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Disability and Rehabilitation

ISSN: (Print) (Online) Journal homepage: www.tandfonline.com/journals/idre20

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To cite this article: Hazel Brown, Ruth van der Looven, Emily S. Ho & Willem Pondaag (2024) Patient reported outcomes in brachial plexus birth injury: results from the iPLUTO world-wide consensus survey, Disability and Rehabilitation, 46:22, 5213-5219, DOI: 10.1080/09638288.2023.2298708

To link to this article: https://doi.org/10.1080/09638288.2023.2298708

DISABILITY AND REHABILITATION
2024, VOL. 46, NO. 22, 5213–5219
https://doi.org/10.1080/09638288.2023.2298708

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RESEARCH ARTICLE

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Patient reported outcomes in brachial plexus birth injury: results from the iPLUTO world-wide consensus survey

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ABSTRACT

Purpose: Brachial plexus birth injuries (BPBI) can have lifelong effects on the development and functional use of the upper extremity. Currently there is no agreement with regards to what patient-reported outcome (PRO) measures should be used. Therefore, the ability to compare the effects of treatment between individuals and institutions is challenging. This study aimed to achieve consensus among clinicians on the use of PRO measures within this patient group to allow for improved comparison of treatments and outcomes in the future.

Materials and Methods: Online, a 3 round Delphi survey was completed by 35 international multi-disciplinary specialist centers.

Results: All respondents (100%) agreed that PRO measures are useful for clinical evaluation and patient treatment. None of the outcome measures scored >75% agreement for ability to assess responsiveness and current state in children with BPBI as most outcome measures were judged as not specific for BPBI. Additionally, participant centers were asked their perspective on the best available PRO option for each of the 3 categories: functional use of the upper limb, quality of life and pain. This resulted in endorsement by the participant centers of the Brachial Plexus Outcome Measure – Self-Evaluation, the Pediatric Quality of Life Inventory, and Visual Analogue Scale/Brief Pain Inventory respectively.

Conclusion: International specialists in BPBI agree that PRO measures are important to use both clinically and in research in children aged 5 years and above.

ARTICLE HISTORY
Received 26 March 2023
Revised 13 December 2023
Accepted 16 December 2023

KEYWORDS
Brachial plexus birth injuries; neonatal brachial plexus palsy (MESH); patient reported outcome measures; clinical utility; consensus methods; survey

> IMPLICATIONS FOR REHABILITATION

Patient-reported outcome measures were judged as useful both in clinic and in research for brachial plexus birth injury (BPBI), according to a panel of specialized centers.

Currently available outcome measures were judged as not specific for BPBI.

The panel endorsed the following measures as best available: the Brachial Plexus Outcome Measure – Self-Evaluation scale for functional evaluation, the Pediatric Quality of Life Inventory for disease-related quality of life and the Faces Pain Scale - Revised/Visual Analogue Scale/Brief Pain Inventory for pain

Abbreviations: BPBI: Brachial Plexus Birth Injuries; BPI: Brief pain inventory; BPOM – SE: Brachial Plexus Outcome Measure – Self-Evaluation scale; CHEQ: Children's Hand-use Experience Questionnaire; FPS-R: Faces Pain Scale- Revised; iPLUTO: international PLexus oUtcome sTudy grOup; HR-QoL: Health-Related Quality of Life; Peds-QL: Pediatric Quality of Life Inventory; PRO: patient reported outcome; VAS: visual analogue scale; WHO-ICF: World Health Organization International Classification of Functioning, Disability and Health

Introduction

Brachial plexus birth injury (BPBI) is a rare condition affecting 1.74 per 1000 live births [1,2]. The injury causes decreased muscle strength and sensibility of the affected arm, but may also lead to joint contractures, dislocations, skeletal malformations, limb length discrepancy and altered posture. In addition to the neuromusculoskeletal problems, BPBI may lead to coordination disorders of the upper limb; which in turn may affect the developmental use of the arm and its integration into gross motor activities [3]. In later life, the children may suffer from joint dysfunction and pain [4,5]. Hence, at different stages of the child's life, the consequences of a BPBI can influence quality of life, may impose a financial burden and have an impact on life choices [6,7].

