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Assessing the impact of parental mental health on child physical health: validation of a measure of carer burden within carers of children with chronic kidney disease

Section A: Does parental mental health impact on a child's physical health? Exploring the relationship between parental mental health and child physical health within chronic paediatric populations

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SECTION B: Assessing Factor Structure, Validity and Reliability of the Paediatric Renal Care Burden Scale (PR-CBS): A measure of carer burden for carers of children with chronic kidney disease

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Summary of portfolio

Section A: provides an overview of the literature investigating the relationship between parental mental health and child physical health within populations of children with chronic childhood illness. Evidence for whether this relationship definitively exists, as well as the potential pathways through which this relationship could operate, is evaluated.

Section B: is an empirical study aiming to assess the validity and reliability of the Paediatric Renal Carer Burden Scale (PR-CBS), a psychometric measure designed to assess carer burden in carers of children with chronic kidney disease. Factor structure of the measure was assessed as internal reliability, construct validity and acceptability to carers was assessed.

Section C: provides a critical evaluation of the research process and reflections from the researcher on learning throughout the process of the study as well as clinical implications and future research directions.

Summary of contents

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Section A:

Does parental mental health impact on a child's physical health?

Exploring the relationship between parental mental health and child physical health within chronic paediatric populations

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Abstract

The impact of a parent's mental health on their child's physical health, when the child has a chronic paediatric condition has, to date, generated little consensus in the literature. Individual studies within specific paediatric populations have resulted in mixed findings. This study aims to provide an overview of the current literature encompassing all chronic childhood conditions, to determine whether a definitive relationship does exist and through which pathways this may potentially operate. Of the 24 articles reviewed, 15 demonstrated a relationship between parental psychological functioning and the child's physical health across several chronic paediatric conditions including chronic kidney disease, those children undergoing kidney or liver transplants and asthma. Poorer parental mental health was associated with measures of the child's physical health including BMI, height, weight, the use of emergency department, parental report of disease morbidity and development and diagnosis of asthma. Support was found for two separate pathways; either by reducing the caregiver's ability to maintain appropriate adherence to medical regimens or through direct physiological pathways which alter the child's immune system and their response to allergens. Further research is needed to validate these findings within other disease groups, explore other potential pathways through which this relationship might operate and determine variables which may modulate the relationship

Keywords: Caregiver stress, carer burden, parental mental health, chronic paediatric conditions, paediatric illness

Introduction

Chronic Paediatric Conditions

Chronic paediatric conditions are defined as a long term illness in a child which lasts (or is predicted to last) for more than 3 months, or requires hospitalisation for over a month (Jessop et al., 1988; Wallander & Varni, 1998). In the UK, data from the Millennium cohort study, which followed 14, 556 children from birth in 2000 to 2010, determined the prevalence of chronic illness within the cohort as being 15.8% (Nikiema et al., 2010), 2.8% of which had conditions which limited the child's ability to participate in typical daily activities.

There is a wide literature base exploring the impact on a family when caring for a child with a chronic condition, with conflicting findings. Some studies have shown these children to place considerable extra strain on families (Grootenhuis & Bronner, 2009; Vrijmoet-Wiersma et al., 2008), particularly mothers, who predominantly fall into the caregiving role (Jessop et al., 1988). For example, two studies found mothers of children with a chronic condition have a significantly higher prevalence of mental health difficulties when compared to the general population (Szabo et al., 2009; Wade et al., 1997). Others studies have concluded the opposite, reporting no difference in levels of parental mental health difficulties within these families when compared with community samples (Fredericks et al., 2007; Walker et al., 1992). A review conducted by Wallander and Varni (1998) concluded that mothers of children with chronic health conditions do have an increased risk of adjustment difficulties; however there was variation within each study sample, suggesting that a reasonable proportion of parents are able to psychologically adjust to the demands of caring for their child. These findings were limited by the fact that many of the studies included in the review tended to capture data on parental psychological functioning at one time point only, not allowing scope to explore a potentially fluctuating and complex relationship between parental mental health and a child's physical illness.

Impact of Parental Mental Health[1] on Children's Physical Well-being in Healthy Populations.

A link between maternal mental health and a child's physical health in non-chronic populations has been demonstrated. Guttman et al. (2003) conducted a population based study in Canada, identifying mothers of adolescents where the child had been hospitalised over the previous year. Using a model which accounted for numerous psycho-social variables, they demonstrated that maternal depression increased the risk of child hospitalisation by almost two fold, greater than any other variable, other than child's current health status. Leiferman (2002) found that mothers who were depressed were more likely to smoke, not put their child in a car seat or give their child daily vitamins, all of which are recognised preventative health practices. Other studies have thrown up somewhat more confusing results. For example maternal depression was shown to be adversely associated with some health promoting practices, such as dental check-ups and regular teeth brushing, but not others such as up to date immunisations or regular medical check-ups (Kavanaugh et al., 2006).

Rationale for this Review.

Currently there appears little consensus as to whether a relationship between parental mental health and a child's physical health in children with chronic conditions exists; no review to date has investigated whether this relationship is robustly present across different chronic conditions. Additionally, there are few theoretical models which propose how a relationship between parental mental health and child physical health might operate. Studying the influences of family characteristics on paediatric asthma, Kaugars et al. (2004) has suggested a unidirectional model with two pathways; either through asthma management behaviours or through physiological factors that impact the child's immune system. Others propose a bidirectional model, where the stress of

1 Within this paper the term 'parental mental health' is used as an overarching concept to encompass several different terms used in the literature including parental stress, distress, depression, burden and strain. Previous articles also using this as an umbrella term have defined it as “the variety and flexibility of emotional response of (the mother) and is measured by the intensity and frequency of certain signs and symptoms. Psychiatric diagnosis is not implied...” (Jessop et al. 1988).

having a child with a chronic illness causes distress in the parent, which then modifies their ability to provide appropriate care (Kazak et al., 2006; Klinnert et al., 2008).

Pless and Pinkerton (cited in Wallander and Varni, 1998) argue that the psychosocial commonalities of caring for a child with a chronic illness warrant studies which take a non categorical approach across conditions. Different areas of paediatric psychology have conducted research within their own medical area, but few studies span illness groups. This current review aims to shed light on two main questions within this area of research: is there a robust relationship between parental mental health and a child's morbidity in children with chronic paediatric illness across conditions and if so, through which pathways could this variable potentially operate?

Literature Search

A literature review was conducted which generated 6 relevant articles (see Appendix A for details). A further 18 were found through hand searching of reference lists of relevant texts, yielding overall 24 articles to review. Articles were included if they directly investigated a relationship between parental mental health and the child's physical health in children with chronic conditions, or provided evidence for a potential pathway through which this relationship could operate. A brief overview of the papers included in this review can be found in Appendix B.

Summary of Findings

Is There a Relationship Between Parental Mental Health and Child Morbidity?

Several studies were identified which aimed to empirically assess any relationship between maternal mental health and child physical health within paediatric populations. Diseth et al. (2010) used the General Health Questionnaire (GHQ) (Goldberg, 1978) to assess psychological functioning in 32 mothers of children who had undergone kidney transplantation and the child's Body Mass Index (BMI), renal function and cardiorespiratory fitness were recorded. Increased BMI was significantly correlated with poorer maternal mental health, but as the study was cross-sectional in

nature, causal relationships could not be determined. Other child physiological variables were unrelated to maternal mental health. Watson (1997) also assessed maternal mental health in mothers of children with kidney disease, this time using the Perceived Stress Scale (Cohen et al., 1983) and the Hospital Anxiety and Depression Scale (Zigmond & Snaith, 1983). 38 mothers completed the measures prior to the child undergoing renal replacement therapy and then at subsequent time points over the following year. The child's weight and height were monitored. A measurement of "Burden of Care" (BCA) was developed, using assessment by four clinicians involved with the family. Parents with higher BCA scores experienced higher levels of stress, anxiety and depression and their children had lower weight and height indices. Though using different methodologies, both studies demonstrate an empirical relationship between maternal mental health and BMI, weight and height, in children with kidney disease. This is significant, as reduced growth parameters are associated with higher rates of morbidity and mortality within this paediatric population (Furth et al., 2002).

Asthma is the most common chronic paediatric condition (Wood et al., 2002) and as such there has been substantial research into the interaction between child morbidity and psycho-social variables. Caregivers with more depressive symptoms, as indicated on the Center for Epidemiological Studies-Depression scale (CES-D) (Radloff, 1977), were shown to have children with higher levels of asthma morbidity, which was determined using a composite measure of physician rated symptomatology and utilisation of health care (Shalowitz et al., 2001). Levels of depression in the parent were shown to be the strongest independent variable to contribute to levels of asthma morbidity (Shalowitz et al., 2001). Wood et al. (2002) used questionnaire data from 386 children with asthma to show that mental health difficulties in parents, as assessed using the five item Mental Health Scale (Ware et al., 1993), were associated with parental report of more asthma symptoms and higher levels of health care use in their children. Using data taken from a national study of 1528 children with asthma, Weil et al. (1999) reported that parents with clinically significant depressive symptoms, measured on the Brief Symptom Inventory (BSI) (Derogatis et al.,

1975), were almost twice as likely to have their child hospitalised for asthma in the 9 months following base line. Similarly, Bartlett et al. (2001) assessed whether there was a relationship between maternal depressive symptoms and emergency department (ED) use, in mothers of children with with asthma. Mothers with high levels of depressive symptoms on the CES-D at baseline were 30% more likely to present their child at the ED during the subsequent six months, which, along with mother's age, was the strongest predictor of ED use. All of these studies demonstrate significant associations between parental mental health, using various measures, and the child's asthma severity, also using a variety of morbidity indicators.

Both Weil et al. (1999) and Bartlett et al. (2001) also investigated the relationship between parent's mental health and asthma morbidity, as based on parental report of symptomatology. Interestingly, despite Bartlett et al. (2001) having found a significant relationship between hospitalisation and parental mental health, neither study found a significant relationship when child morbidity was based on parental report. Other studies have found similarly complex results when one measure of the child's physical health is based on parental report. In a sample of 209 chronically ill children, Jessop et al. (1988) reported that mothers with more psychiatric symptoms, based on a shortened version of the Hopkins Symptom Distress Checklist (Ilfield, 1976), rated their child as being more functionally impaired by their condition. However the child's clinician also completed the Clinicians Overall Burden Index (COBI), a tool designed to be an objective measure of burden which considers the child's health needs, and which was used as an indicator of the child's health. The COBI was found to be unrelated to the psychological functioning of the mother. This suggests that mother's rating higher levels of distress, experience their child as being more unwell and functionally impaired than perhaps a more objective clinician's rating of the child's health. Szabo et al. (2009) also found no significant relationship between parental mental health using the Beck Depression Inventory (BDI) (Beck, 1961) and State-Trait Anxiety Inventory (STAI) (Spielberger et al., 1983), when asthma severity was rated by clinician against World Health Organisation classification. Given these findings, studies where the only measure of child morbidity

is parental report should be interpreted with caution as they may be subjective and incorporate parental bias. Previous studies with chronic paediatric populations have shown that mothers who are more psychologically distressed report their children as having more behavioural problems (Madden et al., 2002).

Several other studies have found no significant relationship between parental mental health and child's physical health, despite using measures other than parental report as an indicator of the child's morbidity. Rodrigue et al. (1997) assessed psychological functioning of 27 mothers of children prior to undergoing various transplant operations, and then reassessed at one and six months post-transplant, using the Parenting Stress Index (Abidin, 1995) and Impact on Family Scale (Stein & Reissman, 1985). Medical parameters recorded were the length of transplant hospitalisation and number of post-transplant hospitalisations. Though parental stress was significantly elevated post operation, neither of the measures related to the child's medical parameters across any time point. Similarly, Kovacs et al. (1995) found there to be no relationship between children who had multiple hospitalisation (classified as three or more over a three year period) and maternal psychopathology measured on the Hopkins Symptom Checklist, in a longitudinal study of 92 children with insulin dependent diabetes mellitus. Finally, Fielding and Brownbridge (1999) used the Leeds Scale for the Self-Assessment of Anxiety and Depression (Snaith et al.,1976) to assess psychological functioning in parents of children in end stage renal failure and found no significant correlation between parents' score on the measure and the number of hospitalisations their child had undergone. Of note is that these three studies used only hospitalisation as the indicator of child health, which tends to be a marker for a severe crisis in the child's condition and might not be representative of the different variations in the child's daily physical health. Rodrigue et al. (1997) suggested the use of potentially more sensitive measures of health fluctuation in future research.

Support for Potential Pathways

Direct alteration of the child's physiology.

The acknowledged genetic risk in the development of childhood asthma (Klinnert et al., 1994) has enabled prospective monitoring of psycho-social variables which might contribute to illness development in those children identified with a genetic vulnerability. Several such studies have been undertaken.

