

VOLUME I

RESEARCH COMPONENT

**LIVING WITH COELIAC DISEASE AND TYPE 1 DIABETES:
THE EXPERIENCES OF ADOLESCENTS AND THEIR
PARENTS**

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A Thesis submitted to

The University of Birmingham

In partial fulfilment for the degree of

Doctorate of Clinical Psychology (Clin.Psy.D.)

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OVERVIEW

This thesis is submitted in partial fulfilment of the requirements for the degree of Clinical Psychology Doctorate at the University of Birmingham. The two volumes included within this thesis comprise of research (Volume I) and clinical work (Volume II).

Volume I

Volume I presents a literature review, qualitative research paper and a public domain paper.

The literature review examines research that explored the links between within-family agreement and disagreement regarding the sharing of responsibility for diabetes management tasks and diabetes outcomes in youth Type 1 Diabetes.

The research study used a qualitative approach to explore adolescents' and parents' experiences of what it is like for a young person to live with a dual diagnosis of Type 1 Diabetes and Coeliac Disease.

The public domain paper presents a summary of the literature review and research paper.

Volume II

Volume II comprises five clinical practice reports reflecting various aspects of clinical work completed across clinical placements:

Clinical Practice Report 1 describes the case of an 8-year old boy with low self-esteem and difficult peer relationships formulated from cognitive and systemic perspectives.

Clinical Practice Report 2 is a service evaluation, conducted in a CAMHS service in the West Midlands, exploring staff perspectives on peer group supervision and discharge that aimed to evaluate discharge processes within a new model of service delivery.

Clinical Practice Report 3 is a single-case experimental design evaluating a cognitive-behavioural intervention to increase the use of memory aids for a 28-year old man with an acquired brain injury.

Clinical Practice Report 4 describes a case study exploration of a 79-year old woman with hallucinations and mild cognitive impairment.

Clinical Practice Report 5 was an oral presentation of a piece of clinical work with a 25-year old male with a learning disability and his family who were attending a community learning disability team. Assessment, formulation and intervention were informed by a systemic approach. An abstract summarising this piece of work is included.

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Paper to be edited for submission to British Journal of Psychology

**INTRA-FAMILIAL AGREEMENT AND DISAGREEMENT
REGARDING FAMILY DIVISION OF DIABETES
RESPONSIBILITIES:
ASSOCIATIONS WITH DIABETES OUTCOMES IN YOUTH
TYPE 1 DIABETES**

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ABSTRACT

Divergence in family member's perceptions of diabetes family responsibility is an emerging area of within-family research in the youth diabetes literature. This review sought to appraise this dyadic family research in relation to three research questions exploring how the distribution of diabetes responsibility is perceived by family members, how agreement and disagreement is associated with diabetes outcomes, and family interventions targeting family sharing of responsibility in improving diabetes outcomes. Thirteen studies were available for inclusion in the review. Ten cross-sectional studies explored the perceptions of various family dyads regarding the distribution of diabetes responsibilities and the associations between different patterns of agreement/discrepancy in family views and diabetes outcomes. Three studies explored the efficacy of family-focused interventions in improving and maintaining family sharing of diabetes responsibility.

Methodological issues were assessed to vary across studies and the implications for generalisability of findings and drawing conclusions are considered. Findings suggest there is limited evidence of strong associations between family agreement/disagreement regarding family diabetes responsibility and diabetes outcomes. Greater parent-child disagreement has been linked to poorer metabolic control and decreased parental psychological wellbeing. Further within-family research, particularly of a longitudinal nature, is required to clarify the influence of family agreement/disagreement regarding distribution of diabetes responsibility on metabolic control, diabetes self-management and family psychosocial functioning.

Keywords: type 1 diabetes, agreement, disagreement, family, responsibility, metabolic control, psychosocial, self-management

INTRODUCTION

Type 1 Diabetes (T1D) is one of the most common chronic health conditions among children and adolescents and accounts for more than 90% of all types of diabetes diagnosed in young people (Craig, Hattersley & Donaghue, 2009).

Treatment for T1D is complex and demanding, with self-care requiring the effective management of a complicated routine including monitoring of blood glucose levels, taking exercise, dietary self-management and administering insulin. These behavioural tasks are fundamental to maintaining healthy blood glucose levels to help prevent short- and long-term diabetes complications.

However, the day to day nature of self-care responsibilities inherent in the management of diabetes can prove very challenging and burdensome for young people and their families (Anderson et al., 2002; Debono & Cachia, 2007; Greening, Stoppelbein & Reeves, 2006), impacting the family system around the adolescent, including family roles, relationships and responsibilities related to diabetes management.

As T1D is commonly diagnosed in childhood, parents naturally assume a significant degree of responsibility for management of their child's chronic health condition, and parents can report struggles with managing their child's diabetes. A recent systematic review (Whittemore, Jaser, Chao, Jang & Grey, 2012) exploring parental psychological experiences of living with children's type 1 diabetes reported prevalence rates for psychological distress (i.e. symptoms of depression and anxiety) between 10 – 74% (with an average of 33.5% at diagnosis and 19% at 1-4 years post-diagnosis). Such distress was associated with several negative outcomes for the child with diabetes, including increased stress and depressive symptoms, behavioural difficulties and lower quality of life.

Parental psychological distress was also negatively associated with tasks that help to optimise family management of diabetes (i.e. reduced frequency of parental monitoring and lower parental self-efficacy for management). However, the relationship between parental diabetes distress and metabolic control requires further exploration (Whittemore et al., 2012).

Adolescence has been identified as a potentially difficult time in the management of diabetes, such that insulin resistance can occur in response to hormonal changes (Amiel, Sherwin, Simonson, Lauritano & Tamborlane, 1986; Goran & Gower, 2001; Moran et al., 1999). Furthermore, a range of biopsychosocial factors can interact with the normal developmental processes of adolescence to complicate self-management and result in poorer metabolic control (McConnell, Harper, Campbell & Nelson, 2001).

Models of the family lifecycle (e.g. Carter & McGoldrick, 1999) describe the stage at which a child enters adolescence as requiring shifts in the parent-child relationship to afford development of the adolescent's autonomy (Carr, 2006). For some families this also represents a juncture at which several other transitions occur concurrently, resulting in increased strain within the family system (Carr, 2006). The developmental course of childhood chronic conditions generates additional complications which may impact the family's ability to negotiate these tasks. During adolescence patterns of responsibility need to shift, by a process of parents relinquishing some responsibility and the adolescent assuming increased responsibility, in order for the child to develop autonomy in diabetes self-care. For parents, this may relate to their role in providing protection and care for their child's needs. For adolescents, achieving the balance between acquiring

autonomy and maintaining parental involvement to support the more complex demands of diabetes management may prove challenging.

Previous research has explored the degree of parental involvement in children's and adolescents' diabetes management and links with diabetes health outcomes. The decline in parental involvement in the tasks of diabetes management during adolescence (Allen, Tennen, McGrade, Affleck & Ratzan, 1983) has been associated with poorer metabolic control and self-management (La Greca et al. 1995; Wysocki et al. 1996). There appears to be a general consensus within the literature advocating the importance of continued parental involvement and shared diabetes family responsibility across adolescence for achieving and maintaining improved diabetes outcomes (Silverstein et al., 2005; Wysocki & Greco, 2006). In the last two decades the focus has expanded to exploration of the distribution of responsibility for diabetes management within families, particularly between parent and child/adolescent.

The transition in family sharing of responsibility for diabetes management that occurs across adolescence may generate uncertainty, and thus divergent perceptions in family members, surrounding each other's responsibility for the performance of self-management tasks. Recognition that divergence in the perceptions of family members involved in diabetes management may represent an important variable related to family functioning in illness management represents a clinically important area of understanding. This is due to the potential detrimental effect that a lack of responsibility for regimen tasks may have on metabolic control and long term health complications. Sood, Shroff-Pendley, Delamater, Rohan & Pulgaron (2012) explored how mother-father discrepancies regarding their child's diabetes self-management related to family functioning

(diabetes family conflict) and diabetes outcomes (metabolic control) for children aged 9-12 years. Mother-father discrepancies related to diabetes self-management predicted poorer metabolic control (for children whose fathers were reported to be less directly involved in diabetes care) and increased frequency of diabetes family conflict. As family sharing of diabetes management responsibility occurs within a relationship context, the divergent perceptions of parents and adolescent may also lead to conflict. Discord between family members regarding diabetes responsibility may lead to ambiguity and conflict in family communication regarding diabetes management. Greater diabetes family conflict has been linked to poorer diabetes self-management and metabolic control (Anderson, 2004).

Family studies within the child and adolescent literature commonly combine family members' data across respondents to create a mean score, or analyse the responses of fathers, mothers and adolescents as separate participant groups. Increasingly, dyadic research designs and complex analytic methods are being employed to examine the extent of agreement and discrepancy between parent (usually mothers) and child/adolescent perceptions of diabetes self-management at a within-family level, and how this links to diabetes outcomes, including metabolic control. This review will attempt to collate and evaluate this dyadic research with regard to diabetes family responsibility.

Self-management of diabetes is experienced at a collective, rather than individual level within families. Therefore, the move towards a family systems framework allows exploration (from multiple levels) of family dynamics which may shape self-management of T1D. Evidence linking poorer parental psychological wellbeing with decreased psychological wellbeing of the child and diminished family management of T1D (Whittemore et al. 2012) lends support to the clinical

relevance and importance of exploring the interrelations between how families function as a system and self-management of diabetes.

Parent and child/adolescent sharing of responsibilities is one aspect of the broad concept of family involvement and collaboration in diabetes self-management (Belendez, De Wit & Snoek, 2010). For the purposes of this review 'diabetes family responsibility' is defined as the responsibility taken by a family member (parent and/or child) for decision-making processes and performance of tasks related to the behavioural and social aspects of diabetes management. Different combinations of family agreement and disagreement (e.g. between mother-child, father-child, mother-father etc.) are considered in this exploration.

AIMS

This review aims to describe and appraise existing evidence in relation to family agreement and disagreement regarding sharing of diabetes responsibilities and the links with diabetes outcomes in youth with type 1 diabetes; specifically, metabolic control (as indexed by HbA1c)¹, diabetes self-management and diabetes family psychosocial functioning. The clinical implications of these findings and recommendations for clinical practice and future research will also be considered. The review seeks to answer the following questions:

- 1) What are mother, father and child/adolescent perceptions of family sharing of diabetes management?
- 2) In what ways do family members' views of the division of diabetes responsibility correspond and differ, and what are the effects of this on HbA1c levels, diabetes self-management and family psychosocial functioning (including family diabetes distress, conflict, adolescent and parental wellbeing)?
- 3) Are family-focused interventions targeted at facilitating family negotiations regarding responsibility for diabetes management efficacious in improving HbA1c levels, diabetes self-management and family psychosocial functioning?

¹ The term HbA1c refers to glycated haemoglobin

METHOD

Search strategy

The following databases were searched: EMBASE (1980 – October 2012), PsycINFO (1967 – October 2012), Web of Science (1960 – 2012) and MEDLINE (1950 – 2012). The following terms (adapted according to the search requirements of each database) comprised the search framework utilised across database searching: (diabete* or “type 1 diabetes mellitus”) AND (child* or adolescen* or “young people” or “young person” or teen* or youth or infan*) AND (parent* or mother* or father* or famil*) AND ((responsibility or accountability) OR (treatment compliance or disease management or illness behaviour) OR (self or dependent or share or sharing or divid* or spread* or care or responsib* or involv* or dut* or task* or job* or administer* or monitor* or injection*)). A detailed example of the search strategy and search terms can be found in Appendix 3. This strategy yielded 353 references, which were subjected to a systematic sifting process, involving title scanning and abstract review (see Figure 1), informed by the inclusion and exclusion criteria stated below. The full text and reference sections of selected articles were also examined to identify any additional papers of relevance. A total of 13 papers were selected for review. Information summarising each study is presented in Table 1.

Inclusion and exclusion criteria

Studies were included if they met the following criteria: 1) included a sample of children or young people (mean age \leq 18 years) with T1D and/or a sample of parents/mothers/fathers, reporting on aspects of family sharing of responsibility for diabetes management, 2) directly explored family agreement

and/or disagreement regarding family division of diabetes responsibility, 3) reported on HbA1c, self-management or aspects of family psychosocial functioning. Quantitative and qualitative studies were included.

Studies that explored family perceptions of diabetes responsibility but did not assess agreement/disagreement or conduct within-family analysis of members' data were excluded. Papers that reported health conditions other than, or as well as, T1D data and did not include a child/young person sample were also excluded along with conference abstracts, books, review articles and papers not published in English.

Figure 1. Flowchart depicting the systematic sifting process

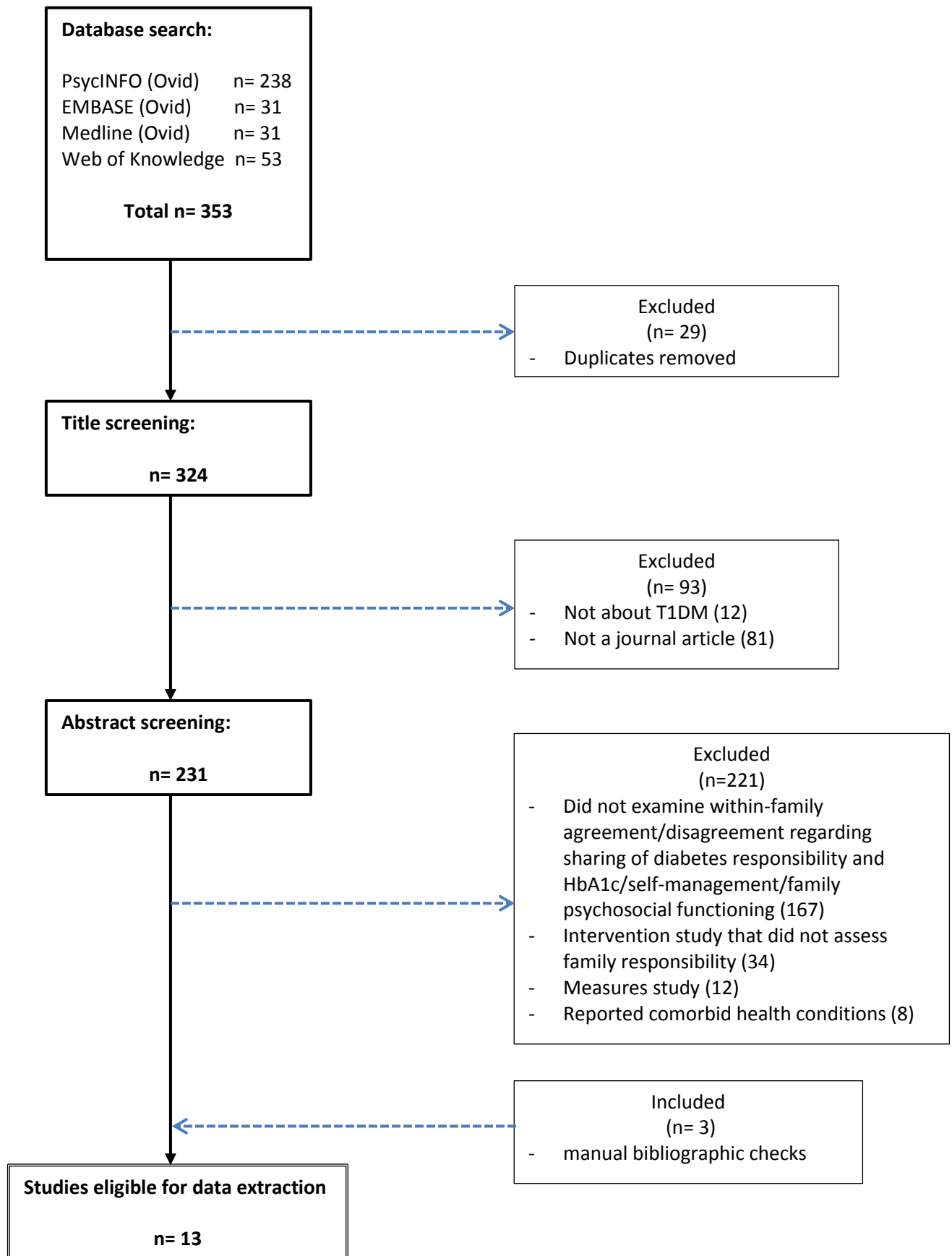


Table 1. Summary table for each study included in the review

Setting	Objectives	Participants	Design	Intervention and/or Outcome measures/data collection	Results (relevant to scope of current literature review)
<i>Country of origin</i>		a) Number of p (% uptake) b) Gender (adolescents) c) Age (mean age)			
Anderson, Auslander, Jung, Miller & Santiago (1990)					
Children's Hospital of a University Medical Centre <i>USA</i>	To examine mother and child sharing of responsibility for diabetes tasks and associations with demographic variables, adherence and metabolic function	a) 121 children and their mothers b) 54% female, 46% male c) 6 – 21 years (13.3)	Cross-sectional Quantitative	<ul style="list-style-type: none"> • DFRQ (developed for this study) • Interviews: demographic information (parent report), mother and child ratings of adherence to treatment • HbA1c levels from blood samples 	<ol style="list-style-type: none"> 1. Both parent and child report that older children assume greater responsibility 2. Higher levels of disagreement (i.e. 'no one takes responsibility' for a task) (including diabetes regimen tasks) were found between younger children (age not identified) and their mothers 3. Multiple regression analyses found higher levels of dyadic scores indicating that no one takes responsibility for tasks (i.e. disagreement) predicted poorer metabolic control
Anderson, Holmbeck, Iannotti, McKay, Lochrie, Volkening & Laffel (2009)					
4 large paediatric diabetes centres (clinical sites) <i>USA</i>	To explore family conflict and parent-child agreement and discordance regarding responsibility for diabetes tasks: associations with glycaemic control	a) 121 children and parents (73%) b) 50% male, 50% female c) 9 – 14 years (12.1) split into 2 groups: 'younger' = <12 years, 'older' = 12 > years	Cross-sectional Quantitative Dyadic (parent-child)	<ul style="list-style-type: none"> • DFRQ • DFCS • HbA1c 	<ol style="list-style-type: none"> 1. No significant effect of age on the frequency of parent-child agreement about diabetes task responsibility 2. Increased frequency of parent-child discordance about responsibility sharing was found in the younger group (9-11 years) 3. Dyadic discordance was not significantly correlated with HbA1c levels for any of the groups (younger, older or combined) 4. Mean diabetes conflict score (parent report) was higher in the older group (12-14 years) than younger group but

					<p>differences were not significant</p> <ol style="list-style-type: none"> Higher levels of conflict (parent report) were significantly associated with higher (poorer) HbA1c for all groups. However, child reported conflict was not significantly associated with HbA1c Increased dyadic agreement was associated with lower (better) HbA1c in the younger group (9-11 years) but not the older group (12-14 years) In regression analyses parent reported diabetes conflict and dyadic agreement were found to be independent predictors of HbA1c: increased dyadic agreement and lower levels of diabetes conflict were associated with lower (better) HbA1c
Beveridge, Berg, Wiebe & Palmer (2006)					
Not reported <i>USA</i>	To explore the concept of 'shared illness ownership' between adolescents and their mothers and relations to joint responsibility for diabetes tasks, diabetes specific stress and age-related change	<p>a) 127 adolescents and 127 mothers (68%)</p> <p>b) 52% male, 48% female</p> <p>c) 10 -15 years (12.85)</p>	<p>Cross-sectional</p> <p>Quantitative</p> <p>Dyadic (mother-child)</p>	<ul style="list-style-type: none"> Responsibility subscale of DRCS Stress and coping interview (separate) Illness ownership interview (joint) 	<ol style="list-style-type: none"> 71% of mothers and 53% of adolescents reported that diabetes was a shared issue, with 50% of mother-child dyads congruent in this view Adolescents and mothers who viewed illness ownership as shared were more likely to report shared (equal) responsibility for diabetes management (compared to those who reported diabetes to be the adolescents own issue to manage) This relationship between diabetes ownership appraisals and diabetes responsibility was also reported in dyads who congruently appraised diabetes ownership to be shared: with mothers and adolescents more likely to report sharing responsibility for management tasks equally Congruence in appraisals of shared illness ownership was not significantly associated with congruence in views of

					<p>diabetes stressful events as mothers and adolescents reported experiencing different types of stressors over the past week</p> <ol style="list-style-type: none"> Metabolic control, food management, injections, BGM and management issues outside of home were the stressors most commonly reported by mothers and adolescents, but they differed in their perspectives of which is most stressful Older adolescents (mean age 13.5 years) reported decreased shared illness ownership (i.e. diabetes as their own issue to manage), however, no age differences were found in mothers' reports Age was not found to be significantly associated with illness ownership congruence in dyads Congruence in reports of stressful events was more likely to occur between younger adolescents and their mothers
Butner, Berg, Osborn, Butler, Godri, Fortenberry, Barach, Le & Weibe (2009)					
<p>Outpatient paediatric diabetes clinics</p> <p>USA</p>	<p>To examine mother, father and child discrepancies in perceptions of adolescent's competence and independence (responsibility) in completing diabetes tasks</p>	<p>a) 185 adolescents, 185 mothers and 145 fathers (66%)</p> <p>b) not reported</p> <p>c) 10 – 14 years (12.52)</p>	<p>Cross-sectional</p> <p>Quantitative</p>	<ul style="list-style-type: none"> SCI (adapted) Responsibility items of the DRCS (revised) SEDMS 2nd treatment problem subscale of Peds-QL Diabetes specific module A measure of adolescent functional autonomy MFPS (independence-encouragement subscale) CDI PWBS (2 subscales- parent report) TCESDS 	<ol style="list-style-type: none"> Although mothers and fathers were consistently discrepant from their adolescent, there was variation in the ways in which they differed across aspects of diabetes management competence and independence Adolescents perceived themselves to be more competent and responsible in diabetes management than did their mothers and fathers Greater adolescent autonomy (adolescents who perceived themselves to be more responsible were more discrepant from their parents' views of them) and parental encouragement of autonomy (greater divergence in views of

				<ul style="list-style-type: none"> LWMAT HbA1c levels 	<p>adolescent' diabetes management in mothers who were perceived to be more encouraging of adolescent independence) were associated with such parent-child discrepancy</p> <p>4. Mother, but not father, discrepancy was associated with HbA1c: greater discrepancy between mother and adolescent was associated with higher HbA1c levels</p> <p>5. Greater discrepancies in perceptions of adolescent competence and independence were associated with poorer glycaemic control and poorer maternal psychological well-being (increased depressive symptoms)</p>
Dashiff (2003)					
<p>Children's Hospital (& participants' homes)</p> <p>USA</p>	To explore congruence of parent-child perceptions of self and dependent-care responsibility for diabetes self-management in two-parent families and relationship to metabolic control	<p>a) 31 adolescents and 31 mothers and 31 fathers (approx. 60%)</p> <p>b) 58% male, 42% female</p> <p>c) 12 – 15 years (13.8)</p>	<p>Cross-sectional</p> <p>Quantitative</p> <p>Dyadic (mother-adolescent, father-adolescent, mother-father)</p>	<ul style="list-style-type: none"> DFRQ (modified to include fathers) HbA1c levels (from medical records) at two intervals 2 months apart 	<p>1. Mother-adolescent perceptions of self and dependent-care responsibility were congruent</p> <p>2. Differences between mother-father and father-adolescent perceptions were found</p> <p>3. Mothers and adolescents viewed the adolescent as having responsibility for the majority of tasks (i.e. more self-care responsibility), however, fathers viewed mothers as primarily responsible for most tasks</p> <p>4. However, mothers were still perceived (by all) to have significant dependent-care responsibility (ranging from a third to half of all management tasks)</p> <p>5. Fathers perceived to have less responsibility (by all), averaging less than one management task. However, fathers perceived themselves to have more responsibility than mothers perceived them as having</p>

					6. No significant associations between parent-child perceptions of diabetes responsibility and metabolic control were found 7. Trends in the data to suggest: 1) adolescents who were previously in poorer glycaemic control perceived their mothers to have more responsibility (and adolescents have less) and 2) fathers perceived mothers to have more responsibility when current metabolic control was worse
Hilliard, Rohan, Carle, Pendley, Delamater & Drotar (2011)					
Children's Hospitals USA	To examine the degree of agreement between mothers' and fathers' perceptions of the amount and helpfulness of paternal involvement in diabetes management. To evaluate three different models of associations between amount and helpfulness of fathers' involvement with adherence and glycaemic control	a) 136 mothers and 136 fathers b) 46% male, 54% female c) 9 - 12 years (10.5)	Cross-sectional Quantitative	<ul style="list-style-type: none"> • DADSS • DSMP • BGM frequency (meter data) • Demographic and medical information • HbA1c 	1. Significant correlation between mother and father ratings of paternal involvement in diabetes management. 2. Mothers and fathers agreed on the amount of parental involvement, but fathers perceived their contributions to diabetes care to be less helpful than mothers did.

