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Original Research

Health-Related Quality of Life Components in Children With Neonatal Brachial Plexus Palsy: A Qualitative Study

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Abstract

Background: Currently, no published, validated patient-reported outcome (PRO) measures of health-related quality of life (HRQOL) exist for use with neonatal brachial plexus palsy (NBPP). NBPP is a debilitating condition that occurs during the perinatal period, resulting in paralysis/paresis and loss of sensation in the affected arm. Commonly used NBPP measures are not comprehensive and do not fully account for clinically meaningful changes in function or progression of the disorder.
Objective: To evaluate important components of HRQOL for children with NBPP and identify where new PRO measures are needed.
Design: Eleven focus groups comprising children with NBPP (4), family members (6), and professional providers (1) to assess HRQOL.
Setting: Brachial plexus clinic.
Participants: Children with NBPP, their parents, and professional providers.
Inclusion Criteria: Children 7-17 years old with NBPP; parents/caregivers at least 18 years of age; professionals with $\geq$2 years' experience providing NBPP clinical care; ability to read and speak English fluently.
Methods: Focus group sessions were recorded, transcribed verbatim, and deidentified. Qualitative frequency analysis identified different aspects of HRQOL relevant to NBPP. This analysis expands on the grounded-theory approach to qualitative analysis, including development of a domain framework, open and axial coding, selective coding, and descriptive analysis. The resulting HRQOL domain framework (and frequency analysis) was then compared to the domain framework for existing PRO measures (PROMIS and Neuro-QoL) to identify components of HRQOL where new PRO measures are needed for NBPP.
Main Outcome Measures: Not applicable.
Results: Although many physical, social, and emotional health domains were captured by existing PRO measures, some significant NBPP-specific topics emerged from qualitative analysis—functionality, sensory, physical appearance, arm/hand compensation and preference, explaining functionality/appearance to others, and self-esteem and body image concerns.
Conclusions: Development of sensitive and specific measures capturing arm/hand function and body image would improve the clinical care of patients with NBPP.

Introduction

Neonatal brachial plexus palsy (NBPP) affects approximately 1.5 per 1000 live births annually in the United States [1,2]. This condition involves stretching of the brachial plexus nerves during the perinatal period, affecting muscle movements of the upper extremity. Although 60%-90% of infants with NBPP recover spontaneously at an early age, the remaining patients experience long-term residual physical impairments, including paralysis/paresis and loss of sensation in shoulder, elbow, wrist, and/or hand [1,3,4]. Such impairments are associated with annual medical and social costs ranging from 1 to 3 billion U.S. dollars [5].

For children with residual upper extremity impairment, customized occupational/physical therapies or surgical treatments often are recommended for regaining upper extremity function; however, treatment outcomes vary depending on palsy extension and involvement [6-9]. The uncertainty of treatment outcomes and bilateral discrepancies in physical appearance might contribute to associated psychosocial

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difficulties in children with NBPP. For example, appearance discrepancies such as muscle atrophy or arm length differences could limit participation in sports/school activities and might isolate children with NBPP from their peers in social settings [10-12].

Because of the complexity of the palsy and the fact that there are no standard guidelines for the treatment of NBPP, it is critical to understand patient expectations and overall health status to achieve maximum treatment efficacy. Patient-reported outcome (PRO) measures reflect patient and family perspectives of the physical, emotional, and social aspects of health-related quality of life (HRQOL). Currently, there are no consensus PRO measures of HRQOL for use in the NBPP population [13]. Existing measures for children with NBPP mainly focus on physical examinations such as muscle power and range of motion without indication of how these physical factors affect daily activities or psychosocial well-being. Furthermore, generic physician-developed measures such as the Child Health Questionnaire and Pediatric Outcomes Data Collection Instrument fail to capture comprehensive NBPP physical, emotional, and social concerns or NBPP-specific issues, such as sensory and body image perception [14-18]. It is critical to understand the patient's perspective of their condition to better understand the natural course of the disease, its impact on HRQOL during child development, as well as the effectiveness of interventions to improve HRQOL.

Recently, the National Institutes of Health launched the Patient-Reported Outcome Measurement Information System (PROMIS) to develop generic measures of HRQOL that are universally applicable to various conditions (http://www.nihpromis.org). Quality of Life in Neurological Disorders (Neuro-QoL) is another federally funded initiative to create, in tandem with PROMIS, measures of HRQOL that are applicable across neurologic conditions. Neuro-QoL, and PROMIS in particular, have been used widely in pediatric populations [19-26] and are designed to sensitively assess self-reported HRQOL. Although these measures have been used in several different pediatric populations, neither PROMIS nor Neuro-QoL has been examined in NBPP.

Thus, the purpose of this study was to explore HRQOL from the perspective of children with NBPP, family members, and clinicians. Specifically, we focused on identifying the relevant aspects of HRQOL that apply to individuals with NBPP, with an eye toward identifying appropriate existing PROs and areas in which new PRO development is needed for use in NBPP.

Methods

Participants

This study used a community-based partnership approach that included key NBPP stakeholders. Specifically, we conducted 2 focus groups of children with NBPP (separate groups for ages 7-8 and 9-17), 6 groups of caregivers/parents (separate groups for parents of children ages 0-4, 5-8, and 9-17), and 1 group of NBPP professionals (eg, clinicians and researchers with more than 2 years' experience working with NBPP) to determine domains and items relevant to HRQOL in individuals with NBPP. All participants were recruited through the Brachial Plexus Palsy clinic. Inclusion criteria for the various groups were as follows: (1) children from 7 to 17 years of age with NBPP; (2) parents/caregivers who were at least 18 years of age at time of participation; and (3) $\geq$2 years' experience providing NBPP clinical care. In addition, because focus group participation requires verbal discussion of HRQOL and participation requires completion of existing PRO measures, children and parent/caregivers were required to read and speak English fluently. Data were collected in accordance with local institutional review boards; participants provided informed consent (and/or assent) before participation.

Data Collection and Analysis

Each focus group discussion was a total of ~90 minutes in length. Two female PhD-level group moderators with extensive focus group experience used broad, open-ended questions designed to allow participants to articulate how having NBPP has affected their (or their child's) overall HRQOL; moderators were not involved in these children's treatment. Follow-up prompts included open-ended prompts to discuss physical, social, and emotional health. Group discussion (~60 minutes) was then followed by the completion of several existing HRQOL measures (PROMIS/Neuro-QOL, ~20 minutes) and a brief discussion of the strengths and weaknesses of these measures (~10 minutes). Discussions were audio-recorded, transcribed verbatim, and deidentified. Qualitative frequency analysis was conducted according to established methodology [27]. Two PhD-level investigators created physical, emotional, and social codebooks for focus group transcript coding. These codebooks were created with the World Health Organization's conceptual framework for quality of life, which includes physical, emotional, and social domains [28]. A list of subdomains was created through a detailed review of the transcripts and was revised collaboratively with the use of input from the study team until a consensus was reached.

Coding was conducted by researchers with either a Bachelor's or Master's degree and supervised by a PhD-level psychologist. Before coding, researchers were required to establish interrater reliability ($\geq$80% agreement), to establish that they were applying the codebook similarly when coding transcripts. Once interrater reliability was established, the 2 raters were required to code the first transcript together to ensure