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Setting a patient-centered research agenda for cerebral palsy: a participatory action research initiative



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Abbreviation

CPRN Cerebral Palsy Research Network

AIM To establish a patient-centered research agenda for cerebral palsy (CP).

METHOD We engaged a large cross-section of the extended community of people living with CP and those providing healthcare to people with CP ('the community') in an educational series and collaborative survey platform to establish an initial list of prioritized research ideas. After online workshops, a facilitated Delphi process was used to select the 20 highest priorities. Select participants attended an in-person workshop to provide comment and work toward consensus of research priorities.

RESULTS A research agenda for CP was developed by the community, which included consumers, clinicians, and researchers interested in advancing the established research agenda. The results included the top 16 research concepts produced by the process to shape and steward the research agenda, and an engaged cross section of the community.

INTERPRETATION It has been shown that proactively engaging consumers with clinical researchers may provide more meaningful research for the community. This study suggests that future research should have more focus on interventions and outcomes across the lifespan with increased emphasis on the following outcome measures: function, quality of life, and participation.

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Patient-Centered Research Agenda for CP *Paul H Gross et al.*

What this paper adds

- Comparative effectiveness of interventions, physical activity, and understanding ageing were leading themes.
- Longitudinal studies across the lifespan, clinical spectrum, and ages were highly ranked.
- Participants reported high value for participation outcomes.
- Participants reported great appreciation for the engagement between consumers and clinician researchers.

[main text]

Cerebral palsy (CP) is an umbrella term for non-progressive, permanent brain disorders that affect development of movement and posture and precipitate a host of secondary and chronic comorbidities.¹ A recent article reports that public funding of CP research between 2001 and 2013 increased but that funding for studies of clinical interventions

comprised only 19 percent of the funding and CP in adulthood comprised only 4 percent.² A survey of an online community of parents and caregivers ($n=1214$) of children with CP conducted by the nonprofit organization CP NOW found that available medical information was judged by survey respondents (parents) to be inadequate to guide medical decision making for children with CP.³ Perceived inadequacy of available information reported by survey respondents may be attributable to the diversity of clinical presentations in this population, the broad array of treatments and medical providers involved in care of persons with CP, the lack of comparative effectiveness research for treatments, and the long-term, progressive nature of CP sequelae. Regardless of cause, perceived inadequacy of information points to the need for a research agenda involving the extended community of people living with CP and those providing healthcare to people with CP that informs medical decision making for persons with CP.

Patient and caregiver involvement in the development of research is vital to producing research that improves the health and well-being of all patient populations,⁴ and, more specifically, for those with disabilities.⁵ However, efforts toward this end are somewhat limited. To date, three groups have published the results of their efforts that included persons with CP and their caregivers in the development of research agendas. Two of these efforts were focused broadly on neurodisability rather than specifically on CP and included caregivers and persons with several neurodevelopmental conditions.^{6,7} The third effort focused specifically on CP in Australia⁸ and may have limited generalizability outside of the Australian healthcare context. In addition, the McIntyre et al.⁸ study was conducted 10 years ago and differed from others in that it did not include an in-person meeting. It is possible that these meetings may foster a deeper understanding of various perspectives⁹ since they facilitate thoughtful information exchanges and the opportunity for creating relationships among stakeholders.

The Patient-Centered Outcomes Research Institute was established to promote comparative clinical effectiveness research that informs healthcare decisions. The Patient-Centered Outcomes Research Institute granted an award to the Cerebral Palsy Research Network (CPRN) and CP NOW to develop a patient-centered research agenda for CP. Called 'Research CP' for short, the effort sought to bring together and to

engage persons with CP and their caregivers, clinicians, scientists, policy makers, and advocates in the development of a patient-centered comparative effectiveness research agenda. The theoretical basis of 'Research CP' builds on the idea of 'nothing about us without us.' Research CP offers two enhancements to improve outcomes of the agenda creation effort: (1) educating the community (consumers, researchers, and clinicians) to give everyone basic information in order to inform the process of research idea generation and prioritization; and (2) developing and confirming the agenda together – having consumers, researchers, and clinicians working together at every step, from development of the concept of Research CP through education, idea generation, the Delphi process of bringing forth best ideas, and then confirmation of priorities. Because the authors believe a research agenda created with consumers would be more important and engaging to the entire community than one developed by professionals alone, we undertook a process to create such an agenda.

