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Towards a definition of neurodisability: a Delphi survey

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AIM The aim of this study was to develop, systematically, a consensus-based definition for 'neurodisability' that is meaningful to health professionals and parents of children with neurological conditions.
METHOD A multidisciplinary group of health professionals was recruited through child development teams and professional societies in the UK; several parents of children with neurological conditions worked with the research team. Professionals participated in three rounds of a Delphi survey. Participants rated their agreement with a proposed definition in each round, and feedback was used to refine the definition. Finally, a perspective was sought from international experts.
RESULTS Responses to the three rounds were as follows: round 1, 245 out of 290 (84.4%); round 2, 242 out of 300 (80.6%); and round 3, 237 out of 297 (79.7%). Agreement with the proposed definition was extremely high in every round (89.0%, 90.1%, and 93.6% respectively). The final version of the definition was widely endorsed among professionals, parents, and a small number of international colleagues. The final definition is as follows: 'Neurodisability describes a group of congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations. A specific diagnosis may not be identified. Conditions may vary over time, occur alone or in combination, and include a broad range of severity and complexity. The impact may include difficulties with movement, cognition, hearing and vision, communication, emotion, and behaviour'.
INTERPRETATION An agreed definition of neurodisability will be useful for conducting research or clinical evaluations with people affected by neurological problems.

'Neurodisability' is a term commonly used in the UK for a range of functional problems and diagnoses of neurological origin. There is a subspecialty of paediatric training devoted to neurodisability;[1] however, the term is not defined and there is no agreement as to which conditions are included. The term is used as a way of describing a group of conditions that share similar problems, including health and educational needs, and that are commonly managed by the multidisciplinary teams skilled in multisystem disabilities. The lack of a definition of neurodisability, or clarity about which conditions are included, can hinder effective communication, especially when considering health outcomes, and planning and evaluating multiprofessional teams and care pathways.

Individually, many conditions that result in a neurodisability are rare, but when grouped together they are common. Based on the Family Resource Survey (2004–05), there are an estimated 952 741 'disabled' children in the UK, which is 7.3% of the population of children aged 0 to 18 years (8.8% male and 5.8% female).[2] However, the survey used a definition that includes any long-term health conditions such as diabetes, arthritis, and asthma. Without a clear and agreed definition, it is not possible to derive a consistent and reliable assessment of the prevalence of neurodisability to ensure appropriate resource planning.

Neurodisability appears to be a UK-specific term; in other English-speaking countries the term 'neurodevelopmental disorders' is used to describe children with similar conditions. For instance, we found a definition of 'neurodevelopmental disorders' as 'disorders where motor, cognitive, behavioural, and/or language functioning are affected by central nervous system impairments, resulting in a variety of challenges associated with ambulation, information processing, self-regulation and communication'.[3] This and perhaps other definitions exist, but, to our knowledge, none is widely known, agreed, or used consistently.

As part of a research study examining what health outcomes should be measured for children affected by neurodisability in the UK NHS,[4] we required a definition that was acceptable and meaningful to both families and health professionals. This study sought to reach a consensus among a multidisciplinary group of health professionals

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and parents in the UK on a definition of neurodisability. In addition, we also consulted international experts on our resulting definition.

METHOD

Family involvement

The Peninsula Cerebra Research Group involves families of disabled children[1] in all aspects of research through a family faculty. An invitation to collaborate in this research was e-mailed to parents on the Peninsula Cerebra Research Group family faculty database; five parents volunteered to be involved in the team for this study. The parents' children had various neurological conditions. The parents were involved in all or some stages by participating at meetings and contributing by e-mail as the project progressed. Parents helped formulate and draft the initial version of the definition; they reviewed each subsequent version revised following feedback from health professionals, and commented on the clarity of the definition and the utility of including examples. Involving parents, and taking into account their feedback, ensured that they felt they could recognize their child's condition in the definition.

