{insert number for local trust}*

### **WILL MY TAKING PART IN THE STUDY BE KEPT CONFIDENTIAL?**

Information that could reveal your identity will be kept confidential to the researchers, medical team and GP. However, if there were worries about your child's safety or someone else's safety then we would need to follow local child protection procedures.

### **EXPENSES AND PAYMENTS**

You will not be given any money for taking part in the research. However, young people who complete the questionnaires will be entered into a free prize draw to win an IPOD shuffle. We will also provide a stamped addressed envelope for you to return completed questionnaires.

### **WHO CAN I CONTACT IF I HAVE ANY QUESTIONS ABOUT THIS STUDY?**

You may ask the study staff any questions you may have about this study at any time. If you would like to discuss any part of this research please contact:

**Name:** Charlotte Tolgyesi  
**Email:** [REDACTED]  
**Telephone:** [REDACTED]  
**Post:** Charlotte Tolgyesi  
Trainee Clinical Psychologist  
School of Psychology  
University of Birmingham  
[REDACTED]

**OR**

**Name:** Dr Ruth Howard

**Email:** [REDACTED]

**Telephone:** [REDACTED]

**Post:** Dr Ruth Howard  
Clinical Director, ClinPsyD  
School of Psychology  
University of Birmingham



Thank you for reading about this study. This is your copy of the information sheet

**Appendix 8: Participant Consent Forms**

**CONSENT FORM FOR YOUNG PERSON**

Research site: .....

Title of Project: **Psychological Impact of Coeliac Disease in Young People**

Participant Identification Number: .....

Researcher: Charlotte Tolgyesi

Please initial box

1. I confirm that I have understood the information sheet dated 28/5/10 (version 2) for the above study. I have had time to think about the information and ask questions about the research. ☐
2. I understand that my participation is voluntary and that I can leave the research at any time, without giving any reason. If I decide not to take part the study my medical care will not be affected. ☐
3. I understand that the data collected during this study will be looked at by the research team at the University of Birmingham to make sure that the analysis is a fair and reasonable representation of the data. ☐
4. I understand that the data collected during the study may be looked at by individuals from the University of Birmingham, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data. ☐
5. I understand that if there were worries about me or someone else then the researcher would need to follow the appropriate guidelines and procedures ☐
6. I agree to my GP being informed of my participation in the study. ☐
7. I agree to take part in the above study. ☐

.....  
Name of participant

.....  
Date

.....  
Signature

I confirm that I am the parent/guardian of name of participant and that I provide consent for name of participant to take part in the above study.

|                                  |               |                    |
|----------------------------------|---------------|--------------------|
| .....<br>Name of parent/guardian | .....<br>Date | .....<br>Signature |
| .....<br>Name of researcher      | .....<br>Date | .....<br>Signature |

## CONSENT FORM FOR PARENT/CARER

Research site: .....

Title of Project: **Psychological Impact of Coeliac Disease in Young People**

Participant Identification Number: .....

Researcher: Charlotte Tolgyesi

Please initial box

1. I confirm that I have understood the information sheet dated 28/5/10 (version 2) for the above study. I have had time to think about the information and ask questions about the research. ☐
2. I understand that my participation is voluntary and that I can leave the research at any time, without giving any reason. If I decide not to take part in the study my child's medical care will not be affected. ☐
3. I understand that the data collected during this study will be looked at by the research team at the University of Birmingham to make sure that the analysis is a fair and reasonable representation of the data. ☐
4. I understand that the data collected during the study may be looked at by individuals from the University of Birmingham, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my child's data and my data. ☐
5. I understand that if there were worries about your child's safety or someone else's safety then the researcher would need to follow local child protection procedures ☐
6. I agree to my GP being informed of my participation in the study. ☐
7. I agree to take part in the above study. ☐

.....  
Name of participant

.....  
Date

.....  
Signature

.....  
Name of researcher

.....  
Date

.....  
Signature

## **Appendix 9: Letter of invitation for young people and parents attending clinic**

*Research site letter head*

*Name of parent/carer*

*Address line 1*

*Address line 2*

*Address line 3*

*Address line 4*

Dear

### **Re: Psychological Impact of Coeliac Disease in Young People**

I would like to invite you and your child to take part in a study to find out what it is like for young people who have Coeliac Disease and their parents. Charlotte Tolgyesi, Trainee Clinical Psychologist is carrying out the research as part of her university doctorate (3 year course) at the University of Birmingham.