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Supplemental data for this article can be accessed online at https://doi.org/10.1080/09638288.2023.2298708.

There is no consensus regarding optimal treatment strategies and algorithms for children with BPBIs. Comparison and pooling of outcome data from different centers is challenging because of variation in the use of outcome measures, follow-up intervals and different patient ages. In an effort to improve comparability of research outcomes, the international PLexus oUtcome sTudy grOup (iPLUTO) project, was initiated with the purpose of defining a universal dataset for the evaluation of upper limb function of children with BPBI [8]. Using a Delphi-derived technique, the first iPLUTO study resulted in consensus on assessment techniques for identifying impairments within the Body Structure and Function domain of the World Health Organization International Classification of Functioning, Disability and Health (WHO-ICF) (Figure 1) [14]. The WHO-ICF encourages that activity and participation into everyday life, as well as environmental and personal factors, should be considered as a part of a holistic assessment. Information with regards to these dimensions can be gained directly from patients and, in the case of young children, caregivers through the completion of self-assessed measures.

The iPLUTO study findings echoed the conclusions of previous systematic reviews which stress the importance of including patient-reported outcome (PRO) measures within the assessment of BPBI [9]. (We chose to abbreviate as PRO measure instead of PROM to avoid confusion with Passive Range Of Motion.) However, consensus was not sought in the previous iPLUTO survey with regards to which PRO measures should be included within a minimal dataset [8]. The use of PRO measures is advocated because they require individuals to reflect upon, evaluate and score their own performance of everyday tasks or severity of symptoms. PRO measures therefore supply unique and meaningful data about the impact of BPBI and its treatment by allowing the patient to self-report their current level of function within their own environment [15,16]. Therefore, they are increasingly viewed as key component of patient-centered care.

When selecting an outcome measures for use within a patient group, consideration needs to be given to their psychometric properties (such as reliability, validity, and responsiveness to change over time) as well as their clinical utility (the feasibility of a tool for use in practice and its usefulness in being able to influence subsequent treatments) [17]. Unfortunately, disease-specific PRO measures with sound psychometric and performance quality evaluations are limited in rare conditions such as BPBI [9,18]. In their systematic review, Bialocerkowski et al. identified no outcome measure with psychometric evidence that suggested they covered the breadth and depth required for clinical and research application in BPBI [18].

Qualitative studies reported perceived unmet needs from both parents/families and patients who have incurred a BPBI [9,19,20]. In particular, the need for improved evaluation HR-QoL [10], pain [11,20], as well as functional status [19] have been highlighted. Therefore, the present study aimed to use a Delphi derived technique to obtain a consensus on the best PRO measures of functional status, HR-QoL and of pain for children and adolescents with BPBI [21].