METHOD

This study utilized a community-based participatory approach combined with consensus building activities. The community included consumers (caregivers and persons with CP), professionals (clinicians and researchers), and advocates. Development of the research agenda included four steps: (1) conduct a series of online webinars to educate stakeholders about clinical trial design and comparative effectiveness research, patient registries, patient reported outcome measures and patient-centered research, the state of publicly funded CP research, collaborative research, and quality networks; (2) elicit from the larger community about research priorities for CP; (3) convene an in-person workshop to set a clinical research agenda that synthesizes elicited perspectives and establishes an engaged team of stakeholders; and (4) broadly disseminate results outlining the community's shared clinical effectiveness research agenda for CP.

Webinar series

Participants ($n=275$) were recruited from advocacy organizations, from the American Academy for Cerebral Palsy and Developmental Medicine, from Facebook groups, via advertising, and from followers of the CPRN and CP NOW websites. Participants

included those who participated in any of five live webinars as well as those who viewed recorded webinars after live presentations. There were five webinars: (1) an overview of the Research CP initiative, outlining the time commitment and objectives of the program, and after the first webinar, successive overviews of (2) methods for clinical comparative effectiveness research, (3) quality improvement, (4) patient-centered research and outcomes, and (5) clinical and patient-reported outcome registries. Active question and answer periods followed each webinar, along with a short posttest designed to reinforce learning and ensure engagement.

Generation, consolidation, and prioritization of research ideas

After the first webinar, those participants were invited to contribute to an iterative online process via Codigital Ltd. Codigital is an online external crowdsourcing intermediary that provided an independent platform for users to openly record, edit, and vote on each other's ideas allowing for the most prominent ideas to emerge naturally. Participants in the generation and prioritization processes are detailed in Table I. Initially, two questions were posed to elicit ideas: (1) What research questions are most important to you? and (2) If you could identify one area to improve in the care or treatment of people with CP, what would that be? These questions were purposefully written to engage researchers/clinicians by asking about research and consumers by asking about desired improvements to care or treatment in a language more familiar to their experience. Subsequently, over the next 20 days, participants received daily email messages notifying them of the opportunity to contribute new questions, edit existing questions, or vote on proposed ideas. Consumers, clinicians, and researchers all saw and participated in creating and voting on new questions in either domain. Overall, 392 ideas were generated, and 26 798 votes were cast in the paired comparison process that Codigital utilized to prioritize ideas elicited from the field.

The Research CP leadership team (six members: two each of consumers, clinicians, and researchers) participated in a facilitated Delphi process to eliminate redundancy and reduce the set of ideas to a number manageable for consideration in the in-person workshop. Several ideas that were more related to advocacy and education, as well as those that suggested guidelines development rather than new

research, were not included in the list for consideration at the workshop. After organization and consolidation, the Delphi process produced 20 highest-ranking, unique ideas to be brought forward for consideration and confirmation by workshop participants. (See Appendix S1, online supporting information, for complete ranked, consolidated, and categorized lists).

In-person workshop

Eighty-three applications were received for the in-person workshop in Chicago in June 2017. Applications were evaluated by the leadership team based primarily on webinar attendance, postwebinar survey completion, and engagement in the idea generation/refinement process. Engagement was measured by Codigital by assigning an engagement rank (Table I). Applicants were then categorized by type (patient/caregiver, provider/researcher, patient advocate), role (e.g. parent, caregiver, person with CP, clinical discipline), age of person with CP, severity of CP, ethnic group, and geographical factors to achieve a representative group of workshop participants. Invitations to the workshop were extended to 43 people including three personal care assistants; 41 could attend. With the leadership team, there were a total of 47 attendees not including the facilitator and a science writer.