Delphi survey

The sample of health professionals was recruited initially through an invitation sent to the lead contacts at child development teams in England, with a request to pass on the invitation to their relevant colleagues. In a subsequent phase of purposive sampling, which was conducted in order to recruit representatives of under-represented professions, several professional societies agreed to forward invitations to their members. Professionals volunteered to take part in the Delphi survey by identifying themselves to the research team.

The views of the multidisciplinary group of health professionals were then gathered using three rounds of an online Delphi survey, an established method for seeking consensus,[5] following recommended procedures.[6] In each round, participants were e-mailed a unique link to an online survey. If they connected, they were presented with a version of the definition and asked to rate their agreement with the definition on a four-point response scale, ranging from strongly disagree to strongly agree. There was a free text box opportunity to make comments and suggestions. Non-responders were sent reminder e-mails, which included an option to withdraw from the survey. A priori, we used a criterion of 67% approval (agree or strongly agree) among participants to consider whether the definition was generally acceptable.[6] Suggestions from participants in each round were categorized and modifications to the definition were considered.

What this paper adds

- A definition of neurodisability for which there appears to be broad agreement among health professionals and parents of children with neurological conditions.
- The definition will enable people with neurodisability health conditions to be grouped for research, evaluation, and other clinical purposes.

Once an acceptable definition was achieved among the survey participants, perspectives from other countries were sought by sending an invitation and link by e-mail to the Delphi survey to (1) country leads for the European Academy of Childhood Disability, and (2) *CanChild* International Collaborators. We sought a rating of agreement or disagreement with the definition, comments, and details of any terms synonymous with neurodisability used in their countries or languages.

RESULTS

In total, 309 health professionals registered their interest in participating in the Delphi survey; registrants identified themselves as being from a range of professions. Although we had principally targeted English health professionals, there were 14 registrants from outside England. Their views on the definition were deemed eligible and they were retained in the survey. Responses to the three rounds were as follows: round 1, 245 out of 290 (84.4%); round 2, 242 out of 300 (80.6%); round 3, 237 out of 297 (79.7%; Table I and Fig. 1). Agreement with the proposed definition was extremely high in every round (89.0%, 90.1%, and 93.6%, respectively), far exceeding the a priori

Table I: Participants at baseline and responding to each round of the survey, by profession

Professional group	Round 1 *n* (%)	Round 2 *n* (%)	Round 3 *n* (%)
Audiologist	1 (0.4)	1 (0.4)	1 (0.4)
Child and adolescent psychiatrist	5 (2.0)	5 (2.1)	5 (2.1)
Child development worker	1 (0.4)	1 (0.4)	1 (0.4)
Neurosurgeon	1 (0.4)	2 (0.8)	2 (0.8)
Nurse	10 (4.1)	10 (4.1)	6 (2.5)
Occupational therapist	14 (5.7)	15 (6.2)	14 (5.9)
Ophthalmologist	0 (0.0)	1 (0.4)	1 (0.4)
Orthopaedic surgeon	6 (2.4)	5 (2.1)	7 (3.0)
Orthotist	20 (8.2)	20 (8.3)	19 (8.0)
Paediatric neurologist	7 (2.9)	9 (3.7)	8 (3.4)
Paediatric oncologist	1 (0.4)	1 (0.4)	1 (0.4)
Paediatric surgeon – other	2 (0.8)	2 (0.8)	2 (0.8)
Paediatrician in neurodisability	4 (1.6)	3 (1.2)	4 (1.7)
Paediatrician	79 (32.2)	75 (31. 0)	75 (31.6)
Physiotherapist	66 (26.9)	64 (26.4)	66 (27.8)
Prosthetist	2 (0.8)	2 (0.8)	1 (0.4)
Psychiatrist	1 (0.4)	1 (0.4)	0 (0.0)
Psychologist	8 (3.3)	8 (3.3)	7 (3.0)
Rehabilitation physician	1 (0.4)	1 (0.4)	1 (0.4)
Speech and language therapist	15 (6.1)	15 (6.2)	15 (6.3)
Teaching assistant	1 (0.4)	1 (0.4)	1 (0.4)
Total	245	242	237

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[1] We use the term 'disabled children' deliberately. Generally we prefer 'person-first language' as it is more appropriate to describe people 'with' or who 'have' specified characteristics, such as impairments or specific diagnoses. However, consistent with the International Classification of Functioning, Disability and Health, disability is created as a consequence of interaction between a person and their environment. Disability cannot be considered as intrinsic to the person. Hence, we believe that people are in fact disabled, and not 'people with disabilities'.

