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CAREGIVER BURDEN IN PAEDIATRIC CHRONIC KIDNEY DISEASE

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Summary

Section A provides an overview of the role of family caregivers of individuals with chronic illness, and describes key conceptualisations and theories posited in the caregiver literature. This is followed by an overview of research conducted with caregivers of children with CKD, a summary of the limitations of this research, and suggestions for future research.

Section B documents a research study conducted to develop a measure of caregiver burden specific to caregivers of children with CKD. An overview of the stages undertaken to develop the measure are outlined, including the completion of interviews with caregivers and healthcare professionals to generate a measure item pool, item reduction, construction of a provisional measure, and a piloting exercise. Based on completion of these stages, the 51-item 'Paediatric Renal Caregiver Burden Scale' (PR-CBS) was developed. Section B concludes with a summary of the findings of the research study, an overview of its limitations, and suggestions for the utility of the PR-CBS.

Section C is a critical appraisal of the conducted research study, and includes an overview of research abilities acquired during its completion, reflections on how the research may have been conducted differently, implications for future clinical practice, and ideas for future research.

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SECTION A

Family caregivers of children with chronic kidney disease (CKD): A review of the literature

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Abstract

Advances in the treatment of paediatric chronic illness, including chronic kidney disease (CKD), have resulted in hospital-based care being increasingly replaced with home-based care, provided largely by family caregivers. While CKD in childhood is relatively rare, its impact can be pervasive, not only for the affected child but also for the family caregiver; children with CKD are dependent on their caregivers for a number of complex and intensive caregiving duties which include medication management, ensuring adherence to a strict dietary and fluid regimen, regular transportation to hospital, and providing dialysis at home where required. There is increasing concern that caregiver demands can have a profound effect on the physical, social and emotional wellbeing of those providing care. The first section of this review provides an overview of the role of family caregivers of children with chronic illness, and describes some of the conceptualisations and theories suggested to account for the effects of, and responses to, caregiving. The second section of this review provides an overview of the existing literature exploring the experiences of caregivers of children with CKD. A literature search was conducted through the MEDLINE(R) (1950-2010), PsycINFO (1806-2010), and EMBASE (1980 – 2010) databases. Of the 24 studies reviewed, common findings included caregiver reports of poor physical health, restlessness, helplessness, uncertainty, preoccupation with the future, impact upon family plans and relationships, loss of social contacts, and stress resulting from caregiving duties. This review concludes with an overview of the limitations of this research and suggestions for future research.

Introduction

Family caregiving and chronic illness

A 'caregiver' has been defined as an unpaid person who helps an individual cope with disease or illness (Hileman, Lackey & Hassanien, 1992). Caregiving in the context of chronic illness has largely been studied in the fields of geriatrics and paediatrics (Raina et al., 2004). While increasing emphasis has been placed on the role of caregivers of elderly family members, particularly in the context of age-related chronic conditions such as dementia, caregiving for a chronically ill family member is not just a late life phenomenon. Indeed, a significant number of children live with complex and continuing health needs (Department of Health, 2004).

The child healthcare paradigm shift

Prior to the 1980s, the prevailing healthcare paradigm for childhood chronic illness comprised hospital-based care, provided largely by health professionals. In line with advances in treatment for paediatric chronic illness however, hospital-based care has been increasingly replaced with home-based care provided largely by family members (Kepreotes, Keatinge & Stone, 2010; Murphy, 2008), with day to day care shifting into the family's domain, that is, the home (Desguin, Holt & McCarthy, 1994). While this shift has provided various benefits for children with chronic illness and their families, it has also brought increased demands for family caregivers (Raina et al., 2004).

Chronic kidney disease in childhood

Chronic kidney disease (CKD) is a term used to describe irreversible kidney damage or reduced kidney function that persists for more than three months. There are five stages of CKD, progressing from near normal kidney function through to end stage renal disease (ESRD; stage 5), where renal function is less than 15% of normal function and dialysis or transplantation is usually required to sustain life. While the most common causes of CKD amongst adults are hypertension, diabetes mellitus and intrinsic kidney (glomerular) diseases, the causes of childhood CKD are more commonly congenital abnormalities of the kidney and urinary tract (CAKUT), genetic or inherited diseases, or diseases acquired later in childhood (Hari et al., 2003; www.aakp.org). Children comprise a relatively small proportion of those with CKD, with a mean annual incidence and prevalence of <100 cases per million children and adolescents (Ardissino, et al., 2003; UK Renal Registry, 2009). The implications of childhood CKD are however nonetheless pervasive, not only for the child, but also for their main caregiver (Aldridge, 2008; Gayomali, Sutherland & Finkelstein, 2008).

Family caregiving and treatment for children with CKD

Despite modern advances in the treatment of CKD, lifelong intervention is required (Snethen, Broome, Bartels, & Warady, 2001). Children with CKD are dependent on their caregivers for complex, continuous and intensive support (Tong, Lowe, Sainsbury & Craig, 2010) with caregiving tasks including medication management, ensuring adherence to a strict fluid and dietary regimen, and regular transportation of the child to hospital, alongside bearing the psychological burden of having a chronically ill child (Gayomali, Sutherland & Finkelstein, 2008).

The kidney has numerous important functions, including the removal of water and waste from the body and the regulation of fluids in the body. For children whose kidney disease has progressed to ESRD (stage 5), dialysis or kidney transplantation is usually required to sustain life. Dialysis comprises the removal of water and body waste from the blood, and is achieved by either haemodialysis (HD) or peritoneal dialysis (PD). During HD blood is removed from the patient (either through an indwelling catheter in the neck, or a needle in a fistula) and pumped through a dialyser membrane, where the blood is essentially cleaned before being returned to the patient; HD conventionally takes place in hospital on a thrice weekly basis for 4-5 hours at a time. While there is an emerging trend to shift HD care into the home, this is still rare amongst children. Alternatively, PD is administered at home by trained family caregivers, usually on a nightly basis. During PD the abdominal lining (the peritoneum), acts as the dialysis membrane and dialysis fluid is cycled in and out of the abdomen via a catheter, indirectly cleaning the blood of toxins. Although PD has the benefit of freedom from frequent hospital attendance, bulky supplies of dialysis fluid are accommodated at the family home, and peritonitis (inflammation of the peritoneum) can be a complication. Dialysis by either HD or PD is usually a temporary bridge until a kidney transplant is available. Living donation is more common in children as parents are often able and willing to donate. While kidney transplantation is a desired goal in the treatment of childhood CKD (owing to increased survival and health related outcomes), it does not signal the end of treatment; the average lifespan of a transplanted kidney is 20 years and future transplantation or dialysis is required for continued survival once the transplanted kidney fails.

The role of caregivers in outcomes for children with CKD

While children are dependent by their nature, and 'caregiving' is therefore a normal part of being a parent, providing care to a child with a chronic illness involves activities above and beyond those required in everyday parenting (Case-Smith, 2004). There has been increasing concern that caregiver demands can have a profound effect on the physical, social and emotional wellbeing of those providing care (Belasco & Sesso, 2002). Friedman (2006) reports that in the context of paediatric CKD a heavy personal toll can be extracted from caregivers. High levels of parental stress have been reported amongst paediatric CKD caregivers, which has been found to influence coping and the quality of care provided (Friedman, 2006; Tsai et al., 2006). Not surprisingly, the quality of care provided by family caregivers of children with CKD is an important determinant in the child's outcome, both medically and developmentally (e.g. Brownbridge & Fielding, 1994; Gerson, Furth, Neu & Fivush, 2004; Reiss, 2005; Watson, 1997). While in paediatric CKD care efforts usually focus on the provision of the medical treatment and wellbeing of the affected child, it has been argued that family caregivers of children with CKD should be considered as a target of intervention "both to benefit the child indirectly, and to make explicit that parents deserve care in their own right" (Tong et al., 2008, p. 358). (For a review of studies examining the experience of being a child with CKD see Darbyshire, Oster & Henning, 2006).

Caregiving for a child with a chronic illness: Conceptual and theoretical overview

The effects of and responses to caregiving

Multiple terms have been used to describe the effects of caregiving, largely delineated into the positive and negative consequences of the caregiver role (Hunt, 2003). While positive conceptualisations of caregiving include caregiver esteem, uplifts of caregiving, caregiver satisfaction, and finding meaning through caregiving (for further details see Hunt, 2003), it is apparent that conceptualisations descriptive of the detrimental impact upon those providing care tend to dominate in the relevant literature.

Caregiver stress

The caregiving experience has commonly been conceptualised in terms of stress (Raina et al., 2004), with the concept of caregiver stress featuring heavily in the caregiver literature. Nolan, Grant and Ellis (1990), define caregiver stress as a response to caregiving resulting from a cognitive imbalance between the perceived nature of a demand and the perceived capabilities of the person to cope with such a demand. Caregiver stress is said to be determined largely by a caregiver's perceived subjective stress, more so than tangible objective stressors (Maurin & Boyd, 1990); stressors in the context of caregiving are defined as the problematic conditions and circumstances experienced by caregivers (Aneshensel, Pearlin, Mullan, Zarit & Whitlatch, 1995).

Despite seemingly similar caregiving circumstances, not all caregivers respond to the caregiving role in the same way. Attempts to understand variations in responses to the caregiving role have tended to focus on what is known about the process of stress (Oyebode, 2003). Lazarus and Folkman (1984), conceptualised stress as being heavily dependent on the cognitive processes of the individual, emphasising the process of the *appraisal* of an event or situation as the precursor of the stress reaction; stress is therefore not inherent in a situation, nor solely attributable to an event, but instead arises from the transaction between an individual and their environment (Lazarus & Folkman, 1984). The concepts of appraisal and coping are central to any psychological theory of stress, whereby appraisal refers to an individual's evaluation of the significance of an event, and coping refers to an individual's efforts in thought and action to manage specific demands (Lazarus, 1993).

The dominant theoretical model of caregiving assumes that the onset and progression of chronic illness is stressful for both patient and caregiver, and as such can be studied within the framework of traditional 'stress-coping' models (Shulz & Martire, 2004). In the context of caregiving, Raina et al. (2004) note that stress-coping models serve to explain the considerable variations that exist in response to seemingly similar caregiving roles. Krulik et al. (1999) have related the factors outlined by Lazarus and Folkman (1984) as important in the appraisal process directly to that of caring for a child with a chronic illness. For example, the degree to which the stressful event can be escaped has been noted to be a key factor in the appraisal process, which in the context of caring for a child with a chronic illness is limited; moreover, a further appraisal factor of relevance in this context is the extent to which a caregiver views their child's condition as disrupting family life (Krulik et al., 1999).

Caregiver burden

Alongside caregiver stress, the concept of caregiver burden is one of the most common concepts in the caregiving literature. First introduced by Grad and Sainsbury (1963) in the context of caring for a family member with mental illness, caregiver burden was initially defined as any cost to the individual's family, and was therefore initially viewed as a unidimensional concept. Caregiver burden was then dichotomised into objective and subjective dimensions, with objective burden defined as the extent of disruptions or changes in various aspects of the caregiver's life, and subjective burden defined as the caregiver attitude or emotional response to the caregiving experience (Montgomery, Gonyea, & Hooyman, 1985). The most recent trend is to consider caregiver burden as a *multidimensional* construct, with Chou (2000) positing a definition of caregiver burden as "an individual's subjective perception of overload in one or more of four perspectives: physical, psychological, social and financial through the caregiving process" (p. 405). Caregiver burden is suggested to result from an imbalance of *perceived* demands and resources, whereby the caregiver will feel burdened by the extent to which they perceive the care-receivers demands or associated demands as outstripping the available resources (Chou, 2003).

Caregiver grief

Doka and Aber (2002) note that grief is an important part of the caregiver experience that is often overlooked. The chronic sorrow model, posited by Olshansky

(1962) regards chronic sorrow as a coping mechanism that allows for periodic grieving; in the context of caring for a child with a chronic illness, a reaction of chronic sorrow is viewed as one of functional adaptation (rather than acceptance of the child's condition), whereby sadness and grief-related feelings occur periodically and in between these episodes the person functions 'normally' (Melynk, Feinstein, Moldenhouer & Small, 2001). Clubb (1991) proposed a time bound model to describe parental responses to childhood chronic illness, postulating that adaptation occurs over a period of time, and parental acceptance of the child's condition is implied in relation to a number of sequential stages, namely: impact, denial, grief, focusing attention, and closure. Melnyk et al. (2002) note that there has been debate in the literature as to whether parents are truly able to achieve closure in this context. Accordingly a 'hybrid model' which contains elements of the chronic sorrow concept and time-bound framework has been proposed, which suggests that parents progress through the stages suggested in the time-bound model but re-experience peaks in the grieving process during developmental transitions and high-risk periods.

The empirical study of caregivers of children with CKD

The objective of the next section of the review is to synthesise and summarise research which has explored the experiences of family caregivers of children with CKD.

Literature search and results

A search of the literature yielded 24 relevant articles; see Appendix 1 for details of the search strategy and inclusion criteria. The scope of the retrieved studies ranged from qualitative explorations of the lived experiences of paediatric CKD caregivers through to studies of mixed or purely quantitative design, seeking to quantify caregiver outcomes such as quality of life.

Overview of included articles

An overview of the retrieved studies is outlined below, with the findings of qualitative studies described first, followed by a summary of studies employing mixed or quantitative methodologies. Table 1 (on page 20) provides a summary of the studies included in this review, including details of the caregiver sample and methodology.

Qualitative studies

A recent review by Tong et al. (2008) serves to provide a comprehensive overview of the qualitative study of the paediatric CKD caregiver experience; see Appendix 2 for a summary of study details. The 16 qualitative studies included in the review by Tong et al. (2008) included caregivers of children across the trajectory of CKD, with the findings classified accordingly by stage of CKD. Common themes amongst caregivers of children in the pre-dialysis stage of CKD were difficulties with blame, depression, and uncertainty about diagnosis and the future. Amongst caregivers of children receiving dialysis (HD and PD) common themes were emotional turmoil, social life restrictions, difficulties with the treatment regimen, hospital visits and communication with clinical staff. Caregivers of children post-transplant reported similar difficulties to those caring for children on dialysis, but expressed additional uncertainty regarding the potential rejection of the kidney transplant, transplant complications and concern over family donor's well-being. Based upon these findings, Tong et al. (2008) developed ten themes grouped into three interrelated clusters: intrapersonal, interpersonal and external issues. Common intrapersonal experiences included shock at initial diagnosis, constant uncertainty about prognosis, lack of confidence in delivering care, and pressures of vigilance and fatigue. Interpersonal experiences included a strengthening of marital relationships or partner neglect, disruptions to family life, sibling jealousy and resentment, issues relating to support from friends and family, and difficulties with parent–staff relationships. External issues included management of the medical regimen, organisation of transport, accommodation and finances, and balancing medical care with domestic responsibilities. On the basis of this review, Tong et al. (2008) concluded that parenting a child with CKD demands "problem solving, information seeking, and financial and practical skills at a time when the capacity to cope is threatened by physical tiredness, uncertainty, and disruption to peer support within and outside the family structure" (p. 349).

Further to this review, Tong, Lowe, Sainsbury and Craig (2010) conducted a qualitative study comprising 20 interviews with parents of children across all stages of CKD, from which four major themes were identified. The first theme 'absorbing the clinical environment' included difficulties with the acceptance of diagnosis and permanence of CKD, forced assimilation into a new environment, experiences of medical procedures, and issues relating to relationships with medical staff. Within the second theme, 'medicalising parenting', the dual roles of parenting and medical caregiving were identified as stressful, exhausting and overwhelming; the physical and emotional challenges of medication adherence, feeding and fluid restrictions, dialysis, monitoring for infection, hygiene precautions, self-blame and management of the child's psychological issues were noted in this theme. Within the third theme, 'disrupting family norms', issues included spousal tension, sibling neglect, impact upon family plans, reduced financial reserves, accommodation of medical equipment at home, and the impact of family decisions about kidney donorship. The final theme, 'coping strategies and support structures', included issues related to dependence and support from health care providers, and the communication of information. Identified internal coping strategies included grieving and reframing the problem, while external coping sources included practical and emotional support from others, including other parents of children with CKD. Tong et al. (2010, p. 555) concluded that "being a parent of a child with CKD was consistently reported as being a pervasive and profoundly negative experience".

Studies using mixed or quantitative methods

Of the studies employing mixed or purely quantitative methodologies, those conducted with caregivers of children receiving a specific CKD treatment modality are presented first, followed by an overview of studies which included caregivers of children across the CKD trajectory.