Klinnert et al. (1994) identified a cohort of 150 unborn children, genetically at risk of asthma as assessed through diagnosis of asthma in either one or both parents. The Family Inventory of Life Events (FILE) (McCubbin et al., 1983) was used to assess levels of stress in the mother prior to the birth. At three weeks old, a score for "Parenting Risk" was determined based on observation of the quality of mother-infant relationship. Mothers who rated poorly on the "Parenting Risk" scale had children who were significantly more likely to develop asthma and a non-significant trend was found towards mothers who reported higher stress prior to birth being more likely to have a child with asthma diagnosis at three years. The theoretical consideration that quality of parenting modulated the risk of stress from the mother to the infant was examined. The cohort was divided into four groups: low stress and high stress with no parenting problems, and low stress or high stress with parenting problems. An interaction between maternal stress and Parenting Risk was found to be a significant predictor of asthma onset; therefore indicating that good parenting acted as a protective factor to stop children developing asthma even when high levels of stress were reported. In a subsequent publication using the same cohort, Mzarek et al. (1999) reported that perinatal depression was significantly related to the "Parenting Risk" score and increased the relative risk of the child developing asthma by 3.9. Though 11 different risk variables were analysed, only four were significant predictors of asthma development; frequent illness, presence of Immunoglobulin-E (an antibody, high levels of which are associated with infection), parenting difficulties and eczema. Early parenting difficulties were shown to independently contribute to the risk of asthma development.

More recently Klinnert et al. (2008) assessed whether maternal mental health and family stress was associated with asthma development, using the Mental Health Inventory (Ware et al. 1985) and a "Family Stress" composite measure with mothers of a cohort of children aged 9-24 months with a history of respiratory illness and episodes of wheezing. Maternal mental health at baseline was unrelated to asthma diagnosis (based on maternal report and medical records) at four years, however higher family stress was associated with asthma diagnosis. The data were further analysed using stepwise regression modelling which showed the "Family Stress" variable attenuated once other variables were entered, though was still significant. This somewhat contradicts the earlier findings of both Klinnert et al. (1994) and Mzarek et al. (1999) and some aspects of the methodology of this more recent paper need to be considered. Firstly, family stress was a composite measure created by the research team using four different scales for which no robust statistical parameters were reported. Secondly, unlike the cohort used in the earlier two studies, the children in this study already reported respiratory illness and wheezing. Already having an unwell child may have an influence on maternal mental health and family stress levels in a more complex way.

Wright et al. (2002) demonstrated that increased levels of stress in caregivers, as measured on the PSS when the child was two-three months old, predicted frequent (two or more) episodes of wheezing at 14 months, in a sample of 436 children. This was independent of other variables shown to contribute to asthma development such as maternal smoking and presence of allergens. Furthermore, the relationship was unidirectional in that wheezing in the infant, assessed at a previous time point, did not predict caregiver stress. A biological pathway has been proposed to explain these results. Wright et al. (2002) suggests that immune response regulation is modified during early childhood when caregiver stress influences infant stress response. Stress responses have been shown to have a physiological impact in adults' immune systems (Herbert & Cohen, 1993) and children with atopic dermatitis have been shown to have attenuated cortisol responses to psycho-social stressors (Buske-Kirschbaum et al., 1997). In asthma this functional change may be

to the airway's sensitivity to allergens and the inflammatory processes that obstruct airways, thus producing wheezing. Wright et al. (2004) later showed that chronic stress in a parent caring for a child genetically at risk of asthma, was associated with increased Immunoglobulin-E expression in the child, which affects cytokine profiles, both of which are markers of alterations in the immune responses.

Wolf et al. (2008) further added weight to this biological pathway hypothesis by comparing inflammatory markers at two time points in both healthy children and children with asthma, and comparing this with parental depression and stress, as measured on the CES-D and PSS. The child's levels of depression and anxiety were also taken into account. Analysis showed that both parental depression and stress predicted increases in the child's inflammatory responses over the subsequent six months and this was independent of the child's psychological functioning. This was found both in children with asthma and healthy children, which suggests that parental mental health exacerbates symptoms and increases the likelihood of developing asthma for those children genetically predisposed. Additionally, it appeared that the two different psychological constructs measured, depression and stress, may influence the inflammatory response in slightly different ways. Parental stress was associated with changes in eosinophil cationic protein (ECP) levels and inter-leukin (IL)-4 production, both of which are implicated in increased inflammatory responses in the body and lead to asthma symptoms, whilst depression was only associated with changes in ECP levels. This suggests that the effects of parental stress may be more robust and chronic in their impact on a child's physical health response than other psychological constructs such as depression.

Five out of six of these mother-infant studies demonstrate that parental stress or depression independently contributes to asthma development. Furthermore, two studies found evidence for a specific biological pathway through which this variable may impact on the child's physiology. The prospective nature of these studies suggests a unidirectional relationship between parental mental health and asthma development.

Poor adherence to treatment regimens.

Psychological factors that are associated with medication adherence is something that has generated research interest in both adult and children with chronic conditions (DiMatteo et al., 2000; Greca et al., 2009). Several studies have shown a link between parental mental health difficulties and adherence to medication regimens within paediatric populations.

Fredericks et al. (2007) assessed psychological functioning in 38 parents of children within 5 years of liver transplantation, using a battery of psychometric measures including the BSI, BDI, Pediatric Inventory for Parents (PIP) (Streisland et al. 2001) and Child Health Questionnaire-Parent Form 50 (Landgraf et al., 1996). Medical regimen adherence was investigated retrospectively and was based on clinic attendance and immunosuppressant levels monitored through blood samples. Health parameters of graft function, frequency of hospitalisation and liver biopsies and episodes of rejection were also recorded. The results provide support for the idea that adherence variables might mediate a relationship between elevated parental stress and child's physical health outcomes. Immunosuppressant adherence was negatively associated with parental emotional functioning as measured on the CHQ-PF 50 and poor clinic adherence was significantly related to higher scores on the PIP. Non-adherence of immunosuppressant and poor clinic attendance in turn both significantly correlated to the number and duration of hospitalisations and liver biopsies. Clinic attendance was also significantly related to number of rejection episodes.

Other studies have demonstrated a similar finding using kidney transplant patients. Gerson et al. (2004) asked parents to complete the PSI and the CHQ-PF, which in this study was used to measure the child’s level of pain and discomfort, prior to transplantation. Patients were classified as either probably adherent (PA) versus probably non-adherent (PNA) to medication, based on electronic monitoring of their dosette box and blood samples. Parental stress at baseline was significantly higher in families that went on to be classified as PNA and these parents also rated their child’s pain and discomfort as being higher. Despite a small sample size of 13 families, and only a moderate correlation between the adherence measures ($r = 0.59$), the prospective nature of

this study adds strength to the argument that parental stress precedes the stress of maintaining the medication regimen. It should be noted however that the measure of the child's health, CHQ-PF, was based on solely parental report, and therefore as discussed before, should be interpreted with caution.

Others studies have also used parental report as a measure of treatment regimen adherence. Using the same sample as Weil et al. (1999), Baumen et al. (2002) showed that caregiver depression, as measured on the BSI, in carers of children with asthma was related to self reported non-adherence by parents and was also related to parental report of asthma morbidity at nine month follow-up. In contrast, Bartlett et al. (2004) found no significant relationship between maternal report of asthma morbidity and non-adherence. They assessed depression in 177 mothers of children with asthma using the CES-D, and then classified them into either high or low depressive symptoms sub-groups. Mothers in the high symptoms group were more likely to report their child had problems using their inhaler and forgot medication regularly, though this was not related to asthma morbidity as based on maternal report.

One study which used a more objective measure of child health outcome was Chisholm et al. (2007). They assessed levels of stress using the PSI in 65 mothers' of children with type 1 diabetes and monitored the child's health by using blood samples to predict level of glycaemic control. However, adherence to treatment regimen was still based on maternal report and scores were based on variables such as injection frequency and diet. However, in contrast to previous findings, parental stress was unrelated to both treatment adherence and child's health outcome. Again, the lack of a significant relationship may be due to the potentially confounding influence of maternal report.

Though interesting preliminary findings, the main methodological issue with the studies discussed so far is the lack of an objective and representative measure of either adherence or child health outcomes. For those that have chosen more objective measures of adherence, such as blood samples, these tend not to correlate particularly well with the other measures of adherence such as

dosette box monitoring, which then cast doubts over the validity of either measure. Whilst those using a child outcome measure based solely on parental report, which has been shown to be subject to parental bias (Jessop et al. 1988), often report conflicted findings (Bartlett et al. 2004).

Other studies, despite demonstrating a significant relationship between treatment adherence and elevated parental distress, failed to include a direct measure of child health outcomes. Using the FILE scale, Foulkes et al. (1993) looked at the relationship between family stress and compliance to three medications after renal transplantation. Compliance was measured in two ways; assessment by a pharmacist of the discrepancy between the amount of medication the patient returned compared with the amount they should have taken over the time period, and measurement of blood samples for levels of immunosuppressant. Stress levels were significantly related to non-compliance of only one of the medications investigated, azathiprine, and regression modelling showed this to account for only 23% of the variance in compliance. It should be noted that compliance was only measured at one time point, and again the correlation between the two measures of compliance was modest ($r=0.44$). Despite other research demonstrating that non-adherence to medication post transplantation has been linked to increase in physical health difficulties, hospitalisation (Fredericks et al., 2007) and morbidity in paediatric liver transplant patients (Shemash et al., 2004) this study would have been significantly strengthened had it included a specific measure of child physical health outcome. Another study with the same methodological weakness is Bourdeau et al. (2007) who used the PSI to investigate the relationship between parental stress and children's ability to follow treatment regimens, such as monitoring glucose levels or taking medications, across various paediatric diseases. Regimen adherence was rated separately by both parent and child. Higher parenting stress was the primary predictor of poorer child self-care as rated by the parent, and there was a trend towards high parenting stress correlating significantly with the child's report of adherence.

Overall, there is tentative evidence to suggest that in some conditions, the relationship between parental mental health and child physical health is mediated by non-adherence to

medication and treatment regimen. However these positive preliminarily results are greatly overshadowed by a host of methodological flaws in many of the studies; the lack of a valid and reliable method with which to assess adherence, the exclusion of any measure or monitoring of the child's physical health in two of the studies and the sole use of parental report in others.

Discussion

Of the 24 articles reviewed in this study, 15 demonstrated a relationship between parental psychological functioning and the child's physical health across several chronic paediatric conditions including chronic kidney disease, those children undergoing kidney or liver transplants and asthma. Poorer parental mental health is associated with measures of the child's physical health including BMI, height, weight, the use of emergency department, parental report of disease morbidity and development and diagnosis of asthma.

Support was found for two separate pathways through which parental mental health can effect a child's physical health; either by reducing the caregivers ability to maintain appropriate adherence to medical regimens (in five studies) or through direct physiological pathways which alter the child's immune system and their response to allergens (in five studies). This second pathway was demonstrated in the asthma literature and shows how parental stress can contribute to development of asthma in children genetically at risk. Within this pathway, a unidirectional relationship was indicated between parental stress and initial asthma development (Wright et al., 2002). Asthma was the only condition which was found to operate through both pathways. This finding supports Kaugars et al. (2004) unidirectional theoretical model of family influences in asthma.

Within other conditions tentative support was found for one pathway through which parental mental health could have an effect a child's physical health; by lowering of parent's motivation or ability to perform medical regimen and health care skills for their child. Poor adherence to medicine and treatment regimens have been shown to be a significant cause of morbidity and

mortality both in transplant patients (Shemesh et al., 2000) and asthma patients (Baumen et al., 2002). Within this review poor parental psychological functioning was shown to have an effect on a child's physical health in kidney and liver transplants patients and asthma patients. Both adherence to medication, clinic attendance and other health monitoring practices were all shown to be significantly associated with the parent's psychological functioning. However two studies did not find a relationship, one with children with diabetes (Chisholm et al., 2007) and the other with asthma (Bartlett et al., 2004), both of which included measures based on parental report either of adherence or the child's morbidity. As most of the studies were retrospective, whether the relationship between parental mental health and child physical health is unidirectional or bidirectional when mediated by adherence to treatment, remains ambiguous. Though one prospective study (Gerson et al., 2004) demonstrated that parental stress was elevated prior to non-adherence to medication, another showed parental stress increased after transplant suggesting that it might be the stress of caring for a chronically ill child that induces stress in the parent (Rodrigue et al., 1997).

The relationship between severity of symptoms in the child, parental report of severity and the parental mental health appears to be a complex one. In this review parental report of morbidity often did not correlate with more objective measures of the child's physical health (Bartlett et al., 2001; Jessop et al., 1988) and therefore studies which relayed solely on its use as a marker of the child's physical health should be interpreted with caution.

Similar to Wallander and Varni's (1994) review findings, there were conflicting reports as to whether mental health difficulties were elevated in parents with children with chronic conditions. For those studies that included the information, some showed significantly higher levels of mental health difficulties when compared to community samples (Diseth et al., 2010; Jessop et al., 1988; Rodrigue et al., 1997) whilst others reported normative levels (Szabo et al., 2009, Wolf et al., 2008, Kovacs et al., 1995). One hypothesis might be that within this population of parents with a chronically ill child are a subgroup of parents with pre-existing mental health vulnerabilities, which

are triggered once the child develops a chronic illness. A study by Spear et al. (2002) would support this hypothesis. They found the level of maternal psychological distress to be independent of the level of child morbidity in parents of neonates admitted to intensive care. Using a neonatal population provides support that it is the parent's own psychological vulnerability as to whether or not they experience stress as a result of a child's illness, not the severity of the condition.

Of the seven studies that failed to find a relationship between parental mental health and a child's physical health, three used hospitalisation as the indicator of the child's physical health status, which has been argued as potentially insensitivity to smaller fluctuations in the child's health (Rodrigue et al., 1997). Two of the other studies were based on maternal report of morbidity or adherence, which as discussed above has not demonstrated itself to be an objective and reliable representation of the child's health status. Future research needs to focus on agreed physical parameters which are not associated with parental subjectivity or insensitivity to smaller health fluctuations. Height, weight, BMI and clinician's reports have all been shown to be useful in these respects.