At the workshop (see agenda in Appendix S2, online supporting information), the facilitator led a process designed to gather more detailed information and perspectives on the 20 highest-ranking research ideas. The facilitator not only guided the agenda but also assured that all opinions were heard throughout the meeting. After introductions, the leadership team reviewed with the full group of meeting participants the idea generation and synthesis processes that took place over the full period of participant engagement. In advance of the workshop, all participants submitted a description of their challenges with CP and desired outcomes for the workshop that was distributed with prereading materials. A subset of participants was asked to share their submissions aloud to set a tone of openness for the workshop. After the group discussion, suggestions on moving the research 'concepts' to actionable research 'questions' were discussed.

The full group was subsequently divided into four subgroups designed to include a diversity of consumer and clinician/researcher perspectives. Subgroups were facilitated by leadership team members. Each subgroup was asked to review a different subset of the highest-ranking research ideas. The groups discussed the intended meaning of the ideas and how they might best be applied in research. After these breakouts, the full group reconvened, and subgroups reported the results of their discussions including redundancies within the list of ideas. Discussion and consolidation produced a final list of highest research priorities.

The meeting facilitator then asked for each participant to summarize his or her most impactful observation from the whole experience. These observations were written on flip charts and consolidated in real time as a group. The meeting concluded with detailed exploration of the top three research concepts, including options to further narrow the concepts as well as study design and implementation issues. Examples of how CPRN infrastructure could accelerate development of the study were discussed. These examples were linked to how Research CP would provide a pool of interested and engaged patient stakeholders to participate in future research and dissemination efforts. In addition, next steps were presented, including preparation of a manuscript detailing the process and results, and how attendees could not only stay engaged in the conversation but also stay abreast of progress of the project and its intended impact on the resultant CP research agenda.

RESULTS

A summary of the process and participants at each stage is illustrated in Figure 1. Table I describes the great diversity in backgrounds and experiences of community participants and professional disciplines represented. There was also diversity in age, level of professional experience, and other factors that created greater representation of varied outlooks in the process.

Table II lists the top-ranked research ideas that resulted from the participatory action research methodology. Lifespan issues and longitudinal studies from childhood to adulthood were prioritized by Research CP participants. The focus on adulthood comes both from our adult participants, who desire more information about their current state,

and participants who are parents of young children who wish to know more about their child's future. These issues included questions related to symptoms such as pain and fatigue, as well as exercise, health, and wellness. There was a strong appreciation for the value of engagement between consumers and clinician researchers to further sharpen this agenda and make resultant research most meaningful and impactful. Participation outcomes were highly valued, particularly by our consumer participants.

Common observations shared by participants at the end of the meeting included the following: a desire to keep the momentum and community partnership moving forward, longitudinal studies across the age span and the clinical spectrum should drive research, the importance of the adult perspective including the cascading loss of function, and emphasizing participation as an important outcome measure.

DISCUSSION

This report describes the development of a research agenda for CP using a highly interactive consumer/clinician/investigator partnership. The study highlights the need to focus on longitudinal research that includes outcomes related to participation and quality of life. To our knowledge, only one other group has published their efforts to establish a prioritized research agenda specific to CP.⁸ That work differed from this study in several ways. Firstly, it involved only Australian participants who receive care in a socialized healthcare system and may have a different outlook on medical care and what it can and should do for individuals with CP. Secondly, the Australian study analyzed themes within two segregated participant groups. For example, Australian consumers identified questions in themes of prevention or cure, quality of life or community participation, and service provision or intervention, whereas intervention researchers or clinicians identified questions in themes of effective outcomes and effective research or services. The Research CP effort reflected group consensus with combined input from all participants to reach its conclusions. Thirdly, Research CP was limited to clinical research questions rather than including basic and translational research topics. Accordingly, results reflect a strong bias toward clinical questions with existing interventions. Finally, the Australian consensus process did not include in-person meetings, which Research CP participants considered an integral part of the

process. A key takeaway noted by many Research CP participants was an increased appreciation and respect of the partnership between consumers and clinician/researchers, which participants were eager to continue going forward.

