

RESEARCH PAPER

## Initial constructs for patient-centered outcome measures to evaluate brain–computer interfaces

Elena M. Andresen<sup>1,2</sup>, Melanie Fried-Oken<sup>1,3</sup>, Betts Peters<sup>1</sup>, and Donald L. Patrick<sup>4</sup>

<sup>1</sup>Institute on Development & Disability, Oregon Health & Science University, Portland, OR, USA, <sup>2</sup>Department of Public Health & Preventive Medicine, and <sup>3</sup>Departments of Neurology, Pediatrics, Otolaryngology, and Biomedical Engineering, Oregon Health & Science University, Portland, OR, USA, and <sup>4</sup>Department of Health Services, University of Washington, Seattle, WA, USA

### Abstract

**Purpose:** The authors describe preliminary work toward the creation of patient-centered outcome (PCO) measures to evaluate brain–computer interface (BCI) as an assistive technology (AT) for individuals with severe speech and physical impairments (SSPI). **Method:** In Phase 1, 591 items from 15 existing measures were mapped to the *International Classification of Functioning, Disability and Health* (ICF). In Phase 2, qualitative interviews were conducted with eight people with SSPI and seven caregivers. Resulting text data were coded in an iterative analysis. **Results:** Most items (79%) were mapped to the ICF environmental domain; over half (53%) were mapped to more than one domain. The ICF framework was well suited for mapping items related to body functions and structures, but less so for items in other areas, including personal factors. Two constructs emerged from qualitative data: quality of life (QOL) and AT. Component domains and themes were identified for each. **Conclusions:** Preliminary constructs, domains and themes were generated for future PCO measures relevant to BCI. Existing instruments are sufficient for initial items but do not adequately match the values of people with SSPI and their caregivers. Field methods for interviewing people with SSPI were successful, and support the inclusion of these individuals in PCO research.

### Keywords

Augmentative and alternative communication, communication disorders, quality of life, severe speech and physical impairments

### History

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### ► Implications for Rehabilitation

- Adapted interview methods allow people with severe speech and physical impairments to participate in patient-centered outcomes research.
- Patient-centered outcome measures are needed to evaluate the clinical implementation of brain–computer interface as an assistive technology.

### Introduction

Jean-Dominique Bauby, the editor of French *Elle* magazine, experienced a brainstem stroke in 1995. He woke up from coma days later with locked-in syndrome (LIS), his entire body paralyzed aside from some movement in his head and eyes. Unable to speak, Bauby learned to communicate by blinking: he could blink to answer yes/no questions, or he could spell words using partner-assisted scanning (a communication partner recited the letters of the alphabet and Bauby blinked to indicate his desired letter). Using this painstaking method, Bauby dictated his memoir, *The Diving Bell & the Butterfly* (later adapted into a film), relating his experiences. He died in 1997.

Bauby's story illustrates the significant challenges faced by people with LIS or other disabling conditions causing severe speech and physical impairments (SSPI). Bauby, like many others

with SSPI, was dependent on a well-trained, patient partner to interpret and relay his messages. In recent years, brain–computer interface (BCI) technology has allowed users to control a computer (for spelling, Internet access or other functions) using only their brain waves, with no motor output required. BCIs are either noninvasive (with externally placed electrodes that detect brain signals through the scalp) or invasive (with implanted electrodes). Several spelling BCIs are available, including the Wadsworth BCI [1], the Berlin BCI Hex-o-spell [2], the IntendIX Speller ([www.intendix.com](http://www.intendix.com)) and the RSVP (Rapid Serial Visual Presentation) Keyboard™ [3]. There has been an explosion of interest in BCI as an assistive technology (AT) in the past decade [4,5]. Research groups around the world are developing BCI systems for AT applications, using a variety of signal acquisition techniques, control signals and user interfaces [6].

The BCI field has moved from the vision of a “thought translation device” for completely paralyzed patients [7] to an AT access method for long-term independent home use [1]. While BCI technology was in its infancy, it was not yet meaningful to ask questions about outcomes. Most BCI research efforts have been devoted to engineering challenges of stability, reliability and

Address for correspondence: Melanie Fried-Oken, PhD, Institute on Development & Disability, Oregon Health & Science University, 707 SW Gaines St., Portland, OR 97239, USA. Tel: +1 503 494 7587. Fax: +1 503 494 6868. E-mail: [friedm@ohsu.edu](mailto:friedm@ohsu.edu)

brain signal classification. However, now that people with disabilities are participating in home-based trials, it is time to start examining the technology from a patient-centered approach, with a focus on outcomes for functional use. Unfortunately, the voices of potential BCI users and other important stakeholders have been largely absent from the conversation guiding the future of this technology. Previous AT research has shown that a lack of patient-centered outcomes research (PCOR) can lead to technology abandonment [8,9]. As BCI continues to be developed and implemented in users' everyday lives, the technology needs to be evaluated using the current state-of-the-science, which is driven by incorporation of PCOR into the BCI field. PCOR requires the creation of a new generation of assessments, or at least the validation and refinement of current communication and AT tools with patient-generated constructs. By beginning this process now, the field will be ready when, in the near future, BCI devices are placed in users' homes for everyday use; both researchers and clinicians will be able to measure functional changes or quality of life (QOL) changes during technology implementation.