I have enclosed an information sheet for you and your son/daughter about the research study. If there is anything that is not clear, or if you would like more information, please contact Charlotte Tolgyesi. Her contact details are on the information sheets enclosed.

Thank you for reading this.

Yours sincerely

**Name of Consultant/Dietician**  
**Title**

## **Appendix 10: Details of procedure**

Three methods of recruitment were employed:

- 3) Young People attending clinic at two hospitals in the West-Midlands during the data collection stage:

A covering letter and information sheets were sent by the local coeliac care team to young people and parents who met inclusion criteria at least one week before their child's coeliac care review. Information sheets invited young people and their parents to take part in the research study. It also explained the rationale for the research, method, confidentiality, consent and their right to withdraw from the study at any point.

These young people and their parents were given the option to discuss the research in more detail with the chief investigator (Charlotte Tolgyesi) after their coeliac review appointment (with their Paediatric Gastroenterologist or Dietitian). Potential participants who made an informed choice to participate in the study were asked to sign a written consent form. If the young person is under 16 years of age, consent was also obtained from parents or from those in loco parentis.

Participants were provided with the option to complete questionnaires either during the clinic or at home. Participants who chose to complete the questionnaires after their medical appointment were provided with a quiet room in outpatients during the clinic to complete them. Questionnaires took approximately 25-30 minutes to complete. The chief investigator was available to answer questions related to the study. Participants who were unable to complete the questionnaires during the clinic, were able to complete the questionnaires at home. A stamped and addressed envelope was provided to return completed questionnaires to the chief investigator. Contact details of the chief investigator and her supervisors were provided for all participants if they had any questions related to the research. However, questions were answered in a manner to minimise bias.

2) Young people attending routine medical reviews for their condition at a hospital in Scotland.

Young people and parents attending medical review appointments were invited to participate in the research by their Consultant Paediatric Gastroenterologist, during a medical review appointment. If young people and/or parents expressed an interest in taking part, they were given a research pack. The research pack contained a letter of invitation, participant information sheets, young person questionnaire booklet, parent questionnaire booklet and written consent forms. Potential participants were asked to take the research pack home and read the information sheet. This ensured that potential participants had time to consider whether they wanted to take part. Contact details of the research staff were provided for all participants, should participants have any questions related to the research. Young people and parents chose if they would like to opt into the research. Those who decided to participate in the research were asked to sign the consent forms, complete the questionnaires and then post completed questionnaires with signed consent forms to the chief investigator (using the stamped and addressed envelope provided). Written instructions for this process were described on the information sheet. If the young person was under 16 years of age, consent was also obtained from parents or from those in loco parentis.

3) Young people not attending a clinic during data collection

Some young people did not attend clinic during the data collection phase of the study. These young people and parents were identified by the child's coeliac medical care team and were sent a research pack by their Consultant Paediatric Gastroenterologist. The research pack contained a letter from their Consultant or Dietitian, participant information sheets, young person questionnaire booklet, parent questionnaire booklet and written consent forms. Contact details of the research staff were provided, for questions related to the research. Young people and parents were able to choose if they wanted to opt into the research. Those who decided to participate in the research were asked to sign the consent forms, complete the questionnaires and then post completed questionnaires with signed consent forms to the chief investigator



(using the stamped and addressed envelope provided). If the young person was under 16 years of age, consent was also provided by parents or from those in loco parentis. Questionnaires took approximately 25-30 minutes to complete.

## Appendix 11: Descriptive Statistics

### Illness Representations

Table 1 displays mean and standard deviations for young people's illness representations, and parental illness representations. To explore the degree of similarity or difference in illness representations, young people and parental illness representations were compared using either paired t-tests or Wilcoxon tests for parametric and non-parametric analysis, respectively.

