Consensus research priorities for cerebral palsy: a Delphi survey of consumers, researchers, and clinicians

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AIM Research funds for cerebral palsy are scarce and competition for them is strong. This study aimed to identify questions for future research that were agreed to be a high priority.

METHOD An expert panel of consumers, researchers, and clinicians was assembled (*n*=127) and surveyed using a Delphi survey comprising three rounds. In round I, participants identified three important research topics. Three parallel surveys were constructed: (1) consumers; (2) intervention researchers and clinicians; and (3) aetiology and prevention researchers. In rounds II and III, participants rated priorities using a seven-point Likert scale. Questions reaching consensus were itemized and those not reaching consensus were discarded.

RESULTS Consumers identified questions in the themes of prevention/cure, quality of life/community participation, and service provision/intervention. Intervention researchers/clinicians identified questions in the themes of effective outcomes and effective research/services. Aetiology and prevention researchers identified questions in the themes of infection/inflammation, focus on timing, haematology, research tools, neuroregeneration, and genetics. Fifty per cent of the consumers' priorities were also identified by professionals.

INTERPRETATION Research priorities change as evidence is established. Phase II of this project is to develop a web portal with international collaboration. As evidence builds for one research question, it will be added to the web portal and unanswered questions will become the priority.

Cerebral palsy (CP) is the most common physical disability in childhood. Despite clinical and research advances, its incidence remains stable.[1] The condition is lifelong with no known cure. There is a need for research in aetiology, prevention, and effective intervention for maximizing potential and optimizing quality of life.

No evidence exists of explicit published prioritization for CP research that is itself developed through a scientific process. Many individual research questions are identified at the conclusion of studies. However, no published study could be identified that provides guidance on the relative importance and ranking of the questions or where best to direct limited research funds to drive the field forward.[2] The World Health Organization has identified that a collaborative, widely consulted, systematic approach to research priority setting is essential.[3] Thus, establishing a unified research agenda for CP and consensus on essential

and urgent research topics may provide the possibility of accelerating breakthroughs.

Key stakeholder groups exist in CP with an interest in a priority-driven research agenda. Consumers (people with CP and their families) have the most vested interests in research, and must be included on an expert panel for developing research agendas.[4] Additional groups include researchers/clinicians who provide intervention for people living with CP, those researching the aetiology/prevention of CP, and policy makers and administrators/senior management of key organizations (Table I).

It is essential to involve as many legitimate stakeholders as possible in the identification and prioritization of research topics.[3,5] Not only does this ensure the interests of all relevant people are considered, but it might also increase ownership of the ensuing research and the

Table I: High-priority questions for consumers and overlap with professionals

Research questions	Median (IQR)	Rank (mean)	Match intervention	Match aetiology/ prevention
Theme 1: prevention and cure				
Aetiology				
How can CP be prevented?	7.00 (1.00)	1 (5.98)	n/a	a
What are the causes of and casual pathways to CP?	6.00 (1.00)	7 (5.80)	n/a	a
Neuroregeneration				
What potential does the brain have to repair injury?	6.00 (1.00)	3 (5.85)	d	a
Can stem cells have a therapeutic effect for CP?	6.00 (1.00)	3 (5.85)	d	d
Theme 2: quality of life and community participation				
Carers				
What policies are needed to improve quality of life for families caring for Australians with severe disabilities?	7.00 (1.00)	8 (5.78)	c	n/a
Lifestyle				
What factors have the greatest impact on improving the lifestyle and quality of life of individuals with CP?	6.00 (1.00)	6 (5.83)	b	n/a
Employment				
What are the barriers to employment that exist for people with CP?	6.00 (1.00)	14 (5.60)	c	n/a
How can people with CP be better trained with the necessary skills to enter the workforce?	6.00 (1.00)	16 (5.44)	b	n/a
Socioeconomics				
What is the relationship between CP and poverty?	6.00 (1.00)	21 (5.08)	d	n/a
Access				
Can people with CP equitably access the community?	6.00 (1.00)	22 (4.85)	d	n/a
Theme 3: service provision and intervention				
Service models				
What can be done to address the mismatch between what service parents and people with CP need and what they actually receive?	7.00 (1.00)	12 (5.68)	d	n/a
What is the optimal intensity of therapy programs?	6.00 (1.00)	9 (5.75)	a	n/a
What is the most efficient service model so that maximal services reach people with CP and their families?	6.00 (1.00)	14 (5.60)	d	n/a
Effectiveness and outcomes				
What are the optimal treatments for CP?	6.00 (1.00)	2 (5.95)	a	b
What are the long-term outcomes of treatments?	6.00 (1.00)	3 (5.85)	a	d

