



Review Article

## A Systematic Review of the Quality of Life of Carers of Children with Cleft Lip and/or Palate

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### ABSTRACT

Caring for an infant or child requires a significant amount of time, energy and resources; this burden is further increased when the infant or child has a chronic condition or disability. Prior research has demonstrated that caregiving for a child with special needs impacts upon parents or carers mental health, well-being and quality of life. This article systematically reviews the literature pertaining to the impact of caring for a child with cleft lip and /or palate upon parental quality of life. A search of four databases was conducted with a number of key terms; the titles, abstracts and finally the whole article were read and assessed for relevance. Only articles written in English were included in the review. The results yielded four relevant articles; that displayed inconsistent results. The results of these articles are reviewed. It was evident that the construct of quality of life was narrowly operationalised in all four articles either being assessed as health-related quality of life or as the impact upon the family. Further all four studies emanated from the same country. The limitations are discussed with recommendations made for future research endeavours.

**KEYWORDS:** Quality of Life, Caregiving, Chronic Illness, Cleft Lip / Palate

### INTRODUCTION

All infants require caregiving; with the amount of caregiving that is required decreasing as the child develops. Caregiving for an infant or child can take a considerable amount of time, energy and resources. This burden of caregiving can impact upon the life of the primary caregiver generally; the parents. However an infant that is developing 'normally' requires less care than an infant or child who has a chronic illness or disability. Additionally it is known that having a child with a chronic condition

or disability results in more familial stress <sup>[1]</sup> as well as more anxieties for the child's future. <sup>[2]</sup> Thus what is the impact of caring for a special needs child upon their primary caregivers?

The notion of impact has been differentially defined and operationalised in prior studies assessing the effect of caregiving for an infant or child with special needs. Previous research has assessed the impact of caregiving upon psychological indices such as depression and anxiety, <sup>[3, 4]</sup> mental health <sup>[5]</sup> and well-being. <sup>[6]</sup>

Although these indices are important they are limited as they only offer a narrow insight into the impact of caring for an infant or child with special needs. That is caring for a child with special needs has a broader effect upon the life of the caregiver than is captured through the aforementioned constructs.

Therefore prior research investigating the impact of caregiving upon the carer has utilised the multi-faceted construct of quality of life (QoL). [3, 4, 7] QoL has been defined by the World Health Organisation QoL Group as an “individual’s perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” [8; p.551]. This definition of QoL highlights that QoL is a subjective evaluation that is contextually based. [8] This conception of QoL incorporates six broad domains namely; physical health, psychological state, levels of independence, social relationships, environmental characteristics and spiritual matters. [7, 8]

However there are a multitude of definitions and operationalisations of QoL in the prior literature [9] with the discussion of which being beyond the scope of the current article. Ideologically in relation to research on the impact of caregiving the construct of QoL is thought to capture the broader more encompassing impact of caregiving upon different facets of an individual’s life.

Previous research has assessed the QoL of parents with children with a range of chronic conditions or illnesses. [4, 7, 10, 11] It is evident that caring for a child with cerebral palsy, [10] epilepsy, [4] cancer, [11] and autism [7] result in poorer QoL for parents. Specifically Davis and colleagues [10] through a qualitative study found that both mothers and fathers of children with cerebral palsy aged 3-18 years had diminished physical, social, family, financial and

freedom related QoL; with no differences evident by the age of the child or their level of impairment. Additionally it was also apparent that this relationship was mediated by levels of parental stress.

Similarly Witt and colleagues [11] found that parents of children with childhood cancer had a poorer QoL than parents of children who did not have an illness or chronic condition. This impact was greatest on mental health and was also mediated by parental stress. Likewise Lv et al. [4] found decreased health-related QoL in parents of children with epilepsy; finding that parents of children whose epilepsy was well controlled exhibited better QoL than their peers who had children unremitting epilepsy.

Conversely a study conducted by Shu [7] found that mothers who expressed more positive feelings about caring for their child with autism reported a better QoL. Thus it is apparent that caring for a child with a chronic condition or illness can result in a decreased QoL for parents and caregivers. It is also evident that QoL is unsurprisingly negatively affected by stress and positively impacted upon by positive feelings.