A potential limitation of the current review is the search strategy employed to identify all relevant studies. As previously acknowledged, few studies span across the range of paediatric conditions, instead tending to focus on single paediatric populations as identified by specific medical presentations. In response to previous criticisms of this medical categorical approach to research within paediatric psychology, (Pless and Pinkerton, cited in Wallander and Varni, 1998) this review aimed to take a non-categorical approach and attempted to review the topic area by presenting evidence from across illness groups and paediatric conditions. However the use of more generalist terms in the search strategy might have risked exclusion of some studies which may only have been identified following search terms that included specific terminology of that paediatric condition.

Though it would not have been feasible within the limitations of this review, in the future a more comprehensive search strategy could be undertaken which utilises the search component of

relevant leading paediatric journals, as well as conducting individual searches using specific illness/paediatric condition terminology. A forward search strategy, whereby articles that have cited those studies which are already identified for inclusion, are also checked for potential relevance, might have served as a way to identify other relevant studies.

Future Research

Other pathways through which parental mental health might operate on a child's physical health need to be considered. Some have proposed that child adjustment may act as another mediating factor between parental mental health and child physical health in these populations (Thompson et al., 1993). In this review Weil et al. (1999) found support for a relationship between child adjustment and the functional impact of their condition, however Wolf et al. (2008) found no evidence that child depression or anxiety mediated the subsequent changes in their immune system. Future research to resolve these conflicting findings would be of value. Similarly, investigation into other variables which might modulate parental mental health difficulties, such as through development of a good quality mother-infant relationship as indicated by Klinnert et al. (1994), is an important direction for further research.

A difficulty when reviewing these studies is the heterogeneity of measures and indicators used for parental mental health, child physical health outcomes and adherence. Firstly, in terms of assessing parental mental health, different studies used measures which assessed different psychological construct. There was some evidence to suggest that different psychological constructs may impact on the child's physical health in slightly different ways. Wolf et al. (2008) showed that parent stress was responsible for elevated levels of two different inflammatory markers in the child, while parent depression was only associated with one. Future research should systematically investigated any association between different psychological constructs such as stress, depression or carer burden, and a child's physical health, choosing appropriate psychometric tools for each construct.

As this review demonstrates an association between parental mental health and child physical health within chronic paediatric populations, there is a need for clinically appropriate tools which identify parents who are experiencing significantly elevated levels of distress. Generic mental health screening measures may underestimate the unique stresses that parents caring for a chronically ill child might experience and so future tools should be designed specifically with this population in mind (Diseth et al., 2010). Similarly, for the reasons previously discussed, there needs to be a consensus within the literature identifying appropriate, valid and sensitive measures of the child's physical health, avoiding indicators such as hospitalisation or solely using maternal report of morbidity. Lastly, if further research regarding adherence to treatment regimens is to be valuable, measures of adherence need to shown to be valid and reliable also.

Considering the sound empirical base for intervention, either psychological or pharmaceutical, within mental health, parental mental health may be one of the few psycho-social variables that is susceptible to intervention. Once these potentially vulnerable families have been identified it would be important to offer support and evidence based interventions to help alleviate the strain that parent is under and allowing them access to resources in various forms, in order to be able to better care for their child, potentially increasing the likelihood of a better medical outcome.

Conclusion

This review has demonstrated a link between parental mental health and a child's physical health across several chronic paediatric conditions, which operates through, for some conditions, two potential pathways; either non-adherence to treatment regimens or through direct physiological alteration of the child's immune system. Future research is needed to further validate this finding within other disease groups, explore other potential pathways through which this relationship might operate and determine variables which may modulate the relationship.

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SECTION B

Assessing Factor Structure, Validity and Reliability of the Paediatric Renal Care Burden Scale (PR-CBS): A Measure of Carer Burden for Carers of Children with Chronic Kidney Disease

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Abstract

Carers of children with Chronic Kidney Disease (CKD) can experience burden and psychological distress, which has been shown to impact on the child's physical health. A 51 item screening tool, the Paediatric Renal Carer Burden Scale (PR-CBS) was developed to assess levels of burden in carers of children with CKD. The current study aimed to assess validity and reliability of the scale. Factor analysis indicated retention of 20 items representing 5 factors; Illness worries (8 items), Impact on self (5 items), Impact on child (3 items), Responsibility (3 items) and Institutional burden (2 items). Together they explain 53% of the total variance. Internal reliability for both the full scale and sub-scales were acceptable. Convergent validity was demonstrated using the Hospital Anxiety and Depression Scale and Caregiver Strain Questionnaire and the scale was reported by respondents as being acceptable to complete. The PR-CBS has been shown to be a valid and reliable scale and as such is a clinically relevant tool with which to identify burdened carers and provide additional psycho-social support so as to ensure best outcomes for both carer and child.

Keywords: Paediatric Chronic Kidney Disease, Carer burden, Factor analysis, Psychometric measure

Introduction

Chronic Kidney Disease

Paediatric chronic kidney disease (CKD) is a life long, progressive condition for which patients are medically managed. There are five stages in the course of the disease. Stage One and Two require observation and monitoring of the kidney function and other associated risks, while Stage Three and Four involve closer monitoring, medication and fluid and dietary restriction. Stage Five, also known as end stage renal failure (ESRF), is when kidney function becomes severely reduced. Progression to ESRF means the child must undergo renal replacement therapy, either haemodialysis, peritoneal dialysis or transplantation. Dialysis patients have the most severe restrictions, both on their fluid and dietary intake and on their time, due to undergoing sometimes daily treatment. Though renal transplantation offers greater rehabilitation back to normal daily routines, it is fraught with medical complications, the uncertainty of rejection, complex immuno-suppresant medication regimens and knowledge that this solution is finite and occasionally short-lived, only lasting a few years for some patients (Mehrabi et al., 2004). Children with CKD also have an increased risk of other health conditions and are particularly vulnerability to cardiovascular disease (Diseth et al., 2011). The prevalence of U.K. children in ESRF was 56.1 per million age related population in 2008 (UK Renal Registry, 2011). The burden of the treatment regimen at ESRF is reported to exceed the other four stages by 50 times (El Nahas & Belo., 2005). Frequent visits to hospital, either for dialysis or due to a change in the child's medical status, are disruptive to the child and family's routine. Though advancements in medical care mean dialysis can be undertaken in the home, thus minimising disruption to routines, this brings with it new burdens such as increased medical responsibility for the carer (Tong et al., 2009)

Burden in Carers of Children with CKD

Since 1980 there has been growing recognition of the unique challenges to families and carers when looking after a child with CKD (Gayomali et al., 2008; Vance et al., 1980). Different treatment

regimens require varying levels of caregiving, but overall the condition is recognised as one which requires complex, continuous and intensive caregiving support from parents (Tong et al., 2009). In interviews with 20 parents of children with CKD, parents felt caring for their child was a pervasive and profoundly negative experience (ibid.). They experienced uncertainty, both in their child's health and in their ability to cope with the caregiving demands, and felt burdened by frequent hospital visits and the technical medical procedures required of them. Tsai et al. (2006) showed that carers of children on peritoneal dialysis have a higher prevalence of depression, and reported poorer quality of life, compared to parents of a healthy control group. They wondered if outcomes of children in ESRF were linked to their carers' mental health.

There are a number of studies which suggest parental mental health does impact on a child's physical health in various paediatric conditions; either through non-adherence to medication regimens (Foulkes et al., 1993; Fredericks et al., 2007; Gerson et al., 2004) or through alterations of the child's physiology and immune functioning (Mzarek et al,. 1999; Wolf et al., 2008; Wright et al. 2002). Several studies have shown a link between a carer's mental health and their child's physical health, specifically in children with CKD.

Watson (1997) demonstrated that increased levels of parental anxiety, depression and stress were associated with higher levels of "Burden of Care" (assessed using a combination of factors including complexity of treatment regimens and socio-economic status) in parents of children undergoing renal replacement therapy. Higher "Burden of Care" was in turn associated with lower growth parameters in the child, both in weight and height. Reduced growth parameters in children with CKD are associated with higher rates of morbidity and mortality (Furth et al., 2002). Maternal psychological distress has also been significantly associated with increased body mass index scores for children with CKD (Diseth et al., 2011). Other studies showed elevated parental stress was associated with non-adherence of medication following paediatric kidney transplantation (Foulkes et al., 1993; Gerson et al., 2004;), an important variable considering non-adherence contributes to 30-70% of graft loss in paediatric renal transplants patients (Gerson et al., 2004).

Despite the acknowledgement of elevated levels of stress and burden in carers of children with CKD, and preliminary findings that these can impact on the physical health outcomes of the child, it has been noted by others that comparatively little has been done in terms of identification and intervention to provide support for carers of children with CKD (Gayomali et al., 2008). It is recognised that carers have varying experiences of caregiving, and give different meanings to their carer roles, a concept that is known as “subjective burden” (Nicholas, 1999). Some carers may have positive associations with the caring role (Nicholas, 1999). However for those who are finding the experience burdensome, psycho-social intervention might be necessary to support best outcomes for both carer and child.

A means to identify these carers would be the initial step. Tsai et al. (2009) called for a measure to screen for psychosocial difficulties in carers of children with CKD, to prevent carer burnout and improve outcomes of children with ESRF. Others highlight the need for screening of parental stress prior to renal transplant (Zelikovsky et al., 2007) considering the implications on adherence post-transplant.

The Need for a CKD Specific Measure

Currently there are few measures designed to assess the impact of the child's illness on carers within paediatric populations. Existing questionnaires focus on posttraumatic stress in the parent (Kazak et al., 1996; Manne et al., 1998), coping (McCubbin et al., 1983) or the impact on the whole family (Stein & Jessop, 1980). Only two measures were identified that aim to assess parental strain due to caring for a child with a physical health condition. The pediatric inventory for parents (PIP) (Steisland et al., 2001) is designed to assess parental stress of caring for a child with an illness, and was validated on parents of children with cancer. To date there has been no factor analysis of this measure to determine the internal structure, though the authors have designed the 42 items to cover four domains of parent experience: 'medical care', 'communication', 'role functioning' and 'emotional functioning'. The parent experience of child illness (PECI) (Bonner et al., 2005) is a 25

item measure of parental adjustment related to caring for a child with a chronic illness and was validated with parents of children with brain tumours. Exploratory factor analysis suggested it contained four factors; 'guilt and worry', 'unresolved sorrow and anger', 'long-term uncertainty' and 'emotional resources'.

Silliman and Sternberg (1988) argued that the caregiving demands related to particular illnesses need to be acknowledged, thus it makes theoretical sense to focus research on single disease groups (Madden et al., 2002). The two measures described above may be able to assess the generic burden placed on a carer of an ill child, but as they were not designed for, or validated on parents of children with CKD, the unique aspects of caring for these children might not be captured (Aldridge, 2008). An important distinction between CKD and other conditions is the chronic, progressive and often fluctuating nature of the condition. Additionally, due to the need for regular renal replacement sessions, complex treatment regimens often in the home, and strict dietary and fluid restrictions, CKD produces unique and burdensome demands of carers. A need exists for a measure that is designed with, and validated with, carers of children with CKD in order to elicit and quantify these stressors.

The Paediatric Renal Carer Burden Scale (PR-CBS) was developed in order to provide quantitative assessment of the level of carer burden a parent of a child with CKD might be currently experiencing. The 51 items on the measure were generated from themes which emerged following thematic analysis of in-depth interviews with parents of children with CKD and healthcare professionals working with these carers and children. These questionnaire items were grouped into nine main themes. Further details of the measure are provided in the methodology section, but for a full description of the initial development and piloting of the PR-CBS please refer to Parham (2011). A copy of the PR-CBS can be seen in Appendix C. Psychometric properties of the PR-CBS are still to be assessed and reported, which is the objective of this study.

Aims and study hypotheses

The aim of this research was to assess the factor structure, reliability and validity of the PR-CBS as a tool to assess carer burden in carers of children with CKD. Specifically this study aimed to:

1. Explore the factor structure of the PR-CBS
2. Reduce the number of items in the PR-CBS current 51 item form, through removal of items which do not load appropriately onto a factor
3. Assess internal consistency of the PR-CBS and that of any factor sub-scales which may emerge
4. Explore the relationship between PR-CBS and an existing carer burden measure
5. Explore the relationship between PR-CBS and an existing measure of psychological distress
6. Assess acceptability of the questionnaire to this population of carers

With respect to the above aims, the following hypotheses were considered:

1. As items of the questionnaire are grouped under nine themes, we would expect nine factors to emerge
2. There will be a significant and positive correlation between the PR-CBS and the other carer burden tool
3. There will be a significant and positive correlation between the PR-CBS and the measure of psychological distress

Method

Participants

In total, 107 participants were recruited. Carers were targeted from across all five stages of CKD, and included all renal replacement modalities; haemodialysis (at home and hospital), peritoneal dialysis and post transplantation. Further information about sample demographics are discussed in the 'Recruitment' section below.

Design

The various aims of this study determined that several quantitative methodologies were required. In order to conduct exploratory factor analysis (EFA) a sample size of at least 100 participants was considered necessary (Ferguson & Cox, 1993). Internal reliability of both the overall measure, and that of factors extracted, was assessed to indicate overall scale and factor internal reliability. Construct validity was determined by assessing correlations between scores on the PR-CBS and other previously validated measures shown to assess either caregiver burden or psychological distress. Acceptability of the PR-CBS was assessed using quantitative content analysis (Krippendorff, 2004) of participant's written responses to three questions designed to elicit whether carers found the scale easy to understand and accessible to use (see Appendix D).