Table I: Continued

Research questions	Median (IQR)	Rank (mean)	Match intervention	Match aetiology/ prevention
What is the effectiveness of alternative therapies for the treatment of CP?	6.00 (1.00)	11 (5.70)	d	n/a
What early intervention (dependent on CP type) will prevent and minimize structural impairments?	6.00 (1.00)	13 (5.67)	c	c
Does physiotherapy benefit people with CP?	6.00 (1.00)	18 (5.25)	d	n/a
What are the most effective methods of pain management so the secondary complications can be reduced?	6.00 (1.00)	19 (5.18)	d	n/a
What is the effectiveness of hydrotherapy for people with CP?	6.00 (1.00)	23 (4.83)	d	n/a
Role of families in intervention				
What are the most effective methods of educating parents to help improve their child's independence and function?	6.00 (1.00)	9 (5.75)	d	n/a
What is the impact of therapy type and duration on parents of a child with CP?	6.00 (1.00)	17 (5.38)	d	d
Is hands-on treatment by therapists a more effective method than guided therapy by parents?	6.00 (1.00)	20 (5.15)	b	n/a

[a]Close match; [b]similar intent; [c]similar focus, different approach; [d]nothing similar; n/a, not applicable. IQR; Interquartile range.

likelihood of the results influencing clinical practice and policy.[3] The more groups and individuals who are involved, however, the greater the potential difficulty in reaching consensus.[6]

The aim of this study was to identify research questions for CP by conducting a three-part Delphi survey. The objectives of the study were as follows: (1) to assess the extent to which a cohort of consumers, intervention researchers/clinicians, and aetiology/prevention researchers could identify high-priority questions for CP research; (2) to determine if consensus for the priorities in CP research could be established by each group; and (3) to identify the extent to which consumers and professionals have similar consensus priorities for research.

METHOD
Design
This study used the well-established Delphi survey, which involves asking experts a recurring progression of questions through a series of questionnaires.[7–9] It is a way of structuring a group communication between experts, where individual participants give feedback that contributes to emerging consensus.[10] Delphi surveys eliminate potential sources of conflict experienced in committees and panels[9] because they are anonymous and they provide the opportunity to revise individual views in response to group trends.[9]

The Delphi method has been used to develop research priorities in many areas of health including midwifery, emergency and critical care nursing, and for general practitioners. We selected it because it is known to be effective when (1) consensus is sought in an area where none previously existed, (2) the research problem does not lend itself to precise analytical approaches but can be illuminated by subjective collective judgments, (3) the study participants have diverse backgrounds in their experience and expertize and therefore consensus cannot easily be reached, (4) more research participants are needed than can effectively interact face-to-face, and (5) frequent meetings of all participants are not feasible.[2] All these indictors were true for CP, where input was sought from a wide variety of

disciplines across the world and from consumers. The consensus-building nature of the Delphi technique combines the rigor of traditional surveys and the collaborative effect of focus groups.[8]

In this study a three-round survey strategy was used, because additional rounds produce minimal change in opinion.[11] The overarching process is summarized in Figure 1. The first step was systematically (literature