Haemodialysis (HD)

A relatively early study conducted by Wolters, Daniels-Wegdam and Donckerwolcke (1980), explored the experiences of caregivers of children receiving hospital-based HD for at least two years. 36 caregivers (18 couples) completed a semi-structured interview and questionnaire developed by the authors to identify caregiver difficulties. Findings indicated that the dialysis regimen had a huge emotional impact for child and parent, and was often accompanied by radical changes in family circumstances (e.g. relocation). Caregivers noted the value of contact with nurses, alongside reluctance to ask questions of medical staff. Further to commencement on HD, increased caregiver physical and psychosocial problems were reported (e.g. physical complaints, irritability, absence from work, preoccupation with the future) alongside difficulties amongst siblings, impact on family leisure time, and issues relating to kidney donation.

Reichwald-Klugger et al. (1984) investigated the psychosocial adaptation of children and their caregivers receiving hospital-based HD (n=10) and home-based HD (n=10). Interviews to elicit information pertaining to daily life, the child's disease, treatment, physical capacities and family life were conducted. Information relating to tolerance of treatment was obtained from questionnaires. Caregivers of children on hospital-based HD reported restlessness, helplessness, being 'bound', and fear of complications. Caregivers of children on home-based HD reported an inability to relax, orientation of family life towards treatment, fear of complications, high treatment responsibility, and guilt from unsuccessful fistula punctures (i.e. being unable to needle the fistula to gain access for haemodialysis). Disruption to social contacts was more notable amongst caregivers of children on home-based HD compared to those receiving hospital-based HD.

Peritoneal Dialysis (PD)

Hulstijn-Dirkmaat and Damhuis (1994) explored parental stress amongst caregivers of children receiving continuous ambulatory peritoneal dialysis (CAPD; 3-4 manual exchanges throughout the day) and continuous automated cyclic peritoneal dialysis (CCPD; 8-10 exchanges overnight co-ordinated by a machine). A questionnaire developed to assess the aspects of caregiving deemed most difficult or to cause greatest burden was completed by 28 parents (14 couples), three times over one year. Increased stress was determined more by psychological aspects (e.g. uncertainty) than medical aspects of caregiving; concerns about the future contributed significantly to caregiver stress. Increased stress was reported amongst caregivers of older children (i.e. those aged 5 years and above), and those with the experience of a failed transplant. Caregiver stress did not fluctuate over the duration of the study.

More recently Tsai et al. (2006) explored depression and quality of life amongst 32 caregivers of children receiving PD and 64 controls (parents of healthy children). Participants completed measures of depression (Taiwanese Depression Questionnaire;

Lee, Yang & Lai, 2000) and quality of life (World Health Organization QOL BRIEF-Taiwan Version; Yao, Chung, & Yu, 2002). Increased rates of depression and reduced quality of life were found amongst caregivers of children on PD compared to healthy controls. Tsai et al. (2006) concluded that caring for children on PD "has a substantial adverse psychosocial effect on caregivers" (Tsai et al., 2006, p.70).

Transplant

Fedewa and Oberst (1996), explored caregiving difficulties amongst three fathers and 17 mothers of children aged 4 to 17 years, at two to fourteen months post-transplant. Caregivers completed measures of caregiving demand and difficulty (Caregiver Burden Scale, CBS; Zarit, Reever & Bach-Peterson, 1980), appraisal (Appraisal of Caregiving Scale, ACS; Oberst, Thomas, Gass, & Ward, 1989), and mood (short form of the Profile of Mood States; POMS-S; Shacham, 1983). The most demanding and difficult tasks were reported as increased household domestic duties, providing emotional support, transportation, monitoring and reporting symptoms, and managing behavioural problems. Caregiving demand and difficulty were greatest amongst carers of adolescents, those with other dependents, and those in employment. Mood dysfunction scores were low, but were associated with increased perceptions of caregiving difficulty.

Hasegawa et al. (2005) investigated quality of life amongst paediatric kidney transplant recipients and their caregivers prior to, and at least six months post-transplant. The SF-36 (Japanese version; Fukuhara, Bito, & Green, 1998), was completed by 54 caregivers, 47 of whom were living donors. Following transplant, while significant improvements were found in the social functioning and mental health of caregivers post-transplant, a significant decrease in physical health amongst caregivers was reported.

Studies across CKD stages and treatment modalities

An overview of studies employing mixed or purely quantitative methodologies conducted with caregivers of children across the spectrum of CKD care is provided below.

Reynolds, Garralda, Jameson and Postlethwaite (1988) compared outcomes in caregivers of 22 children receiving hospital-based HD with those of caregivers of 22 children with less severe chronic renal failure (not yet on dialysis); parents of 31 healthy controls were also included. Structured interviews were completed to obtain information relating to family background, socioeconomic factors, and a description of the child's illness and family impact. Measures of stress (Social Stress and Supports Interview, SSSI; Bailey & Garalda, 1987) and current mental distress (General Health Questionnaire, GHQ; Goldberg, 1978) were completed. Caregivers in both the HD and non-dialysis group reported stress due to financial costs incurred through hospital visits and admissions, and the impact on relationships with other children. Disruptions to family life, impact upon marriage, and difficulties with education were most notable amongst caregivers of children receiving HD. While concerns about the future were reported by both caregiver groups, concerns about growth were more notable amongst caregivers of children on HD. Completion of the SSSI indicated that caregiver stress was not significantly different across the HD and non-dialysis groups.

Brownbridge and Fielding (1991) explored the psychosocial adjustment of children receiving CAPD, HD, or those post-transplant, as well as their caregivers (n = 73). Structured interviews were completed to obtain information about sociodemographic variables, treatment history, and adjustment to treatment, alongside measures of child behavioural disturbance (Rutter A scale; Rutter, Tizard, & Whitmore, 1977), and anxiety and depression (Leeds Scale for Self-assessment of Anxiety and Depression; Leeds SAD; Snaith, Bridge & Hamilton, 1977). No differences were found across treatment groups in marital strain or stress associated with treatment. However, amongst caregivers of children receiving dialysis (CAPD or HD), increased practical difficulties were reported compared to those post-transplant. Increased depression and anxiety was found amongst caregivers of children on HD compared to those receiving CAPD. The authors concluded that kidney transplantation has a number of advantages for the psychosocial adjustment of caregivers of children in end stage renal failure, and that home-based dialysis (CAPD) may have advantages over hospital-based dialysis (HD).

Watson (1997) conducted a longitudinal study to explore caregiver demands amongst 38 caregivers of children commencing PD, HD or receiving a transplant. Measures of stress (Perceived Stress Scale; PSS 10; Cohen, Kamarck, & Mermelstein, 1983), anxiety and depression (Hospital Anxiety and Depression Scale; HADS; Zigmond & Snaith, 1983), information needs, and impact of illness were completed at baseline, 3 months, 6 months, and annually. In addition, a burden of care assessment (BCA) was developed which included domains of information needs, problems between siblings and parents, environment (e.g. poor housing, low income) and demands of treatment; a BCA was completed prior to commencement on dialysis or placement on the transplant waiting list, and every year thereafter. Findings indicated that increased stress, anxiety, and depression was found amongst mothers compared to fathers across all treatment groups, and amongst caregivers of children aged above 10 years compared to those of younger children. BCA scores were greatest (indicating highest burden) amongst caregivers of children commenced on PD.

Zelikovsky, Schast and Jean-Francois (2007) explored predictors of depression in a sample of 86 mothers and 58 fathers of children who were waiting for a kidney transplant; a third of the caregiver sample were caring for a child receiving PD or HD. Caregivers completed measures of parental stress (Pediatric Inventory for Parents, PIP;

Streisand, Braniecki, Tercyak, & Kazak, 2001), coping style (Brief Cope; Carver, 1997), and depression (Becks Depression inventory; BDI—II; Beck, Steer & Brown, 1996). Depression amongst mothers was predicted by reduced family income, increased stress, and the use of avoidant coping strategies, while depression amongst fathers was predicted by increased stress only.

Recently, Weidebusch et al. (2010) examined relationships between psychosocial strains, coping, and health-related quality of life (HRQOL) amongst caregivers of children across the stages of CKD. 105 mothers and 90 fathers (representing 108 families) completed measures of HRQOL (Ulm Quality of Life Inventory for Parents, ULQIE; Goldbeck, & Storck, 2002), psychosocial strains (Impact on Family Scale, IFS; Ravens-Sieberer, et al., 2001), and coping strategies (Social Orientations of Parents of Handicapped Children Questionnaires, SOEBEK; Krause & Peterson, 1998; Freiburg Questionnaire of Coping with Illness, FKV; Muthny, 1989). Caregivers of children undergoing dialysis perceived higher limitations in daily life, reduced quality of life and more psychosocial strains compared to caregivers of children receiving more conservative treatment or those post-transplant. Mothers reported reduced quality of life and increased psychosocial strains compared to fathers across all treatment groups.

Finally, while a relevant review of quantitative studies was not retrieved in the current search of the literature, it should be noted that a review article by Aldridge (2008), included studies exploring the process of adaptation and adjustment amongst families of children with CKD; the inclusion criteria was limited to articles specifying 'adjustment' or 'adaptation'. Of the eleven studies reviewed by Aldridge (2008), seven were deemed relevant to the current review and have been included and outlined accordingly. For information, Aldridge (2008) concluded that high levels of stress, depression, and anxiety were reported by caregivers adjusting to having a child with

CKD, with uncertainty, social isolation, and increased caregiving duties noted to contribute to the burden of providing care.

Summary of the empirical literature

The empirical study of caregivers of children with a chronic illness has tended to delineate the myriad of stressors that caregivers may experience (Brown et al., 2008). This appears particularly true of the qualitative studies conducted amongst caregivers of children with CKD, which have identified numerous potential stressors pertinent to these caregivers, which have been noted to span intrapersonal (e.g. uncertainty), interpersonal (e.g. family relationships) and external levels (e.g. treatment demands) (Tong et al., 2010). Those studies employing mixed and quantitative methodologies in this field serve to provide more evidence of the demands faced by caregivers of children with CKD, with reports of poor physical health, restlessness, helplessness, uncertainty, preoccupation with the future, impact upon family plans and relationships, loss of social contacts, and stress resulting from caregiving duties.

In addition to identifying common stressors, a number of studies have explored factors predictive of paediatric CKD caregiver outcomes, with poorer outcomes reported amongst mothers compared to fathers and amongst caregivers of older children compared to those of younger children. Of those studies exploring caregiver experiences and outcomes across CKD stages, poorer outcomes were found amongst caregivers of children on dialysis (HD or PD) relative to those caring for a child receiving more conservative treatment or those post-transplant; findings in relation to the relative caregiver outcomes for those of children on HD or PD are somewhat contradictory at present.

Table 1: Summary of studies included in the review of the empirical literature

(Studies listed in chronological order)

Author and Date	Caregiver sample (n)	CKD stage/treatment modality	Methodology
Wolters et al. (1980)	36	HD	Mixed (Semi-structured interviews; questionnaires)
Hislop and Lansing (1983)*	20	PD	Semi-structured interviews
Reichwald-Klugger et al. (1984)^	20	HD	Mixed (Structured Interviews; questionnaires)
Reynolds et al. (1988)^	44 (plus 31 controls)	Pre-dialysis and HD	Mixed (Structured interviews; questionnaires)
Waissman (1990)*	15	PD and HD	Semi-structured interviews
Brownbridge & Fielding (1991)^	73	PD, HD, or transplant	Mixed (Structured interviews; questionnaires)
Obrecht et al (1992)*	1	Transplant	In-depth interview
Hulstijn-Dirkmaat & Damhuis (1994)	28	PD (CAPD and CCPD)	Questionnaires
MacDonald (1995)*^	4	PD	In-depth interviews
Fedewa and Oberst (1996)	20	Transplant	Questionnaires
Middleton (1996)*^	8	PD	Focus group
Watson (1997)^	38	Commencing PD, HD or transplant	Questionnaires
Schultz and Farrell (1998)*	10	Predialysis and transplant	In-depth interviews
Nicholas (1999)*	32	PD, HD and transplant	In-depth interviews
Karrfelt et al (2000)*	18	Transplant	Semi-structured interviews
Baines et al (2001)*	7	Transplant	Semi-structured interviews
Cimete (2002)*	31	HD	Focus group
Hasegawa et al. (2005)	54	Transplant	Questionnaires
Tsai et al. (2006)^	32 (plus 64 controls)	PD	Questionnaires
Zelikovsky et al. (2007)	144	All CKD stages	Questionnaires
Aldridge (2008)	425	All CKD stages	Review article (Studies exploring the process of adaptation/adjustment in families of children with CKD)
Tong et al. (2008)~	358	All CKD stages	Review article(Qualitative studies)
Tong et al. (2010)	20	All CKD stages	In-depth Interviews
Weidebusch et al. (2010)	195	All CKD stages	Questionnaires

*Studies included in Tong et al. (2008) review (which were published post 1980, as specified in the search strategy).

^Studies included in Aldridge (2008) review deemed relevant to current review.

Critique of the empirical literature

While the studies conducted to date have provided much information about the experiences of caregiving in this context, these studies are not without their limitations. Firstly, many of the studies included in this review had small samples. While this is the norm amongst studies employing qualitative methodologies, it was notable that seven of the eleven studies using quantitative methods had a sample size of less than 55, thus compromising the validity and generalisability of these studies.

Moreover, very few of the studies reviewed included healthy control groups, with the exception of Reynolds et al. (1997) and Tsai et al. (2006); thus limiting the conclusions that can be made about outcomes amongst caregivers of children with CKD compared to those of healthy children. On a similar thread, the findings of studies conducted with caregivers of children receiving a specific CKD treatment modality are unable to draw conclusions about the potential relative impact upon caregivers of the various CKD stages and associated treatment modalities. Also, few studies included in this review employed a longitudinal design, with the exception of those by Watson (1997) and Hulstijn-Dirkmaat and Damhuis (1994); there is thus a paucity of research exploring the process of adjustment to diagnosis and the progression of CKD for this caregiver population.

All of the studies included in this review were completed in Europe, Australia, US or Canada with the exception of Tsai et al. (2006) and Hasegawa et al. (2005), which were conducted in Taiwan and Japan, respectively. As such, the findings of the studies outlined may only be representative of Western culture. Also, while most of the studies included in this review tended to focus on the caregiving experiences of mothers, which is not surprising since the vast majority of primary family caregivers of children with CKD are mothers, nonetheless an understanding of the perspectives of fathers in this context is lacking.

A further limitation is that the studies conducted in this field to date have been largely atheoretical, with very few studies citing theoretical underpinnings of their investigations. Raina et al. (2004) note that theoretical frameworks in the context of caregiving have often not translated to empirical study, whereby "most of the current broadly-based studies investigating caregiver health in the child health literature do not rely on any specific theoretical frameworks that guide research into an understanding of the mechanisms by which some caregivers experience negative consequences and others do not" (p. 4). The exception in the current review is a study by Fedewa and Oberst (1996) which was guided by existing theory, that is, the cognitive appraisal stress-coping model of stress; the authors note that the assumption underlying their research was that a caregiver's unique appraisal or perception of the caregiving situation is more likely to explain responses to caregiving (e.g. mood disturbance), than demographic or illness characteristics. Conversely, many of the studies included in this review appear to have focused their efforts on exploring the role of demographic or illness factors in the prediction of caregiver outcomes. Indeed, numerous studies in this review have tended to demarcate the experiences and outcomes of caregivers of children with CKD according to factors such as the child's age and stage of CKD or treatment modality (e.g. Reynolds et al. 1988; Brownbridge & Fielding, 1991; Hulstijn-Dirkmaat & Damhuis, 1994; Tsai at el. 2006). While overall the vast majority of studies included in this review did not explicitly state their theoretical underpinnings, it is arguable that their findings nonetheless serve to bolster the concept of caregiver burden as a *multidimensional* construct. That is, the studies included in this review have identified a wide range of difficulties faced by caregivers of children with CKD, which can be delineated into physical, psychological, social, and financial domains, in line with the conceptualisation of caregiver burden posited by Chou (2003).

It was notable that a number of researchers developed their own measures to assess paediatric CKD caregiver difficulties (i.e. Wolters et al., 1980; Hulstijn-Dirkmaat & Damhuis, 1994; Watson, 1997) but did not always report the psychometric properties of these newly developed measures. Moreover, while some studies used existing measures of caregiver stress and burden (e.g. Caregiver Burden Scale, Appraisal of Caregiving Scale), these measures have been largely developed and validated amongst caregivers of elderly family members, specifically those with dementia, rather than caregivers of children with chronic illness (Melnyk et al., 2001). Only one study included in this review used a measure of parental stress which was developed and validated specifically amongst caregivers of children with a chronic illness (i.e. Zelikovsky et al., 2007, who utilised the Pediatric Inventory for Parents). Furthermore, while some studies used measures of depression, anxiety, and quality of life to quantify caregiver outcomes, it has been argued that such 'proxy' measures are unlikely to fully reflect the experience of caregiving for a child with CKD (Aldridge, 2008).