New initiatives have broadened and continue to define what PCOR includes [10–13]. In 2009, the US Food and Drug Administration published guidance for using patient-centered outcomes (PCOs) to evaluate treatment benefits [14]. The rationale for the transition to PCOR is that health care (interventions, medications, AT, etc.) is unlikely to achieve therapeutic goals if it does not match what patients themselves value and want. The ultimate goal of improving people's lives (health, QOL, etc.) is best met when the outcomes goals are those which the target group identifies as most valuable. These guiding principles for PCO measures are particularly challenging for current and future research involving individuals with SSPI. Researchers and clinicians may not understand how to communicate with these individuals, or may perceive such communication as too difficult. Whatever the reason, input from the SSPI population has been missing from instruments currently in use for communication-related outcomes research. Since BCI is a new AT for communication access for people with SSPI, it must be evaluated in terms of its effectiveness as well as its impact on the user's communication and QOL, following a PCOR model to ensure that this technology will meet the needs and wants of users.

Outcomes for AT may be defined as measures of the effects of AT on users' lives and environments [15]. The development of AT outcome measures has lagged behind the growth of the AT industry, in part due to an emphasis by developers on the *performance of the technology itself* rather than *users' performance with the technology* [15]. This pattern is reflected in the BCI field, in which research studies are more likely to report on computer simulations or healthy participants than on patient trials. Among the 186 abstracts presented at the 2013 International BCI Meeting, only 31 included participants with disabilities [16].

Many researchers call for the development and improvement of AT outcome measures, and for the inclusion of AT users in this process. Jutai and colleagues propose a taxonomy for AT outcomes emphasizing the effectiveness, social significance and subjective well-being associated with AT use, and they encourage the use of consistent language for outcomes reporting [17]. Demers and colleagues introduce a framework to address outcomes for caregivers of AT users and advocate for the creation of measures for this population [18]. The Assistive Technology Services Method recommends that the provision of AT services be "ultimately driven by the needs and wants of the [individual with disabilities]", but does not specify how to assess wants and needs, nor how to measure outcomes [19]. Scherer's Matching Person and Technology model supports patient-centered AT assessments, with the goals of finding the best technology for each individual's needs and reducing AT abandonment [9]. Its component scales

can be used during follow-up assessments to measure outcomes and identify any areas for adjustment or improvement, but it is primarily designed to guide professionals in making initial AT recommendations [20]. Researchers in the BCI field are beginning to take an interest in PCO measures, as well. Lorenz and colleagues discuss a holistic assessment of the user experience with BCI, examining both the usability and the appeal of the technology from the user's perspective [21].

The present pilot project was prompted by the need for PCO measures for BCI. The main goal of this project was to produce a research agenda for the generation of instruments that will objectively measure how well this new AT works for individuals with SSPI in functional settings. Secondary objectives were to demonstrate that individuals with the most severe speech and physical disabilities can participate in PCOR, and to propose a methodology for their inclusion. A modified version of the World Health Organization's *International Classification of Functioning, Disability and Health* (ICF) [22] provided an organizing framework for this project. Modifications included: the incorporation of health-related QOL, as proposed by Whiteneck [23]; extension of the participation component to include the concept of opportunity, as in Patrick's model [24] and consideration of values and experiences specifically related to BCI. PCOs domains were developed based on (1) existing instruments and (2) interviews with people with SSPI and their caregivers. In keeping with the principles of PCOR, the project explicitly considered patient-generated information as the "gold standard" for future measures.

## Methods

### Review of existing measures

A review of existing measures was conducted, and included instruments that were deemed important for research on AT devices (particularly for augmentative and alternative communication [AAC]) and communication interventions. Some of these instruments were used by Kübler and colleagues to measure satisfaction of BCI end-users [25]. Using personal knowledge, outreach to experts, literature review and modified electronic database search strategies, a total of 15 instruments were classified as useful for possible items or overall domains relevant to PCOR for BCI. These are listed in Table 1.