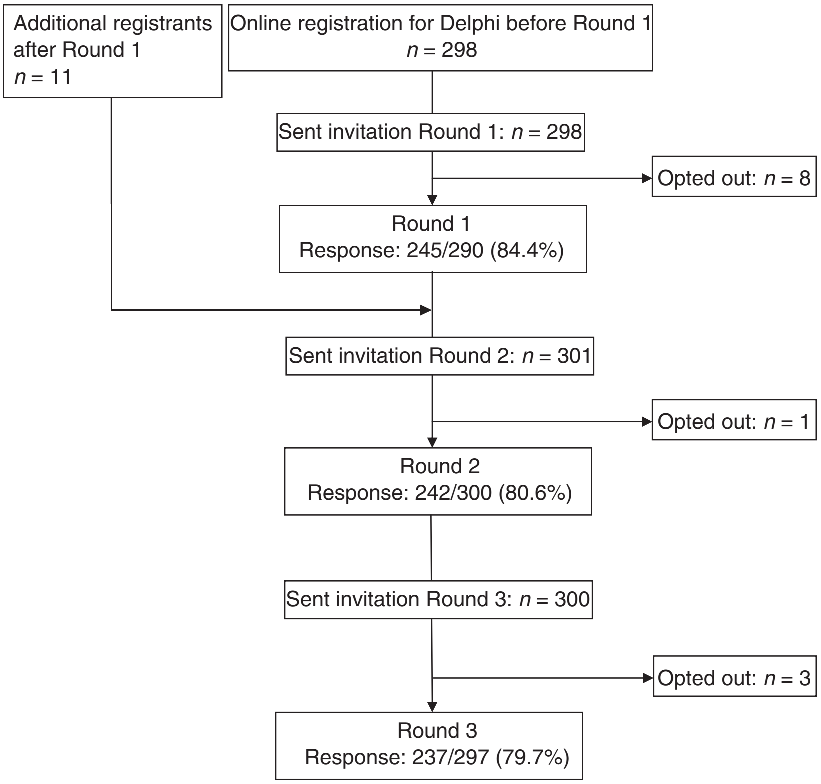

Figure 1: Flow chart showing participants in the Delphi survey at each stage.

expressed criteria of 67%. However, there were a number of queries, comments, and suggestions that informed revisions following each round.

In round 1, participants raised queries about whether both congenital and acquired conditions were included, and requested that it made more explicit that these were long-term conditions with a broad range of severity and complexity, some of which vary over time. Participants were keen that the wording be consistent with the International Classification of Functioning, Disability and Health (ICF);[7] hence, we added that the impairments created 'activity limitations'.

In round 2 we received comments that (1) the revised definition was too long, (2) the inclusion of examples was contested, and (3) the revised definition created uncertainty about whether neuromuscular and sensory conditions were included. Therefore, we revised and presented two versions of the definition in round 3, one with and one without examples, and added that neuromuscular conditions were included. We retained the key concepts from round 2, namely that neurodisability included a group of conditions, congenital or acquired, that were long term, variable, limiting, and often associated with comorbidity, and that undiagnosed neurological conditions were also included.

There was broad endorsement of the version presented in round 3, with 93% of participants agreeing or strongly agreeing. However, in response to a direct question about including example conditions, 75% of respondents favoured their omission. Parents on the team thought the examples may help some parents but also considered that they could be misleading, especially if a child's actual diagnosis was not listed and/or was very different from the examples, or if no specific diagnosis had yet been made. Iterations of the definition are provided as Data S1 (supporting information published online).

