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ORIGINAL ARTICLE

Consensus of physician behaviours to target for early diagnosis of cerebral palsy: A Delphi study

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Aims: Historically, the diagnosis of cerebral palsy has been made after 12 months of age, delaying access to crucial early intervention that optimises functional outcomes. This study aimed to identify and specify priority physician diagnostic behaviours to target in implementation interventions to increase the rate of diagnosis of cerebral palsy under 6 months of age in Australia.

Methods: We conducted a two-round online Delphi study with a multi-professional expert panel of cerebral palsy researchers and clinicians. A reference group identified a six-item list of potential diagnostic behaviours, which were modifiable at the individual level, that could lead to an early cerebral palsy diagnosis. In the first survey, participants rated the importance of each item on a 10-point Likert scale and supplied their reasoning for this, and were able to suggest new behaviours. In the second survey, participants ranked items in order of priority.

Results: All six items reached consensus for inclusion (100%). No new items were added to the list. Ranking identified the top three priorities for online physician implementation interventions: (i) refer for or conduct the General Movements Assessment; (ii) refer for or conduct the Hammersmith Infant Neurological Examination; and (iii) communication of the diagnosis.

Conclusion: An online Delphi method can effectively inform tailored implementation intervention development. A consensus was achieved on the priority physician diagnostic behaviours to target in interventions to lower the age of cerebral palsy diagnosis in Australia.

Key words: cerebral palsy; Delphi technique; early diagnosis; implementation science; physician.

What is already known on this topic

1 Accurate diagnosis of cerebral palsy before 6 months is now possible using a combination of predictive assessments and clinical decision-making skills.
2 Targeted interventions are necessary to improve the uptake of cerebral palsy guidelines and impact on patient outcomes.

What this paper adds

1 To lower the age of cerebral palsy diagnosis in Australia, implementation interventions for physicians should prioritise: (i) Prechtl's General Movements Assessment; (ii) the Hammersmith Infant Neurological Examination; and (iii) communication of the diagnosis.
2 An online Delphi method enables systematic identification and specification of target behaviours for tailored implementation interventions.

Cerebral palsy has a prevalence of 1 in 700 live births.[1] The introduction of predictive infant assessment tools and improvements in neuroimaging now mean accurate and early cerebral palsy detection is possible as early as 12 weeks of age.[2] However, the Australian Cerebral Palsy Register identifies the average age of cerebral palsy diagnosis is 19 months,[1] indicating a research-practice gap for early cerebral palsy diagnosis.

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Clinical trials have shown that early diagnostic-specific intervention before 6 months of age can optimise neuroplasticity.[2] Early intervention and surveillance can improve infants' physical and cognitive outcomes and prevent the development of other complications.[2] Only 26% of infants with cerebral palsy receive a diagnosis before 6 months.[1]

In an effort to reduce the research-practice gap, a clinical guideline for the early diagnosis of cerebral palsy has been published.[2] The variable uptake of guidelines in clinical practice is well known.[3] Tailored implementation interventions, which aim to bridge research-practice gaps[4] and address identified barriers, may effect practice change more than guidelines alone.[5] A guideline implementation barrier analysis, conducted in the Australian context by the Centre of Research Excellence in Cerebral Palsy, identified paediatricians and sub-specialists (hereafter referred to as 'paediatric physicians') as a primary implementation target

audience to enable early diagnosis of cerebral palsy (Appendix S1, Supporting Information). This target audience required targeted implementation interventions, informed by further research prioritisation.

Implementation intervention designs may be more effective when supported by theory.[6] The Behaviour Change Wheel[7] can provide a framework for theory-informed interventions, including defining a problem in behavioural terms. To enable a behavioural analysis to guide future intervention development, we identified a need for expert opinion and systematic methodology to determine the priority behaviours to target for paediatric physicians.

The Delphi technique is an established research prioritisation method for forming a consensus.[8] It is characterised by involvement of experts with varied backgrounds, an anonymous process to avoid bias of a dominant view, an iterative process and design of successive rounds, informed by group responses to previous rounds.[9,10] The primary objective of this study was to reach expert consensus on the three most important behaviours of paediatric physicians to target in an implementation intervention that aimed to improve clinical practice to reduce the age of cerebral palsy diagnosis in Australia.