Measures

Paediatric Renal Caregiver Burden Scale (PR-CBS) (Appendix C).

The PR-CBS is a 51 item self report questionnaire designed to assess burden in carers of children with CKD. Carers rate their responses to statements on a 5 point Likert scale ranging from 1 (“never”) to 5 (“always”) reflecting on how they had been feeling over the previous month. A month was chosen as an appropriate time period as typically outpatient appointments are one month apart, which serves as a tangible time point over which carers could review their recent experiences. It was also felt to be an appropriate time-scale in that it allowed enough time to cover the range of experiences the scale encompasses, whilst also being sensitive to the fluctuating nature of CKD and the potential changes in both the child's health status and treatment regimens. The questionnaire items were designed to capture aspects of the experiences of caring for a child with CKD, which arose during semi-structured interviews with 16 carers and ten relevant healthcare professionals, initially providing a pool of 97 items (Parham, 2011). Through a process of consultation and piloting this was then reduced to 51 items (ibid.). These were broadly classified into the following

nine domains:

- physical
- financial
- social
- emotional/psychological
- caregiver role/identity
- impact on family
- impact on child
- CKD treatment responsibilities
- contact with hospital/medical staff

Caregiver Strain Questionnaire (CGSQ) (Brannan et al. 1997; Appendix E).

The CGSQ is a 21 item self report questionnaire which aims to assess strain in carers of a child with a "serious emotional disturbance" over a six month period. Respondents rate a list of statements on a 5 point Likert scale ranging from 1 ("not at all") to 5 ("very much") which includes one reversed item ("How well do you relate to your child"). Total scores are called "Global Strain Scores", with higher scores indicating greater levels of strain due to the impact of caring for their child.

Exploratory and confirmatory factor analysis of the CGSQ determined the measure to contain three factors; objective burden, internalised subjective burden and externalised subjective burden. The overall scale demonstrated high internal consistency (Cronbach's $\alpha = .93$). Construct validity has been also been demonstrated through significant correlations with scores on the Brief Symptom Inventory (Derogatis & Meliseratos, 1983) and The Family Assessment Device (Epstein et al., 1983).

The CGSQ was chosen to assess construct validity of the PR-CBS as a tool to measure carer strain. Though there are other carer burden measures, the vast majority of these are developed for,

and validated with, carers of individuals with dementia. The CGSQ is different in that it aims to assess strain in a parent looking after a child with additional needs. Though it is acknowledged that it was developed and validated in a population caring for children with emotional and behavioural difficulties, it was felt the majority of items examined constructs of burden and strain that the PR-CBS was also designed to measure. It has demonstrated robust psychometric properties (Brannan et al., 1997) and has previously been used to validate a questionnaire based on a sample of carers of children with brain tumours, the PECI (Bonner et al., 2006) demonstrating its appropriateness for use with paediatric populations. As such it was felt to be an appropriate tool with which to validate the PR-CBS. For the purposes of this study, two items were removed from the CSGQ which appeared to not be applicable to participants of this study, as carers of a child with a chronic illness. These were item 6 (i.e. " Your child getting into trouble with the neighbours, the community or law enforcement?") and item 19 (i.e."How resentful did you feel towards your child?"). Though removal of items might affect the internal consistency of the scale, it was felt that the alienation of participants by potentially irrelevant questions outweighed this concern. This has been done in previous studies, for example when there are concerns that items on a questionnaire might confound results when used with specific populations (Weil et al., 1999).

Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983).

The HADS is a nine item self report scale used as a mental health screening tool to measure psychological distress as reported over a one week time period. It generates a total score which is a measure of general psychological distress (Crawford et al., 2001) and two separate scores from sub-scales measuring anxiety and depression. The HADS was chosen as a measure against which to assess the PR-CBS' relationship to psychological distress, as it has been shown to have psychometrically sound properties and has been used extensively with both clinical and non-clinical samples (Crawford et al., 2001). Previously it has been used to assess caregiver burden in other populations (Grunfield et al., 2004) and as a tool to validate questionnaires measuring distress due

to a physical illness (Mitchell et al., 2010).

Recruitment

Recruitment of carers took place across two national specialist sites in the U.K.. Initially clinicians from each site were asked to distribute the questionnaires to carers when they attended their clinic appointments. This resulted in a response rate of 6% (six out of 100) and was also felt to be susceptible to recruitment bias as clinicians may have been prompted to remember the questionnaire when faced in clinic with those carers who appeared more strained or stressed. A second recruitment method was trialled, whereby the chief investigator attended clinics at the two sites, approaching carers in the waiting room to ask if they would be willing to participate. Clinics were attended on different days in order to capture a sample across the range of CKD phases and treatment modalities which included hospital based haemodialysis, peritoneal dialysis, home haemodialysis and transplant patients and those in Stages one-three. Data collection using this method spanned over a time period of three months and resulted in a 101 questionnaires being completed. Carers were excluded from participating in the study if they were not able to fill in the form due to being unable to understand English, unless there was a friend or family member willing to help them with the translation. Participants had to be the primary carer, which was self defined by the participant. In families where responsibility was jointly shared, parents filled in the form together or chose to do one each (four responses). In total, 107 carers provided data for the study with 62 recruited from site A and 45 from site B.

Procedure

Questionnaire packs were produced containing an information sheet about the study (Appendix F), a consent form (Appendix G), the PR-CBS, CGSQ, HADS and the acceptability questions. Participants had the choice to fill the form in during their wait for the appointment or to take the

forms home and return via post. An overwhelming number or respondents choose the former (94 versus seven).

Ethical considerations

Full ethics approval for the study was granted by the Research Ethics Committee (see Appendix H) along with local research governance approval (Appendix I) for the two data collection sites.

Participants were given the option to fill out the questionnaires anonymously. This was in response to evidence suggesting that some caregivers might find it difficult to acknowledge they are struggling to cope. This is postulated to be particularly problematic for women, due to social and ideological constraints which assume a woman's role as a competent caregiver (Nicholas, 1999). Such constraints might impact on the way in which some mothers would fully engage with the questionnaire or feel able to accurately self report on questions that admit feeling burdened. Some of the questionnaire items also asked carers to disclose potentially difficult feelings about their carer role, frustrations with the medical process or their ability to manage treatment regimens. In an attempt to address this it was felt the option of complete anonymity was appropriate in order to elicit as honest a response as possible. In addition, to reassure participants of anonymity, further demographic information and medical status of the child was not obtained, as this could have led to identification of the family. As the aim of the study was to initially validate the PR-CBS as a tool able to assess strain across the whole of the CKD spectrum, information about the medical status of the child was not considered necessary. Others have acknowledged the importance of gaining a comprehensive understanding of carers experiences across the spectrum of CKD (Tong et al., 2009). During data collection two participants were concerned that their responses might be shared with the team, or information given to social services, should they acknowledge they felt under strain by their child's condition. If participants did become distressed during completion of the questionnaire they had the option to discontinue, which two participants chose to do. When this occurred they

were encouraged to share their feelings or concerns with their nephrology clinician or, if more appropriate, their G.P.

Analyses and Results

Data screening and descriptive statistics

PR-CBS.

107 participants completed the PR-CBS. Data were analysed using PASW version 17.0 (SPSS Inc., 2009). Means and standard deviations were calculated for each item and histograms were scanned, which showed a variety of responses on each item.

Univariate distribution of each item, and the 51 item scale score, was assessed and all were found to be normally distributed as demonstrated by tests for skewness and kurtosis, which generated scores within an acceptable range (skewness = +/- 2; kurtosis = +/- 5) (Kendall & Stuart, 1958). The range of scores on the 51 item PR-CBS was between 71 to 251, out of a potential maximum score of 255. Multivariate normality was assessed through calculation of Mahalanobis distances, none of which exceeded the critical value (87.97, df=51, p=0.001). A summary of the descriptive statistics for the PR-CBS can be seen in Appendix J.

Though it is not strictly necessary for assumptions of either univariate or multivariate normality of distribution to be fulfilled when undertaking factor analysis (Floyd & Widaman, 1995) meeting the assumptions of normality does enhance any factor solution that might be generated (Tabachnick & Fidell, 1996).

Two participants completed only the first page of the PR-CBS. An assumption was made that this was because they did not see the other side of the questionnaire. Analysis of the data both with and without these participants did not change the overall factor solution and so the decision was made to retain their data for items they had provided data for, but exclude it for any analysis which used the full scale score. An expectation maximisation (EM) analysis was used to assess pattern of all missing data. As all EM values generated were non-significant, missing data was

considered to be random and removed from the analysis of that particular item.

Other measures.

The mean and standard deviations for the other psychometric measures can also be seen in Appendix J. Again, using tests for skewness and kurtosis, all were within the limits to meet assumptions of normality. Of note was that carers' reported higher scores on both the HADS depression and anxiety subscales as well as the total score, when compared with population norms (**anxiety scale**: CKD carers mean = 8.87, population norm = 6.14; **depression scale**: CKD carers mean = 6.35, population norm = 3.68; **total HADS**: CKD carers mean = 15.09, population norm = 9.82) (Crawford et al., 2001). Additionally, CKD carers mean score for the anxiety sub-scale fell within the clinical range consider “mild anxiety” (Crawford et al., 2001), indicating that many carers of children with CKD experience levels of anxiety that would be considered clinically significant.

Exploratory Factor Analysis (EFA)

Factor analysis is a data reduction technique which aims to identify latent factors underlying a set of variables. The data for all 107 participants who completed the PR-CBS was subjected to preliminary analysis in order to assess the appropriateness of undertaking EFA. As well as exceeding the sample size recommended by Ferguson and Cox (1993), the results of Kaiser-Meyer-Olkin test, a measure of sampling adequacy, was considered “very good” at .864 (Field, 2005). Bartlett's test of sphericity, which is used as an indication of “factorability” of items when there are less than 5 participants per item, also produced a significant result (Chi-Square = 3734.180, df = 1275). The correlation coefficient matrix of all 51 items was generated and visually screened, which showed no two items correlating either greater than 0.9 or perfectly with any other item, indicating no concerns regarding multicollinearity or singularity of items (Field, 2005). Instead the matrix showed various groupings of moderate correlations between items.

Based on these findings it was considered viable to continue with EFA of the data set. The results of the EFA will be reported using the criterion detailed in Floyd & Widaman (1995) (see Appendix K).

Initial analyses using both principal components analysis (PCA) and principal axis factoring showed no significant difference between solutions. Therefore principal axis factoring was chosen as the favoured extraction method as it has been strongly recommended to be used over PCA as it tends to generalise better to those results obtained in confirmatory factor analysis and gives greater accuracy of estimations of factor loadings and correlations (Floyd & Widaman, 1995).

Initial communality estimates were between 0.64-0.90 for the 51 items. The closer commonalities are to one, the better the factors extracted explain the original data (Field, 2005). Unconstrained principal axis factoring indicated eleven eigenvalues greater than one and a scree plot which showed one large factor which then sloped down without a clear point of inflexion. The 11 factor solution was examined for items with a loading of greater than 0.55, a criterion recommended by Hair (1998) when using samples that are approximately 100. This solution could not be interpreted as item loadings were small; three of the 11 factors had only one, or no items which loaded greater than 0.55. Such low loadings of items suggest factors that are poorly defined and hazardous to interpret (Tabachnick & Fidell, 1996).

Next an analysis was run forcing extraction of nine factors, as this was the number of domains initially indicated during questionnaire development. Again, item loadings onto factors were generally low, with two factors having no items loading greater than 0.55. Thus this solution too could not be meaningfully interpreted.

Further factor analyses were run forcing extraction of six, five and four factors, as closer inspection of the scree plot indicated a slight inflexion between the fifth and sixth factor. The five factor solution was eventually retained as this was the most interpretable in that several items loaded highly on each of the factors, the factors made distinct theoretical sense and it accounted for 57.6% of the variance which is considered reasonable (Streiner, 1994). Initial rotation of the five

factor solution was done using oblique rotation (direct oblimin) as it was considered that the underlying constructs represented by factors might be related to each other. However oblique rotation initially failed to converge on 25 iterations, the default iterations on the statistical package used. Eventually the solution did converge on 37 iterations, but as the correlations between factors were generally low (2.07- 5.33), the decision was made to examine the solution produced by orthogonal rotation (varimax). This produced a solution which converged in nine iterations, explained the same amount of variance as the oblique rotation and generally allowed the same items to load onto the same factors. The only difference between the orthogonal and oblique rotated solutions was that the former allowed for two more items to load onto factor four and a different item to load onto factor one. Additionally, orthogonal rotation has been described as producing solutions with greater generalisability and which allow for ease of interpreting and reporting (Tabachnick & Fidell, 1996). Due to these considerations, and to allow for retention of factor four, the solution chosen to interpret was the five factor model with orthogonal rotation, which converged in nine iterations and explained 53% of the total variance. Table 1 shows the resulting solution with item loadings greater than 0.3. The complete solution, with all loadings can be seen in Appendix L.

From this solution items were retained if their loading on a factor was greater than 0.55. When an item cross loaded onto another factor, it was retained only when there was a difference of at least 0.2 in magnitude between the two loadings. The use of this criterion resulted in the retention of 20 items. Descriptive statistics, including the range and mean, for the 20 item PR-CBS can be seen in Appendix J. Of the other 31 items from the original PR-CBS, 28 failed to load on any factor above 0.55 and three did load on a factor greater than 0.55, but also cross loaded highly onto another.