Law, Walsh, Queralt & Nouwen (2013)					
Outpatient departments of 8 hospitals <i>United Kingdom</i>	To examine how adolescent and parent diabetes distress is associated with perceived consequences of diabetes, dietary self-efficacy and discrepancies in diabetes family responsibility	a) 203 adolescents and 203 parents (primary caregiver) (52% response rate) b) 44% male, 56% female c) 12 – 18 years (14.5)	Cross-sectional Quantitative	<ul style="list-style-type: none"> Demographic information (age, sex, duration of diabetes and current treatment method) PMDQ DSES DFRQ 	<ol style="list-style-type: none"> Parent-adolescent discrepancies regarding family responsibility for diabetes self-management were not significantly associated with adolescent diabetes distress. Parent-adolescent disagreement in the context of both parent and adolescent claiming responsibility for tasks contributed to parental diabetes distress. However, disagreement that related to neither family member assuming responsibility for diabetes self-management did not significantly contribute to parental distress.
Miller & Drotar (2003)					
Clinic at a Children's Hospital <i>USA</i>	To examine the association of discrepancies in mother and adolescent perceptions of diabetes decision-making autonomy with diabetes conflict and regimen adherence	a) 82 adolescents and 82 mothers (63%) b) 55% male, 45% female c) 11 – 17 years (13.9)	Cross-sectional Quantitative Mother-child dyad	<ul style="list-style-type: none"> Demographic questionnaire HbA1c PDS ASI DADTS Conflict subscale of the DRCS SCI Health Care Provider Rating questionnaire BGM frequency (meter data) 	<ol style="list-style-type: none"> Mother-adolescent discrepancies in perceptions of decision-making autonomy were associated with higher diabetes-related conflict (maternal report): mothers reported increased conflict when their adolescent perceived that they had more responsibility for making diabetes decisions, than attributed to them by their mothers. Discrepancies in perceptions of diabetes-related decision-making autonomy were not associated with regimen adherence or glycaemic control.
Palmer, Berg, Wiebe, Beveridge, Korbel, Upchurch, Swinyard, Lindsay & Donaldson (2004)					
Diabetes clinic (Children's medical centre)	To examine how mother and child perceptions of decreased maternal involvement in diabetes	a) 127 children and 127 mothers (68%) b) 52% male, 48% female	Cross-sectional Quantitative	<ul style="list-style-type: none"> Responsibility subscale of DRCS Self-reliance subscale of PMI Mother ratings of fixed-item responses to sentence enquiring about decision to give more responsibility to their child 	<ol style="list-style-type: none"> High correlation between mother-child perceptions of who held responsibility for diabetes tasks, but children reported less maternal involvement than mothers Maternal involvement decreases as adolescents get older Transfer of responsibility when autonomy

USA	management across adolescence relate to adolescent autonomy and pubertal status	c) 10 – 15 years (12.85)		<ul style="list-style-type: none"> Mother ratings of signs of puberty HbA1c values from medical records 	and pubertal status were low was related to poorer glycaemic control
Schilling, Knafl & Grey (2006)					
Not reported USA	To explore youth and parent perspectives on how they divide diabetes management tasks (in the context of patterns of self-management across developmental stages), the degree of youth responsibility for diabetes management and self-management conflict	a) 22 children and 22 parents: 17 mothers and 5 fathers (uptake not reported) b) 13 females and 9 males c) 8 – 19 years (14.5)	Cross-sectional Qualitative Dyads (mother or father-child)	<ul style="list-style-type: none"> Interviews (semi-structured) 	1. <i>Preadolescents</i> , 8-11 years (n=4 dyads): <ul style="list-style-type: none"> parents responsible for majority of diabetes care parent-dominant pattern of self-management mostly low levels of self-management conflict reported 2. <i>Early adolescence</i> , 11-15 years (n=9 dyads): <ul style="list-style-type: none"> parents actively participate and oversee self-management but adolescent responsible for much of diabetes care transitional self-management pattern all parent-adolescent pairs reported conflicts (i.e. over food and BGM) 3. <i>Mid-adolescence</i> , 15-17 years (n=5 dyads): <ul style="list-style-type: none"> Adolescent responsible for the majority of their care with parent oversight of diabetes tasks still considerable in some dyads over half of adolescents showing adolescent-dominant pattern of self-management majority of dyads reported exercise as source of conflict 4. <i>Late adolescence</i> , 17-19 years (4 dyads): <ul style="list-style-type: none"> adolescent independently manages their care adolescent-dominant self-management

					<ul style="list-style-type: none"> dyads reported conflict over food and BGM <p>5. Conflict over food and BGM most commonly reported across developmental stages</p>
Intervention studies					
Anderson, Brackett, Ho & Laffel (1999)					
Diabetes Centre USA	To evaluate an office-based intervention targeting maintenance of teamwork between parent and adolescent in tasks of diabetes management without increasing diabetes-related conflict.	<p>a) 85 young adolescents and parents (not reported whether one or two parents participated) (64%)</p> <p>2 x intervention groups: teamwork intervention (n=28) & attentional control (n=30)</p> <p>control group: standard care (n=27)</p> <p>b) not reported</p> <p>c) 10-15 years (12.6)</p>	<p>RCT: randomisation stratified according to age and gender.</p> <p>Data collection at baseline, 12 months and 24 month follow-up</p>	<p>Teamwork intervention (4 sessions across 12 month period): Focused on the importance of parent-adolescent sharing of responsibility for diabetes tasks and addressing barriers to effective teamwork in diabetes management (i.e. conflict). Family discussions facilitated and written materials provided. A detailed family responsibility sharing plan was negotiated at first session and reviewed and/or renegotiated during subsequent sessions.</p> <p>Attention control (equivalent frequency and duration of sessions to teamwork intervention): Didactic 'traditional' diabetes education provided (e.g. explaining diabetes to others, effects of stress and exercise on diabetes etc). Parent-adolescent responsibility sharing not emphasised, with no plan for family teamwork negotiated.</p> <p>Standard care (quarterly appointments across 12 month period): Routine clinical care from diabetes team.</p>	<ol style="list-style-type: none"> Families in the teamwork intervention maintained shared responsibility (no deterioration in parental involvement) in insulin administration or BGM. Less diabetes-related conflict was reported at 12 months by families who participated in the teamwork intervention No significant difference in HbA1c between groups during the 12-24 month follow-up period, but trends in the data to suggest that more young adolescents in the teamwork intervention showed improvements in glycaemic control (68%), than did those in the comparison group (47%).

				Measures <ul style="list-style-type: none"> • <u>Interview questions to ascertain family division of responsibility</u> for insulin injections and BGM (quantitatively coded); ranging from 1 = adolescent has total responsibility (no parental involvement) to 4 = adolescent has no responsibility (maximum parental involvement) • <u>DFCS</u> (completed at baseline and 12 months) • <u>DFBC</u> • <u>HbA1c</u> (at each session) 	
Laffel, Vangsness, Connell, Goebel-Fabbri, Butler & Anderson (2003)					
Diabetes Centre USA	To evaluate a family-focused intervention designed to improve glycaemic control through negotiated family participation in diabetes management (specifically insulin injections and BGM), including decreasing diabetes-related family conflict and sustaining quality of life	a) 100 children/ adolescents and parents (not reported whether one or two parents participated) (81%) 2 conditions: teamwork intervention (n=50) & standard care (n=50) b) not reported c) 8 – 17 years (12.1)	RCT: randomisation stratified according to age and duration of diabetes. Data collection at baseline and 12 months	Teamwork intervention (4 sessions across 12 month period): Focused on the importance of parent-child sharing of responsibility for diabetes tasks and addressing barriers to effective teamwork in diabetes management (i.e. conflict). Family discussions facilitated and written materials provided. A detailed family responsibility sharing plan was negotiated at first session and reviewed and/or renegotiated during subsequent sessions. Standard care (4 sessions across 12 month period): Brief joint parent and child interview held at quarterly sessions. Families received the same written materials as the teamwork group (given as a complete package at conclusion of the study) but were not engaged in	1. 30% of families who participated in the teamwork intervention maintained or increased family involvement at 12 month follow-up, compared to 14% of families in receiving standard care. 2. HbA1c levels did not deteriorate in teamwork intervention group and were significantly lower (0.5% less=better control) after 12 months, compared to the standard control group. 3. The teamwork intervention and increased frequency of BGM were significant predictors of lower HbA1c at follow-up. 4. Families in the teamwork intervention group did not report an increase in diabetes family conflict or a decrease in quality of life resulting from the increased and maintained family involvement.

				<p>family teamwork discussions.</p> <p>Measures (completed by parent and child at baseline and 12 months):</p> <ul style="list-style-type: none"> • <u>Interview questions to ascertain family division of responsibility</u> for insulin injections and BGM • <u>Peds QL</u> • <u>DFCS</u> • <u>DFRQ</u> • <u>Diabetes adherence rating scale</u> (completed by a clinician at each quarterly visit) • <u>Physical data</u>: growth, pubertal status, insulin therapy and diabetes management (from medical records) • <u>HbA1c</u> (from medical records) 	
Naar-King, Ellis, Idalski, Frey & Cunningham (2007)					
<p>Endocrinology clinic of a children's hospital</p> <p>USA</p>	<p>To determine if multisystemic therapy (MST) is effective in decreasing parental overestimation of adolescents' responsibility for diabetes management tasks</p>	<p>a) 127 adolescents and 127 caregivers (70%)</p> <p>intervention n= 64 control n= 63</p> <p>b) 49% male, 51% female</p> <p>c) 10 - 17 years (13.23)</p>	<p>RCT: randomisation stratified according to HbA1c level (at baseline)</p> <p>Repeated measures, intent-to-treat analyses</p> <p>Data collection at baseline, post-treatment</p>	<p>MST treatment (2-3 MST sessions a week for 6 months, plus standard multidisciplinary care): MST based on 9 treatment principles. Targeted adherence problems across family, peer and community systems. Multisystemic assessment of family strengths and weaknesses informed development of individualised treatment goals and interventions. A range of evidence-based intervention techniques utilised: CBT, parent training, BFST. Development of family routines and communication skills focused on clarification of distribution of responsibility for diabetes tasks.</p>	<ol style="list-style-type: none"> 1. There was a significant reduction in parental overestimation following MST (compared to standard care) and this was maintained at 12 month follow-up 2. Parental overestimation increased in the control group 3. Changes in parental overestimation were not associated with changes in HbA1c 4. MST was effective in decreasing parental overestimation regardless of age, ethnicity and family structure

			and 12 month follow-up	<p>Control (standard multidisciplinary care only): quarterly medical appointments with endocrinologist, nurse and a dietician.</p> <p>Measures (completed by adolescent and caregiver):</p> <ul style="list-style-type: none"> • <u>DFRQ</u>: parental overestimation score derived • <u>HbA1c</u> 	
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Abbreviations: BGM = Blood Glucose Monitoring; DFRQ = Diabetes Family Responsibility Questionnaire; DFCS = Diabetes Family Conflict Scale; DRCS = Diabetes Responsibility and Conflict Scale; SCI = Self-care Inventory; SEDMS = Self-efficacy for Diabetes Management Scale; Peds-QL = Pediatric Quality of Life Inventory; MFPS = Mother-Father-Peer Scale; CDI = Child Depression Inventory; PWBS = Psychosocial Well-being Scale; TCESDS = The Center for Epidemiological Studies of Depression Scale; LWMAT = Locke-Wallace Marital Adjustment Test; DADSS = Dads' Active Disease Support Scale; DSMP = Diabetes Self-management Profile; PMDQ = Personal Models of Diabetes Questionnaire; DSES = Dietary Self-efficacy Scale; PDS = Pubertal Development Scale; ASI = Adolescent Symptom Inventory; DADTS = Deciding About Diabetes Treatment Scale; PMI = Psychosocial Maturity Inventory; DFBC = Diabetes Family Behaviour Checklist; MST = Multisystemic Therapy; CBT = Cognitive-behavioural Therapy; BFST = Behavioural Family Systems Therapy.

APPRAISAL OF STUDY QUALITY

Caldwell, Henshaw & Taylor's (2005) framework for critiquing health-related research was utilised in the assessment of study quality. Acknowledging the potential for inconsistency/inaccuracy that can be created in numerical (and symbol) rating systems (Schunemann, Best, Vist, & Oxman, 2003), and in line with current recommendations (Downs & Black, 1998), a checklist method was utilised. Drawing on other critical appraisal tools (Critical Appraisal Skills programme [CASP] 2006; Downs & Black, 1998), two detailed checklists (qualitative and quantitative versions) were developed to help ensure consistency in the application of quality criteria and are included in Appendices 4-5.

A colour coding appraisal system was developed for the purpose of this review, with the aim of providing the reader with a clear map of quality across the different methodological domains of studies. Figure 2 identifies how each criterion was critiqued. Each study included in the review was subject to this checklist system in assessment of quality criteria, the results of which are presented in Tables 2, 3 and 4 with 'gaps' in quality criteria discussed below.

Figure 2. Colour coding rating system for quality appraisal of studies

Quality rating	Operational definition of quality rating
	<ul style="list-style-type: none">• Complete fulfilment of each criterion, or the majority of criteria have been met• Criteria which have not been met or rated as 'unable to determine' are not considered to pose a significant threat to overall quality of the study
	<ul style="list-style-type: none">• Partial fulfilment of each criterion• Criteria which have not been met or rated as 'unable to determine' are considered to pose minimal threat to overall quality of the study
	<ul style="list-style-type: none">• Insufficient fulfilment of each criterion, or few of the criteria have been met• Those criteria which have not been met or rated as 'unable to determine' are considered to pose a significant threat to overall quality of the study







































































































































		Quantitative studies								
		Anderson et al. (1990)	Anderson et al. (2009)	Beveridge et al. (2006)	Butner et al. (2009)	Dashiff (2003)	Hilliard et al. (2011)	Law et al. (2013)	Miller & Drotar (2003)	Palmer et al. (2004)
Rationale	<i>Clearly outlined?</i>									
Research aim(s)	<i>Clearly stated?</i>									
Ethical issues	<i>Identified & addressed?</i>									
Methodology	<i>Identified & justified?</i>									
Design	<i>Clearly identified? Rationale evident?</i>									
Hypothesis	<i>Stated?</i>									
	<i>Key variables defined?</i>									
Sample	<i>Population identified?</i>									
	<i>Selection described?</i>									
Data collection	<i>Valid & reliable method?</i>									
Data analysis	<i>Valid & reliable method?</i>									
Results	<i>Clearly presented?</i>									
Discussion	<i>Comprehensive?</i>									
	<i>Strengths & limitations identified?</i>									
Conclusion	<i>Justifiable?</i>									

Table 2. Assessment of quality criteria for quantitative studies

















		Qual. study
		Schilling et al. (2006)
Rationale	<i>Clearly outlined?</i>	
Research aim(s)	<i>Clearly stated?</i>	
Ethical issues	<i>Identified & addressed?</i>	
Methodology	<i>Appropriate?</i>	
Philosophical Background	<i>Described?</i>	
Design	<i>Clearly identified? Rationale evident?</i>	
Major concepts	<i>Identified?</i>	
Sample	<i>Population situated?</i>	
	<i>Selection adequately described?</i>	
Data collection	<i>Auditable method?</i>	
Data analysis	<i>Credible & confirmable method?</i>	
Reflectivity	<i>Considered & described?</i>	
Results	<i>Appropriately presented?</i>	
Discussion	<i>Comprehensive?</i>	
	<i>Strengths and limitations identified?</i>	
Conclusion	<i>Justifiable?</i>	

Table 3. Assessment of quality criteria for qualitative study














































		Intervention studies		
		Anderson et al. (1999)	Laffel et al. (2003)	Naar-King et al. (2007)
Rationale	<i>Clearly outlined?</i>			
Research aim(s)	<i>Clearly stated?</i>			
Ethical issues	<i>Identified & addressed?</i>			
Methodology	<i>Identified and justified?</i>			
Design	<i>Clearly identified? Rationale evident?</i>			
Hypothesis	<i>Stated?</i>			
	<i>Key variables defined?</i>			
Sample	<i>Population identified?</i>			
	<i>Selection adequately described?</i>			
Data collection	<i>Valid & reliable method?</i>			
Data analysis	<i>Valid & reliable method?</i>			
Results	<i>Clearly presented?</i>			
Discussion	<i>Comprehensive?</i>			
	<i>Strengths and limitations identified?</i>			
Conclusion	<i>Justifiable?</i>			

Table 4. Assessment of quality criteria for intervention studies

DESCRIPTION OF STUDIES AND OVERVIEW OF STUDY QUALITY

In line with the method of critical appraisal outlined above, methodological issues and discrepancies in quality criteria identified through the appraisal process for all of the studies reviewed are outlined below, set in the context of a synopsis of studies.

Design

The majority of the studies reviewed employed quantitative designs (n=12), one qualitative study was included (Schilling, Knafl & Grey, 2006). Three studies utilised a randomised control design (RCT) to investigate the efficacy of family-focused interventions targeting family collaboration regarding shared responsibility for diabetes management (Anderson, Brackett, Ho & Laffel, 1999; Laffel, Vangsness, Connell, Goebel-Fabbri, Butler & Anderson, 2003; Naar-King, Ellis, Idalski, Frey & Cunningham, 2007). Ten of the studies were cross-sectional in nature (Anderson, Auslander, Jung, Miller & Santiago, 1990; Anderson, Holmbeck, Iannotti, McKay, Lochrie, Volkening & Laffel, 2009; Beveridge, Berg, Wiebe & Palmer, 2006; Butner et al., 2009; Dashiff, 2003; Hilliard, Rohan, Carle, Pendley, Delamater & Drotar, 2011; Law, Walsh, Queralt & Nouwen, 2013; Miller & Drotar, 2003; Palmer, Berg, Wiebe, Beveridge, Korbel, Upchurch, Swinyard, Lindsay & Donaldson, 2004).

There was significant variation in study objectives. Variation was also observed in approaches taken to exploring intra-familial perspectives on division of diabetes management responsibility within this selection of studies. Table 5 outlines the focus on family perspectives (i.e. agreement or disagreement) and associations with diabetes outcomes explored by each study.

Table 5. Responsibility variables and diabetes outcomes explored by each study

	Variable (e.g. <i>agreement/disagreement</i> or <i>family negotiation of responsibility</i>)	Diabetes outcomes (relevant to scope of current review)
Anderson et al. (1990)	<i>Disagreement</i> between mother-child/adolescent indicating no-one takes responsibility	<ul style="list-style-type: none"> • HbA1c
Anderson et al. (1999)	Maintenance of <i>collaboration</i> between parent and child on diabetes tasks	<ul style="list-style-type: none"> • HbA1c • Diabetes family conflict • Shared family responsibility
Anderson et al. (2009)	<i>Agreement</i> and <i>Disagreement</i> between parent-child regarding diabetes task responsibility	<ul style="list-style-type: none"> • HbA1c
Beveridge et al. (2006)	Mother-adolescent <i>congruence</i> in perceptions of joint responsibility for diabetes tasks	
Butner et al. (2009)	<i>Discrepancies</i> between mother-father-adolescent perceptions of adolescent's diabetes task competence and responsibility	<ul style="list-style-type: none"> • HbA1c • Adolescent wellbeing • Parental wellbeing
Dashiff (2003)	<i>Congruence</i> between mother-father-adolescent perceptions of self- (i.e. adolescent) and dependent-care (i.e. parent) responsibility	<ul style="list-style-type: none"> • HbA1c
Hilliard et al. (2011)	Congruence between mother-father perceptions of amount and helpfulness of paternal involvement (responsibility)	
Laffel et al. (2003)	<i>Negotiated</i> family participation in diabetes management	<ul style="list-style-type: none"> • HbA1c • Diabetes family conflict • Adolescent quality of life • Shared family responsibility
Law et al. (2013)	<i>Discrepancies</i> between parent-adolescent regarding family diabetes responsibility	<ul style="list-style-type: none"> • Adolescent and parental diabetes distress
Miller & Drotar (2003)	<i>Discrepancies</i> between mother-adolescent perceptions of diabetes decision-making responsibility	<ul style="list-style-type: none"> • HbA1c • Adolescent diabetes self-management • Diabetes family conflict
Naar-King et al. (2007)	<i>Disagreement</i> between parent-adolescent (i.e. parental overestimation) regarding adolescent responsibility for diabetes management tasks	<ul style="list-style-type: none"> • HbA1c
Palmer et al. (2004)	Congruence between mother-child perceptions of decreased maternal responsibility in adolescents' diabetes management	
Schilling et al. (2006)	Agreement between parent and adolescent regarding distribution of diabetes self-management tasks	<ul style="list-style-type: none"> • Diabetes self-management conflict

Participants

Participant characteristics varied considerably across the studies reviewed and warrant further consideration. Various age ranges and stages of adolescence were examined; for example, Anderson et al.'s (1990) sample of 'children' consisted of young people aged 6 – 21 years (but with mean age of less than 18 years). However, only two studies had an age range that exceeded 18 years (Anderson et al., 1990; Schilling et al., 2006). Some studies employed criteria such as Tanner staging to categorise participants according to their pubertal status/stage when presenting demographic information and collating study results (e.g. Anderson et al., 2009).² Similar ratios of males to females participated in each study (approximately 50:50). Only one study did not differentiate the gender split of the adolescent sample (Butner et al., 2009).

The quality of descriptions of study participants varied: most studies reported sufficient detail regarding child samples, yet very few described the demographics of parent participants. For example, Schilling et al. (2006) identified the number of mothers and fathers who comprised the parental sample, but no additional demographic data was collected. Seven studies detailed efforts to examine whether child samples were representative of the target population through conducting comparisons with non-participants on demographics, diabetes duration and metabolic control etc. (Anderson et al., 1999; Beveridge et al., 2006; Butner et al., 2009; Hilliard et al., 2011; Laffel et al., 2003; Miller & Drotar, 2003; Palmer et al., 2004). However, control groups to evaluate those

² A stage-based scale of the physical development of children, adolescents and adults, based on external primary and secondary sex characteristics: prepubertal (Tanner I), pubertal (Tanner II-IV) and postpubertal (Tanner V).

experiencing good versus poor metabolic control were not utilised in any of the studies reviewed.

A lack of ethnic and cultural diversity exists within the study samples reviewed. However, in Naar-King et al.'s (2007) intervention study, participants reflected a more diverse sub-section of the adolescent population with diabetes, and were reported as urban youth from lower income families with the majority of the sample (62%) described as African American. With the exception of one UK based study, all of the studies included participants originating from the USA. The social class of participants was mostly referenced in terms of parents' level of education and annual household income, with study samples largely categorised as 'middle class'. In the American studies, insurance status is likely to influence access to health care and thus research participation. Several of the studies explicitly acknowledged the limitations of these sample characteristics on the generalisability of their results (Beveridge et al., 2006; Butner et al., 2009; Hilliard et al., 2011; Laffel et al., 2003; Miller & Drotar, 2003; Palmer et al., 2004).

Detail of information regarding the family structure of participants (married, single-parent, step families, co-habitation of parent(s) and child) varied and was not made explicit in four of the studies (Anderson et al., 1999; Hilliard et al., 2011; Law et al., 2013; Schilling et al., 2006). Only one study controlled for family structure in their analyses: Naar-King et al. (2007) investigated whether the effects of a multisystemic therapy intervention on parental overestimation varied by demographic variables, including family composition. Therefore, given the diversity that exists in modern-day family systems, it is difficult to explore consistently the implications of family structure on parenting/caregiving practices; specifically, in the case of this review, family agreement and disagreement

regarding distribution of responsibility in the management of diabetes. In these samples, mothers predominantly participated and were recorded as the primary caregivers (e.g. Anderson et al., 1990, Palmer et al., 2004). However, five studies (Anderson et al., 2009; Butner et al., 2009; Dashiff, 2003; Hilliard et al., 2011; Schilling et al., 2006) did utilise paternal samples to varying degrees and differing in scope. Law et al. (2013) were unable to report on the degree of paternal participation due to missing self-report data on the sex of the participating parent. There are an increasing number of studies with a specific focus on paternal views in relation to child diabetes management and diabetes outcomes emerging within the literature.

Studies differed in how comprehensively they reported participants' management of their diabetes, that is, insulin delivery method (insulin pump or daily injections) and their level of metabolic control (indexed via HbA1c levels). In six of the studies the modes of insulin delivery utilised by participants were mixed, with samples consisting of those on daily injections and others on insulin pumps (Butner et al., 2009; Hilliard et al., 2011; Laffel et al., 2003; Law et al., 2013; Naar-King et al., 2007; Schilling et al., 2006). There appears to be a trend emerging of increased insulin pump use within samples of the more recently published research. Information on treatment delivery method was not reported by Anderson et al. (1990), Anderson et al., (2009) and Dashiff (2003), but it is important to note that insulin pumps were not likely to be an available treatment option for participants in the earlier studies. The mode of insulin delivery may be an important variable for researchers to consider as advancements in technology, such as insulin pump treatment, has significantly influenced methods of diabetes self-management and thus is also likely to have shifted patterns of family sharing

of responsibility. For example, parents of children aged 12-19 years reported withdrawing their responsibility related to insulin administration as their children were perceived to be more familiar with, and thus better skilled in the technological aspects of insulin pump operation (Olinder et al., 2011).

Glycaemic control

HbA1c values are considered to provide a reliable index of the average blood glucose level over the previous 4-8 weeks (Daneman, Wolfson, Becker & Drash, 1981), with current recommendations stating that levels should be below 8% for 13-19 year olds (American Diabetes Association, 2006). As such, HbA1c is considered to provide the most reliable indicator of diabetes self-management, thus a useful clinical research tool for scientific exploration of diabetes management and outcomes. The majority of studies reported HbA1c values for their samples with the exception of Beveridge et al. (2006) and Schilling et al. (2006). Reported mean values ranged between 7.9 – 11.6%, with an overall mean level across studies of 9.3%. Several of the studies' participants had not achieved the recommended level of metabolic control (Anderson et al., 1990; Dashiff, 2003; Law et al., 2013; Miller & Drotar, 2003; Naar-King et al., 2007). However, these values were often obtained at different stages of the research process, for example, at the point of recruitment, retrospectively from medical records, or subsequently at up to 12 months post-study protocol (Palmer et al., 2004). As such, it is not easy to make comparisons between studies and therefore conclude with any certainty whether participants in these samples were achieving adequate metabolic control, limiting generalisability of the results to the wider population of youth with diabetes.

Sample size

Total sample sizes, including all participating family members, ranged from 93 (Dashiff, 2003) to 515 (Butner et al., 2009). Sample sizes for the quantitative studies could be considered generally adequate; six studies had a sample of over 250 participants. The majority of studies recruited participants through convenience sampling via University health and research centres and hospital paediatric diabetes clinics. This may result in a sampling bias; that is, overrepresentation of families who attend health appointments, thus are motivated to seek, and engaged in receiving, help with diabetes management. The majority of the studies were conducted in the USA where health insurance status will determine, to a large extent, the child's access to medical care. An implication of this recruitment strategy is the potential exclusion of young people and their families with limited resources (physical and psychological), who do not receive regular medical attention/support with diabetes management. This may reflect an under-researched sub-section of youth with type 1 diabetes and their families who differ in their level of medical and socioeconomic status and for whom glycaemic control is poorer. Negotiation of responsibility for diabetes tasks may be more challenging for such populations.

Ethical considerations

Limited detail suggested that ethical issues were rarely identified and addressed appropriately among the studies: none reported on responding to potential upset or distress that may result from exploration of family conflict and illness management or risk issues arising from disclosure of risky health behaviours/poor metabolic control. Data collection procedures were not always

consistent or conducive to confidentiality. Beveridge et al. (2006) instructed participants to complete a battery of measures and a stress and coping interview independently but interviewed parent and child dyads together regarding illness ownership, which may have influenced how open participants were in their responses for these different variables.

Recruitment

There was judged to be a lack of sufficient detail regarding methods of recruitment (i.e. sampling and procedure) in the majority of the studies. Detail regarding inclusion and exclusion criteria for child and parent samples was assessed to be insufficient in several of the studies reviewed and was absent for participating parents in all but one study: Anderson et al., (2009) reported eligibility criteria for parent as well as child samples. The potential impact of a parent or other sibling in the family having a diagnosis of diabetes on management and thus distribution of responsibility did not appear to have been considered as this information was not reported in the studies. Diagnoses of parental mental ill-health served as exclusion criterion in Anderson et al. (2009). However, information pertaining to the presence of parental diabetes or other chronic health conditions was not reported in the inclusion/exclusion criteria for parents who participated in any of the studies reviewed. Diabetes duration of at least one year was cited as an eligibility criterion in the majority of studies, although studies were not explicit in explaining their rationale for this.