Methods

We used a two-round online Delphi study with a multi-professional expert panel to identify and prioritise target paediatric physician behaviours. Delphi study methodology followed recommendations of the Conducting and Reporting Delphi Studies[9] and quality indicators for a Delphi study.[10] We followed reporting guidelines recommended by the Standards for Reporting Implementation Studies.[11] The study received approval from The University of Sydney Human Research Ethics Committee (project number 2019/454).

Item list generation

The barrier analysis identified a list of recommendations from the early diagnosis of cerebral palsy guideline that were potential target behaviours for paediatric physicians, modifiable at the individual practitioner level (Table 1).[2] The six-item list was appraised by the research team and an expert advisory group (n = 10) that was established as part of the Australasian Cerebral Palsy Clinical Trials Network, Centre of Research Excellence.

Participants

The Delphi study's multi-professional expert panel (n = 23) was purposively selected from authors of the early diagnosis of cerebral palsy guideline and multi-disciplinary clinical and research leaders in Australia. Following the Behaviour Change Wheel, we identified target paediatric physician behaviours with[7]: (i) likely impact on the age of cerebral palsy diagnosis; (ii) ease of implementation; and (iii) ease of validated measurement.

The study sample size was adequate.[12] The Delphi panel depends on group dynamics rather than statistical power to reach a consensus and stability in the rating of importance was achieved through one survey round.[12]

Table 1 List of behaviours generated from barrier analysis and clinical practice guide

Target behaviours
Use or refer for Prechtl's General Movements Assessment with newborn-detectable cerebral palsy risk factors and under 5 months corrected age
Refer for magnetic resonance imaging with newborn detectable cerebral palsy risk factors before sedation is required
Use or refer for Hammersmith Infant Neurological Examination with newborn detectable (under 5 months corrected age) and infant detectable risk factors (5–24 months of age)
Refer for magnetic resonance imaging with infant detectable cerebral palsy risks where safe and feasible
Refer for cerebral palsy specific early intervention following high-risk of cerebral palsy notification or diagnosis of cerebral palsy
Communicate high-risk of cerebral palsy notification or diagnosis of cerebral palsy to families in a series of tailored, well-planned, face to face conversations

Delphi procedure

Delphi round 1

The six-item list of behaviours was transformed into a survey in which participants could rate the importance of each item (Appendix S2, Supporting Information). Participants were asked to assign a score for each item using a 10-point Likert scale. For each item, we calculated the median and interquartile range, and consensus of agreement. A consensus of agreement that behaviour was important was specified as more than 70% of participants rating an item seven or higher on the scale.[13] A consensus of agreement that behaviour was unimportant was specified as 70% of participants rating a behaviour item as three or below.[13] Participants were informed that behaviour would be excluded from the list and round 2 survey if an agreement was reached that it was unimportant. Lack of agreement was defined as items rated 4, 5 or 6 by more than 30% of participants.[13] Any items with a lack of agreement were taken to the round 2 survey for further iteration. Participants were asked to provide their reasoning or comments for their rating scores in an open-text field.

In the open-text field, participants were also invited to identify any new behaviours they believed were important. New behaviour items would be reviewed by the researchers and included in round 2 if agreement was reached by a majority.[14] Participants were asked to supply demographic details, including their profession and years of experience in cerebral palsy diagnosis.[14]

The survey instrument was piloted (n = 3) to affirm comprehensibility and test for the possibility of bias in judgements. It took approximately 20 min to complete. One reminder email was sent per round, each of which lasted 2 weeks. Round 1 was conducted in November–December 2019.

Delphi round 2

In the second survey, participants were provided with feedback on survey one responses and asked to rank the priority of each behaviour item on the list from 6 for the highest-ranked item to

1 for the lowest-ranked item. An aggregated summary was calculated by summing individual scores for each item and the top three priority behaviour items identified. The round 2 survey was conducted in January–February 2020.

Thematic analysis

Comments about target behaviours were analysed using inductive thematic analysis.[15] Target behaviours were identified as the main themes; data relating to each were analysed separately. Data were coded without a pre-existing coding frame.[15] Equal attention was given to each item.[15] For each target behaviour, categories were identified in the data, which became the subthemes.