	Consumers	Intervention	Aetiology/ Prevention
Investigators conduct a comprehensive literature review to (a) identify literature generated research questions; and (b) identify experts to invite to participate	*n*=50 invited	*n*=180 invited	*n*=80 invited
Round I surveys emailed/sent (and 1 reminder sent) to identified experts requesting: (a) participation; (b) demographic data; and (c) identification of 3 areas that need to be researched for cerebral palsy.	*n*=20 replied 38%	*n*=76 replied 42%	*n*=31 replied 39%
Research problems identified and returned by participants. Investigators review research problems submitted and synthesize these into researchable questions. Questions added to the literature generated questions (bar consumer survey).	140 problems became 50 questions	482 problems became 74 questions	200 problems became 166 questions
Round II survey emailed/sent. Includes all questions proposed by participants and published literature. Participants asked to rate the questions by priority using the 7-point scale. 1 = very low priority and 7 = very high priority	*n*=20 replied 100%	*n*=45 replied 60%	*n*=19 replied 61%
Median responses and inter-quartile ranges to round II questions calculated. All questions with median of 6 or more with an interquartile range of 1 or less were considered to have reached high priority and placed on priority list. Questions with consensus low and low median priority discarded.	22 questions high consensus	16 questions high consensus	3 questions high consensus
Round III survey emailed/sent. Included refined question list with only questions that were a priority but had not yet reached consensus. Participants asked to rate these non-consensus questions using the same 7-point priority scale.	*n*=19 replied 95% / Additional 1 question high consensus	*n*=32 replied 71% / Additional 7 questions high consensus	*n*=18 replied 95% / Additional 7 questions high consensus
All questions that reached high priority consensus (median of 6 or more with an inter-quartile range of 1 or less) were placed on final priority list. Questions with consenus low priority discarded. Research priorities listed in rank order by mean scores.	23 questions high consensus	23 questions high consensus	10 questions high consensus*

* An extra 23 questions kept for aetiology that had median of 6 and interquartile 1-2
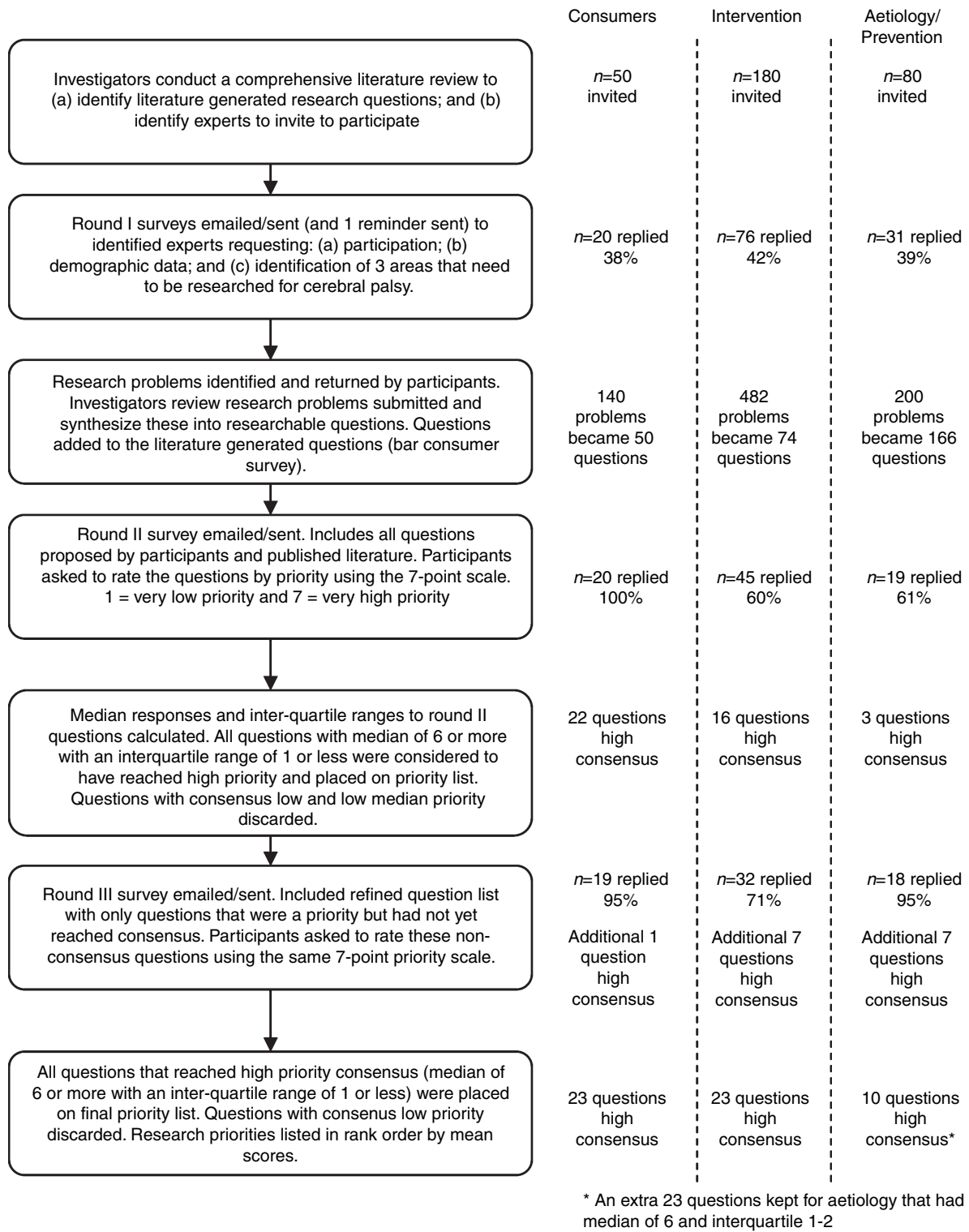
Figure 1: Delphi survey process and analysis