Although numerous chronic conditions and illness have been previously studied others have not received a great deal of attention. Therefore this article systematically reviews the previous literature on the impact of caring for a child with a cleft lip / palate (CL/P) upon their primary caregivers QoL; as CL/P is a condition that can significantly impact upon the capacity to care for an infant or child.

#### ***Cleft Lip and/or Palate***

CL/P is a craniofacial abnormality that affects approximately 1 in 700 live births. The incidence rates vary by sex and type of cleft and occur more frequently among the Indian and Oriental populations. [3] Having an infant or child with CL/P

presents distinct challenges to the caregiver, as children experience multiple difficulties from birth. For example mothers experience difficulties with or it is impossible to breast feed the infant, the children suffer from other complications such as hearing and speech impediments as well as having to undergo in some instances multiple surgeries.

## METHODOLOGY

A systematic search of four electronic databases was conducted namely, Medline (January 1966 – August 2011), CINAHL (January 1982 – August 2011), PsycINFO (January 1887 – August 2011) and Web of Science (January 1981- August 2011) for any articles pertaining to QoL of CL/P carers. Specifically each database was searched four times with the combinations of keywords and the Boolean AND reported in Table 1.

**Table 1. Search Terms**

Search	Keywords
1	Cleft Lip Palate AND Quality of Life AND Caregiver
2	Cleft Lip Palate AND Quality of Life
3	Cleft Lip Palate AND Quality of Life AND parents
4	Cleft Lip Palate AND parents

The title of each of the search results was then read and assessed for relevance; the abstracts of the articles that appeared to be relevant were then read and assessed for relevance. The entire articles of those deemed relevant were then read and further assessed for relevance. The reference lists of the articles deemed relevant were then searched for any additional relevant articles. Only articles that were written in English were included in this review.

## RESULTS

The aforementioned search strategy yielded a total of 4 relevant articles that investigated QoL of parents or carers of children with CL/P. The oldest article was published in 2005 and the most recent in 2009. These articles and their major findings are summarised in Table 2 below. It is apparent that QoL within these studies was narrowly assessed as either health-related QoL or the impact of the child's CL/P upon the family. The results of each article are elaborated on in the section below.

**Table 2. Details of the Studies Included in the Review**

Author	Sample	QoL Measure	Main Outcomes
Weigl et al. <sup>[3]</sup>	50 Mothers of Children with CL/P between 12months – 10 years	Health-Related QoL Short-Form Health Survey (SF-36)	Normal values for the majority of SF-36 domains. Mothers of CL/P displayed better Health-Related QoL than controls in the following domains; personal functioning, bodily pain and general health.
Kramer et al. <sup>[12]</sup>	130 families with children with CL/P between the 6-24 months of age	QoL was assessed by the Impact on Family Scale (IOFS)	Most families exhibited relatively small impacts on all IOFS domains. With impacts being greatest in the domain areas of coping strategies and mastery techniques and personal impacts. Differing impacts were apparent by cleft type.
Kramer et al. <sup>[13]</sup>	147 families with children with CL/P between 5-6 years	QoL was assessed by the Impact on Family Scale (IOFS)	Most families reported low scores on the IOFS, with highest scores on personal impact and coping strategies. Coping was higher in families of children with CL and personal impact was concerned with CLP and CP.
Kramer et al. <sup>[14]</sup>	132 families of school aged children with CL/P with a mean age of 9.6 years	QoL was assessed by the Impact on Family Scale (IOFS)	The impact on family was highest on the IOFS dimensions of personal impact and coping/mastering strategies. Families of children with CL reported better QoL as assessed through the IOFS than families with children with CLP and CP.