Conclusions and future directions

It has been increasingly argued that family caregivers of children with CKD should be considered as a target of intervention (Tong et al., 2008). A common recommendation of the studies included in this review was indeed the need to assess and monitor caregivers of children with CKD with a view to providing targeted intervention and improved outcomes for these caregivers, and ultimately their children (Aldridge 2008; Brownbridge & Fielding, 1994; Hulstijn-Dirkmaat & Damhuis, 1994; Tong et al., 2008; Tsai, et al., 2006; Reynolds, et al., 1988; Watson, 1997; Weidebusch, et al., 2010; Wolters, et al., 1980; Zelikovsky, et al., 2007). However, despite the evident demands faced by these caregivers, and the emphasised need to monitor such demands, there is currently no tool available to measure caregiver burden in this population. While measures of caregiver stress and burden do exist, these have been largely developed for caregivers of chronically ill elderly family members; the authors of measures developed specifically for children and adults with CKD (Bradley, 1997; Goldstein et al., 2006) have highlighted the importance of creating instruments that measure the unique aspects of kidney disease. Aldridge (2008) notes that a measure of carer burden in this population would allow clinicians to qualify areas of stress amongst carers and to monitor response to targeted intervention. Therefore, there is a need for a further exploration of the most appropriate means of quantifying the experiences of caregivers of children with CKD, with a further view to identifying areas of need and providing targeted intervention where necessary. A measure developed specifically in this context could thus serve to support multidisciplinary teams working with children with CKD and their primary caregivers in tailoring a treatment regimen that may best serve not only the child with CKD and their main caregiver, but the family as a whole.

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SECTION B

Development of a measure of caregiver burden in paediatric chronic kidney disease (CKD): Phase One

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Abstract

Background: Despite a recognised need to monitor caregiver burden in caregivers of children with chronic kidney disease (CKD), there is no measurement tool currently available to meet this aim. The present research documents the development of a measure of caregiver burden specific to family caregivers of children with CKD. **Methods**: Interviews were conducted with 16 caregivers of children with CKD and 10 healthcare professionals in order to generate measure items. A provisional version of the measure was developed and piloted with 18 caregivers of children with CKD and five healthcare professionals. **Results**: An initial pool of 97 items was generated from the content of interviews, which was reduced to 60 items following review for item redundancy. A piloting exercise provided preliminary evidence for the usability, readability, and relevance of measure items; adaptations further to piloting resulted in the 51-item 'Paediatric Renal Caregiver Burden Scale' (PR-CBS). **Conclusions**: It is hoped that the PR-CBS will serve to identify areas of need amongst caregivers of children with CKD, and in turn improve outcomes for this caregiver population and children with CKD.

Background

Chronic kidney disease (CKD) in childhood and family caregiving

Chronic kidney disease (CKD) is a term used to describe irreversible kidney damage or reduced kidney function that persists for more than three months. While children and adolescents[1] comprise a relatively small proportion of those with CKD (Ardissino, et al., 2003; UK Renal Registry, 2009), its implications are nonetheless pervasive, not only for the child, but also for their main family caregiver. A 'caregiver' is defined as an unpaid person who helps an individual cope with disease or illness (Hileman, Lackey, & Hassanien, 1992). Advances in treatment for paediatric chronic illness, including CKD, have resulted in hospital-based care provided by healthcare professionals being increasingly replaced by home-based care, provided largely by family caregivers (Kepreotes, Keatinge & Stone, 2010; Murphy, 2008). In the context of paediatric CKD, family caregivers may assume a multitude of caregiving duties including medication management, ensuring adherence to strict dietary and fluid requirements, regular transportation to hospital, and providing dialysis at home where required; duties which occur alongside bearing the psychological burden of having a chronically ill child (Aldridge, 2008; Gayomali, Sutherland & Finkelstein, 2008; Tong, Lowe, Sainsbury & Craig, 2010).

There are five stages of CKD, progressing from near normal kidney function through to end stage renal disease (ESRD; stage 5)[2], where dialysis or transplantation is required to sustain life. Dialysis partly replaces the function of the kidney by removing water and body waste from the blood; this is achieved either by haemodialysis (HD) or peritoneal dialysis (PD). During HD blood is removed from the patient (either through an indwelling catheter in the neck, or a needle in a fistula) and pumped through a dialyser membrane, where the blood is essentially cleaned before it is returned to the patient. HD conventionally takes place in hospital on a thrice weekly basis for 4-5 hours at a time, and while there is an emerging trend to shift HD care into the home, this is still rare amongst children. Alternatively, PD is administered at home by trained family caregivers,

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[1] For the remainder of this paper, 'children' is used to denote children and adolescents aged 0-18

[2] For the remainder of this paper, 'CKD' is used to denote all stages of CKD (stage 1-5, inclusive of ESRD)

usually on a nightly basis. During PD the abdominal lining (the peritoneum), acts as the dialysis membrane and dialysis fluid is cycled in and out of the abdomen via a catheter, indirectly cleaning the blood of toxins. PD has been described as time-consuming and technologically complex, with few children under the age of 14 able to perform PD independently (Tong, Lowe, Sainsbury, & Craig, 2008; Tsai, Liu, Tsai & Chou, 2006). While PD has the benefit of freedom from frequent hospital attendance, bulky supplies of dialysis fluid have to be accommodated at the family home, and peritonitis (an inflammation of the peritoneum) can be a complication. Dialysis by either HD or PD is usually a temporary bridge until a kidney transplant is available. However, while kidney transplantation brings freedom from dialysis and is a desired goal in the treatment of childhood CKD, it does not signal the end of treatment; various complications can ensue post-transplant, particularly during the first six months. Caregivers of children post-transplant monitor their child for indicators of kidney rejection, continue to support their child in attending hospital (initially very frequently), manage a complex medication regimen, and ensure adherence to strict dietary and fluid requirements. Moreover, as the average lifespan of a transplanted kidney is 20 years, future transplantation or dialysis is required for continued survival once the transplanted kidney fails.

In paediatric CKD care, efforts usually focus on the provision of medical treatment and monitoring the wellbeing of the affected child. It has been argued however that family caregivers of children with CKD should increasingly be considered as a target of intervention "both to benefit the child indirectly, and to make explicit that parents deserve care in their own right" (Tong, et al., 2008, p. 358). Friedman (2006) notes that a heavy personal toll can be extracted from caregivers in the context of paediatric CKD. In light of research indicating that increased parental stress is associated with poorer outcomes for children with CKD, both medically and developmentally (e.g. Brownbridge & Fielding, 1994; Fielding et al., 1985; Gerson, Furth, Neu & Fivush, 2004; Reiss, 2005; Watson, 1997), the increased consideration of caregivers as a target of intervention is of paramount importance.

Responses to caregiving

Despite seemingly similar caregiving circumstances, not all caregivers respond to caregiving in the same way (Raina, 2004); the same objective situation can be experienced as difficult by one caregiver and not by another (Braithwaite, 1992). Attempts to understand such variation has tended to focus on what is known about the process of stress (Oyebode, 2003). Lazarus and Folkman (1984) conceptualised stress as heavily dependent on the cognitive appraisal of an event or situation; the concept of caregiver stress features heavily in the caregiver literature, with its presence posited to be determined largely by perceived subjective stress, more so than tangible objective 'stressors' (Maurin & Boyd, 1990). Alongside caregiver stress, caregiver burden is one of the most common concepts in the caregiving literature. Caregiver burden was initially viewed as a unidimensional concept, defined as any cost to the individual's family, but was subsequently dichotomised into its objective dimensions such as the tangible disruptions to the caregiver's life, and subjective dimensions, that is, the caregiver's attitude or emotional response to caregiving (Montgomery, Gonyea & Hooyman, 1985). The most recent trend in the literature is to view caregiver burden as a multidimensional construct, with Chou (2000) positing a definition as: "an individual's *subjective* perception of overload in one or more of four perspectives: physical, psychological, social and financial through the caregiving process" (p. 405).

The empirical study of caregivers of children with CKD

The study of the paediatric CKD caregiver experience has been explored via both qualitative and quantitative methods, with qualitative studies conducted to date tending to identify a multitude of potential stressors pertinent to these caregivers. Based on a review of 16 qualitative studies exploring the paediatric CKD caregiver experience, Tong et al. (2008) outlined three clusters spanning intrapersonal issues (e.g. uncertainty, fatigue), interpersonal issues (e.g. disruptions to family life, difficulties with relationships with medical staff), and external issues (e.g. caregiving demands of the CKD treatment regimen). Further to this review, Tong et al. (2010) conducted an in-depth interview study with 20 parents of children across all stages of CKD, from which four major themes were identified: absorbing the clinical environment (e.g. experiences of medical procedures), medicalising parenting (e.g. dual roles of parenting and caregiving), disrupting family norms (e.g. sibling neglect, impact upon family plans) and coping strategies and support structures (e.g. issues related to dependence on healthcare providers). Tong et al. (2010) concluded that being a parent of a child with CKD was "consistently reported as being a pervasive and profoundly negative experience" (p. 555).

Studies employing quantitative methodologies provide more evidence of the range of difficulties experienced by this caregiver population, with findings commonly including reports of poor physical health, restlessness, helplessness, uncertainty, preoccupation with the future, concern about their child's growth and development, reduced family income, disruptions to family activities and relationships, loss of social contacts and difficulties arising from the CKD treatment regimen (Aldridge, 2008; Fedewa & Oberst, 1996; Hasegawa et al., 2005; Hulstijn-Dirkmaat & Damhuis, 1994; Reynolds, Garralda, Jameson & Postlethwaite, 1988; Tsai, Liu, Tsai, & Chou, 2006; Weidebusch, et al., 2010; Wolters, Daniels-Wegdam & Donckerwolcke 1980; Zelikovsky, Schast, & Jean-Francois 2007). While studies conducted amongst caregivers of children with CKD have been largely atheoretical (with the exception of Fedewa & Oberst, guided by the cognitive appraisal model of stress), it is perhaps arguable that their findings can be understood in terms of the posited concepts and theories in the field of caregiving. For example, the findings relating to the multitude of stressors faced by these caregivers serve to bolster the concept of caregiver burden as a multidimensional construct, with difficulties reported across physical, psychological, social, and financial domains, thus in line with the definition of caregiver burden posited by Chou (2000).

The need for a measure of paediatric CKD caregiver burden

Numerous researchers have highlighted the need to monitor outcomes amongst caregivers of children with CKD, with a view to providing targeted intervention where required and improving outcomes for caregivers and ultimately their children (Aldridge 2008; Brownbridge &

Fielding, 1994; Hulstijn-Dirkmaat & Damhuis, 1994; Tong et al., 2008; Tsai, et al., 2006; Reynolds, et al., 1988; Watson, 1997; Weidebusch, et al., 2010; Wolters, et al., 1980; Zelikovsky, et al., 2007). Despite the recognised demands faced by this caregiver population, and the emphasised need to monitor such demands, there is no specific tool currently available to measure caregiver burden in this population. In the research conducted to date with caregivers of children with CKD, 'proxy' measures of stress, anxiety and depression have tended to be used; it has been argued however that these may not fully capture all of the elements related to how family caregivers adjust to having a child with kidney failure (Aldridge, 2008). Moreover, the 'global' measures of caregiver burden in existence have been largely developed for caregivers of adult relatives, namely those with dementia (Robinson, 1983), stroke (Visser-Meily, Post, Riphagen & Lindeman, 2004), or a mental health diagnosis (Schene, Tessler, & Gamache, 1994). The importance of creating instruments that measure the unique aspects of kidney disease has been noted (Bradley, 1997; Goldstein et al., 2006). In the last decade, measures specific to the renal caregiver population have begun to emerge (e.g. Horsburgh et al., 2008; Schneider, 2010; Teixido et al., 2006); however, these have been developed for caregivers of adults with renal disease, and have tended to focus on operationalising the objective dimension of caregiver burden, or objective/subjective dimensions simultaneously, which has been cited as problematic (this is discussed in more detail below).

Overall, Gayomali et al. (2008) note that "in the pediatric CKD population, there are minimal data regarding the identification of aspects of caregiver burden" (p. 3750). The development of a measure of caregiver burden specific to caregivers of children with CKD could serve to support multidisciplinary teams working with these caregivers to identify those who are in need, and to plan targeted support and tailored treatment programmes where possible. The need to assess for caregiver burden in this caregiver population has increased significance in light of the increasing number of children with CKD who receive home based therapies, where the role of caregivers in the successful delivery of treatment is paramount (Gayomali et al., 2008).

The measurement of caregiver burden

Despite consensus on the relevance of the concept of caregiver burden, its conceptualisation has varied widely. A number of researchers have noted that measurement instruments developed to date often differ in their operationalisation of the subjective and/or objective dimensions of caregiver burden (Gerritsen & van der Ende, 1993; Stommel, Given & Given, 1990; Thompson & Doll, 1982). It has been argued that this lack of consensus has resulted in a lack of progress in the identification of caregivers who may benefit most from intervention (Braithwaite, 1992; Cousins, Davies, Turnbull & Playfer, 2002; Gerritsen and van der Ende, 1994). Gerritsen & van der Ende (1994) argue that "to increase the specificity of the concept, only the subjective dimension should be operationalized, because distress is regarded as the core of agreement on what constitutes burden, i.e. 'worry, anxiety, frustration, depression, fatigue, poor health, guilt and resentment'" (p. 483); a number of caregiver burden researchers concur (e.g. Chou 2000; Poulshock & Deimling, 1984; Stommel et al., 1990).

The present study

The researcher's university supervisors were approached by a consultant nephrologist from a large London children's hospital to initiate a research project that would result in the development of a measure of paediatric CKD caregiver burden. The following recommended phases of measure development were planned in order to meet this aim:

- Determination of the construct to be measured (i.e. specification of the conceptualisation of caregiver burden guiding the development of the measure).
- Generation of an item pool relevant to a measure of paediatric CKD caregiver burden
- Reduction of the generated item pool as appropriate
- Determination of the measurement format (i.e. item wording, response scales)
- Construction of a provisional measure
- Preliminary piloting with a small sample

Method

Design

The study design followed recommended steps of scale development (DeVellis, 1991; Oppenheim, 1996; Rattray & Jones, 2005) which included: the determination of the construct to be measured, item generation, item reduction, determination of the measurement format, construction of a draft measure and preliminary piloting. An extensive evaluation of the psychometric properties of the newly developed measure will be completed by another trainee clinical psychologist at this university as a subsequent Major Research Project.

Materials

To facilitate the generation of an item pool, a semi-structured interview schedule for use with caregivers of children with CKD and renal healthcare professionals was developed. The content of this schedule was based on the existing research conducted with caregivers of children with CKD; the interview schedules for caregivers and healthcare professionals differed slightly in content (Appendix 3 and 4 respectively). The interview schedules were revised in consultation with a caregiver of a child receiving treatment for CKD, two consultant paediatric nephrologists, and two academic research supervisors. The interview schedule included open ended questions querying the main difficulties of caring for a child with CKD, and included prompts relating to the perceived social, financial, physical and emotional impact of caregiving for a child with CKD. The interview schedule also contained prompts to explore issues relating to the impact of CKD and caregiving upon the child and family life, caregiver CKD treatment responsibilities, and interactions between caregivers and medical staff. The interview schedule included a final question querying if there were any issues that had not been discussed or required further exploration.

Participants

Sixteen family caregivers of children with CKD and ten healthcare professionals completed semi-structured interviews during the item generation phase of this research. Eighteen caregivers and five healthcare professionals participated in the piloting phase of the research. Further details of the sample characteristics are outlined in the results section.

Ethics

Ethical approval for this study was granted by the National Research Ethics Service, in addition to local research governance approval (Appendix 5 and 6 respectively). As per the requirements of this approval, a declaration of the end of the study and an accompanying final report were completed (Appendix 7 and 8 respectively); see Appendix 9 for a summary report for participants.