review) and subjectively (round I survey with open-ended questions) to identify topic areas that were considered to be essential. All topic areas identified in both the literature review and the round I survey were included in round II. The second step involved testing consensus using a quasi-experimental design, to verify the topics agreed to be important. The third step was to devise three lists of agreed priorities for CP research and to identify themes from the perspectives of (1) consumers, (2) intervention researchers/clinicians, and (3) aetiology/prevention researchers.[11] The final step was to analyse links between the consumers' and professionals' themes and lists of agreed priorities.

Ethics
Approval for this study was granted by The Spastic Centre of New South Wales Human Research Ethics Committee (HREC EC00402), which is a recognized committee of the National Health and Medical Research Council, Australia.

Participants
A sampling frame consisting of 310 potential participants was identified through purposive sampling.

(1) Consumers who had attended research information sessions provided by the first author over the preceding year, or who had made an enquiry to the research team, were approached. This identified consumers who had a high research interest, to maximize participation and minimize drop out over the three surveys. Gross Motor Function Classification System (GMFCS) levels of adults with CP who participated were not all available. Children of parents who participated had GMFCS levels ranging from I to V.

(2) Potential participants for the intervention survey were identified as those who had multiple publications in CP research or who were clinicians in positions of leadership throughout Australia.

(3) Potential participants for the aetiology/prevention survey were identified as those who had multiple publications in CP aetiology/prevention research.

Analysis
In the analysis of round I, identified research areas were reviewed and developed into mutually exclusive research questions.

Analysis of surveys in rounds II and III used descriptive statistics. Median scores and interquartile ranges (IQRs) were calculated for the groups' responses to each question because the seven-point Likert scale data was ordinal.[9] Responses where the median was 6.00 (high priority) with an IQR of 1.00 were considered important research questions that had reached consensus. This cut-off point was chosen because more than 75% of the group rated the priority as higher than 5.00 (fairly high priority). Final rankings at the completion of round III were assigned using mean scores.

Within this study, consensus was considered to have two key elements, stability and convergence. Stability was the consistency of responses across rounds II and III; convergence was the degree of agreement achieved reflected in descriptive statistics.[8]

At completion of quantitative analysis, the agreed high priorities were categorized into major and minor themes for each of the three surveys. Finally, each question in the consumer survey was compared and contrasted with each question in both the intervention and aetiology/prevention surveys, to identify their level of 'match'. Four categories were developed: (1) close match – the chosen wording was exact or almost exact; (2) similar intent – the content was the same but worded differently; (3) similar focus, different approach – general outcome identified as the same but a different approach identified; (4) nothing similar found; and (5) not applicable – not expected that this group would identify this research question as a priority to them.

RESULTS
Round I
(1) Consumers. Of 50 surveys sent, 20 participants replied, yielding an eligibility fraction for the study of 38%. All participants were from New South Wales, Australia (Table SI, supporting information published online).

(2) Intervention researchers/clinicians. Of 180 surveys sent, 76 participants replied, yielding an eligibility fraction for the study of 42%. The participants were from a diverse range of medical and allied health backgrounds (Table SI). Participants were from Australasia (*n*=68, 89.5%), North America (*n*=4, 5.3%), and Europe (*n*=4, 5.3%).

(3) Aetiology/prevention researchers. Of 80 surveys sent, 31 participants replied, yielding an eligibility fraction for the study of 39%. The participants were from a diverse range of professional backgrounds that commonly research the aetiology of CP (Table SI, supporting information published online). Participants were from Australasia (*n*=17, 55%), North America (*n*=5, 16%), and Europe (*n*=9, 29%).