Measures

Various standardised and non-standardised measures were employed in the assessment of parent-child distribution of diabetes responsibility. The Diabetes Family Responsibility Questionnaire (Anderson et al., 1990) was utilised in six studies (Anderson et al., 1990; Anderson et al., 2009; Dashiff, 2003; Laffel et al., 2003; Law et al., 2013; Naar-King et al., 2007) as a measure to examine responsibility sharing between adolescent and parents. Dashiff (2003) adapted this measure to include fathers as respondents. Three studies (Beveridge et al., 2006; Butner et al., 2009; Palmer et al., 2004) administered the responsibility subscale of the Diabetes Responsibility and Conflict Scale (DRCS). Three studies developed interviews to assess family sharing of diabetes responsibility (Anderson et al., 1999; Laffel et al., 2003; Schilling et al., 2006).

Methods of analyses

A range of data collection, utilisation and analytical methods were used to explore family diabetes responsibility data at a family-level. Of the four quantitative, cross-sectional studies that utilised the DFRQ, three used this measure to derive a dyadic agreement/disagreement score for each family which was then subjected to correlation and regression analyses (Anderson et al., 1990; Anderson et al., 2009; Law et al., 2013), whilst Dashiff (2003) used correlations to examine the congruence of DFRQ scores between dyads. Naar-King et al.'s (2007) intervention study compared adolescent and parent dyad responses on the DFRQ to create a 'parental overestimation of adolescent responsibility' score. Correlations, Analyses of Variance (ANOVAs) and structural equation models were utilised in the remaining quantitative studies (Beveridge et al., 2006; Butner et

al., 2009; Hilliard et al., 2011; Miller & Drotar, 2003; Palmer et al., 2009). In Schilling et al.'s (2006) qualitative study, parent and child interview data were combined following qualitative content analysis.

RESULTS

Question 1. What are mother, father and child/adolescent perceptions of family sharing of diabetes management?

Twelve of the studies explored the parent-child/adolescent division of responsibility for diabetes management by means of eliciting the perceptions of both parent and child regarding their own and the other member's responsibility. The remaining study examined mothers' and fathers' perceptions of paternal involvement in their child's diabetes self-management (Hilliard et al., 2013). The interactions between distribution of diabetes responsibility and a range of demographic and diabetes-related variables were explored across the studies: age; pubertal status; metabolic control; diabetes-specific family conflict; dietary behaviour; blood glucose monitoring; self-efficacy and psychological health.

Both parents and children/adolescents report that older adolescents assume more responsibility than do younger adolescents (Anderson et al., 1990; Palmer et al., 2004; Schilling et al., 2006). Generally, within these family systems, mothers are considered to possess greater responsibility for diabetes self-management, with fathers perceived as having less responsibility (Dashiff, 2003). Both mothers and adolescents (but not fathers), perceived adolescents (aged 12-15 years) as primarily responsible for the majority of diabetes tasks, although mothers were still considered (by all family members) to hold greater responsibility for involvement that supports the adolescent's self-care (Dashiff, 2003). Adolescents also attribute increased competence and self-responsibility to themselves, compared to the amount attributed to them by their mothers and fathers (Butner et al., 2009). Adolescents and their mothers report maternal involvement in diabetes self-management to decrease as the adolescent gets

older, with mothers' intentions to promote the adolescent's maturity and diabetes management skills, as well as to reduce diabetes related disputes and difficulties, cited as instrumental in the process of transferring responsibility (Palmer et al., 2004).

Question 2. In what ways do family members' views of the division of diabetes responsibility correspond and differ, and what are the associations with HbA1c levels, diabetes self-management and family psychosocial functioning (including family diabetes distress, conflict, adolescent and parental wellbeing)?

Agreement and disagreement

In a study exploring the concept of shared illness ownership in families of 10-15 year olds with T1D, mother-adolescent dyads frequently appraised diabetes ownership as shared rather than being solely located within the adolescent (Beveridge et al., 2006). Those dyads who reported shared ownership of diabetes were also more likely to congruently report equal responsibility in the daily tasks of diabetes management. Beveridge and colleagues suggested that working together on tasks of diabetes (joint responsibility) may be the mechanism by which such appraisals are developed and advise clinical recommendations for broadening management interventions to include the family system. Despite sharing ownership of diabetes and its management, there were differences in how mother and child reported experiencing diabetes related stressors. Limitations of this study include illness ownership being assessed by a single question during a joint interview and the data were cross-sectional in nature. Therefore, it is not possible to establish causality and the authors acknowledge that longitudinal

research with additional measures of illness ownership is required to understand the developmental processes of shared illness representation.

The congruence in perceptions between mothers and fathers in parental management of diabetes was explored by two of the studies (Dashiff, 2003; Hilliard et al., 2011). Dashiff (2003) found mothers and fathers differed in their views regarding paternal responsibility, as fathers reported themselves to have greater responsibility than mothers attributed to them. In a separate study, mother and father ratings of paternal involvement in the performance of diabetes management tasks were significantly correlated, with dyads agreeing on the degree to which fathers were involved. However, there were discrepancies between these parents regarding the perceived helpfulness of paternal involvement, with fathers perceiving their efforts to be less helpful than did mothers (Hilliard et al., 2011). Extending exploration of family's perceptions to assess the value, as well as the amount of, member diabetes responsibility may offer additional insight into the relational processes of family agreement/disagreement and inform clinical understanding of how families may be supported in their shared management of the adolescent's diabetes.

Anderson et al. (1990) investigated how children with T1DM and their mothers share responsibilities for diabetes management. The DFRQ was developed specifically for the purposes of this study. This 17-item questionnaire assesses parent and child perceptions of who takes responsibility and to what extent responsibility is assumed, for a range of diabetes management tasks. Three multiple-item subscales were derived from maternal responses: responsibilities related to regimen tasks; general health maintenance; and social presentation of diabetes. The authors argued that analyses demonstrated the

measure to have adequate reliability and validity. The DFRQ has been frequently utilised as a clinical research tool by subsequent studies exploring the distribution of diabetes responsibility between parent and child. The questionnaire's psychometric properties were analysed based upon mother report only, yet the measure is increasingly being administered to include fathers as respondents, without exploration to establish whether use with this population impacts upon the reliability and consistency of the measure.

Patterns of agreement and disagreement in mother and child perceptions of sharing were explored (via mother-child dyadic scores) in relation to demographic variables (age, gender and diabetes duration), adherence and metabolic control. Anderson and colleagues were particularly interested in the response pattern characterised by mother and child disagreement (i.e. each reporting the other to have more task responsibility or one reporting shared responsibility and the other reporting no task responsibility). They found higher levels of discordance between parents and younger adolescents regarding who takes responsibility, the implication being that no one takes ownership for some tasks of diabetes. Furthermore, this pattern did occur for regimen tasks (e.g. administering injections, time management of blood glucose monitoring and awareness of the signs of an insulin reaction), although the authors did not identify the specific regimen items for which higher disagreement was found. Findings showed poorer metabolic control to be associated with dyads reporting higher levels of 'no one takes responsibility'. This is of greater clinical concern due to the detrimental effect on metabolic control and the potential for diabetes-related health complications in the longer term.

However, the process of how participants were categorised as either 'younger' or 'older' adolescents in discussion of the results was not made explicit, thus exact age ranges cannot be reported or compared across studies. Also, the age range of the sample (6-21 years) was extremely broad, encompassing many different stages of childhood, adolescence and early adulthood that are likely to differentially impact family sharing of diabetes responsibility. Later studies have explicitly differentiated between various stages, either developmentally (i.e. the stage of adolescence) and/or according to chronological age, within their data collection and analysis in acknowledgement of the dynamic and transitional nature of family diabetes management across this period. For example, Schilling et al. (2006) explored how parent-child/adolescent (aged 8-19 years) dyads share responsibility for diabetes tasks in the context of four developmental stages: preadolescence (8-11 years), early adolescence (11-15 years), mid-adolescence (15-17 years) and late adolescence (17-19 years). Findings demonstrated the changing patterns of self-management across these stages, from parent-dominant to transitional and then adolescent-dominant. The potential for categorising processes of diabetes management responsibility into stages for families may help to reduce family disagreement by facilitating increased understanding of their current distribution of responsibility and ease the transition in responsibility across adolescence. This may result in increased transparency, and improved negotiation and communication between parent and child and the health care system around them. Furthermore, the degree of support for families could be tailored depending on the adolescent's stage of development (i.e. increased family-centred care).

Mothers and fathers can differ in how they are discrepant from their adolescent's self-perceptions of competence and responsibility in management of T1D, with different implications for adolescents' diabetes outcomes and parental well-being (Butner et al., 2009): adolescents perceived themselves to be more responsible in management of their diabetes than did their parents. Adolescent perceptions of their autonomy in management (responsibility) and parental encouragement of autonomy (adolescent assuming more responsibility) were associated with such discrepancies (Butner et al., 2009).

HbA1c

Across the studies which explicitly explored associations between family agreement/disagreement regarding diabetes management responsibility and metabolic control (Anderson et al., 1990; Anderson et al., 2009; Butner et al., 2009; Dashiff, 2003; Miller & Drotar, 2003), findings were mixed. Anderson et al. (2009) found that increased parent-child dyad agreement was associated with lower (better) HbA1c in a group of 9-11 year olds, but not in an older group of 12-14 year olds. The authors consider that a relationship between dyadic agreement and metabolic control may not have been found in the older group as HbA1c levels may have been more significantly affected by hormonal changes in these adolescents. Parent-reported diabetes conflict and dyadic agreement were demonstrated to be independent predictors of metabolic control, such that increased dyadic agreement and lower levels of diabetes conflict were associated with lower HbA1c.

Increased disagreement between mothers and children was linked to higher HbA1c (Anderson et al., 1990)³. Greater discrepancies between mother-adolescent (but not father-adolescent) perceptions of the adolescent's competence, and independence in performance of diabetes self-management tasks, was associated with poorer metabolic control (Butner et al., 2009). The authors suggest that mothers' greater involvement and responsibility for adolescent's diabetes management may account for the link with HbA1c, as the same pattern was not found for father-adolescent discrepancies.

By contrast, no significant associations between mother-father-adolescent perceptions of the division of diabetes responsibility and glycaemic control were found in Dashiff's (2003) sample of two-parent families. However, there were trends in the data to suggest: 1) that adolescents who were previously in poorer metabolic control perceived their mothers to have more responsibility (and adolescents have less) and 2) fathers perceived mothers to have more responsibility when current metabolic control was worse. Discrepancies in mother's and adolescent's perceptions of diabetes related decision-making responsibility was not found to be significantly associated with metabolic control (Miller & Drotar, 2003).

To summarise, increased parent-child/adolescent agreement regarding diabetes family responsibility has been found to be associated with better metabolic control, whilst disagreement has been linked to poorer metabolic control. Two of the studies did not find any association between concordant or discordant family perceptions of responsibility (for decision-making or performance of diabetes management tasks) and metabolic control (Dashiff 2003; Miller &

³ Disagreement related to neither member assuming responsibility for diabetes tasks.

Drotar, 2003). Findings of age-related and family member dyad (i.e. mother-child vs. father-child) differences in associations with HbA1c suggest that consideration of within-family perspectives may be of importance to understanding metabolic control, but also highlights the multiplicity of variables that likely influence metabolic function.

Diabetes self-management

Mother-adolescent discrepancies regarding responsibility for decision-making on diabetes task responsibility was not found to be significantly associated with the degree of adolescent diabetes self-management (Miller & Drotar, 2003).

Family psychosocial functioning

Increased discrepancies in family perceptions of adolescents' diabetes self-management competence and responsibility was shown to be detrimental to mothers' psychological wellbeing, with mothers reporting increased depressive symptoms (Butner et al., 2009). Law et al. (2013) explored the association between parent-adolescent discrepancies in diabetes family responsibility and adolescent and parent diabetes distress (emotional difficulties specific to diabetes). Dyadic scores were calculated and categorised according to one of three possible combinations of responsibility agreement/disagreement: 1) 'agreed responsibility', 2) 'disagreement/discordance no responsibility taken' and 3) 'disagreement but responsibility taken/partial discordance'. Disagreements of any nature regarding family responsibility were not associated with adolescents' diabetes distress. By contrast, diabetes family responsibility disagreements were associated with parental diabetes distress. That is, disagreement whereby both

parent and adolescent claimed responsibility for tasks of diabetes management ('partial discordance') was linked to greater parental diabetes distress. A hypothesis that parental distress may occur in the context of this type of disagreement due to conflict between parent and adolescent (Law et al., 2013) could not be substantiated as the study did not include a measure of family conflict. Miller and Drotar (2003) found that when mothers and adolescents were more discrepant in their perceptions of the adolescent's diabetes decision-making responsibility, mothers reported experiencing higher diabetes-related conflict. That is, mothers reported more conflict when their adolescent perceived themselves to have to more responsibility for making diabetes decisions whereas their mothers rated them as having less.

These results suggest that parents (particularly mothers) and adolescents may experience the consequences of discrepancies in family responsibility differently in terms of their respective levels of diabetes distress and family conflict, such that parents are more concerned by these relational dynamics in diabetes self-management than are their adolescents. Differences in perceptions between mothers and adolescents, characterised by the adolescent perceiving themselves to have responsibility for diabetes decision making or task implementation, that their mother does not perceive them to have, appears to be particularly detrimental to parental psychological well-being and parent experiences of diabetes family conflict (Butner et al., 2009; Law et al., 2013; Miller & Drotar, 2003). Therefore, parents may benefit from additional support in exploring such discrepancies, and negotiating the sharing of diabetes management tasks, with their adolescent (Law et al., 2013). Further research exploring the associations between family diabetes responsibility discordance and

paternal perspectives of their own well-being and family functioning (e.g. diabetes family conflict) is required to assess in comparison to mothers and adolescents.

Question 3. Are family-focused interventions targeted at facilitating diabetes management negotiations within families efficacious in improving HbA1c levels, diabetes self-management and family psychosocial functioning?

Three studies explored the efficacy of family-focused interventions in improving and maintaining family sharing of diabetes responsibility (Anderson et al., 1999; Laffel et al., 2003; Naar-King et al., 2007). As part of a parent-adolescent 'teamwork' intervention, families negotiated a comprehensive family responsibility sharing plan which was continually reviewed and renegotiated across four sessions in a 12 month period (Anderson et al., 1999). Shared family responsibility for insulin administration and BGM was maintained by those who participated in the teamwork intervention, whereas families who received didactic diabetes education or standard clinical care showed deterioration in parental involvement in these diabetes management tasks. Families reported less diabetes-related conflict post-teamwork intervention. Although HbA1c levels were not found to statistically differ between groups during the 12-24 month follow-up period, 68% of adolescents in the teamwork intervention showed improvements in metabolic control compared to 47% in the comparison group. In a later study, Laffel et al. (2003) replicated a similar design to that of Anderson et al. (1999) but extended their objectives to target optimising metabolic control and maintaining child quality of life in children and adolescents aged 8-17 years. Shared family participation in insulin administration and BGM was maintained, or increased, for 30% of families who participated in the teamwork intervention, compared to 14%

of families of who received standard clinical care. Metabolic control did not deteriorate in the teamwork intervention group and HbA1c was found to be significantly lower at 12months in comparison to the control group. Families who completed the teamwork intervention did not report any increase in diabetes family conflict or decrease in child/adolescent quality of life.

Naar-King et al. (2007) focused on a specific type of parent-adolescent discrepancy in family diabetes responsibility conceptualised as 'parental overestimation'. This form of parent-adolescent disagreement was characterised by parents asserting that the adolescent assumes responsibility for a particular task of diabetes but the adolescent reports not taking this responsibility. A multi-systemic therapy (MST) intervention, adapted for a population of urban youth demonstrating long-term poor metabolic control, aimed to reduce parental overestimation of adolescent responsibility through improving family communication regarding how the tasks of diabetes self-management are allocated. This intervention involved family attendance at two to three MST sessions on a weekly basis for a 6 month period, in conjunction with standard multidisciplinary care. Techniques were informed by principles of CBT, parent training and BFST. Families who participated in the MST intervention demonstrated a significant reduction in parental overestimation, with decreases maintained at 12 months post-intervention. In comparison, parental overestimation was found to increase for families who received standard multidisciplinary diabetes care. However, changes in parental overestimation were not found to be significantly associated with changes in HbA1c levels.

All three intervention studies appear to demonstrate the efficacy of family-focused interventions in facilitating family negotiation of diabetes responsibility.

Also, Naar-King et al. (2007) provided evidence of the potential utility of such approaches for a sample of urban youth in poor metabolic control, suggesting the comparability of these findings to a sub-section of the paediatric diabetic population who are typically under-represented in the literature. The duration, intensity and therapeutic model of interventions did vary. Anderson et al. (1999) and Laffel et al. (2003) delivered very similar intermittent, low-intensity interventions (four sessions over 12 months) designed to be integrated within the routine model of diabetes clinical care (i.e. diabetes clinic sessions), whereas Naar-King et al. (2007) utilised a shorter-term intervention of a more specialist nature and increased intensity (2-3 weekly sessions for 6 months), in addition to the standard clinical care families received.

Findings examining associations between family negotiation of, and disagreements in, responsibility and HbA1c were equivocal. Only one study found a significant association between maintained or improved family sharing of diabetes responsibility and lower HbA1c (Laffel et al., 2003). The systemic intervention that specifically targeted family disagreement regarding family diabetes responsibility was not shown to be linked to HbA1c, but changes in family diabetes management dynamics (i.e. decreased disagreement) were maintained (Naar-King et al., 2007). Maintained or improved family sharing of diabetes responsibility was found to be associated with less diabetes family conflict (Anderson et al., 1999; Laffel et al., 2003) and did not result in reduced child/adolescent quality of life (Laffel et al., 2003). However, the follow-up periods to intervention were relatively short, therefore, it is not possible to assess whether families would benefit from on-going support to help firmly establish changes in

family behaviour patterns of diabetes management responsibility and if associations with HbA1c would increase in significance over time.

DISCUSSION

This review aimed to integrate and systematically examine the existing dyadic research exploring the association of intra-familial agreement and disagreement regarding distribution of diabetes responsibilities with metabolic control, diabetes self-management and family psychosocial functioning in youth type 1 diabetes. Although only a limited number of studies of this nature were available for review at present, there is some evidence to suggest that within-family diabetes responsibility agreements/disagreements may be differentially associated with diabetes outcomes. However, studies presented mixed results.

Overall, within these family systems, parents and children/adolescents viewed the adolescent to assume increased responsibility for self-management as they get older (Anderson et al., 1990; Palmer et al., 2004; Schilling et al., 2006). This finding is consistent with existing developmental family life-cycle models of adolescent's emerging autonomy (Carter & McGoldrick, 1999; Carr, 2006) and the paediatric diabetes literature demonstrating the decline in parental involvement in diabetes management during adolescence (Allen et al., 1983; La Greca et al. 1995; Wysocki et al. 1996). There appeared a strong perception amongst family members that mothers hold significant responsibility for diabetes self-management, or involvement that supports the adolescent's self-care (Dashiff, 2003), although maternal involvement is shown to decline across adolescence (Palmer et al., 2004). Fathers were perceived to hold the smallest proportion of the responsibility distributed within families. Family perceptions regarding the relative degree and perceived helpfulness of maternal and paternal involvement in diabetes management appears to be associated with discordance between family members' perceptions of responsibility. However, to date, the majority of research

has focused on mothers, so exploration of maternal involvement is weighted more heavily within the literature.

The dyadic design of the studies reviewed has provided insight into the divergent perceptions of different members within the same family system and explored the associations with a range of diabetes factors and outcomes. Findings have shown discrepancies in perceptions of diabetes family responsibility between parent and child, but also between mothers and fathers. Evidence of mother-adolescent discrepancies being associated with the adolescent's metabolic control was reported (Anderson et al., 1990; Butner et al., 2009), however, differing perceptions between father and adolescent did not show a similar pattern (Butner et al., 2009). The small number of studies and the methodological issues identified makes it difficult to draw conclusions, based on the strength of this evidence. However, there is evidence within the wider literature to suggest that mothers and fathers may respond differently to their roles in their child's diabetes care and that their perceptions of their involvement may have different associations with diabetes self-management and outcomes (Hanson, Schwartz, Weisbrod, & Taylor, 2012). Mother and father discrepancies may contribute to relational problems that place additional burden on parental efforts to collaborate in family diabetes management (Butner, 2009), corresponding with findings of poorer metabolic control and increased diabetes family conflict when mothers and fathers disagree regarding how frequently their child's engages in diabetes self-management behaviours (Sood et al., 2012).

A range of discrepant family perceptions have been explored providing an initial understanding of the types of disagreement that may be of clinical importance in terms of optimising metabolic control and supporting families with

the challenging regime of diabetes self-management. Most notably, in terms of the current review, two main types of parent-child discrepancies emerged as pertinent: mother and child/adolescent each attributing responsibility to the other person (Anderson et al., 1990) and parents disagreeing with their child's perceptions of the degree of responsibility that the child perceives themselves to have (Butner et al., 2009; Law et al., 2013; Miller & Drotar, 2003). Disagreement between parent and child that potentially results in neither taking responsibility for tasks of diabetes management was linked with higher HbA1c and will be of greater clinical concern to the medical position of diabetes care, given the significant risks poor metabolic control poses to immediate and longer-term health. The latter form of discrepancy was shown to be associated with parental, but not adolescent, psychological wellbeing (i.e. increased distress and depressive symptoms). Identification of the associations between specific patterns of family disagreement regarding diabetes responsibility and diabetes outcomes may help to inform development of targeted family interventions aimed at improving and maintaining diabetes self-management through clearer family communication and negotiation of diabetes responsibility.

Furthermore, the notion that parents appear to be more distressed by disagreements regarding diabetes responsibility suggests that children/adolescents do not internalise such discrepancies in the same way as their parents and as a result they do not appear to be concerned by these dynamics in family diabetes management. Parent-child discrepancies in perceptions of diabetes responsibility may contribute to diabetes family conflict (Miller & Drotar, 2003), with additional studies needed to corroborate this finding

and to explore whether the links with parental wellbeing are mediated by parent-child discrepancy via the route of conflict.

Research into family-focused interventions to facilitate family collaboration in diabetes self-management (including how responsibility is negotiated and distributed) is scarce. Therefore, whilst the potential for small improvements in HbA1c levels and increased or maintained shared family diabetes responsibility have been demonstrated, it is not clear what mediates these associations. Various approaches to improving diabetes self-management have been examined within the paediatric diabetes intervention literature. Hood, Rohan, Peterson & Drotar (2010) conducted a meta-analysis of 15 RCTs of self-management interventions, with interventions comprised of a specific behavioural goal and multiple components (e.g. emotional, social or family targets) found to be most effective. Exploring the efficacy of different systemic interventions for reducing within-family disagreement, and increasing concordance, in comparison to other psychological models of family interventions (e.g. behavioural, cognitive-behavioural, solution-focused) may help to further develop the evidence-base in relation to family division of diabetes responsibility.

The intra-familial approach of the studies reviewed represents a positive shift toward a family model within the youth with diabetes literature. However, it is important to note that the methods of data collection and analysis utilised within the studies resulted in exploration of what may be conceptualised as 'covert disagreements' between family members regarding responsibility for diabetes self-management. That is, the studies' measures of diabetes family responsibility (e.g. DFRQ) were completed by parent and child separately so they may not have been aware of discrepancies between their own perceptions, and those of the other

family member, in relation to the specific tasks of diabetes self-management. Studies did not identify the tasks for which disagreements were found, with the exception of Anderson et al. (1990) who reported disagreements regarding responsibility for 'regimen tasks' but did not identify which specific regimen items were linked with higher levels of disagreement. Therefore, the interrelations between 'who does what with whom' in diabetes self-management was not directly studied, and this remains a gap in the understanding of family management of youth diabetes.

Methodological issues of the studies reviewed

The cross-sectional design of the studies reviewed has afforded consideration of the associations between the diabetes variables under investigation but the causal direction of associations cannot be established. Based on the findings of this review, further evidence is required to establish the mechanisms by which discrepant views of family members influence diabetes outcomes such as self-management, glycaemic control and family psychosocial functioning.

Study methodologies and examination of the variables of interest varied considerably. As a consequence, it is difficult to draw comparisons across the studies and the process of drawing conclusions is limited based on the strength of the evidence. The extent to which the results can be generalised to the wider youth diabetes population was consistently questioned across appraisal of the methodological domains of the studies reviewed. In addition, studies reported crossover in the use of data from samples of other studies being undertaken by the author(s). For example, two studies examined the baseline assessment data

of an on-going intervention study (Anderson et al., 2009; Butner et al., 2009). Beveridge et al.'s (2006) data is drawn from the same sample of participants as Palmer et al. (2004). Multiple use of data sets may be fairly common research practice but serves to further narrow the population from which these findings are ascertained, thus placing additional limitations on the generalisability of results.

The possibility of demand characteristics in the methodology of dyadic study designs has been considered. That is, children and parents may have wanted to present a more socially acceptable 'self' and 'family unit' to the researchers and health care team in terms of family management of the child's condition, such that social desirability influenced participant self-report of who does what and whether they agree on it. Power imbalances within the doctor/researcher and patient/participant relationship are also likely to play a role in research of this nature.

Many of the studies were not clear in stating whether the influence of other family and diabetes variables was controlled for. Family agreement/disagreement related to diabetes responsibility may be affected by multiple factors such as family structure, duration of diabetes and treatment modality (i.e. multiple injections or pump) which future studies should control for within their analyses to help address the risk of potential bias.

A reliance on self-report measures for disclosure regarding distribution of diabetes responsibility is commonly cited as a limitation within the wider diabetes responsibility literature (e.g. Helgeson et al., 2008; Vesco et al., 2010; Ingerski et al., 2010). Introduction of behavioural observation methods into study designs may help to compensate for this in future research.

Clinical implications

Low-intensity office-based interventions⁴ showed promising evidence of improved family management of diabetes and supports the evidence of increased family agreement regarding family sharing of diabetes responsibility to be associated with improved metabolic control (Anderson et al., 2009). Furthermore, the potential utility of integrating such approaches into the existing model of clinic sessions is likely to be of interest, as striking the balance between interventions that are cost and time efficient as well as efficacious continues to present a challenge for clinicians, service managers and organisations currently operating within financially constrained health care services. However, different groups within the paediatric diabetic population may require support of varying intensity and duration across the challenging period of adolescence, in order to achieve and maintain better diabetes outcomes.

Clinicians are well-placed to facilitate conversations with families eliciting their perspectives on family sharing of diabetes responsibility regarding agreement, discordance and the perceived value of family member involvement. This would provide valuable additional information for family-based interventions targeting family communication and negotiation in efforts to improve glycaemic control during adolescence (Anderson et al. 2009). Specifically, as older children (e.g. 9-11 years) transition into adolescence, explicitly discuss and clarify how responsibility for diabetes management tasks will be shared within the family, specific to the dynamics of that family system (Anderson et al., 2009).