Despite these marked differences, there was considerable concordance between the Australian and Research CP efforts. Both efforts highly prioritized areas of quality of life and participation, and service delivery and intervention – specifically dose, alternative treatments, and pain. A major difference between the efforts was the strong value placed in the Research CP effort on the need for longitudinal work that examines outcomes across the lifespan. There were also similarities between outcomes of the Research CP effort and the two reports focused more broadly on neurodisability, including a focus on comparative effectiveness studies.

Outcomes such as decreased tone, improved reach kinematics, or increased range of motion generated much less enthusiasm than participation outcomes in the study, even though those former outcomes are generally the more direct targets of most medical therapies and therapeutic interventions. Although clinicians hope that improved body function and structure will increase participation, this has yet to be demonstrated.¹⁰ Moreover, participation can be targeted directly through adaptation rather than indirectly via attempts to change body structure and function.¹¹ The discussions, both through idea generation and at the workshop, indicate that a focus on more work to determine best practices for improving participation, especially at the intersection of participation and technology, is of paramount importance. Fortunately, there has been a shift from body structure and function measures to participation measures in research related to disability, and future work in CP will need to continue that trend.

Within the overarching priority of longitudinal outcomes, several themes were identified. Firstly, we need better understanding of the etiology and treatment of those issues that are commonly seen in adults with CP. The group clearly identified chronic pain that interferes with life participation, fatigue, and depression as critical problems. There was great interest in exercise and fitness, and early functional loss, all of which are well described in adults with CP.^{12,13} Secondly, more research is needed into pediatric interventions and how they influence adult outcomes. Some work has been done in this area, including studies of selective dorsal rhizotomy^{14,15} and intrathecal

baclofen.¹⁶ Much more research needs to focus on this area, including studies of intensive therapy interventions and other treatments into which families heavily invest their time and resources with the hope of maximizing their child's potential as an adult. Thirdly, there were many questions about physiology of the ageing process combined with a developmental neurologic disorder such as CP. There is evidence of increased risk of chronic cardiovascular and musculoskeletal disease in adults with CP.¹⁷ It is known that muscles, bones, joints, and most body systems are affected by both processes, but the community needs to know more about prevention and treatment. Finally, emphasis on participation suggests that we need greater information about psychosocial issues related to growing up with CP including employment, relationships, and best practices for encouraging independence. The existing literature suggests that much more needs to be done to help adults with CP.^{18,19} This emphasis on lifespan studies (referenced specifically in half of the research ideas, Table II), combined with the desire for work that focuses on participation as noted above, prioritizes the need to validate and implement a set of participation measures for longitudinal study of outcomes for individuals with CP.

Questions such as adult outcomes and relationships between interventions and participation are difficult to answer unless the investigation involves large populations followed over time. Both clinical interventions and adults with CP were identified as underfunded topics by Wu et al.,² which is consistent with what the community prioritized as top research ideas. This consumer driven agenda is different than current directions in CP research, in that it emphasizes prioritization for longitudinal work across the lifespan. A recent review of 2016 funded projects in CP²⁰ demonstrates that only 3 of 74 projects funded (4%) were specific to adults, and none were longitudinal studies or addressed lifespan issues from childhood to adulthood.

The lack of studies addressing lifespan issues is perhaps due to the structure of most public funding mechanisms that typically fund for periods between 1 and 5 years and are not conducive to longitudinal studies across the lifespan. The consumer driven agenda from this study suggests the need to shift the focus and consider different funding mechanisms across a greater number of years to address lifespan and longitudinal research in this population.

Limitations

The Codigital voting tended to favor broad research questions over more actionable narrowly defined questions. Consequently, certain research topics, very important to a smaller slice of participants, may not have been represented in the top 100 even though they are very 'patient-centered'. The leadership group considered bringing some of these 'underserved' questions forward for discussion but decided to honor the ranking system (see Appendix S1, online supporting information).