### ***Quality of Life of Carer's of an Infant or Child with a Cleft Lip / Palate***

As outlined in Table 2 above it is apparent that the results of these studies are inconsistent. Specifically Weigl and colleagues [3] found no impact of caring for a child between 1-10 years of age with CL/P upon mothers health-related QoL in comparison to mothers of 'normal' children. The SF-36 assesses health-related QoL through the following domains; physical functioning, role functioning, bodily pain, general health, vitality, social functioning, emotional functioning and mental health. Surprisingly the results indicated that mothers of children with CL/P had better QoL on the following domains namely; physical functioning, bodily pain and general health. These findings were contrary to the authors' expectations. [3]

Whereas Kramer and colleagues [12, 13, 14] utilising the IOFS to assess QoL of parents of children with CL/P found that QoL was diminished in this group; although the impact was found to be small. Specifically the IOFS assesses the impact of the ill child on the family through the following domains; financial, social, personal, coping and other children.

Kramer and colleagues [12] found relatively small impacts on all dimensions for parents of children with CL/P aged between 6-24 months. Specifically impacts were most evident on the dimensions of coping and personal impact. [12] Similarly Kramer and colleagues [13, 14] found comparable results with the parents of older children with CL/P specifically a mean age of 6.1 [13] and 9.6 [14] years respectively.

It was also evident that impacts differed by type of cleft. Surprisingly parents of children with cleft lip and palate which is more severe reported less impact upon QoL as assessed by the IOFS than parents of children with only cleft lip or palate. [12] The differential effect of type of

cleft was further investigated in a subsequent research endeavour. [13] Finding that impacts on coping were related to cleft lip theorised to be resulting from the influence of aesthetics' upon formulating adequate coping strategies'. [13] Whereas personal impact related to the more severe clefts namely cleft lip and palate and cleft palate this was pertain to the functional impairments of the child. [13] The concern for other children was largest in families with children with the most severe type of cleft; cleft lip and palate. [13]

### **DISCUSSION**

It is apparent from the above review that there is a huge scarcity of literature pertaining to the QoL of parents and carers of children with CL/P. Furthermore it is also apparent that the limited literature reports contradictory results, specifically between the study conducted by Weigl and colleagues [3] and the work of Kramer and colleagues. [12, 13, 14] Specifically Weigl and colleagues [3] found no impact on QoL as assessed by the SF-36 on parents of children with CL/P and Kramer and colleagues [12, 13, 14] found that caring for a child with CL/P did impact upon QoL of carers as measured by the IOFS.

However the SF-36 is ultimately a measure of health status [15] as opposed to being a measure of QoL. Similarly the IOFS was originally created with the objective of assessing the impact of having an ill child upon the family unit [15] which is distinctive to the construct of QoL. Furthermore the IOFS has been found to have dubious psychometric properties; with items loading on more than one factor and low factor loadings. [15] As aforementioned in the introduction the notion of QoL is a broad multi-faceted construct that encompasses more than health status and the familial impact of caring for an ill child. [8]

Therefore it is apparent that perhaps caring for a child with CL/P does impact upon the broader notion of QoL as conceptualised by the WHOQoL Group [8] which is contradictory to the findings of Weigl and colleagues. [3] Equally Kramer and colleagues [12, 13, 14] unsurprisingly found that caring for a CL/P child impacts upon the family unit however this impact does not equate to the construct of QoL. [8] Thus it is apparent that we know at the best very little about the relationship between caring for a child with CL/P and parental QoL.

Another issue pertaining to the current research is that all four articles emanate from the same country. That is all the research was conducted in Germany which extremely limits the generalizability of the findings. [3, 12, 13, 14] In addition as mentioned earlier CL/P is known to be more common in Oriental and Indian populations as oppose to European further limiting the generalizability of findings. [3]

Therefore it is apparent that QoL of parents and carers of children with CL/P is chronically under-researched. The research literature that does exist poorly operationalizes the construct and theoretically does not measure QoL and all the research emanates from the same country.

Thus it is recommended that future research seeks to measure QoL in parents and carers of children with CL/P in order to delineate whether caring for a child with CL/P leads to a diminished QoL. Further these research endeavours should be facilitated by the use of the WHOQoL-BREF QoL scale in order to fully capture the construct of QoL. [8] Such research endeavours should be conducted cross-culturally and with a significant focus upon populations in which CL/P is known to be more evident. Following from the above

recommendations it is also suggested that variables that could potentially moderate the impact of caring for a child with CL/P on parental QoL be investigated.

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