Procedure

Determination of the construct to be measured

DeVellis (1991) notes that a key initial step in the development of a new measure is the determination of the construct to be measured, arguing that scale development researchers "must specify at least a tentative theoretical model that will serve as a guide to scale development, which may be as simple as a well-formulated definition of the phenomenon they seek to measure" (p. 52). Further to a review of the relevant caregiving literature, the current research was guided by the conceptualisation of caregiver burden as a multidimensional construct, defined as "an individual's *subjective* perception of overload in one or more of four perspectives: physical, psychological, social and financial through the caregiving process" (as posited by Chou, 2003, p. 405), thus focusing on the subjective appraisals of caregivers rather than objective facts and circumstances; as suggested by a number of caregiver burden researchers (e.g. Chou, 2000; Cousins, et al., 2002; Gerritsen & van der Ende, 1994; Poulshock & Deimling, 1984; Stommel et al., 1990).

Item Generation

To inform the generation of an item pool, semi-structured interviews were conducted with caregivers of children with CKD and relevant healthcare professionals.

Recruitment for interviews

Primary family caregivers of children aged 18 or younger receiving treatment for CKD were eligible for inclusion in this study. Caregivers were invited to participate via a poster (Appendix 10) and information sheets (Appendix 11) placed in relevant waiting rooms and wards at a London hospital, and in person by the researcher in relevant waiting rooms and wards when in attendance at the hospital. Potential participants were given at least 24 hours to make a decision about participation and were able to express an interest in participating or ask further questions via the contact details listed on the poster and information sheet. The aim was to conduct interviews with approximately three caregivers of children who were either pre-dialysis, receiving haemodialysis (HD), receiving peritoneal dialysis (PD), or were post-transplant, in an attempt to capture caregiver experiences across the trajectory of CKD.

All healthcare professionals involved in the care of children with CKD at the hospital were eligible for inclusion in the study and were invited to participate via email or in person by the researcher; an alternative information sheet was developed for healthcare professionals (Appendix 12). Healthcare professionals spanning a range of disciplines were invited to participate (e.g. doctors, specialist nurses in PD, HD and transplant, members of the psychosocial team).

Interviews

Interviews were scheduled at a mutually convenient time with caregivers and healthcare professionals who expressed an interest in participating in interviews. The same interview procedures were adhered to for both caregivers and healthcare professionals as appropriate. Caregivers who were limited in their ability to meet alone with the researcher for an interview when in attendance at the hospital (due to needing to be with their child at all times) were given the option of completing an interview over the telephone. Interviews conducted at the hospital took place in an isolated room. Prior to interviews taking place, informed written consent was obtained (Appendix 13 and 14) and participants were given information about the interview process. Interviews were conducted using the semi-structured interview schedule and recorded using digital audio recording equipment. Recruitment for interviews continued until interviews had been conducted with a sufficiently diverse sample of caregivers and healthcare professionals and until saturation of the item pool was achieved. Interviews were transcribed verbatim.

Item Pool Generation

Of the caregiver burden measure studies published to date, very few authors have outlined the processes of item generation, with most simply stating that a list of items or phrases were 'generated' (e.g. Schneider, 2006). In the absence of specific guidelines for the generation of measure items from interview transcripts, the use of thematic analysis (TA) to provide a framework for item generation and the tentative categorisation of items was deemed a suitable substitute. The conventional phases of TA as outlined by Braun and Clarke (2006) were utilised as deemed applicable to the process of organising interview data for the purposes of the current research. In line with 'Phase 1' of TA (i.e. familiarisation with the data), immersion in the data was achieved by transcription of the interviews, in addition to repeated readings of interview transcripts. This exercise was followed by 'Phase 2' of TA (i.e. generation of initial codes), during which statements in the transcripts deemed relevant to caregiver burden were identified and highlighted; from these statements, potential measure items were formulated alongside in a right hand column. In the context of the current research, the generation of potential measure items from highlighted statements served as a proxy to the generation of 'initial codes' (as so termed in conventional TA). For an example of this process see Appendix 15. On completion of this phase, the full list of potential measure items were extracted and collated.

Braun and Clarke, 2006 (p.89) suggest that during Phase 2 the focus is on identifying "interesting aspects in the data that may form the basis of repeated patterns (themes)". Thus while conventionally, the process of initial coding of the data serves to organise the data into meaningful groups, for the purposes of the current research this process served to inform the content of a new measure at the item (rather than thematic) level. It is arguable therefore that Phase 1 and 2 comprised the primary task for the purposes of the current research.

Following completion of Phase 1 and 2, the analysis was refocused at the broader level of 'themes', or in this instance, tentative domains of the new measure. In 'Phase 3' of TA (i.e. searching for themes), the emphasis is upon identification of themes in the data, with Phase 3 ending with a collection of 'candidate' themes (Braun & Clarke, 2006). In the current research, Phase 3 thus comprised a review of the list of generated items and their tentative categorisation into a number of domains. 'Phase 4' of TA (i.e. reviewing themes) conventionally involves a rigorous review and refining of the 'candidate themes'. As a subsequent research project is being completed, which will involve an exploration of the factor structure of the newly developed measure via quantitative methods (i.e. item and factor analysis), the completion of a rigorous analysis of the proposed themes (in this instance, domains) was deemed to be a somewhat redundant task; the primary tasks of the current research were thus deemed to have been achieved by completion of Phase 1, 2, and 3 as deemed applicable.

To ensure quality assurance during the procedure outlined, a checklist of criteria listed by Braun and Clarke (2006, p.96) was adhered to as appropriate. That is, the data was transcribed accurately, and each data item was given equal attention in the coding process and there was deemed to be a good fit between what the researcher claimed to set out to do, and what the researcher did; Braun and Clarke (2006) note that "As thematic analysis is a flexible method.. you need to be clear about what you are doing, and what you say you are doing needs to match up with what you actually do" (p. 96). In addition, the procedures through which items were generated and categorised into domains were reviewed by two academic supervisors.

Item Reduction

DeVellis (1991) recommends that for the purposes of item reduction, researchers can eliminate items based on criteria including: lack of clarity, questionable relevance, or undesirable similarity to other items. In order to meet the aims of the item reduction stage of measure development, the generated item pool was reviewed by the lead researcher and the lead supervisor (a consultant paediatric nephrologist) and reduced as appropriate according to the criteria outlined.

Determination of measurement format

Determination of the measurement format was informed by guidance outlining the desired traits of measurement items and response scales (DeVellis, 1991; Oppenheim, 1996). This stage occurred simultaneously with the item reduction stage whereby during the reduction of the generated item pool, items were reworded as statements capable of being used with the response scale: *never, rarely, sometimes, often, always*. Likert scales such as this are the most common measurement response format and increase statistical variation of the overall scale, which is a desirable quality for measurement purposes (as opposed to binary responses such as yes/no) (DeVellis, 1991). The wording of items was based upon the aim of operationalising the subjective (rather than objective) dimension of caregiver burden. For example, rather than wording items to elicit simply whether caregivers were tasked with a particular caregiving duty (e.g. 'I have to make sure that my child has the correct amount of fluid'; that is, an objective fact), items were worded to elicit the caregivers subjective appraisal of events and circumstances (e.g. 'Worrying if my child has had the correct amount of fluid'). Furthermore, on the recommendation of DeVellis (1991), items were worded so that they were neither overly mild, nor overly strong (to avoid too little response variation), and double-barrelled statements were avoided. Further to some experimentation with including both positively and negatively worded items, items were worded consistently in the same (negative) direction; DeVellis (1991) notes that while ordering items in the same direction can increase the risk of agreement bias, it can also avoid confusion.

Piloting

A provisional measure was constructed and piloted with a sample of caregivers and healthcare professionals to serve as a preliminary evaluation of the usability and readability of the newly developed measure, and as a further check of the face and content validity of measure items. Participants taking part in the pilot exercise completed the draft measure in addition to an adapted version of the measure constructed to elicit information about the perceived relevance and importance of the items listed. Moreover, qualitative feedback was sought on the usability and readability of the measure, and on its general content. All caregivers who participated in interviews agreed to be contacted for a future pilot exercise and were sent copies of the measure in the post with a return stamped addressed envelope, or via email where preferred. The measure was also piloted amongst a new sample of caregivers recruited from the waiting rooms of outpatient clinics and relevant wards. Healthcare professionals who participated in interviews also agreed to be contacted to feed back their views on a draft of the measure.

Results

The results are outlined below for each of the steps of measure development undertaken.

Item Generation

Interview Sample

Semi-structured interviews were conducted with 16 caregivers and 10 healthcare professionals. Of the 16 caregiver interviews, ten were conducted in person at the hospital and six were conducted on the telephone; interviews ranged in duration from 20-65 minutes. The characteristics of the interviewed caregivers and their children are shown in Table 1.

In addition to discussing their child's current treatment regime at interview, caregivers also discussed experiences of caring for a child across the trajectory of CKD and their experiences of other treatment modalities, as illustrated in Table 2.

Table 1.

Caregiver interview sample characteristics

Caregiver		
Gender	Female	14
	Male	2
Age (years)	20-29	3
	30-39	6
	40-49	6
	50-59	1
Ethnicity	White	11
	Asian	3
	Black	1
	Mixed	1
Child		
Gender	Female	7
	Male	9
Age (years)	0-1	3
	2-5	3
	6-11	7
	12-18	3
CKD Stage / Treatment	Pre-Dialysis	3
	Peritoneal Dialysis (PD)	2
	Haemodialysis (HD)*	6
	Transplant (Tx)	5
CKD Cause	CAKUT^	7
	Congenital nephrosis	2
	Renovascular disease	2
	Glomerular diseases	4
	Malignancy	1
Time since diagnosis (years)	0-1	4
	2-5	6
	6-10	3
	>10	3

** In all cases HD denotes hospital-based HD (as opposed to home-based HD)*

^ CAKUT = Congenital abnormalities of the kidney and urinary tract

Table 2.

CKD stage / treatment modalities discussed at interview

CKD Stage / Treatment	Current	Discussed at interview
Pre-Dialysis	3	8
Peritoneal Dialysis (PD)	2	9
Haemodialysis (HD)	6	10
Transplant (Tx)	5	7

The 10 healthcare professionals who participated in interviews spanned a range of disciplines and included two consultants, six nurses (inclusive of nurse specialists in PD, HD and transplant), and two members of the psychosocial team. Nine interviews with healthcare professionals were conducted in person at the hospital and one was completed on the telephone; interviews ranged in duration from 20-45 minutes.

Item Pool Generation

Familiarisation with the interview data was achieved through the process of transcription and repeated readings of transcripts; statements within interview transcripts deemed relevant to caregiver burden were highlighted and corresponding potential measure items were noted in a column alongside; an example is illustrated in Figure 1. An example of a full transcript illustrating this process is included as Appendix 15 (written consent to include this transcript as an appendix was obtained).

I live with that all the time, um, if she's got a temperature, I have to ring the hospital, if she's got a cough I have to check that out, or if her bloods aren't right we have to come back here. You know, and I have to make that call.	**I feel overwhelmed by the decisions I have to make about my child's condition**

Figure 1. *Sample section of transcript with potential measure item*

The full list of potential measure items was extracted from each interview and collated into a comprehensive item pool; the item pool was cross referenced with existing relevant literature. On the basis of the recommendation that being overinclusive is desirable during the item generation stage of the measure development process (DeVellis, 1991), a large item pool (n = 97) was initially generated. The generated item pool is listed in the left hand column of Table 3. The items contained in the item pool were organised into nine broad domains, as illustrated in Table 3. Proposed domains included those identified as relevant to caregiver burden as defined by Chou (2000), (i.e. physical, financial, social and emotional/psychological), alongside the emergent domains of caregiver role/identity, impact on family (including practical and interpersonal issues), impact on child, CKD treatment responsibilities, and a domain containing items relating to contact with the hospital/medical staff (including practical and interpersonal issues). It should be noted that these domains are entirely provisional and that the factor structure of the developed measure will be explored via quantitative methods (item and factor analysis) in the research project scheduled to be conducted subsequently.

Item reduction and determination of measurement format

The pool of 97 generated items was reviewed by the researcher and lead supervisor (a consultant paediatric nephrologist) for the purposes of item reduction. This exercise resulted in the reduction of the item pool from 97 to 60 items; this process is outlined in Table 3, whereby generated items were either retained as stand alone items or a number of items were merged due to evident overlap with other items (as illustrated by the bracketing in Table 3).

During the item reduction exercise, the reduced pool of 60 items was subjected to rewording as appropriate for the developed measure instructions, and Likert response scale of: *never (1), rarely (2), sometimes (3), often (4), always (5)*, with increased scores indicative of increased caregiver burden. A draft measure was then constructed (Version 1; Appendix 16). The instructions for the measure and an example section are shown in Figure 2:

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

	Never	Rarely	Sometimes	Often	Always
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**

Figure 2. *Sample section of Version 1 of the measure*

A time window of one month was chosen for the measure. DeVellis (1991) notes that when making a decision regarding the time frame for a measure, it is important to pose the question "Is the phenomenon of interest a fundamental and enduring aspect of an individuals personality, or it is likely to be dependent on changing circumstances?" (p.74). Some measures make no reference to a time frame, implying a universal time perspective, such as in the case of measures of locus of control, which imply an enduring belief about causality (e.g. "If I take the right actions I will be healthy"); which is consistent with the theoretical characterisation of locus of control (DeVellis, 1991). In the context of the current research, caregiver burden is regarded as a transient phenomenon, which may vary over time; what might be considered burdensome at one time may change as the caregiver adapts and finds new ways to cope with the patient's impairment, or conversely new difficulties may arise (Chou 2000). A specified time frame for the current measure was therefore deemed appropriate. Of those measures in existence relevant to the current research, it is apparent that the length of specified time windows has varied widely. While some measures have not specified no fixed time period at all (e.g. Caregiver Burden Scale, CBS; Zarit, Reever & Bach-Peterson, 1980), others have indicated a one month period (e.g. Parent Experience of Child Illness, Bonner et al. 2006; Perceived Stress Scale, Cohen & Williamson, 1988), while some have specified a time frame of six months (Caregiver Strain Questionnaire, Brannan, Heflinger & Bickman. 1997). Consideration was given to a time window of two weeks or one month. Discussion with the consultant nephrologists supervising this research led to the decision to state a time window of one month, with the rationale that outpatient appointments may take place on approximately a monthly basis, and thus this time period is marked in a tangible way, thus aiding accuracy of reflections on their experiences. Moreover, it was felt that a shorter time period may not be sufficient to encompass the range of issues contained within the measure (e.g. hospital attendance, organisation of family leisure activities). Participants in a subsequently planned research study will be asked to comment on their experiences of rating the measure over a one month period.