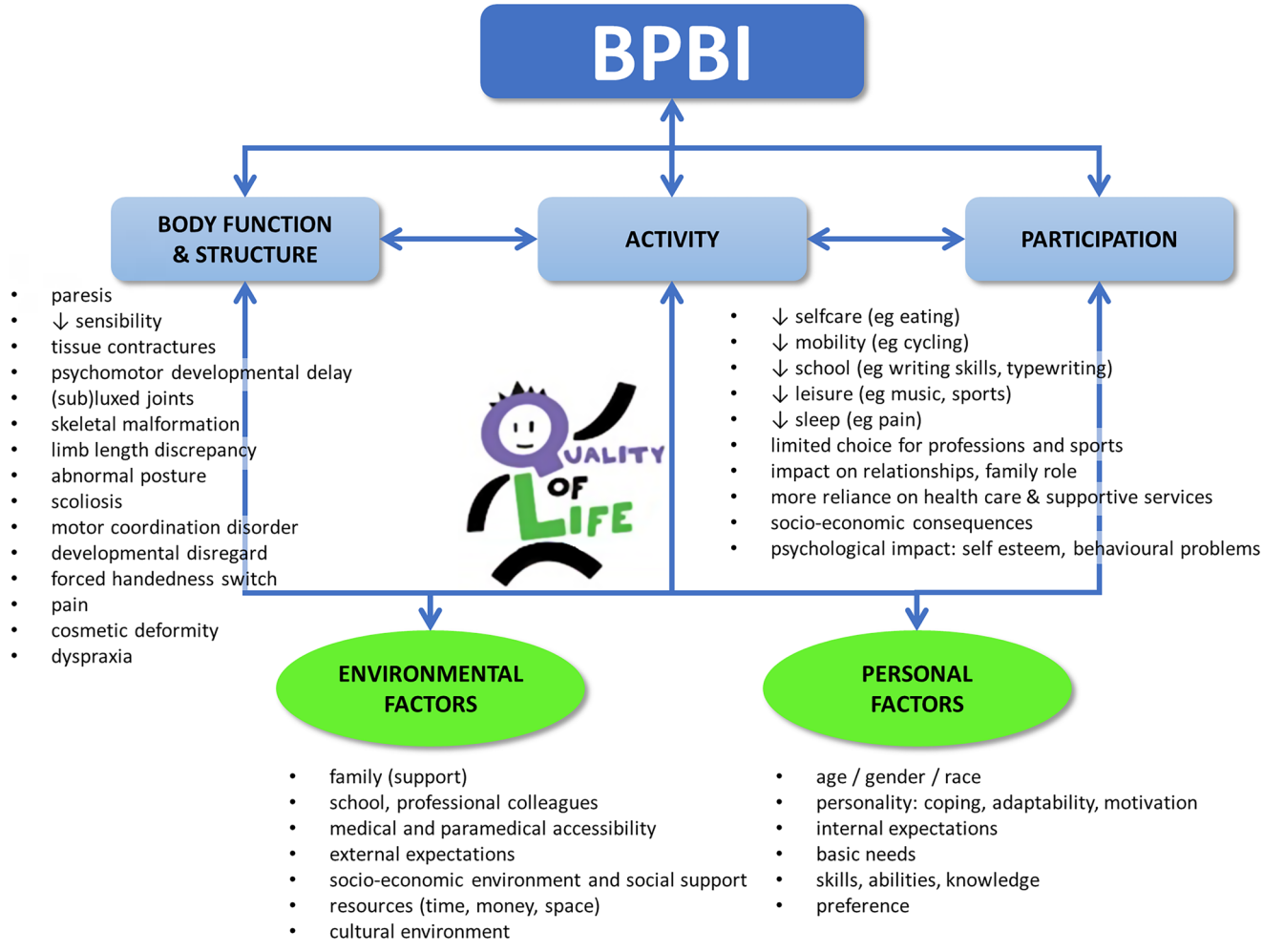

Figure 1. Schematic adaptation of WHO-ICF (2002) related to BPBI from van der Looven R [13].

Materials and methods

A series of structured questionnaires were developed as part of a multistage Delphi technique to gather the opinions of international BPBI experts and achieve group consensus. The study was undertaken in several stages:

Stage 1: Identification of Patient Reported Outcomes

Building upon the systematic reviews published on outcome measures in BPBI [9,18], the four moderators (all four authors) reviewed the literature to ascertain which PRO measures were currently in use and had been published with children and adolescents with BPBI. Basic characteristics of each PRO were collated (e.g., the validated age group, the number of questions, the original language and validated translations) and included within the online survey for participants to consider.

Stage 2: Classification of included Measures

After compiling the list of published PRO measures, the four moderators assigned each PRO to a relevant category: Health-Related Quality of life (HR-QoL) which included 5 PRO measures, Functional Status (11 PRO measures), or Pain (8 PRO measures).