Table 1

Summary of Exploratory Factor Analysis results for the PR-CBS using Principal Axis Factoring using Varimax Rotation with Kaiser Normalisation (n= 107)

Item	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
Difficult feelings due to the uncertainty of my child's condition	**.699**				
Worrying about my child getting ill of dying	**.665**				
Feeling helpless when my child is ill or in pain	**.639**				
Feeling unable to "switch off" when waiting for test results or a phone call from the hospital	**.626**		.333		
Worrying that my child may be admitted to hospital	**.607**				
Feeling overwhelmed by decisions that I have to make about my child's condition.	**.597**	.311		.332	
Worrying about my child during the night	.564	.397			
Worrying about having to deal with unexpected changes in my child's condition (e.g. unexpected hospital admissions)	**.559**		.339		.314

Feeling troubled by difficult memories of when my child was first diagnosed or has been very ill in the past.	**.552**			
Feeling preoccupied with keeping my child safe from illness	.537		.455	
Feeling that I am not able to "switch off to my child's condition	.521	.433		
Worrying about getting medical procedures wrong (e.g. dialysis, injections, tube feeding) or taking measurements incorrectly	.495			.352
Feeling unable to think about my own needs	.492	.441	.396	
Feeling preoccupied with checking my child for signs of illness	.489			
Feeling guilty about spending less time with my child/partner	.465		.410	
Feeling under pressure to be strong for my child and family	.459	.364	.371	
Worrying about the future	.451			
Worrying is my child has had the correct amount of fluid	.424			.423
Worrying about the impact of my child's condition on my other children	.400		.389	

Worrying about getting my child's medicines wrong	.388		.300	.386	
Sadness about not socialising as much as I want because of caring for my child.		**.712**	.386		
Feeling trapped because of caring for my child		**.695**			
Sadness that I can not do the things I used to do because of caring for my child (e.g. work, leisure activities, hobbies)		**.650**	.366		
Feeling that my child's condition has taken over my life	.449	.585	.357		
Worrying about the effect of caring for my child on my own health		**.575**			
Sadness from feeling I am not the person I used to be		.561	.384		
Feeling alone in caring for my child		.535	.453		
Worrying about money due to the costs of my child's care		.508		.406	
Feeling that other people do not understand my situation	.423	.506			
Sadness that I do not have a “normal” relationship with my child		.448	.338	.308	
Difficult feelings due to having no privacy when at the hospital		.417			.363

Sadness about the impact of my child's kidney problems on my relationship with my partner		.392	.386		.372
Arguing with my partner/family about my child's care		.385	.304		.322
Worrying about my child's growth and development			**.644**		
Sadness about the things my child misses out on	.349		**.611**		
Worrying about disruptions to my child's education			**.582**		
Worrying about how my child is coping	.318	.376	.531	.301	
Feeling that I should be doing more for my child			.528	.470	
Feeling overwhelmed by changes in my child's usual treatment	.396		.517	.324	
Feeling overwhelmed by trying to fit family life around my child's condition	.440	.498	.505		.348
Feeling overwhelmed by feeding difficulties (e.g. lack of appetite, managing diet restrictions, vomiting)		.317	.392		
Worrying that I have not understood medical information				**.705**	

Blaming myself if my child gets ill or has bad test results	.304		**.560**	
Feeling uncertain about how to manage my child's emotions and difficult behaviour			**.556**	
Difficult feelings due to my child taking responsibility in their care (e.g. worrying if medicines have been taken)			.475	
Holding back when I disagree with medical staff			.461	.349
Blaming myself for my child's kidney problems			.375	
Feeling frustrated when having to spend time at hospital				**.731**
Feeling bored when having to spend time at the hospital				**.729**
Feeling exhausted in caring for my child	.386	.407		.470
Frustration when dealing with staff who do not know my child				.443

Loadings in bold represent items to be included in each factor

The resulting 20 item scale contained five factors and accounted for 53% of the total variance. The first factor accounted for 15.6% of the variance and was named 'illness worries' (eight items). This factor captures worries and difficult feelings relating specifically to the unique experience of caring for a child with a chronic, life limiting condition and includes the uncertainty of the illness trajectory. The second factor accounted for 12.6% of the variance and was named 'impact on self' (four items). This factor captures the affect of the caring role on the carers own health, both physical and emotional, and other domains of their life. The third factor accounted for 10.8% of the variance and was named 'impact on child' (three items). This factor captures the carers concerns regarding the affect of the child's illness on areas of the child's life, other than their physical health. The fourth factor accounted for 7.6% of the variance and was named 'responsibility' (three items) which captures the carers sense of worry in being responsible in managing both the child's medical regimens and the child's emotions and difficult behaviours. The fifth factor accounted for 6.4% of the variance and was named 'institutional burden' (two items). This factors captures the unique experience of the institutional process and burden of frequent time spent in hospital, both as outpatients and inpatients, that accompanies CKD due to the fluctuating nature and chronicity of the condition.

Internal Reliability

Cronbach's α coefficients were calculated for both the full scale 20 item PR-CBS, and for each factor, in order to assess internal consistency and reliability. The full scale PR-CBS demonstrated good internal consistency ($\alpha = .93$) indicating that all items were measuring the same constructs. No item indicated that it should be deleted.

Internal consistency of the five factors showed them to be within the acceptable criteria of reliability, with each having a Cronbach's α greater than 0.7 (see Table 2) (Kline, 1999 cited in Field, 2005). Removal of any item would not have greatly increased the overall Cronbach's α for

any factor, so all items were deemed acceptable to retain.

Table 2

Internal consistency for each factor as demonstrated by Cronbach's α values

	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
Cronbach's α	0.92	0.84	0.80	0.70	0.78

Assessing Convergent Validity

The relationship between scores on the 20 item PR-CBS and on the CGSQ and HADS was assessed. Pearson product moment correlations were chosen as the test to assess correlations between the measures, as the data had been shown to be normally distributed and all the measures used Likert scales, which are considered to be robust when used in parametric analysis. It was hypothesised that higher scores on the PR-CBS would be related to higher scores on both the CGSQ and HADS, so one-tailed tests were chosen.

Carers' scores on the PR-CBS and CGSQ were positively and significantly correlated (see Table 3) indicating that the theoretical constructs underlying the PR-CBS are related to those measured on the CGSQ; specifically carer strain. The effect size of this correlation was large (Cohen, 1988).

Correlations were also calculated between carers' total scores on the PR-CBS and their score on the HADS, both on the anxiety and depression sub-scales, as well as total score. Again it was hypothesised that higher scores on the PR-CBS would be related to higher scores on all the HADS scales. All correlations were positive and significant (see Table 3), again demonstrating large effect sizes, indicating that the underlying theoretical constructs measured by the PR-CBS are also related to the theoretical constructs of anxiety, depression and general psychological distress.

Table 3

Correlations between the PR-CBS and other psychometric measures

	CGSQ	HADS total	HADS Anxiety	HADS Depression
Correlation with PR-CBS Total Score	0.81*	0.71*	0.70*	0.65*

*Correlations significant at the 0.01 level

Acceptability of PR-CBS to Carers

The acceptability questions were completed by 93 participants. As can be seen in Appendix C this consisted of three questions.

The first question assessed the time taken to complete the questionnaire. The range of time participants reported was between 2-30 minutes, with the mode response for time to complete the questionnaire being ten minutes. It should be noted that these were the times people reported it took them to complete the 51 item version of the PR-CBS; it can be assumed that the reduced 20 item PR-CBS will take even less time.

The results for question two “Did you find the questionnaire alright to answer?” was analysed using qualitative content analysis (Krippendorf, 2004). This methodology was chosen as it has been shown to be a rigorous and replicable method of interpreting written text (Stemler, 2001). After preliminary examination of the data, five emergent category codes were established which were considered to be exhaustive. These were: 'no problems', 'current relevance', 'sadness', 'reflective experience' and 'answer options vague'.

The complete coding frame can be seen in Appendix M. The data were also analysed by a second member of the research team. Inter-rater reliability was determined using Cohen's kappa which demonstrated an “almost perfect” strength of agreement (free-marginal Cohen's kappa = 0.95) (Landis & Koch, 1977). An overwhelming majority of participants (91%) did not identify any

problems with the questionnaire. A small percentage (4-5%) of carers reported that some questions were not relevant at that particular time point; with some saying the answers would have been very different if their child was currently not so medically stable. Two participants (2%) commented on feeling sad whilst completing the questionnaire. One person found the answer options (e.g. “never”, “always”) vague.

In the final question “Were there any questions you didn't understand?”, 85 participants said they were able to understand all questions. Eight participants stated there were either some questions they did not understand, or they had to re-read some questions. However only three of these participants went on to give details of which particular question they were referring to. As none of the questions identified were to be retained in the final 20 item PR-CBS, it was not considered necessary to remove or alter the wording on any of the PR-CBS items.

In order for questionnaires to be of use in busy clinical environments it is important that the measure is brief and accessible to the respondent. The PR-CBS has demonstrated that it is brief, understandable and acceptable to carers which meets the criterion laid out for acceptability and feasibility of questionnaires (Fitzpatrick et al., 1998).

Discussion

Validity and Reliability of the PR-CBS

This study has shown the PR-CBS to be a valid and reliable tool with which to assess carer burden in parents of children with CKD. EFA of the PR-CBS supported the retention of 20 of the original 51 items, and suggested an internal structure of five factors, which together explained 53% of the variance. The full 20 item scale demonstrated very good internal reliability. The five factors, illness worries, impact on self, impact on child, responsibility and institutional burden, each represent unique theoretical aspects of the burden of caring for a child with CKD and have acceptable internal reliability. The reduction of the measure from 51 to 20 items through EFA was an important part of the measure development process. It strengthens the validity of the tool through retention only of

items that are statistically and theoretically highly correlated, whilst also creating a shorter measure which is clinically more useful. Further evidence of the validity of the PR-CBS was demonstrated by strong associations with other measures of carer burden, psychological distress, anxiety and depression. All are theoretical concepts with which carer burden has been associated with (Grunfield et al., 2004; Watson, 1997). The acceptability of questionnaires to respondents is an important part of the validation of a measure to be used in routine clinical use, but appears to often not be included in other measure validation studies (Fitzpatrick et al., 1998). The PR-CBS was shown to be a brief (less than ten minutes) and acceptable questionnaire to carers, with no question retained in the final PR-CBS found to be difficult to understand.

Several of the factors in the PR-CBS have conceptual similarities to those in both the PIP and PECI. Both the 'responsibility' factor on the PR-CBS and the 'medical care' domain on the PIP address issues of making decisions about the child's medical care. Some aspects of the PIP domain 'role functioning' also seemed congruent to the 'impact on self' factor in the PR-CBS, assessing impact on areas of the carer's life. Similarly, the PECI factor 'guilt and worry' appears theoretically similar to the 'illness worries' factor of the PR-CBS, although the PECI also contains 'guilt' items such as "feeling responsible for the child's illness". The PECI 'long-term uncertainty' factor also shares similarities with the 'impact on child' factor of the PR-CBS in that some items make reference to what the child misses out on because of the illness.

The similarity between aspects of the factors in the PIP and PECI with those in the PR-CBS suggests the PR-CBS is also identifying similar underlying constructs related to carer burden when caring for a child with a chronic illness. However a unique feature of the PR-CBS are those items within factors ("worrying about having to deal with unexpected changes in my child's condition e.g. unexpected hospital admissions", "Worrying I have not understood medical information") and whole factors (e.g. 'institutional burden') which capture the different experience of CKD care. The items contained in the PR-CBS are deemed to represent the burdensome and complex demands of caring for a child with a CKD: a chronic and progressive condition with requires adherence to

complex treatment regimens which are recognised as particularly burdensome on caregivers (Aldridge, 2008).

Items on the PR-CBS have been generated through interviews with the target population of carers of children with CKD and the process of assessing validity and reliability again has targeted this specific population. Silliman and Sternberg (1988) have argued that individual caregiver demands related to particular illnesses need to be acknowledged and assessed. The PR-CBS was designed to address this need for carers of children with CKD. As far as the author is are aware this is the only disease specific carer burden scale for a paediatric population, whom both the initial questionnaire development and the validation of the measure have both been conducted with. This study design has allowed for incorporation of carers own experiences and accounts of caring for a child with CKD, and their perspective on the actual measure, throughout development of the PR-CBS.

In this study carers reported levels of depression and anxiety that were elevated when compared with normative samples. Similar findings have been reported in previous studies with paediatric CKD caregivers (Diseth et al., 2011; Tsai et al., 2006). This is of particular clinical relevance as elevated levels of parental depression and anxiety have been shown to negatively correlate with outcomes of child physical health in paediatric populations (Diseth et al., 2011; Foulkes et al., 1993; Gerson et al., 2004; Watson et al., 1997).

Limitations of the Study

The difficulties with return rates, and potential sampling bias when part of the recruitment method involves postal return of questionnaires, is well recorded (Bruce & Chambers, 2002). For those participants who did chose to take the questionnaire home and did not return it, this could be indicative of feeling either under very little strain, making the questionnaire seem irrelevant to them, or under so much strain that filling in the forms would be too time consuming or too

distressing.

A concerted effort was made to access carers from across the various stages of CKD, thus accessing carers at various times in their child's illness course, as well as time from diagnosis and treatment modality. However, as no demographic or clinical variables were recorded it is acknowledged there is a possibility the sample might be skewed in directions the research team would be unaware of.

