

Commentary

Co-building a new landscape in pediatric chronic pain research: Patient partner and researcher perspectives on meaningful patient engagement

Kathryn A. Birnie, Katherine Dib, and Carley Ouellette

“The modern field of pediatric pain was born out of a marriage of science and public concern...Without the science, the public outcry would not have had a knowledge base. Without the public outcry, the knowledge would have languished for years before being adopted” (McGrath, 2011, p. 2458).

Chronic pain affects 11-38% of children and adolescents (King et al., 2011). While researchers have systematically identified important knowledge gaps and priorities for future research in pediatric pain (Caes et al. 2016), the patient and family voice is largely lacking. Estimates suggest that it takes the length of one childhood, or approximately 17 years, for research to change practice (Morris et al., 2011). Despite growing research evidence of effective pediatric pain assessment and management tools, pediatric pain care remains suboptimal (Stevens et al., 2011; Stevens et al., 2012). Lessons learned from the birth of the field of pediatric pain critically identify the value of science-public partnerships to address this.

Broadly stated, patient and public engagement in research is “...research being carried out ‘with’ or ‘by’ members of the public rather than ‘to’, ‘about’, or ‘for’ them” (INVOLVE, 2018). Patient engagement represents an evolution from the traditional view of patients as research subjects or participants to one that empowers patients, otherwise identified as people with lived experience,

as partners and co-builders on research teams. Patient engagement shifts across a continuum from minimally informing patients or enrolling them as research participants (traditional health research) to greater engagement in consultation (ad hoc input), involvement (advisory team member), collaboration (equal partnership), and leadership (patient as leader; Shippee et al., 2015; Amirav et al., 2017). Guiding principles underlying meaningful patient engagement are inclusiveness, support, mutual respect, and co-building (Canadian Institutes for Health Research, 2018). Although the empirical study of patient engagement is in its relative infancy (Shippee et al., 2015; Manafo et al., 2018), existing evidence suggests that engaging patients as collaborators and partners, including children and their families, can enhance the quality, appropriateness, and relevance across all stages of the research process (Brett et al., 2014; Domecq et al., 2014; Thompson et al., 2015; Shen et al., 2017). Specific examples include increased enrolment in studies and decreased attrition, improved data collection tools, improved dissemination and implementation of study findings, greater insight into research area, improved researcher-community rapport, better alignment of research objectives to patient-identified priorities, and improved research effectiveness (Manafo et al., 2018).

Leadership by health research funding agencies, such as the Canadian Institutes of Health Research (CIHR) and the Patient-Centered

Outcomes Research Institute (PCORI; Frank et al., 2014) have promoted patient engagement and co-production through targeted funds and/or requirements to include knowledge users, such as patients or family members, as members of research teams. This welcome progress in the health research landscape has facilitated greater opportunity and empowerment of the expert patient and family voice.

Our goal is to share early reflections from the perspectives of patients and a researcher/care provider engaged in an ongoing patient engagement partnership in pediatric chronic pain in Canada. We hope to encourage a culture shift toward greater patient engagement and co-production in pediatric pain research. We offer learning from our own experience brokering and managing such patient-researcher partnerships, as well as identifying benefits and challenges still to be addressed.

Our Canadian patient engagement partnership in pediatric chronic pain

We are a group of eight researchers and health care providers (nurses, psychologists, physicians) from five multidisciplinary outpatient pediatric chronic pain programs and affiliated institutions (Calgary, Saskatoon, Ottawa, Toronto, and Halifax), in addition to seven (former) patient and parent partners across Canada. Our program team is complimented by collaborators from two patient stakeholder organizations (PainBC and the ILC Foundation), and a government policymaker (from the Ministry of Health and Long Term Care in Ontario, Canada). Our collaboration began with a successful Canadian Institutes of Health Research (CIHR) Strategy for Patient-Oriented Research (SPOR) Collaboration Grant (awarded March 2017), and is additionally supported through funds from The Hospital for Sick Children, the Alberta Children's Hospital Research Institute, the Saskatchewan Health Research Foundation, the Children's Hospital of Eastern Ontario Research Institute, and the Dalhousie University Faculty of Medicine. Our patient engagement project is linked through in-kind support to the Chronic Pain Network, a large scale CIHR-funded pan-Canadian collaboration of patients, researchers, healthcare professionals, educators, industry and government policy advisors to direct new research in chronic

pain, train researchers and care providers, and translate findings into knowledge and policy (www.cpn-rdc.ca).