### ICF coding

The 15 instruments were mapped, item by item, onto the ICF. Each item was mapped to one or more broad ICF components (*b* for body functions and structures, *a* for activities and participation, *e* for environmental factors and *p* for personal factors [personal factors are not specifically coded by the ICF]) and to one or more specific codes when possible. For example, one item from the Quality of Communication Life Scale (QCL), "People include me in conversations", was coded as both *a* and *e* because it was judged to be relevant to the respondent's activities and participation involving conversation, and heavily dependent on others' attitudes and behavior, which are considered environmental factors. In addition, it was mapped to one specific ICF code, "Conversation", and one chapter (indicating a broad category of codes), "Chapter 4: Attitudes". A summary was constructed for each instrument, and for the total item group across all 15 instruments, producing a percentage of items that were mapped to each of the four ICF components.

### Patient-generated qualitative research phase

Three steps were implemented in the PCO methodology for this pilot study: (1) interview three cohorts of stakeholders, namely

Table 1. Mapping of items from existing assistive technology outcomes instruments to International Classification of Functioning, Disability and Health (ICF) components.

Instrument	Total items	Items relevant to each ICF component <sup>a</sup>				
		B	A	E	P	2+
RSVP Keyboard™ User Feedback Questionnaire (adapted from questionnaires by Bates [26])	13	6 46%	13 100%	13 100%	8 62%	13 100%
TLX: NASA Task Load Index [27,28]	6	3 50%	6 100%	0 0%	5 83.3%	6 100%
PIADS: Psychological Impact of Assistive Devices Scale [29,30]	26	0 0%	15 58%	26 100%	26 100%	26 100%
CHIEF: Craig Hospital Inventory of Environmental Factors [31]	25	0 0%	0 0%	25 100%	0 0%	0 0%
FABS-M: Facilitators and Barriers Survey-Mobility [32]	92	0 0%	71 77%	83 90%	0 0%	62 67%
FuNHRQOL: Function-Neutral Health-Related Quality of Life Measure [33]	57	25 44%	18 32%	29 51%	33 58%	35 62%
MQE: Measure of the Quality of the Environment [34]	109	3 3%	0 0%	109 100%	0 0%	3 3%
PARTS/G: Participation Survey/General [35]	14	3 21%	14 100%	11 79%	2 14%	14 100%
QCL: Quality of Communication Life Scale [36]	17	2 12%	15 88%	12 71%	14 82%	15 88%
QUEST 2.0: Quebec User Evaluation of Satisfaction with assistive Technology [37,38]	12	0 0%	0 0%	12 100%	0 0%	0 0%
SPARC: Survey of Participation and Receptivity in Communities [39]	54	3 6%	24 44%	47 87%	4 7%	22 39%
WHODAS 2.0: World Health Organization Disability Assessment Schedule [40]	43	4 9%	39 91%	5 12%	1 2%	5 12%
LwD: Living with Dysarthria [41]	50	39 78%	42 84%	19 38%	18 36%	39 78%
QUIS: Questionnaire for User Interaction Satisfaction [42]	27	0 0%	0 0%	27 100%	27 100%	27 100%
CPIB: Communicative Participation Item Bank [43]	46	0 0%	46 100%	46 100%	46 100%	46 100%
Totals across instruments	591	88 15%	303 51%	464 79%	184 31%	313 53%

<sup>a</sup>B = body functions and structures, A = activities and participation, E = environmental factors, P = personal factors, 2+ = two or more components.

individuals with SSPI who had used BCI and their paid and unpaid (family) caregivers; (2) identify an overall set of constructs and domains that are important to these stakeholders and (3) propose an initial set of constructs for a future family of outcome measures for BCI technology and implementation. The qualitative steps were intended to produce a preliminary list of overarching PCO constructs and component domains, with detailed themes that might generate items or groups of items for future measures.

### Participants

A convenience sample of eight participants with SSPI was selected from a pool of 12 participants in the parent RSVP Keyboard™ BCI study [3,44] funded by the National Institute on Deafness and Other Communication Disorders (NIDCD). Participants with total LIS were excluded because they have no reliable, consistent means of communication and cannot participate in qualitative interviews. Five of the participants were asked to suggest one or more individuals from their support systems who could join the paid or unpaid caregiver participant groups. As a result, three paid caregivers and four unpaid (family) caregivers were identified, for a total of 15 participants. Table 2 presents information on the eight adults with SSPI and their caregivers who participated in the qualitative in-person interviews. A person with SSPI and his or her paid and unpaid (family) caregivers are referred to as a “set” of participants, and are grouped together in the table.

Participants with SSPI included two women and six men, varying in age from 28 to 66 years old. Five individuals presented with neurodegenerative disease (either spinocerebellar ataxia or amyotrophic lateral sclerosis), two had congenital disorders (muscular dystrophy and cerebral palsy) and one had experienced a brainstem stroke. All presented with quadriplegia and required assistance with activities of daily living. Five participants lived in their family homes and three lived in group foster homes. All had been involved in at least one trial session with the RSVP Keyboard™ BCI before the current project interview, and one participant had participated in seven trial sessions. Their primary communication modes or methods varied: some used speech (sometimes in combination with other methods), but most used AAC methods, ranging from printed letter boards to high-tech, eye-controlled speech-generating devices (SGDs).