It was of interest that the sixth item on the original PR-CBS “blaming myself for my child's kidney problems” did not load highly, and therefore was not retained in the final version. Difficulties with caregiver guilt was felt by clinical members of the research team to be of particular relevance for some parents, particularly as there are strong genetic links to certain conditions within CKD. Additionally it was a theme reported by the PECI who retained the item “feeling as though I am in some way responsible for my child's illness”. It is medically acknowledged that certain types of genetically linked CKD conditions have a much higher prevalence in Indo-Asian families (Buck & Feehally, 1997), some of whom might have been potentially under-represented in our sample due to language constraints, leading to a sampling bias which potentially prevented the item from loading higher.

Clinical Implications

The PR-CBS has demonstrated itself to be valid, clinically relevant and acceptable screening measure of caregiver burden of carers of children with CKD. This CKD specific tool can be used as part of the routine clinical evaluation or as part of a more intensive psychological assessment, such as those undertaken prior to transplantation. This self report measure is a way for nephrology clinicians and health psychologists to quickly and easily be alerted to those carers who are experiencing higher levels of burden, something which often might be difficult to elicit or prioritise in busy medical environments.

It should not be assumed that all carers experience feelings of burden and strain when caring for a child with CKD. Research has shown that carers, which in most cases are the mother, have different meanings and experiences of caregiving, many of which are a positive and fulfilling (Nicholas, 1999). Families do adapt to caring for a CKD child (Vance et al., 1980) and we can see from some of the responses to the PR-CBS that parents recognise they feel more burdened and overwhelmed at certain points in their child's illness journey. The results of the present study support that the PR-CBS is a reliable and valid way to screen for levels of burden, and identify a carer quickly, who may be feeling overwhelmed. Often medical staff have neither the time or the psychological knowledge to reliably identify which carer is struggling, as signs may be different for each carer. The PR-CBS can potentially resolve this issue, identifying those who might benefit from a psycho-social intervention in order that the parent feels able to continue caring for the child and the child's own physical health is not jeopardised by an overwhelmed and burdened parent.

The identification of several distinct factors relating to carer burden in children with CKD may have important implications for psycho-social interventions or treatment planning. For example, parental anxiety and depression have been shown to be associated with dissatisfaction with medical care received (Fielding & Brownbridge, 1999). The 'institutional burden' factor on the PR-CBS might allow monitoring of levels of burden specifically related to time spent in hospital, and where both possible and appropriate, support a family with more community based care.

Future Research Directions

There are several key areas of research that need to be undertaken in order to strengthen the validity of the PR-CBS. Predictive validity of the tool should be assessed, potentially through the monitoring of referrals for additional psycho-social support for families, and determining whether an association with higher scores on the PR-CBS is present. Confirmatory factor analysis, with a larger sample size would hopefully reassert the five factor model. Translation of the PR-CBS into other languages, particularly those spoken in communities where there is a higher prevalence of

CKD, and subsequent validation, would allow access to a larger sample of carers who may be experiencing high levels of undetected burden. Finally, the PR-CBS has the potential to discern differences in burden levels between CKD stages, treatment modalities and different age groups of children, all of which would be theoretical and empirically relevant to service planning and provision, and allow for targeted psycho-social support for this carer population.

Conclusion

The results of this study suggest the PR-CBS to be a valid and reliable measure of carer burden in parents of children with CKD. In its final form the PR-CBS is a 20 item self report measure containing five factors representing aspects of particular importance for consideration in caregiver burden in CKD caregiving, and which carers have found acceptable to complete. As such it provides value in helping clinicians identify those carers who are experiencing levels of burden which might be impacting on their ability to care for their child.

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SECTION C:

Critical evaluation of the research process

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What research skills have you learned, what research abilities have you developed from undertaking this project and what do you think you need to learn further?

Factor analysis is a complex procedure (Floyd & Widaman, 1995) which requires both knowledgeable decision making, such as how many factors to retain and method of rotation, along with interpretation of the extracted factors. Originally I had planned to undertake the analysis alone, believing I would be able to understand the complexities of the methodology by reading enough of the literature. However it soon became clear that in order to feel confident and truly understand this type of analysis in order to make informed choices regarding the procedure, I would need some help and so I decided to seek consultation with a statistician. Through this process I learnt that it is just as important to know when to seek expert advice and guidance from others who are more knowledgeable and experienced than youself, and I experienced the benefits that these expertises brought to the analysis. I now have confidence in my ability to undertake this type of research in the future.

This study involved working with a large research team consisting of several senior academic and clinical staff from different institutions, along with another trainee. Working as part of such a big team when conducting research is not something I have had previous experience of. Prior to clinical training, my brief time spent as a research assistant involved me conducting literature reviews for a review article, and working within a team of just three. When working in a large team it can be easy to be swayed by the many different aims for the research and opinions on how things should be done, all valuable and important contributions in their own way. However at times, due to the team's mixture of training backgrounds and clinical perspectives, it felt that the objectives of the research were being pulled in various directions. After a few months I learnt, as the chief investigator, it was important to maintain ownership of the direction of the work and make sure that any suggestion of changes to the project design fit with the overall research objectives, and with what I needed for a thesis project. This was hard, especially as I was the most junior member of the team in terms of both qualifications and experience, but this was an ability I worked on developing

throughout the course of the study.

Holding the tension between being a researcher and at other times in the week, a clinician, was difficult. As the main researcher collecting data, I was often approaching families who were under considerable burden, sitting for long periods in hospital waiting rooms. For some at least, answering the questionnaires did bring to the surface some of the more difficult experiences and strain of caring for their child and when they returned the questionnaire packs to me, they would often share some of their frustrations or worries. To not automatically respond to these disclosures in a therapeutic manner, as I would during my clinical work, was trying, but at the same time the ethical boundaries of the situation dictated that I was there as a researcher and these carers had signed a consent form to consent to research, not a therapeutic encounter. Being able to reflect on and balance the boundaries of this contact was difficult at times, especially considering, unlike in clinical work, supervision is not scheduled weekly to discuss such issues. In future research I would aim to organise more regular research meetings so these dynamics can be thought about and shared.

An ethical dilemma arose during data collection when a parent asked if the information gathered in the PR-CBS would be shared with social services. Perhaps naively, this was not something that had not been considered by the team prior to the study commencing or queried by the NHS ethics panel. We had a protocol in place if parents became upset or overwhelmed following completion of the questionnaire but the idea that the tool could be used to indicate a potential safeguarding risk to the child was not something we had considered. Through discussion with the parent I was able to determine that there were no significant risks, she was just concerned that endorsing items that reflect her feelings of burden would be used to indicate she was not a competent caregiver. She was happy to complete the measures, but stated to me that she would not have her name attached to it. This confirmed to me that we had made the right decision making the questionnaires anonymous, as otherwise we risked excluding those carers who were potentially most burdened. Bringing this incident back for discussion with my research supervisors meant it was thought about with the two clinical members of the research team. We realised there needed to

be further thought and agreement into how any information gathered by the tool is shared, both within clinical teams and with other agencies when risk is indicated. This will be something for the clinical teams who use the measure to decide themselves based on local trust and safeguarding policies. However this encounter highlighted to me how despite best intentions of a thorough protocol, revised by numerous professionals and an external and independent ethics committee, ethical issues and considerations can continue to rise during the project and need to be sensitively and responsibly responded to.

If you were able to do this project again, what would you do differently and why?

One of the limitations of this study is that we were unable to report on demographic information for participants, or the medical status of their child. Originally the focus, both from the research team and the ethics committee, was on making sure that participants were able to respond completely anonymously if they chose to. This was so that parents, and mothers in particular, would feel able to respond to questions openly and honestly. Research has shown that when asking people personal information about their psychological state, anonymity is important in reducing the level of dishonest responses given (Levine et al., 2003). The issue of being a competent caregiver, particularly as a mother, is a constraint which has been shown to impact on this population of CKD carers (Nicholas, 1999). However, the decision to not ask participants to disclose any demographic information about themselves could be viewed as a limitation as it left the study susceptible to an unknown sampling bias. In future I would look for ways to carefully maintain anonymity and participants confidence in that, whilst also getting basic demographics about the carer such as gender and age, and potentially information about the child's medical status. One idea might be to hold a focus group with parents to ascertain their views on whether they would respond honestly if they were able to be identified or what identifying information they would feel comfortable with sharing.

Originally part of the study design had been to assess predictive validity of the PR-CBS by

comparing scores on the measure against those who then went on to be referred for additional psycho-social help. However after liaising with both site's psycho-social team I recognised that there appeared to be no clear way in which these types of referrals were recorded. Though some referrals were recorded more formally on the notes, the teams acknowledged that more often referrals consisted of "corridor chats" between professionals. Thus I recognised that quite a few of the psycho-social team interventions, which might be not more than one off consultation on the ward or with the staff team would not be able to be included, despite being important interventions and indicative of a family or carer who was struggling. In order to gain a more accurate measure of predictive validity there needed to be more time and thought given to how best to capture any psycho-social input. This is something I would have liked to pursue, but given the time constraints it was not possible within this thesis project.

With the prestige of being involved in research with, and collecting data across, two large national specialist prestigious sites also came with difficulties. Both sites were very busy with staff involved in numerous clinics and other duties. As specialist sites they were both heavily involved in other research and the population I was recruiting were often also being asked to participate in other research being lead by members from within the teams. Being a researcher who is out-with the team can make accessing participants difficult. You need team members to give up their time to introduce you to patients and carers and this means making time to build relationship with various key team members. This took me a while to recognise, and had I realised I would have begun building those relationships long before I started trying to collect data, which would potentially have speeded up the process that was somewhat slow at the beginning.

Clinically, as a consequence of doing this study, would you do anything differently and why?

As psychologists we are trained to understand the impact of parental mental state on an infant's mental health, how this affects their ability to regulate their emotions (Gergely & Watson, 1996) and shapes their future relationships (Bowlby, 1969). Though I was interested in the possibility that

parental mental health could also affect the child's physical health, before beginning to explore this topic area for section A, I was genuinely unsure of whether this relationship existed. Reviewing the literature for this study has really illustrated the powerful affects a parent's mental state can have on a child, even more so than before from teaching on clinical training where the focus tends to be on the child's mental health. Post qualifying I hope to continue working in the area of child and adolescent mental health and I will certainly take this knowledge with me when assessing a child and family, and being mindful of the fact that poor parental mental health can impact on both a child's mental and physical well-being.

If you were to undertake further research in this area what would that research project seek to answer and how would you go about doing it?

In the future I would be keen to assess whether the PR-CBS has predictive validity. This would require liaising closely with the nephrology and psycho-social teams at both sites and agreeing with them a way to formalise recording of all contact with the psycho-social team, as well as referrals to G.P. or other agencies for additional psycho-social support for that carer and family. We would then be able to cross-reference this with those carers who did feel able to give their names on the questionnaire, seeing if there is a correlation between scores on the PR-CBS and those who go on to be in need of psycho-social support.

Following on from the literature review I conducted in section A I would be interested in further adding to the emerging evidence base demonstrating a relationship between parental mental health and a child's physical health, within different paediatric populations. It would be particularly interesting to do this within conditions where currently there is little or no research, but the condition has a known genetic predisposition, such as sickle cell disease. Longitudinal prospective studies such as those already under taken within the paediatric asthma population would be able shed further light on the potential pathways through which this relationship might operate. Additionally I would be interested to further investigate potential modulating variables, such as

quality of parenting and the mother-infant relationship. I acknowledge that both of these ambitious areas of research would need a dedicated research team and plenty of funding in order to be viable.

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Section D: Appendix of Supporting Material

Appendix A: Details of the literature search strategy

Databases searched: PsychINFO (1806 to May week 1 2012), MEDLINE (1948 to April week 4 2012), Maternity and Infant Care (1971 to April 2012).

The search was conducted using a combination of the following terms:

Carer* or parent* or maternal AND

Stress or burden or mental health or distress AND

Chronic child* illness or paediatric illness or paediatric condition* or pediatric illness or pediatric condition* or chronic child* condition*

An additional search was also conducted on the search engine "Google Scholar".

The computerised search generated 756 results, the abstracts of which were briefly reviewed and the articles which discussed the investigation of parental mental health as a variable which contributes to child physical health within a chronic paediatric group were retained. This amounted to 41 articles. Duplicates were removed which left 37 articles. Articles were retained if they demonstrated the use of a measure of parental mental health and a measure of child morbidity, in order to investigate whether a relationship between these two variables was present. Additionally, only articles written in English were retained. 6 articles from this original search were included. A hand search of the reference list of the 37 articles generated from the original search was conducted and generated 18 more articles.

This accounts for the final 24 papers reviewed by this article.