The primary aim of our ongoing project is to co-build with patients, families, researchers, policymakers and advocacy groups, a sustainable patient engagement strategy to guide pediatric chronic pain research and practice in Canada. Specifically, our project strives to (1) co-create a Canadian patient engagement registry to link pediatric patients and families who are interested and able to act as partners in research in pediatric chronic pain with researchers seeking such partnerships; (2) facilitate pediatric specific patient engagement training for patients, families, care providers, and researchers; and (3) co-build a Canadian research agenda for pediatric chronic pain with patients, families, and care providers using an established framework for research priority setting partnerships (National Institute for Health Research, 2018).

Patient and parent partners were identified primarily through prior connections with specialized multidisciplinary pediatric chronic pain clinics. They are former pediatric patients who had previously expressed an interest in being involved in future projects or efforts to improve pediatric pain care. Some had existing relationships with project investigators as patient advisors or partners with other research projects.

Patient and parent partner input and expertise are sought throughout project activities from design to dissemination. Examples include co-development of public surveys and public facing materials for research priority setting (e.g. videos, posters), co-analysis of study findings and co-facilitation of the research priority setting process (e.g. reviewing, interpreting, and rephrasing of public comments into testable research questions, co-leading final in-person priority setting workshop, interpretation of qualitative interview data from usability testing to direct changes to patient engagement registry), co-development and co-presentation of project posters and symposia at professional conferences (e.g. Canadian Pain Society, International Forum on Pediatric Pain), and co-authorship of newsletter and peer-reviewed publications (e.g. this paper). Collaborative tools used to facilitate team

communication include telephone calls, emails, online video conferencing, and online collaborative project management platforms. Patients and parent partners are compensated at an hourly wage for their work on project-related activities from project grant funds. This is consistent with new recommendations developed by the Canadian national CIHR-funded SPOR (Strategy for Patient Oriented Research) Networks in Chronic Disease and their patient partners (SPOR Networks in Chronic Diseases and the PICHI Network, 2018). These recommendations identify core values underlying compensation of patient partners, including value (of their time, expertise, experiences, and networks), respect, fairness, transparency, clarity of expectations, representativeness, responsiveness, flexibility, and choice. We have planned evaluation of our partnership using evidence-based tools assessing quality engagement (Abelson et al., 2016).

Reflections from patient partners with lived experience with pediatric chronic pain

Carley Ouellette is a registered nurse working as a research nurse at Hamilton Health Sciences, and is currently completing her Masters of Science (thesis-based) degree in Nursing at McMaster University.

Participating as a patient partner has given a significant sense of empowerment as a young person living with pain. By actively participating in research, I contribute my experiences as expertise to a team that has potential to change how pain is managed, treated, and understood. I think it is crucially important for researchers and patients/families to collaborate in order to effectively address the most important health research questions. This opportunity has provided personal and professional growth through professional communication, collaboration, and experiential learning not only as a nurse but also as a patient living with a chronic condition. I have valued the opportunity to work with my mother who is parent partner on this project, as it has helped us both understand the impact and value of including the perspective of families in pain research, providing us both the platform to advocate as

patient and parent in a constructive, supportive environment.

While great strides have been made through this patient engagement project, some challenges should be considered and acknowledged. The most noted challenge I have considered is in relation to the diversity within our working group, namely noticing similarities among partners, such as the like-mindedness approach to participation, willingness to advocate, education level, familial relationships, and experiences living with pain. Having such eager participants to contribute and step forward is important, however at times, our perspectives lack diversity that may limit the generalizability to those of the greater public. This can result in research being directed by a specific subset of patients and families. With this in mind, it is important to note the efforts of the research team in attempting to recruit a diverse working group by specifically seeking partners to fill obvious gaps on the research team (e.g. fathers and parents of younger children with chronic pain).