**Table 1:** Illness representations and treatment perceptions

| Variable                         | Young Person |                     |          | Parent |                     |          | Young Person and Parent Comparisons |          |                 |
|----------------------------------|--------------|---------------------|----------|--------|---------------------|----------|-------------------------------------|----------|-----------------|
|                                  | N            | Mean (median)       | SD (IQR) | N      | Mean (Median)       | SD (IQR) | N                                   | t (or z) | Sig. (2-tailed) |
| <i>IPQ-R</i>                     |              |                     |          |        |                     |          |                                     |          |                 |
| <i>Identity</i>                  | 39           | (3) <sup>1</sup>    | (9)      | 30     | (3.50) <sup>1</sup> | (6.25)   | 29                                  | (0.123)  | 0.217           |
| <i>Timeline-Acute-Chronic</i>    | 40           | 4.15 <sup>2</sup>   | 0.61     | 32     | (4.58) <sup>2</sup> | (1.04)   | 32                                  | (-1.234) | 0.217           |
| <i>Consequences</i>              | 40           | (3) <sup>2</sup>    | (0.70)   | 32     | 3.18 <sup>2</sup>   | 0.78     | 32                                  | (-0.627) | 0.531           |
| <i>Personal Control</i>          | 40           | (3.67) <sup>2</sup> | (0.67)   | 32     | 3.96 <sup>2</sup>   | 0.66     | 32                                  | (-1.519) | 0.129           |
| <i>Treatment Control</i>         | 40           | 3.57 <sup>2</sup>   | 0.69     | 34     | (4.00) <sup>2</sup> | (1.05)   | 34                                  | (-1.885) | 0.059           |
| <i>Illness Coherence</i>         | 40           | (2) <sup>2</sup>    | (1)      | 34     | (1.80) <sup>2</sup> | (1.1)    | 34                                  | (-0.681) | (0.496)         |
| <i>Timeline Cyclical</i>         | 40           | 2.79 <sup>2</sup>   | 0.86     | 34     | (1.88) <sup>2</sup> | (1.13)   | 34                                  | (-3.358) | <b>(0.001)</b>  |
| <i>Emotional Representations</i> | 40           | 2.60 <sup>2</sup>   | 0.96     | 34     | 3.02 <sup>2</sup>   | 1.29     | 34                                  | -1.886   | 0.068           |

Note: Scores for *identity* range from 0-20. All other scores range from 0-5.

<sup>1</sup> Total median score.

<sup>2</sup> Adjusted mean/median score (sum of scale items divided by number of items).

Overall, young people and their parents believed that CD was a *chronic* condition. They also perceived moderate negative *consequences* and moderate *emotional representations*

associated with CD. Similarly, young people and parents perceived moderate levels of *personal control* over their condition and coherence levels were positive overall (thus, indicating low levels of confusion or puzzlement in regard to CD). In contrast, there was some statistically significant discrepancy between young people and their parents' *timeline-cyclical* beliefs. Namely, young people scored moderately for *timeline-cyclical beliefs*, whereas their parents did not perceive the condition to fluctuate ( $z=1.885$ ;  $p<0.01$ ). In general, parents also believed that CD was controlled by treatment and young people perceived moderate *treatment control*. Differences in these scores were not significant, but there was a trend towards significance.

### *Identity*

*Tummy pain* (n=20), *upset tummy/diarrhoea* (n=17), *feeling tired* (n=16) and *feeling sick* (n=15) were the symptoms that young people most often associated with CD. For parents, *upset stomach/diarrhoea* (n=18), *tummy pain* (n=17) and *loss of strength* (n=13) were the symptoms most often attributed to CD.

### *Treatment Beliefs*

In general, young people scored moderately for self-management *necessity* beliefs (Mdn=3.8, IQR=0.80) and did not report high *concerns* ( $M=2.47$ ,  $SD=0.69$ ) related to dietary self-management.