Stage 3: Survey Development

Mirroring the methodology of the 2018 iPLUTO study [8], a Delphi-derived process was used. Delphi methods are iterative, multistage processes that are designed to establish group consensus [21]. The process involves the presentation of a questionnaire to a panel of informed individuals in a specific field in order to seek their opinion. After completion of the questionnaires, the data are summarized and the overall group response together with the questionnaires are presented to the participants. Participants are asked to reconsider their initial response in the light of the overall opinion. Repeat rounds are carried out until consensus has been reached. Within the present study, centers who are internationally recognized as specialists in BPBI were asked to evaluate the characteristics of each PRO measure by answering a structured questionnaire. A ten-point Likert scale, ranging from 1 (fully disagree) to 9 (fully agree), was used to answer the questions. The option of 0 "no opinion" was also available for selection. To analyze the results, the scores were grouped as follows: 1–3 was interpreted as the participant center being "not in favor/disagree" with the PRO's use in BPBI, 4–6 as "neutral" and 7–9 as being "in favor/agree" with the use of the PRO for BPBI patients. In addition, each center could add comments as free text to clarify their choice. Adapted from the 2018 iPLUTO model [8], the characteristics of the PRO evaluated were: (1) responsiveness to change, (2) ability to assess the current state of the child, (3) ease of application and (4) the specificity of the tool for BPBI. After each round, the questionnaire was returned to each specialist center together with the consolidated group responses of the previous round. As a result, the participating centers were given the opportunity to reconsider their opinion and re-score the PRO measures [21]. The results of all items from the previous rounds were summarized descriptively in table format (See Appendix 1).

It was predetermined that if >75% of the participating centers accepted or rejected a specific item, it would be considered as consensus agreement [21]. In reviewing the available measures and their psychometric evaluations in stage 1 of our methods, it was apparent that achieving consensus for many items would be difficult. Therefore, at the end of each round the participating centers were also requested to select the PRO measure that they thought would be the best to evaluate (a) children and (b) adolescents within the three domains (HR-QoL, functional status and pain). This question was designed to determine which PRO the BPBI specialist centers endorsed as the best available option in each category (i.e., HR-QoL, functional status, pain) of measures. The age groups and time of measurement was based on the 2018 iPLUTO study recommendations (i.e., ages 1,3,5,7 and 15). The online questionnaire was designed with NetQuestionnaires (Survalyzer Nederland B.V, Utrecht, The Netherlands, http://www.survalyzer.com/nl), responses were securely stored on the LUMC hospital server. Example contents of all three questionnaires and the full list of PRO measures are available as Appendices.

Participant recruitment

The participants from the first iPLUTO study were recruited via the XIX's International Symposium on Brachial Plexus Surgery (known as the "Narakas meeting") in February 2016. Additionally, the second iPLUTO study was announced at the Toronto Obstetrical Brachial Plexus Workshop in 2018. Both meetings are considered to bring together leading experts in BPBI from around the world. All participants were invited to participate via email. The combined email list of attendees to both meetings consisted of 359 addresses worldwide.

The first email to announce the start of the project was sent on 7th December 2018. Recipients were strongly encouraged to meet within their teams and designate one representative who would consolidate, record and input the teams' scores for each round. An email reminder was sent after a few weeks if no response was received, or if the questionnaire was not finished completely. During the first and second rounds, two reminders were sent. During the third round three reminders were sent. The software enabled the participating centers to save their answers and continue later.

Rounds

The first round took place from December 2018 to March 2019. Thirty-six centers completed the survey. The 36 centers that completed round 1 were sent an email invitation to participate in the second round between November 2019 and March 2020. It was emphasized that inputted responses should be reflective of the opinions from their whole multi-disciplinary team (MDT). Twenty-six centers responded to round 2. The third round took place between April and June 2020. In this round, participating centers were requested to show a preference between the two remaining PRO measures in each category that had not yet gained consensus. Thirty-five centers responded.