Family and paid caregivers were heterogeneous by demographics and the duration of their relationship with the person with SSPI. The unpaid caregiver group included three parents and one spouse. Paid caregivers reported that they had worked for the person with SSPI for 2–12 years, and were familiar with their communication abilities and needs.

### Settings and procedures

Participants with SSPI and their caregivers were interviewed separately, though individuals with SSPI could choose to have a caregiver present during their interviews to assist with

Table 2. Participant characteristics.

Participant <sup>a</sup>	Age range (in years)	Gender	PSSPI <sup>b</sup> residence	PSSPI <sup>b</sup> diagnosis	PSSPI <sup>b</sup> BCI research sessions prior to interview	PSSPI <sup>b</sup> primary communication strategy
1U	35–64	Male	Family home	Spinocerebellar ataxia	1	Letter board with finger pointing
1F	35–64	Female				
2U	35–64	Male	Family home	Brainstem stroke	4	Partner-assisted scanning
2F	65+	Female				
2C	35–64	Male				
3U	35–64	Female	Group foster home	Cerebral palsy	4	Keyboard-based SGD <sup>c</sup> with hand brace stylus
3C	18–34	Male				
4U	65+	Female	Family home	ALS <sup>d</sup>	7	Tablet-based SGD <sup>c</sup> with foot-controlled trackball
4C	35–64	Female				
5U	35–64	Male	Group foster home	ALS <sup>d</sup>	5	Speech
5F1	65+	Female				
5F2	65+	Male				
6U	18–34	Male	Group foster home	Duchenne muscular dystrophy	5	Speech
7U	35–64	Male	Family home	ALS <sup>d</sup>	5	Tablet-based SGD <sup>c</sup> with head mouse
8U	35–64	Male	Family home	ALS <sup>d</sup>	6	Tablet-based SGD <sup>c</sup> with eye tracking

<sup>a</sup>U = BCI user/person with SSPI, F = family member/unpaid caregiver, C = paid caregiver.

<sup>b</sup>PSSPI = person with SSPI.

<sup>c</sup>SGD = speech-generating device.

<sup>d</sup>ALS = amyotrophic lateral sclerosis.

communication. All interviews were conducted in a location of the participant's choice, commonly at the residence of the person with SSPI. One to three researchers were present at each interview, with one researcher designated as the primary interviewer for each session. Interview questions were provided to all participants in advance. Participants with SSPI were encouraged, but not required, to compose written answers and provide them to the researchers before or during the interview. All participants used their preferred communication methods during interview sessions. For those who used AAC, communication procedures were clarified before or during the interview as needed. For example, some participants appreciated when the interviewer guessed a word or phrase as they were typing, while others preferred to type an entire message without guesses. Some participants used the synthesized speech output on their SGDs, while others preferred an investigator to read text on the device screen. A digital voice recorder was used to record interviews as Waveform Audio Format (WAV) files, which were later transcribed. To ensure that silent forms of communication were represented on the audio recording, an investigator verbalized such utterances when possible (e.g. reading aloud text typed on a screen or speaking yes/no responses given by eye blinks or gestures). Investigators produced field notes describing the context, their observations and summary of the content and procedures within two days of each interview session. Details about non-verbal forms of communication (e.g. facial expressions, gestures) were considered a form of response and captured in field notes. One investigator reviewed all transcripts for accuracy, and to remove names and other identifying information before coding and analysis. The interview protocol was approved by the Institutional Review Board and all participants provided informed consent.

#### Interview structure

The interview protocol was developed based on standards for qualitative interviewing and formative research [45–47]. Interview content was derived from (1) review of existing instruments and their content, (2) review of ICF chapters by

clinical team members and (3) investigators' clinical expertise with the diverse target population of people with SSPI, their family members and paid caregivers. A set of open-ended questions was developed, beginning with broad questions asking participants to describe a "good day" and a "bad day", intended to orient participants to the importance of their experiences and perspectives without explicit constraints on their ideas. Subsequent questions and probes focused on topics directly linked to communication, AT and QOL. Interview questions were modified for use with caregivers, in order to elicit these participants' own opinions rather than speculation on the opinions of the person with SSPI. After review of the first three interview sets, a rating exercise was added to identify which topics were most important to participants with SSPI. One interviewer kept a list of primary issues that emerged in each interview, verifying the list with the participant at the end of the interview. The participant then rated the importance of each issue, from 1 (not at all important) to 7 (extremely important). If participants rated multiple topics as 7, the full list was reviewed, and the participant was asked to identify the most important among these topics.