Table 3.
Item pool generation/Item pool reduction (and provisional domains)

Generated Item Pool (Items = 97)	**Reduced Item Pool (Items = 60)** (Items reworded for Likert response scale and measure instructions)
Physical	**Physical**
1. My health has suffered because of caring for my child	**1.** Worrying about the effect of caring for my child on my health
2. Caring for a child with kidney disease is exhausting	**2.** Feeling exhausted from caring for my child
3. It is very tiring having to be organised about everything	**3.** Feeling exhausted from having to plan everything
Financial	**Financial**
4. I feel angry about the extra costs I have because of my child's condition	**4.** Anger about the extra money that I have to spend
5. We have less money because of my child's condition	**5.** Worrying about money because of my child's condition
Social	**Social**
6. I can't socialise as much as I would like to because of caring for my child 7. I feel sad that I see my friends less because of caring for my child	**6.** Sadness about not socialising as much as I want to because of caring for my child
8. I feel trapped in the house because of caring for my child 9. I can't do anything spontaneously anymore	**7.** Feeling trapped at home because of caring for my child
Emotional / Psychological	**Emotional / Psychological**
10. I worry about the future	**8.** Worrying about the future
11. I have to live with uncertainty	**9.** Difficulties due to feelings of uncertainty
12. I worry that my child could get very ill or die	**10.** Worrying about my child getting very ill or dying
13. I blame myself for my child's kidney problems	**11.** Blaming myself for my child's kidney problems
14. Memories of when my child was diagnosed can play on my mind	**12.** Feeling troubled by memories of when my child was first diagnosed
15. I feel unable to 'switch off' to my child's condition 16. I constantly worry about my child's condition	**13.** Feeling like I cannot 'switch off' to my child's condition
17. I worry a lot about my child during the night	**14.** Worrying about my child during the night
18. I feel very alone in caring for my child	**15.** Feeling alone in caring for my child
19. Nobody understands what I am going through 20. Other people do not understand how difficult it is for me	**16.** Feeling that others do not understand my situation

Caregiver Role / Identity	**Caregiver Role / Identity**
21. My child's kidney disease has taken over my life	**17.** Feeling that my child's condition has taken over my life
22. I am not the person that I used to be	**18.** Feeling that I am not the person that I used to be
23. I am not able to think about my own needs	**19.** Feeling that I cannot think about myself because of caring for my child
24. I have had to give up work/hobbies because of my child's illness 25. I miss doing things that I have had to give up since my child became ill 26. There are things that I cannot do because of caring for my child	**20.** Sadness about things that I have given up or cannot do because of caring for my child (e.g. work, hobbies)
Impact on Family (Practical & Interpersonal Factors)	**Impact on Family (Practical & Interpersonal Factors)**
27. I struggle to fit family life around my child's condition and treatment 28. We can't do as much as a family because of my child's condition 29. It is difficult to plan family holidays / activities as a family	**21.** Feeling overwhelmed trying to fit family life around my child's condition
30. I am not able to use my family as support because I don't trust them / they don't know enough about my child's condition	**22.** Feeling that I cannot use family / friends for support because I do not trust them to do things correctly
31. I feel that my family could support me more sometimes	**23.** Feeling that my friends / family do not support me enough
32. I argue with family members about my child's condition / treatment	**24.** Arguing with my partner / family about my child's care
33. I feel under pressure to hold the family together 34. I feel under pressure to be strong for everyone 35. I feel like I have to stay strong so that my child's needs are met	**25.** Feeling under pressure to be strong for my child / my family
36. I feel guilty that my other children are pushed to one side 37. I feel like I am always at the hospital and away from my family 38. I feel that I am not always there for my other children 39. I feel guilty about being with my child more than other family members 40. I feel torn about where I should be (e.g. hospital with child vs. home)	**26.** Feeling guilty about spending less time with my family
41. I am not able to have a normal relationship with my child	**27.** Sadness that I do not have a 'normal' relationship with my child
42. I feel bad about having to be strict with my child	**28.** Feeling guilty about having to be strict with my child
43. I am unsure how to manage my child's emotions	**29.** Struggling to manage my child's emotional difficulties
44. It is hard when my child behaves in a challenging way	**30.** Struggling to manage my child's difficult behaviour

Impact on Child	**Impact on Child**
45. I feel helpless when I see my child unwell or in pain 46. It is difficult to see my child feeling unwell	**31.** Feeling helpless when my child is ill or in pain
47. It is upsetting to see my child have medical procedures	**32.** Feeling upset seeing my child having medical procedures
48. I worry about disruptions to my child's education	**33.** Worrying about disruptions to my child's education
49. I worry about my child's growth and development	**34.** Worrying about my child's growth and development
50. Seeing my child missing out on things upsets me 51. It is hard to see my child unable to do things that other children can do	**35.** Sadness about the things that my child misses out on
52. I worry about how my child is coping	**36.** Worrying about how my child is coping
CKD treatment responsibilities	**CKD treatment responsibilities**
53. I worry about getting my child's medicines wrong 54. I worry that I may make a mistake with my child's medication	**37.** Worrying about my child's medicines being taken correctly
55. I worry about whether my child has had the right amount of fluid	**38.** Worrying about if my child has had the right amount of fluid
56. I worry about getting my child's food intake correct 57. My child's problems with eating can be overwhelming (e.g. adhering to strict dietary requirements, lack of appetite, vomiting)	**39.** Feeling overwhelmed by feeding difficulties (e.g. strict dietary requirements, lack of appetite, vomiting)
58. Being in charge of carrying out medical procedures (e.g. injections, dialysis, tube feeding) can play on my mind 59. I worry about doing medical procedures correctly	**40.** Worrying about doing medical procedures correctly (e.g. injections, dialysis, tube feeding)
60. I am not sure that I always take measurements from my child in the right way (e.g. temperature, blood pressure, weight)	**41.** Worrying about taking measurements correctly from my child (e.g. blood pressure, temperature).
61. I am always on alert for signs that my child is not well 62. I feel like I am always monitoring my child for signs of illness or infection	**42.** Feeling that I constantly check my child for signs of illness
63. I am preoccupied with making sure that everything is clean 64. I am always looking out for things that may harm my child 65. I feel that it is up to me to prevent illness or infection 66. I feel totally responsible for protecting my child from illness or infection	**43.** Feeling preoccupied with keeping my child safe from infection / illness
67. I worry that something I have done/not done may have caused harm 68. I feel that it is my fault when my chid becomes unwell 69. I feel guilty if my child's results aren't good	**44.** Blaming myself if my child gets ill or has bad results
70. I feel that I should do more for my child	**45.** Feeling that I should be doing more for my child

CKD treatment responsibilities (continued)	**CKD treatment responsibilities (continued)**
71. Adjusting to changes in my child’s treatment can be difficult 72. It is difficult having to adapt to changes in my child’s usual treatment	**46.** Feeling overwhelmed by changes in my child’s usual treatment
73. Adjusting to my child’s condition changing quickly is very difficult 74. I worry about my child getting ill suddenly 75. I can’t make any predictions about when my child is going to get ill next	**47.** Worrying about having to deal with unexpected changes in my child’s condition
76. Making decisions about my child’s care is a big responsibility 77. Having to make decisions about my child’s condition on my own is hard 78. I feel alone in making decisions about my child’s condition	**48.** Feeling overwhelmed by decisions I have to make about my child’s condition
79. I worry about my child taking responsibility for their own care	**49.** Difficulties due to my child’s own responsibilities in their care (e.g. worrying that medicine has not been taken)
Contact with hospital/medical staff: Practical & Interpersonal Factors	**Contact with hospital/medical staff: Practical & Interpersonal Factors**
80. Coming back and forth to the hospital is a strain 81. All the travelling we have to do is exhausting	**50.** Feeling exhausted from travel related to my child’s care
82. I lose a lot of my day when we are at the hospital (e.g. dialysis, waiting for bloods) 83. I feel like things take longer than they need to when I am at the hospital	**51.** Feeling frustrated when I have to wait around at hospital
84. I get very bored when we are at the hospital	**52.** Boredom from waiting around at hospital
85. I feel unable to switch off waiting to hear about test results	**53.** Feeling like I cannot ‘switch off’ when waiting for test results
86. I worry about my child getting ill suddenly and needing to stay at hospital 87. I worry about my child having to be admitted as an inpatient	**54.** Worrying that my child may be admitted to hospital
88. I worry about missing a phone call from the hospital	**55.** Worrying that I might miss an important phone call from the hospital
89. There is no privacy when we are at the hospital	**56.** Feelings of no privacy when we are at the hospital
90. It is hard for me to disagree with a member of the medical team	**57.** Holding back when I disagree with medical staff
91. I feel that I should know more about my child’s condition 92. I don’t always understand the information I am given at the hospital 93. I worry that I don’t understand what the nurses or doctors have told me	**58.** Worrying that I have not understood something that medical staff have told me
94. It is hard for me when new people are involved in my child’s care team 95. I can feel like I am the only person who really knows my child 96. It is hard for me to deal with medical staff who don’t know my child well	**59.** Feeling frustrated from having to deal with medical staff that do not know my child
97. I feel that I am not listened to by medical staff	**60.** Feeling that medical staff do not listen to me

Piloting

Pilot Sample

Version 1 of the measure was piloted with a sample of 18 caregivers and five healthcare professionals. Of the 18 caregivers, ten had participated in the interviews, and eight were a new sample of caregivers. The characteristics of the caregivers who participated in the pilot exercise (and their children) are shown in Table 4. The five healthcare professionals who participated in the pilot exercise had all participated in the interview phase of the research and included five nurse specialists in PD, HD and/or transplant.

Table 4.

Caregiver pilot sample characteristics

Caregiver		
Gender	Female	15
	Male	3
Age (years)	20-29	3
	30-39	6
	40-49	8
	50-59	1
Ethnicity	White	12
	Asian	4
	Black	1
	Mixed	1
Child		
Gender	Female	9
	Male	9
Age (years)	0-1	4
	2-5	3
	6-11	7
	12-18	4
CKD Stage/Treatment	Pre-Dialysis	4
	Peritoneal Dialysis (PD)	3
	Haemodialysis (HD)	4
	Transplant (Tx)	7
CKD Cause	CAKUT	8
	Congenital nephrosis	2
	Renovascular disease	2
	Glomerular diseases	4
	Malignancy	1
	Not specified	1
Time since diagnosis (years)	0-1 years	4
	2-5 years	6
	5-10 years	5
	>10 years	3

Readability, Usability and Item Relevance

Participants completing the pilot exercise (Appendix 17) were asked to comment on the wording of the measure and its ease of completion. No difficulties were reported in interpreting the items or completing the measure, with written and verbal feedback including: "Easy and quick to complete", "Easy to fill in", "Well set out" and "Easy to understand". Moreover, verbal and written feedback from participants indicated that the measure items were relevant to the experience of being a caregiver of a child with CKD. Also, there were very few missing items; also suggestive that the measure items were relevant and easy to complete.

Additional Item Reduction and Addition

Participants taking part in the pilot exercise completed the draft measure (Version 1) in addition to an adapted version of the measure constructed to elicit information about the perceived relevance and importance of listed items, scored on a 5-point Likert scale (ranging from *not at all* to *very*). This exercise served to provide further evidence of the relevance of items and also to inform the exclusion of any potentially redundant items. Based on the piloting exercise some adaptations were made to Version 1 of the measure (detailed in Appendix 18). Five items were deleted due to receiving negligible endorsement (e.g. 'Anger about the extra money that I have to spend') and/or feedback indicating perceived overlap with existing items. Moreover, based on feedback obtained, a number of items were merged which resulted in the overall reduction of a further 6 items from Version 1 of the measure. Moreover, on the basis of feedback from both caregivers and healthcare professionals that the measure would benefit from additional coverage of interpersonal family issues (in particular the impact upon spousal relationships and impact upon siblings), two further items were added: 'Worrying about the impact of my child's condition on my other children'; 'Sadness about the impact of my child's condition on my relationship with my partner'. The resulting 51 items and their categorisation are detailed in table 5. These items were randomised to form the 'Paediatric Renal Caregiver Burden Scale' (PR-CBS) (Figure 3) which will be evaluated further in a subsequent study.

Table 5.

Measure items and provisional domains (further to piloting exercise)

Physical

1. Worrying about the effect of caring for my child on my health
2. Feeling exhausted from caring for my child

Financial

3. Worrying about money due to the costs of my child’s care

Social

4. Sadness about not socialising as much as I want to because of caring for my child
5. Feeling trapped because of caring for my child

Emotional / Psychological

6. Worrying about the future
7. Difficulties feelings due to the uncertainty of my child’s condition
8. Worrying about my child getting very ill or dying
9. Blaming myself for my child’s kidney problems
10. Feeling troubled by difficult memories of when my child was first diagnosed or has been very ill in the past
11. Feeling like I am not able to ‘switch off’ to my child’s condition
12. Worrying about my child during the night
13. Feeling alone in caring for my child
14. Feeling that other people do not understand my situation

Caregiver Role / Identity

15. Feeling that my child’s condition has taken over my life
16. Sadness from feeling that I am not the person that I used to be
17. Feeling unable to think about my own needs
18. Sadness that I can not do things that I used to because of caring for my child (e.g. work, leisure activities, hobbies)

Impact on Family (Practical & Interpersonal Factors)

19. Arguing with my partner / family about my child’s care
20. Feeling under pressure to be strong for my child and family
21. Feeling overwhelmed by trying to fit family life around my child’s condition
22. Feeling guilty about spending less time with my child / partner
23. Sadness about the impact of my child’s kidney problems on my relationship with my partner
24. Sadness that I do not have a ‘normal’ relationship with my child
25. Feeling uncertain about how to manage my child’s emotions and difficult behaviour
26. Worrying about the impact of my child’s condition on my other children

Impact on Child

27. Feeling helpless when my child is ill or in pain
28. Worrying about disruptions to my child’s education
29. Worrying about my child’s growth and development
30. Sadness about the things that my child misses out on
31. Worrying about how my child is coping

CKD treatment responsibilities
32. Worrying about getting my child’s medicines wrong
33. Worrying if my child has had the correct amount of fluid
34. Feeling overwhelmed by feeding difficulties (e.g. lack of appetite, managing diet restrictions, vomiting)
35. Worrying about getting medical procedures wrong (e.g. dialysis, injections, tube feeding) or taking measurements incorrectly
36. Feeling preoccupied with checking my child for signs of illness
37. Feeling preoccupied with keeping my child safe from illness
38. Blaming myself if my child gets ill or has bad test results
39. Feeling that I should be doing more for my child
40. Feeling overwhelmed by changes in my child’s usual treatment
41. Worrying about having to deal with unexpected changes in my child’s condition (e.g. unexpected hospital admissions)
42. Feeling overwhelmed by decisions I have to make about my child’s condition
43. Difficult feelings due to my child taking responsibility in their care (e.g. worrying if medicines have been taken)
Contact with hospital/medical staff: Practical & Interpersonal Factors
44. Feeling frustrated when I having to spend time at the hospital
45. Feeling bored when having to spend time at the hospital
46. Feeling unable to ‘switch off’ when waiting for test results or a telephone call from the hospital
47. Worrying that my child may have to be admitted to hospital
48. Feelings of no privacy when at the hospital
49. Holding back when I disagree with medical staff
50. Worrying that I have not understood medical information
51. Frustration when dealing with staff that do not know my child

Paediatric Renal Caregiver 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)*

Discussion

The present study documents the preliminary stages of the development of a measure of paediatric CKD caregiver burden (PR-CBS), further to the established need for such a measure.

Development and content of the measure

The research was guided by the conceptualisation of caregiver burden as a multidimensional construct, primarily relating to the subjective appraisals of caregivers (rather than objective circumstances) as suggested by a number of caregiver burden researchers (Chou, 2000; Gerritsen & van der Ende, 1994; Poulshock & Deimling, 1984; Stommel et al., 1990). An extensive phase of the current research comprised the completion of a substantial number of interviews with caregivers of children with CKD and healthcare professionals, with the aim of generating an item pool relevant to a measure of caregiver burden for this caregiver population. In line with the conceptualisation of caregiver burden as a multidimensional phenomenon, the generated items were reflective of caregivers subjective appraisals across a number of domains; that is, financial, social, physical, and psychological. In addition, the content of the generated items were also reflective of caregivers subjective appraisal of the impact of their caregiving role upon the family (e.g. guilt in relation to sibling neglect), the impact of CKD and its treatment upon their child (e.g. helplessness when the child is in pain), caregiver CKD treatment responsibilities (e.g. worry about fluid intake), and issues arising from contact with the hospital/medical staff (e.g. frustration from the amount of time spent at hospital). Moreover, the caregivers subjective appraisals of changes in relation to their identity associated with the caregiver role were reflected in the items generated (e.g. sadness about loss of employment).

The content of the generated items correspond with the notion of primary and secondary stressors related to caregiving posited by Pearlin, Mullan, Semple and Skaff (1990), with primary stressors conceived of as emotions involved in caregiving itself, and secondary stressors as the emotional responses to the consequences of caregiving (i.e. indirect effects). In line with this notion of primary and secondary stressors, it has been argued that instruments developed to measure the subjective dimensions of caregiver burden should reflect distress both in providing care as well as distress resulting from the effects of caregiving on one's life (Braithwaite, 1992), which the current developed measure indeed does.

The content of the generated item pool was largely reflective of the findings of studies previously conducted in this field, which have reported poor physical health, helplessness, uncertainty, preoccupation with the future, concern about growth and development, distress arising from the impact of caregiving upon family plans and relationships, social isolation, difficulties resulting from caregiver responsibilities for CKD treatment tasks, and difficulties resulting from interactions with hospitals/medical staff (Brownbridge & Fielding 1991; Fedewa & Oberst, 1996; Hasegawa et al., 2005; Hulstijn-Dirkmaat & Damhuis, 1994; Reynolds, et al., 1988; Tong et al. 2008; Tong, et al. 2010; Watson et al., 1997; Weidebusch et al., 2010; Wolters, et al. 1980).

Involvement of caregivers

The present research benefits from the intensive input of caregivers throughout the development of the PR-CBS; thus aiding attempts to ensure high face and content validity of measure items. Weitzner, Jacobsen, Wagner, Friedland and Cox (1999) report that the development of caregiver burden scales to date largely lack the direct input of caregivers during the item pool generation stage, which is a particular oversight when considering caregiver burden as a subjective paradigm. Consultation with relevant professionals is a common component of the item generation phase of measure development, and indeed proved to be a valuable source of information in the present study. However, much research in this field has tended to rely solely on the input of (often a small number of) relevant professionals to generate an initial item pool. In the present study the first hand accounts of caregivers of children with CKD were used to inform the generation of an initial comprehensive item pool; in addition, caregivers participated in a piloting exercise, with their feedback on the usability, readability and content of measure items incorporated accordingly into the resulting measure (i.e. Version 2; the PR-CBS).