Results

Participant centers

A total of 359 invitations were sent to individuals by email. Thirty-five specialist centers across 6 continents, completed round 3 (see Table 1). A total of 41 representatives participated: 10 contributed to 1 round, 9 contributed to 2 rounds, and 23 contributed to all three rounds. Multiple results were not received from any centers. Full data on the responding specialist centers was available for the 23 participants who completed 3 rounds. These data were already collected during the first iPLUTO survey [8]. All 23 classified themselves as specialized "center" and had a multidisciplinary structure. Teams consisted of 3.1 surgeons on average (range 1–5), 1.8 non-surgical physicians like physiatrist or neurologist (range 0–10) and 3.6 therapists (either physical or occupational therapists, range 1–10). On average each team had 53 new referrals each year; surgical interventions were on average 15 nerve surgeries per year and 18 secondary surgeries per year.

Table 1. Locations of participating centers.

Continent	Round 1	Round 2	Round 3
Africa	1	0	1
Asia	4	3	4
Australasia	1	1	1
Europe	15	9	12
North America	11	10	12
South America	4	3	5
Total	36	26	35

Number of participating centers per continent.

Survey results

Delphi results from rounds 1–3 can be found in Tables 2, 3, 4 *and* Appendix 2. The final clinician preferred PRO measures for use in children and adolescents with BPBI can be found in Table 4.

Usefulness of PRO measure in BPBI practice

Consensus was achieved that PRO measures are useful for clinical and research practice in BPBI. Overall, 67% (mean rating 7.00) of

Table 2. Results for usefulness of PROs for clinical and research use.

	Round 1					Round 2				
	No opinion	1/2/3	4/5/6	7/8/9	Mean	No opinion	1/2/3	4/5/6	7/8/9	Mean
PRO useful for clinic	0	0	*n* = 12	*n* = 24 (67%)	7.00	0	0	0	*n* = 26 (100%)	8.42
PRO useful for research	0	0	*n* = 10	*n* = 26 (72%)	7.08	0	0	0	*n* = 26 (100%)	8.42

Number of rating (n) per Likert category, and mean in Round 1 and Round 2.
(1/2/3: 'not in favor/disagree', 4/5/6: 'neutral', 7/8/9 as 'in favor/agree).

Table 3. Preferred PRO measure, results for rounds 1 to 3.

In your opinion, when taking into account all of the merits and drawbacks, what is the best PRO measure to evaluate **Functional Status** for children (<10)/ adolescents (>10)

Round 1				Round 2				Round 3			
Best PRO for children		Best PRO for adolescents		Best PRO for children		Best PRO for adolescents		Best PRO for children		Best PRO for adolescents	
CHEQ	9	BPOM-SE	10	CHEQ	9	BPOM-SE	16	BPOM-SE	23	*see round 2*	
PODCI	7	PROMIS-UE	6	BPOM-SE	8	DASH	5	CHEQ	12		
HUH	6	DASH	6	HUH	4	PROMIS-UE	5				
BPOM-SE	6	CHEQ	5	PODCI	4						
PROMIS-UE	4	PODCI	2	PROMIS -UE	1						
ABILHAND	3	ABILHAND	3								
DASH	1	PEM-CY	1								
		HUH	1								
		MHQ	1								

In your opinion, when taking into account all of the merits and drawbacks, what is the best PRO measure to evaluate **Health-Related QoL** for children (<10)/adolescents (>10)

Round 1				Round 2				Round 3			
Best PRO for children		Best PRO for adolescents		Best PRO for children		Best PRO for adolescents		Best PRO for children		Best PRO for adolescents	
PedsQL	16	PedsQL	13	PedsQL	12	PedsQL	13	PedsQL	28	PedsQL	22
PROMIS-GH	6	EQ5D-Y	9	PROMIS-GH	10	PROMIS-GH	10	PROMIS-GH	7	PROMIS-GH	13
CHQ	3	CHQ	5	EQ5D-Y	4	EQ5D-Y	3				
EQ5D-Y	10	PROMIS-GH	5								
PROMIS-PR	1	PROMIS-PR	4								

In your opinion, when taking into account all of the merits and drawbacks, what is the best PRO measure to evaluate **Pain** for children (<10)/adolescents (>10)