#### Text and theme coding

Interview transcripts and field notes were coded to identify constructs and domains representing important outcomes to participants. The methods were iterative. Two investigators initially worked independently with documents from one set of participants (i.e. transcripts and field notes from interviews with a BCI user and his or her caregivers). They identified salient quotes from each interview in the set, then coded the quotes according to their important themes. Many quotes were coded with multiple themes. Quotes and their associated codes were then organized into a table to facilitate additional editing during iterative steps. Investigators met and compared their initial codes, also reviewing the common themes from the BCI user rating exercise. Using this initial code list, each investigator independently coded half of the remaining data sets. At a reconciliation meeting, they reviewed each other's work and agreed on consensus codes across the data sets. After creating an initial framework of constructs, domains

and themes, they reviewed their code tables and selected representative text, refining their code list and tables, if necessary. The resulting framework was summarized, with both coders selecting and agreeing on exemplary quotes from the interviews. In the final step, the resulting framework and its components were reviewed by all four research team members.

## Results

### Instrument review

#### *Instrument codes*

Investigators coded 591 items across 15 instruments. Individual instruments varied in length from six items (the NASA Task Load Index [NASA-TLX] [27,28]) to 109 items (the Measure of the Quality of the Environment [MQE] [34]). Each item was mapped to one or more of the four main components of the ICF: body functions and structures, activities and participation, environmental factors and personal factors [22]. More than half of the 591 items (53%) were mapped to more than one ICF component. For example, one item from the QCL [36], “It’s easy for me to communicate”, was mapped to all four components. This pattern of mapping was due to implied associations; ease of communication may depend on body functions and structures (e.g. ability to speak or to access a communication device), activities and participation (e.g. the types of situations in which the individual needs or wants to communicate), environmental factors (e.g. the ease of use or effectiveness of a communication device or the patience and skill of communication partners) and personal factors (e.g. the individual’s personality traits or perspective of what it means for communication to be “easy”). Among the 15 measures, the environmental factors component had more items mapped to it ( $n = 464$ , 79%) than any other component. This was not surprising because the candidate measures were chosen for their focus on AT, which is coded as an environmental factor. Environmental factors were followed by activities and participation (51% of items), personal factors (31%) and body functions and structures (15%). See Table 1 for a summary of how items from each instrument mapped to the ICF components.

Items also were mapped to specific ICF codes, when they captured ICF elements. Many items did not fit easily into the ICF framework, and could be mapped only to “unspecified” categories (e.g. products and technology, unspecified) or to broad domains (e.g. communication). The ICF framework was well suited for mapping items related to body functions and structures (e.g. “Did you experience physical pain?” or “Did you pay attention well?” from the Function-Neutral Health Related Quality of Life [FuNHRQOL] Scale [33]), but less so for other items of great importance to people with SSPI, most notably environmental factors related to AT, QOL, opportunity and personal factors.

Within the environmental factors component, the ICF domain, products and technology, contains a code, assistive products and technology for communication, which could describe BCI-based communication systems or other AAC devices. However, the ICF provides no means of coding specific attributes of such systems, communication effectiveness or user satisfaction with them. Several instruments, including the Psychosocial Impact of Assistive Devices Scale (PIADS) [29,30], the Quebec User Evaluation of Satisfaction with assistive Technology (QUEST) 2.0 [37,38] and the Questionnaire for User Interaction Satisfaction (QUIS) [42] contain items focused on these specific attributes.

### Patient-generated qualitative research phase

A total of 14 interviews were conducted for this pilot project (two unpaid caregivers, parents of the same individual with

SSPI, were interviewed together by their choice). Two participants with SSPI relied on their paid and/or unpaid caregivers during the interview to assist with communication. Most interviews were completed in a single one- to two-hour session, although two participants with SSPI required two sessions each due to fatigue and/or the slow speed of their communication methods. Because of the varied communication methods used by participants with SSPI, even interviews of approximately equal duration resulted in very different text files in terms of their length, as well as the density and complexity of the content. In some cases, interviewers restated participants’ comments and checked for accuracy, especially when the depth or clarity of responses was limited due to slow communication speed. Importantly, one or more researchers experienced in AAC attended each session, and their in-depth understanding of communication technologies helped them to provide specific probes when responses required clarification. For several interviews, participants had prepared text beforehand and these prepared answers were used as a starting point, followed by probes for additional content and details.

#### *Initial constructs*

An iterative process with consensus building resulted in preliminary codes applied to salient quotes from all text data. Two distinct constructs emerged from the coded data: (1) QOL and (2) AT. The QOL construct included seven domains: (1) Social participation and opportunity, (2) Communication, (3) Roles, (4) Relationships, (5) Emotions and attitudes, (6) Environment and (7) Physical health. The AT construct included three domains: (1) Function, (2) Design and (3) Support. Table 3 lists the constructs, domains and component themes.

