Peninsula Cerebra Research Unit
PenCRU Childhood Disability Research

Research Summary

Which questionnaire is best to assess the health and quality of life of children with neurodisability?

This research summary was written by PenCRU and members of the PenCRU Family Faculty

Key findings

- Patient Reported Outcome Measures (PROMs) assess a person's health at a single point in time, and are collected through short, self-completed questionnaires.
- Bringing together scores from PROM questionnaires for groups of patients provides a way to assess whether services, treatment and therapies are improving their health outcomes.
- Our review identified 12 PROMs that had been tested with children with neurodisability in 48 studies. The most common neurodisability conditions in which evaluations were conducted were cerebral palsy, epilepsy, ADHD, autism, and traumatic brain injury.
- There is more evidence in samples of children with neurodisability that the questionnaire called DISABKIDS is a robust measure.
- Overall our review identified a lack of evidence of how well PROM questionnaires perform in children with neurodisability, especially to assess meaningful changes in health.

Who carried out this research and why?

The study was led by the team at Peninsula Cerebra Research Unit (PenCRU) at the University of Exeter Medical School. The National Institute for Health Research (NIHR) funded the research. The NIHR is the Government agency that funds most health research in the UK.

Patient Reported Outcome Measures (PROMs) are short, self-completed questionnaires used to assess a patient's health at a single point in time. Responses to these questions produce a score indicating better or worse health.

In this review we will use the word PROM and questionnaire intermittently.

Bringing together PROM questionnaire scores provide one way to assess whether services, treatments and therapies are improving health outcomes.

They are used in research, clinical audits and as routine outcome indicators in the NHS. It is vital that the measures are robust for purpose.

This study was part of a project looking at how best to measure health outcomes for children with neurodisability using PROMs.

Initially we identified all the currently available questionnaires that could be used to measure children's health and wellbeing. Then we reviewed all the studies that tested them in general populations of children. Next we looked for evidence from studies that tested these questionnaires specifically in groups of children with neurodisability conditions.

There are a number of ways to check how good a measurement is. Examples of relevant and required 'measurement properties' include:

- *Validity* is whether the questionnaire measures what it says it does;
- *Proxy reliability* is whether scores from parent's proxy responses are the same as from children;
- *Test-retest reliability* is whether scores remain the same after a period of time, when no change has occurred during that period;
- *Responsiveness* examines how much scores change when health improves or gets worse.

What did we do?

This type of research is called a systematic review. Systematic reviews bring together the results of all studies addressing the same research question. The aim is to provide a comprehensive and impartial summary of research evidence on a topic.

How did we search for evidence?

We searched online libraries that catalogue published research looking for studies that had tested any measurement properties of PROMs in a sample of children with neurodisability.

We included questionnaires identified in our previous study, all of which are suitable for all children. We also included questionnaires that can be used across children with any chronic health conditions.

We only included studies evaluating English language versions. This is because measurement properties cannot be assumed when questionnaires are translated.

We only included studies that had been published in journals that use peer review as a scientific standard. This means researchers other than the authors should have checked the scientific aspects of the work before publication.

How did we judge the measurement properties?

There are standard criteria for assessing whether a score from a PROM questionnaire is likely to be valid, reliable and responsive to change. There are also standards for judging how well the research was done and reported.

We used these criteria to appraise both the evidence itself - the results of the study; and the quality of the evidence how well the research had been done.

What did we find?

We found 48 research papers that had tested 12 PROM questionnaires. The most common conditions in samples were cerebral palsy (CP), epilepsy, Attention Deficit Hyperactivity Disorder (ADHD), autism, and traumatic brain injury.

The quality of the research was variable. In general, recent studies were reported more completely than older ones and were judged to have used higher quality methods.

None of the questionnaires had been tested across all the relevant and required measurement properties. There were no tests of how well any of the questionnaires measure change, such as how much change in score is important.

Two questionnaires have been evaluated more than other instruments – Pediatric Quality of Life (PedsQL) and Child Health Questionnaire (CHQ). However, evidence for both of them suggests that the PedsQL and CHQ are not robust.

We found most evidence of sound measurement properties in children with neurodisability for a questionnaire called DISABKIDS.

How are the findings useful?

There is keen interest to improve health outcomes for children, so we need ways to measure whether their health has improved. PROM questionnaires are also needed to assess if treatments and therapies are effective to improve children's health.

The technical information from this review and our previous study enhances our understanding of the strengths and limitations of current questionnaires. These reviews identify where further research could be targeted to improve the evidence about how well patient reported outcome measure questionnaires perform in groups of children with neurodisability.

The information also helps those who want to select questionnaires that are likely to produce a robust measurement that is valid and reliable.

From the results from this review and our previous study we recommend that the better questionnaires for use in groups of children with neurodisability are:

- DISABKIDS
- KIDSCREEN
- Child Health Utility (CHU-9D)

However even these PROMs have not been tested thoroughly in groups of children with neurodisability.

What next?

Our work identifies two potential directions for further research. This could be further testing of the existing PROMs in groups of children with neurodisability.

A priority with existing PROM questionnaires is finding out whether they can detect small but important changes in health, and how much change in scores can be considered meaningful.

However, none of the existing PROMs assess all the aspects of health prioritised by young people with neurodisability, parents and clinicians. Hence developing a new questionnaire assessing these aspects of health is also warranted.

The key areas are: communication, emotional wellbeing, pain, sleep, mobility, self-care, independence, mental health, community and social life, behaviour, toileting and safety.

We know there are existing questionnaires that specifically assess some of these key aspects of health. For example there are PROMs that assess only pain, or emotional wellbeing or sleep.

Future research could examine whether those questionnaires are acceptable to children with neurodisability and parents. It would be necessary to also review evidence of their measurement properties. This would be a sensible next step before beginning the development of a new child and/or parent reported questionnaire for children with neurodisability.

Who reviewed our research?

This study is published in a journal called Developmental Medicine & Child Neurology. Before the journal accepted the study to be published it asked independent experts to look at the papers and decide whether the research had been done well and reported properly and whether it was relevant.

The full version of the study is published in the journal Developmental Medicine & Child Neurology
http://onlinelibrary.wiley.com/doi/10.1111/dmcn.12982/epdf
The open access paper is freely available, or contact pencru@exeter.ac.uk for a copy

The team that carried out the research are: Astrid Janssens, Morwenna Rogers, Crispin Jenkinson, Alan Tennant, Stuart Logan and Chris Morris, with support from four parent members from the PenCRU Family Faculty.

Astrid, Morwenna, Chris and Stuart are all part of the Peninsula Cerebra Research Unit and or the NIHR Collaboration for Leadership in Applied Health Research and Care of the South West Peninsula (PenCLAHRC) at the University of Exeter Medical School. Crispin is head of the Health Services Research Unit at the University of Oxford and Alan is member of the Psychometric Laboratory for Health Sciences at the University of Leeds.

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