To cross-check data analysis and ensure transparency, data were coded independently by a second researcher experienced in qualitative methodology.[15] Differences in data interpretation were discussed with the wider research team.

Results

Of the invited experts, 11 respondents (response rate = 48%) completed round 1. Their professions were: paediatrician (n = 1), neonatologist (n = 2), neurologist (n = 1), allied health professional (n = 2), clinician-researcher (n = 6), researcher (n = 4) (some identified as having dual professions). Participants' experience in early diagnosis of cerebral palsy were: 5 years (n = 2), greater than 10 years (n = 3), greater than 20 years (n = 5), never (n = 1). An error in the round 2 participant list resulted in an additional respondent from the original invited experts (making 12 participants), who was included in the results as this was unlikely to compromise the study.

Delphi round 1: Rating of importance

In round 1, all participants indicated their level of agreement for the importance of the target behaviours for paediatric physicians. As outlined in Table 2, all six behaviour items reached consensus for inclusion, with greater than 70% of participants scoring them 7 or more on the 10-point Likert scale of importance. No items reached consensus for exclusion (0%). Validation checks were completed on all data entered. No new behaviours were added to the item list for round 2. Sixty comments from the round 1 rating of importance were included in the thematic analysis.

Delphi round 2: Ranking of priority

In round 2, all participants completed the ranking of behaviour items in order of priority. The ranked order of priority from highest to lowest was: (i) use or refer for General Movements Assessment (n = 69); (ii) use or refer for Hammersmith Infant Neurological Examination (n = 48); (iii) communicate high-risk notification or diagnosis of cerebral palsy to families in a series of tailored, well-planned, face-to-face conversations (n = 42); (iv) refer for magnetic resonance imaging (MRI) with newborn detectable risk factors before sedation is required (n = 41); (v)

Table 2 Round 1 agreement of the importance of identified target physician behaviours

Target behaviour	Maximum score	Minimum score	Median score (IQR)	% rating 7 or more	Consensus of agreement
Use or refer for Prechtl's General Movements Assessment *(GMA)* with newborn-detectable CP risk factors	10	9	10.00 (0)	100	Yes
Refer for *MRI* with *newborn* detectable CP risk factors before sedation is required	10	6	9.00 (3)	90.9	Yes
Use or refer for Hammersmith Infant Neurological Examination *(HINE)* with newborn detectable (under 5 months corrected age) and infant detectable risk factors (5–24 months of age)	10	7	9.00 (2)	100	Yes
Refer for *MRI* with *infant* detectable CP risks where safe and feasible	10	5	8.00 (2)	90.9	Yes
Communicate high risk of CP notification or diagnosis of CP to families in a series of tailored conversations	10	8	10.00 (1)	100	Yes
Refer for CP specific *early intervention* following high risk of CP notification or diagnosis of CP	10	7	10.00 (2)	100	Yes

Legend

GMA Prechtl's General Movements Assessment
MRI Magnetic resonance imaging
IQR Interquartile range item score
% 7 or More Participants rating an item 7 or above on the 10-point Likert scale
Number of participants = 11
CP Cerebral palsy
HINE Hammersmith Infant Neurological Examination
Consensus of Agreement 70% participants rating an item 7 or above on the 10-point Likert scale

CP, cerebral palsy; MRI, magnetic resonance imaging; GMA, Prechtl's General Movements Assessment; HINE, Hammersmith Infant Neurological Examination.

