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Original Article

Caregiver burden of parents of young children with cystic fibrosis

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Abstract

Background: There is a paucity of research examining the impact of informal caregiving on parents of young children with cystic fibrosis (CF). The aim of this study was to examine caregiver burden and identify risk factors associated with high caregiver burden in mothers and fathers of young children with CF.
Methods: This was a cross-sectional study of parents of young children with CF. A total of 213 families were invited to complete the CarerQoL questionnaire, a validated tool composed of two parts: (i) the CarerQol-7D which describes the care situation in terms of the negative and positive effects of caregiving and (ii) the visual analogue scale (VAS) which measures happiness on a scale from 0 to 10 (0 = completely unhappy and 10 = completely happy). The utility score (US) is a weighted average of the subjective burden derived from the CarerQol-7D (0 − 100); higher US indicates reduced burden. Differences in mother-father dyad median utility scores were examined using Wilcoxon signed rank test. Generalised linear mixed models were used to identify factors associated with high caregiver burden.
Results: At least one parent from 195 families completed the questionnaire (130 mother-father dyads, 189 mothers and 137 fathers). Fathers had a significantly higher median utility score than mothers [(89.2 (IQR 79.6–96.5) vs. 84.7 (74.5–88.0) $p < 0.001$]. Factors found to be significantly associated with higher caregiver burden were increasing child age (OR 1.02; CI: 1.00–1.04), having a child ever positive for *Pseudomonas aeruginosa* (Pa) (OR 2.48; CI: 1.30–4.73) and being a mother (OR 1.65; CI: 1.02–2.65).
Conclusions: This study contributes new findings to the sparse literature on caregiver burden of parents of young children with CF. Increasing child age and infection with Pa, associated with higher morbidity, were linked to greater parental burden.

Keywords: Caregiver burden; CarerQol; Cystic fibrosis; Parents

1. Introduction

Informal caregivers perform an important economic and social function in society. The economic value of informal care is estimated at £132 billion per year in the UK and $470 billion per year in the US [1,2]. The importance of informal care is set to increase due to demographic changes, the increase in the aging population and growing pressure on healthcare services [1].

There have been significant advances in CF care in the past decade and as a result CF patients are living longer into adulthood with a median life expectancy of 40 years for children born in 2010 [3]. According to Sawicki et al. [4] for adult patients with CF there is a high level of treatment activity each day, irrespective of age and disease severity. CF patients are required to take pancreatic enzymes multiple times each day, monitor their nutritional intake, perform airway clearance and administer inhaled and oral medications to slow the progression of the disease. Adults with CF are generally independent with their caring needs; however young children and adolescents require a higher level of assistance from their caregivers. Informal caregivers like parents of young children with CF are required to adapt quickly to the caregiving role. They take on more responsibility with this role as young children and infants require assistance with administration of medication and treatment management. Caring for a child with a

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chronic illness can present significant challenges for caregivers. Challenges for caregivers of young children with CF include; ambiguity about disease progression, financial strain due to direct and indirect costs associated with care, adherence to complicated treatment schedules, frequent outpatient clinic visits, disruption to family life due to hospitalisation and complexity with making plans due to uncertainty about changes in their child's health status.

There has been a growing interest in measuring health related quality of life (HRQOL) in the past few decades. HRQOL is now frequently measured in clinical trials along with medical outcomes as the importance of HRQOL has been increasingly recognised. The Food and Drug Administration (FDA) and the European Medicines Agency (EMA) frequently request that HRQOL be measured as part of clinical trials [5]. Numerous studies have measured quality of life (QOL) of CF patients [6–8]. A disease-specific quality of life instrument has been developed for use with CF patients, the Cystic Fibrosis Questionnaire (CFQ), which is valid for use in children 8 years and over [9]. However, there is only one disease-specific QOL questionnaire for use in CF carers; this questionnaire has only been used in two studies to date [10,11].