A measure specific to paediatric CKD

The impetus for the current research was the lack of a measure of caregiver burden specifically developed for caregivers of children with CKD, with measures of caregiver burden in existence largely developed for caregivers of older adults or those with mental health diagnoses (Robinson, 1983; Schene, Tessler & Gamache, 1994; Visser-Meily, Post, Riphagen & Lindeman, 2004). The application of nonspecific burden measures to different populations of caregivers has hindered progress in caregiver burden research (Chou, 2000); it is encouraging that disease-specific measures of the impact of caregiving are increasingly being developed (e.g. Sepúlveda, Whitney, Hankins & Treasure, 2008). It is hoped that the development of the current measure will serve to enhance an understanding of caregiver burden in the context of paediatric CKD.

Limitations

While the current study benefits from the inclusion of caregivers of children across the whole trajectory of CKD, it should be acknowledged that in the interview sample there were slightly fewer caregivers of children who were currently pre-dialysis or receiving PD, compared to those of children currently on HD or post-transplant. However, this was not deemed to compromise the generation of items relevant to caregivers of children pre-dialysis or receiving PD, as many caregivers discussed their experiences of a variety of CKD treatment modalities, past and present, during the interviews (e.g. nine of the 16 caregivers interviewed relayed experiences of caring for a child on PD). Moreover, interviews were conducted with healthcare professionals specialising in the care of children across the trajectory of CKD, and indeed saturation of the item pool was deemed to have been achieved in relation to issues specific to the various CKD modalities available.

The caregiver sample for both the interview and piloting exercise was predominantly comprised of mothers. Male caregivers are largely under-represented in CKD caregiver research (Aldridge, 2008); just two of the 16 caregivers interviewed in the current study were fathers. Previous research has reported some differences in the psychosocial outcomes of mothers compared to fathers of children with chronic illness (e.g. Noll et al., 1995; Sloper, 2000) and it could therefore be speculated that the issues pertinent to the experience of caregiver burden amongst fathers relative to mothers of children with CKD may differ. It should be acknowledged however, that it is much more common for mothers than fathers to assume the role of primary family caregiver (Brown 2008; Coffey, 2006).

Moreover, only two of the caregivers interviewed were part of single-parent families (both single mothers); single-parent families are under-represented in the study of caregivers of children with chronic illness (Brown 2008). As such, there may be issues of relevance to the experience of caregiver burden in single parents of children with CKD which have not been captured adequately in the current research. (It has been suggested that single-parent families of children with chronic illness may be those who are most at risk of caregiver burden; Brown, 2008).

A further limitation of the present study is the lack of diversity in the ethnicity of the caregiver sample. As the sample was predominantly white, it is possible that the generated item pool may not be sufficiently generalisable to the variety of minority ethnic groups present in the paediatric CKD caregiver population. Furthermore, while efforts were made to enable those who do not speak English as a first language to participate in the study, the resources to provide translation services were not available; two of the 16 caregivers interviewed in the current study did not speak English as a first language. While the developed measure contains an item reflecting worry related to poor understanding of medical staff, it should be acknowledged that issues relevant to caregiver burden arising from communication difficulties may be exacerbated amongst those who cannot speak English as first language; this may particularly true in the context of a highly medicalised setting and the complex CKD treatment regimen.

It was not within the scope of the current research study to provide more than a preliminary evaluation of the psychometric properties of the developed measure. While it could be argued that the completion of the initial phases of the development of this measure (i.e. item generation) were somewhat laboured, it is evident that some researchers who have developed measures of caregiver burden have not invested in the care required during the item generation phase; arguably the most critical phase of measure development (Hinkin, 1995). Establishing the content validity of a new measure is regarded as the minimum psychometric requirement for measurement adequacy (Schriesheim, Powers, Scandura, Gardiner, & Lankau, 1993).

Future Directions

The study of caregiver burden amongst carers of family members with CKD is largely undeveloped, particularly compared to the study of caregivers of relatives with dementia and cancer (Teixido et al., 2006). La Greca and Lemanek (1996) have previously noted a general lack of relevant, reliable and valid assessment tools for ill children and their families. The present study documents the initial phases of the development of a measure of caregiver burden, specific to paediatric CKD. A subsequent research project is scheduled to be completed to evaluate the psychometric properties of this measure. It is essential that a newly developed measure can demonstrate reliability, validity and acceptability for use amongst its target population; the current proposed aims of the subsequent research study are to provide an evaluation of the internal consistency reliability, factor structure, and convergent validity of the newly developed PR-CBS. To achieve these aims, the subsequent study will be completed with a larger sample (100 participants or above) across at least three children's hospitals in the UK. In order to evaluate the internal consistency of the measure, Cronbach's alpha coefficients will be calculated; to evaluate the factor structure of the measure, exploratory factor analysis will be completed (it is worth noting that Chou (2003) notes that factor analysis provides important information on the dimensionality of burden); to explore the convergent validity of the measure, caregivers will complete the newly developed measure alongside an existing measure of caregiver burden and/or measures of caregiver outcomes which have previously been found to correlate with caregiver burden (e.g. measures of anxiety and/or depression). To evaluate the acceptability of the new measure, qualitative data to gain insight into caregiver's views about the acceptability of the new measure will be collected.

It is hoped that the multidimensional nature of the PR-CBS will lend itself to the identification of areas for targeted intervention; according to Chou (2003), "multidimensional measures of caregiver burden give a sensitive reading of caregivers' feelings and a sophisticated picture of caregivers' response to the demands of care" (p.80). It is hoped that the newly developed PR-CBS will serve to enhance an understanding of caregiver burden in this population, and moreover, serve as a tool to aid the identification of caregivers who may benefit most from support intervention. The identification of those who may be most in need of additional support (and thus potentially compromised in their caregiving duties), may be particularly important in light of advances in treatment, with care increasingly shifting into the family home (Gayomali, 2008). Assessment is after all, a necessary preliminary to intervention (Cousins et al., 2002). There is currently very limited research exploring support interventions for caregivers of individuals affected by CKD (Tong, Sainsbury & Craig, 2008). It is hoped that the PR-CBS will be a useful tool in future studies which aim to explore the development and evaluation of support interventions in this caregiver population.

Conclusion

In the UK, the National Health Service (NHS) views the experiences of caregivers as a priority (Department of Health, 2004). It has been increasingly argued that family caregivers of children with CKD should be considered as a target of intervention, with a view to improving outcomes for these caregivers, and ultimately their children (Tong et al., 2008). This is the first study which has sought to develop a measure of caregiver burden specific to caregivers of children with CKD; this study benefits from the completion of a large number of interviews with family caregivers and healthcare professionals in order to inform the content of the measure. Moreover, this study is an example of a collaborative research relationship between nephrologists and clinical psychologists. It is hoped that use of the PR-CBS in future research will enhance an understanding of caregiver burden in the paediatric CKD caregiver population, and ultimately serve as a tool in efforts to improve outcomes for caregivers of children with CKD, as well as their children.

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SECTION C

Critical Appraisal

WORDCOUNT: 1994

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

This was my first experience of designing and conducting a research project that aimed to develop a new measurement tool. As such, I have learnt about the stages which should ideally be completed during the development of a new measure, and in doing so have developed skills in the design and execution of each of these stages. For example, I have learnt about methods which are most suited to ensuring the generation of an item pool which has face and content validity, and I have also learnt about the process of reducing a generated item pool where required (and the criteria upon which such decisions should be based). Moreover, I have developed further skills in reviewing and critiquing the existing literature in a given field to inform the most valuable design of ongoing research (e.g. in this instance, ensuring that the caregiver sample were an active component of the item generation stage).

Completion of this research project has also equipped me with core skills in the design of materials commonly required when undertaking research (i.e. information sheets, study posters, consent forms, interview schedule). Moreover, completion of this study has improved my confidence in making decisions regarding the most appropriate methods of sampling and recruitment. Through the completion of this project I have also developed further knowledge and skills required for obtaining successful ethical and local research governance approval and feel that I will be much less daunted when approaching this task in the future.

Due to the nature of this research project, which involved active engagement with family caregivers and healthcare professionals to inform the development of a new measure, I have learnt about the value of consulting with the research population during various stages of research. I have learnt skills in liaison and consultation with service-users for the purposes of research, and have developed skills in the incorporation of feedback gained through consultation and piloting exercises. I plan to carry forward this increased awareness of the value of consulting with the target research population when undertaking research in the future; the importance of service-user consultation was highlighted as an area of importance in the Research Governance Framework for Health and Social Care (Department of Health, 2001). The completion of this research project has also enabled me to grow in confidence when collaborating with professionals across a variety of disciplines for the purposes of research.

Overall, while I feel that I have developed a variety of research skills during the completion of this project (including those required for carrying out interviews for the purposes of developing a new measurement tool), I am keen in the future to develop a working knowledge of methodologies which may be most suited to other research aims (e.g. Interpretative Phenomenological Analysis).

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

While interview participants were aware of the purpose of the scheduled interviews, it may have been beneficial to create more opportunities for participants to reflect on issues relevant to caregiving for a child with CKD prior to the interviews taking place; Fischer (2009) recommends asking participants to give a written account of experiences prior to interview, to give participants time to reflect on their experience. Despite the lack of encouragement for participants to reflect on their experiences before interviews were conducted, the interviews were however felt to be rich in their content, and I feel that the items generated were a valid representation of the experiences of this caregiver population.

The completion of individual interviews comprised a substantial phase of the research project; this was a lengthy process. On reflection, I wonder if there may have been alternative, less 'labour-intensive' methods of generating potential measure items for the measure. For example, focus groups have previously been utilised to inform the generation of items for a new measure. However, based on the interviews conducted I feel that the richness of the content of the generated item pool may have been compromised if focus groups had been used. For example, some caregivers described during interviews that it could be challenging at times to acknowledge and discuss difficult feelings in relation to providing care for your own child, and moreover to admit that you may be having difficulties in fulfilling your caregiver duties. Alternatively, it may have been feasible to have relied more on the relevant previous literature to inform the content of the measure. However, while there is an existing body of research exploring the experiences of caregivers of children with CKD, the current research project was the first study to date which has been conducted specifically with the aim of developing a measure of paediatric CKD caregiver burden. As such, relying solely on the findings of previously conducted research (which have had a variety of competing aims), may have compromised the validity of the current research. Overall, while conducting focus groups or relying more heavily on previously conducted literature may have meant that the progress of the development of the measure would have been quicker, I believe that the absence of individual interviews with caregivers caring for children across the trajectory of CKD and healthcare professionals spanning a number of specialities, would have been a major omission; and indeed an error, which I believe, has compromised the validity of much of the caregiver burden measurement studies conducted to date.

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

Prior to conducting this project, I already had an existing interest in the impact of chronic illness upon affected individuals. Completion of this project has given me the opportunity to gain insight into the impact of caring for individuals affected by chronic illness, particularly from the perspective of family caregivers of children with a chronic illness. In the future I am keen to gain experience of working in services which provide support to children affected by chronic illness or disability and their families, and as such I feel that this project has provided me with a broad awareness and understanding of a range of issues which are pertinent to this caregiver population. While this research was specific to the experience of caring for a child with a chronic illness, I feel that completing this study will have highlighted issues that are relevant to caregivers of individuals affected by chronic illness across the life span. When working as a clinical psychologist it is very likely that I will work with individuals who provide care to family members affected by physical illness or disability. In the UK, 1 in 8 adults are carers, with the organisation 'Carers UK' noting that "625,000 people suffer mental and physical ill health as a direct consequence of the stress and physical demands of caring" (www.carersuk.org). The experience of completing this research project with therefore serve to enhance my clinical skills in capturing information which may be relevant to the assessment, formulation and subsequent intervention with caregivers of individuals affected by chronic illness or disability.

It was apparent during the completion of this research that caregivers were keen for their voices to be heard, with a number of caregivers citing this as the reason for their participation in the research – to raise awareness of the difficulties that they face. It is evident that the support needs of informal caregivers are often unaddressed, and consequently remain unmet (particularly in the context of physical healthcare settings, where the medical needs of individuals affected by chronic illness are often prioritised). In light of this, I will aim to carry forward an increased awareness of the potential needs of this 'invisible but invaluable' population in my future clinical work, particularly if I am working in services based within a physical healthcare setting. I feel that addressing and attending to the support needs of family caregivers is particularly important in light of the increasing body of research which highlights an association between improved outcomes for caregivers and improved outcomes for care recipients.

4. 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?

During the completion of interviews I was struck by the resilience of the caregivers that I spoke to, often in the face of ongoing challenges in fulfilling their role as caregivers, and with the provision of very limited support. The purpose of the current research was to develop a measure of caregiver burden, not only with a view of assisting the identification of caregivers who may be having difficulties, but ultimately with a view to providing additional support and intervention. There is currently however very limited research exploring support interventions for caregivers of individuals affected by CKD (Tong, Sainsbury & Craig, 2008); as such, an established need exists for the development and evaluation of services that respond to the support needs of caregivers of individuals with CKD (Tong et al., 2008).

A number of the caregivers noted during interviews that they had benefited hugely from developing friendships and maintaining ongoing contact with other family caregivers of children with CKD (friendships often established through informal introductions in clinic waiting rooms). In addition, a number of the caregivers of children who had received a kidney transplant noted that they had valued attendance at the annual 'British Transplant Games' and had particularly valued the opportunity to share experiences with other caregivers of children post-transplant. On the flip side, one mother of a child who was currently receiving HD described how she had no contact with other caregivers of children with CKD and felt that during previous attempts to maintain her child on PD at home (which was subsequently unsuccessful) that she would have benefited from contact with a caregiver with experience of caring for a child on PD.

Therefore, I feel that there is a paediatric CKD caregiver virtual community in existence at the hospital where I completed this project, who may benefit from the establishment of a formal network to facilitate access to each other in order to provide a forum for support and resources (e.g. an online forum, a caregiver mentoring system, etc). The possible benefits of establishing a support network for caregivers of children with CKD, and the suggestion that this may be something which may meet some of the support needs of this caregiver population is obviously speculative, and based purely on anecdotal indicators from interviews conducted as part of the current study. Therefore, a future line of research which I would be interested in would comprise a study to explore the perceived support needs of caregivers of children with CKD; and ultimately to further explore the types of support interventions which this caregiver population perceive that they require and would gain most benefit from.

Tong et al. (2008) have previously suggested the use of participatory action research methods to extend beyond an understanding of the needs and perspectives of informal caregivers; this 'empowering methodology' allows participants to be involved in the planning, implementation and evaluation of interventions, and participants essentially become co-researchers. Based on the completion of the current research, I particularly valued the opportunity to engage with the caregiver population in consultation and piloting exercises, and to incorporate their feedback accordingly into the measure that was developed. Equally, I feel that the caregiver participants valued the opportunity to be an integral component of the development of materials which, after all, were intended to be reflective of their experiences. As such, if I was to conduct a future research project to explore the perceived support needs of this caregiver group, I would use a similar design comprising largely a qualitative design, whereby caregivers would be interviewed as an in-depth exploration of their perceived support needs, with a view to using this information to inform the content of future potential support interventions.

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SECTION D

Appendices

Appendix 1: Literature Search Methodology

A literature search was conducted through the MEDLINE(R) (1950-2010), PsycINFO (1806-2010), and EMBASE (1980 – 2010) databases up to March 20th 2011. To increase the relevance of the articles retrieved, terms denoting the possible negative effects of caregiving were included (e.g. stress, strain, burden). Some of the search terms were specified to appear within the abstracts of articles (denoted by 'ab.' in the search strategy), as outlined below:

Search Strategy:

child$.ab. *or* adolescen$.ab. *or* paediatric$.ab. *or* pediatric$.ab.

and

parent$.ab. *or* mother$.ab. *or* father$.ab. *or* carer$.ab. *or* caregiver$.ab.

and

chronic kidney disease$.ab. *or* CKD.ab. *or* kidney disease$.ab *or* renal disease$.ab

or

predialysis$.ab. *or* kidney failure$.ab. *or* renal replacement therap$.ab. *or*
peritoneal.ab. *or* haemodialysis$.ab. *or* hemodialysis.ab. *or*. kidney transplant$.ab.

and

stress$.mp. *or* strain$.mp. *or* burden$.mp. *or* mental health$.mp.
or psycholog$.mp. *or* emotion$.mp. *or* social.mp.