Round 1				Round 2				Round 3			
Best PRO for children		Best PRO for adolescents		Best PRO for children		Best PRO for adolescents		Best PRO for children		Best PRO for adolescents	
FPS-R	17	BPI	9	FPS-R	22	BPI	11	*see round 2*		VAS	18
BPI	5	VAS	8	VAS	2	VAS	10			BPI	17
PROMIS-P_interf	4	APPT	8	BPI	2	APPT	5				
VAS	4	NRS	7								
APPT	2	PROMIS-P_qual	2								
PROMIS-P_intens	2	PROMIS-P_interf	1								
NRS	2	PROMIS-P_intens	1								

Number of votes pre PRO measure.
PRO - Patient reported outcome measure; CHEQ - Children's Hand-use Experience Questionnaire; PODCI - Pediatric Outcomes Data Collection Instrument; HUH - Hand Use at Home; PROMIS-UE - Patient-Reported Outcomes Measurement Information System – Upper Extremity; BPOM-SE - Brachial Plexus Outcome Measure – Self Evaluation; ABILHAND – ABILHAND; PEM-CY - Participation and Environment Measure – Children and Youth; DASH - Disability of the Arm Shoulder and Hand; MHQ - Michigan Hand Questionnaire; WOSI - Western Ontario Shoulder Instability; PROMIS-GH - Patient-Reported Outcomes Measurement Information System – General Health; PROMIS-Peer Rel - Participation and Environment Measure – Peer Relationships; PedsQL - Pediatric Quality of Life Inventory; EQ5D-Y - EuroQol5D – Youth; CHQ - Child Health Questionnaire; VAS - Visual Analogue Scale; NRS - Numerical Rating Scale; PROMIS-P_qual - Patient-Reported Outcomes Measurement Information System – Pain quality; PROMIS-P_interf - Patient-Reported Outcomes Measurement Information System – Pain interference; PROMIS-P_Intens – Patient Reported Outcomes Measurement Information System – Pain Intensity; APPT - Adolescent Pediatric Pain Tool; FPS-R - Faces Pain Scale – Revised; BPI – Brief Pain Inventory.

Table 4. Clinician preferred PRO measures.

	Children	Adolescents
Function	BPOM-SE*	BPOM-SE
HR-QOL	Peds-QL **	Peds-QL
Pain	FPS-R**	BPI
		VAS

Final matrix for PRO measures; HR-QoL - health related quality of life; BPOM-SE - Brachial Plexus Outcome Measure Self Evaluation scale; FPS-R - Faces Pain Scale-Revised; BPI - Brief Pain Inventory; VAS - Visual Analogue Scale; Peds-QL - Pediactric Quality Of Life* 5 Years old and above; ** 7 Years old and above.

participating centers in round 1 and 100% (mean rating 8.42) in round 2 agreed that PRO measures are useful for clinical evaluation and patient treatment. Seventy-two percent (mean rating 7.08) of participating centers in round 1, and 100% (mean rating 8.42) in round 2 agreed that PRO measures are useful for scientific evaluation (i.e., research practice) (Table 2).

Age of administration of PRO measure in BPBI practice

Based on the consolidation of data of all the PRO measures, participating centers thought that functional status PRO measures were most appropriate to administer between ages 5 to 15 years (52 – 65% agreed), and HR-QoL and pain PRO measures between 7 and 15 years (55 – 61% agreed) (Table 4).

Functional status PRO measure

The results of rounds 1 and 2 on functional status PRO measures indicate that consensus on a measure that is responsive to change and descriptive of the current functional status in BPBI was not achieved (Appendix 2). Consensus was achieved that the BPOM Self-Evaluation scale is feasible to administer in the clinic setting (77% agreed). In round 2, the BPOM Self-Evaluation scale was ranked highest as the best available functional status PRO measure for adolescents (16 votes, 62% agreed). The best available functional status PRO measure for children was not determined in round 2. Therefore, participating centers were asked to choose between the Children's Hand-use Experience Questionnaire (CHEQ) and BPOM Self-Evaluation scale; the two measures that had the most votes as the best available functional status measures for children in Round 2. In Round 3, 23 (66% agreed) respondents voted the BPOM Self-Evaluation scale as the best available functional status measure for children among the two remaining measures from Round 2 (Table 3).