⁴ Term used by Anderson et al. (1999) and Laffel et al. (2003) to describe family-centred interventions designed to be incorporated with the routine clinical care of youth diabetes. That is, delivering the intervention during the regular clinical ('office-based') appointments attended by children and their families.

Findings of increased parental distress and depressive symptoms associated with family member discrepancies suggests the need for routine screening of parental psychological wellbeing as part of clinical care in order to support parents in their fundamental role in family management of diabetes.

Future research

The direction of the association between family discrepancies in sharing of diabetes responsibility and the range of variables explored is not possible to determine from these cross-sectional studies. Longitudinal research is required to examine the causal and directional relationships between family perceptions and diabetes variables (Miller & Drotar, 2003). The evidence-base for family-based diabetes interventions may benefit from longitudinal research by helping to determine the intensity and duration of support that families require in order to establish lasting changes in family patterns of interactions and behaviour related to family management of diabetes, and whether this varies across demographic and family variables. The inclusion of more diverse participant groups will increase the representation of the wider spectrum of youth with diabetes population within the literature.

Additional research could usefully combine qualitative methodology and within-family designs to conduct more in-depth explorations of the specific forms of family disagreement regarding diabetes responsibility that this review has highlighted to be potentially pertinent to metabolic control and parental psychological wellbeing. More clarity regarding what diabetes family responsibility should entail would help to develop understanding and compare findings, as the construct is measured and examined differently within the existing literature.

Limitations of the review

The very specific focus in exploring family agreement and disagreement regarding sharing of diabetes responsibility at a within-family data level meant that a small number of studies were available for review. This clearly represents a limitation. Generalisability of the findings to the wider adolescent with T1D population represents a major limitation of the studies and may be further compounded by the relatively wide age-range of child/adolescent participants across studies included in this review. However, only two of the thirteen studies included participants aged over 18 years (Anderson et al., 1990; Schilling et al., 2006).

CONCLUSION

The links between diabetes family responsibility agreement/disagreement and HbA1c are tentatively presented as few studies have reported significant results. The review has highlighted interesting findings related to parental psychological wellbeing and family psychosocial functioning. That is, parents appear to experience increased psychological distress and perceive greater diabetes family conflict in response to discrepancies regarding diabetes family responsibility, compared to their adolescent. Furthermore, there is evidence to indicate that a specific type of discrepancy between mother and adolescent is linked with decreased parental psychological well-being and increased diabetes family conflict. Diabetes health care professionals should explore divergent family perspectives regarding diabetes family responsibility that may have a detrimental effect on diabetes outcomes. Additional research with more robust designs, including within-family data-collection and analytic methods, is required to further develop the evidence-base concerning associations between within-family agreement/disagreement and diabetes outcomes for the adolescent and family system.

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LIVING WITH COELIAC DISEASE AND TYPE 1 DIABETES: THE EXPERIENCES OF ADOLESCENTS AND THEIR PARENTS

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ABSTRACT

Objective

Type 1 diabetes (T1D) and coeliac disease (CD) are chronic health conditions commonly diagnosed in youth, and a small population of adolescents live with both of these conditions. Management of T1D and CD occurs in a relational context within the parent-child relationship and wider family system. This study aims to develop a better understanding of how a group of adolescents and their parents make sense of the challenges associated with self-management of the dual diagnosis, specifically how these two conditions are experienced as a dual-diagnosis, from a family perspective.

Method

This study utilised a qualitative design informed by the principles and methods of an interpretative phenomenological approach. Eight individual semi-structured interviews were conducted with four adolescents with a dual diagnosis of type 1 diabetes and coeliac disease, and their parent.

Results

Dyadic analysis of the interviews of adolescents and their parents was guided by interpretative phenomenological analysis (IPA). Two super-ordinate themes emerged from the accounts of adolescents and their parents: 'Perpetual protection and loss' and 'Duality: together but separate'.

Conclusions

The findings illustrated how adolescents living with a dual diagnosis of T1D and CD, and their families, are tasked with negotiating the continual and fluctuating challenges of management of these two conditions. Families appear to focus on T1D and its management but losses in the relationship with food and eating emerged as the most prominent theme within families' experiencing of CD. The chronicity and variability in management of T1D and CD appears to be burdensome for families, particularly parents.

Keywords: Type 1 Diabetes, Coeliac Disease, dual diagnosis, adolescents, parents

INTRODUCTION

Type 1 Diabetes (T1D) is one of the most common chronic health conditions among children and adolescents. Treatment is multi-faceted and demanding, requiring a high-degree of self-management. Effective management of T1D requires monitoring and controlling of carbohydrate intake, monitoring blood glucose, administering insulin, engaging in physical exercise and adjusting insulin levels. Such complexity can be overwhelming and burdensome for young people and their families (Anderson et al., 2002; Debono & Cachia, 2007; Greening, Stoppelbein & Reeves, 2006). Deteriorating metabolic control in adolescents has been associated with physiological factors associated with puberty (Amiel, Sherwin, Simonson, Lauritano & Tamborlane, 1986), poorer self-care behaviour (Anderson et al., 1997) and psychosocial variables (Helgeson, et al., 2010).

Findings in the literature exploring possible psychosocial difficulties in adolescents with diabetes are mixed. Helgeson et al. (2007) found no differences in psychological distress or behavioural problems between adolescents with and without diabetes, but adolescents with diabetes reported increased social difficulties and disturbed eating. Parental psychological distress in parents of children with diabetes has been found to be associated with negative child outcomes including: increased stress and depressive symptoms, behavioural difficulties and lower quality of life (Whittemore, Jaser, Chao, Jang & Grey, 2012). The presence of depressive or anxiety symptomatology in parents can mediate the impact of parental distress on family diabetes management. That is, parental depressive symptomatology was associated with reduced frequency of parental monitoring of diabetes related self-management. Conversely, parental anxiety

was linked with higher parental involvement but lower parental self-efficacy for diabetes management. Thus, parental psychological distress may produce additional barriers to successful family management of diabetes, however, the relationship with metabolic control remains unclear (Whittemore et al., 2012).

Coeliac disease (CD), an autoimmune condition, causes the body to attack itself in response to ingestion of gluten. Antibodies are produced which cause damage to the small intestine and a range of long-term health complications are associated with continued exposure to gluten such as infertility, compromised bone strength (e.g. osteoporosis) and increased risk of gastrointestinal lymphoma (Coeliac UK, 2013). Adoption of a strict life-long gluten-free diet (GFD) is required for treatment of CD. Adolescence appears to be a particularly difficult time for dietary self-care and levels of dietary self-management have been shown to vary depending on the social situation the individual is in (e.g., Saukkonen et al., 2002). Evidence suggests that young people are more likely to eat food containing gluten when with friends (Ljungman & Myrdal, 1993), or when eating out, in order to avoid social difficulties (Mayer, Greco, Troncone, Auricchio & Marsh, 1991). Poor dietary self-management has been associated with lower quality of life, increased physical health problems, greater burden of illness, more family problems and difficulties in leisure time, compared to those reporting good self-management (Wagner et al., 2008).

Olsson, Hornell, Ivarsson & Sydner (2008) qualitatively explored adolescents' experiences and perspectives of managing CD through a GFD. Adolescents' differing views of everyday life with CD and the GFD were linked to their varied approaches to following the GFD. Poor availability of gluten-free food and a sense of social embarrassment and stigma about their condition were

identified as challenges to successful management of the GFD. Themes relating to managing their identity as a young person with CD and ambivalent relationship with food have also emerged from a qualitative study exploring the experiences of young people with CD (Theodosi, 2009).

The prevalence of CD is greater in children with T1D, than in the general population (Saukkonen et al., 1996) and is estimated to range between 1 – 10%. This association has been observed since the late 1960s with evidence of genetic factors in both conditions, related to the system that mediates autoimmune diseases. As children with T1D are known to be at higher risk for CD, routine screening for CD as part of their medical care is becoming increasingly widespread. National Institute for Clinical Excellence (NICE) guidelines recommend serological screening for CD at diagnosis of T1D (NICE, 2004). Some young people with both conditions do not show the common noticeable symptoms (i.e. are asymptomatic) for CD, and in such cases, the CD is usually identified by routine serological testing for coeliac antibodies. To date, there has been a focus within the literature, exploring T1D and CD in children and adolescents, on debating the challenges and controversies in screening and diagnosing CD in this group of young people (see Sud et al., 2010 for a review of this literature).

Psychosocial issues have been shown to be fundamental to young people's experiences of living with either T1D or CD as single diagnoses. Therefore, it could be expected that a dual diagnosis of both conditions may lead to additional difficulties. However, research exploring the impact of both T1D and CD in children and adolescents is limited. Emerging literature has mainly employed quantitative techniques when exploring the impact of CD and the GFD on factors

such as quality of life, growth and glycaemic control in this population (Sanchez-Albisua, Wolf, Neu, Geigert, Wascher and Stern, 2005; Saukkonen et al., 2002; Sud, Marcon, Assor, Daneman & Mahmu, 2012), with mixed findings. Families have reported experiencing difficulties with introducing and maintaining the GFD, identifying limited availability of gluten-free food and the complexity of ensuring appropriate dietary provisions outside of the home environment as challenges to effective dietary management (Saukkonen et al., 2002).

Sud et al., (2012) quantitatively explored the impact of managing CD and T1D on quality of life in children and adolescents, with a focus on self-management of the GFD and metabolic control. Twenty-eight children with confirmed diagnoses of CD (> 6 months) and T1D (>1 year) were compared to a control group of forty children with T1D (age range: 8-18 years). Child and parental self-report of the child's general and diabetes-specific quality of life were assessed. Symptoms at the time of CD diagnosis and self-management of the GFD were also explored. Results showed there was a high rate of adherence (79% as assessed by a dietician) with the GFD reported at the time of quality of life assessment, and that there were no significant differences in quality of life between children with CD and T1D and those with T1D alone. Age at CD diagnosis, CD duration and adherence with a GFD were not shown to be associated with quality of life. The authors argued these findings demonstrate a minimal impact on quality of life of an additional diagnosis of CD in children with T1D. As dietary self-care is an existing feature of diabetes management it was proposed that the introduction of a GFD may not result in additional burden. However, the parents of children with a dual diagnosis did report lower social functioning scores for their children compared to the T1D group. Significant

differences between parent and child ratings were found for some quality of life items. Parents of children with CD and T1D scored their child's psychosocial health and social functioning lower than their child, indicative of poorer functioning.

There are difficulties in comparing findings of the existing literature in this area due to small sample sizes (e.g. Saukkonen et al., 2002) and the lack of validated measures utilised to explore the impact on quality of life (Sanchez-Albisua et al., 2005).

In acknowledgement that adolescence has been identified as a difficult time in the management of both conditions, the potential for an increased sense of burden and negative psychosocial experience could be usefully explored in greater depth in adolescents managing a dual diagnosis of T1D and CD. Furthermore, initial findings of differing perceptions between parents and adolescents regarding aspects of self-management and adolescent psychosocial functioning in T1D and CD (Sud et al., 2012) suggest that within-family perspectives may represent an important focus of future research in this area, given the role parents play in management of their child's conditions.

Rationale

As much is already known about the psychosocial issues associated with these two conditions as single diagnoses, the aim of this study is to develop a richer understanding of the experiences of living with a dual diagnosis of T1D and CD, from the perspective of adolescents and their parents. This will include a broad interest in understanding the challenges faced in everyday life associated with T1D and CD, perceptions of T1D and CD as a dual diagnosis and whether there is a greater focus on management of one condition over the other. This

study will attempt to address the lack of research exploring a dual diagnosis of T1D and CD through an in-depth qualitative exploration of the experiences of these adolescents and their parents. The focus on the dual diagnosis of T1D and CD combined with a qualitative approach to gaining intra-familial perspectives on living with these conditions offers an innovative exploration of this area.

It is hoped that this research will help to increase clinical and theoretical understanding about what it is like to live with these two chronic health conditions and improve how health professionals support adolescents and their families to stick to the demanding treatment regimes in the management of both conditions.

METHOD

Design

This in-depth qualitative study utilised an Interpretative Phenomenological Analysis approach. The commitment of this approach to capturing and attempting to understand the 'lived world' (social and psychological) of the participant and the meaning they make of their experiences (Smith, Flowers and Larkin, 2009) through its methods of data collection and analysis was considered appropriate in developing understanding of how adolescents and their parents experience a dual diagnosis of T1D and CD. That is, combining phenomenological methodology with a dyadic design to explore, and understand, what is different and similar about the experience of living in a family where there is a child with both T1D and CD. This exploratory process of intra-familial perspective-taking and meaning-making is undertaken from the viewpoint of the adolescent with T1D and CD, and their parent.

The phenomenological approach is transparent in its focus on understanding the meaning individuals make of their experiences within the complexity of their context, with an acknowledgement of the role of the researcher in influencing the interpretations of that individual's experience through their own theoretical and experiential 'viewpoint'. This is referred to in terms of a 'double hermeneutic': the researcher making sense of the individuals 'sense-making' (Smith et al., 2009). This analytic process requires a balance between the researcher's use of 'self' (their thoughts, feelings and experiences) for reflexive engagement with the data and the importance of the analysis primarily being about the participant. An intense focus on a small number of cases is considered

advantageous in studies of this nature, thus, sample sizes are typically small (Smith et al., 2009).

Context

Young people living with a dual diagnosis represent a relatively small sub-group within the wider youth T1D and CD population, posing additional challenges to optimising the opportunities for recruitment. Families were approached from a specialist paediatric dual diagnosis clinic and through local charitable T1D and CD support groups. The clinics were developed specifically for young people with a dual diagnosis of T1D and CD to provide specialist advice and support with the management of these two conditions. Clinics are held quarterly at a children's hospital and facilitated by a consultant gastroenterologist and a consultant diabetologist, specialist diabetic nurses and a dietician. Local branches of the charitable organisations Coeliac UK and Diabetes UK, whose members include young people with both T1D and CD and their families, provided an alternative environment from which to approach people who may wish to share their experiences.

Participants

A total of eight participants took part in the study, consisting of four pairings of parent and adolescent. The adolescents, two male and two female, were aged 12 – 18 years (mean age = 15.8) and had confirmed diagnoses of T1D and CD, with mean duration since diagnosis of 9.8 (SD = 5.9) and 6.8 (SD = 4.9) years, respectively. Three of the adolescents were on insulin pump treatment whilst one adolescent administered daily insulin injections. Three of the adolescents had

received the T1D diagnosis prior to that of CD, the fourth child was first diagnosed with CD and then T1D three weeks later. For two of the adolescents their CD is asymptomatic, meaning that they do not experience noticeable immediate physical symptoms as a consequence of gluten consumption. Objective data regarding current level of metabolic control (as indexed by HbA1c values obtained from medical records) was only available for the two young people recruited from the specialist dual diagnosis clinic: 8.0% and 9.6%. An objective measure of GFD self-management (such as serological test results for coeliac antibodies) was not available for any of the adolescents who participated. This demographic and contextual information as it relates to each individual adolescent is outlined in Table 1. Names of all participants have been changed to protect anonymity. The parent-adolescent pairs were: Susan and Sarah, Caroline and Charlie, Mary and Michael, Richard and Rachel.

The parent sample comprised three mothers and one father, all of whom lived with their child with a dual diagnosis. Parents were aged 42-48 years (mean age 45.5 years). Three of the parents were married, one was divorced. Three parents' occupations were of a professional background, one parent was a full-time carer for their child with the dual diagnosis. All of the families were of White British origin. Although information pertaining to social class was not explicitly requested of those who participated, families were subjectively rated as 'middle class'.

Table 6. Adolescent demographic information and condition status

Adolescent (<i>participating parent</i>)	Age	Gender	Treatment	Age at diagnosis		CD symptomatic/ asymptomatic
				T1D	CD	
Sarah (<i>Susan</i>)	17	Female	Insulin pump	18 months	6 years	Asymptomatic
Charlie (<i>Caroline</i>)	12	Male	Insulin pump	9 years	9 years	Symptomatic
Michael (<i>Mary</i>)	18	Male	Insulin pump	11 years	16 years	Asymptomatic
Rachel (<i>Richard</i>)	16	Female	Insulin injections	2 years	5 years	Symptomatic

Measures

Parent and child interview topic guides were developed (see Appendices 6 & 7) affording a flexible framework to the interview process and serving as prompts to the researcher. Question development was informed by review of relevant literature and covered several areas related to the psychosocial issues of living with T1D and CD, with a focus on facilitating exploration of nuances in participants' experiences specifically related to living with both of these conditions. Questions covered diagnosis, explaining the conditions to others, impact on activities and relationships, management of the conditions and concerns/worries. Topic guides were reviewed by a specialist clinician experienced in working with this population and a parent and young person representative of this demographic who did not participate in the study.

Procedure

Ethical approval was obtained through an NHS Research Ethics Committee and full Research and Development approval was granted by the NHS organisation which provides the dual diagnosis clinic (See Appendix 8). Eligible

families attending the dual diagnosis clinic were approached during clinic sessions by their consultant who informed them of the potential to participate in the study and provided them with age-appropriate information sheets: parent (Appendix 9), 11-13 year old (Appendix 10) and young person (Appendix 11). The researcher was present at the clinic site to meet with families who expressed an interest in participating. Across the two dual diagnosis clinics that coincided with the research timetable, five families were invited to participate and 2 families agreed to participate (3 families declined for reasons unknown).

Volunteer co-ordinators of two local T1D and CD support groups were contacted and forwarded parent and young person letters of invitation to participate (Appendices 12 & 13) and information sheets to group members via email. Those who wished to express an interest in participating were advised to contact the researcher directly. One of the groups held a database of families who have previously consented to be approached regarding participation in research. However, due to issues of confidentiality and it not being possible to target email correspondence to those members specifically with a dual diagnosis, one group's global mailing list was utilised. Thus, it is not possible to determine exact numbers of how many potential participants were approached and offered the opportunity to share their experiences.

Inclusion criteria for young people and their participating parent are presented in Figure 3.

Figure 3. Inclusion criteria

Inclusion criteria
<ul style="list-style-type: none">• Young people between the ages of 11-18 years• Confirmed clinical diagnoses of CD and T1D, including symptomatic and asymptomatic cases of CD• Clinical diagnosis confirmed via serological screening (T1D and CD) and biopsy (CD)• Diagnosis for both conditions must have been made for at least one year (as a recent diagnosis may impact understanding of experience of and management of the conditions)• The parent being interviewed does not have a single or dual diagnosis of CD and/or T1D (as this would likely have a significant impact on the meaning and experience of the dual diagnosis for the young person and their family)• No co-morbidity of other long-term physical or mental health conditions for the young person• Participants demonstrate an understanding of, and ability to speak, English

All interviews were conducted by the author and took place in participant's homes. In order to facilitate confidentiality, parent and child interviews were conducted separately in rooms which afforded as much privacy as possible. Data was collected via semi-structured interviews, which lasted approximately 60 minutes per participant. Interviews were digitally audio-recorded for the purposes of transcribing verbatim as required for the selected method of analysis.

Ethical considerations

Informed consent was obtained prior to participation; parents gave written consent for themselves and for their children aged under 16 years, whilst young people aged 16 years and over gave their written assent (see Appendices 14 and 15 for parent and young person consent forms). Whilst this study aimed to explore the experiences of a dual diagnosis of T1D and CD from a family perspective, the presence of the other participating family member during interviews may have influenced the interviewee's responses. In order to minimise

this and to encourage full and honest accounts of their experiences, it was a condition of participation that parent and child interviews would be conducted separately.

It was expected that issues relating to the treatment and management of CD and T1D would be raised by parent and child when discussing their experiences. This had the potential for disclosures of risk behaviour concerning dietary and insulin self-management. Appropriate safeguarding and information sharing pathways were approved as part of the governance processes in obtaining ethical approval for this study. Strategies to manage any significant emotional distress that may arise as a result of discussing personal issues of a sensitive nature related to these health conditions were also in place (additional information and sign-posting to further support systems). The therapeutic skills of the author were also utilised within the research context and appropriate to the role of IPA researcher.

Despite all reasonable precautions being taken (i.e. conducting parent and child interviews separately), the pairing of parent and adolescent interview data posed a threat to anonymity. The potential for participants to identify their parent's/adolescent's responses within the final report was made explicit in the study information sheets and reiterated by the researcher during the pre-interview consultation. Further, participants were sent a copy of their transcripts and afforded the opportunity to identify any quotes of a sensitive nature that they wished to have removed from analysis or inclusion in the final report. None of the participants requested for any of their responses be withdrawn.

Analysis

Interview data were analysed using IPA, guided by a six-step process suggested by Smith, Flowers and Larkin (2009). This analytic process is presented in Figure 4 below. These stages are not proposed as a prescriptive method for a linear process, rather an iterative and inductive cycle (Smith, 2007) to ensure interpretation is stimulated by, and tied to the text. Discussions with academic and clinical supervisors supported this interpretative process.

Figure 4. Six-step analytic process of IPA (as described by Smith et al., 2009)

Step 1 - *Ensuring the participant and their experience becomes the focus:*

- Immersion in the data through listening to the audio-recording whilst engaging in repeated readings of the interview transcript.

Step 2 – *Deeper exploratory examination of content and language:*

- Comprehensive noting of interesting aspects of the participant's accounts with commenting focusing on the:
 - *Descriptive*: things that matter to the participant (e.g. relationships, values, events etc.) and what these things are 'like' for the participant
 - *Linguistic*: how language is used to convey content and meaning, attending to pauses, laughter, repetition, tone and fluency of speech etc.
 - *Conceptual*: moving analysis towards the interpretative, development of provisional conceptual questions and deconstruction
- This process is conducted on a line-by-line basis, with comments recorded in the left-hand margin of the transcript.

Step 3 - *Developing emergent themes:*

- Focus shifting from core transcript data to mapping of the patterns between initial noting and generation of emergent themes (sequentially ordered as they have arisen in the data).

Step 4 – *Drawing together of emergent themes*

- Exploring patterns and connections between the emergent themes via:
 - *Abstraction*: generation of a super-ordinate theme from grouping of similar emergent themes
 - *Subsumption*: an emergent theme unifies a group of similar themes and becomes super-ordinate
 - *Polarisation*: noticing divergence which indicates oppositional relationships between emerging themes
- Use of visual aids (i.e. themes presented on post-it notes) to facilitate the process of making connections between themes

Step 5 – *Progressing to the next case*

- Analysis of each participant on an individual (idiographic) level, striving for subsequent analysis not to be constrained by the influence of ideas that have arisen in the accounts of others, so that new themes may emerge with each case.
- Use of reflexive journal to facilitate this process

Step 6 – *Exploring patterns of themes across cases*

- Patterns of themes identified across adolescents, parents and adolescent-parent pairs and development of super-ordinate themes
- Use of colour-coding to facilitate easy identification of adolescent-parent dyads

Reflexivity

As a young child it was discovered that I had significant allergies to specific artificial additives that are commonly found in major food and drink groups. Although the consequences of my allergy did not pose any major threat to my immediate or long-term health and did not place significant limitations upon me, I can recall my own experiences of certain food and drink becoming 'forbidden' and of feeling 'different' to my siblings and peers. Within my wider network of friends and family I know of those who live with T1D or CD as separate diagnoses and have witnessed the challenges they continually face in management of their conditions and how a lack of knowledge and understanding in others and wider society can be stigmatising.

Soon after I began meeting with families to conduct the interviews I discovered that I was pregnant. I became aware of how my emerging identity as a mother may influence how I connect with some of the parenting 'scripts' (spoken and unspoken) expressed within the parents' stories of caring for their child who is living with these chronic health conditions. Recording my thoughts and reflections in a journal across this process helped me be mindful of, and to 'hold', these influences across data collection and analysis. Use of a reflexive journal to reflect upon the experiences, values and assumptions I brought to this process is considered to be 'good practice' within IPA (Brocki & Wearden, 2006).

I am aware that these experiences and my emerging professional interests have likely influenced my interest in this area of research. Within IPA, the experiences and views of the researcher are an expected and accepted influence in the interpretive 'sense-making' process of understanding people's experiences, rather than being considered a source of bias in analysis (Smith et al., 2009).

RESULTS

Analysis of these families' accounts of their experiences of living with a dual diagnosis of T1D and CD resulted in the emergence of two super-ordinate themes: 'perpetual loss and protection' and 'duality: together but separate'. At the sub-ordinate level, a total of six themes were found to emerge and are presented in Table 7. The selection of these themes reflected their prevalence and perceived significance to the adolescents and their parents. Although the majority of sub-ordinate themes represented shared experience between parents and adolescents, the theme of 'Living in the present vs. connecting with future threat' was unique to the adolescents.

The individual presentation of these themes is not intended to imply mutual exclusivity, rather there was found to be significant overlap between themes due to the complex and dynamic nature of the families' experiences. Whilst the majority of sub-ordinate themes were embodied within the accounts of all of the adolescents and parents, some were not.

Table 7. Super-ordinate and sub-ordinate themes

Super-ordinate	Sub-ordinate
Perpetual loss and protection	
<i>Parent & Adolescent</i>	<ul style="list-style-type: none"> • Threat to hopes and expectations for future health and family lifecycle • Protection from consequences of T1D and CD • Loss in the relationship with food and eating
<i>Adolescent</i>	<ul style="list-style-type: none"> • Living in the present vs. connecting with future threat
Duality: together but separate	
<i>Parent & Adolescent</i>	<ul style="list-style-type: none"> • CD as a 'consistent constant' vs. T1D as a 'variable constant' • Focus on T1D vs. T1D and CD

Diagnosis

Adolescents' and parents' experiences of living with both T1D and CD as a dual diagnosis are situated as the central focus of analysis within this report. However, in order to place these experiences in context it is important to first consider these families' stories of how the adolescent received their diagnoses. Each interview commenced with the participant being invited to share their understanding of the 'events' surrounding the adolescent's diagnosis of both conditions.

The onset and progression of the conditions (typically T1D) was experienced to be very sudden with symptoms manifesting quickly and intensely such that one day the child was 'healthy' and the next they were 'different', evoking a sense that this represented the start of irreversible change for the adolescent and their family system. For Mary, this difference in her child, Michael, was so remarkable that he was almost unrecognisable to her:

he went off on a school trip which my husband went on as well and he stepped off the coach, Michael did, and I just thought that's not my child; he'd lost so much weight and he came over to me. Normally Michael was very affectionate and I was expecting him to come and have a hug or something and he came up to me and said, "Mum, let's go home." Like that and it was all a bit of a shock really. He came home and he was quite, he just wasn't himself. He was quite aggressive, I mean verbally and just not himself. We put it down to tiredness for a few days but it just didn't get any better (Mary)

Adolescents described vivid recollections of painful symptoms associated with onset of the conditions and the subsequent invasive medical procedures that they were subjected to as part of the diagnostic process. They were also very

aware of their parents' worry which generally exceeded that which they themselves were experiencing. This was especially true for those who were of a young age when they were diagnosed, as they recognise that they did not fully understand what was happening at the time or the implications of what their diagnoses would mean for their lives.