The search resulted in 276 articles which were reduced to 174 upon removal of duplicate articles. The search criteria were limited further to articles published in the English language, resulting in 138 articles. As the healthcare paradigm shift to home-based care provided by family members is largely limited to the last three decades, the publication date of articles was limited to 1980 onwards. The abstracts of the 129 identified studies were examined and articles were retained for inclusion in the review if they met the following criteria:

1. Experiences of caregivers of children with CKD was explored
2. Published in a peer-reviewed journal

Once these criteria were applied, 21 articles were retained for inclusion in the review. Examination of the reference lists of these articles resulted in the inclusion of 3 further studies.

Appendix 2: Summary table of Tong et al. (2008) review of qualitative studies

Author (Year)	**Caregiver Experiences Explored**	**Predialysis** *n*	**PD** *n*	**HD** *n*	**Transplant** *n*	**Total no. of patients**	**Interview/ Focus Group**
Korsch et al (1973)	Psychosocial attributes and impact on long-term rehabilitation; family functioning	0	0	0	35	35	Semistructured
Raimbult (1973)	Psychological issues	30	0	39	0	69	In-depth
Sampson (1975)	Social and emotional adaptation	0	0	8	14	22	Interview
Crittenden et al (1977)	Coping with stress of treatments	0	0	0	17	17	Interview
Klein and Simmons (1979)	Impact of kidney disease and transplant	—	—	—	—	65	Semistructured
Hislop and Lansing (1983)	Comparing home PD modalities	0	4	0	0	4	Semistructured
Reichwald-Klugger et al (1984)	Psychosocial adaptation	0	0	20	0	20	In-depth/ Semistructured
Waissman (1990)	Doctor–parent interactions; family relationships and social life	0	11	4[b]	0	15	Semistructured
Obrecht et al (1992)	Managing child's care	0	0	0	1	1	In-depth
MacDonald (1995)	Meaning assigned to child's illness by mothers	0	4	0	0	4	In-depth
Middleton (1996)	Psychosocial issues	0	8	0	0	8	Focus group
Schultz and Farrell (1998)	Everyday experience and attributed events and meanings	4	0	0	6	10	In-depth
Nicholas (1999)	Experiences and meanings attributed to maternal caregiving	0	14	10	8	32	In-depth
Karrfelt et al (2000)	Decision-making about donation	0	0	0	18	18	Semistructured
Baines et al (2001)	Parent donor and child recipient relationship	0	0	0	7	7	Semistructured
Cimete (2002)	Stress factors; coping strategies	0	0	31	0	31	Focus group

Appendix 3: Caregiver Interview Schedule

1. Age of child now: _____years______months

2. Age of child when first diagnosed: _____years_____months

3. Treatment child is receiving at the moment: _______________________________

4. i) Can you tell me about the main difficulties of caring for your child?

5. ii) Can you tell me about the most difficult parts of their current treatment? [unprompted at first]

<u>Prompts</u>:

- o *Practical issues / Treatment related*
  [e.g. hospital attendance, managing medications, monitoring dietary/fluid restrictions]

- o *Impact upon family life; relationships between carer and partner/ other children*
  [e.g. disruption of family norms, strain on relationship with partner, loss of time with other children]

- o *Impact upon social and leisure activities*
  [e.g. restrictions on social/leisure activities, isolation]

- o *Impact upon finances/ financial support received* (if carer open to discussing this)

- o *Physical effects of providing care*
  [e.g. fatigue]

- o *Psychological effects of providing care/ having a child with CKD*
  [e.g. stress, low mood, living with uncertainty]

- o *Impact of CKD upon child*
  [e.g. education, friendships, emotional and behavioural issues]

- o *Issues relating to involvement with hospital staff*
  [e.g. 'surrendering' control of child to staff, communication]

- o *Other: anything else that you feel is important that we haven't discussed yet?*

6. How does your child's current treatment compare with previous treatments (if applicable)? Is this more or less difficult, and in what ways?

7. Can you tell me about the ways that you cope with the demands of caring for your child? What is most helpful?

[Thank participant for their time. Interview could be emotive - check how parent is feeling and debrief where necessary. Provide information about how researcher can be contacted if any other issues arise that they want to discuss / where to access support].

Appendix 4: Healthcare Professional Interview Schedule

1. Can you tell me what you feel are the main difficulties which a family carer may experience when caring for a child with CKD? [unprompted at first]

   <u>*Prompts*</u>:
   - *Practical issues / Treatment related*
     [e.g. hospital attendance, managing medications, monitoring dietary/fluid restrictions]

   - *Impact upon family life; relationships between carer and partner/ other children*
     [e.g. disruption of family norms, strain on relationship with partner, loss of time with other children]

   - *Impact upon social and leisure activities*
     [e.g. restrictions on social/leisure activities, isolation]

   - *Impact upon finances/ financial support received* (if carer open to discussing this)

   - *Physical effects of providing care*
     [e.g. fatigue]

   - *Psychological effects of providing care/ having a child with CKD*
     [e.g. stress, low mood, living with uncertainty]

   - *Impact of CKD upon child*
     [e.g. education, friendships, emotional and behavioural issues]

   - *Issues relating to involvement with hospital staff*
     [e.g. 'surrendering' control of child to staff, communication]

   - *Other: Is there anything else that you feel is important that we haven't discussed yet?*

2. What aspects of each of the different treatment modalities do you think are most difficult for carers?

   - pre-dialysis
   - peritoneal dialysis
   - haemodialysis
   - post-transplant

3. Do you think these difficulties are dependent on the child's age? In what ways?

4. In what ways do you think carers cope with the demands of caring for their child?

[*Thank participant for their time. Provide information about how researcher can be contacted if any other issues arise that they want to discuss*].

Appendix 5: Ethical Approval

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Appendix 6: Local Research Governance Approval

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Appendix 7: Declaration of end of study (Ethics)

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Appendix 8: Final Report (Ethics)

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Appendix 9: Summary Report (Participants)

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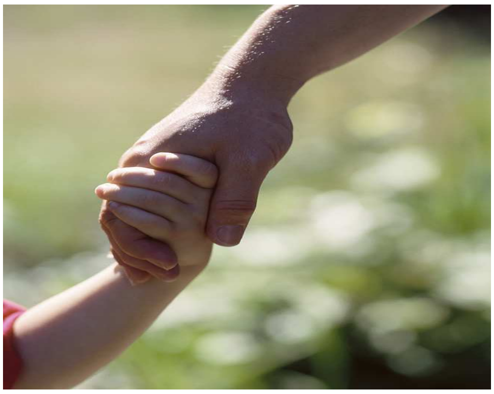
Appendix 10: Study Poster

Canterbury
Christ Church
University

Are you a parent or carer of a child or teenager with kidney disease?



Research study: Development of a measure of caregiver burden in chronic kidney disease (Phase 1)

I am inviting parents and carers of children and teenagers who receive treatment at the Renal Unit at ___________ Hospital to take part in a research study. The aim of the study is to find out more about the difficulties of caring for a child with kidney disease.

Taking part in this study will involve being interviewed for about 20-30 minutes about caring for a child with kidney disease. Interviews will take place at the Renal Unit, or over the phone if you prefer – at a time which is convenient for you.

For more information please pick up one of the information sheets which are in the waiting room. If you would like to know any more about this study or if you would like to take part please contact me on: rsp3@canterbury.ac.uk
or tel: 01892 507673 (please state that the message is for Rhian Parham and leave your contact details).

Many thanks, Rhian Parham

(Trainee Clinical Psychologist,
Canterbury Christ Church University)

Canterbury
Christ Church
University

Parent/Carer Information Sheet

Research study: Development of a measure of caregiver burden in chronic kidney disease

Are you a parent or carer of a child or teenager with kidney disease?

You are being invited to take part in a research study. Before you decide to take part it is important that you understand why this study is being done and what it will involve.

Background

My name is Rhian Parham - I am a Trainee Clinical Psychologist at Canterbury Christ Church University and I am carrying out this research as part of my training. This study will be supervised by two supervisors at the University, as well as Dr _______ and Dr _____ who work at the Renal Unit at ____________ Hospital. This study has received approval from an NHS ethics committee.

What is this study about?

Previous research has shown that caring for a child with kidney disease can be demanding at times. This study aims to explore the experiences of family carers of children with kidney disease.

Why have I been invited?

We are inviting family carers of children who are being treated for kidney disease at __________ Hospital to take part in this study.

What are the possible benefits?

It is hoped that the study will lead to a better understanding of the experiences of family carers of children with kidney disease. The results of this study will allow us to develop a questionnaire to measure the most important issues for family carers. We hope in the future that this questionnaire will be used to assess the experiences of caring for a child with kidney disease.

What will happen if I take part in the study?

If you decide to take part in this study I will interview you about your experiences of caring for your child. Interviews will take place at ____________ Hospital at a time which is convenient for you and will last about 20-30 minutes. I will interview you about the things that you can find difficult about caring for your child and also about what has been helpful. You do not have to talk about anything which you do not feel comfortable talking about. Recordings of interviews will be copied straight to password protected CDs and then deleted from the recording equipment. The interviews will then be written up into documents which will be stored securely. All interviews will be given an ID number so your name will not be linked with the interview in any way. All information collected is strictly confidential, unless there are any concerns about yourself or your child - this will be discussed with you if this is the case.

Do I have to participate?

No, you do not have to participate in the study. It is completely optional and taking part or choosing not to take part will not affect your care or your child's care in any way.

What happens with the results?

The findings of this study will be written up and submitted to Canterbury Christ Church University as part of my doctoral training in clinical psychology. It is also hoped that the findings will be published in a research journal. You will also be sent a summary report if you would like to a copy.

What now?

If you have any more questions about this study or you have decided that you would like to take part, please feel free to speak to me in person when I am at the hospital, or **email: rsp3@canterbury.ac.uk** or **tel: 01892 507673** (please state that the message is for Rhian Parham and leave your contact details).

Many thanks, Rhian Parham (Trainee Clinical Psychologist)

Thank you for taking the time to read this information sheet.
Your help in this research would be greatly appreciated.

Canterbury
Christ Church
University

Staff Information Sheet

Research study: Development of a measure of caregiver burden in chronic kidney disease

You are being invited to take part in a research study. Before you decide to take part it is important that you understand why this study is being done and what it will involve.

Background

My name is Rhian Parham - I am a Trainee Clinical Psychologist at Canterbury Christ Church University carrying out this study as part of my doctoral training. This study will be supervised by two supervisors at the University as well as Dr Hothi and Dr Marks who work at the Renal Unit at ___________Hospital. This study has received approval from an NHS ethics committee (approval number: ___________).

What is this study about?

Previous research has shown that caring for a child with kidney disease can be demanding at times. This study aims to explore the experiences of family carers of children with kidney disease. As part of this study I am also conducting interviews with staff at that Renal Unit.

Why have I been invited?

I am inviting staff at the Renal Unit who work with children who are being treated for kidney disease at __________ Hospital to take part in interviews.

What are the possible benefits?

It is hoped that the study will lead to a better understanding of the experiences of carers of children with kidney disease. We plan to develop a questionnaire to measure the most important issues for carers of children with kidney disease – we hope in the future that this questionnaire will be used to assess the experiences of caring for a child with kidney disease.

What will happen if I take part in the study?

If you decide to take part in this study I will arrange interviews to take place at the Renal Unit at a time which is convenient for you; interviews will last approximately 20-30 minutes. Recordings of interviews will be copied straight to password protected CDs and then deleted from the recording equipment. The interviews will then be written up into documents which will be stored securely. All interviews will be given an ID number so your name will not be linked with the interview in any way. All information collected is strictly confidential.

Do I have to participate?

No, you do not have to participate in the study. It is completely optional and your decision to participate or not in no way relates to your position at the hospital.

What happens with the results?

The findings of this study will be written up and submitted to Canterbury Christ Church University as part of my training. It is hoped that the findings will be published in a research journal. The Renal Unit at __________ Hospital will receive a summary report of the findings.

What now?

If you have any more questions about this study or you have decided that you would like to take part, please feel free to speak to me in person when I am at the hospital, or contact me on **rsp3@canterbury.ac.uk** or **tel: 01892 507673** (please state that the message is for Rhian Parham and leave your contact details).

Many thanks, Rhian Parham (Trainee Clinical Psychologist)

Thank you for taking the time to read this information sheet.
Your help in this research would be greatly appreciated.

Appendix 13: Parent/Carer Consent Form

Canterbury
Christ Church
University

Date: ………………………

Please read the statements below and tick where appropriate:

1. I confirm that I have read and understand the information sheet (dated 15.05.10) and that I have had an opportunity to ask questions. [ ]

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

3. I consent to a recording of the interview being made. [ ]

4. I consent to anonymous quotes from my interview being used in the write up of this study with my permission. [ ]

Participant Name: ………………………………………………

Signature: ………………………………………………………….

Researcher Name: ………………………………………………

Signature: ………………………………………………………….

Appendix 14: Healthcare Professional Consent Form

Canterbury
Christ Church
University

Date: ……………………..

Please read the statements below and tick where appropriate:

1. I confirm that I have read and understand the information sheet (dated 15.05.10) and that I have had an opportunity to ask questions. [ ]

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

3. I consent to a recording of the interview being made. [ ]

4. I consent to anonymous quotes from my interview being used in the write up of this study with my permission. [ ]

Participant Name: ………………………………………………
Signature: …………………………………………………………..

Researcher Name: ………………………………………………
Signature: …………………………………………………………..

Appendix 15. Example interview transcript

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Appendix 16: Version 1 of the measure

CAREGIVER QUESTIONNAIRE – CHILD CKD

Below is a list of things that parents of children with kidney disease have said can be a 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.

How much of a difficulty has this been for you during the last month:

	Never	**Rarely**	**Sometimes**	**Often**	**Always**
Worrying about my child's medicines being taken correctly	**1**	**2**	**3**	**4**	**5**
Arguing with my partner / family about my child's care	**1**	**2**	**3**	**4**	**5**
Holding back when I do not agree with medical staff	**1**	**2**	**3**	**4**	**5**
Worrying about my child during the night	**1**	**2**	**3**	**4**	**5**
Feeling under pressure to be strong for my child / family	**1**	**2**	**3**	**4**	**5**
Worrying that my child may get very ill and need to be admitted to hospital	**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**
Feelings of no privacy when we are at the hospital	**1**	**2**	**3**	**4**	**5**
Worrying about my child's growth and development	**1**	**2**	**3**	**4**	**5**
Feeling that my family / friends do not support me enough	**1**	**2**	**3**	**4**	**5**
Feeling like I cannot 'switch off' to my child's condition	**1**	**2**	**3**	**4**	**5**
Feeling overwhelmed trying to fit family activities (e.g. leisure, holidays) around 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 overwhelmed by changes in my child's usual treatment	**1**	**2**	**3**	**4**	**5**
Feeling exhausted from travel related to my child's care	**1**	**2**	**3**	**4**	**5**

How much of a difficulty has this been for you during the last month

	Never	Rarely	Sometimes	Often	Always
Feeling upset seeing my child having medical procedures	1	2	3	4	5
Worrying about disruptions to my child's education	1	2	3	4	5
Feeling frustrated when I have to wait around at hospital	1	2	3	4	5
Feeling exhausted from caring for my child	1	2	3	4	5
Feeling overwhelmed by feeding difficulties (e.g. strict dietary requirements, lack of appetite, vomiting)	1	2	3	4	5
Difficulties due to my child's own responsibilities in their care (e.g. not trusting my child to take medicines)	1	2	3	4	5
Boredom from waiting around at hospital	1	2	3	4	5
Blaming myself if my child gets ill or has bad results	1	2	3	4	5
Feeling that others do not understand my situation	1	2	3	4	5
Worrying about taking measurements correctly from my child (e.g. blood pressure, temperature).	1	2	3	4	5
Worrying about if my child has had the right amount of fluid	1	2	3	4	5
Feeling helpless when my child is ill or in pain	1	2	3	4	5
Feeling like I cannot 'switch off' when waiting for test results	1	2	3	4	5
Worrying that my child may be admitted to hospital	1	2	3	4	5
Feeling that my child's condition has taken over my life	1	2	3	4	5
Blaming myself for my child's kidney problems	1	2	3	4	5
Worrying about my child getting very ill or dying	1	2	3	4	5
Worrying about the effect of caring for my child on my health	1	2	3	4	5
Feeling overwhelmed trying to fit family life around my child's condition	1	2	3	4	5
Feeling alone in caring for my child	1	2	3	4	5
Worrying about how my child is coping	1	2	3	4	5
Feeling guilty about having to be strict with my child	1	2	3	4	5
Worrying about doing medical procedures correctly (e.g. injections, dialysis, tube feeding)	1	2	3	4	5