Table 3 Illustrative quotations

Behaviour	Sub-theme	Quotations with participant numbers
Use or refer for GMA with newborn-detectable CP risk factors and under 5 months corrected age	Accurate	'Evidence for General Movements is overwhelmingly strong, and the tool should form part of standard clinical care for any high-risk infant populations'. Participant 8
	Feasible	'The tool is easy to administer and there are many trained clinicians in this tool and this number is increasing'. Participant 11
	Enables early intervention	'Can be performed remote to health service'. Participant 2. 'Parents want it and it helps the baby access early intervention which is likely to improve the outcomes and decrease complications'. Participant 5
Use or refer for HINE with newborn detectable (under 5 months corrected age) and infant detectable risk factors (5–24 months of age)	Predictive additional assessment	'This assessment provides valuable additional information relating to asymmetries that can indicate hemiplegia, which can be missed on GMs (as is known to be less accurate in detecting milder motor phenotypes and hemiplegia). It also provides excellent information that triangulates evidence of neurological dysfunction, topology, and reasonable prediction of motor outcome.' Participant 2
	Clinically useful	'This is a simple tool that should be widely implemented in at-risk populations.' Participant 8
Communicate high risk of CP notification or diagnosis of CP to families in a series of tailored, well-planned, face to face conversations	Communication takes time for families to understand risks, ask questions, adjust to the diagnosis and know what to do	'Information and understanding are essential for families to adjust to the possibility of a diagnosis of CP.' Participant 1. 'Must be supported environment in appropriate language, and to key carers in an infants' life.' Participant 2
		'Families have reported that this discussion needs to be done carefully with time to digest the information and ask questions.' Participant 6
	Improvement in diagnostic communication skills are needed	'Part of clinical care which needs to be done better.' Participant 5
		'I think that this is often done poorly and can have long lasting ramifications for relationships between health care workers and families.' Participant 11
Refer for MRI with newborn detectable CP risk factors before sedation is required	Assists with early diagnosis and prognosis	'Ideal in early life as can provide both prognostications for severity as well as comorbidity.' Participant 2
	Not always feasible or possible	'MRI is really important but there can also be long wait lists and expensive. Important to do but if other assessments can be done before, such as GMs, this would save resources.' Participant 7
		'May not always be possible due to child's health needs.' Participant 2
	Uncertainty in the interpretation of findings	'Sometimes a repeat MRI with a general anaesthetic is needed to look for white matter changes not evident at 6–12 weeks of age which is the time that an infant may

(*Continues*)

Table 3 (Continued)

Behaviour	Sub-theme	Quotations with participant numbers
		be scanned with sedation alone.' Participant 1
Refer for CP-specific early intervention following high risk of CP notification or diagnosis of CP	Early intervention evidence is promising, more research is needed in CP outcomes and key ingredients	'The whole purpose of early detection is to then provide early interventions which are aimed at improving outcomes Although we still need more evidence on what these interventions should be and what 'ingredients' they should contain. Early detection evidence is now very strong. Evidence for early intervention is promising.' Participant 8 'Early intervention works but it should be targeted.' Participant 9

CP, cerebral palsy; MRI, magnetic resonance imaging; GMA, Prechtl's General Movements Assessment; HINE Hammersmith Infant Neurological Examination.

refer for cerebral palsy specific early intervention following high-risk notification or diagnosis (n = 28); and (vi) refer for MRI with infant detectable risks where safe and feasible (n = 24).

Free-text comments

Participants' comments on the round 1 survey were frequently clarifications or justifications of rating decisions. Quotations that are illustrative of themes and sub-themes are in Table 3. Thematic analysis facilitated further understanding of participants' beliefs about the target behaviours in Australia. For example, Prechtl's General Movement Assessment, which was ranked as the highest priority, was considered accurate, feasible and enabling early intervention.

Discussion

The multi-professional experts in the field of cerebral palsy reached a high level of consensus on the priority behaviours that paediatric physicians need to reduce the age of cerebral palsy diagnosis in Australia. Ranking identified the top three priorities of: (i) refer for or conduct Prechtl's General Movements Assessment; (ii) refer for or conduct Hammersmith Infant Neurological Examination; and (iii) communication of diagnosis to families.

