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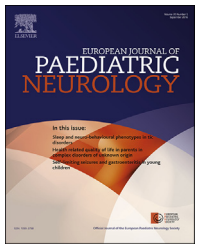

Original Article

Parental quality of life in complex paediatric neurologic disorders of unknown aetiology

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ABSTRACT

Complex paediatric neurology (CPN) patients generally present with non-specific symptoms, such as developmental delay, impaired movement and epilepsy. The diagnostic trajectory in these disorders is usually complicated and long-lasting, and may be burdensome to the patients and their parents. Additionally, as caring for a chronically ill child can be stressful and demanding, parents of these patients may experience impaired health-related quality of life (HRQoL). This study aims to assess parental HRQoL and factors related to it in CPN.

Physical and mental HRQoL of 120 parents was measured and compared to the general population using the SF-12 questionnaire. Parents also completed this questionnaire for the measurement of patient HRQoL. Additional questionnaires were used to measure parental uncertainty (Visual Analogue Scale) and worry phenomena (Penn State Worry Questionnaire), and to obtain socio-demographic data. A linear mixed model with random effect was used to investigate which of these variables were associated with parental HRQoL.

As compared to the general population, HRQoL of these parents appeared diminished. Fathers showed both lowered physical (51.76, $p < 0.05$) and mental (49.41, $p < 0.01$) HRQoL, whereas mothers only showed diminished mental (46.46, $p < 0.01$) HRQoL. Patient HRQoL and parental worry phenomena were significantly correlated with overall and mental parental HRQoL.

List of abbreviations: CPN, complex paediatric neurology; e.c.i., e. causa ignota; HRQoL, health-related quality of life; MCS, mental component score of the SF-12; PCS, physical component score of the SF-12; PCQ, productivity cost questionnaire; PSWQ, Penn State Worry Questionnaire; SF-6D, Short Form 6 Dimension; SF-12, 12-item Short Form health survey; VAS, visual analogue scale.

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The reduction in parental mental HRQoL is alarming, also because children strongly rely on their parents and parental mental health is known to influence children's health. Awareness of these problems among clinicians, and supportive care if needed are important to prevent exacerbation of the problems.

1. Introduction

Complex paediatric neurologic disorders for which patients are referred to academic paediatric neurology departments are a heterogeneous group of chronic disorders. Often, these have a genetic origin and patients present at young age with non-specific symptoms, ranging from developmental delay to epileptic seizures and motor symptoms. Because of the non-specificity of the symptoms, the diagnostic trajectory in complex paediatric neurology is usually long, and conclusive diagnoses are established in a minority of patients.[1]

Chronic paediatric neurologic conditions, such as epilepsy, cerebral palsy and pervasive developmental disorders, are known to affect the health-related quality of life (HRQoL) of both patients and caregivers, which are in these disorders usually the parents.[2–4] Previous studies show that parents of children with developmental disabilities experience increased stress,[5,6] impaired mental health,[7] a sense of devaluation and self-blame,[8] impaired physical functioning, and fatigue or exhaustion.[4,9,10] Moreover, parents of children with cerebral palsy indicate that caring for a child with a neurologic disorder has an impact on physical health, involves disrupted sleep and limited time, and puts pressure on both marital and other social relationships.[11]

In complex paediatric neurology, many children remain currently undiagnosed despite intensive diagnostic trajectories.[1] The high frequency of hospital appointments (on average, 15.6 in this patient population) and (invasive) diagnostic testing (on average 30.0 imaging, neurophysiologic, genetic and other diagnostic tests) is known to cause both child and parental distress.[1,12] Moreover, diagnostic uncertainty may result in less effective coping and poorer adaptation, as parents have no information on prognosis and no ability to tap into established support networks. Therefore, complex paediatric neurologic disorders are expected to affect parental HRQoL.[12,13]

As chronically ill children demand parental participation and adaptation, and rely on their parents for health care decisions, careful evaluation of parental HRQoL is important.[14] Furthermore, determining factors associated with parental HRQoL might allow for early detection of parental health problems and provision of supportive care when needed.

Parental HRQoL might be affected by patient-related factors. For example, more severely affected patients who need more care causing a higher burden for parents are expected to impact their parents' HRQoL more strongly than less severely affected patients. The HRQoL of the patient is therefore expected to be positively correlated with parental HRQoL. Patient age on the other hand, is expected to be negatively correlated with parental HRQoL. As patients usually present with symptoms at very young age, patient age is assumed to be a proxy measure for the duration of the diagnostic trajectory of the patient. A long-lasting diagnostic trajectory is expected to reduce parental HRQoL. Not only is this diagnostic trajectory expected to result in more stress over time due to the numerous trips to the hospital and many (invasive) diagnostic procedures, care giving requirements for children with neurological symptoms might also increase over time, as pointed out by Carmichael et al. and Macias et al.[12,15]

In addition to patient factors, parental characteristics might also be associated with their HRQoL.[16–18] Lower education and less hours of employment are known predictors for parental stress.[19] Therefore, higher education and employment are expected to be positively correlated with parental HRQoL.[18] Moreover, Macias et al. showed that older mothers of children with spina bifida report greater stresses related to medical and legal concerns than younger mothers.[15] Additionally, HRQoL is known to generally decrease with aging.[20,21] Hence, parent age is expected to be positively correlated with parental stress and therefore negatively correlated with parental HRQoL. Also, Lenhard et al. state that uncertainty around the aetiology of disease constitutes a determinant of long-lasting emotional burden for parents.[22] Therefore, the uncertainty parents experience regarding the aetiology of disease is expected to reduce parental HRQoL. Finally, since parental worry is known to be related to parental anxiety and stress, we expect parents with high levels of trait worry to have a lowered HRQoL.[16,17,23]

The aims of this study are (1) to assess parental HRQoL in complex paediatric neurology and (2) to gain insight in the relation between parental HRQoL and several patient and parental variables.

2. Methods

2.1. *Study population and design*

This cross-sectional study was conducted at the Department of Paediatric Neurology at the Radboud university medical center in Nijmegen, the Netherlands. Two experienced paediatric neurologists consecutively enrolled 100 eligible patients and their parents between August 2013 and January 2015. Patients were children $\leq$ 18 years, presenting with complex neurologic problems e causa ignota (e.c.i.), of