How much of a difficulty has this been for you during the last month:

	Never	Rarely	Sometimes	Often	Always
Worrying about the future	**1**	**2**	**3**	**4**	**5**
Feeling exhausted from having to plan everything	**1**	**2**	**3**	**4**	**5**
Worrying about money because of my child’s condition	**1**	**2**	**3**	**4**	**5**
Feeling overwhelmed by decisions I have to make about my child’s condition	**1**	**2**	**3**	**4**	**5**
Feeling frustrated from having to deal with medical staff that do not know my child	**1**	**2**	**3**	**4**	**5**
Feeling that I constantly check my child for signs of illness	**1**	**2**	**3**	**4**	**5**
Feeling troubled by memories of when my child was first diagnosed	**1**	**2**	**3**	**4**	**5**
Feeling that medical staff do not listen to me	**1**	**2**	**3**	**4**	**5**
Difficulties due to feelings of uncertainty	**1**	**2**	**3**	**4**	**5**
Feeling trapped because of caring for my child	**1**	**2**	**3**	**4**	**5**
Worrying that I might miss an important phone call from the hospital	**1**	**2**	**3**	**4**	**5**
Feeling that I cannot think about myself because of caring for my child	**1**	**2**	**3**	**4**	**5**
Worrying that I have not understood something that medical staff have told me	**1**	**2**	**3**	**4**	**5**
Sadness that I do not have a ‘normal’ relationship with my child	**1**	**2**	**3**	**4**	**5**
Struggling to manage my child’s emotional difficulties	**1**	**2**	**3**	**4**	**5**
Feeling that I cannot use family / friends for support because I do not trust them to do things correctly	**1**	**2**	**3**	**4**	**5**
Feeling guilty about spending less time with my partner / family	**1**	**2**	**3**	**4**	**5**
Struggling to manage my child’s difficult behaviour	**1**	**2**	**3**	**4**	**5**
Feeling that I am not the person that I used to be	**1**	**2**	**3**	**4**	**5**
Anger about the extra money that I have to spend	**1**	**2**	**3**	**4**	**5**
Sadness about things that I have given up or cannot do because of caring for my child (e.g. work, hobbies)	**1**	**2**	**3**	**4**	**5**
Feeling that I should be doing more for my child	**1**	**2**	**3**	**4**	**5**

Appendix 17: Piloting Exercise

RESEARCH PROJECT

Development of a measure of kidney disease caregiver burden

Dear Parent/Carer,

My name is Rhian Parham - I am a Trainee Clinical Psychologist at Canterbury Christ Church University and I am carrying out a research project at _________ Hospital as part of my training. The aim of this research is to develop a questionnaire to measure the experiences of caring for a child with kidney disease.

During the first part of this project, parents/carers were interviewed about their experiences of caring for their child. For the next part of this project, we are asking parents and carers for their views on a questionnaire which was developed from these interviews.

Please complete parts 1 and 2 of the questionnaire over the page.

This should take about 10-15 minutes.

This study is being supervised by two supervisors at Canterbury Christ Church University, as well as Dr ______ and Dr ______ who work at the Renal Unit at _________Hospital. This study has received approval from an NHS ethics committee.

PART 1: CAREGIVER QUESTIONNAIRE

Please can you first answer the following questions:

1. How old is your child? [_________]

2. How old was your child when they were diagnosed with kidney problems? [_________]

3. What is the cause of your child's kidney problems (if known)?
[____]

4. What treatment does your child *currently* receive? [please tick as appropriate]:
Medication [ ]
Peritoneal Dialysis [ ]
Haemodialysis (at hospital) [ ]
Haemodialysis (at home) [ ]
Post-transplant [ ]

Below is a list of things that parents of children with kidney disease have said can be a 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:

How much of a difficulty has this been for you during the last month:

	Never	**Rarely**	**Sometimes**	**Often**	**Always**
Worrying about my child's medicines being taken correctly	**1**	**2**	**3**	**4**	**5**
Arguing with my partner / family about my child's care	**1**	**2**	**3**	**4**	**5**
Holding back when I do not agree with medical staff	**1**	**2**	**3**	**4**	**5**
Worrying about my child during the night	**1**	**2**	**3**	**4**	**5**
Feeling under pressure to be strong for my child / family	**1**	**2**	**3**	**4**	**5**
Worrying that my child may get very ill and need to be admitted to hospital	**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**
Feelings of no privacy when we are at the hospital	**1**	**2**	**3**	**4**	**5**
Worrying about my child's growth and development	**1**	**2**	**3**	**4**	**5**
Feeling that my family / friends do not support me enough	**1**	**2**	**3**	**4**	**5**
Feeling like I cannot 'switch off' to my child's condition	**1**	**2**	**3**	**4**	**5**
Feeling overwhelmed trying to fit family life around 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 overwhelmed by changes in my child's usual treatment	**1**	**2**	**3**	**4**	**5**
Feeling exhausted from travel related to my child's care	**1**	**2**	**3**	**4**	**5**

How much of a difficulty has this been for you during the last month:

	Never	Rarely	Sometimes	Often	Always
Feeling upset seeing my child having medical procedures	1	2	3	4	5
Worrying about disruptions to my child's education	1	2	3	4	5
Feeling frustrated when I have to wait around at hospital	1	2	3	4	5
Feeling exhausted from caring for my child	1	2	3	4	5
Feeling overwhelmed by feeding difficulties (e.g. strict dietary requirements, lack of appetite, vomiting)	1	2	3	4	5
Difficulties due to my child's own responsibilities in their care (e.g. not trusting my child to take medicines)	1	2	3	4	5
Boredom from waiting around at hospital	1	2	3	4	5
Blaming myself if my child gets ill or has bad results	1	2	3	4	5
Feeling that others do not understand my situation	1	2	3	4	5
Worrying about taking measurements correctly from my child (e.g. blood pressure, temperature).	1	2	3	4	5
Worrying about if my child has had the right amount of fluid	1	2	3	4	5
Feeling helpless when my child is ill or in pain	1	2	3	4	5
Feeling like I cannot 'switch off' when waiting for test results	1	2	3	4	5
Worrying that my child may be admitted to hospital	1	2	3	4	5
Feeling that my child's condition has taken over my life	1	2	3	4	5
Blaming myself for my child's kidney problems	1	2	3	4	5
Worrying about my child getting very ill or dying	1	2	3	4	5
Worrying about the effect of caring for my child on my health	1	2	3	4	5
Feeling overwhelmed trying to fit family life around my child's condition	1	2	3	4	5
Feeling alone in caring for my child	1	2	3	4	5
Worrying about how my child is coping	1	2	3	4	5
Feeling guilty about having to be strict with my child	1	2	3	4	5
Worrying about doing medical procedures correctly (e.g. injections, dialysis, tube feeding)	1	2	3	4	5

How much of a difficulty has this been for you during the last month:

	Never	Rarely	Sometimes	Often	Always
Worrying about the future	1	2	3	4	5
Feeling exhausted from having to plan everything	1	2	3	4	5
Worrying about money because of my child’s condition	1	2	3	4	5
Feeling overwhelmed by decisions I have to make about my child’s condition	1	2	3	4	5
Feeling frustrated from having to deal with medical staff that do not know my child	1	2	3	4	5
Feeling that I constantly check my child for signs of illness	1	2	3	4	5
Feeling troubled by memories of when my child was first diagnosed	1	2	3	4	5
Feeling that medical staff do not listen to me	1	2	3	4	5
Difficulties due to feelings of uncertainty	1	2	3	4	5
Feeling trapped because of caring for my child	1	2	3	4	5
Worrying that I might miss an important phone call from the hospital	1	2	3	4	5
Feeling that I cannot think about myself because of caring for my child	1	2	3	4	5
Worrying that I have not understood something that medical staff have told me	1	2	3	4	5
Sadness that I do not have a ‘normal’ relationship with my child	1	2	3	4	5
Struggling to manage my child’s emotional difficulties	1	2	3	4	5
Feeling that I cannot use family / friends for support because I do not trust them to do things correctly	1	2	3	4	5
Feeling guilty about spending less time with my partner / family	1	2	3	4	5
Struggling to manage my child’s difficult behaviour	1	2	3	4	5
Feeling that I am not the person that I used to be	1	2	3	4	5
Anger about the extra money that I have to spend	1	2	3	4	5
Sadness about things that I have given up or cannot do because of caring for my child (e.g. work, hobbies)	1	2	3	4	5
Feeling that I should be doing more for my child	1	2	3	4	5

PART 2: YOUR VIEWS ON THE QUESTIONNAIRE YOU HAVE JUST COMPLETED

We would like to know how well the items in the questionnaire you have just completed fit with your experience of being a carer of a child with kidney disease.

Please could you complete the questionnaire again, this time <u>circle how relevant or important this item</u> is as a caregiver of a child with kidney disease. Also, please feel free to write down any thoughts you have on the questionnaire as you look through it - there are some questions and space for comments on the back page.

How relevant or important is this item as a cause of difficulty for you:

	Not at all	A Little	Somewhat	Quite a Bit	Very Much
Worrying about my child's medicines being taken correctly	1	2	3	4	5
Arguing with my partner / family about my child's care	1	2	3	4	5
Holding back when I do not agree with medical staff	1	2	3	4	5
Worrying about my child during the night	1	2	3	4	5
Feeling under pressure to be strong for my child / family	1	2	3	4	5
Worrying that my child may get very ill and need to be admitted to hospital	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
Feelings of no privacy when we are at the hospital	1	2	3	4	5
Worrying about my child's growth and development	1	2	3	4	5
Feeling that my family / friends do not support me enough	1	2	3	4	5
Feeling like I cannot 'switch off' to my child's condition	1	2	3	4	5
Feeling overwhelmed trying to fit family life around 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 overwhelmed by changes in my child's usual treatment	1	2	3	4	5
Feeling exhausted from travel related to my child's care	1	2	3	4	5

How relevant or important is this item as a cause of difficulty for you:					
	Not at all	**A Little**	**Somewhat**	**Quite a Bit**	**Very Much**
Feeling upset seeing my child having medical procedures	**1**	**2**	**3**	**4**	**5**
Worrying about disruptions to my child’s education	**1**	**2**	**3**	**4**	**5**
Feeling frustrated when I have to wait around at hospital	**1**	**2**	**3**	**4**	**5**
Feeling exhausted from caring for my child	**1**	**2**	**3**	**4**	**5**
Feeling overwhelmed by feeding difficulties (e.g. strict dietary requirements, lack of appetite, vomiting)	**1**	**2**	**3**	**4**	**5**
Difficulties due to my child’s own responsibilities in their care (e.g. not trusting my child to take medicines)	**1**	**2**	**3**	**4**	**5**
Boredom from waiting around at hospital	**1**	**2**	**3**	**4**	**5**
Blaming myself if my child gets ill or has bad results	**1**	**2**	**3**	**4**	**5**
Feeling that others do not understand my situation	**1**	**2**	**3**	**4**	**5**
Worrying about taking measurements correctly from my child (e.g. blood pressure, temperature).	**1**	**2**	**3**	**4**	**5**
Worrying about if my child has had the right amount of fluid	**1**	**2**	**3**	**4**	**5**
Feeling helpless when my child is ill or in pain	**1**	**2**	**3**	**4**	**5**
Feeling like I cannot ‘switch off’ when waiting for test results	**1**	**2**	**3**	**4**	**5**
Worrying that my child may be admitted to hospital	**1**	**2**	**3**	**4**	**5**
Feeling that my child’s condition has taken over my life	**1**	**2**	**3**	**4**	**5**
Blaming myself for my child’s kidney problems	**1**	**2**	**3**	**4**	**5**
Worrying about my child getting very ill or dying	**1**	**2**	**3**	**4**	**5**
Worrying about the effect of caring for my child on my health	**1**	**2**	**3**	**4**	**5**
Feeling overwhelmed trying to fit family life around my child’s condition	**1**	**2**	**3**	**4**	**5**
Feeling alone in caring for my child	**1**	**2**	**3**	**4**	**5**
Worrying about how my child is coping	**1**	**2**	**3**	**4**	**5**
Feeling guilty about having to be strict with my child	**1**	**2**	**3**	**4**	**5**
Worrying about doing medical procedures correctly (e.g. injections, dialysis, tube feeding)	**1**	**2**	**3**	**4**	**5**

How relevant or important is this item as a cause of difficulty for you:	Not at all	A Little	Somewhat	Quite a Bit	Very Much
Worrying about the future	**1**	**2**	**3**	**4**	**5**
Feeling exhausted from having to plan everything	**1**	**2**	**3**	**4**	**5**
Worrying about money because of my child’s condition	**1**	**2**	**3**	**4**	**5**
Feeling overwhelmed by decisions I have to make about my child’s condition	**1**	**2**	**3**	**4**	**5**
Feeling frustrated from having to deal with medical staff that do not know my child	**1**	**2**	**3**	**4**	**5**
Feeling that I constantly check my child for signs of illness	**1**	**2**	**3**	**4**	**5**
Feeling troubled by memories of when my child was first diagnosed	**1**	**2**	**3**	**4**	**5**
Feeling that medical staff do not listen to me	**1**	**2**	**3**	**4**	**5**
Difficulties due to feelings of uncertainty	**1**	**2**	**3**	**4**	**5**
Feeling trapped because of caring for my child	**1**	**2**	**3**	**4**	**5**
Worrying that I might miss an important phone call from the hospital	**1**	**2**	**3**	**4**	**5**
Feeling that I cannot think about myself because of caring for my child	**1**	**2**	**3**	**4**	**5**
Worrying that I have not understood something that medical staff have told me	**1**	**2**	**3**	**4**	**5**
Sadness that I do not have a ‘normal’ relationship with my child	**1**	**2**	**3**	**4**	**5**
Struggling to manage my child’s emotional difficulties	**1**	**2**	**3**	**4**	**5**
Feeling that I cannot use family / friends for support because I do not trust them to do things correctly	**1**	**2**	**3**	**4**	**5**
Feeling guilty about spending less time with my partner / family	**1**	**2**	**3**	**4**	**5**
Struggling to manage my child’s difficult behaviour	**1**	**2**	**3**	**4**	**5**
Feeling that I am not the person that I used to be	**1**	**2**	**3**	**4**	**5**
Anger about the extra money that I have to spend	**1**	**2**	**3**	**4**	**5**
Sadness about things that I have given up or cannot do because of caring for my child (e.g. work, hobbies)	**1**	**2**	**3**	**4**	**5**
Feeling that I should be doing more for my child	**1**	**2**	**3**	**4**	**5**

Do you have any comments about the wording of this questionnaire?

Did you find it difficult to complete?

Do you think that that the questionnaire is a good overview of the experiences and difficulties of caring for a child with kidney disease?

Do you feel that anything of importance is missing? Please comment if so.

If you have any further comments - on anything at all - please write them below.

Thank you for your time. Your help is much appreciated.

Canterbury
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Appendix 18: Item reductions (deleted/merged) and additions further to piloting exercise

DELETED ITEMS (n = - 5)	
Minimal endorsement of item	
Anger about the extra money that I have to spend	
Feeling that medical staff do not listen to me	
Minimal endorsement /perceived overlap with existing item	
Feeling that my family / friends do not support me enough Feeling that I cannot use family / friends for support because I do not trust them to do things correctly	(Feeling alone in caring for my child)
Feeling exhausted from travel related to my child's care	(Feeling exhausted from caring for my child)

MERGED ITEMS (n = - 6)	
Feedback that two items should be merged together	
Struggling to manage my child's emotional difficulties Struggling to manage my child's difficult behaviour	Feeling uncertain about how to manage my child's emotions and difficult behaviour
Worrying that my child may be admitted to hospital Worrying that my child may get very ill and need to be admitted to hospital	Worrying that my child may have to be admitted to hospital
Feeling overwhelmed trying to fit family life around my child's condition Feeling overwhelmed trying to fit family activities (e.g. leisure, holidays) around my child's condition	Feeling overwhelmed trying to fit family life around my child's condition
Feedback that item should be merged with an existing item	
Worrying that I might miss an important telephone call from the hospital	Feeling unable to 'switch off' when waiting for test results or *a telephone call from the hospital*
Worrying about taking measurements correctly from my child (e.g. blood pressure, temperature)	Worrying about getting medical procedures right (e.g. dialysis, injections, tube feeding) or *taking measurements correctly*
Feeling upset seeing my child having medical procedures	Feeling helpless when my child is ill or in pain

ADDED ITEMS (n = +2)	
Worrying about the impact of my child's condition on my other children'	
Sadness about the negative impact of my child's condition on my relationship with my partner	