I know I would not be who I am today, pursuing my current professional path, without my pain. My pain has transformed how I work with patients, my personal self-care, and how I view the world. With my challenges, I have focused my energy on how I can contribute and enhance research for the next generations of young people with pain. This is why patient engagement is vitally important in addressing real-world problems. I have also learned that it is crucial that patients are engaged with meaningful consideration to one's collaboration and contribution to avoid tokenism, such as seeing our suggestions come to fruition in the research design and conduct. Moreover, as a young trainee, I value the importance of the patient/family perspective, and furthermore, know that as a young researcher, patient engagement will be an inherent partnership I will always strive to achieve in my own program of research.

Katherine Dib is currently an undergraduate student studying neuroscience at Dalhousie University.

To me, meaningful engagement is contributing value to a research project and having the project add value to your own life. The project has given me confidence by focusing on my abilities and the value I can bring. For example, I have provided feedback to ensure that the wording used in our surveys with families is understandable and resonates with young people with chronic pain. This makes it easier for them to share their ideas, in turn giving us more responses for more accurate research priority setting. This is amazing because there are many activities I cannot do because of chronic pain. There are many ways to promote meaningful engagement in the project but I believe the best way to ensure it is assessing along the way with all team members. To keep the team engaged and interested, it is important that we all feel like our contributions are valued.

The greatest part of this project is having so many other patient partners and parent partners on the team. It is common to hear many different perspectives from the patient and parent partners that would not be there if it was just one or two of us. We mostly communicate with each other and with the project lead through email, online discussion forums, and videoconference to co-produce, provide feedback, and guide project-related activities typically initiated by the project lead. I like that our team has one researcher project lead who is primarily responsible for managing the patient partner contributions as it gives us all a chance to feel comfortable and I don't feel an obligation to be too formal. This is great not only to bring forth my ideas, but also to adjust the workload or type of work if needed. The downside is that I don't always know what the researchers are working on with the project. When there is collaboration with the whole team it is less intimidating due to having multiple patient and parent partners on the calls.

The experience the project is giving me and the recognition that comes along with it is very valuable to me. The project leader reaches out individually to the patient partners to find out what

our interests are and if we would like to take on extra projects. This strategy is helpful because it allows me to choose if I want to strengthen certain skills like writing and public speaking, and ensures that I have a platform to do so. Sharing my experience as a patient partner on this project as part of a patient engagement panel and co-presenting a poster at the 11th International Forum on Pediatric Pain was empowering. Having the chance to be a co-author on presentations and publications shows that I am a valued member of the team. There are so many opportunities and ways we are recognized with this project, and each vary in how meaningful they are to patient partners. It is also important to note that these forms of recognition come at different points in the project. I like this because it continuously reminds me of the benefits of the project rather than only at the beginning or the end. I also receive compensation for the project, which I appreciate because I had to previously stop working because of my pain.

I also have the opportunity as part of this project to network with incredible researchers. Being on a team of researchers and health care providers who understand my pain journey gives the sense of having a community behind you. We are all working towards the same goal and that is clear in the work that we are doing. I never thought I would have the opportunity to play such a crucial role in a research project.

Reflections from a pediatric pain researcher and care provider

Kathryn Birnie, PhD, CPsych is a psychologist and postdoctoral fellow at the University of Toronto and The Hospital for Sick Children. She leads this national Canadian patient engagement project in pediatric chronic pain.

No researcher wants their efforts to take an estimated 17 years to achieve real world impact (Morris et al., 2011). Engaging and empowering the patient and family voice in directing, implementing, and sharing pediatric pain research feels like a powerful step towards ensuring benefit and usefulness of our work. Overall, I have found patient engagement adeptly pulls together my roles as a care provider and as a scientist in an enjoyable,

rewarding, and meaningful way. While this work takes time and effort, it is a privilege to empower the voice of patients and their parents when their journey towards chronic pain care has often been fraught with disbelief and denial of their symptoms by others.