### Bivariate correlations

A series of inter-correlations between IPQ-R subscales for young people (see table 2) and parents (see table 3) were identified.

**Table 2:** Young People's illness representations

| Young Person Variable        | 1        | 2             | 3      | 4              | 5      | 6              | 7   |
|------------------------------|----------|---------------|--------|----------------|--------|----------------|-----|
| 1. Identity                  | -        |               |        |                |        |                |     |
| 2. Timeline (acute/chronic)  | (.22)    |               |        |                |        |                |     |
| 3. Consequences              | (.31)    | (-0.2)        |        |                |        |                |     |
| 4. Personal Control          | (.02)    | (-.13)        | (0.2)  |                |        |                |     |
| 5. Treatment Control         | (.06)    | -.18          | (0.28) | <b>(.61**)</b> |        |                |     |
| 6. Illness coherence         | (.24)    | -.22          | (.25)  | (-.22)         | (-.36) |                |     |
| 7. Timeline cyclical         | (0.49**) | <b>-.62**</b> | (.08)  | (-0.40)        | .08    | (.40)          |     |
| 8. Emotional Representations | (.38)    | <b>-.48**</b> | (.40)  | (-.17)         | -.30   | <b>(.49**)</b> | .36 |

Note: \*\*p<0.01

Inter-correlations using Spearman's rho are displayed in brackets.

Inter-correlations indicated that young people who believe their CD is a chronic condition (i.e. *timeline acute-chronic*) were less likely to believe their CD fluctuates (i.e. *timeline-cyclical*) and had less negative *emotional representations* associated with their condition. In contrast, young people who did not feel their condition made any sense (i.e. *illness coherence*) were more likely to report negative *emotional representations*. *Personal control* and *treatment control* were positively correlated with each other.

Spearman's rho showed that *age* was not significantly correlated with any dimensions of illness representations. However, young people who had been diagnosed with CD for longer, were more likely to believe that their condition was chronic (*timeline acute/chronic*:  $r_s=.45$ ,  $p<0.01$ ) and more likely to perceive CD to come and go in cycles (*timeline-cyclical*:  $r_s=-.441$ ,  $p<0.01$ ). No other illness representations were associated with duration of CD.

**Table 3:** Parental illness representations

| <i>Parent Variable</i>          | <i>1</i> | <i>2</i> | <i>3</i> | <i>4</i>       | <i>5</i> | <i>6</i> | <i>7</i> |
|---------------------------------|----------|----------|----------|----------------|----------|----------|----------|
| 1. Identity                     |          |          |          |                |          |          |          |
| 2. Timeline<br>(acute/chronic)  | (-.19)   |          |          |                |          |          |          |
| 3. Consequences                 | (-.01)   | (-0.90)  |          |                |          |          |          |
| 4. Personal<br>control          | (.10)    | (.14)    | .03      |                |          |          |          |
| 5. Treatment<br>control         | (.06)    | (.07)    | (-.04)   | <b>(.61**)</b> |          |          |          |
| 6. Illness<br>coherence         | (.13)    | (-.40)   | (.14)    | (-.05)         | (-.14)   |          |          |
| 7. Timeline<br>cyclical         | (.05)    | (-.15)   | (.30)    | (-.24)         | (-.36)   | (.40)    |          |
| 8. Emotional<br>representations | (0.5)    | (.15)    | -.37     | -.09           | (.16)    | (.24)    | (.20)    |

Note: \*\* $p<0.01$

Inter-correlations using Spearman's rho are displayed in brackets.

As Table 5 shows, only parental beliefs measuring perceived *personal control* and *treatment control* were positively correlated with each other.

## Self-efficacy

Young people were asked to rate their self-efficacy to stick to a gluten-free diet across a variety of situations. The most common situations where young people reported low levels of confidence (rated below 5 out of 10) in their ability to stick to their gluten-free diet, were *eating out at my favourite restaurant* (27%, n=11), *when I want more dietary variety* (27%, n=11) and *when faced with appealing foods that are not gluten-free in a supermarket, vending machine or cafe* (24%, n=10). A Mann-Whitney test showed that total self-efficacy in young people did not differ between boys and girls.