The round I questionnaire asked participants to list three problem areas associated with CP that might be addressed by research. In parallel, the investigators conducted a literature review of research priorities in CP. For the intervention and aetiology/prevention surveys, the participants' data on research topics was combined with research questions identified from literature. Eight hundred and twenty-two research questions, topics, and ideas

were generated. Two investigators with expertize in CP research and content analysis reviewed each research question, topic, or idea using the grounded theory approach. Through a process of content analysis to the level of open coding, the items were categorized into themes.[12] Reliability was excellent, with 97% agreement for assignment to topic themes.[8]

This process allowed the initial 822 responses to be reduced to 290 mutually exclusive research areas. The topics were reworded into the format of research questions for use in the round II surveys.[2] The consumer survey contained 50 items, all generated by consumers, with no additional literature-based questions included. The intervention survey contained 74 questions and the aetiology/prevention survey contained 166 questions (Fig. 1).

Round II

The round II questionnaire was sent to all participants in round I. It was returned by 20 consumers, 45 intervention researchers/clinicians, and 19 aetiology/prevention researchers, yielding response rates of 100%, 60%, and 61% respectively. Participants rated the perceived priority of each of the research questions using a seven-point Likert scale (1, very low priority; 7, very high priority).

(1) Consumers. (a) Twenty-two research questions reached a consensus high priority and these were added immediately to the final priority list. (b) No questions reached a consensus low priority and therefore none were discarded. (c) Twenty-eight questions were overall rated high (i.e. median greater than 5) but had not reached consensus (i.e. IQR greater than 1).

(2) Intervention researchers/clinicians. (a) Sixteen research questions reached a consensus high priority. (b) Twenty-two questions reached a consensus low priority and were discarded. (c) Thirty-six questions were rated high overall.

(3) Aetiology/prevention researchers. (a) Three research questions reached a consensus high priority. (b) Seventy-three questions reached a consensus low priority and were discarded. (c) Ninety questions were rated high priority overall (Fig. 1).

Round III

The round III questionnaire was sent to all round II participants and returned by 19 consumers, 32 intervention researchers/clinicians, and 18 aetiology/prevention researchers, yielding response rates of 95%, 71%, and 95% respectively.

Round III only included items that were considered a priority but had not yet reached consensus. The questionnaire also provided feedback on the round II group median response for these research questions.[2] This enabled respondents to reflect on colleagues' scores, as well as their own, to help develop consensus.

(1) Consumers. One additional question reached consensus high priority. The final list included 23 high-priority research questions. Questions were ranked in order of mean scores, and categorized into three major themes: (a) prevention and cure; (b) quality of life and community participation; and (c) service provision and intervention, with minor themes in each (Table I).

(2) Intervention researchers/clinicians. Seven additional research questions reached consensus high priority. The final list included 23 research questions. Questions were ranked and categorized into two major themes: (a) effective outcomes; and (b) effective research and services, with minor themes in each (Table SII, supporting information published online).

(3) Aetiology/prevention researchers. Seven additional research questions reached consensus high priority. The final list included 10 research questions. Owing to the low numbers of high-priority consensus questions, it was decided to report on a further 23 questions that were agreed to be a high priority but did not reach the consensus definition of IQR=1 (Table SIII, supporting information published online). Research questions on the priority list were ranked and categorized into six major themes: (a) infection and inflammation; (b) gestation; (c) haematology; (d) neuroregeneration; (e) research tools; and (f) genetics.

When comparing the consumer priority list with the professionals' priority lists there was nothing similar for 11 of 23 questions (48%), a similar focus but different approach identified for 3 of 23 questions (13%), and a similar intent or close match for 9 of 23 questions (39%).

DISCUSSION

All of the study's objectives were answered by the findings. The first objective was to assess the extent to which a population of consumers, intervention researchers/clinicians, and aetiology/prevention researchers could identify high-priority areas of concern about CP research needs. The results of this study demonstrate that each group identified a wide range and large number of research questions needing answers.