A few of the parents' narratives were dominated by a 'fight' or 'battle' to receive the diagnosis of one or both of the conditions and a sense of having had their concerns dismissed or minimised by healthcare professionals. This inevitably added to their distress and sense of helplessness as they struggled to receive help in finding answers whilst their child's symptoms persisted and their health deteriorated further. A range of emotions felt by parents were named or expressed when recounting their experiences of their child's diagnoses: anger, shock, blame and guilt. The intensity of emotional experiencing and apparent 'reliving' of the diagnoses for parents was not only expressed through their language but was also evident in other linguistic nuances such as how that language was used.

The immediate aftermath of diagnosis signified a point at which family-life changed irrevocably. It also represented the beginning of a new way of being for these families that would persist up to the present day, characterised by 'just getting on with it' and focusing on the practicalities of day to day self-management. This overview sets the scene for contextualising the experiences that families share in the subsequent sections.

Super-ordinate theme: *Perpetual loss and protection*

A sense of overwhelming permanence related to the chronicity of both conditions and their management appeared to underlie parents' and adolescents' stories of the past, present and future. The understanding of this 'chronicity' developed further to represent the repeated experiences of 'loss' and continual need for 'protection' against the consequences of T1D and CD that is felt by parents and adolescents as a result of living with the dual diagnosis.

Threat to hopes and expectations for future health and family lifecycle

The perceived threat posed by T1D and CD to the hopes and expectations that parents and adolescents hold for the adolescent's long term health and future life events was a shared theme for both parent and adolescent, but relates to their experiences in different ways.

For parents, processes of loss and grief seem to have originated from their earlier experiencing of the child's loss of health at the point of diagnosis and then recurred in relation to what the losses associated with T1D and CD mean for their expectations and hopes for their adolescent's future. Mary describes grieving the loss of a healthy child, portraying a sense of cruelty in believing that her role in protecting her son's health had been fulfilled, only for his health to be taken away from him by T1D:

In a funny way bereaved; and I know that sounds a bit strong but you know I'd had this healthy child till then and you know the first few years you are backwards and forwards to the doctors and other people's children are getting meningitis and all these things and you think you've survived that and you've had chicken pox and you sort of think once you get them to the eight to ten mark that they've had all the

horrible things they are going to have and you've got this healthy child and everything is going to be fine. So when you find out your child's got a life-threatening disease, which is what diabetes is, if it's not treated that's it isn't it? So I did feel bereaved for Michael and all the things because you want your child to do everything and it was hard to feel that he'd had his health taken away from him

(Mary)

Sarah was only 18months old when she was diagnosed with T1D and Susan reflected on her grief that T1D denied her the 'normal' experiences of parenting, such as the very special time in the parent-child relationship whereby the parent delights in their young child:

yeh in a way I sort of grieved for that lost you know, when they were little that lovely time when you could, when you don't have any, you can just enjoy them and I never really, I feel as though I missed out on that a bit because... I feel that a lot of that time was taken up with worrying about the diabetes and you know, pricking fingers and getting them to eat and so yeh yeh (pause) (Susan)

Susan's experiences of loss were particularly emotive to bear witness to, as she explained that her dreams of having more children were given up due to fear that they may also have to endure ill-health and how living with the conditions has challenged her values of how she wanted to parent her children:

before I had children I vowed I wouldn't be one of these mums who's like "oh no, you've got to eat all that and you've got to.." you know I thought I am not going to be one of those but I did end up being one of those just because you have to be yeh, yeh because you have to be (Susan)

In terms of their adolescent's future, parents are concerned by the implications of the long-term health consequences associated with T1D and CD

for their adolescent's chances of experiencing the typical family lifecycle stages, such as becoming parents themselves. Susan is concerned about the long-term effects of poor self-management on Sarah's fertility, but recognises that this is probably not something a 17 year old is too concerned by:

I sort of try to talk to her about things like, you know, fertility, I say "well you know you don't want a baby now Sarah but in 10 years' time when you come to, if you're not really careful it might not happen" but I think, you know, when you are 17 you are not thinking about like that are you (Susan)

However, adolescents are aware of the potential threat their conditions pose to things that they may wish to experience as part of their futures. Rachel expressed her enthusiasm to travel the world when she is older but is uncertain "it's going to work" because of her conditions. She has given thought to having children but finds it difficult to contemplate the idea of them going through what she does should they develop the conditions.

Despite holding some concerns regarding their futures, this was balanced by a few of the adolescents who were also able to describe their future potential with more hope. That is, viewing the barriers of the conditions as things that can be overcome. Sarah finds it annoying, that compared to her peers, she has to take additional precautions to travel but is determined to find a way to circumvent the obstacles:

it does sometimes feel a bit like, ok, fine but everyone else can just do what they want but I don't, like, if everyone else decides you know to go travelling coz we've got four months where we can just travel but everyone else can just take a backpack and go where they wanna go whereas I can't take four months' worth of insulin, set changes, you know, dextrose in a backpack with me, I've got to take

seven suitcase which is going to, you know, be a pain um so in some ways it can like limit but there's always ways around it, always ways around it, like if its just an inconvenience or whatever you can always find something to do it a bit differently so like I am taking a year's worth of insulin... and we are going to get round it somehow (laugh)...I don't think I'd ever let it stop me doing anything, I think im too stubborn for that (laugh) to be fair like yeh its always going to come second to whatever I want to do. (Sarah)

Charlie spoke of how receiving both of his diagnoses in relatively quick succession and having lots of time to think while he was staying in hospital initially left him feeling despondent about the impact on his future. He looks to famous people with diabetes, such as Sir Steve Redgrave, as positive role models for what can still be achieved:

...because it had a huge impact on my life and it... before I found out about everyone else who had achieved lots of great things with diabetes, uh, I thought that all my plans for life were just going to be ruined. But ((slight pause)) they weren't really... (Charlie)

In contrast to the losses, Michael shared examples of situations whereby his T1D and CD resulted in 'special treatment' from others, such as being served a meal from first class on a plane journey as a gluten free option was not available. He and Mary also explained that through his T1D Michael has engaged with meaningful activities and Mary reflects on how she perceives this to have possibly had a positive influence on Michael's self-confidence and determination:

Well, the weekends I go on where I look after the children, I mean that's great because you're looking after these kids for a weekend and they've got diabetes and it feels really rewarding to be able to give them a good fun weekend and

watch them sort of... And they almost forget about their diabetes for a weekend sometimes, without their parents just fussing over them all the time, they sort of... They grow up a little bit I suppose with their diabetes which I quite like. (Michael)

((pause)) Well I think it's given him confidence in a funny sort of way because he was always quite quiet and quite, a bit of a Mummy's boy really, I think he was before he got his diabetes. But because he's had to deal with it, I mean he might have changed anyway and I think he likes, it gives him something to, he's never been scared of talking about it to people, which I think is good. He's never hidden it and I know when he does volunteering, he's happy to be around other people with the same issues and it has given him confidence and also probably a bit of a determination that he is going to go and do things and it's not going to stop him as well, which is good. It's hard to say whether he would have done that anyway, its really hard to explain (Mary)

Protection from consequences of T1D and CD

This theme covers a broad spectrum of 'protection' and conveys the efforts of parents and adolescents to try and prevent or limit the negative consequences associated with the conditions and their management. It connects to parents and adolescents differentially due to their respective positions in the parent-child relationship and responsibilities in relation to self-management of T1D and CD.

Parental over-protection and hyper-vigilance to self-management appears to represent an extension of the normal protective role of parents and may have served as a strategy for coping with the 'near loss' experience of these parents when their child was so unwell at the point of diagnosis. Mary's extract below

illustrates her strong protective instincts towards her child, such that she would sacrifice a part of her own body in order for Michael to have his full health:

but I mean obviously if someone said to me, "Would you like Michael to be fit and healthy again, to have the diabetes and coeliac taken away?" Of course I would, I'd do anything, I'd give him my pancreas, I'd do anything because you want your child to be healthy, you want your child to have everything and to have a good life and everything so I would. (Mary)

Parents expressed fears of their adolescent being exposed to risk or harm when responsibility for the protection of their adolescent's health needs is in the hands of others. Susan voiced her concerns about what may happen if Sarah becomes ill when travelling abroad and "nobody else recognises it". Caroline takes active steps to keep Charlie 'safe' when she is not around, if she feels that she cannot fully entrust responsibility to others:

I programme the pump, I programme it differently when he goes to his dads to what I would do at home just to run it a little bit higher, um, to keep him out of danger. Um, his dad does now get up once in the night to do a check on his blood. That took a long time to get him to do that. He'd forget to set his alarm, or he'd be too tired to do it. Um, so yeah. But I send him with a flow chart and an instruction thing to fill in for each of his blood tests and meals and to what to do in the night and what to press, what buttons to press, and what to do if this, what to do if that.

(Caroline)

Furthermore, some parents find it difficult to relinquish some of their responsibility for protection and struggle with the loss of control over self-management as their adolescent gets older and seeks more autonomy. A sense of parental 'stuckness' is engendered in a few of the parents' accounts which

seems related to having held responsibility and shared illness ownership for so long. Susan explained that T1D has had such a consuming role in her life that “for so long it’s been my diabetes in a way”. Below she reflects on how this causes her to struggle with ‘letting go’ as Sarah becomes more independent:

it’s hard because you know, it’s always sort of been as much my diabetes really, in a way, so it is hard you know stepping back and letting her get on with it...um yeh it is hard because you want to, you sort of want to let go and let them get on with it but then sometimes when you do that it all goes to pot a bit you know they just don’t, don’t do it, sometimes I just think I don’t know if my way is the right way or whether I am too, I’m too controlling with it and I should just sort of take you know massive steps back...you know when they are little it’s hard in another way then when they get to be teenagers ‘cause they’re going off and doing their own thing it’s you know, it throws up other problems really so you know, I don’t know, maybe I am just too controlling (laughs) ooooh I just, I want it all to be perfect but then you know it, you just want them to be well really (Susan)

Mary describes how her involvement in Michael’s self-management has gradually shifted from being shared to Michael becoming more self-reliant, although she still engages in some tasks out of habit and will always be there for him should he need her to be:

Probably not as much as I used to because obviously he deals with it himself now but when he was first diagnosed it was very much a joint thing where I’d tell him to check his blood sugars and help him with, he did the injection himself but we’d decide between us how much insulin he needed and when we were going out I’d make sure I’d got hypo stuff and things like that, which I still carry some but I expect Michael to think about it now...Because I used to order everything but I

don't now, I let Michael do it, so I suppose I've got less to do with that really. I suppose I do tend to do things, I do it without really thinking about it now, weighing pasta and rice so he knows how much he's got; even though he's not here now it's just got into a habit that I weigh the rice and the pasta out.. I take him to his clinic appointments because he hasn't got a car but it's definitely got less and obviously if he needed me I'm here. (Mary)

Parents are aware that their efforts can be perceived negatively or as 'fussing' by the adolescent. The extract below illustrates how tensions can arise between parent and adolescent. Richard acknowledges how the loss of control over self-management causes him to struggle with finding ways to support Rachel with the aspects of self-management that worry him:

I think Rachel actually needs reminding from time to time, as now, being a teenager, it's more, I find, I find it's more difficult now in this age range, personally, so I worry a lot about her and tend, as well, not to sleep at night at times, worrying about her. I do remind her, if not every night, most nights, whether she's taken her long-acting insulin, because to me that's a very, very important injection that's going to affect the following, so that injection does worry me and there has been the odd occasion where, you know, being, you know, teenagers I think can, do, do forget, so it's difficult. So from that point of view, I've been finding it very, very difficult in terms of trying to help her. I view that myself as trying to help her by reminding her, but it can work in a negative way as well, in that Amy can get, you know, there's a wrong reaction from her about, um, just being reminded, really
(Richard)

Rachel acknowledges her father's support and is aware of his concerns when she is away from her parents but she can become annoyed by persistent reminders as she knows what she has to do:

he helps me, like explain it to me, and he always checks before I go to bed if I've taken my insulin, so then I'm not ill, and if I'm taking the right amount, and he cares a lot and he always worries about me... Yeah, it's just a bit annoying after a while, because my Dad keeps on asking me before I go to bed, and I know that I'm going to take it, and it just gets annoying after a while, and he's saying, 'Oh, take your insulin, take your insulin,' and it's annoying because I know I should take it and I wouldn't forget to take it, yeah. (Rachel)

The overall balanced way in which adolescents can appear to perceive the threat and limitations associated with their conditions suggests that parents' efforts have been psychologically 'protective' for the adolescent and have been incorporated into some of their attitudes and beliefs in how they respond to, and cope with, their conditions. For example, a 'script' of never letting T1D and CD prevent Sarah from doing anything was shared by Susan and Sarah:

...I know it's serious and whatever but I don't see it as life-debilitating or any of things coz we've never let it stop us do anything (Sarah)

Recognition of the substantial thinking, planning and preparation that is a continual requirement of T1D and CD across all aspects of life was present for all parents and adolescents. Michael spoke of having to be "*double prepared for everything in some cases*" to ensure that he has the supplies that he needs and to deal with the continual threat of "*in case something goes wrong*". Caroline illustrates how every aspect of Charlie's forthcoming day needs to be considered and accounted for prior to leaving the house:

Every time he leaves the house making sure he's got supplies of everything that he might need whilst he's out plus a mobile phone, plus all the supplies that need to be left at school. Um, every bit of every day has to be managed. And then you've got the added bonus of he might not just be able to find food when you're out because you've got coeliac disease so he's got to carry, or I've got to carry whenever we go out, all of his food for the day unless we know for definite that there's somewhere that we can eat whilst we're out. Um, if we're going to school he's got to carry every snack, um, lunch everything in his bag plus all of his diabetes supplies and blood testing stuff, and hypo treatment, so yeah it's a lot to think about before you can leave the house to make sure you've got everything.

Parents have worked hard to protect their child from exclusion as a result of their conditions. However, despite their best efforts through planning and preparation, adolescents have experienced their T1D and CD to be excluding. The challenges associated with this were reflected in the accounts of parents and adolescents. Adolescents told of being excluded from interests and activities due to the restrictions imposed by management of their conditions. They also shared concerns about potential future exclusion connected to the themes of threat to future plans (e.g. travel) and a sense of being punished because of their conditions. Rachel spoke of her sadness at being told that she was not allowed to go on school trips abroad as her self-management requirements would prove too problematic:

That's probably, that's one of the reasons why I haven't been allowed, I haven't been able to go to Hungary and Spain and Peru and all these other places that my school have organised, because of my diabetes and coeliac, so I feel like it

restricts me to do things that I really want to do, but hopefully I will be able to go and travel the world when I'm older, and that will be good. (Rachel)

Charlie has withdrawn himself from some of his interests due to being self-conscious of his symptoms and how they can prevent him from fully engaging in the activity in the way that he used to, but exclusion can also be imposed by others:

Um, he was picked for the football team his first year at high school but then missed out on matches because the one member of the PE staff who'd been trained in diabetes and his insulin pump wasn't able to go to the football matches on the particular night when he was picked for the team so he wasn't...so he was dropped from the team. Um, comments at school like, "By the time you've tested your blood and you've had something to eat and de de de da it's hardly worth you coming to the practice at lunchtime." So I think it has affected him. He wanted to go and play for the local football team that all his friends are in, I asked my friend whose son plays whether she could ask and the answer was no because he's got that insulin pump and the other kids are rough and we wouldn't...couldn't guarantee that he wouldn't get hurt. So yes it has impacted on the physical side, exercise stuff that he's wanted to do. And we have to kind of steer away and find something different to do instead. (Caroline)

The previous extract indicates that in some cases parents' planning and preparation can only go so far and that exclusion can often result if the people or systems involved are unable or not willing to accommodate the adolescent's needs. Caroline finds this particularly difficult and is aware of how much harder it is getting as Charlie gets older and is directly exposed to the 'rejection':

I think ((crying)) you try to plan and prepare, organise everything so that he can still join in everything knowing what to do and raise him to, to believe that having these conditions won't impact with the right planning and organising, and although it's a pain if we do this right you can still join in and do everything just like everybody else. And the reality of it is that you can only do that if the people who you want to entrust his care to are willing to, to do what they need to do as well. And people tell him everything will be alright and try to arrange it all. Unfortunately he's now of an age where people say directly to him, "No. You can't come" or "you can't go it's because of your diabetes." So it's cruel really. You try to protect him from knowing there's things that he might be prevented from doing as he gets older and you try and make everything work but, um, it's not always possible.

(Caroline)

Whilst the protectiveness of parents towards their adolescent dominates this theme, the notion of protection relates to the adolescent's feelings of responsibility for 'limiting' the family. That is, how the consequences of self-management are experienced by other members of the family (e.g. preventing them from eating gluten food, or decisions about activities/holidays being determined by T1D or CD). Adolescents described feeling 'bad' for their conditions having caused other family members to feel worried or guilty and for placing burden upon them. Charlie recognises how hard Caroline works to care for him and that "she's basically wearing herself into the ground to do it". This is difficult for the adolescent as they are not able to exert much control over the impact of their self-management on others but they try to minimise it as much as possible.

The extract below illustrates how perceptions of responsibility and protection for the feelings of other family members interact for Mary and Michael:

I guess my sister might have been a bit annoyed that things she was eating, probably she'd have to eat less, but I never said don't eat anything, because I think my family gets on with it, I don't really care what they eat, it's up to them, it doesn't bother me. Mum seems to think it bothers me a lot when she eats things with gluten in front of me and stuff, she's always like, 'oh, I don't want to eat this, Michael, but I really don't care. And she just... yeah, she's always like, 'oh, we can't have this, we can't go out for this meal because it won't be fair on you,' and I'm like, 'just go and do it, ((laughs)) it really doesn't bother me.' So, yeah, I feel a bit guilty sometimes about Mum worrying, but Mum worries about everything..... I don't feel guilty as a rule, it's just when Mum does say things like, 'oh, I don't want to eat this,' I think, well why do you feel like you have to feel like that, because I've told you it doesn't bother me and I just feel like she should just carry on, ((smilingly)) I don't want them to stop doing anything for me. (Michael)

If I eat something he can't have I feel guilty and he tells me to stop being silly but that's just the way I am, I'd just rather we all ate the same and if he can't have something we can't have something and that probably sounds really silly as well.

(Mary)

Loss in the relationship with food and eating

This represented the most prominent aspect of the narratives associated with CD and was most powerful for adolescents, although the whole family's relationship with food and eating is affected to some degree. The diagnosis of CD

was associated with a major loss in the relationship with food (i.e. immediate removal of a significant food group) and change related to eating as a social activity:

But you say eating social activities you don't realise how much of life revolves around food until there's a problem in getting it, you don't kind of think about how frequently that crops up whether it's just going round to a friend's house and the kids are playing outside and they come in and the mum goes, "Do you want, um..." you don't realise just how much food is involved. Because when he first got the coeliac diagnosis I just went, "Oh that's okay I'll just, you know, it's free-from stuff and we'll do that" until it becomes something that's limited or something that you can't have, you don't realise just how much of life revolves around food.

(Caroline)

For the adolescent, this loss is experienced in terms of either the previous or ideal relationship with food, depending on how long their eating behaviour has been defined by the GFD. Rachel was diagnosed with CD at age 5 years but she talked of appreciating how “daunting” it must be to be diagnosed with CD in teenage years due to the sudden change to your diet and exclusion of gluten foods that you are so used to eating.

This sense of loss was encapsulated in several of the adolescents’ and parents’ accounts. In response to his diagnosis of CD Charlie was left questioning whether there was anything he would be able to eat as his previous diet and favourite foods largely contained gluten. His options for eating out also became severely restricted although this has started to improve. Caroline reflected on Charlie’s process of loss related to his loved foods and eating out, but after her

struggle with getting a diagnosis there was some relief for her in knowing that a GFD was the way to proceed with treatment of the condition:

He was gutted at the Coeliac diagnosis because he knew all the things that my cousin couldn't eat. So we went through all that, "I'll never have Jaffa cakes again, I'll never have this again, I'll never have that again" because he had an idea of what coeliac disease was and the fact that when you go out to eat there's nothing on the menu you can have. So he was gutted; I was relieved to know that there was a physical reason and that we could just sort it out. (Caroline)

Michael also felt similar loss but the lead up to his CD diagnosis in receiving positive results of routine screening for a few years prior to diagnosis being confirmed by biopsy helped him to 'prepare' psychologically for the loss of food he liked:

well I sort of knew it was coming because I'd already had the one blood test and then the second one and they'd all come back and I thought, oh this is going to happen really. So I had a build up to it and at school I was always like oh, well this could be my last biscuit, like joking with my friends and stuff, but I mean... Yeah, I wasn't... It wasn't as bad when it actually came round to it as it was the first time I had the blood test come back positive. The first time I was really upset about it, because I thought, oh I'm not going to be able to eat any of these things anymore, the second time I was kind of ready for it, I wasn't so worried about it because, yeah, I guess I was just more prepared.(Michael)

This loss is exacerbated in social situations when the adolescent has to observe others eating the food they once enjoyed:

...you know everybody comes down in the morning and they've got their croissants and the choc au pain and that nice crusty French bread and poor old

Sarah can't have any of it you know and she'd been used, before that she'd been used to having nice bread and teacakes and macaroni cheese and everything.

(Susan)

Parents and adolescents talked of the family home becoming a largely gluten-free environment:

I suppose Mum's more in charge of all of that at home really because she does all the shopping, so we don't have anything really with gluten in the house, we have a special cupboard where we keep all the stuff that's got gluten in it, which only Mum and [sister's name] really go in. They sometimes hide things from me in there, so I have to check up occasionally just to see what they're hiding from me, but yeah, so most of the things in the house, I mean we only have gluten free flour and things and it's just easier that way I think because I know that everything pretty much in the kitchen I can eat. And that's nice of them to do that (Michael)

When we looked at things it was easy to adapt things and the kitchen now is gluten free apart from one cupboard. [sister's name] got one cupboard that we actually put crisps and things and biscuits, you know, anything like that Michael can't eat goes in the gluten cupboard because we just decided it was easier to have the rest of the kitchen gluten free and we all eat the same. The only things we tend to eat different are bread and cereals. I cook family meals and they are gluten free and that's just the way we do it now and it's much easier than everybody eating different things. Michael's got his own toaster, he's got his own margarine in the fridge with a sticker on so that he knows it's his and people can't get crumbs in it and I would have said we were a 95% gluten free household really

(Mary)

An image of a special cupboard that contains 'forbidden food', which has to be hidden, engenders sense of 'secrecy' around family access to gluten food at home, borne out of wanting to protect the feelings of the adolescent.

Like Michael, Rachel appreciates the family's sacrifice in not having 'tempting' gluten foods at home and explained that she would encourage parents of a young person who has just been diagnosed with CD to introduce the GFD at home as it would help them to feel 'normal' around their family.

Caroline's account below encapsulates how CD and T1D has changed her relationship with food in many ways, including her weight, eating in secrecy and how it has elevated food from something that used to be an automatic feature of everyday life to yet another issue to have heightened awareness of in the management of these conditions:

I think so yeah because I've put three stone on since he's ((laughingly)) been diagnosed. And I know a lot of that is from stress and just shoving things in my mouth to stay awake with having to get up and do night testing and stuff. But also because I eat, um, in secret because I might be eating stuff that he can't have and you ((laughingly)) kind of go up to the kitchen and eat this. But you still eat with them for the social aspect of it. So yeah. ((laughs))... Obviously I'm not blaming that on my weight gain and I could quite easily not eat the stuff but it is, it kind of heightens the just everything about food in general that you would have to constantly plan and prepare and think about stuff that you just never really gave much of a thought to until it was mealtime or you're going shopping for. (Caroline)

Another element to this theme appears to be how T1D and CD interact to complicate dietary management and heighten the burden surrounding food and

eating, which families try to manage through careful consideration and preparation.

it makes life more difficult like for me it made life more difficult by far just because um... [pause] quite a lot of the coeliac products put extra sugar in just to try and like improve the taste of stuff so sometimes that can be difficult um eating out becomes a nightmare sometimes [laugh], if you're out with friends and stuff coz you have got to be very careful of where you are eating and what you are eating and you can feel like a bit of a burden sometimes which isn't nice um but to be fair like I find the coeliac becoming more of a problem socially than the diabetes can because um with eating out you have got to be really careful with the coeliac but you can just bolus or inject or whatever for the diabetes side of it. (Sarah)

It confuses a lot of people because you go, "Yeah he's got Type 1 diabetes so that doesn't mean he's banned from eating anything he can have whatever he likes it's not managed with diet." And then you go, "But he has got coeliac disease so he can't have..." and I think that's where the confusion comes in a lot with having the two conditions. (Caroline)

I think that's the added complication, or one of the added complications with having coeliac disease is obviously being diabetic you've got to be able to get food. And coeliac disease means that you might not be able to get it so you've got to take everything with you all the time. (Caroline)

Living in the present vs. connecting with future threat

Adolescents are aware of the long term risks and health complications associated with T1D and CD. However, the accounts of some reflected a tension in connecting with this 'threat' in the present moment in the ways that maybe their parents or professionals involved in their care expect or want them to. This presented as a particular struggle for Michael and Sarah as their CD is asymptomatic, seeming to place additional conflict between 'knowing' (the risks) but not 'feeling' (the consequences):

I mean it's like the diabetes I worry about the long term things with that more, because you think there's all these long term complications that could happen if I don't control it well enough. The coeliac, there are long term complications, but because I've never had the symptoms I've never really worried... Well, I have worried, I have worried because I've thought, well you know, I don't have any symptoms, I could be eating whatever and I wouldn't know and that could lead to long term things. (Michael)

I don't get symptoms if I've eaten something, um I know the long term effects of it um... which are not good but I don't know, I don't feel anything, and it's like me and mum have said before, I think if I had symptoms I'd take it more seriously, if I felt unwell afterwards so that it, you know, I know there's a knock-on effect then I would take it more seriously, but I don't, and sometimes it's like "well why can't I eat it when nothing happens?", do you know what I mean, it feels like, there's, you're being told something without a reason, even though I know there's a reason... um, at this stage it's quite hard to see you know 60 years into the future

and it can feel like, oh just a biscuit won't hurt or you know and whatever um...

(Sarah)

Although this tension was more present in the stories of some adolescents than others, it connects with the theme of hopes and expectations for future health that is shared between parent and adolescent, whereby parents acknowledged how the challenges of normal adolescence are complicated by these conditions and can make it difficult for adolescents to see further into their future health. This challenge felt sufficiently pertinent as self-management behaviours and attitudes that are laid down across adolescence may be carried into adulthood and have implications for longer-term health. It may also be possible that a future-focused thinking style regarding the threat of serious health complications is incongruent with, and therefore threatens, adolescents' hopes and expectations for their life goals that were expressed earlier.