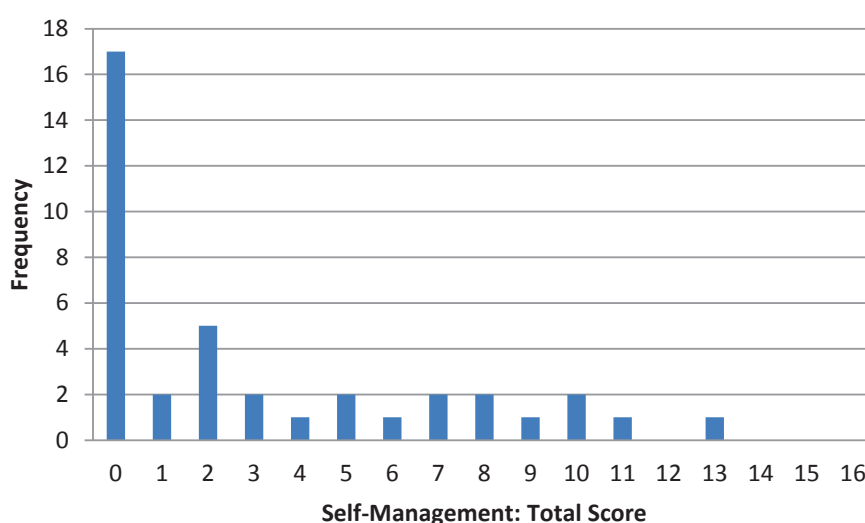
Young people's self-efficacy was negatively and moderately correlated with their own timeline-cyclical beliefs ( $r_s = -0.596$ ,  $p < 0.01$ ), but not to other illness representations. This indicates that young people with higher levels of self-efficacy were less likely to believe that their CD fluctuates.

## Dietary Self-Management

A dietary self-management total score was generated from four items: the frequency of eating gluten in the last two weeks (at home and away from home), as well as young people's perception of how well they stick to the gluten-free diet generally (at home and away from home). Figure 3 shows self-management total scores, with lower scores indicating better dietary self-management. Overall, 44% (n=17) of young people reported good self-management, indicated by a score of 0 (i.e. did not knowingly eat foods containing gluten in the last two weeks and in general stick to their gluten free diet extremely well). More specifically, 60% (n=24) of young people reported that they had not eaten foods containing

gluten at home (in the last two weeks). This frequency increased to 65% (n=26) while away from home.

In contrast, 13% (n=5) of young people reported that in the last two weeks they had eaten gluten *daily* or *all the time* while at home (10% while away from home). 15% (n=6) of young people reported that in general they did not stick to their gluten-free diet very well while at home (10% away from home).



**Figure 1:** Self-Management total scores

Mann-Whitney tests showed that self-management (self-report and parent report) was not significantly different between males and females. Furthermore, a Wilcoxon test showed no significant difference between parents' and child's self-management ratings (see Table 4). This provides some indication of inter-rater reliability across self-management ratings.

**Table 4:** Self-Management descriptive data and comparison scores

| Variable               | Young Person |        |      | Parent |        |      | Young Person and Parent Comparisons |       |                 |
|------------------------|--------------|--------|------|--------|--------|------|-------------------------------------|-------|-----------------|
|                        | N            | Median | SD   | N      | Median | SD   | N                                   | z     | Sig. (2-tailed) |
| <b>Self-Management</b> | 39           | 2.00   | 6.00 | 34     | 1.00   | 4.50 | 33                                  | -1.49 | 0.14            |

Note: range 0-16

Higher score = poorer self-management

## Well-being

### *Well-being of Young People*

Well-being of young people was assessed using the SDQ (self-report and parent versions) and KIDSCREEN. Overall, SDQ mean total scores indicated that young people were in the “normal” range (see Table 5). More specifically, 77% (n=31) of self-report and 82% (n=28) of parent-report SDQ scores were in the “normal” range, 10% (n=4) of self-report and 3% (n=1) of parent scores were classified as “borderline”. “Abnormal” scores were identified in 10% (n=4) of self-report questionnaires and 15% (n=5) of parent-report questionnaires.