Health-related quality of life (HR-QoL) PRO measure

The results of rounds 2 and 3 on HR-QoL PRO measures show that consensus on a measure that is responsive to change, feasible, and specific to BPBI was not achieved (Appendix 2). In round 2, PROMIS Global Health (GH) and PedsQL were ranked highest as the best available HR-QoL PRO measures. In round 3, participating centers were asked to choose between these two measures and the PedsQL was ranked highest as the best available HR-QoL PRO measure for both children (28 votes, 80% agreed) and adolescents (22 votes, 63% agreed) (Table 3).

Pain PRO measure

During rounds 1 and 2 consensus on a measure that is responsive to change as well as specific and descriptive of current pain status in BPBI was not achieved (Appendix 2). However, consensus was achieved that both the VAS (92% agreed) and FPS-R (77% agreed) were feasible PRO measures of pain. The FPS-R was subsequently ranked highest (22 votes, 85% agreed) as the best available pain PRO measure for children. In round 3, participating centers were asked to choose between the VAS and BPI, as their choice of best available pain PRO measure for adolescents. A clear choice between these measures was not achieved: VAS received 18 votes (51% agreed) and BPI received 17 votes (49% agreed) (Table 3).

Discussion

This study presents the results of the latest iPLUTO Delphi consensus study on the PRO measures that should be used to serially assess outcomes in BPBI. It extends the 2018 attempt to reach consensus on a core-set of outcome measures for BPBI. During the 2018 iPLUTO study no consensus was found with regards to the inclusion of specific PRO measures into a minimal dataset [8]. It is of note, that when questioned, only 19% of participants in the first iPLUTO study judged themselves as capable to evaluate and rate different PRO measures. Such unfamiliarity may have led to the lack of consensus and may have resulted from the respondents' background; as all but two respondents were surgeons and therefore PRO measures may not feature in their day-to-day practice [8]. Therefore, the present study specifically requested the inclusion of the whole multi-disciplinary treatment team (MDT) when considering and scoring each PRO measure. None of the submitted responses were duplicated within centers (i.e., only one response per round was received from each center despite the original list of participant emails containing individuals who resided in the same specialist center). As a result, the present study was able to infer that MDT specialist centers caring for children with BPBI strongly agree (100%) that the use of PRO measures in both clinical and research practices is advantageous.

Importantly, consensus was also obtained with regards to the best age in which PRO measures should begin to be administered: the agreed age to begin PRO measures of function was at 5 years, and 7 years for HR-QoL and pain.

Qualitative studies have provided great insight into the needs of children and families with health conditions by providing forum for their "voices" to be heard. In recent years many studies have reported perceived unmet needs from both parents/families and patients who have a lived experience of BPBI [9,19,20]. In particular, the need for improved evaluation HR-QoL [10], pain [11,20], as well as functional status [19] have been highlighted.

Achieving consensus on which PRO measures were best to evaluate HR-QoL, Functional Status, and Pain within this study was challenging due to the lack of established disease-specific PRO measures with regards to the BPBI population [15]. Respondents may have had difficulty rating the PRO measures presented in the survey because previous studies that use established PRO measures to assess BPBI cohorts are varied and no studies have formally evaluated the validity, reliability, and responsiveness to change of these measures as the child matures [15]. These limitations were echoed within the respondents' comments within the present study. Free text comments from some specialist centers reflected that the existing tools had poor specificity to the condition; so were not able to fully measure the functional ability or impairments within all of the joints of the upper limb. Furthermore, it is recognized that some tools focus on reach (which evaluates upper brachial plexus function), while others evaluate hand function (which is more seriously affected in children with a total plexus lesion). Finally, functional status can be evaluated in terms of capacity, capability, or performance; each with its own benefits and drawbacks [12]. The ideal

evaluation tool that covers all aspects of daily functioning does not seem to exist presently, but should be an important focus of further development in the field. Consequently, overall consensus relating to the descriptive and responsiveness to change properties of measures was not achieved. It was observed that tools which were short and easy to implement such as the FPS-R and BPOM SE were agreed upon due to their feasibility. This may also suggest a desire for tools which have good clinical utility and are easily administered within a busy clinic environment. However, in practice, the specific research question that is to be addressed will be decisive to which specific PRO should be employed.