The care-related quality of life instrument (CarerQoL) is a newer instrument that measures caregiver burden and QOL [21]. The CarerQoL describes the caring circumstances in terms of the negative and positive effect of caregiving [12], and provides an assessment for the overall impact of informal care. The CarerQoL questionnaire was developed by Brouwer et al. [12] and is based on the EuroQoL. The CarerQoL has been used in studies to measure the QOL of parents of children with muscular dystrophy, haemophilia and children with craniofacial malformations [13,14]. Five validation studies have been conducted which showed good psychometric properties for the CarerQoL [12,14–17].

A literature gap exists regarding the caregiver burden of parents of young children with CF and therefore little is known about the QOL of carers of young children with CF. The main aim of this study was to examine the caregiver burden and QOL of parents of young children with CF using the CarerQol questionnaire, and to examine factors associated with increased caregiver burden.

2. Methods

This was a cross-sectional study of mothers and fathers of young children with CF which was part of a larger study (the Irish Comparative Outcomes Study (ICOS)) of CF examining clinical outcomes of children with CF in Ireland. The ICOS Study is a national historical cohort study comparing children clinically diagnosed with CF (clinical cohort) and those diagnosed via newborn bloodspot screening for CF (NBS cohort). Informed consent was obtained from parents to participate in the study. A total of 213 families enrolled in the ICOS study were invited to complete the CarerQoL questionnaire. If both parents were not currently living together then only the principal carer was asked to complete the CarerQoL questionnaire. Parents were contacted by telephone to complete two questionnaires (the CarerQoL and a costs questionnaire). The costs questionnaire was used to collect demographic information from parents (age, marital status, employment status, educational status, nationality and possession of private health insurance).

The CarerQol questionnaire was completed by parents (both mothers and fathers) in one of three ways:

- Via telephone consultation with a member of the study team
- By self-completion when they were recruited in the outpatient CF clinic (questionnaire returned to a member of the study team while in the clinic)
- By self-completion at home and returned by post.

The CarerQoL was administered initially to all parents recruited to the study (*Baseline questionnaire*), the questionnaire was also administered a second time (*year 4*) to parents in the NBS cohort only when the child reached age four years to facilitate an age-comparable group with the clinical cohort. This was necessary because the clinical cohort was significantly older at the time of recruitment (median age at study enrolment was 17.2 months for the NBS cohort and 64.2 months for the clinical cohort).

Permission was obtained from the authors to use the CarerQoL questionnaire for this study. There are two components to the questionnaire, the CarerQol-7D and the visual analogue scale (VAS). The CarerQol-7D is comprised of five negative (relational problems, mental health problems, problems combining daily activities with care-tasks, financial problems, physical health problems) and two positive (fulfilment from caregiving, social and family support when needed) dimensions of providing informal care. A utility score (US) can be obtained from the CarerQoL7D; this is a weighted average of subjective caregiver burden. Possible scores are from 0 to 100 with a higher score indicating a reduced burden. Utility scores based on relative utility weights for the UK population were used for this study [18]. The second component is the VAS which measures happiness (subjective wellbeing) on a scale of 0 to 10, where 0 is equal to a state of complete unhappiness and 10 is equal to a state of complete happiness.

2.1. Data analysis

To examine if mothers and fathers were responding to the seven components of the CarerQoL7D in a similar way the McNemar-Bowker test of symmetry was used. A comparison of mother-father dyads with regard to median utility score and median visual analogue scale was conducted using the Wilcoxon signed rank test and the Sign test as data were not normally distributed. This data was also split into two sub-groups, based on mean age of the children at completion of the questionnaire ($\geq$/$<$40 months); this was done to examine any effect the child's age might have on caregiver burden. Mother-father dyads that had another child in the study were excluded from this analysis (the parents could not be stratified into either group as they had one child in the screen detected group and one in the clinically diagnosed group). The baseline data was used for this comparison.

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