Some domains in the QOL construct (e.g. relationships) also appear in other QOL and health-related QOL measures (e.g. the WHOQOL measures [48,49]). However, the domains generated are considerably different from the measures reviewed previously (Table 1). Some domains overlapped with ICF chapters though a majority of themes that make up these domains are not coded in the ICF, or are coded in terms that do not allow for detailed description of how these domains affect QOL. For example, the ICF includes fairly coarse groupings for participation elements, while the framework emerging from this study comprises separate domains for social participation and opportunity, roles and relationships, each of which incorporates a far richer set of themes than are coded within the ICF. Representative quotes for two domains are presented below.

#### *Social participation and opportunity*

Component themes in this domain focused on various types of social participation, such as recreation, spirituality, community involvement and entertainment. Many participants discussed the importance of social participation, and how their opportunity and level of participation is affected by their communication abilities or limitations. One man related how his social life has changed now that he is living with a disability, and how a good day involves “talking about stuff we see on TV and the news, and what’s going on in my parents’ life and my wife’s life. ‘Cause I kind of live through other people, pretty much”. A spouse described how she spends quality time with her husband now that he has difficulty in communicating, “Sometimes we go for a ride in our little golf cart and enjoy the sunshine, or we sit on our patio and enjoy water features, you know, have a beer, have an adult beverage... We’ve always liked gardens, and landscapes, we like outdoor stuff, so we could sit... And we don’t need to talk”.

Table 3. Summary of constructs, domains and component themes derived from interviews with people with SSPI and their caregivers.

Constructs	Domains	Component themes
Quality of life	Social participation & opportunity	Recreation
		Spiritual
	Communication	Community
		Entertainment
		Expressive
		Self-expression
		Independence
	Roles	Social interaction
		Interaction with healthcare providers
		Professional/volunteer
Civic		
Relationships	Family/parenting	
	Caregiving	
	Home	
	Family	
Emotions & attitudes	Friends	
	Caregivers	
	Intimate	
	Self-worth	
	Motivation	
	Attitudes (self)	
Environment	Attitudes (others)	
	Affect symptoms	
	Safety	
	Privacy	
	Finances	
	Living situation	
	Caregivers & support	
Physical & health	Pain	
	Variability	
	Personal care	
	Function	
	Transition	
	Reliability	
	Communication speed	
	Applications	
	Ease of use	
	Portability	
Assistive technology	Function	Appearance
		Software (user interface, output, etc.)
	Design	Invasive versus noninvasive
		Comfort
		Hardware & setup requirements
	Support	Technical support
		Training (user)
Training (caregiver)		

### Communication

Interviews touched on many aspects of communication, including the identified component themes of expressive communication (communication performance), self-expression (the ability to express one's personality or views), independence with communication, social interaction and interaction with healthcare providers. Participants recounted both benefits and drawbacks to their AAC systems; one man said, "Even though my current device allows me to communicate with my kids, it only brings out a very minimal portion of who I really am". Another participant stated, "Holding a normal conversation would be ideal... It takes spontaneity. Obviously, [my SGD] helps a lot with my communication. But it takes time". The role of other people in effective communication was another common topic. As one participant stated, "If you ask the question, then you must be patient with me and wait for me to type the answers for you. Many people don't have the patience to wait. Doctors are the worst at waiting".

### Adaptation and response shift

Aspects of the theory of response shift (a theory about how people's views and expectations related to health and QOL change over time as they adapt to their condition [50]) also appeared among several domains. For example, during one interview with a participant and his parent, both noted that he had little interest in independent communication during the time shortly after his acute event (a brainstem stroke). However, over the years his desire for independence in communication had increased, and he suggested that this issue should be reassessed regularly in people with SSPI to see if their interest or motivations had changed. Another participant responded to a probe about his relationships by saying, "Since my disease came later in life, [the change in my ability to communicate] has not had any effect on [my relationships with family and friends]. If I had started life without the ability to communicate, well, it would be just part of my life... [now] it's harder to make new friends. Not because of the communication. But people are more standoffish when you have

Table 4. Results from an interview rating exercise with a man with SSPI.

Topic, domain, or theme (emerging code assignments)	Rating (1–7) <sup>a</sup>	Most important topic
Independent communication	7	
Participation with family	7	X
Work role	7	
Privacy	3	
Social civic participation	5	
Isolation	4	
Choices/control for reaching out	6	
Finances & choices	7	

<sup>a</sup>1 = ‘not at all important’, 7 = ‘extremely important’.

something like this [disease]... Perception is the truth to he or she that perceives’. While this quote also fit with themes about relationships and attitudes, it demonstrated the potential importance of further exploring adaptation and response shift concepts in a future PCOR research effort.