Superordinate theme: *Duality: together but separate*

The similar but opposing positions held by T1D and CD was clearly evident in parents' and adolescents' perceptions of the dual diagnosis. That is, the conditions appear to be experienced 'jointly' in terms of biological (autoimmune) status and in requiring complex management, but also 'separately' in how the adolescents and parents identify with them. This separation related to the state of 'flux' within the conditions and precedence of either T1D or CD, as reflected in the themes of 'CD as a 'consistent constant' vs. T1D as a 'variable constant' and 'Focus on T1D vs. T1D and CD' which are described below.

CD as 'consistent constant' vs. T1D as 'variable constant'

Linked to the concept of overwhelming permanence introduced in the previous theme of perpetual loss and protection, this theme explores the how the conditions contrast within their similarities. That is, T1D and CD are constant in that they are both life-long conditions and require continual self-management...: *so the coeliac's more difficult to get round sometimes and I think it gets in the way more but as sort of like an everyday thing it just becomes tiring being diabetic to be fair because its... its constant... its six blood tests a day, its you know I'm on the insulin pump so I've got to carry that round with me all the time, like even choosing an outfit in the morning becomes an issue [small laugh] its just like 'ok where am I going to put this' 'where I am going to carry everything' yeh it can just become tiring sometimes. (Sarah)*

If he's had a bad experience, if someone's said something to him and he's upset about it then it's obviously harder because you can't ever say we'll have a day off from it today. So no matter what he's feeling about it or how naffed off he is there's no option to not do it. You've still got to do those blood tests, you've still got to do those, you know you can't leave it on holiday or have a night off, go out and leave it at home it's always there. (Caroline)

...but within this, the conditions' state of 'constant' is also experienced to differ in that CD can be relatively consistent whereas diabetes can fluctuate considerably and be more unpredictable. This relates to the conditions' symptoms and self-management and was a recurring theme found to be present within all adolescents' and parents' accounts.

It's just the diabetes, I just take exactly the units as I have done, unless suddenly my units change, and it's annoying if you have to wake up in the middle of the night, like last night I had to, I woke up feeling hypo and I was 3.1, so I had to stay awake for 15 minutes, to wait for my blood sugar level to go up before I could go back to sleep again, and that was like four o'clock in the morning! ((laughingly)) So I was up at four o'clock in the morning, waiting for my blood sugar level to go up, and then I ate a sandwich to get it up and took some Lucozade, and then, and then it went up to 5 with that amount. And then when I woke up at about nine, it, I checked my blood sugar level again and I was 3.2, and I thought to myself, 'Why are you hypo, Rachel, you've woken up and you've got yourself up to the correct level?' So I'm going to have to sort that out. And it's annoying when things like that happen, because you've got into a routine of taking the amounts, and you have to go and change it all, and change your insulin sensitivity and things like that, but the coeliac disease is easier because it's just all the same food, unless the manufacturer changes the recipe, yeah.(Rachel)

The consistency of CD management (i.e. the relatively uncomplicated principle of the GFD) can make it easier to manage in comparison to T1D:

Well, it, coeliac is a lot easier to learn about and ((slight pause)) I suppose in a way it's just keep everything separate and make sure it hasn't got this, this and this in it, and you should be all right. Whereas diabetes with all the equipment and all the possibilities of stuff going wrong and all the eventualities that could come out it takes a lot more training, and it takes a lot more brain power to work out what to do.(Charlie)

Caroline elaborates on Charlie's comments regarding the complexity of T1D management and describes how despite her best efforts to adopt a

consistent approach to T1D this does not always lead to the same outcome for Charlie and *“you can do the same thing on the same day, same set of circumstances, and get a different, total different set of blood results, it's just one of those kind of illnesses”*:

Within this narrative of negotiating the consistent state of flux within and between T1D and CD, there emerged a strong perception that both adolescent and parent can experience the conditions as punitive in relation to their efforts in self-management. This notion developed out of a sense that both parent and adolescent can perceive that they are considered to be at fault (e.g. “I can never do right”) when blood glucose or coeliac antibody levels fluctuate or worsen. These dynamics appear to occur in several relationships within the system around the adolescent: between adolescent and clinicians, parent and clinicians and adolescent and parent. Sarah talks of clinic below:

it does become (slight laugh) a period of high stress in my house just before and just after clinic because we always know that there is going to be something that we've done wrong and it can feel sometimes like you are getting a telling off, for something that you don't want to deal with in the first place, and something that um you, you know, it's not your fault or whatever but you can feel like that you're getting a telling off for something that you didn't choose, which again is annoying (laughs). (Sarah)

I did get referred to see a dietician six months after Charlie had been diagnosed because his coeliac levels were still high and so the diabetes consultant said that I needed to see a dietician because obviously I didn't understand and I wasn't feeding him the right things. So I went to ((laughingly)) see the dietician who went,

"Oh it's you!" Because I'd just met her on a course for carbohydrate counting and she went, "But you know about coeliac disease don't you?" I said, "Yeah but I've been sent because the diabetes consultant says his levels are too high so clearly I don't." So she just went through a few things and said, "Well that's we've ticked the box now haven't we because I know you know what coeliac disease is". I did phone Coeliac UK on the back of these, the high levels still after six months and they said that's fine because they are now half what they were undiagnosed, because his levels were so high undiagnosed, it's going to take a long time for the gut to repair and that damage to be completely undone, and it could take up to two years for it to be back down within those limits. And obviously the diabetes consultant didn't know that he just saw a raised level and then said that I obviously didn't know what I was doing, so sent me to the dietician. (Caroline)

Power dynamics within relationships and interactions appears to play a role in feeling penalised. Sarah strongly articulated how her experiences of clinic can leave her feeling 'ganged up on' and that she lacks control and choice in these situations. She reflected on the frustration at being "told what to do" without acknowledgement and understanding of how challenging self-management is:

I feel like they're not giving me a choice and they are just telling me what to do like the nurses aren't diabetic, they're not coeliac, you know, they don't know and yet they are quite happy to tell me "oh well you need to do 12 blood tests a day" Well you try doing it, you know, if you knew what it was like then I might have a little bit more respect for you but if its someone who doesn't have a clue and literally just, just knows the medical side of it then I am not interested in listening to them, at all... [pause] because they don't understand how that extra six blood tests a day is going to have an impact, and it is going to have an impact. (Sarah)

However, adolescent's experiences did vary and Rachel's described her experiences of clinic to feel less medically orientated and more humorous and conversational.

Sarah experiences increased frustration when only the 'negative' (what is going 'wrong' with management) becomes the focus whilst positive aspects of her efforts with self-management are minimised or overlooked:

um, like my mum felt the need to call up me at my friend's the other day and sort of have a real go at me down the phone about finding a chocolate bar wrapper in my bin, which is gluten free, and I have proved it to her on the internet but you know she felt the need to call me up and sort of, you know, have a go at me about it when its, its one thing, um, and I think sometimes like one thing like eating the wrong thing, even sometimes if it is not accidentally, you know, eating the wrong thing can overshadow all the other things I am doing that are right, you know, like blood testing constantly or um you know keeping my blood sugars under control or eating sensibly most of the time, that can all be overshadowed by one thing and I think sometimes people don't understand that, there's quite a lot to it that um you have to work around... and they (laugh) see every little thing as quite a big deal whereas I see it as "ok there is one little thing, but look at all these things that are going well and they don't seem to see it like that, my parents and the nurses.

(Sarah)

This feels like it invalidates Sarah's efforts and may reduce her motivation for self-management.

Focus on T1D vs. T1D and CD

The predominance of T1D across adolescents' and parents' experiencing of these conditions and their management was salient and reflected consistently throughout the interview process in terms of content and intensity. Furthermore, this focus on T1D appears to transcend the immediate family system and to be a message that is also received through interactions with the clinical care system around the adolescent's dual diagnosis. Although it is important to be aware that these families' experiences of clinic will differ depending on the types of services they access in terms of their medical care, there were some commonalities in parents' and adolescents' descriptions of how CD is monitored and supported. Rachel describes how her clinical appointments centre round her T1D with intermittent review of her CD on "*coeliac day*" when a dietician or representatives from gluten-free food companies are present in clinic.

Parents have also experienced their involvement with professionals in relation to management of CD to be brief in terms of contact and guidance provided:

we saw a dietician when Charlie was originally diagnosed before we were discharged from hospital. She said, "Do you know what coeliac disease is?" I said, "Yes." She said, "Good. Have you got any questions?" I said, "No" she said, "Good." (Caroline)

Michael describes similar experience but acknowledges that, from his perspective, the GFD is relatively straightforward and that attending an additional clinic for support with CD would not be preferable.

Connected to the dominance of T1D within families' narratives, and the perceived severity of health consequences (compared to CD), was the impression

that the dual diagnosis can be experienced as 'T1D putting CD into perspective'. That is, T1D and CD not existing as two equal conditions that are appraised and managed in equal weighting, but the T1D being 'prioritised' and influencing the perceptions of, and attitudes towards, CD and its management. This concept was most clearly articulated by Caroline when describing her perception that whilst a single diagnosis of CD would carry burden and inconvenience it would be more manageable and less serious in comparison to T1D. She expressed this 'perspective-taking' in terms of the relative threat and consequences of T1D compared to CD (i.e. threat of losing consciousness contrasted with feeling hungry):

And also having coeliac disease if you went out and you couldn't get something to eat you'd just have to stay hungry until you found something you could eat. But if you're diabetic you can't run that risk of... You can't be... It's not just about being hungry it's about being unconscious so you can't... No the diabetes is the one that is, um, the biggest life changer. If you've just got coeliac disease you can always take a bag of crisps or a biscuit or something and it doesn't matter if you're a bit hungry does it? But if you're diabetic it matters if you don't treat that straight away.

(Caroline)

The ever-present focus on T1D can be difficult for some of the adolescents as they contemplate important transitional events that they would like to experience at this stage in their lives, such as education and travel. Michael spoke of struggling to maintain this focus on his T1D whilst he is in a good period of self-management. Sarah is planning to travel as part of a gap year. She reflected on not wanting the trip to be overshadowed by her T1D, and of a desire

for her self-management to become a 'background issue' so that she may fully embrace this experience:

I know it's serious, but at the same time I don't want that to be the front issue of my trip I want the trip to sort of lead itself to be fair without having that being the main issue, if that, if that makes sense...im still going to take care of myself and whatever, but I don't want that to be the main, the main point so (pause). (Sarah)

This defining influence of T1D over the adolescents' day to day living may generate tensions as these adolescents enter a crucial normative stage of development, aspiring for their experiences, rather than their health, to define their lives.

However, T1D's influence is not considered to be solely located within the adolescent's lived world as its presence is sensed by all family members and becomes part of the family and family life. Parents' and adolescents' accounts engendered an impression of T1D as an 'unwanted' member of the family who is resented but eventually 'accepted' as part of a "new normal". Rachel accepts that it is necessary for T1D to hold this position in her family as it is so important to maintaining her health:

...the diabetes is like a big part of our life. It shouldn't really be a small part, because if it is, then you wouldn't, you wouldn't be very well, so that would be stupid. (Rachel)

Mary reflected on how T1D becomes integrated into the identities and ways of being for herself, Michael and the family at an individual and collective level:

It just becomes part of the way things are and you find yourself, even when Michael's not with you, reading labels on things and checking to see what's available because it is just part of everyday life now and it's part of him, the way

he is, it's part of him and how he deals with things. He wouldn't be the same person if he hadn't got it and I don't think we would be either. (Mary)

Similar meanings did not appear to be constructed within parents and adolescent's accounts of CD.

Whilst the general impression regarding the centrality of T1D compared to CD was quite marked and consistent between some dyads, at other points during the interviews parents and adolescents expressed apparent difficulties in distinguishing between T1D and CD. This mainly related to whether the focus of management and perceived limitations of T1D and CD was weighted more heavily towards one of the conditions in relation to the other. Mother and daughter, Sarah and Susan, both focus on managing T1D over CD:

I focus on managing the diabetes more than the coeliac because I think, because if something goes wrong with the diabetes I'll have a symptom like I'll feel unwell or you know I won't feel very good um whereas if something happens with the coeliac then nothing happens, so the diabetes takes priority, definitely...over the coeliac. (Sarah)

yeh probably with me it's the diabetes. I think with the coeliac I make sure everything she eats while she's here is gluten free um but yeh probably, probably that. (Susan)

For Sarah, this focus reflects whether immediate or noticeable consequences will result from her self-management behaviours: the fact that her T1D is symptomatic whereas her CD is not. From Susan's perspective, her role in supporting the GFD can be more straightforward so emphasis is placed on T1D.

That said, Susan is aware the CD may actually place greater limitations on Sarah in terms of the stage of life she is at:

well, I don't know really I mean the coeliac limits her in that you know she's not free to you know, maybe socially it limits her as well if you are going out for a meal you know things like that but um I don't think, like with the diabetes it limits her, it doesn't stop her doing anything you know she does everything that everybody else does um whereas the coeliac might be more limiting in that way um but yeh so probably the coeliac being 17 you know, probably limits her more than the diabetes does I mean she's got to make sure she boluses every time she eats and everything but apart from you know having this um pump attached to her all the time probably doesn't limit, doesn't stop her doing anything, no. (Susan)

Perceptions of the limitations posed by the dual diagnosis emerged as a more complex dynamic, determined by the interaction between T1D, CD and the situation in question. The restrictive nature of the GFD can elevate CD above T1D in terms of the limitations generated on an everyday basis. This appears to represent balancing food restriction and accessibility in CD against T1D being less 'excluding' but requiring self-management to be adapted accordingly. However, as Michael and Mary's comments illustrate, this is a complex interplay that requires continual negotiation by both parent and adolescent:

Yeah, the coeliac I think, because it's... Diabetes doesn't actually stop you doing anything, I mean it makes you think about things maybe a bit more, you know, you have to think am I going to have...? Like I was thinking of going volunteering in Africa or whatever, and you have to think about how much supplies you're going to need to take and whether there's going to be a hospital nearby just in case and things like that. And insurance, diabetes makes a difference to the insurance, but

coeliac, there are things that I'd like to do that just really are a bit impractical with coeliac. I'm trying to think of one now. This is what I thought when I got it anyway. I mean things like drinking beer, I don't miss it too much, because I've never really tried it to be honest, but it would be nice to be able to, I don't know, talk about beers with my friends or whatever, just... I mean it's stopping me doing less things I think than it used to, because restaurants are getting more onboard and things. So yeah, it's getting better, but I would say coeliac still stops me more, diabetes just makes you think a bit more before you do things. (Michael)

I think coeliac is more awkward from the point of view that I can't really get such as gluten-free bread in town and so I have to plan and make sure that if I want anything; So I suppose it is the coeliac really from day-to-day because it's making sure we've got the right ingredients and things in whereas the diabetes and things Michael deals with...But it's hard to say really, it's very hard to say, but probably the coeliac. (Mary)

Other parents and adolescents held similar views, describing CD to be more restrictive and socially limiting. For Rachel, the limiting effect of the GFD on eating as a social activity is powerful and presents her with a dilemma as to which condition is most challenging:

I'd prefer to have just the diabetes, I don't want, I don't like, well, I'd prefer not to have either, but if I had to choose, I'd prefer to just have the diabetes, because then I could eat normal food with everyone else. No, I don't know actually, I wouldn't like either. It's not, because being diabetic is hard, and being coeliac is hard, so you can't really choose between them. I don't know. (Rachel)

These conversations appeared to be asking adolescents and parents to connect with, and conceptualise their experiences of the dual diagnosis in ways they may not have considered before. This engendered a sense of these families having had little opportunity to reflect on or explore their experiences in any depth, evidenced by the following extracts from Susan and Rachel:

it's useful doing something like this just chatting about 'cause it you know I suppose it you know makes you sort of think about what's gone before and everything and sort of how you get through it (Susan)

I've never spoken to anyone like this about my diabetes or coeliac disease, never. I've just spoken to people, explaining what it is, I've never spoken about how it affects me and how I feel about it. (Rachel)

DISCUSSION

Through the use of a phenomenological approach, this research aimed to conduct an in-depth exploration of how families with an adolescent with T1D and CD experience and make sense of what it is like to live with both of these conditions as a dual diagnosis. This included a broad curiosity in understanding the challenges encountered in day-to-day life, from a psychosocial perspective. In terms of existing research into chronic health conditions in childhood, this represents an as yet little-understood area within the youth T1D and CD literature. Two main overarching themes emerged: 'Perpetual loss and protection' and 'Duality: together but separate'. These themes encompass an understanding of the families' dynamic journeys from diagnosis to the present day, characterised by paradoxical experiences of T1D and CD separately, and as a dual diagnosis, alongside living with continual threat of further loss and need for protection against the consequences of these conditions.

Although experienced in similar and different ways the majority of the themes that emerged were shared across the accounts of these parents and adolescents, demonstrating the importance of considering these conditions from a broader perspective. That is, not viewing T1D and CD, and their self-management, as being located solely within the adolescent but as a 'shared' process between parent and adolescent. Mother-adolescent dyads have been found to frequently appraise diabetes ownership as shared and to report engaging in the tasks of diabetes self-management collaboratively (Beveridge, Berg, Wiebe & Palmer, 2006). Comparisons may be drawn with the concept of shared illness ownership and this would also support the current study's findings of parents' difficulties in relinquishing responsibility and involvement due to having 'owned'

the conditions and responsibility for their self-management for so long. Further, it appears to be difficult for these parents to 'ease' their protective instincts as their adolescent gets older. Research advocates the importance of sustained parental involvement and collaboration in adolescents' self-management for improving diabetes outcomes in youth T1D (Silverstein et al., 2005; Wysocki & Greco, 2006) but families may require additional support in renegotiating their roles in order for the adolescent to develop autonomy in self-management during the challenging period of adolescence. This may benefit from incorporating an explicit understanding of how parents view and experience their 'protective' role in relation to these health conditions.

Within the theme of 'Perpetual loss and protection' parents and adolescents have experienced 'loss' in various ways as a result of T1D and CD and they live with the knowledge that further losses to the adolescent's health and family lifecycle may yet be suffered, particularly if the conditions are not 'well-managed'. In response, parents have engaged in highly protective practices, channelled through their involvement in self-management, in order to try and shield their child from further harm (physical and psychological) and limitation or exclusion. Analogy can be drawn with systemic ideas and practice found within the intellectual disability literature: that is, when families are challenged with how to grieve for lost hopes and expectations when a family member is diagnosed with an intellectual disability (Goldberg et al., 1995). Through their experiences of family therapy work, Goldberg et al. (1995) recognised how families strive to protect each member from additional grief related to the disability or the perceived 'dangers' associated with its consequences. The authors advocate providing families with therapeutic space to experience a shared grieving process for past, current and

future losses. This may offer useful insight into potential intervention options for supporting families living with childhood chronic health conditions who may be experiencing struggles related to loss and grief in this context.

Counter to the overall predominance of T1D across accounts, losses in the relationship with food and eating, related to the social limitations placed on adolescents, emerged as the most prominent theme within families' experiencing of CD. This is consistent with existing findings in the CD literature of challenges associated with the GFD, such as poor availability of gluten-free food (Roma et al., 2010), sense of social stigma and discomfort (Olsson et al., 2008) and ambivalent relationship with food (Theodosi, 2009). Furthermore, families' descriptions of how the GFD can interact with T1D to complicate dietary self-management are consistent with reported difficulties in managing the complex dietary requirements of T1D and CD outside of the home (Saukkonen et al., 2002) and indicate this to be an additional limitation associated with the dual diagnosis. Thus, the families' experiences of the changed relationship with food and eating appear to be shaped by both CD and T1D, creating complications in dietary self-management which are experienced as 'twofold'.

In the theme of 'Duality: together but separate', the relationship between T1D and CD as a dual diagnosis was found to be experienced in complex and sometimes contradictory ways. That is, the chronicity of the conditions and their management are perceived similarly and experienced as being 'constant'. However, T1D generally dominated the narratives of parent and adolescent and was experienced to be more 'dangerous' and inconsistent in its manifestation of symptoms and response to self-management. The focus on T1D appears to exist beyond the family system and to be communicated within the clinical care families

receive. Support from health care providers regarding the practical side of CD self-management can be delivered relatively succinctly but there appears to be little follow-up or exploration of how the psychological and social side of the GFD impacts the adolescent and family over the longer-term.

The question of whether a dual diagnosis of T1D and CD results in additional burden is not a straightforward one. The findings of the current study suggest the interplay between T1D and CD self-management to be complex and variable. Nevertheless, based on the experiences of these parents and adolescents, factors that are specific to managing both T1D and CD appear to place additional strain on families. For example, families in this study described T1D and CD to interact in complicating dietary self-management, which does not support Sud et al.'s (2012) conclusions of a minimal impact of the GFD on quality of life in children with T1D and CD. However, findings of lower social functioning (parental report) in adolescents (Sud et al., 2012) were supported as both adolescents and parents in this study reflected on the social limitations imposed by T1D and CD. The focus of Sud et al.'s (2012) study was specific to examining the impact of the GFD on child quality of life and impact on parental quality of life was not assessed.

The sense of burden that emerged within the current study may be conceptualised in terms of chronicity and variability in the conditions. Whilst present in the accounts of both parent and adolescent, this burden appeared to manifest more noticeably in the accounts of parents. In the case of the present study, an accumulative effect of the continual and complex demands of self-management (particularly the inconsistent nature of T1D) and threat associated with T1D and CD may be placing a longer-term burden on parents. Mothers and

fathers have reported finding the long-term health concerns associated with T1D to be most burdensome (Haugstvedt, Wentzel-Larsen, Rokne, & Graue, 2011).

Clinical implications

This study highlights how the management of youth T1D and CD operates within a relational context and thus illustrates the clinical importance of understanding how the family system, particularly parent and child, experience day to day life with these conditions and collaborate in their self-management. Family work (drawing on systemic approaches) would be well-placed to support these families through its potential to offer a forum for families to explore what matters to them about the dual diagnosis and to facilitate conversations that may help negotiate some of the emotional, social and behavioural challenges of self-management.

Additionally, adopting a wider family perspective in supporting families with the management of the dual diagnosis should include eliciting parental perceptions of their psychological wellbeing. Parents may benefit from additional support alongside the practical side of self-management in the immediate aftermath of diagnosis so that losses and emotional impact may be processed and not carried through into future experiences.

At present, provision of integrated psychological and medical support in paediatric and adolescent diabetes services is uncommon (Christie & Martin, 2012) and even less is known about any specialist provisions for the dual diagnosis. Increased psychological input to augment clinical care, with specific consideration to a dual diagnosis of T1D and CD and their management, may help

to address some of the psychosocial issues associated with managing these life-long conditions.

Future research

Research into the psychosocial experience of a dual diagnosis of T1D and CD is scarce and warrants further exploration. More within-family dyadic research of a quantitative and qualitative nature may help to develop this evidence base and provide richer understandings of how families manage the dual diagnosis. It may be interesting to extend qualitative methodologies to include interviewing all key members in the family system (e.g. parents, adolescent and siblings) and to explore the perceptions of health care professionals involved in the care of young people with a dual diagnosis of T1D and CD.

Methodological considerations

This study was innovative in combining a qualitative approach with dyadic analysis of parents' and adolescents' responses in conducting an in-depth exploration of the experiences of families living with a dual diagnosis of youth T1D and CD, as the existing literature has mainly employed quantitative methodology in examination of the dual diagnosis.

Sample size was in line with advice for IPA and afforded the idiosyncratic commitment of this approach to be fulfilled (Smith et al., 2009) through allowing the researcher space to engage in comprehensive analysis of the accounts of this small number of parents and adolescents. The sample could be considered homogenous with respect to all of the adolescents live with a dual of diagnosis, but diversity in participant characteristics inevitably existed: for example, age and

duration of diagnoses, use of insulin pump in self-management of T1D and asymptomatic CD. The potential for such variables to differentially influence experiences of self-management should be considered. Also, objective measurement of HbA1c levels and coeliac antibodies could not be comprehensively reported for the entire sample and meant that it was not possible to establish current levels of metabolic control and self-management of the GFD for this group of adolescents. This precludes situating the sample in terms of how well their T1D and CD are currently self-managed and the experiences they have described.

Therefore, it is not proposed that this group of parents and adolescents is representative of every adolescent and family living with a dual diagnosis of T1D and CD. However, the prevalence of shared themes across the families serves to counter some of the limitation placed on the potential transferability of the findings.

CONCLUSION

Adolescents living with a dual diagnosis of T1D and CD and their families are tasked with negotiating the continual and fluctuating challenges of two complex and variable chronic health conditions. Previous research has proposed there to be a minimal impact of an additional diagnosis of CD in T1D (Sud et al., 2012), however, findings of this study suggest the chronicity and variability in condition management to be burdensome for families. Further intra-familial research, particularly of a longitudinal nature, is required to deepen our understanding and inform how families of young people with T1D and CD may be supported with the self-management of these life-long conditions.

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PUBLIC DOMAIN BRIEFING DOCUMENT

**Intra-familial agreement and disagreement regarding family division of
diabetes responsibilities:**

Associations with diabetes outcomes in youth type 1 diabetes

&

**Living with Coeliac Disease and Type 1 Diabetes: The experiences of
adolescents and their parents**

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PUBLIC DOMAIN BRIEFING DOCUMENT

This paper summarises two original research documents relating to two common chronic health conditions found in children and young people: Type 1 Diabetes and Coeliac Disease. Firstly, the literature review was interested in examining research on how the agreement and disagreement between family members regarding the sharing of responsibility for diabetes management tasks is linked to other aspects of diabetes. Secondly, the research study used a qualitative approach to explore adolescents' and parents' experiences of what it is like for a young person to live with both Type 1 Diabetes and Coeliac Disease.

Literature review

Introduction: Type 1 Diabetes (T1D) is one of the most common chronic health conditions among children and adolescents. Treatment of T1D is complicated and demanding, including insulin injections, monitoring blood glucose levels and management of diet and families can find this challenging to cope with (Anderson et al., 2002; Debono & Cachia, 2007; Greening, Stoppelbein & Reeves, 2006). Parents and children often share these tasks between them and exploring the divergence in family members' perceptions of responsibility for these tasks is a growing area of within-family research.

Method: Research databases were systematically reviewed to identify family research in relation to three research questions exploring how the distribution of diabetes responsibility is perceived by family members, how agreement and disagreement is associated with diabetes outcomes and family interventions

targeting family sharing of responsibility in improving diabetes outcomes. Thirteen papers were available for inclusion in the review.