Appendix B: Table containing overview of the 24 articles reviewed in section A

Table 1

Table to summarise all studies included in the review

Paper	Chronic condition	Parental psychological measures used	Child physical health monitored	Association present ?
Diseth et al. (2011)	Kidney transplantation	General Health Questionnaire (GHQ) (Goldberg & Williams, 1988)	Body Mass Index (BMI) & renal functio	Yes
Watson et al. (1997)	Renal replacement therapy	Perceived stress scale (PSS) (Cohen et al., 1983) Hospital Anxiety and Depression Scale (HADS) and Burden of Care (authors own measure)	Height and weight	Yes
Shalowitz et al. (2001)	Asthma	Centre Epidemiological Studies-Depression Scale (CES-D) (Radloff, 1977)	Composite measure of asthma morbidity	Yes
Wood et al. (2002)	Asthma	5 item Mental Health Scale (Ware et al., 1993)	Parent report of asthma symptoms and health care use	Yes
Weil et al. (1999)	Asthma	Brief Symptom Inventory (BSI) (Derogatis & Spencer, 1982)	Hospitalisation and parental report of asthma morbidity	Yes

Bartlett et al. (2001)	Asthma	CES-D	Emergency department use and parental report of asthma morbidity	Yes
Jessop et al. (1988)	Various chronic conditions	Psychiatric Symptom Index (Ilfield, 1976)	Maternal report functional of impairment and clinician report of burden	Yes
Szabo et al. (2009)	Asthma	Beck Depression Inventory (BDI)(Beck et al., 1961) and State Trait Anxiety Inventory (STAI) (Spielberger et al.,1971)	World Health Organisation asthma severity rating	No
Rodrigue et al. (1997)	Various transplantation groups	Parenting Stress Index (PSI)(Abidin, 1995); Impact on Family Scale (Stein & Reissman, 1985)	Number of hospitalisations	No
Kovacs et al. (1995)	Diabetes Mellitus	Hopkins Symptom Checklist (Derogatis et al, 1973)	Hospitalisation	No
Fielding & Brownbridge (1997)	End stage renal failure	Leeds Scale for the Self-Assessment of Anxiety and Depression (Snaith et al, 1977)	Hospitalisation and child self report of functional impairment	No

Klinnert et al. (1994)	Asthma	The Family Inventory of Life Events (FILE) (McCubbin et al, 1983); Clinician rated score of 'Parenting Risk'	Development of asthma	Yes
Mzarek et al. (1999)	Asthma	Perinatal Depression Scale (no reference provided) Clinician rated score of 'Parenting Risk'	Development of asthma	Yes
Klinnert et al. (2008)	Asthma	Rand Mental Health Inventory (Ware, Veit & Donald, 1985); 'Family Stress' (authors own composite measure)	Diagnosis of asthma	No
Wright et al. (2002)	Asthma	PSS	Prevalence of wheezing	Yes
Wright et al. (2004)	Asthma	PSS	Immunoglobulin-E levels	Yes
Wolf et al. (2008)	Asthma	PSS and CES-D	Increased inflammatory responses	Yes

Fredericks et al. (2007)	Liver transplantation	Paediatric Inventory for Parents (PIP) (Streisand et al, 2001); BSI; BDI; CHQ-PF50	Medication adherence, clinic attendance, hospitalisation, liver biopsies	Yes
Gerson et al. (2004)	Kidney transplantation	Child Health Questionnaire-Parent Form 50 (CHQ-PF50) (Landgraf et al, 1996) and PSI	Adherence to medication and parental rating of child pain and discomfort	Yes
Bauman et al. (2002)	Asthma	BSI	Non-adherence and parental report of asthma morbidity	Yes
Bartlett et al. (2004)	Asthma	CES-D	Adherence to asthma regimen and parental report of asthma morbidity	Yes
Chisholm et al. (2007)	Diabetes	PSI	Adherence to treatment	No
Foulkes et al. (1993)	Kidney transplantation	FILE	Adherence to medication	Yes (with one medication)
Bourdeau et al. (2007)	Various chronic conditions	PSI	Treatment adherence-parental and child report	Yes

Appendix C: Paediatric Renal-Carer Burden Scale (PR-CBS)

Below is a list of things that parents of children with kidney disease have said can cause difficulty. Please read each item and circle the number that best describes how much of a problem this has been for you over the last month.

	Never	Rarely	Sometimes	Often	Always
Worrying about the effect of caring for my child on my health	1	2	3	4	5
Feeling that I am not able to 'switch off' to my child's condition	1	2	3	4	5
Worrying about the future	1	2	3	4	5
Feeling trapped because of caring for my child	1	2	3	4	5
Worrying if my child has had the correct amount of fluid	1	2	3	4	5
Blaming myself for my child's kidney problems	1	2	3	4	5
Feeling preoccupied with checking my child for signs of illness	1	2	3	4	5
Worrying about money due to the costs of my child's care	1	2	3	4	5
Feeling helpless when my child is ill or in pain	1	2	3	4	5
Frustration when dealing with staff that do not know my child	1	2	3	4	5
Feeling troubled by difficult memories of when my child was first diagnosed or has been very ill in the past	1	2	3	4	5
Worrying about my child during the night	1	2	3	4	5
Feeling that my child's condition has taken over my life	1	2	3	4	5
Worrying about the impact of my child's condition on my other children	1	2	3	4	5
Worrying about my child getting very ill or dying	1	2	3	4	5
Feeling that other people do not understand my situation	1	2	3	4	5
Blaming myself if my child gets ill or has bad test results	1	2	3	4	5
Feeling guilty about spending less time with my child / partner	1	2	3	4	5
Difficult feelings due to the uncertainty of my child's condition	1	2	3	4	5
Feeling unable to think about my own needs	1	2	3	4	5
Sadness about not socialising as much as I want to because of caring for my child	1	2	3	4	5
Feeling overwhelmed by trying to fit family life around my child's condition	1	2	3	4	5
Difficult feelings due to having no privacy when at the hospital	1	2	3	4	5
Worrying about getting medical procedures wrong (e.g. dialysis, injections, tube feeding) or taking measurements incorrectly	1	2	3	4	5
Feeling under pressure to be strong for my child and family	1	2	3	4	5

	Never	Rarely	Sometimes	Often	Always
Sadness that I can not do things that I used to do because of caring for my child (e.g. work, leisure activities, hobbies)	1	2	3	4	5
Feeling alone in caring for my child	1	2	3	4	5
Worrying that my child may have to be admitted to hospital	1	2	3	4	5
Feeling overwhelmed by decisions that I have to make about my child's condition	1	2	3	4	5
Sadness about the things that my child misses out on	1	2	3	4	5
Feeling unable to 'switch off' when waiting for test results or a telephone call from the hospital	1	2	3	4	5
Arguing with my partner / family about my child's care	1	2	3	4	5
Worrying about how my child is coping	1	2	3	4	5
Feeling uncertain about how to manage my child's emotions and difficult behaviour	1	2	3	4	5
Sadness from feeling that I am not the person that I used to be	1	2	3	4	5
Worrying about disruptions to my child's education	1	2	3	4	5
Feeling overwhelmed by changes in my child's usual treatment	1	2	3	4	5
Sadness that I do not have a 'normal' relationship with my child	1	2	3	4	5
Worrying about my getting my child's medicines wrong	1	2	3	4	5
Feeling bored when having to spend time at the hospital	1	2	3	4	5
Holding back when I disagree with medical staff	1	2	3	4	5
Feeling overwhelmed by feeding difficulties (e.g. lack of appetite, managing diet restrictions, vomiting)	1	2	3	4	5
Worrying about having to deal with unexpected changes in my child's condition (e.g. unexpected hospital admissions)	1	2	3	4	5
Feeling frustrated when having to spend time at the hospital	1	2	3	4	5
Worrying about my child's growth and development	1	2	3	4	5
Feeling that I should be doing more for my child	1	2	3	4	5
Worrying that I have not understood medical information	1	2	3	4	5
Feeling exhausted from caring for my child	1	2	3	4	5
Difficult feelings due to my child taking responsibility in their care (e.g. worrying if medicines have been taken)	1	2	3	4	5
Feeling preoccupied with keeping my child safe from illness	1	2	3	4	5
Sadness about the impact of my child's kidney problems on my relationship with my partner	1	2	3	4	5

Figure 3. *Version 2 of the measure - adapted further to piloting (n = 51 items)*

Appendix D: PR-CBS acceptability questions

Please answer the following questions thinking about the Paediatric Renal Caregiver Burden Scale (PR-CBS)

How long did it take you to fill out this questionnaire?
Did you find the questionnaire alright to answer?
Were there any questions you didn't understand?

Appendix E: Care Giver Strain Questionnaire (CGSQ)

This has been removed from the electronic copy

Appendix F: Participant information sheet

Participant Information Sheet

Developing a measure of burden and adjustment in carers of children with chronic kidney disease.

We would like to invite you to take part in our study. This sheet is to give you some information about the study before you decide to take part.

What is the purpose of the study?

We are designing a questionnaire to help indicate how carers cope with any stress that may come with caring for a child with chronic kidney disease.

Why have I been invited to take part?

We are collecting data from carers of children undergoing all types of treatment for chronic kidney disease from site A and site B. All carers of children with chronic kidney disease from these sites are being asked to participate in this study.

Do I have to take part?

It is up to you to decide to join the study. We will describe the study and go through this information sheet. If you agree to take part, we will then ask you to sign a consent form. You are free to withdraw at any time, without giving a reason. This would not affect the standard of care you receive from the renal team at this unit.

What will happen if I decide to take part?

If you decide to take part we will ask you to fill in a questionnaire about your experiences of being a carer for someone with chronic kidney disease. We would also like you to fill in two other questionnaires that will help us understand if the questionnaire we have designed are measuring what we expected. These should only take about 20-30 minutes to fill out and can be done while you wait for your appointment. Alternatively, you could take them home and post them back to us in the envelope provided.

What will happen to the information in the questionnaires?

We will use the information in the questionnaires to help finalise the design of a new measure that is aimed at measuring stress in carers of children with chronic kidney disease. The questionnaires will be stored securely and destroyed once the study is finished. All information collected will be kept confidential to the research team and will not be used for any other purpose than already stated. We are also asking you to write down a few sentences about how accessible you found the questionnaire to fill in. We may use some quotes from this section in the write up of the research findings, but these will remain anonymous.

Why do I have to put my name on one of the questionnaires?

We are hoping to understand whether the questionnaire we have designed is able to identify carers who are experiencing high levels of burden. In order to do this we would be interested to know about participants who in the future get a referral from the renal team for extra psychological support. We need to have the names of participants who do get a referral in order to match these to the participants score on the original questionnaire. The chief investigator will be asking the renal team for the names of participants who they had referred for extra psychological support over the next 6 month period. Outside of the renal team, it will only be the chief investigator who will have this access to this information, and the names will be destroyed once the data has been recorded. If you are happy for this to happen, please write your name at the top of the carer burden questionnaire form.

If you do not wish for any future psychological referrals for you to be shared with the chief investigator, you can still fill out the questionnaire confidentially, just do not put your name on the top of the questionnaire.

Are there any risks to taking part?

It is not anticipated that there are any risks to filling out these questionnaires, however some questions deal with sensitive issues such as levels of stress, anxiety and depression. If you feel uncomfortable after filling

out any of the questionnaires please talk to either your healthcare professional from the renal team or your GP. You could also contact the researcher xxxxxxxxxx, who would be happy to answer any of your questions about the research that you may have. You can email them on xxxxxxxxxxx.

What will happen to the results of the research?

The results will hopefully give us a way to identify carers of children with kidney disease who are feeling burdened and stressed, and help staff to identify in the future families that may need additional help or support. It is hoped that results from this study will be published in a peer reviewed journal, but as stated before, your contribution will be anonymous. We may use some quotes from the short written answer section to illustrate if people found the questionnaire easy to fill in, but these will again be kept anonymous.

If you would like to receive a short summary of the findings of this research then please feel free to email xxxxxx, Chief Investigator, at xxxxxxxxxxxxx.

Appendix G: Participant consent form

Carer Identification Number for this trial:

CONSENT FORM

Title of Project: Developing a measure of burden and adjustment in carers of children with chronic kidney disease.

Name of Researcher: Nicola Jacyna

1. I confirm that I have read and understand the information sheet dated.....
for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

2. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.

3. I agree to take part in the above study.

Name of Carer :

Date :

Signature :

Appendix H: NHS Ethics approval

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Appendix I: NHS Site specific approval letters

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Appendix J: Table containing descriptive statistics for respondent's score on each item of the PR-CBS as well as total scores on all measures

	N	Mean	Standard deviation	Skewness	Kurtosis
Item 1	107	2.52	1.35	0.46	-0.94
Item 2	105	3.50	1.30	-0.50	-0.79
Item 3	106	4.01	0.98	-0.76	0.14
Item 4	107	2.42	1.31	0.59	-0.73
Item 5	102	3.64	1.18	-0.50	-0.56
Item 6	105	2.26	1.32	0.63	-0.74
Item 7	106	2.99	1.03	0.70	-0.24
Item 8	106	2.86	1.43	0.13	-1.24
Item 9	106	3.78	1.23	-0.81	-0.21
Item 10	106	2.94	1.26	0.08	-0.86
Item 11	106	2.99	1.16	0.17	-0.68
Item 12	107	2.87	1.22	0.26	-0.82
Item 13	107	2.84	1.43	0.19	-1.29
Item 14	103	2.76	1.33	0.21	-1.06
Item 15	107	3.27	1.34	-0.15	-1.1
Item 16	106	3.15	1.25	-0.17	-0.81
Item 17	106	2.33	1.14	0.49	-0.51
Item 18	106	2.79	1.15	0.23	-0.45
Item 19	106	3.07	1.14	-0.21	-0.67
Item 20	107	2.83	1.23	0.17	-0.74
Item 21	107	2.29	1.27	0.74	-0.45
Item 22	107	2.6	1.27	0.38	-0.83
Item 23	107	2.17	1.19	0.78	-0.24
Item 24	106	2.43	1.24	0.63	-0.34
Item 25	107	3.06	1.25	-0.11	-0.93
Item 26	105	2.42	1.17	0.46	-0.57
Item 27	105	2.38	1.27	0.53	-0.78
Item 28	104	3.28	1.11	-0.18	-0.52
Item 29	105	2.86	1.16	0.17	-0.77
Item 30	104	3.34	1.31	-0.44	-0.72
Item 31	104	3.23	1.21	-0.09	-0.82
Item 32	105	2.21	1.17	0.73	-0.19