#### Rating exercise

The rating exercises completed by five participants with SSPI provided another source of data for identifying themes and domains. Table 4 shows results of the rating exercise for one man with SSPI. Among four topics the participant rated as extremely important with a numerical score of 7 (independent communication, participation with family, work role and finances & choices), he chose ‘participation with family’ as the most important for him. All items from these lists were examined during coding, and some common issues were expected to be rated highly. A few participants did not name a single top choice, instead identifying multiple issues as being of the utmost importance. Table 5 summarizes all issues that participants rated as 7, noting items that were chosen as the most important. Several participants talked about communication device issues, including future BCI technology issues, as among their most important, underscoring the need for a separate construct measuring AT outcomes. The majority of important issues revolved around personal factors (e.g. attitude and motivation), the importance of family and other caregivers, and their roles. The heterogeneity of issues and ratings was substantial among these five participants.

#### Discussion

This pilot project generated a preliminary set of constructs, domains and themes for future PCOs research relevant to BCI as an AT. Investigators found that existing instruments do not contain sufficient detail and content related to these areas of interest. In particular, personal factors (a component not coded by the ICF) were often important to participants’ communication, AT use and QOL. Aspects of participation, while present in the ICF and in a number of other instruments, are represented in vague terms that do not sufficiently capture outcomes important to people with SSPI. Some of the existing instruments reviewed in this pilot study are well-suited for measuring one or more domains proposed in the preliminary set. For example, several instruments included items relevant to the Environment domain, including the Craig Hospital Inventory of Environmental Factors (CHIEF) and the MQE. Measures such as the Communicative Participation Item Bank (CPIB) and QCL, evaluate factors within the Communication domain. The PIADS and the Quebec User Evaluation of Satisfaction with assistive Technology (QUEST 2.0) may be useful for examining AT-related outcomes. While these existing tools are valuable in current research and clinical practice, and may fit partially within the constructs and domains

Table 5. Summary of participant-generated issues rated 7 (‘extremely important’ on a scale from 1 to 7) by five people with severe speech and physical impairments (SSPI).

Participant	Topics rated 7 (‘extremely important’)
4U	<b>Physical health</b> <b>Mental health</b> Intimate conversation Caregiver support Getting out of the house Personal (SSPI) communication effort
5U	<b>Participation with family</b> Independent communication Work role Finances & choices
6U	<b>BCI ease of setup</b> BCI portability, long battery life
7U	<b>Family</b> Personal attitude Reliability of communication device
8U	<b>Family</b> <b>Paid caregivers</b> <b>Parenting</b> Independence Getting out of the house Work role Direct (SSPI) care Communication ease Basic functions like food Household role

Issues in bold font are those each participant chose as the most important of all issues with a 7 rating.

identified here, they do not adequately match the values of people with SSPI and their caregivers. New constructs and domains have been identified and will be refined and developed further. New instruments are required to fully address all areas that are important to the various stakeholders of BCI technology.

The results suggest a number of future steps. First, there must be a larger and heterogeneous sample of people with SSPI to generate sufficient qualitative data to provide a more complete framework for the relevant constructs, domains and the content of PCO measures. Coded data generated some repetition, but included content that was not duplicated among the eight participants. The heterogeneity of the issues raised by participants and the variation in ratings were substantial, suggesting that these pilot findings have not reached a point of saturation (sufficient repetition of common themes) [12,51]. A future project with a larger participant pool will ensure that saturation is achieved, and may suggest areas that require additional inquiry for the final battery of PCO measures. Elements representing the theory of response shift [50] also emerged, and could be explored in a larger study or developed as a separate domain for a future battery of PCO measures. The adapted rating exercise was very useful in gauging potentially critical content for future measures, and should be continued in an expanded study on this topic. In addition, future research should identify what people with SSPI consider to be an important change in these critical elements to set measurement metrics for PCOR. A research sample from several sites conducting similar research would be needed to generate a sufficient number of participants with SSPI for future formative PCOR efforts. Importantly, a large pool of participants will be required for the quantitative phases of testing draft instruments.

Secondly, additional data are needed from paid and unpaid caregivers, as well as other BCI stakeholders. Besides the patients themselves, family and paid caregivers provided exceptionally useful data, but the sample size in this preliminary study was too

small to ensure saturation or obtain sufficient details for constructs, domains and themes. Two important groups were not interviewed in this pilot study: providers and BCI system developers (engineers). Healthcare providers knowledgeable about SSPI will be the future prescribers of BCI, and have an important perspective. While the content must be patient-centered and generated, clinical goals have been the drivers of most current outcome measures, and will continue to be valuable for future PCO measures. Engineering and development perspectives also provide key information about future outcome measures because design features will be based, in part, on BCI user feedback. Qualitative sessions with BCI engineers might be more successful with a more directed forum reporting on summaries of prior qualitative analysis from interviews with people with SSPI and their caregivers. The feasibility of meeting BCI user expectations might be a focus of these sessions.