Contributions to the agenda through Codigital were influenced by the number and engagement of groups of participants. Amongst clinicians, some disciplines were more heavily represented and had more impact on the ideas and rankings. This may have occurred because of differences in recruiting efforts between disciplines, and other factors beyond the control of the leadership team. It is notable, however, that the rehabilitation disciplines that spend significant time with patients with CP over the lifespan (e.g. physical therapy, occupational therapy, physiatry) contributed significantly to the list of priorities. The differential contribution of the various professionals was balanced by the fact that the consumers contributed more than 60 percent of the resultant agenda. See participant description (Table I) to see relative contribution of various audiences.

Promotion of the initiative and engagement in the process required frequent email access and initial contact via e-mail or social media. It is likely that consumer participants represented a selection bias common to social media outlets targeted for participants (trending toward white, middle-income women). While there were parents of teens participating, the authors are not aware of any teens participating directly so that important voice was missing. Funding limitations of the workshop may have reinforced this bias by bringing together only those who were most engaged in the digital portion of the process.

Next steps and conclusion

The CPRN registry, created as a result of development of the National Institutes of Health Strategic Plan for CP,²¹ captures a breadth of patient characteristics,

interventions, and patient reported outcomes. Use of these evolving data sets to provide unique insights into long-term outcomes from interventions, and effects of various medical issues and environmental factors for people with CP throughout their lifespan, are encouraged in two recent National Institutes of Health strategic plans.^{21,22} CPRN's community registry provides a platform to continue longitudinal studies focused on outcomes after transition from a pediatric to adult care center. It enables linkage of those long-term patient reported outcomes to clinical registry data collected from the point of their diagnosis with CP. It will provide an important tool for investigators to answer questions identified in this effort as highest priority to consumers and clinicians/researchers.

Potential benefits of patient-centered research were strongly reinforced by the Research CP initiative. The partnership formed between consumers and clinician researchers was invaluable not only to set the research agenda but also for the potential to develop relationships that will enable collaboration in the development of studies that stem from these priorities. This study provides a guide for important research for CPRN and the field of CP to improve care and outcomes for individuals within this population.

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Supporting information

The following additional material may be found online:

Appendix S1: Complete list of research ideas

Appendix S2: Workshop agenda

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Table I: Participant description in Research CP initiative

Type of participant	Webinar participants	Codigital participants	% contribution ^a	Workshop participants
Parent or caregiver of a person with CP	99	72	31.91	15
Person with CP	38	33	27.60	6
Clinician, therapist, or researcher	132	93	38.76	23
CP community advocate	6	4	1.73	3 ^b
Total	275	202	100	47
GMFCS level ^c				
I	N/A	4	3.41	0
II	N/A	35	13.78	3
III	N/A	19	15.64	5
IV	N/A	18	10.95	9
V	N/A	25	12.42	3
Unknown GMFCS	N/A	4	1.08	1
Average/median age of person with CP	N/A	46/48	N/A	52.2/49
Average/median age of person with CP	N/A	13/10	N/A	13.6/14

CP represented by parent/caregiver				
Clinician/researcher disciplines				
Complex care pediatrics	2	1	0.01	0
Developmental pediatrics	10	6	0.36	3
Neurology	2	2	2.72	2
Neurosurgery	3	2	0.82	2
Nursing	3	2	5.56	0
Occupational therapy	11	9	5.08	1
Ophthalmology	1	1	0.18	0
Orthopedic surgery	11	8	1.97	2
Orthotics	1	1	0.43	0
Physical medicine and rehabilitation	17	9	6.39	3
Physical therapy	50	37	11.90	4
Research	17	10	3.01	5
Speech and language therapy	4	0	0.00	1
PT/research administration	0	1	0.05	0
Speech-language pathology and audiology	0	2	0.17	0
Speech language/feeding therapy	0	1	0.04	0

^aThe overall percentage contribution is calculated from (1) the contribution to the content of the top ideas and (2) the contribution to the voting that put those ideas to the top. Codigital imposed limits on voting to prevent a participant from having too great an impact on rankings. The percentages sum to 100% so this column demonstrates how much impact any one cohort had on the final research agenda. ^bAll three CP community advocates were also parents of children with CP. ^cGMFCS level was both for the people with CP that participated, and those people represented by parents and caregivers. CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; PT, physical therapy.