The finding that referring for Prechtl's General Movements Assessment was the highest priority is not surprising as this predictive motor assessment tool has become the gold standard for early detection of cerebral palsy, with established validity and inter-rater reliability,[16] sensitivity as high as 98% and specificity as high as 91% in early months.[17] The General Movements Assessment is a simple video that enables remote and smartphone application. Training and certification are widely available and established clinical networks provide support and referral pathways for physicians.[18] Participants' comments that this test enables early intervention relates to the accuracy of the General Movements Assessment in the detection of early motor dysfunction and neurological disability, and certainty in the need for early intervention with infants identified with an abnormal result. Prior to the clinical application of the General Movements Assessment, the previous cerebral palsy diagnostic approach of history-taking, clinical observation and awaiting missed milestones resulted in late physician referrals for intervention to the detriment of infants and parents.[2]

The Hammersmith Infant Neurological Examination was ranked as the second-highest priority behaviour. This was a significant finding, considering the most accurate method for early detection of cerebral palsy is a combination of the General Movements Assessment, MRI and history-taking about risk factors.[2] The Hammersmith Infant Neurological Examination is recommended where the General Movements Assessment or MRI is not available, or feasible.[2]

The ranking of the Hammersmith Infant Neurological Examination above MRI in priority for Australian physicians may be explained through free-text comments. Participants reported that an MRI was not always feasible or possible, and identified the need to balance the use of MRI with risk of sedation. These findings point to future directions for implementation research, including the role of MRI usage in cerebral palsy diagnosis and evidence-based testing for cerebral palsy differential diagnosis in high, middle and low-income contexts. In contrast, free-text comments identified the Hammersmith Infant Neurological Examination as a predictive additional assessment that was clinically useful. No comments related to risks or feasibility concerns in Australia. The Hammersmith Infant Neurological Examination is a scoreable, standardised assessment that can be used between 2 and 24 months of age.[19] It provides additional information on the severity and distribution of cerebral palsy and other aspects of neurological function other than motor.[19]

The combined diagnostic accuracy of all three instruments (General Movements Assessment, MRI and the Hammersmith Infant Neurological Examination) and their use in isolation has been established.[20] When all three tests are not feasible, the use of two tests is recommended to increase clinical diagnostic certainty.[2,20]

Of significance, the third-highest priority identified was the communication of high-risk notification or diagnosis of cerebral palsy to families. This, again, was ranked higher than MRI in order of priority, suggesting that change in practice in when and how communication of diagnosis is provided to families may impact patient outcomes. Free-text comments that communication takes time and diagnostic communication skills need to improve are aligned with best practice recommendations.[21]

All six behaviours were identified as important, acknowledging the role of the combination of all standardised assessments with strong predictive validity for an early and accurate diagnosis in clinical practice and prompt referral for cerebral palsy specific early intervention as soon as the clinical criteria for high-risk of cerebral palsy are met.[2] Targeted physician education on the best available evidence[2] and early diagnosis of cerebral palsy clinical decision-making skills training are warranted (Appendix S1, Supporting Information). Given cerebral palsy is a complex diagnosis, with heterogeneous clinical presentations, it is likely that tailored knowledge translation strategies will be needed to address implementation barriers over and above predictive tool use.

The response rate for survey one (48%) was acceptable compared to other research prioritisation studies and the retention rate was good. The error in the participant list for the round 2 survey is a potential limitation. The views of the Delphi panellists may not represent the views of all early diagnosis of cerebral palsy experts[13] and the high rate of dual trained clinician-researchers may narrow the profile of responses; however, the participant criteria are reproducible and the panel was comprised of multi-disciplinary experts from diverse settings.

Conclusion

This Delphi study identified target behaviours for early diagnosis of cerebral palsy implementation intervention research in Australia. An early diagnosis facilitates early intervention, optimises infant neuroplasticity and provides access to psychological supports for parents. An early and accurate clinical diagnosis of cerebral palsy may be expedited in Australia by physician referral for two highly predictive standardised tests of movement and neurological function. The importance of communicating the diagnosis of cerebral palsy has been highlighted, indicative of the perceived impact on mental health outcomes for families and likely spill-over effects on access to funding and early intervention in Australia. The priorities identified can be used to inform future tailored interventions for the early clinical diagnosis of cerebral palsy.

The systematic design allows replication of this study in other high-, middle- and low-income countries. Future implementation interventions to lower the age of cerebral palsy diagnosis in Australia should prioritise referral for Prechtl's General Movements Assessment, referral for the Hammersmith Infant Neurological Examination and communication of diagnosis.

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Supporting Information

Additional Supporting Information may be found in the online version of this article at the publisher's web-site:

Appendix S1. Guideline implementation barrier analysis.
Appendix S2. Delphi study surveys.