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USING QUESTIONNAIRE DESIGN TO EXPLORE REPORTING DIFFERENCES FOR SPASTICITY-RELATED PAIN IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

Manjari Quintanar-Solares,¹ Mona L. Martin,¹ Andrew Bryant,¹ Friedrich Asmus,² Thorin L. Geister²

¹HRA, Inc., Seattle, WA, USA; ²Merz Pharmaceuticals, GmbH, Frankfurt, Germany

INTRODUCTION

Cerebral Palsy (CP) is a motor impairment condition caused by abnormal brain development or damage to the brain during birth or in early childhood. CP-related disorders include limb spasticity, as well as impaired cognition, behavioral problems, and sensory deficits. [Aisen et al., 2011]. Developing a spasticity-related pain (SRP) Patient Reported Outcome (PRO) instrument for this diverse population poses specific challenges in addition to the factors considered in developing a pediatric PRO (reading levels, vocabulary, recall period).

The United States Food and Drug Administration (FDA) has issued guidance on the development of PROs in the adult population listing challenges to be considered in the development of PROs in the pediatric population [USDHHS, 2009]. The ISPOR Task Force has addressed some of these challenges in its report of good practices for pediatric PROs [Matza, (in press)], but as the authors describe in the report, several aspects of the design and formatting of these PROs require further research, in particular in special populations.

During the development of the Questionnaire on Pain Caused by Spasticity (QPS), initial (unpublished) qualitative interview data from children/adolescents with CP and their parents/caregivers documented instances in which children/adolescents reported intentionally hiding their pain from their parents/caregivers. This raised the concern that self-reported pain data would not be trustworthy in child self-reports. Both child/adolescent-reported and parent/caregiver-reported modules were developed for the QPS.

OBJECTIVES

- (1) To use progressive sequencing in questionnaire design in order to identify spasticity-related pain that the children initially denied
- (2) To compare the differences between the children/adolescents and their parents/caregivers regarding the reported presence of pain

METHODS

- Cognitive interviews with children/adolescents with CP and SRP and their parent/caregivers were scheduled in the US to evaluate the newly developed QPS.
- Children/adolescents with CP were eligible to participate in the study if they were between 5 and 17 years old, had upper or lower limb SRP, had any level of functional ability as determined by a Gross Motor Function Classification System (GMFCS) score, and were able to communicate sufficiently to respond to interview questions. In the case of children 2-4 years old, only their parent/caregiver was scheduled for an interview.
- During the interview, children/adolescents answered the draft questionnaire (either self- or interviewer-administered) and then were asked about their understanding of the individual items. Parent/caregiver interviews followed the same procedure. Interviews were audio recorded and transcribed, and the answers to both child/adolescent and parent/caregiver questionnaires were captured.
- Questions in the child/adolescent draft measure assessed presence (Yes/No) and severity (Wong-Baker Faces scale [Baker & Wong, 1987]) of SRP. The first item was general (*Think about today and the last 6 days. Did your hip, leg or foot hurt when it got tight during any of that time?*). The following items were all related to specific, progressively difficult activity situations (at rest, during usual daily activities, during active mobilization, and during physically difficult activities). Questions in the parent/caregiver draft measure assessed presence (Yes/No) and frequency (5-item Likert-type scale) of observable signs of SRP, in the same sequence.
- Each child/adolescent questionnaire was compared with that of his or her parent/caregiver, to identify differences in responses about SRP.

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Table 1 Children/Adolescent Demographic Characteristics

Children/Adolescents		Interviewed N=8 (100%)
Age (Years):	- Mean (SD)	12.1 (3.2)
	- Median	12.5
	- Range	7-16
Gender:	- Male	4 (50.0%)
	- Female	4 (50.0%)
Highest Grade Completed:	- 0 (No school)	0 (0%)
	- Grade 1	1 (12.5%)
	- Grade 2	1 (12.5%)
	- Grade 6	2 (25.0%)
	- Grade 7	1 (12.5%)
	- Grade 8	1 (12.5%)
Racial and Ethnic Group:	- White/Caucasian (Non-Hispanic)	5 (62.5%)
	- White/Caucasian (Hispanic)	2 (25.0%)
	- Hispanic/Latino	1 (12.5%)

Table 2: Clinical Characteristics

Children/Adolescents		Interviewed N=8 (100%)
Does child have a cognitive deficit?	- No	7 (87.5%)
	- Yes	1 (12.5%)
	- Able to communicate	1 (100% of 1)
	- Unable to communicate	0 (0.0% of 1)
	- Of 1 child with cognitive deficit, estimate of child's cognitive age (as assessed by clinician)	7 years
Gross Motor Function Classification System* (GMFCS) levels:	- GMFCS Level I	1 (12.5%)
	- GMFCS Level II	1 (12.5%)
	- GMFCS Level III	3 (37.5%)
	- GMFCS Level IV	1 (12.5%)
	- GMFCS Level V	2 (25.0%)
Pain Localization:	- Both Lower Limbs Only	3 (37.5%)
	- Upper and Lower Limbs on Both Sides	5 (62.5%)

RESULTS

- Ten children and 11 parent/caregivers were interviewed. However, data from 2 children were excluded from the analysis because even though they were able to communicate through "Yes/No" answers, they were not able to comprehend the faces scale to provide a reliable answer. Clinicians confirmed at enrollment that all subjects had SRP at least once per week. Table 1 shows the demographic characteristics of the 8 children/adolescents considered in the analysis, and Table 2 provides a summary of descriptive clinical characteristics.
- Evaluation of the questionnaire responses showed discrepancies between children/adolescents' and the corresponding parents/caregivers' responses in 7 out of the 8 sets analyzed. In most cases, children/adolescents responded that they didn't have SRP either in general or related to a specific activity, while the parent/caregiver reported observing signs of pain in their child in that given situation.
- Results also showed that 4 of the 8 children/adolescents reported having no pain on the initial general SRP item, yet they proceeded to acknowledge their pain in the subsequent items that asked them either to rate the severity of their pain, or asked them about presence of pain in a specific activity situation. Figure 1 shows an example of this finding. In 3 of these 4 cases, the parents/caregivers reported observing signs of pain in their child.

Figure 1: Example of the responses in a child/adolescent questionnaire

1. I think about today and the last 6 days. Did your hip, leg or foot hurt when it got tight during any of that time? Circle your answer.

YES NO

2. I think about today and the last 6 days. How much did your hip, leg or foot hurt during any of that time? Circle your answer.

3. Think about your today and the last 6 days. When you were just sitting, watching TV, or trying to sleep, did your hip, leg or foot hurt when it got tight? Circle your answer.

YES NO

CONCLUSION

The results from this study address an aspect of pediatric PRO design that is particularly relevant for children facing condition-specific challenges. The findings suggest that combining a self-reported and an observer-reported assessment produces complementary results that facilitate a more accurate picture of the spasticity-related pain experienced by children with CP. Results also suggest that in the assessment of chronic conditions in which children/adolescents may tend to accommodate to or deny their symptoms for any reason, the use of progressive sequencing of questions to inquire about the same concept might facilitate discovery of symptoms that might otherwise be denied.

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