Table II: Top 16 research ideas^a

Number	Research ideas	Codigital question	Codigital rank
1	Research the issues around ageing with CP, to understand not only how to treat adults now, but also to update our treatments & therapies with children who have CP to prevent some of the secondary impairments such as pain, fatigue, and functional loss.	Care Treatment ^b	1
2	What are the best long-term exercise/strength training strategies to improve activity, participation and health, minimize pain, and maximize function in each GMFCS category across the lifespan?	Research ^c	1
3	Which interventions (surgeries, injections, medications and therapies [orthotics, equipment, training]) are associated with better functional outcomes (important to child/family) controlling for GMFCS level, age, and comorbidities	Research	2
4	Increasing age is related to pain and fatigue in people with CP, regardless of GMFCS level. What variables are important to monitor/treat early on in life to prevent the development of pain and fatigue later on in life?	Research	4
5	What are the best methods and ways to reduce pain, falling, lack of stamina, and deterioration of function that can have a negative impact on the quality of life for people with CP especially in adulthood?	Research	5
6	Research effective ways to build and maintain	Care	6

	strength, flexibility, and endurance health in children and adults with CP. How can we find better ways to successfully integrate these into daily living?	treatment	
7	Develop and test effective methods for exercise and increased physical activity for individuals who are less ambulatory (some GMFCS III, and GMFCS IV and V) including techniques, proper dosing, and information on effects on strength and health.	Research	7
8	How can we best leverage the brain's neuroplasticity to retrain neural pathways for improved motor function, speech function, and mobility?	Research	8
9	Have large-scale studies that follow children with the various types of CP throughout adulthood to discover how the ageing process affects individuals with different types of CP and severity levels.	Care treatment	11
10	What are effective treatment methods to address differential outcomes in adolescents and adults related to pain, fatigue, and early functional loss?	Research	11
11	Study the outcomes of complementary and nontraditional therapies, i.e. hyperbaric oxygen, hippo, swimming/aquatic, Feldenkrais, massage, yoga, Tai Chi, music, recreational, acupuncture, etc. Review efficacy, costs, insurance support probabilities.	Care treatment	12
12	Research the effectiveness of intensive physical therapy programs, bursts of services, combined protocols (i.e. botulinum neurotoxin with intensive PT, etc.). Evidence of efficacy is needed to get ALL insurers to fully cover the treatment options that work.	Research	13
13	How do we best maximize functional independence	Care	15

	and life participation of children and adults with CP?	treatment	
14	Identify biomarkers (neuroimaging, blood, CSF, amniotic fluid) to help determine which individuals respond best to which interventions (therapy, medical, surgical) so that treatment approaches can be tailored to each individual person with CP.	Care treatment	18
15	Quality of life is an important goal for a number of questions related to CP. How do we quantify that so that we can really answer which interventions produce the greatest benefit to quality of life?	Research	18
16	Not much work has been done on the cognitive impairments including difficulties with math and any subject with spatial orientation.	Research	21

^aTop 16 research ideas table lists equally ranked ideas from Codigital in alphabetical order of top level questions, i.e. care treatment ideas of equal rank to research ideas are listed first. ^bCare treatment 1 refers to the top ranked (based on votes) research idea in answer to the question: If you could identify one area to improve in the care or treatment of people with CP what would that be? ^cResearch 1 refers to the top ranked (based on votes) research idea in answer to the question: What research questions are most important to you? CP, cerebral palsy; GMFCS, Gross Motor Function Classification System; PT, physical therapy; CSF, cerebrospinal fluid.

[figure legend]

Figure 1: Flow chart showing the process and numbers of participants and results at each stage

Figure 1 Flow chart showing the process and numbers of participants and results at each stage