A key strength of the present study is the large number of participating centers and their wide geographical distribution. In addition, all participants in round 1 through to round 3 reported that they resided in "brachial plexus centers." In all centers both surgical and allied health disciplines were represented; giving confidence that the whole MDT inputted their opinions into the question rounds.

Study limitations

The present study, however, is subject to some limitations. Although the initial invitation to participate was sent to attendees of highly specialized BPBI meetings, the first limitation is that participating centers were explicitly selected as specialist centers. However, presently no formal criteria exist to determine what constitutes to a center being deemed "specialist." The Canadian consensus guidelines advise that teams at multidisciplinary centers should include at least a dedicated therapist with experience in the assessment and treatment of BPBI and a peripheral nerve surgeon with experience in microsurgical repair of BPBI [22]. In this respect, all centers fulfilled these criteria. Equally determining "expertise" is also subjective and is usually denoted by how long an individual has worked within a particular field, their publication record and their previous caseload containing a particular condition [23]. The recruitment for the present study originated from special interest meetings which are attended by those with knowledge and experience of BPBI. However, there is no certainty that those completing the present Delphi would be considered "experts" by others. On the other hand, the authors are confident that respondents who registered themselves as a participating specialist center in the iPLUTO survey were residing in specialist centers with MDTs, which was confirmed by the reported patient workloads.

Secondly, it is recognized that despite specific and repeated instructions during each round emphasizing participating centers to include the whole MDT in the assessment process for the PRO measures, we did not verify that the survey responses were reflective of input of the whole MDT. Response-rate is difficult to discern for the first round as one participant from each center was asked to represent their colleagues and input data. There may have been several people from one center within the original contact list. However, response-rate for the second round was 72% and the third 97%. Sending automated reminders to the participants proved powerful.

The decision was made for the final round to ask participating centers to choose between the highest ranked PRO measures as opposed to the true Delphi scoring method due to the difficulty attaining consensus within the individual PRO scores. It was apparent that the respondent responses to the questions regarding current state, responsiveness to change, feasibility of use, and specificity to BPBI were too lofty to evaluate in the available measures to clearly indicate a preference. In retrospect the moderators could have been less ambitious in seeking consensus on specific PRO measure characteristics and minimized the questions that were included within the study at an earlier stage. Additionally, data collection on the disciplinary composition of each participating MDT would have supported the strength of our survey based on multidisciplinary opinions.

Finally, it is a limitation of this study that patients, families and patient organizations were not consulted. This would be of great benefit in future research.

Conclusions

This world-wide consensus survey aimed to produce a minimal dataset for PRO measures in young people who have a BPBI. The results of this study indicate that specialist clinicians agree that PRO measures are important to use both clinically and in research. We recommend the use of the FPS-R (VAS or BPI in adolescents), PedsQL and BPOM SE, as the best available practice. However, research to establish their validity for this patient group is necessary. Measures should begin to be implemented for children aged 5 years and above. The study identified that the existing PRO measures are insufficient in examining the multi-modal presentation of BPBI. To overcome this limitation, we feel that the development of a psychometrically sound, BPBI specific PRO measure that evaluates whole arm function, HR-QOL and pain is needed.

Acknowledgements

The authors thank all the centers and clinicians who participated in this study.

Disclosure statement

The authors report there are no competing financial or non-financial interests to declare.

Funding

The author(s) reported there is no funding associated with the work featured in this article.

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