While leading this project, I have encountered a continual need for flexibility. This has included making space for patient partner pain flares that require time away from project responsibilities, the need to identify new patient partners when priorities shift, and an openness to fluctuating levels of involvement depending on the project activity. Undoubtedly, engaging patient and parent partner relationships takes additional time. This project has opened opportunities to educate and champion patient engagement with others at all levels within and outside our institutions, with families, and with other researchers. We have navigated the concern about potential dual relationships between care providers on our team and current or past patients as partners by having myself as a central project lead, with no clinical relationship (former or current) with any of the patient partners or their families. I serve as the main liaison and project contact for all patient and parent partners. We have also worked with our institutional research ethics boards to provide accessible research ethics and privacy training to our patient and parent partners (e.g. through a brief 1-hour online presentation covering the Canadian Tri-Council Policy Statement on Ethical Conduct for Research Involving Humans; Canadian Institutes for Health Research et al., 2014).

The rapid growth in patient engagement resources and initiatives at the project, local institution, national, and international level is striking. This is an area of great interest and potential, but one that remains in need of continued rigorous empirical inquiry regarding the value of such partnerships to the research process and its outcomes. Patients and families are interested in these partnerships, but we must ensure that we, as health care providers and researchers, are willing and adequately prepared to engage in these partnerships as well. This may mean that we start before we're ready and learn together, but do so with a spirit that authentically values the patient and parent expertise of lived experience. Practical

considerations include adequate time and resources, as well as personal alignment with patient engagement values (Canadian Institutes for Health Research, 2018). Above all else, it is critical to avoid any potential for tokenism. This can be done by involving patient partners in all phases of research, ensuring clear expectations of their roles, facilitating and encouraging patient partner contributions, and providing specific and necessary information and training to do so, as well as appropriately recognizing their contributions (Kirwan et al., 2017). Researchers considering this work may find it useful to consult emerging guidelines and practices for patient engagement in research from leading groups and organizations listed in the Appendix and peer-reviewed publications cited herein (e.g. Kirwan et al., 2017; Manafo et al., 2018).

Conclusions and future hopes

By enhancing pediatric patient and family engagement in pediatric chronic pain research, we believe that research will be more effective, efficient, and meaningful to pediatric patients and their families. This will increase the likelihood that research will be taken up in clinical care, lead to better patient outcomes, and avoid wasting research funds or effort in areas that are not of importance to patients. To achieve this, we believe it is imperative to engage pediatric patients and family members as co-producers in the preparation, execution, and translation of research in pediatric chronic pain.

We hope that our efforts will inspire others about the value of patient engagement, while also considering the challenges and work still to be done. We recognize that this culture shift opens new roles and ways of relating for all involved - patients, parents, families, clinicians, researchers, and policymakers. This exciting yet daunting venture demonstrates the need for further scholarly activity regarding best practices and value of patient engagement.

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Acknowledgement

We would like to acknowledge other current and past members of our project team including Dr. Jennifer Stinson, Mary Anne Dib, Kimberly Nelson, Matthew and Bill Dawson, Nikki Cooke, Dolores Pahtayken, Dr. Krista Baerg, Dr. Jill Chorney, Dr. Paula Forgeron, Dr. Christine Lamontagne, Dr. Patricia Poulin, and Dr. Melanie Noel. We would also like to acknowledge supporting staff and students for this project, including Lauren Harris, Cynthia Nguyen, Yalinie Kulandaivelu, Casey McMahan, and Adena Cox. K.A. Birnie is funded by a CIHR Fellowship.

Appendix: Patient Engagement Resource List

There is an ever-growing list of resources for members of the public, care providers, and researchers to guide patient engagement in research. Here are a few websites we have found useful, many including practical guidelines, tools, and recommendations:

- CIHR – Patient Engagement (www.cihr-irsc.gc.ca/e/45851.html)
- Patient-Centered Outcomes Research Institute (PCORI; www.pcori.org/engagement)
- INVOLVE, National Institute for Health Research (www.invo.org.uk)
- Institute for Patient- and Family-Centered Care (<http://ipfcc.org>)
- Canadian Foundation for Healthcare Improvement (CFHI) Patient Engagement Resource Hub (www.cfhi-fcass.ca/WhatWeDo/PatientEngagement/PatientEngagementResourceHub.aspx)
- The Alberta SPOR SUPPORT Unit (<http://www.aihealthsolutions.ca/initiatives-partnerships/spor/>)
- Centre of Excellence on Partnership with Patients and the Public (<https://ceppp.ca/en/>)

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