**Table 5:** SDQ scores

| SDQ              | Self-report Version |       |      | Parent Version |      |      |
|------------------|---------------------|-------|------|----------------|------|------|
|                  | N                   | Mean  | SD   | N              | Mean | SD   |
| SDQ: Total Score | 39                  | 11.31 | 5.45 | 34             | 8.47 | 6.66 |

Note: Scores range 0-40.

Mean and median KIDSCREEN subscale scores were also in the average range (see Table 6). More specifically, only 8% (n=3) of young people were in the “low” or “very low” range for *physical well-being*, 5% (n=3) for *psychological well-being*, 8% (n=3) for *parent relations and autonomy*, 8% (n=3) for *social support* and 3% (n=1) for *school environment* (see Table



7). Levels of well-being did not differ significantly between boys and girls, when measured by KIDSCREEN (Mann-Whitney) and SDQ total scores (Independent t-test).

**Table 6:** KIDSCREEN descriptive data.

| <b>KIDSCREEN Subscale</b>                         | <b>Boys</b> |                      |                 | <b>Girls</b> |                      |                 |
|---|-------------|----------------------|-----------------|--------------|----------------------|-----------------|
|   | <b>N</b>    | <b>Mean (Median)</b> | <b>SD (IQR)</b> | <b>N</b>     | <b>Mean (Median)</b> | <b>SD (IPR)</b> |
| <sup>2</sup> <i>Physical Wellbeing</i>            | 9           | (19.00)              | 5.50)           | 29           | (19.00)              | (5.00)          |
| <sup>3</sup> <i>Psychological Well-being</i>      | 9           | 29.44                | 3.09            | 29           | 27.79                | 4.25            |
| <sup>3</sup> <i>Parent Relations and Autonomy</i> | 9           | 27.66                | 5.07            | 29           | 28.31                | 4.71            |
| <sup>1</sup> <i>Social Support and Peers</i>      | 9           | 16.67                | 3.16            | 29           | 16.48                | 3.16            |
| <sup>1</sup> <i>School Environment</i>            | 9           | 15.44                | 2.70            | 29           | 15.41                | 3.16            |
| <sup>a</sup> <i>KIDSCREEN total Score</i>         | 40          | 107.00               | 14.70           | 29           | 105.83               | 15.95           |

Note: <sup>2</sup>= 5 items; <sup>3</sup>=7 item; <sup>1</sup>= 4 items, <sup>a</sup>=27 items

**Table 7:** KIDSCREEN scores

|                                | Gender       | Very low | Low | Low average | Average | High average | High | Very high |
|--------------------------------|--------------|----------|-----|-------------|---------|--------------|------|-----------|
| Physical Wellbeing             | Girls (n=30) | 7%       | 0%  | 7%          | 27%     | 37%          | 23%  |           |
|                                | Boys (n=9)   | 11%      | 0%  | 11%         | 44%     | 33%          | 0%   |           |
| Psychological well-being       | Girls (n=31) | 3%       | 3%  | 13%         | 48%     | 19%          | 13%  |           |
|                                | Boys (n=9)   | 0%       | 0%  | 33%         | 56%     | 11%          | 0%   |           |
| Parents relations and Autonomy | Girls (n=31) | 0%       | 6%  | 13%         | 39%     | 32%          | 10%  |           |
|                                | Boys (n=9)   | 0%       | 11% | 11%         | 56%     | 11%          | 11%  |           |
| Social Support and Peers       | Girls (n=30) | 3%       | 7%  | 13%         | 43%     | 33%          |      |           |
|                                | Boys (n=9)   | 0        | 0   | 22%         | 33%     | 44%          |      |           |
| School Environment             | Girls (n=29) | 3%       | 0%  | 17%         | 38%     | 28%          | 14%  |           |
|                                | Boys (n=9)   | 3%       | 0%  | 11%         | 67%     | 0%           | 22%  |           |

Note: high score = positive well-being

As expected, *SDQ: Total Score* was negatively correlated with *KIDSCREEN: total score* ( $r=-0.48$ ,  $p<0.01$ ). This is logical given that a higher SDQ scores indicates greater emotional and behavioural difficulties, whereas higher scores on the KIDSCREEN suggest better well-being. Correlations between the KIDSCREEN and SDQ are shown in Table 8.