From our international survey we received 22 responses from a variety of professions and one parent (Table II); 60% of participants had English as their native language. Participants endorsed the definition generally with 18 out of 22 agreeing or strongly agreeing. Suggestions of English-language terms in use included 'neurodevelopmental disorder or conditions' and 'neurological disorders or conditions'. Similar terms are apparently used in other languages including 'handicap neurologique' (French), 'neurologische entwicklungsstörung' (German), and 'deficiência neurológica' (Portugese [Brazil]). There were indications that no terms were consistently used or widely agreed upon. The most common feedback from international colleagues was to use terminology consistent with the ICF.[7]

The final recommended definition from this process is as follows: 'Neurodisability describes a group of congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations. A specific diagnosis may not

Table II: Participants responding in the international survey, by profession and country

Participant characteristics	*n* (%)
Profession	
Occupational therapist	4 (18.1)
Paediatrician	4 (18.1)
Paediatric neurologist	3 (13.6)
Parent	1 (4.5)
Physiatrist	2 (9.1)
Physiotherapist	4 (18.1)
Psychologist	1 (4.5)
Speech and language therapist	1 (4.5)
Social science researcher	1 (4.5)
Social work	1 (4.5)
Total	22
Country	
Australia	3 (13.6)
Belgium	1 (4.5)
Canada	8 (36.4)
France	1 (4.5)
Israel	1 (4.5)
Macedonia	1 (4.5)
Switzerland	1 (4.5)
The Netherlands	2 (9.1)
Turkey	1 (4.5)
UK	1 (4.5)
USA	2 (9.1)
Total	22

be identified. Conditions may vary over time, occur alone or in combination, and include a broad range of severity and complexity. The impact may include difficulties with movement, cognition, hearing and vision, communication, emotion, and behaviour'.

DISCUSSION

Contributions from parents and professionals have helped to produce a definition of neurodisability that appears to be clear and generally, if not universally, approved. Developing the definition was an unanticipated part of the research that we had originally set out in our protocol, but we feel it is an output that may be useful in the future for a wide range of applications including evaluation of health services and relevant research studies. We hope that the definition helps to discriminate between children affected by neurodisability and those with other long-term conditions, such as impairments of the endocrine, respiratory, or musculoskeletal systems. Although children with such conditions share some characteristics with children with neurodisability, we believe that there are sufficient differences to make this an important distinction.

Our definition was developed in a systematic step-wise process, incorporating the views of health professionals and parents of children affected by neurodisability. We remain unaware whether any definitions of terms such as 'neurodevelopmental conditions', or others, have been endorsed as valid and acceptable to health professionals and parents. International colleagues indicated that the terms such as 'neurodevelopmental conditions' are used in some places, but also that no agreed terms are used consistently.

Early versions of our definition included examples of three of the most common neurological conditions; on balance it was felt that including these was not helpful and could be misleading. We debated whether neuromuscular conditions such as Duchenne muscular dystrophy should be included, as the defining impairment may not strictly be considered as neurological. Nevertheless, it was our view that neuromuscular conditions should be included, and survey participants supported inclusion of this term in the definition. Including the specific comment that a 'specific diagnosis may not be identified' was a consequence of parents pointing out that some children undergo extensive diagnostic tests and investigations, yet are not given a named condition or syndrome. The final version of the proposed definition does not include the terms 'children' or 'young people', and could be applied to adults with the same conditions, although the validity of this consensus supporting this definition has so far been based on the contributions of largely paediatric professionals and parents.

Some argue that 'disability' is a wholly socially constructed phenomenon.[8] Retaining a 'label' of 'neurodisability' could be viewed as problematic if it implied that having a neurological impairment inevitably created disability, rather than disability being a consequence of interactions between a person and his or her environment. Our own position is that functioning and disability are largely mediated by environmental factors, consistent with the ICF.[7] Others have set out criteria for definitions of 'disability', with which we would agree.[9] We emphasize that our proposed definition is a grouping of health conditions and certainly not intended as a definition of disability per se.