Finally, review of an expanded set of existing outcomes and health-related QOL measures may continue to build the item base that can serve as an underlying pool of potential items matching patient-generated content. A recent analysis comparing and contrasting the domains and extensive item banks of the Patient Reported Outcomes Measurement Information System (PROMIS) with the ICF may serve as a useful resource [52]. The review identified key deficits in PROMIS coverage of some ICF content, notably for the ICF chapter related to communication, but PROMIS item banks may provide some important content to introduce into future emerging outcome measures for people with SSPI. Suggestions for additional instruments for review should be solicited from an expert panel of outcomes researchers familiar with the ICF, as was accomplished in the PROMIS-ICF analysis [52]. In this pilot study, patient-generated themes were not directly matched to the pool of items examined. The initial item pool and ICF coding from this study can serve as a future resource when augmented with a future larger set of candidate items. Item-generating methods used by Krahn and colleagues [33] might be extended to compare and contrast existing items with the content generated by people with SSPI and other BCI stakeholders.

### Broader implications for PCOR

Preliminary findings from this pilot study have implications beyond the realm of PCO measures for BCI. They indicate the effectiveness and utility of including people with SSPI in PCOR. Eight individuals with SSPI, with a variety of ability levels and communication methods, participated in qualitative interviews and provided valuable data that will help to shape the future of BCI as an AT. They offer different perspectives than their caregivers. Soliciting their viewpoints is the only way to ensure truly patient-centered outcomes for this population. This pilot study demonstrated that typical interview procedures can be modified to accommodate people with SSPI and elicit their full participation. Primary interview questions should be provided in advance, with an option to prepare typed or written answers. This reduces the time constraints on participants during the interview itself, and may encourage longer, more detailed responses. Increasing the length of each interview session and planning for additional sessions should be standard procedures when interviewing people with SSPI. When feasible, interviews with people who use AAC should include an investigator familiar with AAC devices and techniques. Non-verbal responses, such as facial expressions, head nods and gestures, must be acknowledged and accepted. When relying on audio recordings of interviews, these responses must be translated into audible answers. Investigators should clarify the interviewee's preferences regarding communication etiquette. Some individuals with SSPI prefer that their communication partners

complete their words or predict phrases or letters as they form messages. Others expect communication partners to patiently wait until a message has been completely formed. This process must be discussed and agreed upon before the interview takes place. Interviewers should be prepared to verbalize silent responses (e.g. yes/no gestures or blinks), read aloud text that is produced on a screen but not spoken by a computer, serve as a facilitator for partner-assisted communication methods and ask appropriate follow-up questions or probes for added detail. Family members or other caregivers who are familiar with the interviewee's communication methods may facilitate effective communication, if the person with SSPI is agreeable to their presence. Video recording may be more suitable than audio recording for data collection with this population, as it preserves the inaudible response modalities. If video recording is not possible, investigators should keep detailed notes of all participant responses.

### Limitations

Only participants with SSPI who have a consistent, reliable means of expression could participate in the qualitative interviews. Two of the initial participants, unfortunately, did not meet this inclusion criterion. Even with modified interview techniques, some potential BCI users, such as those with total LIS, are unable to provide input on the development of PCOs. At this time, investigators must rely on their proxies, including family members and paid caregivers, for insight and information. It is hoped that BCI may one day provide a reliable means of communication for the total LIS population so that they too can participate in PCOR.

Due to the limited time and resources available during this pilot study, the authors could not interview caregivers for three of the eight participants with SSPI. These caregivers may have shared different experiences, opinions and priorities than the seven caregivers who were included. Future expansion of this project will include interviews with a larger set of paid and unpaid caregivers to ensure a robust set of results and saturation of data to help to guide the design of new PCO measures.

Other BCI stakeholders who were not included in this pilot study would offer insights for PCOR, as well. Health care providers who evaluate and treat individuals with LIS, especially physicians and therapists, may describe important domains that have not been addressed yet. Other family members, such as siblings, may have experiences, opinions and priorities that have not been stated yet. The inclusion of these stakeholders will strengthen the validity of PCOR for BCI and the instruments that eventually are derived from this work.

### Conclusion

The PCOR framework generated from the qualitative interviews with people with SSPI and caregivers has face validity as a starting point for a future battery of PCO measures for use in BCI research. At present, the framework suggests two overarching constructs (QOL and AT), each of which includes component domains. These domains might generate either subscales of an overall measure (e.g. a multi-item and multi-scale measure of QOL), or require further development as independent measures (e.g. a measure on social participation and roles with subscales on key component themes). It is clear that the voices of people with SSPI and their caregivers can and should be included in BCI research.

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