Item 33	104	3.35	1.18	-0.20	-0.67
Item 34	105	2.92	1.24	-0.04	-0.87
Item 35	104	2.48	1.30	0.45	-0.84
Item 36	102	3.46	1.38	-0.44	-1.00
Item 37	104	2.34	1.05	0.63	-0.05
Item 38	105	1.77	1.1	1.48	1.54
Item 39	105	2.13	1.11	1.15	1.06
Item 40	105	2.77	1.31	0.17	-1.03
Item 41	104	2.15	1.1	0.93	0.46
Item 42	105	2.65	1.43	0.31	-1.2
Item 43	105	3.07	1.16	0.55	-5.29
Item 44	105	2.93	1.17	0.24	-0.65
Item 45	104	3.35	1.25	-0.23	-0.89
Item 46	104	3.00	1.24	0.15	-1.1
Item 47	103	2.21	1.12	0.80	0.24
Item 48	103	2.76	1.39	0.24	-1.07
Item 49	101	2.64	1.35	0.33	-1.00
Item 50	104	2.88	1.36	0.06	-1.19
Item 51	102	2.16	1.35	0.88	-0.47
Total 51 item PR-CBS score	105	141.91	39.27	0.31	-0.44
Total 20 item PR-CBS score	105	58.20	15.63	0.19	0.57
CGSQ	100	2.57	0.82	0.37	-0.82
HADS Total	95	8.87	4.52	0.12	-0.85
HADS Anxiety	95	6.35	4.59	0.66	-0.26
HADS Depression	95	15.09	8.51	0.4	-0.50

Appendix K: Floyd and Widamen (1995) factor analysis criteria

Information to include in when reporting exploratory factor analysis:

- Principal component analysis or common factors analysis
- Initial communality estimates (common factor analysis)
- Method of factor extraction
- Criteria for retaining factors
- Eigenvalues, percentage of variance accounted for by the the unrotated factors
- Rotation method (and rationale)
- All rotated factor loadings
- Factor inter-correlations (oblique solutions)
- Variance explained by factors after rotation.

Appendix L: Table detailing all item loadings on to factors following exploratory factor analysis of the PR-CBS using Principal Axis Factoring with Varimax Rotation with Kaiser Normalisation (n= 107)

Item	Factor 1	Factor 2	Factor 3	Factor 4	Factor 5
19. Difficult feelings due to the uncertainty of my child's condition	.699	.178	.189	.243	.136
15. Worrying about my child getting ill of dying	.665	.272	.254	.252	.101
9 Feeling helpless when my child is ill or in pain	.639	.173	.211	.018	.102
31 Feeling unable to "switch off" when waiting for test results or a phone call from the hospital	.626	.152	.333	.214	.101
28 Worrying that my child may be admitted to hospital	.607	.299	.265	.218	.255
29 Feeling overwhelmed by decisions that I have to make about my child's condition.	.597	.311	.206	.332	.243
12 Worrying about my child during the night	.564	.397	.108	.090	.082
43 Worrying about having to deal with unexpected changes in my child's condition (e.g. unexpected hospital admissions	.559	.229	.339	.114	.314
11 Feeling troubled by difficult memories of when my child was first diagnosed or has been very ill in the past.	.552	.113	.082	.161	.164
50 Feeling preoccupied with keeping my child safe from illness	.537	.163	.455	.157	.244
2 Feeling that I am not able to "switch off to my child's condition	.521	.433	.135	.134	.073
24 Worrying about getting medical procedures wrong (e.g. dialysis, injections, tube feeding) or taking measurements incorrectly	.495	.059	.251	.352	.278
20 Feeling unable to think about my own needs	.492	.441	.396	-.036	.060
7 Feeling preoccupied with checking my child for signs of illness	.489	.246	.156	.155	.062
18 Feeling guilty about spending less time with my child/partner	.465	.279	.410	.124	.174
25 Feeling under pressure to be strong for my child and family	.459	.364	.371	.169	.193
3 Worrying about the future	.451	.272	.008	.289	-.040

5 Worrying is my child has had the correct amount of fluid	.424	.113	.127	.423	-.096
14 Worrying about the impact of my child's condition on my other children	.400	.218	.389	.008	.197
39 Worrying about getting my child's medicines wrong	.388	.017	.300	.386	.262
21 Sadness about not socialising as much as I want because of caring for my child.	.262	.712	.386	.030	.177
4 Feeling trapped because of caring for my child	.199	.695	.124	.240	.105
26 Sadness that I can not do the things I used to do because of caring for my child (e.g. work, leisure activities, hobbies)	.191	.650	.366	.183	.286
13 Feeling that my child's condition has taken over my life	.449	.585	.357	.023	.198
1 Worrying about the effect of caring for my child on my own health	.238	.575	-.038	.082	.122
35 Sadness from feeling I am not the person I used to be	.238	.561	.384	.232	.103
27 Feeling alone in caring for my child	.292	.535	.453	.156	.255
8 Worrying about money due to the costs of my child's care	.260	.508	.024	.406	.086
16 Feeling that other people do not understand my situation	.423	.506	.259	.132	.112
38 Sadness that I do not have a "normal" relationship with my child	.144	.448	.338	.308	.159
23 Difficult feelings due to having no privacy when at the hospital	.219	.417	.246	.227	.363
51 Sadness about the impact of my child's kidney problems on my relationship with my partner	.284	.392	.386	.154	.372
32 Arguing with my partner/family about my child's care	.147	.385	.304	.243	.322
45 Worrying about my child's growth and development	.175	.074	.644	.113	.178
30 Sadness about the things my child misses out on	.349	.248	.611	.218	.068
36 Worrying about disruptions to my child's education	.177	.197	.582	.136	.190
33 Worrying about how my child is coping	.318	.376	.531	.301	-.013
46 Feeling that I should be doing more for my child	.286	.178	.528	.470	-.001
37 Feeling overwhelmed by changes in my child's usual treatment	.396	.239	.517	.324	.185

22 Feeling overwhelmed by trying to fit family life around my child's condition	.440	.498	.505	.007	.348
42 Feeling overwhelmed by feeding difficulties (e.g. lack of appetite, managing diet restrictions, vomiting)	.201	.317	.392	.083	.278
47 Worrying that I have not understood medical information	.087	.039	.118	.705	.176
17 Blaming myself if my child gets ill or has bad test results	.304	.207	.110	.560	.097
34 Feeling uncertain about how to manage my child's emotions and difficult behaviour	.115	.269	.293	.556	.057
49 Difficult feelings due to my child taking responsibility in their care (e.g. worrying if medicines have been taken)	.088	-.071	.245	.475	.129
41 Holding back when I disagree with medical staff	.018	.159	-.113	.461	.349
6 Blaming myself for my child's kidney problems	.158	.178	.024	.375	-.043
44 Feeling frustrated when having to spend time at hospital	.253	.285	.139	.070	.731
40 Feeling bored when having to spend time at the hospital	.028	.065	.268	.161	.729
48 Feeling exhausted in caring for my child	.386	.407	.224	-.041	.470
10 Frustration when dealing with staff who do not know my child	.291	.196	.123	.163	.443

Appendix M: Content analysis coding framework

Code	Summary of theme
Fine	No problems identified with any aspect of completing the PR-CBS
Current relevance	Answers to questions on the PR-CBS would have been different at other times over the course of their child's condition or are not relevant to their current situation
Sadness	Acknowledgment of feeling sad or emotional while completing the PR-CBS
Reflective experience	Answering the questionnaire allowed for an opportunity to think back over the experience of living with their child's condition
Answer options vague	Suggestion of the difficulty of reducing a number of complex and varying experiences into a single number

Appendix N: Annual report of progress to NHS ethics committee

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Appendix O: Submission guidelines for Journal of Pediatric Psychology

MANUSCRIPT PREPARATION

Instructions to Authors

The Journal of Pediatric Psychology is an official publication of the Society of Pediatric Psychology, Division 54 of the American Psychological Association. JPP publishes articles related to theory, research, and professional practice in pediatric psychology.

Types of Manuscripts:

- Original research, including case studies
- Review articles
- Commentaries

Manuscript preparation: General Instructions

Full instructions for uploading data and files etc. are given on Manuscript Central at the website under Instructions for online submission:http://www.oxfordjournals.org/our_journals/jpepsy/for_authors/submission_online.html

Organization of manuscripts

Manuscript Central will guide authors through the submission steps, including: Abstract, Keyword selection, and the Manuscript. The manuscript must contain an Introduction, Methods, Results, Discussion, Acknowledgements and Reference List.

Length of manuscript: Original research articles should not exceed 25 pages, in total, including title page, references, figures, tables, etc. In the case of papers that report on multiple studies or those with methodologies that necessitate detailed explanation, the authors should justify longer manuscript length to the Editor in the cover letter. Case reports should not exceed 20 pages. Review articles should not exceed 30 pages. Commentaries should not exceed 4 pages. The Journal of Pediatric Psychology no longer accepts brief reports but will accept manuscripts that are shorter in length than the 25 page manuscripts.

Manuscripts (text, references, tables, figures, etc.) should be prepared in detailed accord with the Publication Manual of the American Psychological Association (6th ed.). There are two exceptions:

(a) The academic degrees of authors should be placed on the title page following their names, and

(b) a structured abstract of not more than 150 words should be included. The abstract should include the following parts:

(1) Objective (brief statement of the purpose of the study);
(2) Methods (summary of the participants, design, measures, procedure);
(3) Results (the primary findings of this work); and
(4) Conclusions (statement of implications of these data).

Key words should be included, consistent with APA style. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch). Authors should remove all identifying information from the body of the manuscript so that peer reviewers will be unable to recognize the authors and their affiliations. E-mail addresses, whenever possible, should be included in the author note.

Informed consent and ethical treatment of study participants. Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s). Authors will also be asked to sign a statement, provided by the Editor that they have complied with the American Psychological Association Ethical Principles with regard to the treatment of their sample.

Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Terminology should be sensitive to the individual who has a disease or disability. The Editors endorse the concept of "people first, not their disability." Terminology should reflect the "person with a disability" (e.g., children with diabetes, persons with HIV infection, families of children with cancer) rather than the condition as an adjective (e.g., diabetic children, HIV patients, cancer families). Nonsexist language should be used.

Special instructions for types of manuscripts

(1) Treatment studies/Randomized controlled trials: If you are submitting a manuscript of a randomized clinical trial to JPP, you are required to submit a flowchart of your research showing the steps found in the Consort E-Flowchart. This should be submitted as a figure. The Consort E-Flowchart and a checklist of items to be included when reporting a randomized trial can both be found on http://www.consort-statement.org

(2) Case reports should not exceed 20 pages. Case reports are appropriate to document the efficacy of new treatment applications; to describe new clinical phenomena; to develop hypotheses; to illustrate methodological issues, difficult diagnoses, and novel treatment approaches; and to identify unmet clinical or research needs. Guidelines for case study submissions can be found in Drotar, D. (2009). Editorial: Case Studies and Series: A Call for Action and Invitation for Submissions, Journal of Pediatric Psychology, 34, 795-802.

Guidelines for Single Subject Studies: Please read Rapoff, M. & Stark, L. (2008).Editorial:Journal of Pediatric Psychology Statement of Purpose: Section on Single-Subject Studies.

(3) Measurement development and validation articles: For additional guidance please read, Holmbeck, G. & Devine, K. (2009) Editorial: An Author's Checklist for Measure Development and Validation Manuscripts.

(4) Review articles: Please consult Checklist for Preparing and Evaluating Review Articles Scholarly reviews should not exceed 30 pages.

(5) Commentaries: Commentaries are invited on all topics of interest in pediatric psychology, and should not exceed 4 pages, including references.

Additional Guidance:

The following links provide additional guidance for authors and reviewers. Editorial Policy, Authors' Checklist, Guidelines for Reviews, Mentoring Policy, Suggestions for Mentored Reviews, "People First,", NIH policy, Replication of research, Duplicate and redundant policies Conflict of interest

See the following articles for detailed guidance concerning preparation of manuscripts:Editorial: Thoughts in Improving the Quality of Manuscripts Submitted to the Journal of Pediatric Psychology: How to Write a Convincing Introduction. ; Methods: Editorial: How to Report Methods in the Journal of Pediatric Psychology; Results and Discussion: Editorial: How to Write an Effective Results and Discussion Section for the Journal of Pediatric Psychology.

Funding

Details of all funding sources for the work in question should be given in a separate section entitled 'Funding'. This should appear before the 'Acknowledgements' section.

The following rules should be followed:
- The sentence should begin: 'This work was supported by …'
- The full official funding agency name should be given, i.e. 'the National Cancer Institute at the National Institutes of Health' or simply 'National Institutes of Health', not 'NCI' (one of the 27 subinstitutions) or 'NCI at NIH'(full RIN-approved list of UK funding agencies)
- Grant numbers should be complete and accurate and provided in parentheses as follows: '(grant number xxxx)'
- Multiple grant numbers should be separated by a comma as follows: '(grant numbers xxxx, yyyy)'
- Agencies should be separated by a semi-colon (plus 'and' before the last funding agency)
- Where individuals need to be specified for certain sources of funding the following text should be added after the relevant agency or grant number 'to [author initials]'.

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Seehttp://www.oxfordjournals.org/for authors/repositories.html for details. Authors must ensure that manuscripts are clearly indicated as NIH-funded using the guidelines above

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