The second objective was to establish if consensus for priorities in CP research could be established by each group. Although 23 research questions were agreed as high priorities for consumers and intervention researchers/clinicians, a significantly lower number of questions (n=10) reached consensus in the aetiology/prevention survey. This was not surprising given (1) there are multiple pathways to CP, (2) many specialties are needed to

research each pathway, and (3) it is not expected that any one pathway will be the dominant cause. We therefore believed it was valid and imperative to include all aetiology/prevention questions that were agreed to be a high priority (n=33, IQR=2) but did not reach the consensus definition of IQR=1.

The third objective was to identify the extent to which consumers and combined professionals (intervention researchers/clinicians plus aetiology/prevention researchers) have similar areas of concern and consensus priorities for research. When comparing the responses of the two groups, there was only one priority that overlapped: focusing on the need for CP registers. When comparing the consumers' results with those of the combined professionals, 50% of the questions had overlap (Table I). There were four areas identified by consumers that did not appear in the results of either groups of professionals: the relationship between CP and poverty, access in the community, effectiveness of alternative therapies, and pain management.

There were several significant findings when analysing the consumers' results on their own and in comparison with the professionals' results. Consensus was reached for 22 of the 23 questions in round II, suggesting a high level of stability between surveys for this group. The intervention and aetiology/prevention survey had a slightly lower degree of stability. Convergence was also highest for consumers as the range of scores for all questions was lowest in this group. The aetiology/prevention survey had several questions with the full range of possible answers, suggesting low convergence. Consumers ranked preventing CP as their highest priority for research and were the only group to identify exploring the therapeutic impact of stem cells. This particular group of consumers provided a balanced continuum of priorities, ranging from prevention to cure and effective interventions across the lifespan.

No literature-based questions were used in the consumer survey, to keep ideas purely as consumers' points of view. Yet, the only additional themes that appeared in the intervention survey that did not appear in the consumer survey were 'measurement and methodologies' and 'relationships' (Table SII). It was not expected that consumers would consider the research tools required to answer their questions. Relationships were ranked highly but did not reach consensus, so were not reported in the final consumers' results.

Half of the consumer-generated questions were reflected in the professionals' priority questions. Important lessons can be learnt from close examination of consumers' priorities that were not identified by professionals. Pain is an important focus for consumers; so too is the effectiveness of individual interventions including alternative therapies, physiotherapy, and parent education. The efficiency and effectiveness of service models, improving access, and understanding the complex relationship between CP and poverty were consensus high priorities. Consumers were interested in policies at a societal level that may improve their quality of life, whereas professionals were still focused on health interventions aimed at improvements in families' quality of life.

The potential limitations of this study are known weaknesses of the Delphi technique and include the subjective process used for defining expert panellists for the sampling frame, the representativeness of the panel assembled, and the panellists' ability to remain impartial in light of others' views when rating items on multiple rounds. Policy makers and administrators were included as participants, but not analysed as separate groups, and 'society at large' was not included at all. The Delphi technique is also limited by whether the anonymous nature influences accountability and response rates.[2] The consumer survey was limited by a small sample size. However, those who participated were highly involved, and response rates were higher than the other surveys. Small numbers also meant that multivariate statistics could not be used to identify whether GMFCS levels predict type of research questions seen as a high priority by consumers. Consensus, however, was highest for consumers; this suggested that the consumers involved considered the full range of disability in CP when prioritizing research.

The strengths of this study included acceptable response rates for rounds II and III, ranging from 60 to 100%,[13] and the sample being both multidisciplinary and international, so increasing the likelihood that the findings were representative of those who research CP. The most important strength of the study was that the consumers in this study were equal members on the expert panel.

The process of documenting research priorities in CP is critical to building the knowledge base for best-practice intervention. These findings provide direction for future CP research based on the consensus views of consumers and internationally renowned researchers and clinicians. The quest for answers to the research questions identified by Delphi panellists is both urgent and imperative.[2] It is acknowledged that priorities for research change as evidence builds to answer the questions. Phase II of this project will be to develop a web portal. With international collaboration, outcomes of research will be added to it, and unanswered questions will become the next priorities.

SOURCES OF SUPPORT

SUPPORTING INFORMATION

Additional supporting information may be found in the online version of this article:

Table SI: Participants (one person in this profession was predominantly working in policy[a] or senior[b] management/administration at the time of the study).

Table SII: High-priority questions for intervention researchers and clinicians.

Table SIII: High-priority questions for aetiology/prevention researchers.

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