Findings: Ten cross-sectional studies explored the perceptions of various family members (e.g. parent and child, mother and father) regarding the distribution of diabetes responsibilities and the associations between different patterns of agreement/disagreement in family views and diabetes outcomes (such as metabolic control, diabetes self-management and family psychosocial functioning). Three studies explored the effectiveness of family-focused interventions in improving and maintaining family sharing of diabetes responsibility. Findings suggest there is limited evidence of strong associations between family agreement/disagreement regarding family diabetes responsibility and diabetes outcomes. However, greater parent-child disagreement has been linked to poorer metabolic control and decreased parental psychological wellbeing (increased distress and depressive symptoms). There is also evidence to indicate that a specific type of disagreement between mother and adolescent is linked with parental psychological well-being and diabetes family conflict, that is, parents disagreeing with their child's perceptions of the degree of responsibility that the child perceives themselves to have.

Clinical and research implications

Diabetes health care professionals could help families to explore differing family perspectives regarding diabetes family responsibility that may have a detrimental effect on diabetes outcomes. Additional within-family research is required to

further develop the evidence-base on the impact of family agreement/disagreement regarding diabetes responsibility on diabetes outcomes.

Research study

Introduction: A growing number of adolescents live with both of Type 1 Diabetes (T1D) and Coeliac Disease (CD). Coeliac disease (CD) is an autoimmune condition that causes the body to attack itself in response to ingestion of gluten. Adolescence is acknowledged as a particularly challenging time in self-management of these conditions. There is a lot of research about the psychosocial issues associated with these two conditions as single diagnoses, however, much less is known about what it is like for adolescents to live with both T1D and CD. One study has suggested there to be a minimal impact of an additional diagnosis of CD in T1D (Sud, Marcon, Assor, Daneman & Mahmu, 2012). The current study aimed to develop a deeper understanding of how a group of adolescents and their parents make sense of the challenges associated with self-management of the dual diagnosis, specifically how these two conditions are experienced as a dual-diagnosis, from a family perspective.

Method: Eight individual semi-structured interviews were conducted with four adolescents with a diagnosis of both T1D and CD, and their parent. Interviews were analysed in-depth using interpretative phenomenological analysis (IPA).

Findings: Two super-ordinate themes emerged from the accounts of adolescents and their parents: 'Perpetual loss and protection' and 'Duality: together but separate'. These themes present an understanding of the families' dynamic

journeys from diagnosis to the present day; this has included experiences of various 'losses' as a result of T1D and CD and striving to protect against the perceived future threat to the adolescent's health and family lifecycle if the conditions are not managed effectively. There was a focus on T1D and its management but losses in the relationship with food and eating emerged as the most prominent theme within families' experiencing of CD. The findings illustrated how adolescents living with a dual diagnosis of T1D and CD, and their families, are tasked with negotiating the continual and fluctuating challenges of management of these two conditions. The chronicity and variability in management of T1D and CD appears to be burdensome for families, particularly parents.

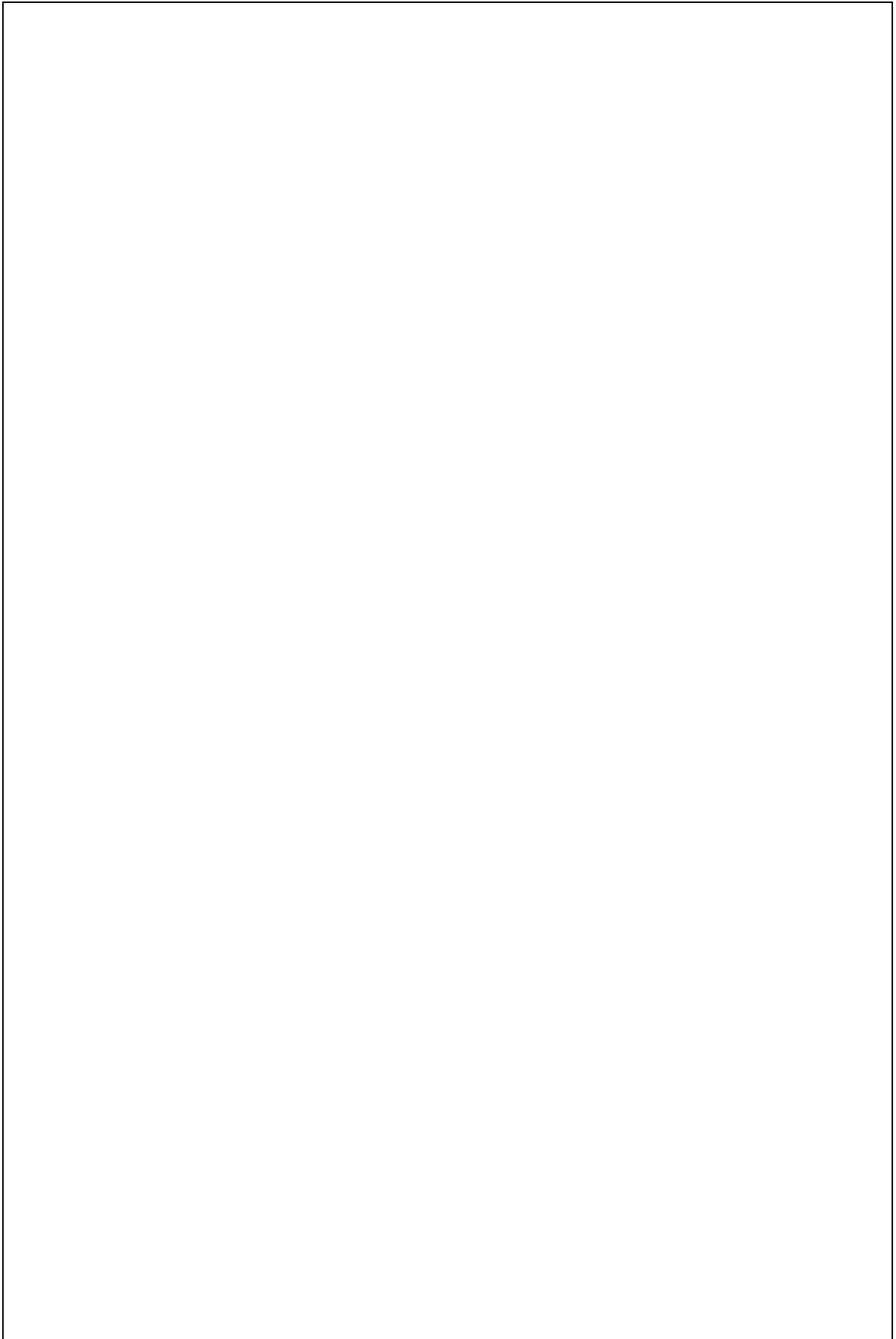
Clinical and research implications: At present, provision of integrated psychological and medical support in paediatric and adolescent diabetes services is uncommon (Christie & Martin, 2012). Introducing psychological input alongside clinical care, with specific consideration to both T1D and CD and their management, may help to address some of the psychosocial issues associated with managing these life-long conditions. Further within-family research is required to deepen our understanding and inform how families of young people with T1D and CD may be supported with the self-management of these life-long conditions.

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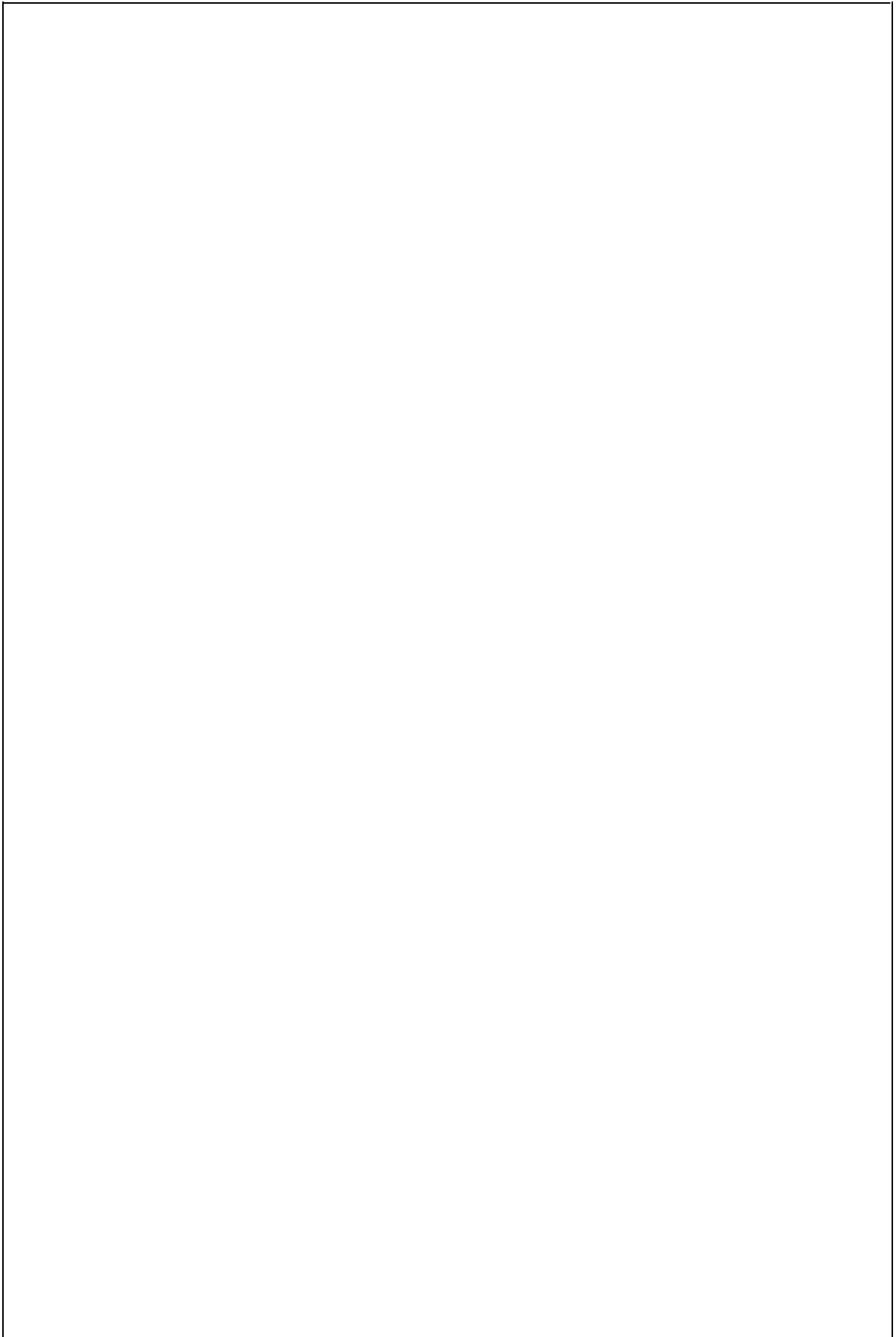
Appendix 1: Instructions for authors – British Journal of Psychology

Copyright Material



Appendix 2: Instructions for authors – Qualitative Health Research

Copyright material



Appendix 3: Search strategy and search terms

Search strategy and search terms

#	Searches
1	diabete* or “type 1 diabetes mellitus”
2	(child* or adolescen* or “young people” or “young person” or teen* or youth or infan*)
3	(parent* or mother* or father* or famil*)
4	(self or dependent or share or sharing or divid* or spread*) adj3 (care or responsib* or involv* or dut* or task* or job* or administer* or monitor* or injection*)
5	(responsibility or accountability) OR (treatment compliance or disease management or illness behaviour)
6	(treatment compliance or disease management or illness behaviour)
7	4 or 5 or 6
8	1 and 2 and 3 and 7

Appendix 4: Quality criteria checklist for quantitative studies

Quantitative Studies		
<u>Quality criteria</u>	<u>Questions to guide appraisal of criteria</u> ☑ or ☒	<u>Rating</u>
1. <i>Is the rationale clearly described?</i>	<input type="checkbox"/> Is the current evidence base relevant to the study reflected and described comprehensively within the introduction? <input type="checkbox"/> Are gaps or inconsistencies within the evidence base identified? <input type="checkbox"/> Is justification of the need for this area of research presented?	
2. <i>Are the research aims clearly stated?</i>	<input type="checkbox"/> Does the author(s) clearly describe what they plan to research?	
3. <i>Are all ethical issues identified and addressed?</i>	<input type="checkbox"/> Is it stated whether ethical approval has been sought? <input type="checkbox"/> Is there adequate discussion of ethical issues pertinent to the study? (i.e. informed consent, right to withdraw, confidentiality, responding to upset or distress, safeguarding etc.)	
4. <i>Is the methodology identified and justified?</i>	<input type="checkbox"/> Is a quantitative approach and methodology appropriate to the aims of the research?	
5. <i>Is the study design clearly identified and is the rationale for choice of design evident?</i>	<input type="checkbox"/> Is the design of the study clearly stated and justified (e.g. cross-sectional, longitudinal, RCT)?	
6. <i>Is the experimental hypothesis stated?</i>	<input type="checkbox"/> Does the author(s) clearly state what they expect to find?	
7. <i>Are the key variables identified?</i>	<input type="checkbox"/> Are the main variables to be investigated by the study clearly identified?	
8. <i>Is the population from which the sample is drawn identified?</i>	<input type="checkbox"/> Is the description of the sample adequate? (e.g. gender, age, relationship between child/adolescent and caregiver, metabolic control, duration since diagnosis, treatment delivery method etc.)	
9. <i>Is the selection of participants adequately described?</i>	<input type="checkbox"/> Is the context from which the sample was recruited clearly identified? <input type="checkbox"/> Is the method of recruitment to the study adequately described? <input type="checkbox"/> Are the inclusion/exclusion criteria identified (for child/adolescent and parent/caregiver)?	
10. <i>Is the method of data collection valid and reliable?</i>	<input type="checkbox"/> Are the measures suitable for the population? <input type="checkbox"/> Are the measures appropriate to the aims of the study (i.e. Do they measure the desired constructs)? <input type="checkbox"/> Are the psychometric properties of the measures described (e.g. reliability and validity)?	
11. <i>Is the method of data analysis valid and reliable?</i>	<input type="checkbox"/> Are the statistical tests used in analysis of the data stated/described? <input type="checkbox"/> Are the statistical tests appropriate for the data? <input type="checkbox"/> Is the impact of extraneous variables (e.g. gender, age at diagnosis etc.) considered and	

	<p>controlled for within the analysis?</p> <p><input type="checkbox"/> Is evidence of the statistical findings presented (e.g. figures, tables and within the text)?</p> <p><input type="checkbox"/> Are the levels of significance stated?</p>	
<i>12. Are the findings presented clearly and appropriately?</i>	<p><input type="checkbox"/> Are the findings explicitly stated?</p> <p><input type="checkbox"/> Is the statistical data presented clearly?</p> <p><input type="checkbox"/> Are significant and non-significant findings clearly differentiated?</p>	
<i>13. Is the discussion comprehensive?</i>	<p><input type="checkbox"/> Is there a summary of the main findings?</p> <p><input type="checkbox"/> Are the findings linked back to the research aims?</p> <p><input type="checkbox"/> Are the findings linked to the existing/current literature and/or psychological theory?</p> <p><input type="checkbox"/> Is the clinical usefulness of the findings considered?</p>	
<i>14. Are the strengths and limitations of the study identified?</i>	<p><input type="checkbox"/> Are the limitations of the research identified (e.g. sample size, recruitment strategies, generalisability, methods of data collection etc.)</p> <p><input type="checkbox"/> Are the strengths of the research identified (e.g. clinical usefulness, implications for future research etc.)</p>	
<i>15. Are the conclusions made justifiable?</i>	<p><input type="checkbox"/> Are the conclusions supported the discussion of the findings?</p>	

Appendix 5: Quality criteria checklist for qualitative studies

Qualitative Studies		
<u>Quality criteria</u>	<u>Questions to guide appraisal of criteria</u> <input checked="" type="checkbox"/> or <input type="checkbox"/>	<u>Rating</u>
1. <i>Is the rationale clearly described?</i>	<input type="checkbox"/> Is the current evidence base relevant to the study reflected and described comprehensively within the introduction? <input type="checkbox"/> Are gaps or inconsistencies within the evidence base identified? <input type="checkbox"/> Is justification of the need for this area of research presented?	
2. <i>Are the research aims clearly stated?</i>	<input type="checkbox"/> Does the author(s) clearly describe what they plan to research?	
3. <i>Are all ethical issues identified and addressed?</i>	<input type="checkbox"/> Is it stated whether ethical approval has been sought? <input type="checkbox"/> Is there adequate discussion of ethical issues pertinent to the study? (i.e. informed consent, right to withdraw, confidentiality, responding to upset or distress, safeguarding etc.)	
4. <i>Is the methodology identified and justified?</i>	<input type="checkbox"/> Is a qualitative approach and methodology appropriate to the aims of the research?	
5. <i>Is the philosophical background identified?</i>	<input type="checkbox"/> Is the philosophical background identified and described?	
6. <i>Is the study design clearly identified and is the rationale for choice of design evident?</i>	<input type="checkbox"/> Is the design of the study clearly stated and justified (e.g. grounded theory, IPA etc.)?	
7. <i>Are the major concepts identified?</i>	<input type="checkbox"/> Are the key concepts to be explored by the study defined?	
8. <i>Is the sample population situated?</i>	<input type="checkbox"/> Is the description of the sample adequate? (e.g. gender, age, relationship between child/adolescent and caregiver, metabolic control, duration since diagnosis, treatment delivery method etc.)	
9. <i>Is the selection of participants adequately described?</i>	<input type="checkbox"/> Is the context from which the sample was recruited clearly described? <input type="checkbox"/> Is the method of recruitment to the study adequately described? <input type="checkbox"/> Are the inclusion/exclusion criteria identified (for child/adolescent and parent/caregiver)?	
10. <i>Is the method of data collection auditable?</i>	<input type="checkbox"/> Are the methods of data collection described (e.g. semi structured interviews, where data collection took place who collected the data etc.)? <input type="checkbox"/> Are the interview topic areas described, is a copy of the questions made available to the reader?	
11. <i>Is the method of data analysis credible and confirmable?</i>	<input type="checkbox"/> Is the analysis process transparent/clearly described? <input type="checkbox"/> Is made clear how issues of credibility and confirmability were addressed during the analysis and interpretation stages (e.g. external reviewers, dual coders etc)	

	<input type="checkbox"/> Are sufficient quotes/data extracts provided in support of the themes/findings discussed? <input type="checkbox"/> Do the quotes/extracts appear relevant to the themes identified?	
<i>12. Is reflectivity considered and described?</i>	<input type="checkbox"/> Is there acknowledgement of the influence of the research process and the presence of the researcher (including potential biases in analysis and selection of the data, assumptions and experiences etc) on the data collected?	
<i>13. Are the findings presented clearly and appropriately?</i>	<input type="checkbox"/> Are the findings explicitly stated (e.g. themes and sub-themes identified)? <input type="checkbox"/> Are the themes and sub-themes clearly differentiated?	
<i>14. Is the discussion comprehensive?</i>	<input type="checkbox"/> Is there a summary of the main findings? <input type="checkbox"/> Are the findings linked back to the research aims? <input type="checkbox"/> Are the findings linked to the existing/current literature and/or psychological theory? <input type="checkbox"/> Is the clinical usefulness of the findings considered?	
<i>15. Are the strengths and limitations of the study identified?</i>	<input type="checkbox"/> Are the limitations of the research identified (e.g. sample size, recruitment strategies, transferability, methods of data collection etc.) <input type="checkbox"/> Are the strengths of the research identified (e.g. clinical usefulness, implications for future research etc.)	
<i>16. Are the conclusions made justifiable?</i>	<input type="checkbox"/> Are the conclusions supported the discussion of the findings?	

Appendix 6: Parent interview topic guide

Parent's topic guide

1. Can you tell me about how you found out that your child had diabetes and coeliac disease?

Possible prompts (consider in relation to CD and T1DM separately as well as together):

- How old was your child when they were diagnosed?
- What symptoms did they have?
- Who told you?
- What did you think?
- How did you feel then; how do you feel now?
- How did you react?
- Who was with you; what did they think?
- How did others react?
- Did you know that your child was being tested for CD as well as T1DM?

2. How do you explain to others what diabetes and coeliac disease are?

Possible prompts:

- Teachers
- Friends
- Family
- Staff in restaurants, on holidays etc
- How do they react? How does this make you feel?

3. Can you describe how you have been managing your child's diabetes and coeliac disease?

Possible prompts:

- What do you do to manage their diabetes? Tell me about monitoring blood glucose, injecting insulin?
- What do you do to manage their coeliac disease? Tell me about the GFD?
- Do you focus on managing one of the conditions more than the other; if so, in what ways?
- Does one of the conditions limit your child more than the other; if so, in what ways?
- How much do you do to help manage these (e.g. choosing and checking food, telling people, checking blood sugar, injecting insulin)?
- How much do other people do to help manage your child's conditions (e.g. family members, friends, health professionals)? How does this make you feel?
- How do the rest of your family deal with your child's conditions?
- Who helps you to manage the most (doctors, family, friends, teachers, dietician)?
- What would you say to the parents of a child who has just been diagnosed?

4. Can you tell me about how things have changed, if at all, since you found out that your child has diabetes and coeliac disease?

Possible prompts:

- What changes have you had to make to your child's diet, eating behaviour?
- How has the T1DM and CD affected your child's health (physical, psychological, emotional)?
- How are your family's and your child's social activities affected?
- Have your relationships changed (friends and family)?

- What is it like for your child at school?
 - What is it like when you are at home?
 - What is it like when you are away from home? (e.g. family outings/events/holidays)?
 - As a parent, how do you feel about these changes?
 - Do you think these have changed the way your child thinks about them self? How?
 - How would you like things to be different?
5. What is most difficult about your child having both diabetes and coeliac disease?
- Possible prompts:*
- What makes it harder; what makes it easier?
 - Recent example; how did you cope?
 - What concerns you the most; what concerns you the least?
6. What is ok about your child having diabetes and coeliac disease?
- Possible prompts:*
- Do you pick your child's food items? If you are already careful about diet is it much different managing the T1DM too?
 - Are there situations that are really easy?
 - What do these feel like?
7. How do you think other people feel about your child's diabetes and coeliac disease?
- Possible prompts:*
- Friends?
 - Family?
 - How can you tell what these people feel about it?
 - Who worries the most; who worries the least (you, your spouse, your other children)?
 - What is it that makes you think that they worry the most/least?

Appendix 7: Young person's interview topic guide

Young person's topic guide

8. Can you tell me about how you found out that you have diabetes and coeliac disease?

Possible prompts:

- How old were you when you were diagnosed?
- What symptoms did you have?
- Who told you?
- What did you think?
- How did you feel then; how do you feel now?
- How did you react?
- Who was with you; what did they think?
- How did others react?
- Did you know that you were being tested for both CD and T1DM?

9. How do you explain to others what diabetes and coeliac disease are?

Possible prompts:

- Teachers
- Friends
- Family
- Staff in restaurants, on holidays etc
- How do they react? How does this affect how you feel about yourself?

10. Can you describe how you have been managing your diabetes and coeliac disease?

Possible prompts:

- What do you do to manage your diabetes? Tell me about monitoring blood glucose, injecting insulin?
- What do you do to manage your coeliac disease? Tell me about the GFD?
- Do you focus on managing one of the conditions more than the other; in what ways?
- Does one of the conditions limit you more than the other; in what ways?
- How much do you do to help manage these (e.g. choosing and checking food, telling people, checking blood sugar, injecting insulin)?
- How much do other people do (e.g. parents, family members, friends)? How does this make you feel?
- How do your family deal with your conditions?
- Who helps you to manage the most (doctors, parents, family, friends, teachers, dietician)?
- What would you say to someone who has just been diagnosed?

11. Can you tell me about how things have changed, if at all, since you found out that you have diabetes and coeliac disease?

Possible prompts:

- What changes have you had to make to your diet, eating behaviour?
- How has the T1DM and CD affected your health (physical, psychological, emotional)?
- How are your social activities affected?
- Have your relationships changed (friends and family)?
- What is it like at school?

- What is it like at home?
- What is it like away from home (e.g. family outings/events/holidays)?
- How do you feel about these changes?
- Have these changed the way you think about yourself?
- How would you like things to be different?

12. What is most difficult about having both diabetes and coeliac disease?

Possible prompts:

- What makes it harder; what makes it easier?
- Recent example; how did you cope?
- What concerns you the most; what concerns you the least?

13. What is ok about having diabetes and coeliac disease?

Possible prompts:

- Do you pick your food items? If you are already careful about diet is it much different having T1DM too?
- Are there situations that are really easy?
- What do these feel like?

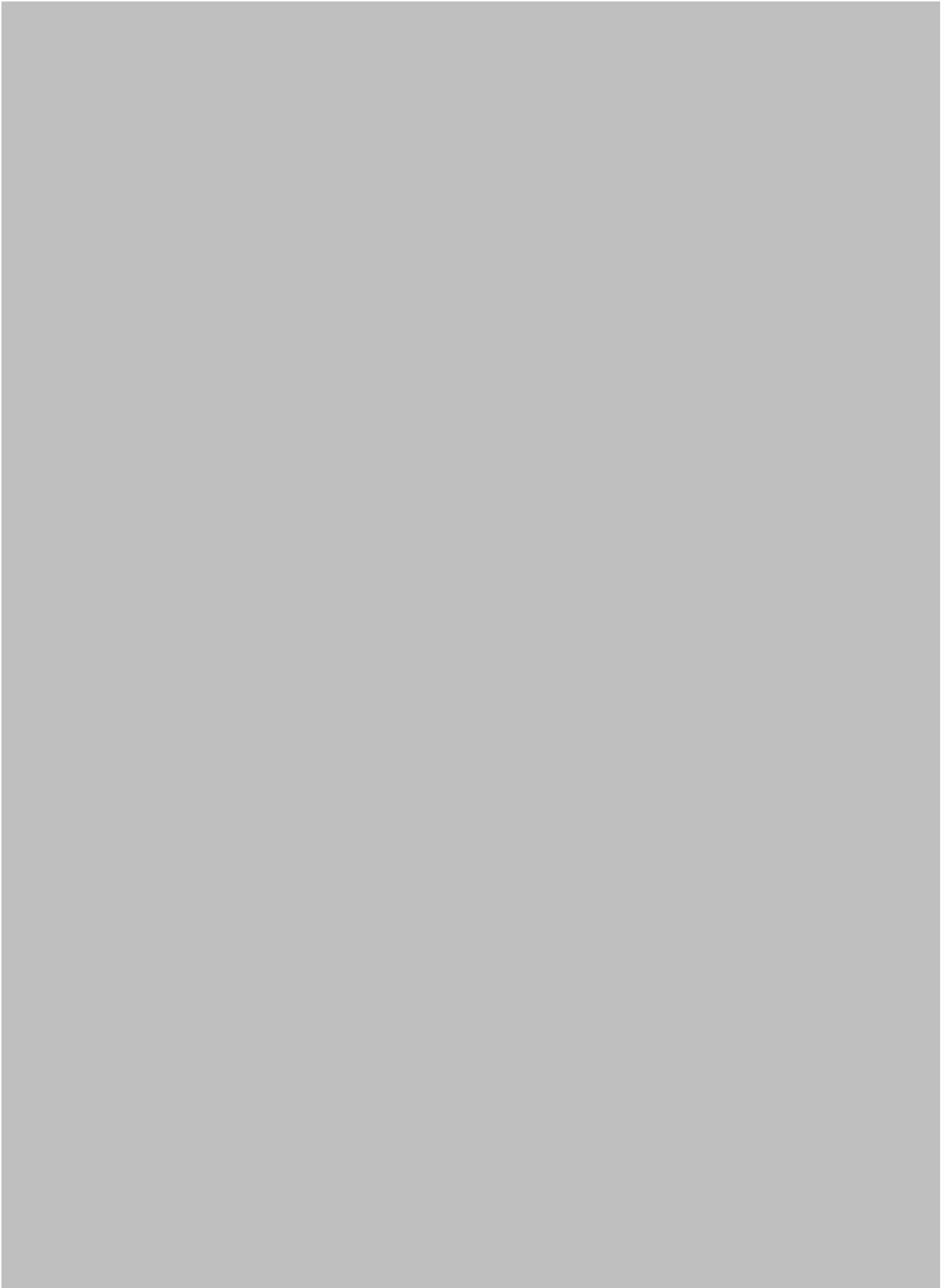
14. How do you think other people feel about your diabetes and coeliac disease?