A strength of this study is the multidisciplinary sample that participated, composed of paediatricians, paediatric neurologists, paediatric surgeons, and a wide range of allied health professionals all working with children affected by neurodisability. The fact that they were motivated to volunteer and participate in the study may mark them as different to their colleagues who did not participate, so their views may or may not represent the views of the broader population of health professionals. Our international participants are small in number but come from a broad range of countries and professions and included a parent from France. On such small numbers we would not claim 'international consensus', but we feel that these international perspectives added to the refinement of the wording, and suggests wider utility for a definition.

The involvement of parents throughout the development of our definition increases the likelihood that the definition will be meaningful and understandable to other parents. We concede that there were only a small number of parents, and that they were a self-selecting group who had volunteered to work with the research team. It would have been useful to engage the opinions of a larger group of parents. However, interestingly, parents on a steering group for a research priority-setting exercise for children affected by neurodisability,[10] who had not been involved in developing the definition, readily endorsed using the proposed definition for that work. We also recognize that the definition should be meaningful for other professions who work closely with people affected by neurodisability, such as social care and educational professionals, who were not targeted in our survey and are therefore under-represented.

Although there was large agreement with the definition in the Delphi survey, with 93% of participants agreeing or strongly agreeing, there were a small number of people who disagreed. One reason for disagreeing was inconsistency with the ICF terminology. We addressed this to some extent in the final version by using 'impairment' in the first sentence, but we preferred 'functional' rather than 'activity' limitations in the last sentence because of the broader interpretation of 'functioning', although some inconsistency with the ICF remains. Nevertheless, we were reluctant to become 'instructive' about ICF terminology at the expense of clarity. One could be critical of the lack of detail regarding 'causality' within the definition; we would emphasize that our definition is intended to be descriptive of a phenomenon, rather than an explanatory model. One could also suggest that there should be more explicit detailed inclusion and exclusion criteria, which we did debate. Ultimately, we prioritized producing a concise definition that would be accessible by any of our intended users, many of whom would not be familiar with the ICF or medical terminology.

It could be argued that finding a definition of neurodisability is an issue parochial to the UK. However, our international survey identified that similar grouping of conditions is made commonly in other countries, but that terminology is applied inconsistently. We believe that there is a need for international consensus, as people with neurodisability represent a group with particular difficulties who are often vulnerable and require advocacy. Therefore it is important to be able to identify people with neurodisability, and then to identify their needs, and the needs of their carers and families. Hence, working towards international agreement for a definition is desirable, to improve clarity of communication to enable the sharing of knowledge and experiences, and the use of findings from research.

One of the uses for a definition is to estimate prevalence, and the wording of definitions is influential; Blackburn et al.[11] demonstrated how the prevalence of disabled children in the UK was observed to vary from 5% to 18% depending on the definition and/or measure used. Developing and agreeing on definitions is often a problematic process, as demonstrated by the debates about a definition of cerebral palsy,[12] and the classification of autistic spectrum disorders in the DSM-5.[13] The systematic approach we have taken provides support for the 'face' and 'content' validity of the proposed definition; however, our work has not examined the reliability of whether different professionals, parents, or people affected by neurodisability would classify conditions as within the definition consistently.

This study suggests that there is considerable consensus around the resulting definition of neurodisability among professionals and parents in the UK, and some support internationally for its usefulness. It will be important to explore whether this definition is meaningful for young people affected by conditions included within this category, and explore their preferences for an appropriate 'label'. We have found the definition useful in informing our National Institute for Health Research-funded project examining the potential for using patient-reported outcomes measures for children affected by neurodisability, and in a research priority-setting exercise for children affected by neurodisability.[10] We hope that others will also find this a helpful tool, enabling children with neurodisability to be grouped for research, evaluation, and other clinical purposes.

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SUPPORTING INFORMATION

The following additional information may be found online:

Data S1: Version iterations of the definition at each round.

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