**Table 8:** Bivariate correlations: Well-being of young people

|                             | 1             | 2              | 3              | 4            | 5            | 6            |
|-----------------------------|---------------|----------------|----------------|--------------|--------------|--------------|
| 1. SDQ: total score         |               |                |                |              |              |              |
| 2. Kidscreen: total score   | <b>-.48**</b> |                |                |              |              |              |
| 3. Physical Wellbeing       | (-.28)        | <b>(.71**)</b> |                |              |              |              |
| 4. Psychological Well-being | <b>-.51**</b> | <b>.89**</b>   | <b>(.64**)</b> |              |              |              |
| 5. Parent relations         | -.36          | <b>.82**</b>   | <b>(.41**)</b> | <b>.44**</b> |              |              |
| 6. Social Support           | -.15          | <b>.68**</b>   | (.35)          | <b>.54**</b> | <b>.44**</b> |              |
| 7. School                   | <b>-.55**</b> | <b>.81**</b>   | <b>(.43**)</b> | <b>.72**</b> | <b>.62**</b> | <b>.47**</b> |

Note: \*\*  $p<0.01$

Inter-correlations using Spearman's rho are displayed in brackets

### *Well-being of Parents*

The Depression, Anxiety and Stress Scale (DASS) measured parents' psychological well-being. Overall, the DASS indicated levels of psychological wellbeing (as measured by symptoms of depression, anxiety and stress) in the "normal" range (see Table 9). More specifically, 94% were in the "normal" range for depression (  $n=32$ ), anxiety (88%,  $n=30$ ) and stress (88%,  $n=30$ ) (see Table 10).

**Table 9:** Parent's well-being descriptive data

| <b>Parents Well-being</b> | <b>N</b> | <b>Median</b> | <b>Interquartile range</b> |
|---------------------------|----------|---------------|----------------------------|
| DASS: Depression          | 34       | 0.50          | 2.00                       |
| DASS: Anxiety             | 34       | 0.00          | 1.00                       |
| DASS: Stress              | 34       | 1.00          | 3.25                       |
| GWBI: Total Score         | 34       | 23.5          | 7.00                       |

**Table 10:** DASS scores

| <b>DASS</b>      | <b>Normal</b> |          | <b>Mild</b> |          | <b>Moderate</b> |          | <b>Severe</b> |          | <b>Extremely Severe</b> |          |
|------------------|---------------|----------|-------------|----------|-----------------|----------|---------------|----------|-------------------------|----------|
|                  | <b>N</b>      | <b>%</b> | <b>N</b>    | <b>%</b> | <b>N</b>        | <b>%</b> | <b>N</b>      | <b>%</b> | <b>N</b>                | <b>%</b> |
| DASS: Depression | 32            | 94%      | 0           | 0%       | 1               | 3%       | 1             | 3%       | 0                       | 0%       |
| DASS: Anxiety    | 30            | 88%      | 0           | 0%       | 2               | 6%       | 2             | 6%       | 0                       | 0%       |
| DASS: Stress     | 30            | 88%      | 1           | 3%       | 0               | 0%       | 2             | 9%       | 0                       | 0%       |

Inter-correlations between measures of well-being showed that parents with high levels of anxiety (DASS: Anxiety) were also more likely to have higher levels of depression (DASS: Depression) ( $r=.51$ ,  $p<0.01$ )

## **Appendix 12: Copies of letters relevant to ethical review and approval**

[not available in electronic copy of thesis]

### **Appendix 13: Copies of Research and Development Approval Letters**

[not available in electronic copy of thesis]