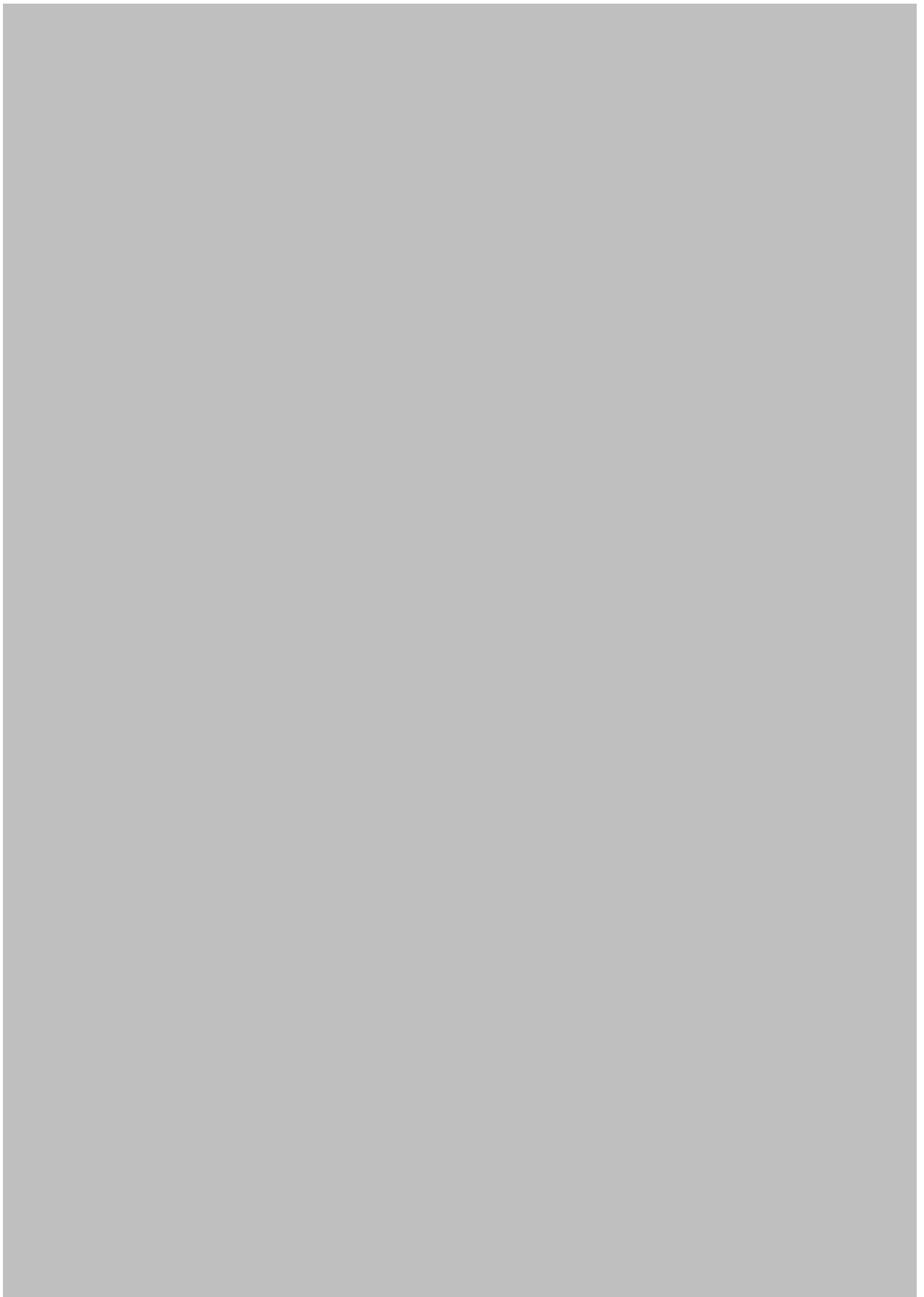
Possible prompts:

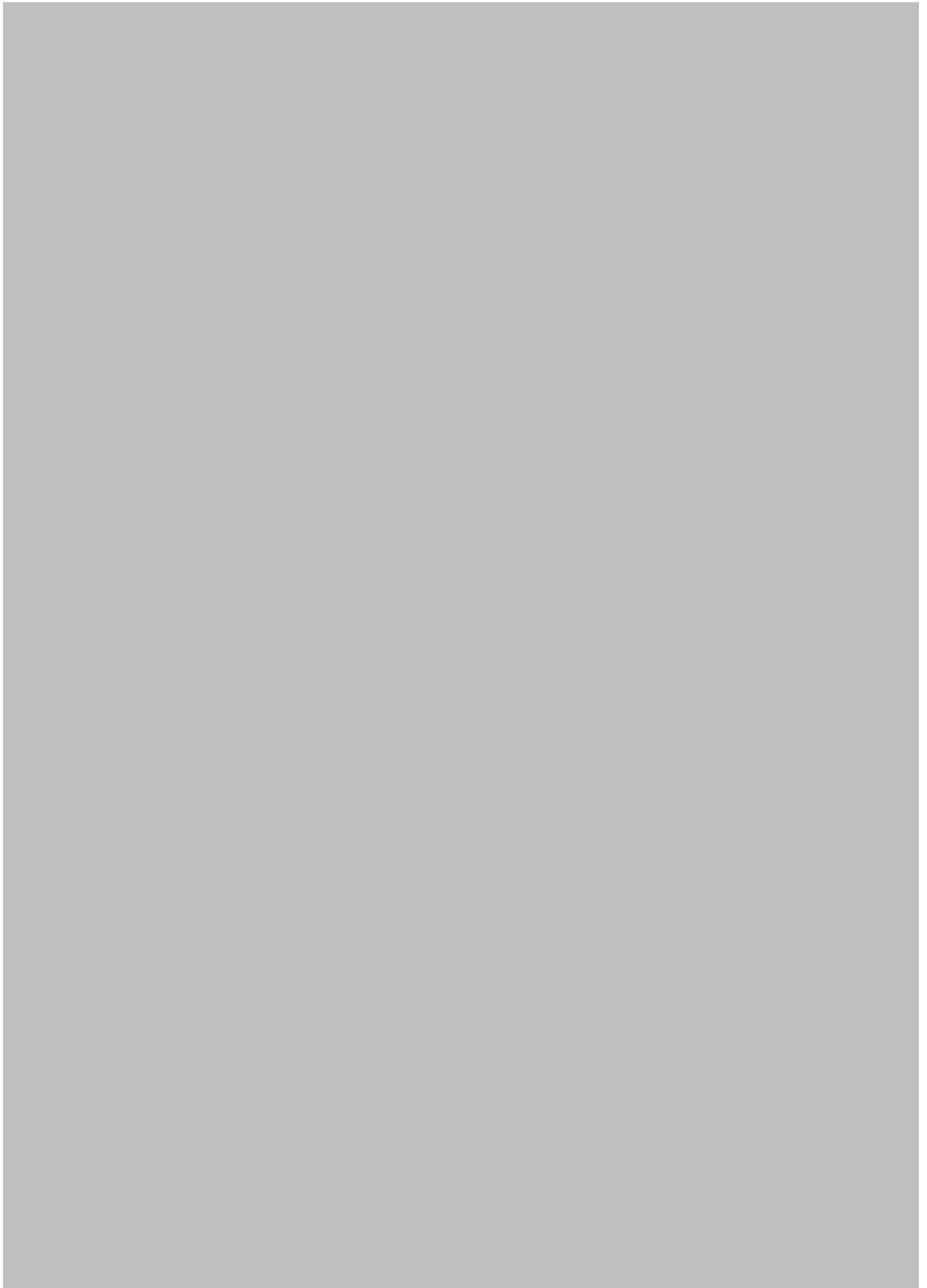
- Parents?
- Friends?
- Family?
- How can you tell what these people feel about it?
- Who worries the most; who worries the least (you, mum, dad, brother, sister)?
- What is it that makes you think that they worry the most/least?

Appendix 8: Ethical and R&D approval letters









Appendix 9: Parent information sheet

Information sheet for parents/guardians

Project Title:

Living with coeliac disease and type 1 diabetes: The experiences of young people and their parents



Hello, thank you for taking the time to read this leaflet, it will invite you to take part in some new research. Before you decide that you would like to take part, it is important for you to read this information carefully so that you understand what the research is about and what you will be asked to do if you take part.

Who are we?

We are researchers at the School of Psychology, University of Birmingham. My name is Maria Love, I am a student at the University and I am completing this research as an important part of my training to become a clinical psychologist. I am being supervised by Dr Ruth Howard and Dr Gary Law who also work at Birmingham University. I will meet with them regularly to make sure that the project runs smoothly.

What is this research about?

We are really interested in finding out about the experiences of young people with both type 1 diabetes and coeliac disease. We would also like to understand more about what parents/guardians feel and think about how living with these two conditions affects your children's hobbies, their time at school, their friendships, and doing things with their family etc.

Why is this research being done?

More and more young people are being diagnosed with both type 1 diabetes and coeliac disease and we would like to know more about how these conditions affect things in everyday day life, like your feelings, what you do and your relationships. It is hoped that the findings of this research will help us to understand more about what it is like for young people to live with both diabetes and coeliac disease together. This may also help to improve how health professionals like doctors, dieticians and nurses support young people and their families with managing these conditions.

Why have you and your child been invited to take part?

We are inviting you to take part because you are a parent of a young person with both diabetes and coeliac disease and your child attends the specialist clinic at [Hospital name].

If I want to take part, what would I have to do....

It is entirely up to you if you want to take part and your decision will not affect the care that your child receives in any way. Before deciding you may like to talk about this with your child. If you do decide that you would like to take part, I will meet with you and your child to talk more about the project and to get your written consent.

... and then what would happen?

Then I will make a time to meet with you somewhere that is convenient for you so I can ask you some questions about your experiences of your child's diabetes and coeliac disease. This will be in the style of an *interview* but the questions will not be hard, it is more like a conversation between us and it will probably last about an hour. I will also meet with your child at a different time to ask them some similar questions. What we say during the interview will be confidential. The interviews will be tape-recorded so that I can write down exactly what we both say. This will help me look for patterns in what people say about their experiences of diabetes and coeliac disease. I will then send this information to you, as it is important that you check my work to make sure that I understood what you said correctly.

Will other people be told what I say?

I will not tell other people, or your child, what you say. I will not tell you what your child has said. However, because I am interviewing both you and your child there is a possibility that you might

recognise something that each other has said when I write up your responses. When I send you the information about what you have said I will ask you to tell me if there are any sentences which you don't want me to write in the final report. If you or your child tell me something that makes me worried that some harm may come to you or someone in your family I would have to discuss this with my supervisor and possibly certain other people too as your safety is very important.

Who else will know that I am taking part?

No other people will be informed of your participation. A letter will be sent to your child's GP to say that your child is taking part in the project. This is normal procedure for projects like this and the letter will just explain what the research is about and that your child has agreed to take part. The doctor will not be told what your child says and we will not send them any further information. The people who are involved in your child's care at the hospital will also know that you and your child are taking part, but again we will not tell them what you or your child have said.

Are there any advantages to taking part in the project?

This project does not offer any treatment but sometimes people find it helpful to talk about what it feels like as a parent of a child with health conditions. If your child chooses to take part they will be given a £10 Amazon voucher. It is hoped that the findings of this research will be made widely available and help improve how young people with diabetes and coeliac disease, and their families, are supported in future.

Are there any potential disadvantages to taking part?

As the project only involves talking about things that you feel comfortable to talk about, it is not expected that there will be any disadvantages to you taking part. If anything about the interview makes you worried then you can talk to me about it. You can also talk to the staff at the hospital who support you and your child with their diabetes and coeliac disease.

What will happen to the results of the project?

The project will be written up as part of my doctorate and bound in my thesis. The work will hopefully be published in a journal that is interested in this area. The final report might include some sentences from our interview but I will change the details so no-one will be able to identify you.

What if I change my mind about taking part?

You can decide to stop taking part, without giving a reason, until the answers from your interview have been written up.

I have some questions....

If you would like to know more about this project please contact either myself (Maria), Ruth or Gary at the University of Birmingham Clinical Office on 0121 4147124. For advice from somebody who is not directly involved with the project please contact the Patient Advice and Liaison Service (PALS) on 0121 333 8403.

If you are unhappy with any aspect of the research and wish to make a complaint or raise a concern please contact the complaints department of [*Hospital name and telephone number*] for advice and support.

Thank you for your time and we would really value your involvement in this research.

Appendix 10: 11-13 year old information sheet

Information sheet for young people aged 11 - 13 year olds



Project Title:

Living with coeliac disease and type 1 diabetes: The experiences of young people and their parents

Hello, my name is Maria Love and I would like to invite you to take part in a research project to find out what it is like for young people to have coeliac disease and diabetes.

Before you decide that you would like to take part, please read this information carefully so that you understand what the project is about. You may like to read this leaflet with your parent/guardian.

Why is this research being done?

We want to find out ways of helping young people with coeliac disease and diabetes by understanding more about how they affect things like your friendships, school, hobbies and going out.



Why have I been invited to take part and do I have to?

You have coeliac disease and diabetes and come to clinics at [Hospital name] and we would like to understand more about how these things affect you.



It is entirely up to you if you want to take part and it will not affect the care you receive from your doctors and nurses.

If I want to take part, what would I have to do....

I will meet with you and your parent/guardian to talk more about the project and to get you and your parent/guardian's agreement in writing. Then I will meet with you to ask you some questions about your coeliac disease and diabetes. It will be like a conversation between us and will probably last about an hour. I will also meet with your parent/guardian at a different time to ask them some similar questions.

The interviews will be tape recorded so that I can write down exactly what we both say. I will send you this information so that you can check my work to make sure that I understood what you said correctly.

Will other people be told what I say?

I will not tell people what you say and I will not tell you what your parent/guardian has said. There is a chance that you and your parent/guardian might recognise something that each other has said, so I will ask you to tell me if there are any sentences which you don't want me to write in the final report.

Also, If you tell me something that makes me worried that some harm may come to you or someone in your family I would have to discuss this with certain people.

Who else will know that I am taking part?

A letter will be sent to your GP, this is just to let them know that you are taking part in the project. The people who are involved in your care at the hospital will also know that you are taking part.

What are the possible benefits of taking part?

This project will not give any treatment but sometimes people find it helpful to talk about what it is like with their health conditions. If you chose to take part you will be given a £10 Amazon voucher. We hope that the results of the project will help improve how young people with coeliac disease and diabetes, and their families, are supported in future.

Is there anything to be worried about?

As the project only involves talking about things that you feel comfortable to, we do not think there is anything to be worried about from taking part.

What will happen to the results of the project?

The project will be written up and hopefully made public so that other people interested in coeliac disease and diabetes can read it. The report might include some of the sentences that you and your parent said but I will try and make sure that people cannot work out who said them.

What if I change my mind about taking part?

You can decide to stop taking part, without giving a reason, until your answers have been written up.

I have some questions....

If you would like to know more about this project please contact either myself (Maria), Ruth or Gary at the University of Birmingham Clinical Office on 0121 4147124. For advice from somebody who is not directly involved with the project please speak to the Patient Advice and Liaison Service (PALS) on 0121 333 8403.

If you are unhappy with any aspect of the research you can make a complaint by calling the complaints department of [Hospital name and telephone number]. Your parent/guardian could do this for you (with your agreement).



Thank you for your time and for any help you may decide to give.



Appendix 11: Young person info sheet

Information sheet for young people

Project Title:

Living with coeliac disease and type 1 diabetes: The experiences of young people and their parents



Hello, thank you for taking the time to read this leaflet, it will invite you to take part in some new research. Before you decide that you would like to take part, it is important for you to read this information carefully so that you understand what the research is about and what you will be asked to do if you take part. You may like to read this leaflet with your parent/guardian.

Who are we?

We are researchers at the School of Psychology, University of Birmingham. My name is Maria Love. I am a student at the University and I am completing this research as an important part of my training to become a clinical psychologist. I am being supervised by Dr Ruth Howard and Dr Gary Law, who also work at Birmingham University. I will meet with them regularly to make sure that the project runs smoothly.

What is this research about?

We are really interested in finding out about the experiences of young people with both type 1 diabetes and coeliac disease. Particularly, we would like to understand more about how living with these two conditions affects things like your hobbies, school, your friendships, doing things with your family, etc. We would also like to know what your parents/guardians feel and think about those things too.



Why is this research being done?

More and more young people are being diagnosed with both type 1 diabetes and coeliac disease and we would like to know more about how having both these conditions affects things in everyday day life, like your feelings, what you do and your relationships. It is hoped that the findings of this research will help us to understand more about what it is like for young people to live with both diabetes and coeliac disease together. This may also help to improve how health professionals like doctors, dieticians and nurses support young people and their families with managing these conditions.

Why have I been invited to take part?

We are inviting you to take part because you come to clinic at [Hospital name] and you are a young person with both diabetes and coeliac disease.

If I want to take part, what would I have to do....

It is entirely up to you if you want to take part and your decision will not affect the care you receive in any way. Before deciding you may like to talk about this with your parent/guardian or doctor. If you do decide that you would like to take part, I will meet with you and your parent/guardian to talk more about the project and to get your agreement in writing. I will also ask you if it is ok for me to access some information about your diagnosis and treatment from your medical notes. This information will help me to understand how your experiences may be different or similar to other young people with both coeliac disease and diabetes who take part in the research.



... and then what would happen?

Then I will make a time to meet with you somewhere that is convenient for you so I can ask you some questions about your diabetes and coeliac disease. We call this an *interview* but the questions will not be hard and this is not a test, it is more like a conversation between us and it will probably last about an hour. I will also meet with your parent/guardian at a different time to ask them some similar questions. What we say during the interview will be confidential. The interviews will be tape-recorded so that I can write down exactly what we both say. This will help me look for patterns in what people say about their experiences of diabetes and coeliac disease. I will then send this information to you, as it is important that you check my work to make sure that I understood what you said correctly.

Will other people be told what I say?

I will not tell other people, or your parent/guardian, what you say. I will not tell you what your parent/guardian has said. However, because I am interviewing both you and your parent there is a possibility that you might recognise something that each other has said when I write up your responses. When I send you the information about what you have said I will ask you to tell me if there are any sentences which you don't want me to write in the final report.



Also, if you tell me something that makes me worried that some harm may come to you or someone in your family I would have to discuss this with my supervisor and possibly certain other people too as your safety is very important.

Who else will know that I am taking part?

A letter will be sent to your GP to say that you are taking part in the project. This is normal procedure for projects like this and the letter will just explain what the research is about and that you have agreed to take part. The doctor will not be told what you say and we will not send them any more letters. The people who are involved in your care at the hospital will also know that you are taking part, but again, we will not tell them what you say in the interview.

Are there any advantages to taking part in the project?

This project does not offer any treatment but sometimes people find it helpful to talk about what it is like with their health conditions. If you chose to take part you will be given a £10 Amazon voucher. It is hoped that the findings will be made widely available and help improve how young people with diabetes and coeliac disease, and their families, are supported in future.

Are there any disadvantages to taking part?

As the project only involves talking about things that you feel comfortable to talk about, it is not expected that there will be any disadvantages to you taking part. If anything about the interview makes you worried then you can talk to me about it, or to your parent/guardian. You can also talk to the staff at the hospital who support you with your diabetes and coeliac disease.

What will happen to the results of the project?

The project will be written up as part of my doctorate and bound in my thesis. The work will hopefully be published in a journal that is interested in this area. The final report might include some sentences from our interview but I will change the details so no-one will be able to identify you.

What if I change my mind about taking part?

You can decide to stop taking part, without giving a reason, until the answers from your interview have been written up.

**I have some questions....**

If you would like to know more about this project please contact either myself (Maria), Ruth or Gary at the University of Birmingham Clinical Office on 0121 4147124. For advice from somebody who is not directly involved with the project please contact the Patient Advice and Liaison Service (PALS) on 0121 333 8403.

If you are unhappy with any aspect of the research and wish to make a complaint or raise a concern please contact the complaints department of [Hospital name and telephone number] for advice and support.



Thank you for your time and we would really value your involvement in this research.

Appendix 12: Parent letter of invitation (support groups)

Letter of invitation to participate in research

UNIVERSITY OF
BIRMINGHAM

Would you like to help us learn more about the experiences of young people with both type 1 diabetes and coeliac disease?

Living with coeliac disease and type 1 diabetes:
The experiences of young people and their parents

We hope that this new research will help us to improve how we support young people and their families to manage these conditions.

As you are a parent of a young person with both diabetes and coeliac disease I am offering you the opportunity to take part in a conversation where I will ask you questions about what you feel and think about what it is like for your child to have diabetes and celiac disease.

We will also invite your child to take part as we would like to know what they feel and think about these things too.

Would you like to know more...?

I have enclosed an information sheet for you and your child explaining more about the project and what taking part would involve. You may want to read and discuss this information with your child.

Thank you for reading this letter, your involvement with this research would be really valuable. If you are interested in participating please contact me via one of the ways listed below:

- Email (for direct contact with Maria): MXL074@bham.ac.uk
- Telephone (University of Birmingham Clinical Office): 0121 4147124
- Fill in the slip below and return it to me in the stamp addressed envelope provided

***** A £10 Amazon gift voucher will be given to the young people who participate*****

After I have heard from you I will contact you to talk more about the project

Thank you



I have discussed this with my child and I would/would not (please delete as appropriate) be interested in taking part

Parent's name: Your child's name:

Contact telephone number:

Please indicate the best day(s)/time(s) to contact you: Mon/Tues/Weds/Thurs/Fri/Sat/Sun
am/pm/evenings

Appendix 13: Young person letter of invitation (support groups)

Letter of invitation to participate in research

UNIVERSITY OF
BIRMINGHAM

Would you like to help us learn more about the experiences of young people with both type 1 diabetes and coeliac disease?

Living with coeliac disease and type 1 diabetes:
The experiences of young people and their parents

This new research aims to help us learn more about the experiences of young people with both type 1 diabetes and celiac disease so that we can try to improve how we support young people and their families to manage these conditions.

As you are a young person with both diabetes and coeliac disease I am offering you the opportunity take part in a conversation where I will ask you questions about what it is like for you to have these conditions.

I will also invite your parent/guardian to take part as we would like to know what they feel and think about these things too.

Would you like to know more...?

I have enclosed an information sheet for you and your parent/guardian explaining more about the project and what taking part would involve. You may want to read and discuss this information with your parent/guardian.

Thank you for reading this letter. If you are interested in participating please contact me via one of the ways listed below, or you could ask your parent to do this for you:

- Email (for direct contact with Maria Love): MXL074@bham.ac.uk
- Telephone (University of Birmingham Clinical Office): 0121 4147124
- Fill in the slip below and return it to me in the stamp addressed envelope provided

***** A £10 Amazon gift voucher will be given to the young people who participate*****

After I have heard from you I will contact your parent to talk more about the project

Thank you



I have discussed this with my parent and I would/would not (please delete as appropriate) be interested in taking part

Name of young person: Age of young person:

Name of parent:

Contact telephone number:.....

Please ask your parent to indicate the best day(s)/time(s) to call them:

Mon/Tues/Weds/Thurs/Fri/Sat/Sun am/pm/evenings

Appendix 14: Parent consent form

UNIVERSITY OF
BIRMINGHAM

Living with coeliac disease and type 1 diabetes: The experiences of young people and their parents

Consent Form

This section should be completed by the participant:

Please tick the box if you agree
with the statements below

I confirm that I have read the information leaflet about the project

☐

The project has been explained to me by the researcher

☐

I have had the chance to ask questions about the project

☐

I have understood all of the answers to my questions

☐

I understand what the project is about

☐

I understand that I can stop taking part without having to give a
reason

☐

I understand that my interview will be recorded and that some of
what I say might be written in the final report

☐

I would like to take part in this project

☐

Please sign here if you want to take part and have ticked all of the boxes above.

Name:

Signature:

Date:

I would be happy to be contacted for future research

☐

To be completed by the researcher:

The participant has been provided with an information sheet and I have explained the project to them. He/she has indicated his/her willingness to participate.

Name of researcher: Signature:

Date:

Appendix 15: Young person consent form

UNIVERSITY OF
BIRMINGHAM

*Living with coeliac disease and type 1 diabetes: The experiences of young people
and their parents*
Consent Form



This section should be completed by the young person:

Please tick the box if you agree
with the statements below

I have read the information leaflet about the project

☐

Maria has explained the project to me

☐

I have had the chance to ask questions about the project

☐

I have understood all of the answers to my questions

☐

I understand what the project is about

☐

I understand that I can stop taking part without having to give a
reason

☐

I understand that my interview will be recorded and that some of
what I say might be written in the final report

☐

I would like to take part in this project

☐

I understand that relevant sections of my medical notes and data collected
during the study may be looked at by individuals from 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 records.

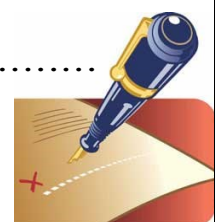
☐

Please sign here if you want to take part and have ticked all of the boxes above.

Name: Signature:

Date:

I would be happy to be contacted for future research

☐

Please turn over...

If you are under 16 years old your parent or guardian must also sign this form if they are happy for you to take part in the project

I give my consent for my son/daughter (name) to take part in this project.

Name of parent/guardian:

Signature:

Date:

To be completed by the researcher:

The participant has been provided with an information sheet and I have explained the project to them. He/she has indicated his/her willingness to participate.

Name of researcher:

Signature:

Date: