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Healthcare Quarterly

Engaging Patients in Research Evidence Uptake: The Ontario Experience

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From the Guest Editor

The Growing Imperative for Patient and Caregiver Partnership

Anne Wojtak

This special edition of *Healthcare Quarterly* (*HQ*) has been developed through a novel partnership between the Ontario Strategy for Patient-Oriented Research (SPOR) SUPPORT Unit (OSSU) and Longwoods Publishing. The concept for this edition emerged from an alignment of interests between the partners. OSSU's mandate is to support patient-oriented health and health services research in Ontario and to facilitate the uptake of research evidence to improve health policy and decision making. *HQ*'s mission is to recognize, nurture and champion excellence in the Canadian healthcare system by sharing leading practices in health services delivery and policy development. As we look toward the future of healthcare and health system transformation, leading practices will be increasingly defined by the degree to which they are designed and implemented in full partnership with patients and caregivers. This perspective, shared by both OSSU and Longwoods, is at the heart of this publication.

OSSU's Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Awards support patient- and caregiver-oriented research that has had a wide impact in Ontario. The 15 EMPOWER grant winners featured in this edition exemplify patient and caregiver empowerment and co-design. Growing evidence underscores the imperative for patient and caregiver partnership not just in research but in every aspect of healthcare design and implementation, from the development of individual patient care plans to macro-level policy and governance of health systems. Longwoods' recent edition of *HQ* on "Resetting the Future of Healthcare Leadership" (Wojtak and Stuart 2021) emphasized this construct, including perspectives on what true patient partnership looks like and how it changes our systems.

The common learnings across the EMPOWER research projects, including the importance of addressing health equity, being flexible in tackling new challenges and evaluating patient engagement, do not only apply to research. Furthermore, the application of integrated knowledge translation, as discussed in the Introduction (Gill et al. 2022), can guide the broader spheres of healthcare policy and leadership, whereby end-users of the system are engaged in policy, strategy and governance to optimize system performance and achieve improvements in healthcare outcomes. In other words, while the focus of OSSU's EMPOWER Awards is patient-oriented research, what is highlighted in these articles extends well beyond that.

In creating this special edition of *HQ*, the combined editorial team from OSSU and Longwoods worked collaboratively over several months to review the submissions, discuss the findings and identify the core themes. Patients and caregivers were equal partners in all aspects of this effort as members of the individual research teams, co-authors, participants in the editorial board and commentary providers. This partnership was important not only because it was aligned with the EMPOWER approach but also because it markedly improved our editorial process and outcome. It is our hope that readers of this special edition, whether they are researchers, policy makers, clinicians or patients, can apply the tools and knowledge on partnership and co-design from these pages to help improve our health system regardless of where they sit within it.

References

Gill, P.J., D. Urajnik and R. Ganann. 2022. Empowering and Accelerating Impacts of Patient-Oriented Research. *Healthcare Quarterly* 24(Special Issue): 5–9. doi:10.12927/hcq.2022.26781.

Wojtak, A. and N. Stuart. 2021. Resetting the Future of Healthcare Leadership. *Healthcare Quarterly* 24(3): 1–3. doi:10.12927/hcq.2021.26627.

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Empowering and Accelerating Impacts of Patient-Oriented Research

Peter J. Gill, Diana Urajnik and Rebecca Ganann

Abstract
Patient partnerships require adequate planning, support and funding to mobilize knowledge and accelerate impact. We outline the themes and foreground ways in which the Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach teams have advanced patient-oriented research in Ontario.

Introduction

At its core, the aim of health research is to improve patient care and outcomes. Yet, for decades, patients have not been active members of the research enterprise. How can we ensure that healthcare research is relevant and applicable to the ultimate end-users without involving them from the outset? The growing recognition of the need to partner with patients has led to large-scale initiatives to put patients at the centre by engaging them in research teams and decisions. Canada has been a global leader with the launch of the Strategy for Patient-Oriented Research (SPOR) in 2015 (CIHR 2019). Patients should be involved in all stages of research, including the development of the research question, applying for and securing funding, conducting the study, interpreting the results, co-authoring knowledge products and disseminating information (Gill and Cartwright 2021). As we seek to address a key priority of the *CIHR Strategic Plan 2021–2031* to "Pursue Health Equity through Research" (CIHR 2021: 4), researchers need to pursue equity through patient-oriented research (POR) aims and through the composition of teams and engagement of patient partners throughout the research enterprise.

Parallel to the greater awareness and emphasis of patient involvement and health equity in research is the increased recognition of effective knowledge translation approaches. Simply conducting research and assuming that new information will naturally infuse into clinical practice is naive and wasteful. Knowledge translation refers to activities that help raise awareness of research findings (e.g., presentations, lay summaries, etc.) (CIHR 2016). In contrast, integrated knowledge translation (IKT) involves knowledge users, including patients and decision makers, as research team members from the outset. IKT is collaborative, with a commitment to co-production that increases the applicability and relevance of final products (Banner et al. 2019). Previous research has shown that projects incorporating an IKT approach, including integrating patients as partners, leads to optimized health system performance and improvements in health outcomes (CIHR 2015).

Patient partnership and IKT require adequate planning, support and funding. Recognizing the importance of knowledge mobilization, the Ontario SPOR SUPPORT Unit launched the Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Awards in 2019 (OSSU 2021). These awards aimed to provide additional end-of-grant funding to POR projects to empower wider research impacts in Ontario.

EMPOWER Goals and Outcomes

Canada's Strategy for Patient-Oriented Research (SPOR) was created by the Canadian Institutes for Health Research to engage patients, researchers, clinicians, policy makers, industry representatives and other health system professionals. It is a national collaboration of federal, provincial and territorial partners dedicated to enhancing the integration of research into care. The goal of SPOR is to integrate funding, research and expertise with users' perspectives and experience to foster evidence-informed healthcare to ensure greater quality, accountability and accessibility and inform policy and practice.

The Ontario SPOR SUPPORT Unit (OSSU) is a collaboration of leading health research centres, research initiatives and a coordinating centre dedicated to supporting patient-oriented research (POR) in Ontario. The coordinating centre facilitates, connects and integrates activities across the network, including the facilitation, implementation and mobilization of SPOR in Ontario.

The Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Awards launched by OSSU in 2019 aim to provide additional end-of-grant funding to recently completed POR projects to enhance and broaden the uptake and impact of research evidence. In particular, the goal is to promote and enable patient-partnered research teams to build relationships with patient partners and push evidence to those who can use it to inform their healthcare policy and decision making.

The articles in this special issue outline the experiences of the 15 EMPOWER Grant Award winners from 2019 to 2021. The special issue is divided into main themes based on the patient-partnering methods and tools used by the EMPOWER teams: (1) priority setting and best practices; (2) equity in patient partnerships; (3) co-designing interventions and tools; (4) tools for patient engagement; and (5) patient- or community-driven projects. This article will introduce these themes and foreground the ways in which the EMPOWER teams have advanced POR in Ontario. Each article is led or co-authored by patient partners, and the authors collectively describe how engagement was initiated and sustained, the insights gained through engagement and the impacts achieved.

Priority Setting and Best Practices

When conceptualizing research, an important question often arises: what topics or priorities should be researched? Identifying priorities provides a set of specific, concrete tasks to achieve over time. They are also strategic to focus limited time and energy. There are numerous ways that research networks, organizations, funding agencies and foundations can identify priorities. It can be done informally, such as based on strategic skills, or formally, such as based on burden of illness, cost and variation in care, or formally by using consensus methods (e.g., Delphi surveys) (Manafò et al. 2018). In alignment with POR principles, patients should be actively involved in identifying research priorities; James Lind Alliance Priority Setting Partnerships are a commonly used approach to engage patients, caregivers and clinicians in identifying and prioritizing unanswered research questions that are of shared importance (Nygaard et al. 2019).

After identifying the top 10 patient-oriented priorities in pediatric chronic pain (Birnie et al. 2019), Birnie et al. (2022b), in their 2020 EMPOWER project, partnered with youth and parents to maximize the impact of the priorities in research, care and policy. Building on the strong patient involvement strategies used to set the priorities, the team partnered with the pediatric chronic pain community to generate awareness, facilitate collaborative dialogue and innovation to address the priorities and engage the broader pediatric pain community to integrate and adopt the priorities into clinical practice, policy and advocacy.

Another example of patient-oriented priority setting was led by Ristevski et al. (2022b). These authors identified the top 10 research priorities in Canada for retinoblastoma (Flegg et al. 2020), a serious and rare cancer of the eye. Recognizing the challenges of mobilizing action on the priorities, they developed a number of knowledge dissemination tools and processes. One strategy entailed the unique approach of hiring a "parent in research" to lead the development and dissemination of the priorities. The Birnie et al. (2022b) and Ristevski et al. (2022b) projects illustrate that identifying priorities alone is insufficient – a multi-pronged approach is needed to ensure that these priorities result in further research and actual system change.

Equity in Patient Partnerships

The integration of equity and diversity considerations in patient partnership is essential for ensuring maximal impact of health research. Patient partnership, however, has been challenged as a result of the COVID-19 pandemic, along with exacerbation of social inequities in health (Sayani et al. 2021). While equity is a recurring theme in this special issue, the Birnie et al. (2022a) and Vanderhout et al. (2022) projects emphasize inclusivity and equity for patient engagement and research impacts in response to the pandemic.

One of the major impacts of COVID-19 restrictions in healthcare was the transition from in-person visits to virtual care. Recognizing that certain marginalized communities (e.g., People of Colour, Indigenous Peoples) were excluded from pain services and research, combined with the potential of virtual care to exacerbate health inequities, Birnie et al. (2022a) engaged with a diverse group of youths living with chronic pain and their families to develop best practice recommendations for virtual care. While the project identified opportunities for virtual care to reduce stigma, bias and discrimination, it also described key gaps, including lack of reliable Internet access and challenges associated with virtual delivery environments (e.g., access to private settings for visits). Key messages for patient-oriented care included the need for joint decisions with families about whether appointments should be virtual or in person.

The pandemic also had direct impacts on children and their families. The COVID-19 Study of Children and Families sought to better understand these impacts, building on the well-established TARGet Kids! Primary care research network. Vanderhout et al. (2022) describe the parent engagement in the development of the study, including co-developing documents and attending meetings as grant co-applicants. While parent engagement was described as successful overall, key shortcomings included difficulties recruiting fathers and parents who represent the ethnic and sociodemographic diversity of the target population.

Identifying priorities provides a set of specific, concrete tasks to achieve over time.

Co-Designing Interventions and Tools

With an emphasis on mobilizing knowledge from their existing patient-oriented studies, several projects engaged patient partners in co-designing interventions and tools to advance patient-oriented care. Collectively, these teams sought to promote POR in intervention studies, advance equity and demonstrate important returns achieved through investments in patient partnerships.

Parry et al. (2022), in partnership with Clinical Trials Ontario, aimed to widen the effects and reach of digital POR decision aids for patient partners and researchers. These capacity-building tools were created to support learning about POR, set priorities, expand understanding and evaluation of patient engagement in clinical trials, assess readiness and make decisions. The team engaged wide stakeholder audiences to mobilize these tools to end-users across Ontario. Parry et al. (2022) demonstrate the application of relevant frameworks to POR, patient-partner leadership roles in multi-faceted dissemination strategies and advocacy and offer insights into benefits and challenges associated with partner engagement in longer-term projects.

Similarly, Kuluski et al. (2022) built on their early engagement work to co-design an intervention to address delayed care transitions and improve care quality. The team co-designed a communication guide to engage and support patients and caregivers in transitional care communication, decisions and plans. Through meaningful partnerships, this team amplified patient and caregiver voices and advanced advocacy roles, by engaging their patient and caregiver council in creative knowledge mobilization that centralized lived experience perspectives.

The *MyDiabetesPlan* by Pan et al. (2022) aimed to evaluate the implementation and impacts of patient engagement in a large-scale innovative digital health trial. Impacts were explored from diverse stakeholder perspectives qualitatively. Their engagement evaluation showcases meaningful engagement and the mechanisms to achieve this. Notably, the team details the impacts of engaging with the *MyDiabetesPlan* tool and ensuring alignment with the specific needs of the target population. Pan et al. (2022) also offer critical insights into contextual factors that enabled and hindered engagement impacts.

Tools for Patient Engagement

Beyond co-designing interventions, the next theme focuses on patient-led projects that developed tools for patient engagement. Naylor et al. (2022) sought to increase access to kidney transplantation and living kidney donation; together, they developed the Transplant Ambassador Program (TAP), a volunteer patient-led program where patients with kidney disease are connected with someone who received a kidney transplant or who donated a kidney. Unlike busy healthcare professionals, these volunteers can dedicate more time, offer personal insights and provide inspiration to patients with kidney disease. TAP is part of a larger research network and trial, providing an example of patients informing important research questions and initiatives.

Identifying priorities and launching studies is by itself insufficient if research participants are not diverse or reflective of the population impacted. To address these concerns and building on the top 10 priorities in retinoblastoma research, Ristevski et al. (2022a) sought to catalyze patient involvement in research by developing a *Retinoblastoma Research and You!* booklet. The project, led by patients, used a human-centred design methodology to co-create a patient engagement booklet with healthcare professionals and researchers. The booklet helps explain what is involved and the benefits of partnering with research and has accelerated peer-to-peer recruitment efforts, particularly during the pandemic.

Patient engagement can be particularly fraught when it relates to marginalized communities; one such population is people with intellectual and developmental disabilities (IDD). Jiwa et al. (2022) illustrate the value of tools that can introduce patients to their providers and help patients prepare and document what happens during a visit. But more importantly, the project illustrated the importance of including patients with IDD, not just their family or workers, despite challenges such as those in comprehending verbal and written communication. For example, one patient advisor interacted with 60 physicians at a conference, sharing his experience and perspective with each individual, which ultimately challenged stereotypes.

As the science of patient engagement advances in Canada, researchers are challenged to push boundaries to integrate patients into innovative study designs and stages of research that have less frequently engaged patient partners in their design and implementation. Fox et al. (2022) offer one such example as they describe the powerful roles of patient partners in the development of selected tools to support early-phase clinical trials. The article highlights an innovative visual consent tool and non-technical summary, as well as an online peer-support model for trial participants. Through patient partnerships, the team gained critical insight into considerations that would not otherwise have been identified.

Patient- or Community-Driven Projects

Moving along the spectrum of patient participation to empowerment, the future of research is one in which research-based decision making is in the hands of patients and patient- or community-led projects. Four of the EMPOWER team articles showcase projects that were community- or patient-driven from the outset.

Tingley et al. (2022) – building on work that led to the recent development of core outcome sets for two rare inherited metabolic diseases (phenylketonuria and medium-chain acyl-CoA dehydrogenase deficiency) (Pugliese et al. 2021) – recognized the importance of identifying barriers and facilitators to implementing these outcome sets in future research. Two patient/family partners led the patient engagement strategy, which identified novel insights to guide future research (e.g., the importance of consent and privacy). Similarly, Syan et al. (2022) describe youth-led development and dissemination of the Youth Wellness Quest resource, which helps youth navigate mental health services. The youth-led team recognized the importance of developing an online version of the Youth Wellness Quest and led the grant application and subsequent implementation to develop a resource that is freely accessible (https://moodle8.camhx.ca/moodle/course/view.php?id=20).

Mulder et al. (2022) describe ongoing work related to the First Nations Aging Study funded by the Canadian Institutes of Health Research, which identified important health differences between First Nations people and non-First Nations people in Ontario. The authors set out to raise awareness about the key findings of the study, with key individuals working with First Nations people to better understand the impact of prior knowledge translation initiatives. They describe the importance of adding the principle of action to the traditional set of core principles of First Nations ownership, control, access and possession (OCAP®).

Finally, Sayani et al. (2022) outline the participatory co-design that led to the development of EMPaCT (Equity-Mobilizing Partnerships in Community), a community-designed patient partnership that aims to identify key areas for action in health equity for maximal impact. They provide unique insights about co-designing a scalable and sustainable model of equitable patient partnership. This team engaged in participatory co-design using an equity-oriented intersectional lens and trauma-informed approaches to engagement. This paper makes a unique contribution to POR by highlighting advancements in equitable patient partnerships, in particular, as they relate to sustainable and scalable models of patient engagement.

Conclusion

The diversity and range of projects supported by the EMPOWER Award illustrate the value of involving patients. They include examples of new partnerships formed to improve outcomes or innovative projects building off existing patient partnerships. The COVID-19 pandemic accentuated the critical inequities in our healthcare system and the communities disproportionately impacted by both SARS-COV-2 directly and the indirect impacts of lockdowns. Several EMPOWER Grant Award winners explicitly aimed to address health inequities in Ontario using a range of approaches, including stakeholder engagement with impacted communities.

While the style and structure of the articles vary, each project team outlines how the project started and how they sustained meaningful engagement and shares the experiences of both the research team and patients. Furthermore, they describe the overall value added through patient involvement and describe the evaluation of impacts, in particular, on patient partners, research, researchers and other key stakeholders. Teams also reflect upon real-world implementation challenges and how these challenges were mitigated.

What these projects have in common is the shared sense of purpose, where patients are placed at the centre. Ultimately, this special issue underscores that the key to making an impact in healthcare is valuable input from and uptake by end-users. These exemplary projects help to illustrate the "how to" for decision makers and other stakeholders so that they can catalyze research into impact and support the shift to meaningful engagement and involvement. HQ

References

Banner, D., M. Bains, S. Carroll, D.K. Kandola, D.E. Rolfe, C. Wong et al. 2019. Patient and Public Engagement in Integrated Knowledge Translation Research: Are We There Yet? *Research Involvement and Engagement* 5: 8. doi:10.1186/s40900-019-0139-1.

Birnie, K.A., K. Dib, C. Ouellette, M.A. Dib, K. Nelson, D. Pahtayken et al. 2019. Partnering for Pain: A Priority Setting Partnership to Identify Patient-Oriented Research Priorities for Pediatric Chronic Pain in Canada. *CMAJ Open* 7(4): E654–64. doi:10.9778/cmajo.20190060.

Birnie, K.A., T. Killackey, G. Backlin, F. Gavin, C. Harris, I. Jordan et al. 2022a. Ensuring Equity and Inclusion in Virtual Care Best Practices for Diverse Populations of Youth with Chronic Pain. *Healthcare Quarterly* 24(Special Issue): 25–30. doi:10.12927/hcq.2022.26778.

Birnie, K.A., C. Ouellette, J. Marianayagam, F. Campbell, C. Lamontagne, P. Forgeron et al. 2022b. Partnering with Youth and Parents for the Greatest Impact of Top Patient-Oriented Priorities in Pediatric Chronic Pain Research, Care and Policy. *Healthcare Quarterly* 24(Special Issue): 18–23. doi:10.12927/hcq.2022.26779.

Canadian Institutes of Health Research (CIHR). 2015, March. Guide to Knowledge Translation Planning at CIHR: Integrated and End-of-Grant Approaches. Retrieved October 30, 2021. <https://cihr-irsc.gc.ca/e/45321.html#f1>.

Canadian Institutes of Health Research (CIHR). 2016, July. About Us – Knowledge Translation. Retrieved November 30, 2021. <https://cihr-irsc.gc.ca/e/29418.html>.

Canadian Institutes of Health Research (CIHR). 2019, May. Strategy for Patient-Oriented Research – Patient Engagement Framework. Retrieved August 20, 2021. <https://cihr-irsc.gc.ca/e/48413.html>.

Canadian Institutes of Health Research (CIHR). 2021, February. *CIHR Strategic Plan 2021–2031*. Retrieved November 30, 2021. <https://cihr-irsc.gc.ca/e/documents/cihr-strategic-plan-2021-2031-en.pdf>.

Flegg, K., M.J. Gelkopf, S.A. Johnson, H. Dimaras; for the Canadian Retinoblastoma Research Advisory Board Priority Setting Steering Committee. 2020. The Top 10 Retinoblastoma Research Priorities in Canada as Determined by Patients, Clinicians and Researchers: A Patient-Oriented Priority-Setting Partnership. *CMAJ Open* 8(2): E420–28. doi:10.9778/cmajo.20190221.

Fox, G., D.A. Fergusson, M. Foster, T. Hawrysh, S. Dupont, D.J. Walling et al. 2022. Building a Platform for Meaningful Patient Partnership to Accelerate "Bench-to-Bedside" Translation of Promising New Therapies. *Healthcare Quarterly* 24(Special Issue): 74–79. doi:10.12927/hcq.2022.26770.

Gill, P.J. and E. Cartwright. 2021. Partnering with Patients in the Production of Evidence. *BMJ Evidence Based Medicine* 26(3): 73–76. doi:10.1136/bmjebm-2020-111339.

Jiwa, M.I., J. Durbin, V. Pereira, E. Streisslberger, L. Steel and Y. Lunsky. 2022. "This Is about My Health": Partnering with Patients and Families to Share Knowledge and Tools about Health Care Communication for Adults with Intellectual and Developmental Disabilities. *Healthcare Quarterly* 24(Special Issue): 98–104. doi:10.12927/hcq.2022.26766.

Kuluski, K., I. McLaughlin, L. Bennett, G. MacGregor, L. Bilotta, B. Farrell et al. 2022. In Pursuit of Better Care Transitions: Lessons Learned from a Co-Designed Project. *Healthcare Quarterly* 24(Special Issue): 49–54. doi:10.12927/hcq.2022.26774.

Manafò, E., L. Petermann, V. Vandall-Walker and P. Mason-Lai. 2018. Patient and Public Engagement in Priority Setting: A Systematic Rapid Review of the Literature. *PLoS ONE* 13(3): e0193579. doi:10.1371/journal.pone.0193579.

Mulder, C., D. Debassige, M. Gustafson, M. Slater, E. Eshkawkogan and J.D. Walker. 2022. Research, Sovereignty and Action: Lessons from a First Nations–Led Study on Aging in Ontario. *Healthcare Quarterly* 24(Special Issue): 93–97. doi:10.12927/hcq.2022.26767.

Naylor, K.L., S.Q. McKenzie, A.X. Garg, S. Yohanna and J.M. Sontrop. 2022. Partnering with Patients to Enhance Access to Kidney Transplantation and Living Kidney Donation. *Healthcare Quarterly* 24(Special Issue): 69–73. doi:10.12927/hcq.2022.26771.

Nygaard, A., L. Halvorsrud, S. Linnerud, E.K. Grov and A. Bergland. 2019. The James Lind Alliance Process Approach: Scoping Review. *BMJ Open* 9(8): e027473. doi:10.1136/bmjopen-2018-027473.

Ontario SPOR SUPPORT Unit (OSSU). 2021. OSSU EMPOWER Awards. Retrieved November 30, 2021. <https://ossu.ca/for-researchers/work-we-support/ossu-empower-awards/>.

Pan, R.Y., K. Zhang, A. Sivakumar, D. Choi, A.H. Wang, P. Wijeyesekera et al. 2022. Evaluation of Experiences and Impact of Patient Engagement on e-Health Research: A Qualitative Study. *Healthcare Quarterly* 24(X): 44–48. doi:10.12927/hcq.2022.26775.

Parry, M., D.P. Richards, D. Wells, A. Najam, S. Hemani and S. Marlin. 2022. Building Capacity for Patient-Oriented Research: Utilizing Decision Aids to Translate Evidence into Practice, Policy and Outcomes. *Healthcare Quarterly* 24(Special Issue): 37–43. doi:10.12927/hcq.2022.26776.

Pugliese, M., K. Tingley, A. Chow, N. Pallone, M. Smith, P. Chakraborty et al. 2021. Core Outcome Sets for Medium-Chain Acyl-CoA Dehydrogenase Deficiency and Phenylketonuria. *Pediatrics* 148(2): e2020037747. doi:10.1542/peds.2020-037747.

Ristevski, I., J. Kiew, M. Hendry, M. Prunier, R. Noronha, M. Al-Hammadi et al. 2022a. The Retinoblastoma Research Booklet: A Catalyst for Patient Involvement in Retinoblastoma Research. *Healthcare Quarterly* 24(Special Issue): 61–68. doi:10.12927/hcq.2022.26772.

Ristevski, I., J. Robert, R. Baddeliyanage, R. Noronha, M. Gelkopf, K. Flegg et al. 2022b. EMPOWER Retinoblastoma: Engaging Patient Partners in Solving the Top 10 Priorities for Eye Cancer Research in Canada. *Healthcare Quarterly* 24(Special Issue): 11–17. doi:10.12927/hcq.2022.26780.

Sayani, A., A. Maybee, J. Manthorne, E. Nicholson, G. Bloch, J.A. Parsons et al. 2021. Building Equitable Patient Partnerships during the COVID-19 Pandemic: Challenges and Key Considerations for Research and Policy. *Healthcare Policy* 17(1): 17–24. doi:10.12927/hcpol.2021.26582.

Sayani, A., A. Maybee, J. Manthorne, E. Nicholson, G. Bloch, J.A. Parsons et al. 2022. Equity-Mobilizing Partnerships in Community (EMPaCT): Co-Designing Patient Engagement to Promote Health Equity. *Healthcare Quarterly* 24(Special Issue): 86–92. doi:10.12927/hcq.2022.26768.

Syan, A., J.Y. Lam, L.D. Hawke, K. Darnay and J. Henderson. 2022. The Youth Wellness Quest: A Comprehensive Online Mental Health Literacy and Self-Advocacy Resource Developed by Youth for Youth. *Healthcare Quarterly* 24(Special Issue): 55–59. doi:10.12927/hcq.2022.26773.

Tingley, K., M. Smith, N. Pallone, P. Chakraborty and B.K. Potter. 2022. Patient Engagement in a Multi-Stakeholder Workshop to Plan the Collection of Patient-Oriented Outcomes for Children with Inherited Metabolic Diseases. *Healthcare Quarterly* 24(Special Issue): 81–85. doi:10.12927/hcq.2022.26769.

Vanderhout, S.M., C.S. Birken, P.D. Wong, S. Weir, J. Batten and J.L. Maguire. 2022. Parent Engagement in a COVID-19 Cohort Study of Children and Families: Successes, Challenges and Next Steps. *Healthcare Quarterly* 24(Special Issue): 31–34. doi:10.12927/hcq.2022.26777.

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> “The greatest impact of patient involvement on the project was the symbiotic relationship that developed among patients, researchers and health professionals.”
>
> *– p. 15*

EMPOWER Retinoblastoma: Engaging Patient Partners in Solving the Top 10 Priorities for Eye Cancer Research in Canada

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Abstract

While it is recognized that research priorities should reflect and integrate the perspectives and needs of patients along with those of health professionals and researchers, it remains challenging to actualize such priorities into tangible research projects. Targeted dissemination is required to catalyze research on these priorities. To create awareness of and inspire action toward actualizing the top 10 retinoblastoma research priorities in Canada, Canadian Retinoblastoma Research Advisory Board (CRRAB) members developed a wide range of dissemination tools and processes. These resources, co-produced with patients, were instrumental to CRRAB sharing the top 10 priorities internationally to mobilize action toward solving them.

Key Points

- The Canadian Retinoblastoma Research Advisory Board (CRRAB) serves to facilitate involvement of patients in all aspects of retinoblastoma research. One of the first initiatives of CRRAB was to jointly determine the top 10 retinoblastoma research priorities in Canada.
- To increase awareness of and inspire action toward solving the top 10 retinoblastoma research priorities, CRRAB members developed and implemented a wide range of dissemination tools and processes.
- The dissemination tools were used to reach key researchers, as well as members of the public, and they continue to be utilized through the CRRAB website.

Introduction

Retinoblastoma is a rare cancer of the retina, diagnosed in 25 children per year in Canada (Dimaras et al. 2015). Children with retinoblastoma develop tumours in one or both eyes, and about half carry a gene variant that causes retinoblastoma and results in increased risk of a second cancer, as well as the chance of passing it on to their offspring (Dimaras et al. 2015). While 96% of Canadian patients survive (Selvarajah et al. 2021), survivors live with consequences of the tumours and/or treatment, such as low or no vision in one or both eyes, cosmetic effects or other disruptions such as frequent lifelong follow up.

Given the lifelong disease implications, there is a recognized need to involve patients in retinoblastoma clinical care and research. The Canadian Retinoblastoma Patient Engagement Strategy (https://lab.research.sickkids.ca/dimaras/research/engagement/strategy/) was created to meet this need, involving patients (defined as those with lived experience of retinoblastoma and caregivers) in all aspects of retinoblastoma research. It aims to (1) share research results with those affected by retinoblastoma, (2) include a diverse group of people in retinoblastoma research and (3) promote research

P = Patient with lived experience of retinoblastoma. R = Researcher with expertise in retinoblastoma. H = Health professional with expertise in retinoblastoma.

created and led by individuals affected by retinoblastoma. The Canadian Retinoblastoma Research Advisory Board (CRRAB) was created to govern and sustain the Patient Engagement Strategy (White et al. 2019). Aligned with the Canadian Strategy for Patient-Oriented Research (SPOR) (CIHR 2014), CRRAB is a multidisciplinary group of retinoblastoma patients, researchers and health professionals that aims to shift the focus from research created "about" patients to "with" patients. This approach has many potential benefits – for example, evaluation of studies that involved patients as partners indicates that study findings are often more applicable, credible and transparent, which is beneficial with regard to patient autonomy, dignity and self-worth (Boyko 2015; Kovacs Burns et al. 2014). In a formal evaluation of CRRAB, members perceived that they similarly achieved a meaningful impact on retinoblastoma research and improved accessibility to evidence-based retinoblastoma practices (Gelkopf et al. 2020).

Research priority settings benefit from patient engagement as they allow for the identification of research questions that are considered most relevant and valued by those that the research is intended to benefit. Historically, funded research has not aligned with the priorities of patients and health professionals, reducing its impact (Crowe et al. 2015; Tallon et al. 2000). The first project led by CRRAB was to determine the top 10 retinoblastoma research priorities in Canada (hereafter referred to as "top 10 priorities") (Flegg et al. 2020).

However, having a list of priorities is not enough, and targeted dissemination methods are required to raise awareness and catalyze research (Lavis et al. 2003). Furthermore, there is a lag, often years, between conducting research and its translation to clinical applications (Morris et al. 2011). To overcome this lag, the field of knowledge translation has identified ways to improve dissemination and uptake (Azimi et al. 2015). A major focus of CRRAB is to harness its network to meaningfully disseminate research findings to key stakeholders. Key factors for effective dissemination include formulating a written plan, involving patients in the process and using a combination of traditional and innovative dissemination tools (Schipper et al. 2016).

With the above-mentioned points in mind, our team began to brainstorm and co-create dissemination tools and draft a dissemination strategy for the top 10 priorities. We applied to and were granted an Ontario SPOR SUPPORT Unit Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach award to support these efforts. Thus, the overall objective of this project was to develop and implement a dissemination plan to create awareness of and inspire action toward actualizing the top 10 priorities.

Patient Engagement Methods and Outputs

Leadership and operations

While an independent entity spanning all of Canada, CRRAB is directed from within the Retinoblastoma Program of the Hospital for Sick Children (SickKids). A novel research role was created within the SickKids Retinoblastoma Program for an individual with lived experience of retinoblastoma. Called a "parent in research," they incorporate their experiences as a parent of a retinoblastoma survivor into research and serve as the co-leader of CRRAB and its activities. In general, the parent in research works within the research team to coordinate and advise on various research projects and CRRAB activities and serves as a liaison between the health research and patient communities. As a reliable point person within CRRAB and within the leading retinoblastoma research institution in Canada, their role helps sustain patient engagement in research.

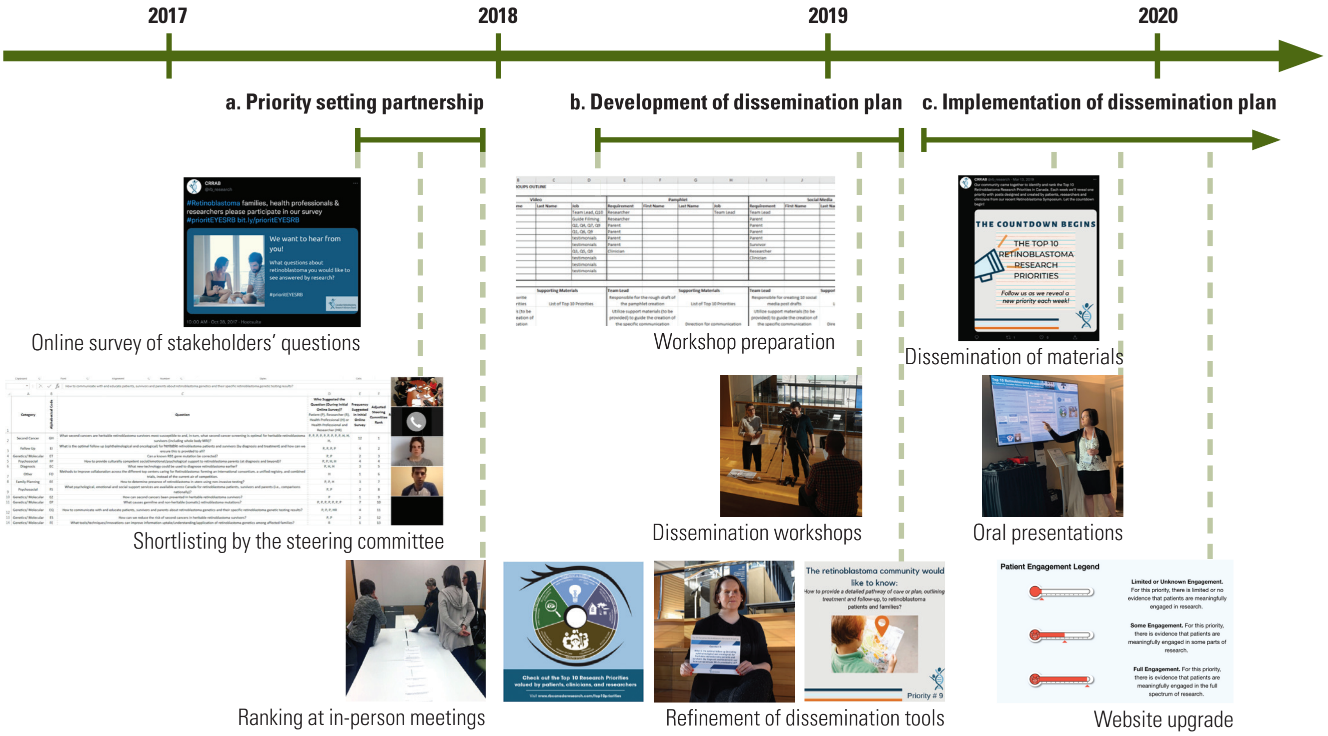
At its inception, the CRRAB structure was composed of a steering committee and three working groups, each led by a patient and non-patient co-chair. CRRAB members attended an annual general meeting to set goals and evaluate annual progress. Throughout the year, the working groups met online every four to six weeks. The priority setting working group was responsible for supervising the priority setting activity that developed the top 10 priorities in 2017 (Figure 1a) (Flegg et al. 2020). The parent in research led the dissemination of the top 10 priorities and coordinated communications and cooperation among patient, researcher and health professional stakeholders. The consistent nature of CRRAB meetings, the schedule of which is decided by member consensus, established a regular routine that supported sustained stakeholder involvement in this and other projects.

Recruitment and composition of the dissemination team

Attendees of the 2019 CRRAB annual general meeting were invited to participate in a workshop focused on developing dissemination plans and materials for the top 10 priorities (Figure 1b). The workshop was subdivided into three breakout groups focused on distinct dissemination methods. In advance of the meeting, participants indicated their preference for breakout groups based on individual interest and skill set. The parent in research facilitated recruitment by identifying and inviting potential workshop leads and matching participant interests and prior CRRAB experience to the dissemination goals.

For each breakout group, we aimed to include a diverse sample of patients, researchers and health professionals. In the patient community, indicators of diversity included relationship

FIGURE 1.
Timeline of priority setting partnership and dissemination plan



(a) The priority setting partnership established the top 10 retinoblastoma research priorities in Canada.
(b) The dissemination plan was developed and then finalized in a workshop, resulting in the creation of (i) an educational pamphlet, (ii) video testimonials and (iii) social media content to publicize the top 10 priorities.
(c) The dissemination materials were implemented in a targeted dissemination campaign to raise awareness and inspire action among patients, researchers and funders.

to retinoblastoma (e.g., parent, survivor, etc.), diagnosis (e.g., unilateral, bilateral, heritable, non-heritable, etc.), time since diagnosis and the caregiver's role (e.g., mother, father, grandparent, etc.). For health professionals, these included clinical roles (e.g., physician, social worker, nurse, etc.) and for researchers, field of study (e.g., basic science, clinical trials, etc.). Final groups were composed of eight to nine participants with an approximate patient to non-patient ratio of 1:1.

Workshop

The parent in research (IR) and lead scientist (HD) formulated a dissemination plan and designed a workshop with breakout sessions (Figure 1b) modelled on the concept of human-centred design (Hasso Plattner Institute of Design at Stanford 2010). Each breakout session had a patient and non-patient facilitator to provide support and ensure deliverables were met. Following the in-person workshop, online meetings were held to develop and refine dissemination tools and processes.

First workshop breakout group: Development of an infographic and educational pamphlet

This workshop breakout session was dedicated to the development of an infographic and educational pamphlet. Participants first discussed methods, imagery and key words that could concisely highlight each of the top 10 priorities in an easily accessible way. While the group brainstormed ideas, two patient CRRAB members with professional experience in visual communication, web design and media arts drafted preliminary sketches of an infographic. The resulting infographic summarized the top 10 priorities and grouped them by common thematic areas to present the information in a way that was easily understood and visually appealing. With the draft infographic as the central focus, participants jointly developed additional messages to draft a complete one-page educational pamphlet.

Following the preliminary work completed in the breakout session workshop, the parent in research worked with a graphic designer to complete the educational pamphlet.

The final pamphlet included the co-designed infographic and key messages on how to learn more about the top 10 priorities and how to connect to CRRAB. The pamphlet was optimized for digital distribution and included in dissemination activities.

Second workshop breakout group: Video testimonials

This breakout group was focused on developing short videos to raise awareness and provide context about the top 10 priorities. Ahead of the workshop, participants identified the top three priorities for which they would be willing to share a personal story. Based on the selected priorities, the parent in research assigned one or two priorities to each participant based on their selections. Prior to the breakout session, participants were asked to write a brief script, a one-sentence summary and key words pertaining to their assigned priorities.

During the workshop, the session leads were responsible for providing support through idea generation and script refinement. All participants collaborated and engaged in discussion during the session to further inform the content of the videos. During the session, video footage was captured by student volunteers, and in the end, one video for each of the top 10 priorities was created. The content of the videos depicted a CRRAB member voicing their unique perspective. Patients shared their lived experience and perspectives on the importance of addressing their assigned priority. Health professionals shared their clinical expertise working with retinoblastoma patients and how addressing their priority could impact health outcomes, while researchers shared their scientific knowledge and the significance of their priority. The videos were edited by a lab volunteer and were disseminated through social media avenues and the CRRAB website. One video outlining all of the top 10 priorities was created by the SickKids' creative services team.

Third workshop breakout group: Social media campaign

This group developed social media content and a dissemination plan to reveal the top 10 priorities to the public. A brainstorming session helped develop clear messaging, a unifying colour scheme and appealing imagery for posts. Each priority was represented by two social media posts: one depicting a CRRAB member "championing" the priority and another entailing a graphical representation of the priority. CRRAB social media volunteers helped create the posts using Canva, an online graphic design software. The posts were shared on CRRAB social media platforms (i.e., Instagram, Facebook and Twitter) in a reverse countdown fashion to stimulate audience engagement and interest in the content.

Website development

Situational analysis and e-mail campaign

A situational analysis was conducted to uncover any current progress toward solving the top 10 priorities and identify researchers whose interests may align with them. The analysis identified 61 ongoing and completed projects. To promote the actualization of the priorities, targeted communications were sent to the identified researchers alerting them of the top 10 priorities and offering support in incorporating patient engagement methods in their work.

Website design and content

The CRRAB website was upgraded with a new design and domain name (www.rbcanadaresearch.com) and was updated to publicize the top 10 priorities (Figure 1c). The upgrade was led by a patient CRRAB member, a software developer, working in collaboration with the parent in research to gather feedback from monthly CRRAB working group meetings. CRRAB members suggested that the website follow the design and content of the infographic. A landing page featuring the infographic summarized all the priorities, and separate webpages were developed for each priority to add further detail, feature the video testimonials and highlight the ongoing research projects related to each priority that were uncovered in the situational analysis.

Additionally, a patient engagement "thermometer" was developed to visually depict the degree to which patients are involved in actualizing the top 10 priorities. The situational analysis determined if there was full, some or limited/unknown patient engagement (Figure 2).

Presentation at an international conference

The parent in research (IR) and the scientific lead (HD) attended the 2019 International Society for Genetic Eye Disease and Retinoblastoma Congress (Ristevski et al. 2019), where several retinoblastoma researchers, patient advocacy groups and funders were in attendance. The parent in research presented a poster about the top 10 priorities, distributed the educational pamphlet and engaged with relevant individuals to discuss how their work can help actualize the priorities.

Challenges and Mitigating Strategies

Engaging a diverse group of stakeholders comes with its own challenges. Each person comes to the table with different interests and ways they want to be meaningfully engaged. In the process, our patient partners indicated that they were more willing to contribute to tasks where they could use their professional skills or interests. To mitigate such challenges, it was important to be flexible and enable the participants to self-select their level and area of participation.

FIGURE 2.
Patient engagement in the top 10 priorities

Top 10 priorities	Full engagement	Some engagement	Limited or unknown engagement
Priority 1: Early Diagnosis		✓	
Priority 2: Second Cancer Screening			✓
Priority 3: Psychosocial Support		✓	
Priority 4: Follow up & Follow Through			✓
Priority 5: New Treatments			✓
Priority 6: Life with Vision Loss			✓
Priority 7: Second Cancer Prevention			✓
Priority 8: Improved Collaboration		✓	
Priority 9: Pathway of Care			✓
Priority 10: Access to Care		✓	

A situational analysis was performed to identify related research, scientists and the degree of patient engagement involved in each. The level of patient engagement was classified as "limited or unknown engagement" (i.e., limited or no evidence that patients are meaningfully engaged in research), "some engagement" (i.e., evidence that patients are meaningfully engaged in some parts of research) or "full engagement" (i.e., evidence that patients are meaningfully engaged in the full spectrum of the research).

Furthermore, many participants with full-time careers and busy family lives found it difficult to consistently attend meetings. Also, including patients and families from across Canada created challenges to find common meeting times across the different time zones. To mitigate this, the parent in research was able to reach out to members to provide them alternative means of participating – for example, by reviewing items on their own time and having the parent in research relay feedback at the regular meetings.

Finally, owing to the nature of retinoblastoma, some of our patient partners experience and live with significant visual impairment or blindness. We have adopted practices to facilitate engagement of this group, such as ensuring that electronic documents are screen reader–friendly or visual aids are verbally described and meeting materials are sent well in advance to facilitate advance preparation.

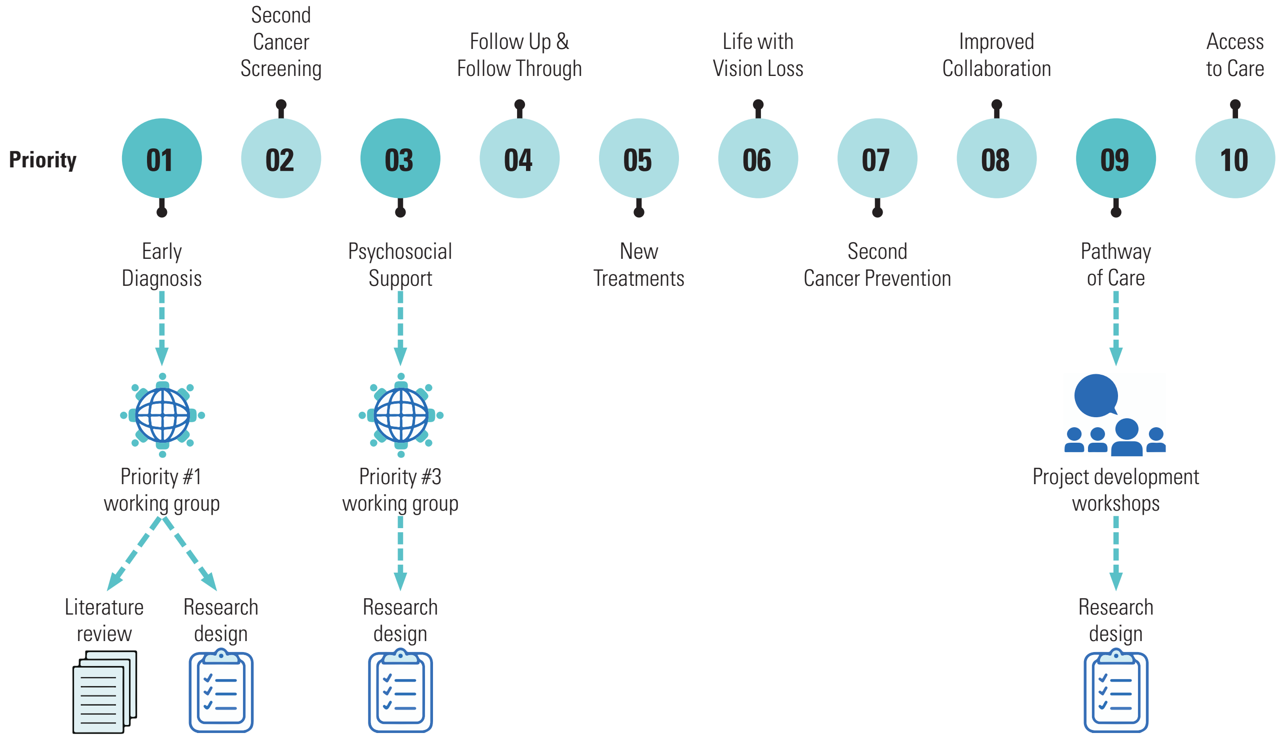
Discussion

The creation of a wide range of dissemination materials resulted in a broad reach and international awareness of the top 10 priorities. These activities generated interest among researchers and health professionals who were inspired to conduct patient-partnered research to solve one of the top 10 priorities. Since the implementation of this project, three working groups have launched to focus on priorities 1, 3 and 9. Each working group comprises patients, researchers and health professionals, both Canadian and international, who collaborate in design research, apply for funding and implement studies (Figure 3).

The top 10 priorities were successfully identified through collaboration and consensus among patients, health professionals and researchers. It was imperative to continue that approach to develop their dissemination strategy. All of the activities described actively involved key stakeholders from the retinoblastoma community generating meaningful and innovative materials to raise awareness of and inspire action toward solving the top 10 priorities.

The greatest impact of patient involvement on the project was the symbiotic relationship that developed among patients, researchers and health professionals. Each was required to learn and develop a more holistic understanding of retinoblastoma research. Through this work, a sense of community and connectedness emerged and continues to grow, creating an interconnected, pan-Canadian retinoblastoma research community inclusive of patients, survivors and their families. There is a shared desire and ownership to find evidence-based answers and solutions to the research priorities so that the care and outcomes of those affected by retinoblastoma continue to improve.

FIGURE 3.
Development of patient-partnered research projects



Three working groups have emerged focused on three priorities (Priority 1: Early Diagnosis, Priority 3: Psychosocial Care and Priority 9: Pathway of Care). Working groups meet on an *ad hoc* basis to co-design research, apply for funding and implement studies.

Specifically, our patient partners indicated that participating in the research workshops offered a chance to apply their lived experience to make the cancer journey easier for future patients. It also offered the unique opportunity to build more personal connections with researchers and health professionals; for some, this made interactions with clinical teams at subsequent follow-up visits easier simply by knowing that their lived experiences had been heard and understood.

Overall, it took longer than we had hoped to finalize the dissemination products, but, in the end, they were more robust and captivating as a result of the multi-stakeholder approach. The parent in research role, in partnership with the research team, was, and continues to be, a critical enabler to coordinate and spur momentum on the dissemination strategy and its outputs by continually developing and building relationships with patients and families, steadfastly creating multiple avenues for patient partners to provide their feedback and input.

The dissemination strategy is a continued priority of CRRAB, and sustaining momentum and specific patient partnerships requires ongoing, dedicated effort. Continual recruitment and engagement is critical as the availability of existing patient partners to engage in the work ebbs and flows. Practical considerations include the ongoing maintenance and updates of the website and ongoing nurturing of the new research groups to ensure robust patient participation and involvement in the research process. The social media channels continue to share dissemination materials periodically, to remind the community about the top 10 priorities. CRRAB meetings remain the medium via which the top 10 priorities are consistently reviewed and working groups are encouraged to form around yet unaddressed priorities.

CRRAB was able to successfully share the results of the priority setting exercise on the international stage, leveraging the dissemination tools and processes that were developed by and for patients, researchers and health professionals. Action toward solving the top 10 priorities has begun as a result, and a Canada-wide retinoblastoma research community continues to grow.

References

Azimi, A., R. Fattahi and M. Asadi-Lari. 2015. Knowledge Translation Status and Barriers. *Journal of the Medical Library Association* 103(2): 96–99. doi:10.3163/1536-5050.103.2.008.

Boyko, J.A. 2015. Evidence-Informed Health Policy Making in Canada: Past, Present, and Future. *Journal of Evidence-Based Medicine* 8(4): 215–21. doi:10.1111/jebm.12169.

Canadian Institutes for Health Research (CIHR). 2014. *Strategy for Patient-Oriented Research: Patient Engagement Framework*. Retrieved September 9, 2021. <https://cihr-irsc.gc.ca/e/documents/spor_framework-en.pdf>.

Crowe, S., M. Fenton, M. Hall, K. Cowan and I. Chalmers. 2015. Patients', Clinicians' and the Research Communities' Priorities for Treatment Research: There is an Important Mismatch. *Research Involvement and Engagement* 1: 2. doi:10.1186/s40900-015-0003-x.

Dimaras, H., T.W. Corson, D. Cobrinik, A. White, J. Zhao, F.L. Munier. 2015. Retinoblastoma. *Nature Reviews Disease Primers* 1: 15021. doi:10.1038/nrdp.2015.21.

Flegg, K., M.J. Gelkopf, S.A. Johnson and H. Dimaras. 2020. The Top 10 Retinoblastoma Research Priorities in Canada as Determined by Patients, Clinicians and Researchers: A Patient-Oriented Priority-Setting Partnership. *CMAJ Open* 8(2): E420–28. doi:10.9778/cmajo.20190221.

Gelkopf, M. J., I. Avramov, R. Baddeliyanage, I. Ristevski, S.A. Johnson, K. Flegg and H. Dimaras. 2020. The Canadian Retinoblastoma Research Advisory Board: A Framework for Patient Engagement. *Research Involvement and Engagement* 6(1): 7. doi:10.1186/s40900-020-0177-8.

Hasso Plattner Institute of Design at Stanford. 2010. *An Introduction to Design Thinking: Process Guide*. Retrieved September 9, 2021. <https://web.stanford.edu/~mshanks/MichaelShanks/Files/509554.pdf>.

Kovacs Burns, K., M. Bellows, C. Eigenseher and J. Gallivan. 2014. 'Practical' Resources to Support Patient and Family Engagement in Healthcare Decisions: A Scoping Review. *BMC Health Service Research* 14: 175. doi:10.1186/1472-6963-14-175.

Lavis, J.N., D. Robertson, J.M. Woodside, C.B. McLeod, J. Abelson and Knowledge Transfer Study Group. 2003. How Can Research Organizations More Effectively Transfer Research Knowledge to Decision Makers? *The Milbank Quarterly* 81(2): 221–48. doi:10.1111/1468-0009.t01-1-00052.

Morris, Z.S., S. Wooding and J. Grant. 2011. The Answer Is 17 Years, What Is the Question: Understanding Time Lags in Translational Research. *Journal of the Royal Society of Medicine* 104(12): 510–20. doi:10.1258/jrsm.2011.110180.

Ristevski, I., K. Flegg, M. Gelkopf, S. Johnson and H. Dimaras. 2019. The Top 10 Retinoblastoma Research Priorities in Canada as Determined by Patients, Clinicians and Researchers. In "2019 ISGEDR Meeting – 29th to 31st August". Retrieved March 2, 2022. <https://isgedr.com/giessen-2019/>.

Schipper, K., M. Bakker, M. De Wit, J.C.F. Ket and T.A. Abma. 2016. Strategies for Disseminating Recommendations or Guidelines to Patients: A Systematic Review. *Implementation Science* 11(1): 82. doi:10.1186/s13012-016-0447-x.

Selvarajah, A., K. Flegg, W. Sim, J.B. Hu, B.L. Gallie, F. Shaikh et al. 2021. Clinical Audit of Retinoblastoma Management: A Retrospective Single-Institution Study. *Canadian Journal of Ophthalmology* S0008–4182(21): 00163-0. doi:10.1016/j.jcjo.2021.04.019.

Tallon, D., J. Chard and P. Dieppe. 2000. Relation between Agendas of the Research Community and the Research Consumer. *The Lancet* 355(9220): 2037–40. doi:10.1016/s0140-6736(00)02351-5.

White, E., R. Baddeliyanage, F. Shaikh and H. Dimaras. 2019. Meaningful Patient Engagement in Research: Lessons from Retinoblastoma. *Pediatrics* 143(6). doi:10.1542/peds.2018-2166.

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Partnering with Youth and Parents for the Greatest Impact of Top Patient-Oriented Priorities in Pediatric Chronic Pain Research, Care and Policy

Kathryn A. Birnie, Carley Ouellette^{P}, Justina Marianayagam^{P}, Fiona Campbell, Christine Lamontagne, Paula Forgeron, Jennifer Stinson and the Partnering For Pain Priority Setting Partnership Team

Abstract

Our original patient-oriented research project identified the top 10 priorities for pediatric chronic pain research and care in Canada from the perspective of people with lived experience (patients), their family members and healthcare professionals through a modified James Lind Alliance Priority Setting Partnership. We undertook subsequent knowledge translation activities with youth, families, healthcare professionals, decision makers and researchers to (1) generate awareness and interest in the top 10 priorities and our partnership process, (2) facilitate collaborative dialogue and open innovation and (3) integrate and adopt the top 10 priorities into stakeholder activities. This paper describes our knowledge translation activities, outcomes and impact.

Introduction

Chronic pain affects 11–38% of children and adolescents (King et al. 2011) or approximately one to three million youth in Canada, and costs C$40 billion annually (Health Canada 2021). Youth with chronic pain experience poor quality of life and impairment in physical, sleep, social, family- and school-related functioning (Eccleston et al. 2021; Forgeron et al. 2010; Groenewald et al. 2020; Lewandowski et al. 2010; Valrie et al. 2013). Pediatric pain researchers have identified priorities for future research (Caes et al. 2016; Liossi et al. 2017), but prior to our work, the patient and family voice was largely absent from guiding what research should be prioritized to improve chronic pain for youth in Canada and their families.

Our original patient-oriented research (POR) project sought to identify the top 10 priorities for pediatric chronic pain research and care in Canada from the perspective of people with lived experience (patients), family members and healthcare professionals (Birnie et al. 2018; Birnie et al. 2019). We modelled our methods to identify the top priorities after the internationally established James Lind Alliance Research Priority Setting Partnership (James Lind Alliance Priority Setting Partnerships 2018). The James Lind Alliance methodology is recognized for its equitable and rigorous approach to identify what matters most to people who need to use the research in their everyday lives (Manafò et al. 2018). We have previously published on our partnership (Birnie et al. 2018) and our national multi-stage engagement process to reach the final top 10 list, which was completed in December 2018 (Birnie et al. 2019).

Key Points

- Partnership with youth with chronic pain and parents led to a greater impact of knowledge translation activities.
- A focus on knowledge translation and patient partnership enabled our work to move beyond knowledge sharing to behaviour and policy change.
- Challenges in patient engagement and knowledge translation were overcome through inclusion of multiple youth and parent partners.

P = Patient partner.

BOX 1.
Top 10 patient-oriented research priorities in pediatric chronic pain in Canada

1	What treatments or strategies effectively prevent acute pain from becoming chronic in children and adolescents?
2	What is the impact of living with chronic pain on children's and adolescents' academic performance and educational attainment, and what strategies best support vocational planning for children and adolescents with chronic pain?
3	What physical and psychological treatments are effective for improving pain and function in children and adolescents with chronic pain (e.g., functional outcomes including quality of life, depression, fatigue, sleep, acceptance, concentration, resilience, coping and self management)?
4	What strategies improve access and delivery of evidence-based treatments, and coordination of care for all Canadian children and adolescents with chronic pain and their families with a view to reduce disparities?
5	What strategies effectively increase healthcare providers' training, knowledge, recognition, beliefs, attitudes and communication about the validity and risk of chronic pain in children and adolescents, and its evidence-based treatments?
6	What strategies effectively increase governmental and healthcare organizational financial support for evidence-based pediatric chronic pain care in Canada?
7	What strategies for educating school personnel about pediatric chronic pain effectively increase their awareness, understanding and recognition of the validity, impact and treatment of pediatric chronic pain?
8	What interventions are effective for managing acute pain flares in children and adolescents with chronic pain?
9	What is the interaction between chronic pain and mental health symptoms in children and adolescents, and when and how can co-occurring chronic pain and mental health symptoms be most effectively diagnosed and treated?
10	When are treatments for chronic pain in children and adolescents most effective (e.g., after medical investigation is complete, during variation by type of treatment modality or with the readiness of the child/adolescent or family to engage in treatment)?

Source: Birnie et al. 2019.

See Box 1. Since then, our national team of youth with lived experience, parents, healthcare professionals and researchers have been working in partnership to ensure these top 10 priorities are being mobilized for the greatest impact.

Patient Partnership

Our approach to patient engagement recognizes and values the expertise of lived and living experience of youth with chronic pain and their family members. Youth and parent partners were engaged equally as members of the project team (Birnie et al. 2018), including as members of the steering committee overseeing the Priority Setting Partnership (Birnie et al. 2019). They were also responsible for co-producing and mobilizing all knowledge translation activities. Youth and parent partners were identified through known relationships to project team members (e.g., former patients and family members), as well as through postings to patient engagement registries (e.g., Saskatchewan Centre for Patient-Oriented Research). Youth and parent partners were diverse in race, geography/rurality, type of chronic pain and experience of chronic pain care. Youth and parent partners identified important areas of representation to ensure that diverse participants from underrepresented groups engaged in the priority setting partnership (e.g., age, sex and gender, chronic pain experience, geography/rurality, family member relationship, family system, healthcare setting and discipline of healthcare professionals). Youth and parent partners were co-authors on resulting publications, co-facilitators and co-presenters. Youth partner experiences with the project (Birnie et al. 2018), as well as evaluation of our patient engagement process (Birnie et al. 2019), have been previously reported.

Knowledge Translation Goals and Activities

With support from the Ontario SPOR (Strategy for Patient-Oriented Research) SUPPORT Unit EMPOWER award, our team set out three primary knowledge translation goals to act on the top 10 patient-oriented priorities for pediatric chronic pain research and care in Canada. Together, we undertook diverse activities aligned with our goals to reach various key audiences including patients and families, healthcare professionals, researchers, research funders, decision makers and policy makers.

First goal: To generate awareness about and interest in the top 10 priorities in pediatric chronic pain and the partnership process undertaken to identify these priorities

Activities

We co-created a suite of materials outlining the top 10 priorities. Three key outputs included (1) a one-page infographic available in English and in French outlining the priority-setting methodology and results to enable quick communication; (2) a brief video taken at the final priority-setting consensus meeting that includes interviews with youth, parents and healthcare professionals engaged in the priority-setting activities and outlines the top 10 list; and (3) a co-developed patient journey map that graphically outlines one family's lived experience with chronic pain from early childhood to late adolescence, including highlights. The patient journey map is intended to meaningfully articulate the lived experience impact of the top 10 priorities. A "Partnering For Pain" logo and website were created to further facilitate communication and awareness through brand recognition and centralized access to outputs (https://partneringforpain.com/portfolio/top-10-priorities-for-pain/). Additional activities to generate awareness about the top 10 priorities include articles written for the Canadian Institutes of Health Research (CIHR) Institute of Musculoskeletal Health and Arthritis (Birnie 2019) targeting researchers and health research funders and in *RELIEF* – a publication by the International Association for the Study of Pain (Sukel 2020) – targeting the public, and a podcast episode with Pain BC's (2020) *Pain Waves* targeting healthcare professionals and the public.

Impact

Increased awareness and interest in the top 10 priorities in pediatric chronic pain and the partnership process is reflected through widely reached key audiences who have read, listened to and attended the knowledge translation activities. In 2020, the Partnering For Pain Priority Setting Partnership received the Pain Awareness Award from the Canadian Pain Society, as well as a CIHR Institute of Human Development Child Youth Health Video Prize (https://partneringforpain.com/portfolio/top-10-priorities-for-pain/).

Second goal: To facilitate collaborative dialogue among patients, families, researchers, healthcare professionals and decision makers to address the top 10 priorities in pediatric chronic pain

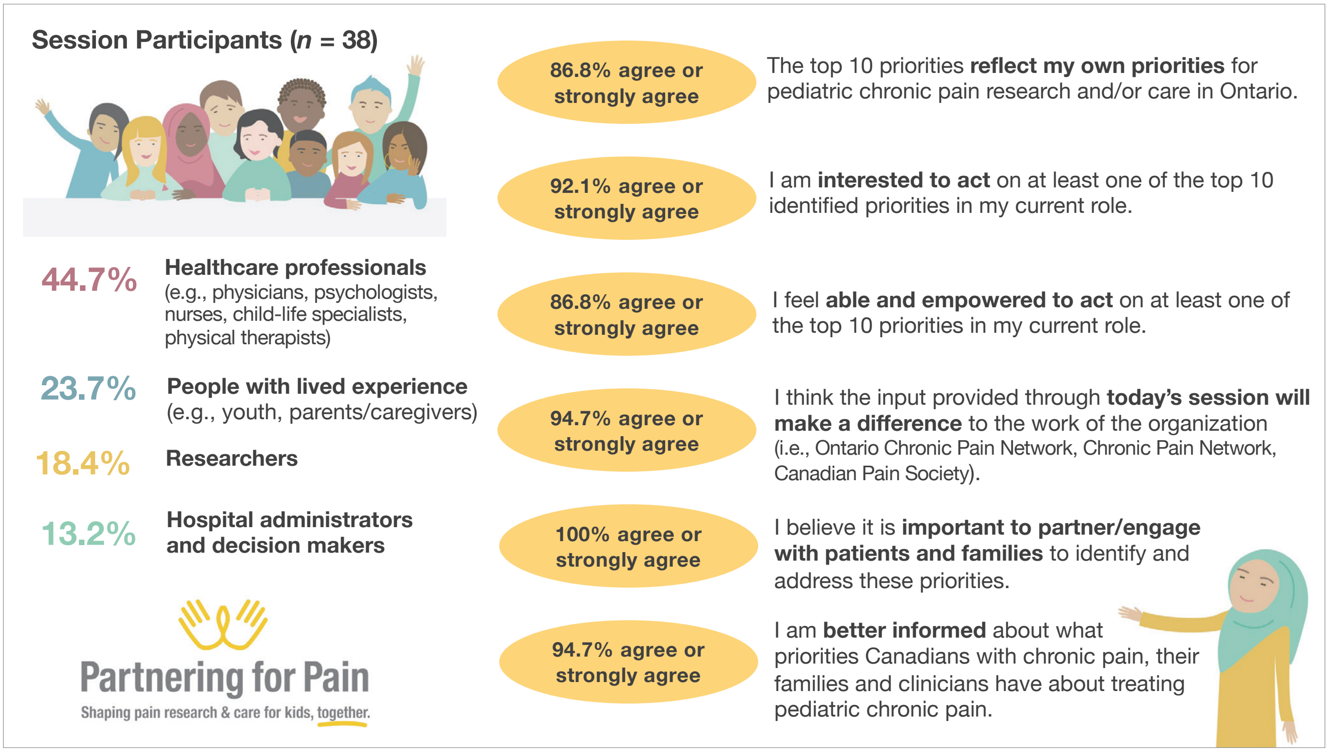
Activities

Research team members and youth and parent partners co-developed and co-facilitated round-table discussions of the top 10 priorities with multidisciplinary groups of healthcare professionals and researchers at the Ontario Chronic Pain Network annual meeting, the Chronic Pain Network (CIHR-funded SPOR) and the Canadian Pain Society annual meeting.

Impact

Evaluation from the round-table dialogue sessions about the top 10 priorities are shown in Figure 1.

FIGURE 1.
Evaluation from the round-table dialogue sessions about the top 10 priorities in pediatric chronic pain



Session Participants (*n* = 38)

44.7% **Healthcare professionals** (e.g., physicians, psychologists, nurses, child-life specialists, physical therapists)

23.7% **People with lived experience** (e.g., youth, parents/caregivers)

18.4% **Researchers**

13.2% **Hospital administrators and decision makers**

86.8% agree or strongly agree	The top 10 priorities **reflect my own priorities** for pediatric chronic pain research and/or care in Ontario.
92.1% agree or strongly agree	I am **interested to act** on at least one of the top 10 identified priorities in my current role.
86.8% agree or strongly agree	I feel **able and empowered to act** on at least one of the top 10 priorities in my current role.
94.7% agree or strongly agree	I think the input provided through **today's session will make a difference** to the work of the organization (i.e., Ontario Chronic Pain Network, Chronic Pain Network, Canadian Pain Society).
100% agree or strongly agree	I believe it is **important to partner/engage with patients and families** to identify and address these priorities.
94.7% agree or strongly agree	I am **better informed** about what priorities Canadians with chronic pain, their families and clinicians have about treating pediatric chronic pain.

Source: Image reprinted with permission from Partnering for Pain.

Third goal: To engage pediatric pain researchers, healthcare professionals, government decision makers and healthcare and patient organizations to integrate and adopt the top 10 priorities in pediatric chronic pain research, clinical practice, policy and advocacy

Activities

Knowledge translation activities also focused on enabling action on the top 10 priorities. Following consultation with the Ontario SPOR SUPPORT Unit, our team created an Evidence and Gap Map, which mapped existing evidence from systematic review and clinical practice guidelines for any treatment for pediatric chronic pain to the top 10 priorities (Birnie et al. 2020). Evidence and Gap Maps provide visual summaries of the scope and quality of available evidence to enable informed decisions for policy makers (Saran 2020; Snilstveit et al. 2017). A research team member and youth partner evaluated whether each scientific article included in the Evidence and Gap Map addressed each of the top 10 priorities. This work identified POR priority areas with sufficient and high-quality evidence for immediate action by healthcare professionals, decision makers and policy makers (i.e., psychological interventions for pediatric chronic pain), as well as areas needing further evidence for prioritization by health research funders and researchers (e.g., strategies for educating school personnel about pediatric chronic pain).

Co-presentations with youth and parent partners of the top 10 priorities and our partnership process included posters, webinars and talks targeting researchers (e.g., Canadian Pain Society, International Association for the Study of Pain, International Symposium on Pediatric Pain, Chronic Pain Network), healthcare professionals and families (Children's Healthcare Canada, Pediatric Pain Project ECHO, Alberta SPOR SUPPORT Unit Patient Engagement platform) and decision makers and policy makers (Canadian Academy of Health Sciences). Two youth partners were also highly engaged in additional activities targeting policy makers as members of the External Advisory of Health Canada's Canadian Pain Task Force and as part of a lobby day on Parliament Hill in Ottawa, ON, in partnership with Solutions for Kids in Pain (SKIP)– a Networks of Centres of Excellence knowledge mobilization network focused on pediatric pain.

Impact

A great success for our knowledge translation activities was the integration of the top 10 priorities and patient partnership approach into the following: reports from Health Canada's Canadian Pain Task Force (Canadian Pain Task Force 2019), the Canadian Academy of Health Sciences' development of a national strategy for chronic pain (CAHS 2018) and a recent Lancet Child & Adolescent Health Commission in Paediatric Pain (Eccleston et al. 2021). These documents guide priorities and recommend actions for transformative change to improve pain care for youth and their families in Canada and around the world. Following completion of the Priority Setting Partnership, the Partnering For Pain POR program was developed at the University of Calgary, led by Katie Birnie, which seeks to action all of the identified top 10 priorities.

Key Messages and Implications

The OSSU EMPOWER award and patient partnership enabled the creation of novel, creative and accessible knowledge translation activities and outputs to better reach different audiences beyond other pediatric pain researchers (i.e., the public, patients and families, healthcare professionals, decision makers and policy makers). A focus on knowledge translation and patient partnership enabled our work to move beyond knowledge sharing to behaviour change.

Challenges included some turnover of youth and parent partners due to the length of the project, as well as the high level of engagement required. This was overcome through inclusion of multiple youth and parent partners to decrease potential burden and create flexibility over type and amount of engagement. Additional challenges included the geographical location of project team members across the country, thereby requiring various methods – including virtual team meetings, phone calls and use of an online discuss forum – to build strong relationships and meaningful connections between team members. Lastly, having one main project lead and contact for youth and parent partners helped to mitigate any potential dual relationships and power differentials for youth partners who were former pediatric patients of healthcare professional team members.

All project and knowledge translation activities were undertaken in partnership with youth with lived experience and parents. It is our belief that patient partnership achieved greater reach and impact of the top 10 priorities by powerfully combining lived experience with rigorous scientific methodology. Our patient partners took the projects' findings to new conversations and new audiences, particularly with decision makers and policy makers. Lived experience narratives are a powerful knowledge translation tool that supports developing policies, shared decision making, strategies and research in healthcare through increased awareness of patient experiences (Park et al. 2021; Rose et al. 2016). Collaboration and advocacy by team members who are also researchers was needed to overcome challenges for youth and parent partners, including securing compensation for invited presentations with external partner organizations.

The national scale and scope of this project's high-quality and meaningful patient partnership serves as a key exemplar to other researchers, thus enabling more researchers as well as youth and families to engage in patient partnerships, particularly in the areas of child health and pain. HQ

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References

Birnie, K.A. 2019. Partnering For Pain: A Priority Setting Partnership to Establish Patient-Oriented Research Priorities in Pediatric Chronic Pain. Canadian Institutes of Health Research. Retrieved October 18, 2021. <https://cihr-irsc.gc.ca/e/51558.html>.

Birnie, K.A., K. Dib and C. Ouellette. 2018, October. Co-Building a New Landscape in Pediatric Chronic Pain Research: Patient Partner and Researcher Perspectives on Meaningful Patient Engagement. *Pediatric Pain Letter* 20(3): 21–27. Retrieved October 18, 2021. <http://ppl.childpain.org/issues/v20n3_2018/v20n3_birnie.pdf>.

Birnie, K.A., K. Dib, C. Ouellette, M.A. Dib, K. Nelson, D. Pahtayken et al. 2019. Partnering For Pain: A Priority Setting Partnership to Identify Patient-Oriented Research Priorities for Pediatric Chronic Pain in Canada. *CMAJ Open* 7(4): E654–64. doi:10.9778/cmajo.20190060.

Birnie, K.A., C. Ouellette, T. Do Amaral and J.N. Stinson. 2020. Mapping the Evidence and Gaps of Interventions for Pediatric Chronic Pain to Inform Policy, Research, and Practice: A Systematic Review and Quality Assessment of Systematic Reviews. *Canadian Journal of Pain* 4(1): 129–48. doi:10.1080/24740527.2020.1757384.

Caes, L., K.E. Boerner, C.T. Chambers, M. Campbell-Yeo, J. Stinson, K.A. Birnie et al. 2016. A Comprehensive Categorical and Bibliometric Analysis of Published Research Articles on Pediatric Pain from 1975 to 2010: *PAIN* 157(2): 302–13. doi:10.1097/j.pain.0000000000000403.

Canadian Academy of Health Sciences (CAHS). 2018, November 22. *CAHS Major Forum 2018: Chronic Pain Summary Report Arising from Forum*. Retrieved October 18, 2021. <https://cahs-acss.ca/wp-content/uploads/2018/11/Forum-2018-Proceedings-Report.pdf>.

Canadian Pain Task Force. 2019, June. *Chronic Pain in Canada: Laying a Foundation for Action*. Health Canada. Retrieved October 18, 2021. <https://www.canada.ca/content/dam/hc-sc/documents/corporate/about-health-canada/public-engagement/external-advisory-bodies/canadian-pain-task-force/report-2019/canadian-pain-task-force-June-2019-report-en.pdf>.

Eccleston, C., E. Fisher, R.F. Howard, R. Slater, P. Forgeron, T.M. Palermo et al. 2021. Delivering Transformative Action in Paediatric Pain: A *Lancet* Child & Adolescent Health Commission. *The Lancet Child and Adolescent Health* 5(1): 47–87. doi:10.1016/S2352-4642(20)30277-7.

Forgeron, P.A., S. King, J.N. Stinson, P.J. McGrath, A.J. MacDonald and C.T. Chambers. 2010. Social Functioning and Peer Relationships in Children and Adolescents with Chronic Pain: A Systematic Review. *Pain Research and Management* 15(1): 27–41. doi:10.1155/2010/820407.

Groenewald, C.B., S.W. Tham and T.M. Palermo. 2020. Impaired School Functioning in Children with Chronic Pain: A National Perspective. *The Clinical Journal of Pain* 36(9): 693–99. doi:10.1097/AJP.0000000000000850.

Health Canada. 2021, May. *An Action Plan for Pain in Canada*. Retrieved October 18, 2021. <https://www.canada.ca/content/dam/hc-sc/documents/corporate/about-health-canada/public-engagement/external-advisory-bodies/canadian-pain-task-force/report-2021-rapport/report-rapport-2021-eng.pdf>.

James Lind Alliance Priority Setting Partnerships. 2018. JLA Guidebook. Retrieved December 1, 2018. <https://www.jla.nihr.ac.uk/jla-guidebook/>.

King, S., C.T. Chambers, A. Huguet, R.C. MacNevin, P.J. McGrath, L. Parker et al. 2011. The Epidemiology of Chronic Pain in Children and Adolescents Revisited: A Systematic Review. *PAIN* 152(12): 2729–38. doi:10.1016/j.pain.2011.07.016.

Lewandowski, A.S., T.M. Palermo, J. Stinson, S. Handley and C.T. Chambers. 2010. Systematic Review of Family Functioning in Families of Children and Adolescents with Chronic Pain. *The Journal of Pain* 11(11): 1027–38. doi:10.1016/j.jpain.2010.04.005.

Liossi, C., A.-K. Anderson, R.F. Howard, On behalf of NIHR CRN-C CSG in Pain and Palliative Care. 2017. Development of Research Priorities in Paediatric Pain and Palliative Care. *British Journal of Pain* 11(1): 9–15. doi:10.1177/2049463716668906.

Manafò, E., L. Petermann, V. Vandall-Walker and P. Mason-Lai. 2018. Patient and Public Engagement in Priority Setting: A Systematic Rapid Review of the Literature. *PLoS ONE* 13(3): e0193579. doi:10.1371/journal.pone.0193579.

Pain BC. 2020. How Two Canadian Initiatives Are Advancing Children's Pain Research and Practice. *Pain Waves*. Retrieved February 29, 2020. <https://poddtoppen.se/podcast/1169792513/pain-waves/how-two-canadian-initiatives-are-advancing-childrens-pain-research-and-practice>.

Park, E., M. Forhan and C.A. Jones. 2021. The Use of Digital Storytelling of Patients' Stories as an Approach to Translating Knowledge: A Scoping Review. *Research Involvement and Engagement* 7(1): 58. doi:10.1186/s40900-021-00305-x.

Rose, R., S. Chakraborty, P. Mason-Lai, W. Brocke, S.A. Page and D. Cawthorpe. 2016. The Storied Mind: A Meta-Narrative Review Exploring the Capacity of Stories to Foster Humanism in Health Care. *Journal of Hospital Administration* 5(1): 52. doi:10.5430/jha.v5n1p52.

Saran, A. 2020. Evidence and Gap Maps. *Campbell Systematic Reviews* 16(1): e1075. doi:10.1002/cl2.1075.

Snilstveit, B., R. Bhatia, K. Rankin and B. Leach. 2017, February. *3ie Evidence Gap Maps: A Starting Point for Strategic Evidence Production and Use*. Working Paper 28. International Initiative for Impact Evaluation. Retrieved June 9, 2018. <https://www.3ieimpact.org/sites/default/files/2019-01/wp28-egm.pdf>.

Sukel, K. 2020, July 8. Managing Pain in Kids: What We Know and What We Don't Know [Previously accessed]. *Relief: Pain Research News, Insights, and Ideas.*

Valrie, C.R., M.H. Bromberg, T. Palermo and L.E. Schanberg. 2013. A Systematic Review of Sleep in Pediatric Pain Populations. *Journal of Developmental and Behavioral Pediatrics* 34(2): 120–28. doi:10.1097/DBP.0b013e31827d5848.

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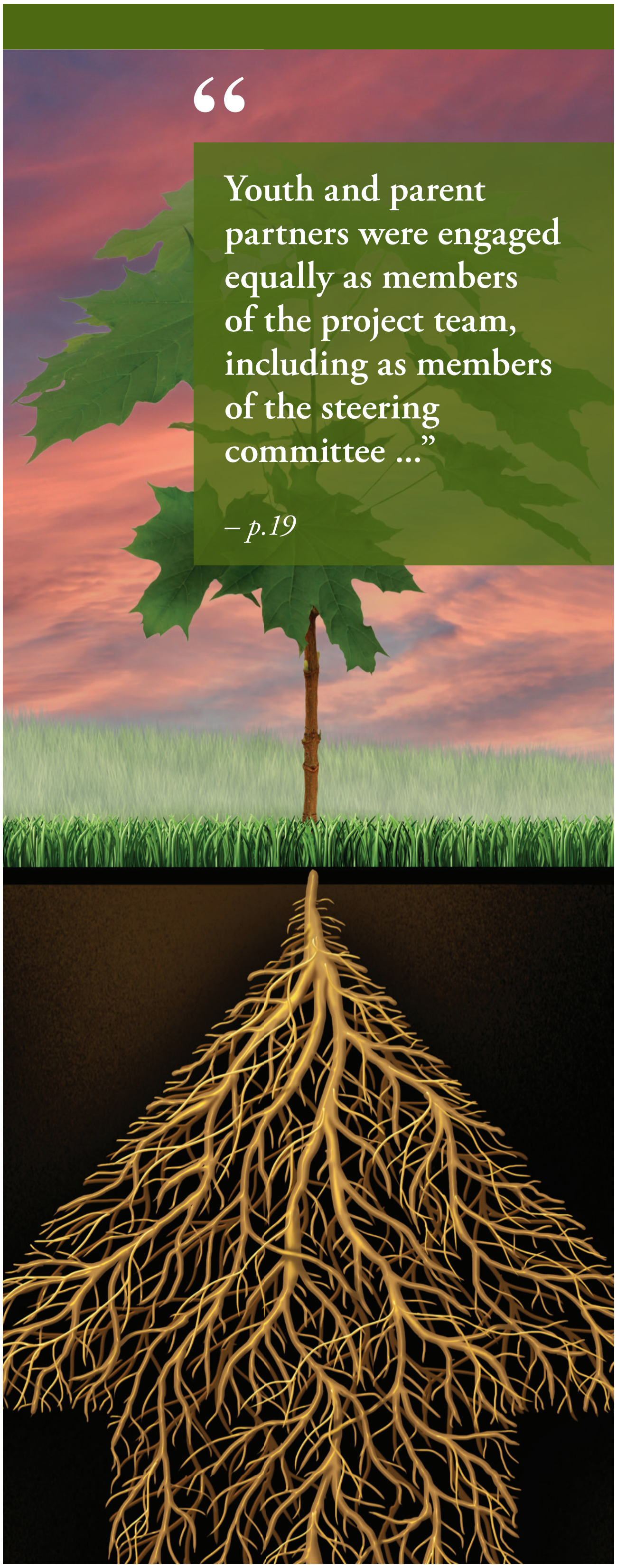

> "Youth and parent partners were engaged equally as members of the project team, including as members of the steering committee ..."
>
> *– p.19*

EQUITY IN PATIENT PARTNERSHIPS

> “Feedback from patient partners reflected the team’s success in achieving meaningful, inclusive, accessible and collaborative partnership.”
>
> *– p. 27*

Ensuring Equity and Inclusion in Virtual Care Best Practices for Diverse Populations of Youth with Chronic Pain

Kathryn A. Birnie, Tieghan Killackey, Gillian Backlin[P], Frank Gavin[P], Christine Harris[P], Isabel Jordan[P], Laesa Kim[P], Justina Marianayagam[P], Jenna Swidrovich[P], Corinne Lalonde, Lanre Tunji-Ajayi, Tim Oberlander, Melanie Kirby-Allen, Simon Lambert, Hal Siden, Jaris Swidrovich, Melanie Noel, Chitra Lalloo and Jennifer Stinson

Abstract

Poor access to care is a top patient-oriented research priority for youth with chronic pain in Canada, and the COVID-19 pandemic has exacerbated these concerns. Our patient-oriented project team engaged with marginalized and racialized youth with chronic pain (Black youth with sickle cell disease, Indigenous youth and youth with complex medical needs) and their families to ensure that best practice recommendations for virtual care are inclusive and equitable. Input provided through virtual round-table discussions improved recommendations for leveraging, implementing and selecting best platforms for virtual care for youth with chronic pain and identified new gaps for future research, practice and policy change.

Key Points

- Partnership is key to equitable, diverse and inclusive engagement, particularly when engaging with populations or population groups that are marginalized.
- Virtual activities both facilitated and hindered equitable, diverse, inclusive and accessible engagement.
- Patient engagement offered an opportunity to critically expand on and refine learnings from the scientific literature – in this case, on a rapidly emerging widespread need for virtual care for pain during the COVID-19 pandemic.

Introduction

The COVID-19 pandemic necessitated a rapid and wide-scale pivot to virtual care across all areas of health, including for youth living with chronic pain (Bokolo 2020; D'Alessandro et al. 2020; Eccleston et al. 2020). Poor access to care is a top patient-oriented research priority for youth with chronic pain in Canada (Birnie et al. 2019), and the COVID-19 pandemic has exacerbated these concerns with the closure of clinics and therapies (Cohen et al. 2020; Eccleston et al. 2020; Killackey et al. 2021; Shanthanna et al. 2020). Early treatment of pain in youth is critical to prevent persisting pain, mental health and substance use issues into adulthood (Groenewald et al. 2019; Kashikar-Zuck et al. 2014; Murray et al. 2019; Walker et al. 2010, 2012), and the COVID-19 pandemic has only reinforced that necessity. In 2020, members of our team received a Canadian Institutes of Health Research (CIHR) rapid COVID-19 Knowledge Synthesis Grant to review existing scientific literature to identify recommendations for best practices for virtual care for youth with chronic pain and their families (Birnie et al. 2021a). The 16 scientific articles included in our scoping review highlighted a range of recommendations related to leveraging and implementing virtual care, selecting best virtual care platforms and identifying limitations and considerations for remaining research priorities for pediatric chronic pain. These articles also underscore the immense potential for harnessing virtual care to address

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P = Patient partner.

this growing population. These themes were summarized in a one-page infographic available in English and in French (https://partneringforpain.com/) (Birnie et al. 2021a). This review was intended to guide healthcare professionals and decision makers in evidence-informed practice and policy.

However, in undertaking this scoping review, our team recognized the omission of diverse populations or population groups in the included studies that identified best practices for virtual care for youth with chronic pain. In general, pain services and research disproportionately exclude individuals who are marginalized, such as people who are Indigenous, Black or Persons of Colour, or people who cannot communicate verbally, among others, despite being vulnerable to a higher prevalence of chronic pain and less likely to be able to access virtual care (e.g., poorer Internet access; Craig et al. 2019; Crawford and Serhal 2020; Latimer et al. 2018). According to the International Association for the Study of Pain, populations can be considered vulnerable due to (i) physical, psychological and/or verbal impairments, (ii) social circumstances or (iii) shortcomings in healthcare and are at risk of being under-assessed and undertreated for pain (IASP n.d.). It is imperative that recommendations for virtual care include the perspectives of diverse and vulnerable pain populations that are underrepresented in existing clinical care and scientific literature. Evidence-based and emerging virtual treatments exist for pediatric pain generally (Birnie et al. 2021b), but implementation and access are sparse.

Project Objectives and Overview

The aim of our project was to engage with diverse youth with chronic pain and their families to ensure that recommendations for best practices for virtual care are inclusive and equitable. We engaged Black youth with sickle cell disease, Indigenous youth (including all of First Nations [status and non-status], Métis and Inuit) and youth with complex medical needs (including youth with brain-based developmental disabilities), their parents/caregivers and healthcare professionals in collaborative dialogue regarding recommendations for virtual care for chronic pain identified in our original scoping review (Birnie et al. 2021a). These populations were specifically selected because they are susceptible to inequities in pain care (Craig et al. 2019) and healthcare access (Kuo et al. 2014), experience stigmatization (Jenerette and Brewer 2010; Wakefield et al. 2017) and systemic barriers (i.e., systemic racism; Anastas et al. 2020; Latimer et al. 2018; Wylie et al. 2019) or may be impacted by socio-economic factors that further exacerbate existing inequities (Ambrose 2020), all of which can ultimately impact virtual care delivery. Virtual care is here to stay, and we must ensure that it meets the needs of diverse youth and their families. Creating space for patient and family voices to direct decisions regarding virtual care best practices in Canada is both timely, given the COVID-19 pandemic, and long overdue, given their integral role as partners in virtual care.

Knowledge Translation Goals and Activities

With support through an EMPOWER (Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach) award from the Ontario SPOR (Strategy for Patient-Oriented Research) SUPPORT Unit (OSSU), our team set three primary knowledge translation goals:

1. to facilitate collaborative dialogue regarding recommendations for virtual care for diverse chronic pain populations – specifically Black youth with sickle cell disease, Indigenous youth and youth with complex medical needs – and their families;

2. to engage diverse stakeholders and rights holders in contributing to recommendations for virtual care best practices in pediatric chronic pain; and

3. to share findings related to identified recommendations for virtual care with relevant stakeholder and rights holder groups and explore how these recommendations may be tailored to best address the needs of diverse chronic pain populations.

Knowledge translation goals were achieved through co-facilitation of virtual interactive round-table discussions and engagement conducted separately with each group (Black youth with sickle cell disease, Indigenous youth and youth with complex medical needs, including youth with a brain-based developmental disability) and stakeholder or Indigenous rights holder type (youth, parents/caregivers). Each session was tailored based on conversations with relevant patient partners and partner organizations, with some conducted in groups and others via individual interviews.

Patient Partner Roles

Our team used the term “patient partner” according to the CIHR definition to refer to individuals with personal experience of a health issue (i.e., chronic pain during childhood) and informal caregivers, including family and friends (CIHR 2018). Our team included nine patient partners with lived experience relevant to one of the three population groups of interest, as well as pediatric pain researchers, healthcare professionals and partner organization collaborators (Complex Care for Kids Ontario [CCKO], CHILD-BRIGHT Network – National Youth Advisory Panel [NYAP], Sickle Cell Awareness Group of Ontario [SCAGO], Network Environments for Indigenous Health Research [NEIHR] National Coordinating Centre and Solutions for Kids in Pain [SKIP]).

Patient partners were included as equal members of the research team, and partnership was sustained through regular virtual meetings and e-mail communication altogether, and separate communication was maintained for each population group of interest as well. This structure enabled continuity as well as flexibility for each population group to design and carry out engagement in ways that increased accessibility and equity for patient partners and community members. Interactive round-table discussions were co-designed for each population group in collaboration with patient partners and relevant partner organizations. Patient partners were centrally involved in facilitating all the round-table discussion sessions, including how engagement created a safe space for open dialogue and selecting questions for session evaluation. At each interactive round-table discussion, research and health professional team members were also present to provide a brief overview of the previously completed scoping review (Birnie et al. 2021a). Patient partners also contributed to synthesis and presentation of what we learned, including infographics and co-presentation of project findings.

The team's collaborative approach was reflected by a parent/caregiver patient partner:

> Among the many things I appreciated was that we – the three parents – had quite a bit of choice about how and how much we would be involved … More importantly, we were encouraged to provide suggestions about every aspect of the sessions and saw all of them taken up and applied. For instance, we recommended that there be two sessions rather than one (with the same group of participants), since that would give participants a chance to reflect over the course of a week on what they had heard and said and perhaps make it easier for the less-quick-to-speak parents to contribute more to the discussion. We also provided suggestions on the recruiting materials, the land acknowledgement, the "ice-breaker" and the questions or prompts themselves. And the "wrap-up" session was a chance not only to review the process but to add our own further thoughts.

Feedback from patient partners reflected the team's success in achieving meaningful, inclusive, accessible and collaborative partnership. As shared by a youth patient partner:

> First and foremost, I would like to thank the team for allowing me to partake in this project through co-facilitating and sharing my story along with the other youth. I was impressed by their eagerness to learn from me and each other participant about their experiences. Their genuine interest in creating a fruitful and accessible discussion for all made this process comfortable and enjoyable from start to finish. I really appreciated that accessibility was always prioritized. They thought of everything, from time zones to content format. The team was very approachable and a pleasure to work with.

Project Outcomes

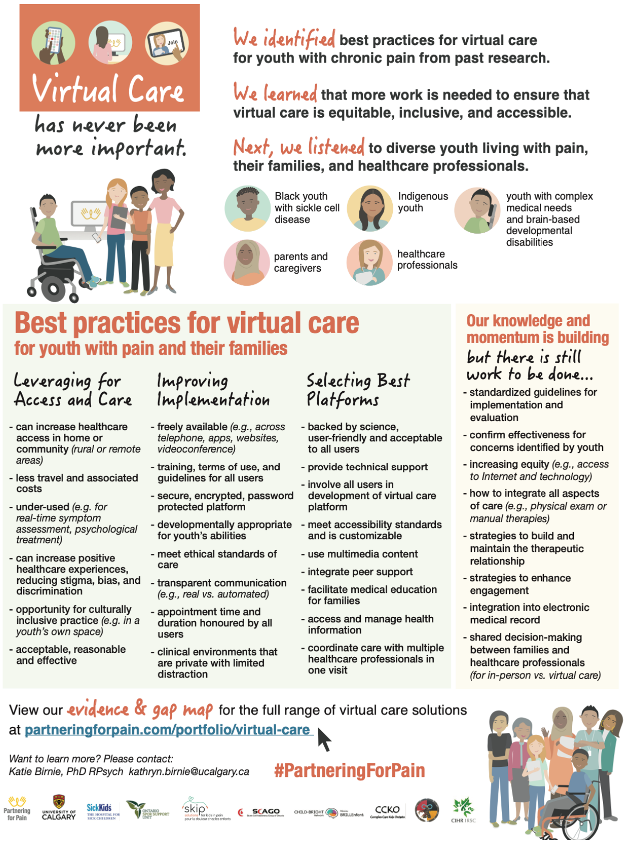
Interactive round-table discussions were held via Zoom from November 2020 to January 2021. In total, 11 Black youth with sickle cell disease and seven parents/caregivers, two Indigenous youth with chronic pain and one parent/caregiver, and three youth with complex medical needs and four parents/caregivers were engaged.

Each virtual engagement session was evaluated using four questions adapted from the Patient and Public Engagement and Evaluation Tool module for one-time engagement activities (Abelson et al. 2016; Abelson and The PPEET Research-Practice Collaborative 2018). Of the 11 round-table discussion participants who completed the post-session evaluations, all agreed that they felt that their views were heard, that the goals of the sessions and questions presented were clear and that the engagement initiative was a good use of their time. All but one (91%) agreed that they were confident that the input provided through the engagement initiative would be used.

Suggestions made during the round-table discussions revealed additions or modifications to the best practices for virtual care for youth with chronic pain and their families not identified in the previous scoping review (Birnie et al. 2021a):

- *Leveraging virtual care:* (1) It may be an opportunity to increase positive healthcare experiences, reducing stigma, bias and discrimination in the healthcare system, thereby encouraging those in the 2SLGBTQ+ (Two-Spirit, Lesbian, Gay, Bisexual, Trans, Queer+) and BIPOC (Black, Indigenous and People of Colour) communities to access healthcare. (2) It will allow patients and families to access care in their own environment. (3) It will provide more opportunity for culturally inclusive practices (e.g., Indigenous youth may smudge in their own environments before an appointment).
- *Improving implementation of virtual care:* (1) Clinical environments should be arranged to support virtual care (e.g., limited distractions, private environment). (2) Appointment time and duration should be respected as much as possible by healthcare professionals, patients and families with consideration given to building trust and connection.
- *Selecting best platforms for virtual care:* (1) The virtual care platform should facilitate virtual medical education, teaching and demonstration for patients, families and caregivers (e.g., how to flush a line). (2) It should allow patients, families and caregivers to easily access and manage health information. (3) It should be able to

FIGURE 1.
Best practices for virtual care for diverse youth with chronic pain and their families (English-language version)



Virtual Care has never been more important.

We identified best practices for virtual care for youth with chronic pain from past research.

We learned that more work is needed to ensure that virtual care is equitable, inclusive, and accessible.

Next, we listened to diverse youth living with pain, their families, and healthcare professionals.

Black youth with sickle cell disease

Indigenous youth

youth with complex medical needs and brain-based developmental disabilities

parents and caregivers

healthcare professionals

Best practices for virtual care
for youth with pain and their families

Leveraging for Access and Care

- can increase healthcare access in home or community *(rural or remote areas)*
- less travel and associated costs
- under-used *(e.g. for real-time symptom assessment, psychological treatment)*
- can increase positive healthcare experiences, reducing stigma, bias, and discrimination
- opportunity for culturally inclusive practice *(e.g. in a youth's own space)*
- acceptable, reasonable and effective

Improving Implementation

- freely available *(e.g., across telephone, apps, websites, videoconference)*
- training, terms of use, and guidelines for all users
- secure, encrypted, password protected platform
- developmentally appropriate for youth's abilities
- meet ethical standards of care
- transparent communication *(e.g., real vs. automated)*
- appointment time and duration honoured by all users
- clinical environments that are private with limited distraction

Selecting Best Platforms

- backed by science, user-friendly and acceptable to all users
- provide technical support
- involve all users in development of virtual care platform
- meet accessibility standards and is customizable
- use multimedia content
- integrate peer support
- facilitate medical education for families
- access and manage health information
- coordinate care with multiple healthcare professionals in one visit

Our knowledge and momentum is building
but there is still work to be done...

- standardized guidelines for implementation and evaluation
- confirm effectiveness for concerns identified by youth
- increasing equity *(e.g., access to Internet and technology)*
- how to integrate all aspects of care *(e.g., physical exam or manual therapies)*
- strategies to build and maintain the therapeutic relationship
- strategies to enhance engagement
- integration into electronic medical record
- shared decision-making between families and healthcare professionals *(for in-person vs. virtual care)*

View our **evidence & gap map** for the full range of virtual care solutions at partneringforpain.com/portfolio/virtual-care

Want to learn more? Please contact:
Katie Birnie, PhD RPsych kathryn.birnie@ucalgary.ca

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accommodate care with multiple healthcare professionals in one visit.

- *Identfying gaps in virtual care:* (1) Solutions are required to increase equity across Canada (e.g., reliable access to the Internet and technology required for virtual care). (2) Decisions for the type of care (virtual vs. in-person) should be made in collaboration with patients and families.

These findings are being integrated into a policy brief and revised infographic (see Figures 1 and 2 for the English- and French-language versions, respectively). These are shared on our project website: www.partneringforpain.com/portfolio/virtual-care. We have previously shared a poster about our project at the CHILD-BRIGHT virtual symposium, which was co-presented by patient partners. Members of our team have also been invited to engage with national decision makers and policy makers, such as Canada Health Infoway and Health Canada, working to improve virtual care.

FIGURE 2.
Best practices for virtual care for diverse youth with chronic pain and their families (French-language version)

Les soins virtuels n'ont jamais été aussi importants.

Nous avons identifié les meilleures pratiques en matière de soins virtuels pour les jeunes vivant avec la douleur chronique, en nous appuyant sur les recherches antérieures.

Nous avons compris qu'il faut poursuivre le travail pour faire en sorte que les soins virtuels soient équitables, inclusifs et accessibles.

Nous avons ensuite écouté le point de vue de divers jeunes souffrant de douleur chronique, de leur famille et de professionnels de la santé :

Jeunes noirs ayant l'anémie falciforme

Jeunes Autochtones

Jeunes ayant des besoins complexes et des troubles du développement d'origine cérébrale

Parents et aidants

Professionnels de la santé

Pratiques exemplaires en soins virtuels
pour les jeunes souffrant de douleur chronique et leur famille.

La mobilisation pour l'accès aux soins :

- accroître l'accès aux soins à domicile ou dans la communauté *(régions rurales ou éloignées)*;
- diminuer les besoins en transports et les coûts qui y sont rattachés;
- accroître l'accès aux soins sous-utilisés *(en particulier pour l'évaluation des symptômes en temps réel et le traitement psychologique)*;
- accroître les expériences de soins de santé positives, réduire la stigmatisation, les préjugés et la discrimination;
- favoriser les pratiques culturellement inclusives *(ex. dans un lieu réservé aux jeunes)*;
- donner accès à des soins acceptables, raisonnables et efficaces.

Améliorer la mise en œuvre :

- services offerts gratuitement sur toutes les plateformes technologiques *(téléphone, applications, sites Web, visioconférence);*
- formation, conditions d'utilisation et lignes directrices pour tous les utilisateurs;
- services sécuritaires *(chiffrement, protection par mots de passe)*;
- adaptés au développement des capacités des jeunes;
- conformes aux normes de soins éthiques;
- communication transparente *(communications réelles plutôt que communications automatisées)*;
- respect de l'heure et la durée des rendez-vous par tous les utilisateurs;
- environnement clinique privé avec distraction limitée.

Choix des meilleures plateformes :

- fondées sur les connaissances, conviviales et adaptées aux utilisateurs;
- offrir du soutien technique;
- impliquer tous les utilisateurs dans le développement de plateforme de soins virtuels;
- conformes aux normes d'accessibilité et adaptables;
- utilisation de contenu multimédia;
- intégrant du soutien par les pairs;
- facilitant l'éducation médicale des familles;
- accès et gestion de l'information sur la santé;
- coordination des soins avec différents professionnels de la santé en une seule visite.

Nos connaissances s'améliorent et nous sommes sur une bonne lancée...
mais il y a encore du travail à faire...

- lignes directrices uniformisées concernant la mise en œuvre et l'évaluation;
- confirmer l'efficacité pour les préoccupations soulevées par les jeunes;
- accroître l'équité *(ex. accès à Internet et aux technologies)*;
- intégration de tous les types de soins *(ex. examen physique ou thérapies manuelles)*;
- stratégies pour établir et maintenir les relations thérapeutiques;
- stratégies pour favoriser l'engagement;
- intégration aux dossiers médicaux informatisés;
- prise de décision partagée par les familles et les professionnels de la santé *(pour le choix des soins en personne ou des soins virtuels)*.

Consultez notre carte **« des données probantes et des lacunes »** pour voir l'ensemble des solutions de soins virtuels à : partneringforpain.com/portfolio/virtual-care.

Vous souhaitez en savoir plus?
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Katie Birnie, PhD RPsych kathryn.birnie@ucalgary.ca

#PartenairesPourLaDouleur

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Key Messages and Implications

An important reflection from our team is that partnership is key to equitable, diverse and inclusive engagement, particularly when engaging with populations or population groups who are marginalized (Craig et al. 2019). Strong partnership is created when each person can contribute their unique expertise and see their expertise both used and respected. Specifically, our team worked closely to empower patient partners to co-design and lead facilitation of engagement sessions to ensure that participants felt safe and respected. Contributions from patient partners led to the expansion of round-table discussions to two sessions instead of one for each group and also resulted in more clearly worded questions to guide the conversation. Working with each partner organization was critical for effectively reaching out and engaging with each population group. There was a need to be flexible within the engagement process to meet the different needs of each population group and address emerging challenges, such as shifting from group to individual and family-based engagement with Indigenous youth with chronic pain and their families.

Our interactive round-table discussion sessions were conducted using Zoom. Virtual activities both facilitated and hindered equitable, diverse, inclusive and accessible engagement. For example, going virtual made it more possible to engage with people across the country, but it likely limited our ability to reach those with poorer access to the Internet and communities where in-person community-based engagement is valued (e.g., with Indigenous communities).

The round-table discussion sessions provided new insights to improve equity and accessibility of best practices for virtual care. Patient engagement offered an opportunity to critically expand on and refine learnings from the scientific literature, in this case on a rapidly emerging widespread need for virtual care for pain during the COVID-19 pandemic.

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References

Abelson, J., K. Li, G. Wilson, K. Shields, C. Schneider and S. Boesveld. 2016. Supporting Quality Public and Patient Engagement in Health System Organizations: Development and Usability Testing of the Public and Patient Engagement Evaluation Tool. *Health Expectations* 19(4): 817–27. doi:10.1111/hex.12378.

Abelson, J. and The PPEET Research-Practice Collaborative. 2018, August. *Public and Patient Engagement Evaluation Tool (PPEET) Version 2.0*. McMaster University. Retrieved December 1, 2020. <https://healthsci.mcmaster.ca/docs/librariesprovider61/default-document-library/ppeet-complete-set-final.pdf?sfvrsn=d1617fe6_2>.

Ambrose, A.J.H. 2020. Inequities during COVID-19. *Pediatrics* 146(2): e20201501. doi:10.1542/peds.2020-1501.

Anastas, T.M., M.M. Miller, N.A. Hollingshead, J.C. Stewart, K.L. Rand and A.T. Hirsh. 2020. The Unique and Interactive Effects of Patient Race, Patient Socioeconomic Status, and Provider Attitudes on Chronic Pain Care Decisions. *Annals of Behavioral Medicine* 54(10): 771–82. doi:10.1093/abm/kaaa016.

Birnie, K.A., K. Dib, C. Ouellette, M.A. Dib, K. Nelson, D. Pahtayken K. et al. 2019. Partnering for Pain: A Priority Setting Partnership to Identify Patient-Oriented Research Priorities for Pediatric Chronic Pain in Canada. *CMAJ Open* 7(4): E654–64. doi:10.9778/cmajo.20190060.

Birnie, K.A., T. Killackey, J. Stinson, M. Noel, D.L. Lorenzetti, J. Marianayagam et al. 2021a. Best Practices for Virtual Care to Support Youth with Chronic Pain and Their Families: A Rapid Systematic Review to Inform Health Care and Policy during COVID-19 and Beyond. *PAIN Reports* 6(2): e935. doi:10.1097/PR9.0000000000000935.

Birnie, K.A., M. Pavlova, A. Neville, M. Noel, I. Jordan, E. Jordan et al. 2021b. Rapid Evidence and Gap Map of Virtual Care Solutions across a Stepped Care Continuum for Youth with Chronic Pain and Their Families in Response to the COVID-19 Pandemic. *PAIN* 162(11): 2658–68. doi:10.1097/j.pain.0000000000002339.

Bokolo, A. 2020. Use of Telemedicine and Virtual Care for Remote Treatment in Response to COVID-19 Pandemic. *Journal of Medical Systems* 44(7): 132. doi:10.1007/s10916-020-01596-5.

Canadian Institutes of Health Research (CIHR). 2018. Patient Engagement. Retrieved December 1, 2020. <www.cihr-irsc.gc.ca/e/45851.html>.

Cohen, S.P., Z.B. Baber, A. Buvanendran, L.T.C.B.C. McLean, Y. Chen, W.M. Hooten et al. 2020. Pain Management Best Practices from Multispecialty Organizations during the COVID-19 Pandemic and Public Health Crises. *Pain Medicine* 21(7): 1331–46. doi:10.1093/pm/pnaa127.

Craig, K.D., C. Holmes, M. Hudspith, G. Moor, M. Moosa-Mitha, C. Varcoe et al. 2019. Pain in Persons Who Are Marginalized by Social Conditions. *PAIN* 161(2): 261–65. doi:10.1097/j.pain.0000000000001719.

Crawford, A. and E. Serhal. 2020. Digital Health Equity and COVID-19: The Innovation Curve Cannot Reinforce the Social Gradient of Health. *Journal of Medical Internet Research* 22(6): e19361. doi:10.2196/19361.

D'Alessandro, L., S.C. Brown, F. Campbell, D. Ruskin, G. Mesaroli and J.N. Stinson. 2020. Rapid Mobilization of a Virtual Pediatric Chronic Pain Clinic in Canada during the COVID-19 Pandemic. *Canadian Journal of Pain* 4(1): 162–67. doi:10.1080/24740527.2020.1771688.

Eccleston, C., F.M. Blyth, B.F. Dear, E.A. Fisher, F.J. Keefe, M.E. Lynch et al. 2020. Managing Patients With Chronic Pain during the COVID-19 Outbreak: Considerations for the Rapid Introduction of Remotely Supported (eHealth) Pain Management Services. *PAIN* 161(5): 889–93. doi:10.1097/j.pain.0000000000001885.

Groenewald, C.B., E.F. Law, E. Fisher, S.E. Beals-Erickson and T.M. Palermo. 2019. Associations between Adolescent Chronic Pain and Prescription Opioid Misuse in Adulthood. *The Journal of Pain* 20(1): 28–37. doi:10.1016/j.jpain.2018.07.007.

International Association for the Study of Pain (IASP). (n.d.). Pain in the Most Vulnerable. Retrieved December 1, 2020. <https://www.iasp-pain.org/advocacy/global-year/pain-in-the-most-vulnerable/>.

Jenerette, C.M. and C. Brewer. 2010. Health-Related Stigma in Young Adults with Sickle Cell Disease. *Journal of the National Medical Association* 102(11): 1050–55. doi:10.1016/S0027-9684(15)30732-X.

Kashikar-Zuck, S., N. Cunningham, S. Sil, M.H. Bromberg, A.M. Lynch-Jordan, D. Strotman et al. 2014. Long-Term Outcomes of Adolescents with Juvenile-Onset Fibromyalgia in Early Adulthood. *Pediatrics* 133(3): e592–600. doi:10.1542/peds.2013-2220.

Killackey, T., M. Noel, K.A. Birnie, M. Choinière, M.G. Pagé, L. Dassieu et al. 2021. COVID-19 Pandemic Impact and Response in Canadian Pediatric Chronic Pain Care: A National Survey of Medical Directors and Pain Professionals. *Canadian Journal of Pain* 5(1): 139–50. doi:10.1080/24740527.2021.1931069.

Kuo, D.Z., A. Goudie, E. Cohen, A. Houtrow, R. Agrawal, A.C. Carle et al. 2014. Inequities in Health Care Needs for Children with Medical Complexity. *Health Affairs* 33(12): 2190–98. doi:10.1377/hlthaff.2014.0273.

Latimer, M., S. Rudderham, L. Lethbridge, E. MacLeod, K. Harman, J.R. Sylliboy et al. 2018. Occurrence of and Referral to Specialists for Pain-Related Diagnoses in First Nations and Non–First Nations Children and Youth. *CMAJ* 190(49): E1434–40. doi:10.1503/cmaj.180198.

Murray, C.B., C.B. Groenewald, R. de la Vega and T.M. Palermo. 2019. Long-Term Impact of Adolescent Chronic Pain on Young Adult Educational, Vocational, and Social Outcomes. *PAIN* 161(2): 439–45. doi:10.1097/j.pain.0000000000001732.

Shanthanna, H., N.H. Strand, D.A. Provenzano, C.A. Lobo, S. Eldabe, A. Bhatia et al. 2020. Caring for Patients with Pain during the COVID-19 Pandemic: Consensus Recommendations from an International Expert Panel. *Anaesthesia* 75(7): 935–44. doi:10.1111/anae.15076.

Wakefield, E.O., J.M. Popp, L.P. Dale, J.P. Santanelli, A. Pantaleao and W.T. Zempsky. 2017. Perceived Racial Bias and Health-Related Stigma among Youth with Sickle Cell Disease. *Journal of Developmental and Behavioral Pediatrics* 38(2): 129–34. doi:10.1097/DBP.0000000000000381.

Walker, L.S., C.M. Dengler-Crish, S. Rippel and S. Bruehl. 2010. Functional Abdominal Pain in Childhood and Adolescence Increases Risk for Chronic Pain in Adulthood. *PAIN* 150(3): 568–72. doi:10.1016/j.pain.2010.06.018.

Walker, L.S., A.L. Sherman, S. Bruehl, J. Garber and C.A. Smith. 2012. Functional Abdominal Pain Patient Subtypes in Childhood Predict Functional Gastrointestinal Disorders with Chronic Pain and Psychiatric Comorbidities in Adolescence and Adulthood. *PAIN* 153(9): 1798–806. doi:10.1016/j.pain.2012.03.026.

Wylie, L. and S. McConkey. 2019. Insiders' Insight: Discrimination against Indigenous Peoples through the Eyes of Health Care Professionals. *Journal of Racial and Ethnic Health Disparities* 6(1): 37–45. doi:10.1007/s40615-018-0495-9.

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Parent Engagement in a COVID-19 Cohort Study of Children and Families: Successes, Challenges and Next Steps

Shelley M. Vanderhout, Catherine S. Birken, Peter D. Wong, Shannon Weir[P], Jennifer Batten[P] and Jonathon L. Maguire

Abstract

This article describes the methods, successes and challenges of engaging parents while studying the impacts of COVID-19 on healthy children and families. Parent partners in a Parent and Clinician Team (PACT) informed study aims, supported feasibility and recommended changes to enhance participation. PACT members stated that they felt a sense of connectedness and purpose by contributing to COVID-19 research. Engagement increased by parents acquiring new roles, attending more frequent meetings and co-creating alternative methods of engagement. Recruiting new PACT members was challenging, likely due to limited time and resources available to parents of young children during a pandemic.

Key Points

- Parents co-developed child and family COVID-19 research by informing study aims, supporting feasibility, recommending changes to enhance participation and helping researchers adapt to evolving family concerns, priorities and challenges.
- Researchers perceived parent engagement to increase among those who had partnered prior to the pandemic but recruiting new, diverse parent partners during COVID-19 was challenging.
- The next steps include increasing the diversity of parent partners, co-presenting findings alongside parents, recruiting youth partners and evaluating our patient engagement approach.

Background

Patient engagement in health research can increase the acceptability, relevance and reach of the evidence it produces (Domecq et al. 2014) while allowing patient communities to have an active voice in the planning, conduct and dissemination of research (Frank et al. 2015). Best practices for engaging patient partners in research suggest including patients as early as possible (Kirwan et al. 2017). During the COVID-19 pandemic, child health researchers developed new studies to understand the physical, social, emotional and developmental effects of COVID-19 on children's health and well-being, creating an opportunity to partner with parents and families to conceptualize, design and conduct research.

Working alongside parent partners, we describe how support provided by an Ontario SPOR SUPPORT Unit (OSSU) EMPOWER award supported patient engagement in a study about the impacts of COVID-19 on healthy children and their families. Reflecting on the methods we used and the successes and challenges of engaging parents, we explore next steps for meaningfully engaging parents in child health research during the COVID-19 pandemic.

P = Patient partner.

TARGet Kids! COVID-19 Study of Children and Families

In March 2020, the TARGet Kids! COVID-19 Study of Children and Families was developed within the TARGet Kids! (Carsley et al. 2015) primary care research network to prospectively follow healthy children and their families throughout the pandemic. The objectives of the study are to describe the key epidemiological characteristics, risk factors and short- and longer-term outcomes of both COVID-19 infection and the effect of the COVID-19 pandemic on healthy children and families in Toronto, ON. TARGet Kids! is a collaboration of applied health researchers at The Hospital for Sick Children and St. Michael's Hospital, primary care providers from the Departments of Paediatrics and Family and Community Medicine at the University of Toronto and families. Children participating in TARGet Kids! are recruited from multiple primary care practices; anthropometric, dietary, lifestyle, developmental and biochemical data are collected at well-child visits from birth to adolescence. Families participating in the TARGet Kids! COVID-19 Study of Children and Families completed brief biweekly online questionnaires about family routines, mental health, public health preventive behaviours, COVID-19 infection and overall well-being.

The PACT

The Parent and Clinician Team (PACT) (Vanderhout et al. 2021) was formed in 2018, with support from the OSSU and the Canadian Institutes of Health Research Strategy for Patient-Oriented Research (CIHR SPOR) and serves as the core patient engagement team for all TARGet Kids! research. Parents of children participating in TARGet Kids! were invited to partner in the planning, conducting and sharing of research results. E-mail invitations to join the PACT were sent to all parents of the study's participants who had agreed to be contacted by e-mail. Typically, 10 to 12 parent partners attended semi-annual meetings, which were held in person until March 2020, after which meetings switched to a virtual format on Zoom. Parent partners were provided training via the Patient-Oriented Research Curriculum in Child Health Research (PORCCH n.d.) modules and compensated for their time according to the CIHR SPOR considerations when paying patient partners in research (CIHR 2019). With the support of an OSSU EMPOWER award, a part-time patient engagement coordinator with expertise in working with patients and families in research was hired to provide structure and coordination for patient engagement activities in the PACT and education for patient partners. This coordinator oversaw communication between study investigators and PACT members, conducted ongoing needs assessments, matched PACT members to ongoing tasks based on skills and interests and managed administrative duties.

Outcomes of Parent Engagement

Our OSSU EMPOWER award enabled us to sustain patient engagement we had established through the PACT in the COVID-19 Study of Children and Families. PACT members were invited to help co-develop the study during the protocol design stage, inform the study aims by voicing their suggestions and research interests, support study feasibility and recommend changes to enhance participation. They co-developed questionnaires and reviewed outcome measurement instruments, indicating what was relevant and appropriate to them. PACT members also provided key logistical and technical feedback about online versus paper questionnaires, which had been used in TARGet Kids! before the COVID-19 pandemic. PACT members volunteered to serve as patient-partner co-investigators on research funding applications. The frequency of virtual PACT meetings was increased to every other month. It was essential to achieve a balance of maintaining consistent communication, not overburden parents and allow for feedback to be rapidly incorporated into the study procedures and materials. Parent partners provided input to questionnaires, such as adding questions about mask use and vaccination, and modified questions about school and child care to suit current restrictions and routines. At each meeting, members were briefed on how their input from the previous meeting was incorporated into the study, and available findings were shared.

Parent partners were consistently willing to participate, possibly because they were curious about how COVID-19 would affect their families ...

Successes

Parent engagement in TARGet Kids! created an opportunity for parents to share their lived experience in the production of children's health research and communication of COVID-19 research findings with their communities (Vanderhout et al. 2020). PACT members were enthusiastic and promoted team building over Zoom, which was more accessible to parents with small children and resulted in better attendance than pre-pandemic meetings. New ways of contributing to meetings were introduced using the chat function in Zoom. Online collaborative documents were used to obtain feedback on questionnaires between meetings, which parents found quick and easy to use. Active participation appeared to be the most effective mode of engagement (i.e., meetings), but offering PACT members other forms of engagement (i.e., remote review of protocols, forms, questionnaires, etc.) allowed for flexibility and continuous feedback throughout the project.

Parent partners were consistently willing to participate, possibly because they were curious about how COVID-19 would affect their families, hopeful to express their concerns about the pandemic and interested in developing solutions to the challenges many children and families were facing. As one parent partner said, "It was an amazing outlet to have in such a stressful time."

Parent partners may have also felt a sense of security and connectedness, where their relationships with peer parents and researchers allowed them to keep abreast of emerging COVID-19 evidence and recommendations for children and families. Another parent said:

> I felt a sense of purpose and control I may not otherwise have, being part of the PACT. Knowing that we were in this together and that there was a group of researchers working to ensure we leveraged what we could to help our future selves really gave me a sense of control in a really uncertain time.

The evolution of the pandemic may have fostered ongoing creative thinking and input from PACT members as concerns and questions shifted over time from transitioning from school and childcare to absence of friends, family and regular activities to the introduction of vaccinations and variants of concern. Finally, PACT engagement during the pandemic strengthened parents' engagement in the TARGet Kids! study overall. By attending regular PACT meetings, parent partners were able to learn about opportunities to be involved in other studies in the cohort, which served to broaden parent engagement activities across a variety of ongoing projects.

Researchers' experiences of engagement were positive. Gaining an understanding of PACT members' experiences, concerns and priorities at different stages throughout the pandemic allowed the research team to direct resources, prioritize research questions and adjust the frequency and length of questionnaires families were asked to complete accordingly. Though it was evident that parents were facing multiple demands at home, their enthusiasm and willingness to contribute to research was unhindered. Though encouraged by this, the research team felt unprepared at times to provide sufficient opportunities for parents to contribute meaningfully in the midst of fast-paced funding calls, protocol development deadlines and changing public health guidelines.

Areas for Improvement

It was challenging to recruit new parent partners who represented the ethnic and sociodemographic diversity of the study's participants. TARGet Kids! participants' primary care visits during the pandemic were reduced and research assistants were removed from the clinics, limiting face-to-face interaction between the research team and parents. Increased social stress on families or a lack of resources among unengaged families, such as Internet access or knowledge about patient engagement in research, were other limitations. As a result, accessing hard-to-reach individuals was increasingly difficult. Other limitations of our study were the inability to recruit fathers and gender imbalance within the team. We have explored options such as "fathers only" meetings (as the group initially comprised only mothers), different types of engagement (document review or online surveys instead of meetings) and alternating parent attendance to PACT meetings. Due to the fast-paced nature of the pandemic and COVID-19 research, it was challenging to engage parent partners and solicit feedback before each step in the research process. While PACT members were consulted, they were not always actively involved in the research design stage owing to rushed timelines for COVID-19 grant applications. Active participation and collaboration would have been preferred had timelines been less rushed and if more preparation was possible. Parent partners were keen to be involved, but having the foresight to arrange this was not always possible. We also did not implement a formal evaluation process, which would have been helpful to understand the qualitative and quantitative impacts of parent engagement in this study.

Next Steps

PACT involvement in TARGet Kids! COVID-19 research has encouraged us to consider new research questions about COVID-19 vaccination and long-term implications of the pandemic for children and families, and parents are now involved in developing proposed methods for examining these key questions. As results become available from initial analyses of the TARGet Kids! COVID-19 Study of Children and Families, PACT members will be invited to co-present findings at webinars and virtual conferences and co-author publications. PACT members are currently helping to develop knowledge translation tools and knowledge mobilization strategies such as a new website and regular e-mail newsletters, which will be distributed to the PACT between meetings. TARGet Kids! research assistants are increasingly interacting with families virtually over Zoom, which is aimed at supporting relationships and opportunities for future engagement. Support from the OSSU EMPOWER award has allowed for training on patient engagement for research assistants, which is intended to facilitate recruitment of new PACT members who are representative of the entire cohort (i.e., from a variety of neighbourhoods and ethnic and racial backgrounds) as they return to in-person research roles. Recruitment of fathers is growing through a variety of approaches including alternating parents who attend meetings, peer referrals and targeted recruitment at TARGet Kids! participants' primary care visits. To promote more active participation of PACT members, we are leveraging support from the OSSU EMPOWER award to designate small teams of parent partners based on their interests and skills – for example, re-designing the website, creating new recruitment and retention strategies for TARGet Kids! and engaging in grant writing. Two PACT members who serve as champions for engaging fathers are leading innovative ideas for recruitment and creative involvement of fathers. Youth engagement in the PACT is also a priority for future development, where young people will help advise on research priorities and developing child-centred interventions. Going forward, we are considering different meeting formats that will best serve PACT members. Though in-person meetings can facilitate relationship building, virtual meetings have introduced a number of benefits to the structure and conduct of PACT collaboration. A mixed approach may be beneficial to continue to engage parents with varied preferences, resources and availability. Finally, we will ask PACT members to complete the Patient Engagement in Research Scale (Hamilton et al. 2021) to assess our approach to parent engagement in TARGet Kids! and develop strategies to address limitations we identify through this evaluation.

Conclusion

Engaging parents as partners in a study about COVID-19 in healthy children and families allowed family perspectives, priorities and insights to be incorporated into the design, conduct and dissemination of the research. Building on trusting relationships and maintaining consistent contact with parent partners allowed for dynamic and productive teamwork to suit the fast-paced nature of the COVID-19 pandemic. Parents' active engagement served to co-build the PACT in the TARGet Kids! cohort, which we foresee will support future child health research. Going forward, we are committed to increasing the diversity of parent partners and conducting evaluations that will improve our approach to patient engagement. HQ

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References

Canadian Institutes of Health Research (CIHR). 2019, September 15. Considerations When Paying Patient Partners in Research. Government of Canada. <https://cihr-irsc.gc.ca/e/51466.html>.

Carsley, S., C.M. Borkhoff, J.L. Maguire, C.S. Birken, M. Khovratovich, B. McCrindle et al. 2015. Cohort Profile: The Applied Research Group for Kids (TARGet Kids!). *International Journal of Epidemiology* 44(3): 776–88. doi:10.1093/ije/dyu123.

Domecq, J.P., G. Prutsky, T. Elraiyah, Z. Wang, M. Nabhan, N. Shippee et al. 2014. Patient Engagement in Research: A Systematic Review. *BMC Health Services Research* 14(1): 89. doi:10.1186/1472-6963-14-89.

Frank, L., L. Forsythe, L. Ellis, S. Schrandt, S. Sheridan, J. Gerson et al. 2015. Conceptual and Practical Foundations of Patient Engagement in Research at the Patient-Centered Outcomes Research Institute. *Quality of Life Research* 24(5): 1033–41. doi:10.1007/s11136-014-0893-3.

Hamilton, C.B., A.M. Hoens, A.M. McKinnon, S. McQuitty, K. English, L.D. Hawke and L.C. Li. 2021. Shortening and Validation of the Patient Engagement in Research Scale (PEIRS) for Measuring Meaningful Patient and Family Caregiver Engagement. *Health Expectations* 24(3): 863–79. doi:10.1111/hex.13227.

Kirwan, J.R., M. de Wit, L. Frank, K.L. Haywood, S. Salek, S. Brace-McDonnell et al. 2017. Emerging Guidelines for Patient Engagement in Research. *Value in Health* 20(3): 481–86. doi:10.1016/j.jval.2016.10.003.

PORCCH. n.d. Patient-Oriented Research Curriculum in Child Health. Retrieved August 10, 2021. <https://porcch.ca/>.

Vanderhout, S.M., C.S. Birken, P. Wong, S. Kelleher, S. Weir and J.L. Maguire. 2020. Family Perspectives of COVID-19 Research. *Research Involvement and Engagement* 6(1): 69. doi:10.1186/s40900-020-00242-1.

Vanderhout, S.M., C.S. Birken, M. Zaccaria Cho and J.L. Maguire. 2021. Forming a Parent and Clinician Team (PACT) in a Cohort of Healthy Children. *Research Involvement and Engagement* 7(1): 47. doi:10.1186/s40900-021-00293-y.

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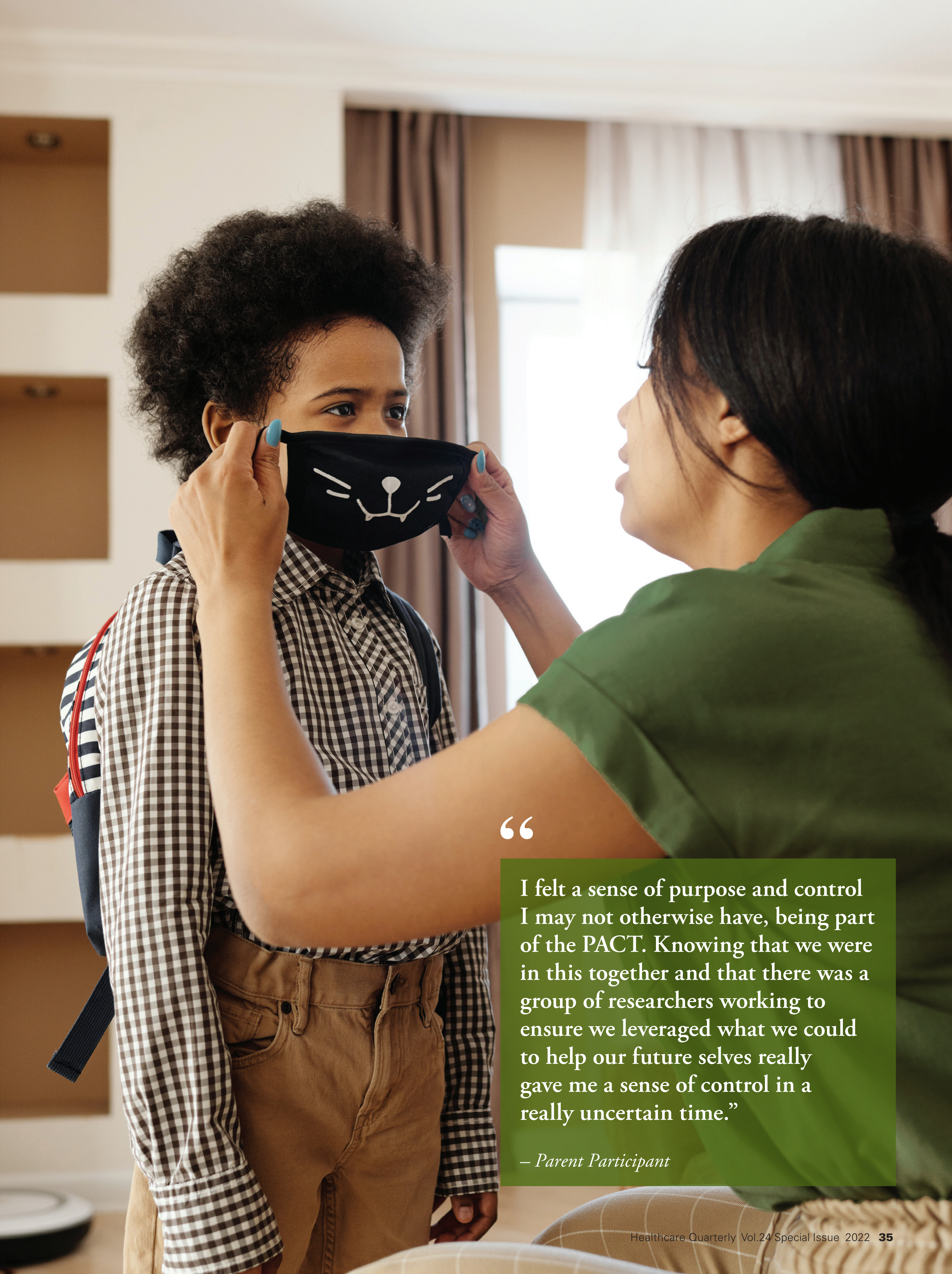


> “I felt a sense of purpose and control I may not otherwise have, being part of the PACT. Knowing that we were in this together and that there was a group of researchers working to ensure we leveraged what we could to help our future selves really gave me a sense of control in a really uncertain time.”
>
> *– Parent Participant*

> “Researchers found it valuable when patients provided first-hand experience and input as it validated their work and grounded it in real patient perspectives.”
>
> *– p. 47*

Building Capacity for Patient-Oriented Research: Utilizing Decision Aids to Translate Evidence into Practice, Policy and Outcomes

Monica Parry, Dawn P. Richards, David Wells^P^, Adhiyat Najam^P^, Salima Hemani and Susan Marlin on Behalf of the Patient-Oriented Research Decision Aids Investigative Team

Abstract

Background: The aim of this project was to engage with patient partners to translate knowledge about the decision aids and develop a scaling-up strategy for wider effects and reach.

Method: This project was guided by the World Health Organization and IDEAS (Integrate, Design, Assess and Share) frameworks for design thinking (e.g., ideating creative strategies), dissemination (e.g., sharing locally and widely) and scalability.

Results: We engaged 132 stakeholders in six webinars, had 321 total page views of the decision aids and conducted 16 interviews to determine revisions to the design of the decision aids before scalability.

Conclusion: Patient-partner collaborations assisted with design thinking, dissemination and scalability.

Key Points

- Commitment to research projects can be difficult. Patient partners need to feel safe enough to disclose the challenges they face, and research team members need to be respectful and responsive to the needs of the patient partner.
- Key stakeholders have collaborated to co-design innovative web-based open-access patient and investigator decision aids to support patient-oriented research (POR).
- Funding agencies should consider making POR training mandatory for all investigators and patient partners (e.g., decision aid completion) before making POR funding decisions.

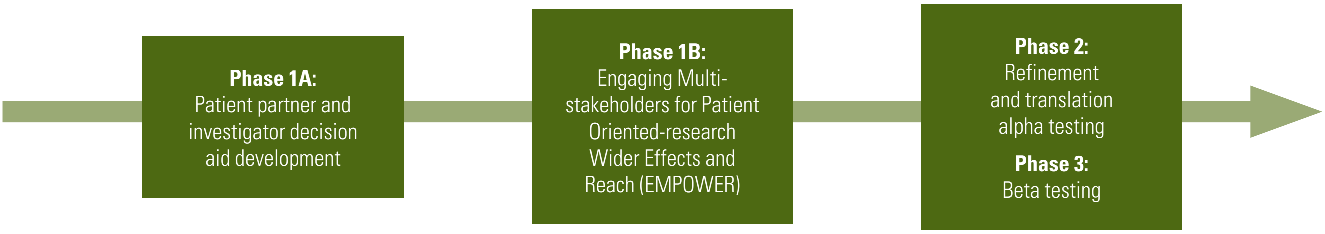
Introduction

Building partnerships, improving research quality and impact and developing best practices underpin values that impact patient engagement in research (Haywood et al. 2017). Patient partnerships aptly generate patient-reported outcome measures (Staniszewska et al. 2012), and the strongest predictor of patient engagement and partnership in research is investigators' attitudes (Cary et al. 2015). Guidelines for establishing research partnerships with patients suggest that the following are essential: (1) helpful organizational policies, (2) supportive investigator attitudes that are grounded in shared goals and strong communication practices, (3) principles of trust, respect and co-learning, (4) patient-oriented research (POR) training for all team members, (5) tools/resources for successful patient engagement and (6) value for patient partnerships across various stages of the research cycle (Kirwan et al. 2017).

This program of research consists of multiple phases guided by the Strategy for Patient-Oriented Research (SPOR) capacity development framework (CIHR 2015a) and the SPOR Patient Engagement Framework (CIHR 2015b). In Phase 1A (Figure 1a), two decision aids (patient and investigator) were developed to build capacity and increase engagement of patients with clinical trials (Parry et al. 2020). The decision aids are

P = Patient partner.

FIGURE 1a.
Study implementation plan



Phase 1A: Patient partner and investigator decision aid development

Phase 1B: Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER)

Phase 2: Refinement and translation alpha testing
Phase 3: Beta testing

FIGURE 1b.
Patient and investigator decision aid core functionalities

Introduction Get the facts

What Matters Your priorities

Learn More Resources

My Readiness Compare benefits/risks with what matters

My Decision Results and next steps

designed to help patients and investigators decide if they are ready to engage with each other on a research team. Each decision aid has five core functionalities: (1) *Introduction* (get the facts on POR), (2) *What Matters* (where patient partners can be engaged, including levels of engagement), (3) *Learn More* (resources to plan, engage and evaluate patient partnerships), (4) *My Readiness* (comparing personal priorities with perceived benefits and risks) and (5) *My Decision* (decisions and next steps, such as finding a patient partner or finding a research team) (Figure 1b).

Decision aids provide information for improving knowledge about potential expectations (Coulter et al. 2013), benefits and risks (Coulter et al. 2013) and options and outcomes (Elwyn et al. 2006). They assist users to gain skills in assessing uncertainties and help to clarify personal priorities about the benefits and risks of engaging in a decision (Elwyn et al. 2006; Volk and Coulter 2018). Most decision aids to date have been designed to provide support in making decisions about health treatments or screening, with evidence that indicates their usefulness in improving knowledge (Feldman-Stewart et al. 2013; Stacey et al. 2014, 2017) and offering choices about what matters most to patients (Stacey et al. 2014).

We collaborated with Clinical Trials Ontario (CTO) in Phase 1B to Engage Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) of Phase 1A of our research to impacts in Ontario. The aim of Phase 1B was to engage with patient partners to translate knowledge about the decision aids and develop a scaling-up strategy for wider effects and reach. We also aimed to use knowledge gained from Phase 1B to inform Phases 2 and 3 of this research program. POR principles and practices were used throughout Phase 1B and reported using the Guidance for Reporting Involvement of Patients and the Public–Short Form (GRIPP2–SF) (Staniszewska et al. 2017).

Materials and Methods

Patient partners were meaningfully engaged in all aspects of the research process as defined by the SPOR patient engagement framework (CIHR 2015a): (1) identifying research priorities, (2) designing the project for wider effects and reach, (3) developing the funded grant proposal, (4) co-designing and co-delivering all webinar presentations, (5) collecting, analyzing and interpreting interview data, (6) disseminating results through a presentation and a publication and (7) making recommendations for further refinements to the design of each decision aid. Patient partners were compensated for their level of engagement (i.e., commitment, responsibility and scope) based on recommendations provided by the SPOR Chronic Disease Networks and the Primary and Integrated Health Care Innovations (SPOR Networks in Chronic Diseases and the PICHI Network 2018). We engaged racially and ethnically diverse men and women as patient partners to ensure that knowledge translation and dissemination strategies for wider effects and reach were delivered through a lens of equity, diversity and inclusion (EDI).

The IDEAS (Integrate, Design, Assess and Share) (Mummah et al. 2016) and the World Health Organization's (WHO 2010) frameworks for disseminating and scaling up innovations were also used to inform our project. The IDEAS framework uses a flexible repetitive approach to design thinking (e.g., ideate creative strategies) and dissemination (e.g., share locally and widely), supporting a multi-stakeholder method for the development and evaluation of the decision aids. The WHO framework considers the elements (e.g., innovation, user organization, environment, and the resource team) and strategies (e.g., dissemination and advocacy, organizational process, costs/resources, monitoring/evaluation) of scalability and is guided by four principles: systems thinking, sustainability, defining scalability a priori and respect for gender, equity and human rights principles.

Data collection and analysis

Changes in study protocol and delays in projects due to the COVID-19 pandemic are reported per recommendations suggested by Perlis et al. (2021). The original project methods included an initial four-hour in-person brainstorming workshop with multi-stakeholders. *User organizations* defined by the WHO (2010) would have included organizations expected to adopt and implement the decision aids on a large scale. Intended outputs of the brainstorming workshop, aligned with the WHO (2010), included strategies for *dissemination and advocacy* (e.g., training, policy dialogues and briefs, cultivating champions), the *organization process* (e.g., identifying the number and type of "other" organizations for future scale-up), *costs/resource mobilization* (e.g., ensuring adequate budgetary allocation and linking future [beyond six months] scaling-up to other funding mechanisms) and *monitoring and evaluation* (e.g., a phone "hotline" and Google analytics). We then planned to offer three-hour face-to-face decision aid training webinars to interested user organizations identified in our brainstorming workshop. As a result of the pandemic, slight modifications to the study protocol included presenting one-hour synchronous and interactive online decision aid training webinars to interested organizations. Social media (i.e., Twitter) assisted with wider effects and reach. Instead of the four-hour brainstorming workshop, a series of interviews were planned with interested multi-stakeholders who attended the decision aid training webinars. A phone hotline was used to address questions, and Google analytics was used to track decision aid page views. Semi-structured interviews were intended to achieve similar outputs aligned with the WHO framework (WHO 2010) that would have been achieved in the initial brainstorming workshop. Interviews were not audiotaped but field notes were made. All data were anonymized and identified only by type of stakeholder. A thematic analysis informed the overall qualitative analytical framework (Braun et al. 2012). The original field notes were revisited regularly to ensure that codes and themes were grounded in the data (Kvale 1996).

Results

The aim of the Phase 1B EMPOWER project was to engage with patient partners to translate knowledge about the decision aids and develop a scale-up strategy for wider effects and reach. Additionally, knowledge gained from Phase 1B would inform Phases 2 and 3 of this research program. Two patient partners co-designed and co-delivered the webinars. They were given access to sufficient and appropriate resources to facilitate engagement (i.e., task/role definitions, training, appropriate time allocation and compensation). Patient partners also collaborated in the development of interview questions and assisted in analyzing and interpreting interview data. They were actively engaged in making priority decisions regarding revisions to the decision aid designs before scalability. Patient engagement details for Phase 1B using GRIPP2–SF are described in Table 1.

A total of 132 international multi-stakeholders engaged in six interactive webinars from June to November 2020; 59 attendees self-identified as having lived experience (i.e., patients), and the remainder were trainees (*n* = 9), investigators (*n* = 29), decision makers (*n* = 23) and individuals from charitable or patient organizations (*n* = 9) and SPOR SUPPORT Units (*n* = 3). A separate webinar was recorded and is available on the CTO YouTube channel (https://www.youtube.com/watch?v=H3XIchDrz2E), which received 109 views during the project. No calls were made to the hotline. Google analytics indicated a total of 321 decision aid page views (landing pages of both the patient and the investigator decision aids) from August to November 2020. The "Introduction" and "Learn More" pages were the most frequently accessed sections of both decision aids. A total of 16 interviews were completed with three policy makers, two investigators and 11 patient partners. Themes were aligned with the IDEAS framework (Mummah et al. 2016) and the WHO framework (WHO 2010) (Table 2). Overall, the feedback on design suggested that the decision aids were clear, logical and visually appealing. Suggestions for refinement (Phase 2) included larger font size; adding hyperlinks, glossary and bookmarks; enabling a certificate of completion or profile for sharing; adding culturally sensitive language, including videos/visuals from racial and ethnically diverse populations; and increasing availability through hard-copy versions (i.e., printable patient-partner decision aid).

TABLE 1.
Patient engagement in Phase 1B (GRIPP2–SF)

Section and topic	Item
1. Aim	To Engage Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) of Phase 1A of our research
2. Method	To ensure that principles of EDI were upheld, one patient partner (woman) was a co-principal investigator and three patient partners (two women, one man) were co-investigators. Gender and racial diversity in patient partnership helped to ensure that knowledge translation and dissemination strategies for wider effects and reach were delivered through an EDI lens. Patient partners engaged in all aspects of the research process: (1) identifying research priorities, (2) designing the project for wider effects and reach, (3) developing the grant proposal, (4) co-designing/co-delivering all the webinars, (5) collecting, analyzing and interpreting interview data, (6) disseminating results through a conference presentation/publication and (7) making recommendations for refinement of decision aids. Three patient partners contributed to editing this paper and are listed as co-authors/members of our Patient-Oriented Research Decision Aids Investigative Team. One of our patient partners (a woman) could not continue in the project due to certain life events.
3. Results	Two patient partners took the lead for co-designing and co-delivering all the webinars. These same patient partners had collaborated in Phase 1A so they were able to seamlessly explain the patient decision aid to webinar participants and answer questions. Patient partners collaborated in the development of interview questions and assisted in analyzing and interpreting interview data. Patient partners offered feedback to prioritize the integration of the refinements suggested to the design of the patient and investigator decision aids in the interviews. They were also instrumental in providing recommendations for sharing, dissemination and advocacy. Patient partners also led and co-delivered presentations to the Diabetes Research Excellence Cluster at the University of British Columbia and at the Canadian Women's Heart Health Alliance Virtual Summit.
4. Discussion	Patient collaboration in Phases 1A and 1B has been instrumental to the success of this project and the overall research program in building capacity for patient engagement in research. Although the decision aids were initially developed for use by patients and investigators engaging in clinical trials, ongoing engagement and dissemination strategies highlight the importance of the decision aids across all types of research designs. Patient partners co-authored the publication (Parry et al. 2020) detailing Phase 1A during the tenure of the Phase 1B project. Moreover, additional funding was secured from the Canadian Institutes of Health Research to make refinements to the decision aids based on feedback from Phase 1B – to translate both decision aids to French and conduct alpha (e.g., usability) and beta (e.g., field) testing of the decision aids (Figure 1).
5. Reflections	One challenge included the withdrawal of a patient partner due to time constraints and the extenuating circumstances caused by the COVID-19 pandemic. This experience emphasized the importance of maintaining awareness of patient-partner needs throughout the project. It was imperative that all POR team members recognized that both short- and long-term commitment to a project could be difficult for some patient partners. Empowering patient partners to feel safe to disclose challenges was essential to the success of this project. Investigators and other POR team members needed to maintain respect and be responsive to patient-partner needs. This challenge highlighted the value of engaging more than one patient partner on a POR team.

We created a PEP-CT Twitter account (@pep_ctTOOLS), shared the decision aids via presentations (the Canadian Women's Heart Health Alliance Virtual Summit and the Diabetes Research Excellence Cluster at the University of British Columbia [invited]) and a publication (Parry et al. 2020) and created a Policy Brief (Appendix 1, available online at www.longwoods.com/content/26776) for wider effects and reach.

Discussion

Funding from the Ontario SPOR SUPPORT Unit (OSSU) EMPOWER award grant allowed us to translate knowledge about the decision aids to at least 562 multi-stakeholders via webinars and web pages and develop a scale-up strategy for wider effects and reach using the IDEAS (Mummah et al. 2016) and WHO (2010) frameworks. Impacts of patient engagement in this project included (1) effective patient recruitment to webinars and interviews, (2) increased translation and dissemination of results through presentation and publication, (3) improved decision aid uptake in the community (i.e., decision aid page views), (4) informed patient and decision aid enhancements, (5) heightened POR knowledge dissemination to investigators and (6) evidence for a policy brief (Appendix 1). The web-based patient and investigator decision aids are the first to provide POR knowledge and decision support beyond the traditional aids used for health screening and/or treatment decisions (The WHO framework, the *Innovation*). Although our work focused on building capacity

TABLE 2.
Themes aligned with the IDEAS and WHO frameworks

IDEAS framework			
Integrate	**Design**	**Assess**	**Share**
WHO framework			**Dissemination and advocacy**
Race- and ethnicity-specific content	Acceptable literacy, videos/visuals to reflect EDI and translation to French	Detailed analytics	Marketing strategy (e.g., social media, conferences, meeting invitations, online discussions, publications, educational programs, etc.)
Larger font, less text	Hyperlinks/glossary and hard-copy versions (i.e., printable patient partner decision aids)	Access to decision aids across all populations, including those from racially and ethnically diverse people in Canada	National reach through patient-partner and non-profit organizations, industry, etc.
More visuals, videos and transitions (e.g., infographics)	Algorithms based on knowledge gaps. Visuals and videos will include racially and ethnically diverse populations in Canada (e.g., South Asians, black Canadians, etc.)	Access to decision aids on all devices (e.g., iPads)	Mandatory training modules for POR funding
Sample timelines for research projects	Bookmarks		Sustainable policy
Negotiating payments with organizations/ reimbursement strategies	Creating a shareable patient profile/ report/certificate		
Clear messaging and direction after *My Decision*	Hyperlinks to connect patient partners and investigators		
Investigator responsibilities for patient-partner training (e.g., screening, data extraction)			
Usefulness of tool for investigators already committed to POR (i.e., helpful information, checklists and links)			

for POR in clinical trials, the decision aids are applicable to patient partners and investigators involved in other demonstration and research projects. For example, in the "Learn More" resource section, the *Research Process* provides a launching point to learn more about engaging as/with patients in the *Design* of a research project, *Development of the Grant Proposal* and *Dissemination* of results.

We have secured further Canadian Institutes of Health Research funding to refine, translate and conduct alpha (usability) and beta (field) testing for each of the decision aids (Phases 2 and 3). Refinement and evaluation of the decision aids to improve patient partnerships will be guided by the International Patient Decision Aid Standards (Elwyn et al. 2006; Volk and Coulter 2018), user-centred design (Abras et al. 2004) and the Ottawa decision-support framework (The Ottawa Hospital 2015). The International Patient Decision Aid Standards provide an evidence framework for the content, development, implementation and evaluation of decision aids (Elwyn et al. 2006; Stacey et al. 2017). In addition, user-centred designs optimize end-user experience (Plaisance et al. 2018; Witteman et al. 2015) and web-based platforms facilitate development (Hoffman et al. 2014). Relevant design feedback from Phase 1B (i.e., including those prioritized by patient partners and investigators) will be incorporated into refinements of decision aids, and then each decision aid will be translated to French, refined through iterative cycles of usability testing with patients and investigators (Phase 2) and evaluated using a pragmatic pre–post pilot study (Phase 3).

Conclusion

Patient partners were actively engaged in translating knowledge about the decision aids and developing a scale-up strategy for wider effects and reach. The IDEAS framework provided a flexible repetitive approach to design thinking (e.g., ideating creative strategies) and dissemination. The WHO framework helped us identify strategies to increase decision aid impacts for research and policy through a gender, equity and human rights lens – for example, use of the decision aids (1) for all research designs (i.e., not only for clinical trials), (2) as a repository of essential POR and sex/gender information for new and experienced patient partners and investigators and (3) for mandatory online training for POR funding applications. Patient engagement in this project and in this research program is a necessity – patients are the heart of SPOR (CIHR 2015b) and our team's patient partners were actively engaged in Phase 1A and continued to collaborate and co-lead Phase 1B. The decision aids will contribute to Canada's strategy for POR to support the collaborative efforts of patients and investigators in building a sustainable, accessible and equitable healthcare system. HQ

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References

Abras, C., D. Maloney-Krichmar and J. Preece. 2004. User-Centered Design. In W. Bainbridge, Ed., *The Encyclopedia of Human-Computer Interaction* (pp. 1–14). Sage Publications.

Braun, V. and V. Clarke. 2012. Thematic Analysis. In H. Cooper, P.M. Camic, D.L. Long, A.T. Panter, D. Rindskopf and K.J. Sher, Eds., *APA Handbook of Research Methods in Psychology, Vol. 2. Research Designs: Quantitative, Qualitative, Neuropsychological, and Biological* (pp. 57–71). American Psychological Association.

Canadian Institutes of Health Research (CIHR). 2015a. Capacity development framework. Strategy for Patient-Oriented Research Capacity Development Framework. Retrieved July 23, 2019. <http://www.cihr-irsc.gc.ca/e/49307.html>.

Canadian Institutes of Health Research (CIHR). 2015b. Strategy for Patient-Oriented Research – Patient Engagement Framework. Retrieved July 23, 2019. <http://www.cihr-irsc.gc.ca/e/48413.html>.

Cary, M.S., J.D. Rubright, J.D. Grill and J. Karlawish. 2015. Why Are Spousal Caregivers More Prevalent than Nonspousal Caregivers as Study Partners in AD Dementia Clinical Trials? *Alzheimer Disease and Associated Disorders* 29(1): 70–74. doi:10.1097/WAD.0000000000000047.

Coulter, A., D. Stilwell, J. Kryworuchko, P.D. Mullen, C.J. Ng and T. van der Weijden. 2013. A Systematic Development Process for Patient Decision Aids. *BMC Medical Informatics and Decision Making* 13(Suppl 2): S2. doi:10.1186/1472-6947-13-S2-S2.

Elwyn, G., A. O'Connor, D. Stacey, R. Volk, A. Edwards, A. Coulter et al. 2006. Developing a Quality Criteria Framework for Patient Decision Aids: Online International Delphi Consensus Process. *BMJ* 333(7565): 417. doi:10.1136/bmj.38926.629329.AE.

Feldman-Stewart, D., M. O'Brien, M.L. Clayman, B.J. Davison, M. Jimbo, M. Labrecque et al. 2013. Providing Information about Options in Patient Decision Aids. *BMC Medical Informatics and Decision Making* 13(2): S4. doi:10.1186/1472-6947-13-S2-S4.

Haywood, K., A. Lyddiatt, S.J. Brace-McDonnell, S. Staniszewska and S. Salek. 2017. Establishing the Values for Patient Engagement (PE) in Health-Related Quality of Life (HQoL) Research: An International, Multiple-Stakeholder Perspective. *Quality of Life Research* 26(6): 1393–404. doi:10.1007/s11136-016-1465-5.

Hoffman, A.S., H.A. Llewellyn-Thomas, A.N.A. Tosteson, A.M. O'Connor, R.J. Volk, I.M. Tomek et al. 2014. Launching a Virtual Decision Lab: Development and Field-Testing of a Web-Based Patient Decision Support Research Platform. *BMC Medical Informatics and Decision Making* 14: 112. doi:10.1186/s12911-014-0112-8.

Kirwan, J.R., M. de Wit, L. Frank, K.L. Haywood, S. Salek, S. Brace-McDonnell et al. 2017. Emerging Guidelines for Patient Engagement in Research. *Value in Health* 20(3): 481–86. doi:10.1016/j.jval.2016.10.003.

Kvale, S. 1996. *Interviews: An Introduction to Qualitative Research Interviewing*. Sage Publications.

Mummah, S.A., T.N. Robinson, A.C. King, C.D. Gardner and S. Sutton. 2016. IDEAS (Integrate, Design, Assess, and Share): A Framework and Toolkit of Strategies for the Development of More Effective Digital Interventions to Change Health Behavior. *Journal of Medical Internet Research* 18(12): e317. doi:10.2196/jmir.5927.

The Ottawa Hospital. 2015. Ottawa Decision Support Framework (ODSF). Retrieved July 23, 2019. <https://decisionaid.ohri.ca/odsf.html>.

Parry, M., A.K. Bjørnnes, K. Toupin-April, A. Najam, D. Wells, A. Sivakumar et al. 2020. Patient Engagement Partnerships in Clinical Trials: Development of Patient Partner and Investigator Decision Aids. *Patient* 13(6): 745–56. doi:10.1007/s40271-020-00460-5.

Perlis, R.H., S.J.P.A. Haneuse, G.D. Rubenfeld, S.D. Fihn and F.P. Rivara. 2021. Reporting Clinical Studies Affected by the COVID-19 Pandemic: Guidelines for Authors. *JAMA Network Open* 4(1): e2036155. doi:10.1001/jamanetworkopen.2020.36155.

Plaisance, A., H.O. Witteman, A. LeBlanc, J. Kryworuchko, D.K. Heyland, M.H. Ebell et al. 2018. Development of a Decision Aid for Cardiopulmonary Resuscitation and Invasive Mechanical Ventilation in the Intensive Care Unit Employing User-Centered Design and a Wiki Platform for Rapid Prototyping. *PLoS ONE* 13(2): e0191844. doi:10.1371/journal.pone.0191844.

SPOR Networks in Chronic Diseases and the PICHI Network. 2018. *Recommendations on Patient Engagement Compensation*. Retrieved July 23, 2019. <https://diabetesaction.ca/wp-content/uploads/2018/07/TASK-FORCE-IN-PATIENT-ENGAGEMENT-COMPENSATION-REPORT_FINAL-1.pdf>.

Stacey, D., F. Légaré, N.F. Col, C.L. Bennett, M.J. Barry, K.B. Eden et al. 2014. Decision Aid for People Facing Health Treatment or Screening Decisions. *Cochrane Database of Systematic Reviews* 28(1): CD001431. doi:10.1002/14651858.CD001431.pub4.

Stacey, D., F. Légaré, K. Lewis, M.J. Barry, C.L. Bennett, K.B. Eden et al. 2017. Decision Aids for People Facing Health Treatment or Screening Decisions. *Cochrane Database of Systematic Reviews* 4(4): CD001431. doi:10.1002/14651858.CD001431.pub5.

Staniszewska, S., J. Brett, I. Simera, K. Seers, C. Mockford, S. Goodland et al. 2017. GRIPP2 Reporting Checklists: Tools to Improve Reporting of Patient and Public Involvement in Research. *BMJ* 358: j3453. doi:10.1136/bmj.j3453.

Staniszewska, S., K.L. Haywood, J. Brett and L. Tutton. 2012. Patient and Public Involvement in Patient-Reported Outcome Measures: Evolution Not Revolution. *Patient* 5(2): 79–87. doi:10.2165/11597150-000000000-00000.

Volk, R.J. and A. Coulter. 2018. Advancing the Science of Patient Decision Aids through Reporting Guidelines. *BMJ Quality and Safety* 27(5): 337–39. doi:10.1136/bmjqs-2017-007657.

Witteman, H.O., S.C. Dansokho, H. Colquhoun, A. Coulter, M. Dugas, A. Fagerlin et al. 2015. User-Centered Design and the Development of Patient Decision Aids: Protocol for a Systematic Review. *Systematic Reviews* 4(1): 11. doi:10.1186/2046-4053-4-11.

World Health Organization (WHO). 2010. Nine Steps for Developing a Scaling-Up Strategy. Retrieved July 23, 2019. <https://www.who.int/reproductivehealth/publications/strategic_approach/9789241500319/en/>.

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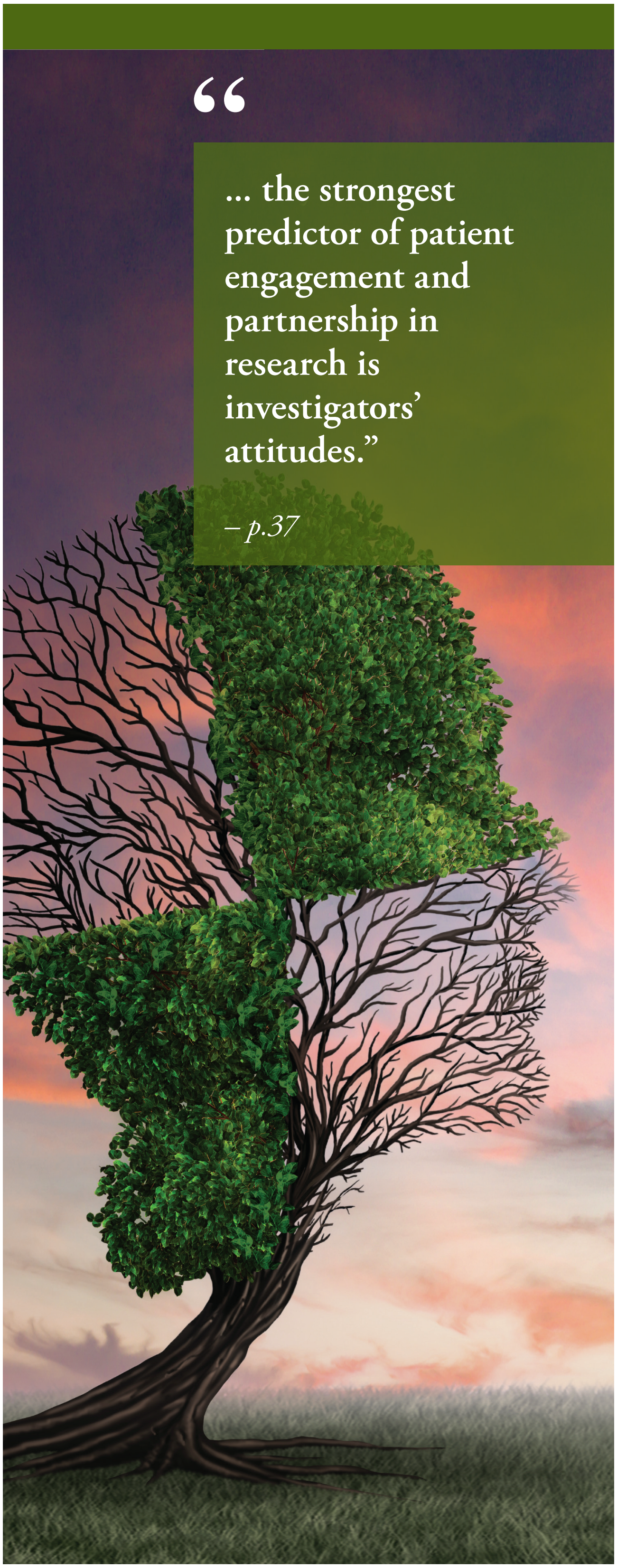
The **Patient-Oriented Research Decision Aids Investigative Team** is composed of senior scientists/investigators (**Anne Ellis**, **Ian Gilron**), mid-career scientists/investigators (**Monica Parry**, **Dawn P. Richards**, **Karine Toupin**), early career scientists/investigators (**Ann Kristin Bjørnnes**), decision makers (**Susan Marlin**), trainees (**Salima Hemani**) and patient partners (**David Wells**, **Adhiyat Najam**, **Tina Ceroni**) who are actively engaged in providing scientific and practical knowledge across all phases of this research program.



> “… the strongest predictor of patient engagement and partnership in research is investigators’ attitudes.”
>
> *– p.37*

Evaluation of Experiences and Impact of Patient Engagement on e-Health Research: A Qualitative Study

Rachel Y. Pan*, Kendra Zhang*, Arani Sivakumar, Dorothy Choi, Angel H. Wang, Pauline Wijeyesekera^P and Catherine H. Yu

Abstract

Background: Health technology has increasingly moved toward adopting a "user-centred design" approach to include the user/patient throughout the innovation and design process; however, few studies have evaluated the patient's experience of such an engagement.

Objective: The aim of this study was to explore the role of patient engagement (PE) within e-health innovation research.

Method: Using qualitative descriptive methodology, semi-structured interviews were conducted with eight participants (patient partners and research/development team members).

Findings: Key themes were centred on enablers of, challenges to and methods of improving PE.

Conclusion: PE must be prioritized from study conception, explicitly programmed into study conduct and valued by integrating patient partner input.

Key Points

- This study provided a multi-stakeholder perspective on patient engagement (PE), revealing that stakeholders' perspectives differed and must be considered separately when planning for engagement.
- This study identified strategies to improve PE during the development of an e-health tool, including providing support for patients throughout the process, ensuring thorough team communication, making patients feel valued for their contributions and fostering a positive research environment.
- Overall, PE helped ensure the relevance, usability and appropriateness of an e-health tool for different patient demographics; however, the process of PE should pay special attention to recruiting representative patient populations.

Introduction

Patient engagement (PE) is a meaningful and active collaboration in governance, disease diagnosis and management, priority setting, conducting research and knowledge translation (CIHR 2014). With the shift away from paternalism in healthcare, PE in health research has increasingly become a key research priority in Canada (Manafo et al. 2018). E-health is a category within digital health that denotes the wide range of information, expertise and communication technologies that support health-related fields, including health surveillance, education and healthcare delivery (Baker et al. 2014). Despite its many potential benefits to patients, existing e-health services are largely technology-driven rather than being personalized and user-centred, resulting in tools that fail to address individualized patient goals and that, thus, cannot be widely adopted (Cowie et al. 2013). In response, *MyDiabetesPlan* (*MDP*), a web-based shared decision making and priority-setting patient decision aid, was developed by engaging patients in the research process of e-health tool development in order to create a more user-centred e-health toolkit (Yu et al. 2014).

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* Lead co-authors.

P = Patient, caregiver and knowledge user.

There is a lack of investigation into PE within e-health, which, in today's digital climate, is a deficiency (Leung et al. 2019). Furthermore, few studies have explored patients' perspectives of and experiences in such an engagement. Existing studies tend to leave research topics unspecified and predominantly report only on academic researchers' experiences (Bhati et al. 2020). Moreover, PE within research has only recently garnered attention in North America and warrants findings specific to local populations (Bhati et al. 2020). With the role of patient partners (PPs) in improving research feasibility, acceptability, rigor and relevance, this Engage Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Award project explored the experiences and perspectives of the PPs and researchers from the initial research project to guide PE in the future (Forsythe et al. 2019).

The purpose of this study was to explore the role of PE within Canadian e-health innovation research using our experience with *MDP* as a case study. We aimed to identify how patients were engaged, the impact and benefits of this engagement, challenges that were faced and how PE can be more effective moving forward. The PE in research framework was used to guide this line of inquiry (Hamilton et al. 2018). By involving both PPs and research team members, we obtained a multi-stakeholder perspective.

Method

Our previous work developed and trialled *MDP* in a multicentred randomized controlled trial (Yu et al. 2020). Recently, through the EMPOWER grant, we explored patients' and clinicians' experiences regarding its integration into clinical care and interprofessional stakeholder involvement to assess the implementation potential of *MDP*. Thus, our study acts as an extension of the broader *MDP* project (Sivakumar et al. 2021). PPs were consulted frequently as members of the research team throughout all phases of the research program, from innovation ideation, development and testing to implementation. In the development of this evaluation of PE study, one of the authors (PW) was consulted to arrive at the most relevant research question from a PP's perspective.

We used qualitative descriptive methodology as it offers an in-depth understanding of participants' experiences and impact of PE. We adhered to the Guidance for Reporting Involvement of Patients and the Public–Long Form (GRIPP2–LF) reporting checklist (Staniszewska et al. 2017).

To date, *MDP* has involved a multi-disciplinary team that consisted of PPs ($n = 3$), software development team members ($n = 2$) and research team members ($n = 13$). In the EMPOWER study, these same members in addition to Ontario Ministry of Health participants were end-user participants. In order to gain the experiences of all those who worked on *MDP*, recruitment occurred by contacting PPs and team members through e-mail and obtaining informed consent.

Data were obtained through semi-structured interviews. The interview guides (one each for PPs and research team members) explored experiences with PE in *MDP*, challenges that were faced and how PE in research can be more effective moving forward, the contents of which were based on the GRIPP2 framework. Interviews were conducted virtually via Zoom due to the COVID-19 pandemic by research team members trained in qualitative interviewing (RP and KZ). All interviews were audiotaped, transcribed verbatim and annotated. Content analysis was carried out in an iterative fashion alongside data collection (Elo and Kyngäs 2008; Korstjens and Moser 2018).

Results

We interviewed eight participants (Table 1).

We describe participants' views on ways to sustain PE and the impacts, enablers and hindrances of PE. See Tables 2 and 3 for representative quotes (available online at www.longwoods.com/content/26775).

TABLE 1.
Participants

Stakeholder group	Participant
PPs	Patient living with type 1 diabetes, has been a PP from October 2015 to present. Patient living with type 2 diabetes, has been a PP from February 2016 to present. Patient living with type 2 diabetes, has been a PP from December 2017 to present.
Research team members	Physician and site lead Physician and site lead Registered nurse Physician Research coordinator/assistant
Development team member	Software developer

PPs' experiences and perspectives

Ways in which meaningful PE was sustained

PPs felt valued and "listened to" through integration into the entirety of the research process. They appreciated providing input for, testing and evaluating the e-health tool and being consulted on the selection of study outcomes and data collection tools. This was enabled by frequent meetings with the research team.

They reported that their sentiment of feeling respected, valued and able to contribute (despite initially thinking that their own non-medical backgrounds would be a limitation) helped sustain PE. They felt empowered to apply their unique skills, coming from different cultural and ethnic backgrounds, thus contributing to a collaborative team environment. They also identified the creation of opportunities that demonstrated the value of PE; for example, PPs were given the opportunity to present at conferences alongside research team members.

Positive and negative results of PE and its impacts on the research and individuals involved

PE provided first-hand patient perspectives and experiences, which helped ensure the tool's relevance to patients and guided the direction of the research. PPs helped refine the tool's user interface and ensured patient understandability of the tool; they also helped contextualize the tool in terms of language barriers, generational gaps and cultural nuances. PPs themselves were able to get the perspective and support of other individuals with diabetes while learning more about the condition. Furthermore, learning about the time and effort that goes into conducting research was paradigm-shifting for the PPs, and they developed a greater appreciation for the process. PPs felt empowered and confident by the act of providing their perspectives and creating impactful change, and experienced fulfillment from long-term involvement with the research project.

Contextual and research process factors that enabled the impact of PE

PPs' prior interest in diabetes-related topics and their motivation to self-educate was a contextual factor that spurred their engagement. In terms of the research process, PPs felt comfortable and confident voicing their concerns in the team environment and were supported if needed. Having differing contributions be valued helped create a positive experience for the PPs and, in particular, a trusting relationship with team members before and throughout engagement further promoted the same.

Contextual and research process factors that hindered the impact of PE

PPs noted that their comprehension and insecurity about what they bring to the table were potential hindrances to full engagement. A contextual challenge was encountered by patients who spoke English as a second language, which at times acted as a language barrier. Similarly, PPs were sometimes challenged and had difficulty understanding everything discussed at meetings, especially medical terminology. Regarding the research process, PPs found it difficult to maintain motivation during periods when there was no communication from the researchers.

A contextual challenge was encountered by patients who spoke English as a second language, which at times acted as a language barrier.

Researchers' perspectives and experiences

Ways in which meaningful PE was sustained

Throughout the research process, researchers appreciated that PPs were engaged consistently alongside research members. Researchers also valued PPs as the main "experts" on diabetes, and their personal experiences and inputs were valued equally to inputs by other research members.

Positive and negative results of PE and its impacts on the research and individuals involved

The first notable impact patients made was providing diverse perspectives and experiences. These insights not only helped to identify discrepancies between researchers' assumptions about patient knowledge and opinions and the reality for patients; these insights also would sometimes change the direction of the research itself. Specifically, in considering the development of an e-health innovation, patients played a critical role in ensuring the relevance, practicality and usability of the tool, as well as other patient-facing research materials, for patients. Researchers particularly appreciated that patient input improved the efficiency of the research. However, researchers noted that a somewhat negative impact of PE included increasing the amount of time needed to conduct research and also that it was important to take this additional time to ensure that the final outcome was appropriate and justified.

Contextual and research process factors that enabled the impact of PE

Firstly, institutional support and resource allocation to PE was a facilitator to *MDP* as there was a large amount of support, but researchers noted that it was a challenge within the research community, overall. Additionally, researchers noted that relationship building between the research team and the PPs both before and during the project was critical to fostering engagement and successful partnership. Similarly, the effort that researchers took to speak the same "language" as patients was identified as an enabler of PE.

Contextual and research process factors that hindered the impact of PE engagement

Researchers expressed several challenges to PE. Ensuring that PPs constantly feel valued and engaged was identified as a challenge; however, within the context of *MDP*, researchers felt that PPs were enthusiastic and dedicated. More broadly, researchers held concerns regarding the process of PE, which included the inherent power differential between PPs and research team members and how our sample of PPs may not be representative of the whole patient population of interest.

Discussion

Prior studies have shown that the usability, understandability and relevance that PPs bring to e-health tools lead to increased uptake of new technologies (Lupton 2013).

Our PPs reported a predominantly positive experience with PE, which was attributed to feeling valued by the research team, finding a community of other diabetes patients, learning about diabetes and feeling empowered about disease management. Many of these positive findings are consistent with a study by Forsythe et al. (2019), which analyzed PE across 126 studies that described the contribution(s) of engagement to their project; however, our study is one of the few that investigated challenges and enablers expressed by PPs themselves specifically within the context of e-health. Age, language, cultural background and prior knowledge impacted the ease with which patients could engage with the research and the tool being developed. The PPs' motivation to self-educate and the research team's supportiveness facilitated their engagement; however, an area of improvement was the need for increasing the regularity of communication with patients.

Researchers found it valuable when patients provided first-hand experience and input as it validated their work and grounded it in real patient perspectives. Their feedback on experiencing the disease, financial constraints and lack of knowledge, education and accessibility to family physicians was insightful. Researchers found that *MDP* engaged patients meaningfully, with patients being extensively involved throughout the research process. Within the literature, one of the concerns that researchers have for PE is its potential tokenistic usage of patients (Carroll et al. 2017); our findings demonstrate that treating patients as an equal member of the team and a previous positive relationship with a core research member may mitigate this concern. Other facilitators to engagement included increased institutional support and using accessible language. Researchers emphasized the challenge of PPs not being representative of the whole population of interest, which is consistent with the literature (Carroll et al. 2017). Although our PPs had different cultural backgrounds, there should be more measures to involve a wide range of patient populations and uphold equity, diversity and inclusion. A centralized process for patient recruitment and engagement may help address this; however, the difficulty lies in doing so in a meaningful and ethical way (Bishop et al. 2018).

Limitations included our limited sample size, which may not be representative of the diabetes patient population or other e-health research teams.

Conclusion

The value-add of PE was its role in creating an e-health tool that is indeed beneficial and sensitive to end-users' needs. Our work suggests the need for more centralized processes for PE that also involves more diverse patient populations while ensuring adequate relationship building between the research teams and partners; strategies to improve PE during the development of an e-health tool include providing support for patients throughout the process, ensuring thorough team communication, making patients feel valued for their contributions and fostering a positive research environment. HQ

References

Baker, T.B., D.H. Gustafson and D. Shah. 2014. How Can Research Keep Up with eHealth? Ten Strategies for Increasing the Timeliness and Usefulness of eHealth Research. *Journal of Medical Internet Research* 16(2): e36. doi:10.2196/jmir.2925.

Bhati, D.K., M. Fitzgerald, C. Kendall and S. Dahrouge. 2020. Patients' Engagement in Primary Care Research: A Case Study in a Canadian Context. *Research Involvement and Engagement* 6(1): 65. doi:10.1186/s40900-020-00238-x.

Bishop, A.C., M.J. Elliott and C. Cassidy. 2018. Moving Patient-Oriented Research Forward: Thoughts from the Next Generation of Knowledge Translation Researchers. *Research Involvement and Engagement* 4(1): 23. doi:10.1186/s40900-018-0110-6.

Canadian Institutes of Health Research (CIHR). 2014. *Strategy for Patient-Oriented Research – Patient Engagement Framework*. Government of Canada. Retrieved February 28, 2022. <https://cihr-irsc.gc.ca/e/documents/spor_framework-en.pdf>.

Carroll, S.L., G. Embuldeniya, J. Abelson, M. McGillion, A. Berkesse and J.S. Healey. 2017. Questioning Patient Engagement: Research Scientists' Perceptions of the Challenges of Patient Engagement in a Cardiovascular Research Network. *Patient Preference and Adherence* 11: 1573–83. doi:10.2147/PPA.S135457.

Cowie, M.R., C.E. Chronaki and P. Vardas. 2013. E-Health Innovation: Time for Engagement with the Cardiology Community. *European Heart Journal* 34(25): 1864–68. doi:10.1093/eurheartj/ehs153.

Elo, S. and H. Kyngäs. 2008. The Qualitative Content Analysis Process. *Journal of Advanced Nursing* 62(1): 107–15. doi:10.1111/j.1365-2648.2007.04569.x.

Forsythe, L.P., K.L. Carman, V. Szydlowski, L. Fayish, L. Davidson, D.H. Hickam et al. 2019. Patient Engagement in Research: Early Findings from the Patient-Centered Outcomes Research Institute. *Health Affairs* 38(3): 359–67. doi:10.1377/hlthaff.2018.05067.

Hamilton, C.B., A.M. Hoens, S. McQuitty, A.M. McKinnon, K. English, C.L. Backman et al. 2018. Development and Pre-Testing of the Patient Engagement in Research Scale (PEIRS) to Assess the Quality of Engagement from a Patient Perspective. *PLoS ONE* 13(11): e0206588. doi:10.1371/journal.pone.0206588.

Korstjens, I. and A. Moser. 2018. Series: Practical Guidance to Qualitative Research. Part 4: Trustworthiness and Publishing. *European Journal of General Practice* 24(1): 120–24. doi:10.1080/13814788.2017.1375092.

Leung, K., D. Lu-McLean, C. Kuziemsky, R.G. Booth, S.C. Rossetti, E. Borycki et al. 2019. Using Patient and Family Engagement Strategies to Improve Outcomes of Health Information Technology Initiatives: Scoping Review. *Journal of Medical Internet Research* 21(10): e14683. doi:10.2196/14683.

Lupton, D. 2013. The Digitally Engaged Patient: Self-Monitoring and Self-Care in the Digital Health Era. *Social Theory & Health* 11(3): 256–70. doi:10.1057/sth.2013.10.

Manafo, E., L. Petermann, P. Mason-Lai and V. Vandall-Walker. 2018. Patient Engagement in Canada: A Scoping Review of The 'How' and 'What' of Patient Engagement in Health Research. *Health Research Policy and Systems* 16(1). doi:10.1186/s12961-018-0282-4.

Sivakumar, A., R.Y. Pan, D. Choi, A. He Wang and C.H. Yu. 2021. *"If We Got a Win-Win, You Can Sell It to Everybody"*: A Qualitative Study Employing Normalization Process Theory to Identify Critical Factors for eHealth Implementation and Scale-up in Primary Care [Epub]. *Canadian Journal of Diabetes*. doi:10.1016/j.jcjd.2021.08.005.

Staniszewska, S., J. Brett, I. Simera, K. Seers, C. Mockford, S. Goodlad et al. 2017. GRIPP2 Reporting Checklists: Tools to Improve Reporting of Patient and Public Involvement in Research. *BMJ* 358: j3453. doi:10.1136/bmj.j3453.

Yu, C., D. Choi, B.A. Bruno, K.E. Thorpe, S.E. Straus, P. Cantarutti et al. 2020. Impact of MyDiabetesPlan, a Web-Based Patient Decision Aid on Decisional Conflict, Diabetes Distress, Quality of Life, and Chronic Illness Care in Patients with Diabetes: Cluster Randomized Controlled Trial. *Journal of Medical Internet Research* 22(9): e16984. doi:10.2196/16984.

Yu, C.H., D. Stacey, J. Sale, S. Hall, D.M. Kaplan, N. Ivers et al. 2014. Designing and Evaluating an Interprofessional Shared Decision-Making and Goal-Setting Decision Aid for Patients with Diabetes in Clinical Care – Systematic Decision Aid Development and Study Protocol. *Implementation Science* 9(1): 16. doi:10.1186/1748-5908-9-16.

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In Pursuit of Better Care Transitions: Lessons Learned from a Co-Designed Project

Kerry Kuluski, Ida McLaughlin[P], Lisa Bennett[P], Gordon MacGregor[P], Lucy Bilotta[P], Bernadette Farrell[P], Murray Powell[P] and Monika Syed[P]

Abstract

In this commentary, we reflect on our experience of co-designing an intervention to address challenges due to delayed hospital discharge (known as alternate level of care in Canada). Through a series of focus groups and co-design sessions, we identified common challenges with delayed discharge (including a lack of services while waiting for discharge and poor communication with the care team). In co-designing service improvements, we (1) amplified the voices of patients and caregivers, which helped them feel unified in their experience and (2) developed tools that aim to improve patient, caregiver and provider experiences. In this commentary, we reflect on these impacts along with the key lessons learned.

Key Points

- We co-created a strategy (i.e., components of an intervention) to address challenges with delayed hospital discharge (a care quality issue experienced by health systems worldwide).
- Starting at the very beginning of the project, shared leadership (i.e., shared power) among all stakeholders was essential to create a safe space to open up.
- Projects can lose momentum if participants do not stay connected to people (i.e., decision makers) who have the power to make the change required for the co-designed activity/intervention to be adopted and implemented.

Background

On June 11, 2018, a group of mostly strangers (a researcher, a patient and caregivers) met in the governor's tea room of the historic Don Jail (now Bridgepoint Hospital's administrative building) for the first time. Our group came together through a common interest: improving delayed hospital care transitions (referred to as alternate level of care [ALC] in Canada). Three years later, with an additional patient member, we are still going and eager to share our journey through this commentary.

Part One: The Evolution, Purpose and Impact of Our Work Together

It started over coffee (lead author's experience)

As a researcher fairly new to the patient engagement space, meeting my first caregiver partners was akin to blind dating. We met over coffee in a neutral, safe and comfortable atmosphere. They chose the place, and I met them there. These coffee chats were an effective way to meet, share and assess the next steps. Other patients/caregivers who would eventually join

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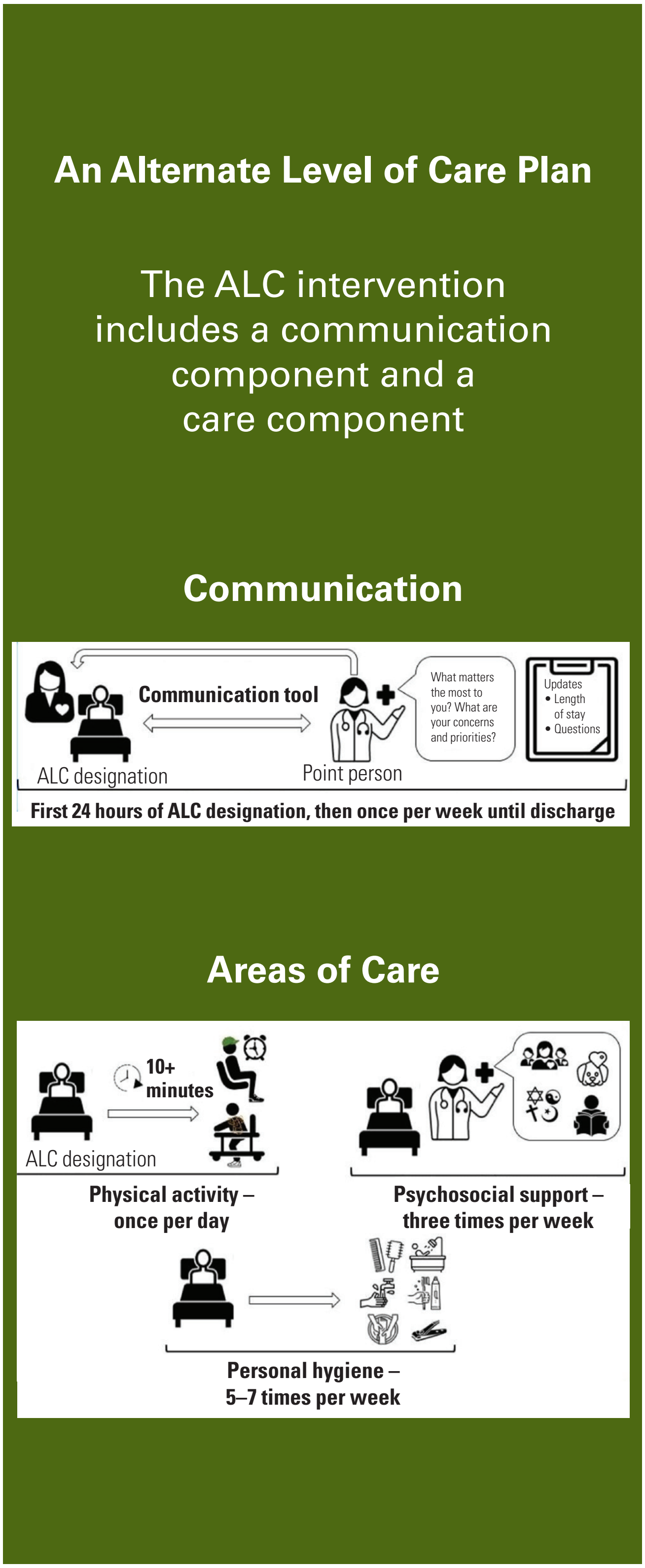
P = Patient or caregiver partner.

our council/team were already members of patient and family advisory councils at other hospitals. They responded to flyers shared in hospitals by members of a regional ALC task force (an initiative led by health system leaders who recognized that we needed to hear the voices of patients and caregivers). After a few council meetings, we identified the need to find another patient partner, and a hospital social worker introduced us to a current ALC patient who joined our team.

FIGURE 1.
Components of the ALC Intervention



We focused on improving delayed care transitions (i.e., ALC)

All patients and caregivers on our team had experienced a delayed care transition and wanted to see systemic changes. Care transition refers to moving from one sector of the healthcare system to another (i.e., hospital to home or to long-term care [LTC]). Sometimes, this transition in care is delayed (Walker 2011), resulting in negative personal and system-level outcomes. Transition delays (i.e., ALC) are experienced in hospitals worldwide (Amy et al. 2012; Challis et al. 2014; Costa and Hirdes 2010; Costa et al. 2012; Gaughan et al. 2017; McCloskey et al. 2015; Rojas-Garcia et al. 2018; Sutherland and Crump 2013; Tan et al. 2010). Across Canada, the number of beds occupied by ALC patients exceeds 13% almost daily (Sutherland and Crump 2013). ALC patients and their caregivers report a drop (or absence) of care and little communication about next steps. Patients experience functional decline (Bender and Holyoke 2018; Kortebein et al. 2007; McCloskey et al. 2014; Swinkels and Mitchell 2009; Wilson et al. 2014), patients and their loved ones experience confusion and stress (Cressman et al. 2013; Everall et al. 2019; Kuluski et al. 2017) and resources are not optimized (Burr and Dickau 2017; Ministry of Health and Long-Term Care 2019; Sutherland and Crump 2013; Walker 2011). Although the number of patients with an ALC status decreased during the first wave of the COVID-19 pandemic, ALC rates have fluctuated over time and are rising again (Howlett 2020; Roberts 2020; Sibbald 2020; Zeidler 2020). During the COVID-19 pandemic, outbreaks and concerns about safety and quality of care in LTC created an additional obstacle for safely transitioning people out of hospital (Grant and Ha 2020).

When our research team scanned the literature for strategies to improve these delayed transitions, we did not find any examples that were co-designed with patients, families and their care providers (Cadel et al. 2021). Our team of researchers, research staff, patients and caregivers came together to address this gap.

In an effort to co-design a strategy with patients and caregivers, we formed the ALC Patient and Caregiver Advisory Council. To identify key "pain points" during ALC, we conducted two focus groups (one with patients and caregivers and a separate one with care providers) and three co-design sessions with a mix of patients, caregivers and care providers across Ontario. Altogether, 61 people were engaged. We began our work in Toronto, ON, with funding from the Canadian Institutes of Health Research. Through the Ontario SPOR SUPPORT Unit (OSSU) Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach Award (EMPOWER), we expanded our co-design sessions across Ontario, and designed a website and a video to share our work on a wider scale. Council members' roles included creating terms of reference and other administrative activities related to council meetings, advising on project goals, sharing perspectives and, later, facilitating focus groups and co-design sessions. Rather than formally assessing the impact of working together, we held council meetings in between co-design sessions, allowing time to reflect on and revise our intervention (Figure 1). Together, we identified components of an intervention to improve the ALC experience, including a communication guide (a set of questions for patients, caregivers and providers to ask each other) and a care component (core set of services).

The impact of our work together

First impact: We amplified the voices of patients and caregivers

By being part of the council and project activities, patient and caregiver partners noted that they "felt less alone."

> [We had the] opportunity to speak and find out [about] other people [who] were in the same boat [allowing us to] build strength and overcome fears. (Lucy Bilotta, caregiver partner)

Ida McLaughlin, a caregiver and council chair, noted the importance of "bringing [patients and caregivers] to the table right from the start. [In doing so] you're more likely to have something that works, otherwise they [providers/leaders] come to the table with one-sided experience."

Lucy added that involving patients and caregivers is important because "patients and caregivers [are the ones] that have to live with the change."

> What strikes me, as a caregiver, is [that] we know the one we are looking after … but the healthcare people don't know what they [patients] like, what they don't like … Part of our job is to tell them. (Gordon [Gord] MacGregor, caregiver partner)

Ida reflected on the importance of being *beyond* her caregiver experience (her great aunt has passed away) and of having the ability to see the bigger picture: "I have watched patients and caregivers interact with hospital workers and have noticed that people are very emotional in the moment but here we are in a committee, and we can step back … Yes we bring our emotions to the table, but we can analyze things really clearly."

Gord, who shared his story in a podcast as one of our project outputs, said, "I really enjoyed doing the podcast. That was a big thing for me. Talking about my experiences and sharing them. I don't mind getting up and speaking to people."

The council still exists because of the members' commitment and desire to raise awareness on what ALC means and its implications.

> Nobody knows this is happening. You hear comments about hallway medicine … people think you are waiting a couple of days for a bed [but] it's more than that." (Ida McLaughlin)

Lisa Bennett added, "Other people are reaching out to me to learn more about my experiences and learn from what we learn from each other."

Second impact: We developed tools to improve patient, caregiver and provider experiences

In co-designing the communication guide, Lucy noted: "It opens a two-way street. Now we both have tools, not just providers."

Monika Syed concurred: [It is] "not adequate to keep what worked before. You need to get both sides. Having a group like this opens up the opportunity to change for the better [and] overcome roadblocks."

In addition, Lisa felt value in working in partnership with healthcare providers. She shared that by "having health professionals, you understand the other side of the coin," and it is "equally important to understand [their] obstacles." In reference to care providers, Lucy added, "Their hands are tied too. They only have the tools they have."

In reflecting on our co-designed activities with patients, caregivers and care providers across Ontario, Ida noted: "We've gone to others to verify that these are things that we've all gone through – common problems." Ida added that we can say that "these recommendations can help."

In addition to supporting patients and caregivers, Ida noted that addressing ALC issues could "free up a lot of beds."

As hospitals continue to deal with capacity issues (including ALC) and the COVID-19 pandemic, Lisa noted: "Now we've got two problems, and neither one of them will go away."

Alongside the above-mentioned impacts, our work together informed a set of leading practices for ALC for Ontario hospitals. For example, strategies to include patients and caregivers in care planning and decision making are part of the leading practices, along with our co-designed communication guide. The ALC leading practices will be circulated to hospitals across Ontario in the near future.

Part Two: Key Lessons Learned and Insights

Having accommodations in place so that no one feels excluded

To reduce barriers to participation in our co-design sessions, we offered accommodations for people with sensory impairments, medical needs and language barriers. We were also mindful about *where* we held the sessions. Our preplanning session with the council and an external expert facilitator in co-design helped us think through ways to engage people based on their specific needs and plan the sessions. During the co-design sessions, a volunteer verbalized steps of the co-design process and made notes on behalf of a council member experiencing blindness. We did not resort to traditional forms of knowledge sharing (Microsoft PowerPoint) and described any visuals we shared. We also hired an interpreter to work with a Mandarin-speaking patient during one of our co-design sessions. In-patient participation was made easier by holding focus groups and co-design sessions within three Ontario-based hospitals (Sinai Health System, Toronto; Health Sciences North, Sudbury; Trillium Health Partners, Mississauga). We kept sessions small (10–12 participants) and divided participants into smaller groups for deeper discussions. Also, in providing honorariums, we considered impacts on participants' financial circumstances (i.e., we divided payments between fiscal years).

One limitation of our project is that we just scratched the surface in terms of our attention to equity and diversity. While the council itself has a mix of men and women from different social locations and with experiences related to living with disability, only one of our members and a few of our co-design participants were from visible minority groups. Moving forward, development of approaches to engage a more diverse group of patients and caregivers and attention to race, gender, ethnicity and other social factors and how these shape co-design strategies will be essential.

Deliberately sharing leadership

The term *sharing leadership* is what the council members chose to use instead of the concept *sharing power*. The first step of shared leadership is giving people the space to talk. At our first meeting, we set the foundation by each council member sharing their story – from Gordon's wife, who was stuck in hospital, battling mental health challenges to Ida's elderly aunt, stuck on a stretcher, frightened and confused, waiting for a bed to open up. Instead of coming to the table as a "know-it-all" researcher, I (lead author) admitted that working in partnership with patients and caregivers was new to me and that I was excited to learn and receive guidance from the group. I asked people to call me out if I engaged in "research speak." As a result of sharing leadership with patients/caregivers, we were able to design something reflective of their needs and experiences.

We had co-design sessions that were fully led by council members. Other sessions were co-led by council members and researchers. Patient and caregiver partners liked both formats. I (lead author) was worried about having council members in facilitation roles during co-design sessions as I had thought they might not be comfortable (which later turned out to be my assumption). From this, I recognized that I continue to be influenced by traditional conceptions of the "researcher as leader." I also learned that partnership need not entail handing all power to patients and caregiver partners; rather, it means sharing leadership (or power) in ways that work for everyone. As noted by a patient partner and council member, Murray Powell: "Knowledge and involvement of the patients in their care path is, in fact, power, and accommodating patient needs provides a means to that end."

Finding ways to capture more patient voices is important

Patient voices are critical but seldom captured in projects that focus on ALC. The switch to virtual council meetings and co-design sessions during the pandemic made it even more difficult to engage people while they are in hospital. We engaged a few patients through council and co-design activities, but caregivers were the main source of insights on both patient and caregiver needs. While it is possible to garner patient experiences through the caregiver, it is not the same; although, we must note that in some cases it was the only way to gather information about the patient experience.

Partnering with decision makers to maximize the possibility of work getting implemented

A fundamental principle of co-design is to include people *impacted* by the problem as well as those who have the power to *address* the problem (Bammer 2013). We were missing the latter. While the project itself was borne out of the interest of a local task group of decision makers tackling ALC challenges, this group disbanded during the early stages of the project. We eventually found other practice communities to share our work; however, more deliberate engagement with decision makers throughout the project would have helped us position our work for greater uptake.

Rediscovering purpose between projects

Although our project has ended, the ALC Patient and Caregiver Advisory Council still has bimonthly Zoom meetings. We are applying for additional funding and looking for hospital partners to implement and evaluate our intervention in practice. In the interim, we have invited those doing similar work to our council meetings. For example, we have learned about new care units at Trillium Health Partners and Health Sciences North that aim to prevent ALC-related discharge delays and keep people activated during care transitions. These may be sites for

TABLE 1.
Guidance for reporting involvement of patients and the public, version 2, short form

Section and topic	Item	Page numbers
Aim	Report the aim of PPI in the study	49–50
Methods	Provide a clear description of the methods used for PPI in the study	50–51
Study results and outcomes	Report the results of PPI in the study, including both positive and negative outcomes	50–52
Discussion and conclusions	Comment on the extent to which PPI influenced the study overall. Describe positive and negative effects	52–53
Reflections/critical perspectives	Comment critically on the study, reflecting on the things that went well and those that did not	52

PPI = patient and public involvement.

our implementation and evaluation work.

Part Three: Dissemination Activities and Outcomes

Funding from OSSU helped us share our co-design work with patients, caregivers and care providers across Ontario and build on our findings. We published our findings in an international peer reviewed journal, *Health Expectations* (Kuluski et al. 2020), and presented our work virtually at two international conferences: Academy Health (Boston, MA) and the International Conference on Patient- and Family-Centered Care (Nashville, TN). The Council also created a website (https://www.bettercarejourney.com/), which outlines our work and shares tools and structured guidance for different audiences. Audio-visual content about the ALC experiences of some council members is included. Stakeholders wanting to test these tools in practice can use the contact information on our website to reach our team.

So what was the overall impact of this work?

Patients and caregivers had a platform to articulate their lived experience and co-designed an intervention that aims to improve care and communication during a delayed discharge. The broader system impact of our work remains to be seen as we strengthen and develop partnerships and look for opportunities to implement and evaluate our intervention in practice. See Table 1 for guidance on reporting involvement of patients and the public. HQ

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References

Amy, C., B. Zagorski, V. Chan, D. Parsons, R. Vander Laan and A. Colantonio. 2012. Acute Care Alternate-Level-of-Care Days due to Delayed Discharge for Traumatic and Non-Traumatic Brain Injuries. *Healthcare Policy* 7(4): 41–55. doi:10.12927/hcpol.2013.22883.

Bammer, G. 2013. *Disciplining Interdisciplinarity: Integration and Implementation Sciences for Researching Complex Real-World Problems.* ANU Press.

Bender, D. and P. Holyoke. 2018. Why Some Patients Who Do Not Need Hospitalization Cannot Leave: A Case Study of Reviews in 6 Canadian Hospitals. *Healthcare Management Forum* 31(4): 121–25. doi:10.1177/0840470418755408.

Burr, E. and S. Dickau. 2017. Leading Practices in Alternate Level of Care (ALC) Avoidance: A Standardized Approach. *Healthcare Quarterly* 20(2): 44–47. doi:10.12927/hcq.2017.25227.

Cadel, L., S.J.T. Guilcher, K.M. Kokorelias, J. Sutherland, J. Glasby, T. Kiran et al. 2021. Initiatives for Improving Delayed Discharge from a Hospital Setting: A Scoping Review. *BMJ Open* 11(2): e044291. doi:10.1136/bmjopen-2020-044291.

Challis, D., J. Hughes, C. Xie and D. Jolley. 2014. An Examination of Factors Influencing Delayed Discharge of Older People from Hospital. *International Journal of Geriatric Psychiatry* 29(2): 160–68. doi:10.1002/gps.3983.

Costa, A.P. and J.P. Hirdes. 2010. Clinical Characteristics and Service Needs of Alternate-Level-of-Care Patients Waiting for Long-Term Care in Ontario Hospitals. *Healthcare Policy* 6(1): 32–46. doi:10.12927/hcpol.2010.21899.

Costa, A.P., J.W. Poss, T. Peirce and J.P. Hirdes. 2012. Acute Care Inpatients with Long-Term Delayed-Discharge: Evidence from a Canadian Health Region. *BMC Health Services Research* 12: 172. doi:10.1186/1472-6963-12-172.

Cressman, G., J. Ploeg, H. Kirkpatrick, S. Kaasalainen and C. McAiney. 2013. Uncertainty and Alternate Level of Care: A Narrative Study of the Older Patient and Family Caregiver Experience. *Canadian Journal of Nursing Research* 45(4): 12–29. doi:10.1177/084456211304500403.

Everall, A.C., S.J.T. Guilcher, L. Cadel, M. Asif, J. Li and K. Kuluski. 2019. Patient and Caregiver Experience with Delayed Discharge from a Hospital Setting: A Scoping Review. *Health Expectations* 22(5): 863–73. doi:10.1111/hex.12916.

Gaughan, J., H. Gravelle and L. Siciliani. 2017. Delayed Discharges and Hospital Type: Evidence from the English NHS. *Fiscal Studies* 38(3): 495–519. doi:10.1111/j.1475-5890.2017.12141.

Grant, K. and T.T. Ha. 2020, May 20. How Shoring Up Hospitals for COVID-19 Contributed to Canada's Long-Term Care Crisis. *The Globe and Mail*. Retrieved January 2, 2021. <https://www.theglobeandmail.com/canada/article-how-shoring-up-hospitals-for-covid-19-contributed-to-canadas-long/>.

Howlett, K. 2020, April 5. Ontario Hospital Scramble to Open More Beds as They Brace for Surge in Coronavirus Cases. *The Globe and Mail*. Retrieved April 10, 2020. <https://www.theglobeandmail.com/canada/article-ontario-hospitals-scramble-to-open-more-beds-as-they-brace-for-surge/>.

Kortebein, P., A. Ferrando, J. Lombeida, R. Wolfe and W.J. Evans. 2007. Effect of 10 Days of Bed Rest on Skeletal Muscle in Healthy Older Adults. *JAMA* 297(16): 1772–74. doi:10.1001/jama.297.16.1772-b.

Kuluski, K., J.W. Ho, L. Cadel, S. Shearkhani, C. Levy, M. Marcinow et al. 2020. An Alternate Level of Care Plan: Co-Designing Components of an Intervention with Patients, Caregivers and Providers to Address Delayed Hospital Discharge Challenges. *Health Expectations* 23(5): 1155–65. doi:10.1111/hex.13094.

Kuluski, K., J. Im and M. McGeown. 2017. "It's a Waiting Game" a Qualitative Study of the Experience of Carers of Patients Who Require an Alternate Level of Care. *BMC Health Services Research* 17(1): 318. doi:10.1186/s12913-017-2272-6.

McCloskey, R., P. Jarrett and C. Stewart. 2015. The Untold Story of Being Designated an Alternate Level of Care Patient. *Healthcare Policy* 11(1): 76–89. doi:10.12927/hcpol.2015.24364.

McCloskey, R., P. Jarrett, C. Stewart and P. Nicholson. 2014. Alternate Level of Care Patients in Hospitals: What Does Dementia Have to Do with This? *Canadian Geriatrics Journal* 17(3): 88–94. doi:10.5770/cgj.17.106.

Ministry of Health and Long-Term Care. 2019, January. *Hallway Health Care: A System Under Strain. 1st Interim Report from the Premier's Council on Improving Healthcare and Ending Hallway Medicine*. Retrieved February 26, 2020. <http://www.health.gov.on.ca/en/public/publications/premiers_council/docs/premiers_council_report.pdf>.

Roberts, D. 2020, April 1. Sudbury Hospital to Move ALC Patients to Hotel as First COVID-19 Patient Admitted. *Northern Ontario CTV News*. Retrieved June 30, 2020. <https://northernontario.ctvnews.ca/sudbury-hospital-to-move-alc-patients-to-hotel-as-first-covid-19-patient-admitted-1.4877995>.

Rojas-Garcia, A., S. Turner, E. Pizzo, E. Hudson, J. Thomas and R. Raine. 2018. Impact and Experiences of Delayed Discharge: A Mixed-Studies Systematic Review. *Health Expectations* 21(1): 41–56. doi:10.1111/hex.12619.

Sibbald, B. 2020. What Happened to the Hospital Patients Who Had "Nowhere Else to Go"? *CMAJ* 192(22): E614– 15. doi:10.1503/cmaj.1095873.

Sutherland, J.M. and R.T. Crump. 2013. Alternative Level of Care: Canada's Hospital Beds, the Evidence and Options. *Healthcare Policy* 9(1): 26–34. doi:10.12927/hcpol.2013.23480.

Swinkels, A. and T. Mitchell. 2009. Delayed Transfer from Hospital to Community Settings: The Older Person's Perspective. *Health and Social Care in the Community* 17(1): 45–53. doi:10.1111/j.1365-2524.2008.00796.x.

Tan, W.S., W.F. Chong, K.S.G. Chua, B.H. Heng and K.F. Chan. 2010. Factors Associated with Delayed Discharges after Inpatient Stroke Rehabilitation in Singapore. *Annals of the Academy of Medicine of Singapore* 39(6): 435–41.

Walker, D. 2011, June 30. *Caring for Our Aging Population and Addressing Alternate Level of Care. Report Submitted to the Minister of Health and Long-Term Care*. Government of Ontario. Retrieved September 4, 2020. <https://www.niagaraknowledgeexchange.com/wp-content/uploads/sites/2/2014/05/Caring_for_Our_Aging_Population.pdf>.

Wilson, D.M., J. Vihos, J. Hewitt, N. Barnes, K. Peterson and R. Magnus. 2014. Examining Waiting Placement in Hospital: Utilization and the Lived Experience. *Global Journal of Health Science* 6(2): 12–22. doi:10.5539/gjhs.v6n2p12.

Zeidler, M. 2020. Thousands of Hospital Beds in B.C. Cleared to Make Room for COVID-19. *CBC News*. Retrieved April 2, 2020. <https://www.cbc.ca/news/canada/british-columbia/bc-hospital-beds-covid-19-1.5505356>.

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The Youth Wellness Quest: A Comprehensive Online Mental Health Literacy and Self-Advocacy Resource Developed by Youth for Youth

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Abstract

A lack of mental health literacy may impact youths' ability to advocate for themselves as they seek to access and navigate the mental healthcare system. Recognizing this, members of the National Youth Action Council at the Centre for Addiction and Mental Health in Toronto, ON, developed the Youth Wellness Quest resource. This health literacy resource informs youth of possible available services, increasing their capacity to make informed mental healthcare decisions. The youth-led process of creating this resource, from development to dissemination, is described within this paper, showcasing how youth can lead the development of tools designed for youth.

Key Points

- A youth team led the development and evaluative study of a mental health literacy resource for youth, along with its transformation into an engaging online resource.
- All activities were youth-led, with support from experienced researchers, a youth engagement coordinator, education specialists and web developers.
- This online youth-led resource is now an online platform, complemented by a brief downloadable version that provides youth with relevant information to better understand the mental healthcare system and service options, empowering youth to advocate for themselves and make informed decisions about their mental health and substance use care.

Introduction

In a recent survey of Canadian youth, 59% of respondents stated that they did not know where to go for help regarding mental health issues (Jing 2019). This is concerning, especially as youth – defined as those aged 15 to 29 years by Statistics Canada (2019) – have reported worse mental health than older Canadians (Garriguet 2021). Understanding the types of services available and how to access them is a key component of mental health literacy; poor mental health literacy can be a significant barrier for youth seeking help, many of whom prefer to be self-reliant and advocate for themselves (Bowers et al. 2013; Gulliver et al. 2010; Mehra et al. 2021). Most youth accessing the mental health system for the first time find it difficult to navigate (James 2007; McGorry et al. 2013). Youth with higher levels of mental health literacy may access help earlier, leading to better outcomes (Kelly et al. 2007).

Youth Wellness Quest

Recognizing the issue noted above, members of the National Youth Action Council (NYAC) at the Centre for Addiction and Mental Health (CAMH) in Toronto, ON, developed the Youth Wellness Quest (YWQ) resource (www.youthwellnessquest.com). NYAC members are Canadian youth with lived experience of mental health and/or substance use difficulties who understand struggles with

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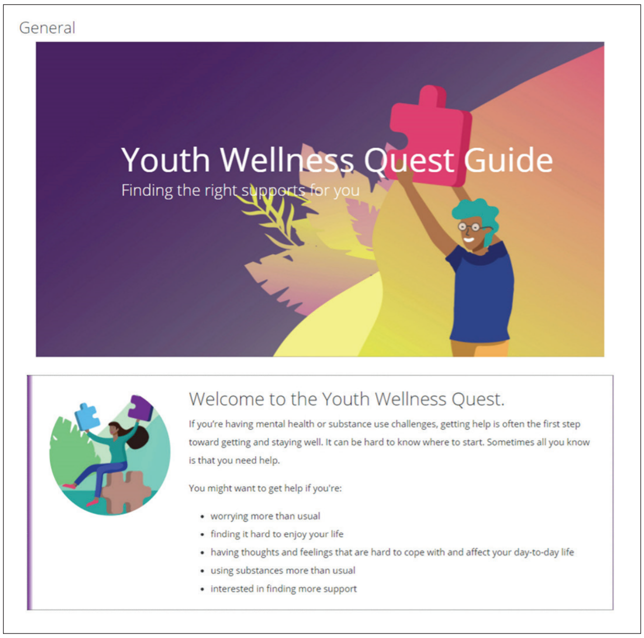
P = Youth partner.

*Co-lead authors.

the mental healthcare system and who used their lived experience to develop YWQ. The YWQ tool is a health literacy and self-advocacy resource for youth seeking services for any type of mental health or substance use concern. It is composed of a guide (Figure 1) and a checklist (Figure 2). The guide helps youth identify the issues most important to them and explains how these may be addressed using youth-friendly language in an engaging, easy-to-navigate format (Figure 3). It lists potential types of services and treatment partners rather than specific treatment providers, guiding youth in how to find such services in their own communities. It suggests questions that youth may ask service providers to be well informed and engaged in the treatment process. Youth may document their needs and preferences on the checklist as part of developing their own treatment plan, potentially alongside their service providers. As the ultimate goal of YWQ is to move youth from the role of patient to partner in their own mental healthcare, the YWQ resource was developed by youth for youth.

In developing the YWQ, NYAC members conducted a study to understand youths' perspectives on a draft version, involving five focus groups and an online survey (Syan et al. 2021). Participants lived across Canada, were aged 14 to 26 years and had diverse gender identities, cultural backgrounds and sociodemographic characteristics. Study participants were asked about the resource's content, presentation, utility and ability to promote self-advocacy. Overall, youth evaluations of the draft YWQ were positive, with some suggestions for content expansion. Participants suggested that the resource be available as both a comprehensive online version and a shorter print version for use with service providers in healthcare and mental health organizations, as well as educational institutions. A youth-led peer-reviewed journal article provides further details (Syan et al. 2021).

FIGURE 1.
Sample excerpt of the YWQ guide



General

Youth Wellness Quest Guide

Finding the right supports for you

Welcome to the Youth Wellness Quest.

If you're having mental health or substance use challenges, getting help is often the first step toward getting and staying well. It can be hard to know where to start. Sometimes all you know is that you need help.

You might want to get help if you're:

- worrying more than usual
- finding it hard to enjoy your life
- having thoughts and feelings that are hard to cope with and affect your day-to-day life
- using substances more than usual
- interested in finding more support

Funding Application

To develop an online version of the YWQ resource, a knowledge translation grant application was co-written by NYAC members, including the youth project lead, a youth advisor and allied researchers, then submitted to the Ontario SPOR SUPPORT Unit (OSSU). The youth project lead and youth advisor referenced the YWQ research project to frame the application. Through multiple team discussions, the youth lead and advisory team co-designed the knowledge translation plan, including the steps, the timeline, the youth leadership and engagement plan and the dissemination plan. While an allied researcher with grant-writing experience supported them and refined the final application for submission, youth contributed the core substance. OSSU funding was awarded to the team.

FIGURE 2.
Sample excerpt of the YWQ checklist

Youth Wellness Quest

Finding the right supports for you

Welcome to your Youth Wellness Quest! Below is a checklist that describes different parts of mental health and substance use care. You can use the checklist to note how important each part is for your own care. If you don't know what something is or want to learn more, you can find more information in the **Youth Wellness Quest Guide**. The guide also provides other useful notes, like tips and questions you might want to ask your service provider.

By the end of the checklist, you will have an idea of the types of services and supports that are important for you. You can use your completed checklist to find the services that suit you best. This can help you advocate for your mental health and substance use needs on your Youth Wellness Quest!

Section 1: Types of Services

Different types of services I am interested i

Is this important to me when I am getting support for my mental health or substance use?	How important is this to me?			
	Very	A bit	Not	Does not apply
Services that interest me				
Help only when I need it (e.g., walk-in counselling)				
Regularly scheduled appointments (outpatient programs)				
Services offered daily (day treatment)				
Help for mental health emergencies (crisis support)				
Living at the place where I will get help (inpatient/residential programs)				
What I want the service to offer				
One-on-one counselling				
Group counselling				
Support for my family				
Online/virtual options				
Support for challenges with my substance use				
Support/accommodation for disabilities (e.g., hearing or visual impairment, physical disabilities)				
Gender, sexuality and sexual orientation support (including support for 2SLGBTQA+ youth)				
Religious/spiritual support				
Cultural support (including services for Indigenous, Black, immigrant and refugee youth)				
Language/interpreter services				

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Project Execution

Funding was used to develop a comprehensive online version of the YWQ resource. Per study findings, this involved building an engaging and accessible youth-friendly resource for Canada-wide dissemination. The project was led by youth, using the McCain Model of Youth Engagement (Heffernan et al. 2017). One youth served as the primary lead, with six additional NYAC youth advisors supporting the project; these youth varied in the amount and type of lived experience they had, as well as in socio-economic and demographic backgrounds. Experienced researchers, a youth engagement coordinator, education specialists and web developers also provided support. The youth team was paid for their contributions.

The youth lead began by updating the content based on the research results, with input from the youth advisors. This included implementing suggestions to change some of the language and add information about services for historically marginalized populations. New sections were added, such as self-help, confidentiality and privacy. The finalized core content was adapted to an online educational software platform by adding links, click navigation, fillable fields, images, infographics and a short introductory video, making it engaging and easy to navigate. Some new content was added to increase online interactivity, such as worksheets and self-reflection activities. Through multiple virtual meetings and e-mail consultations with six youth advisors and other team members, the team enhanced the design, look and feel of the resource.

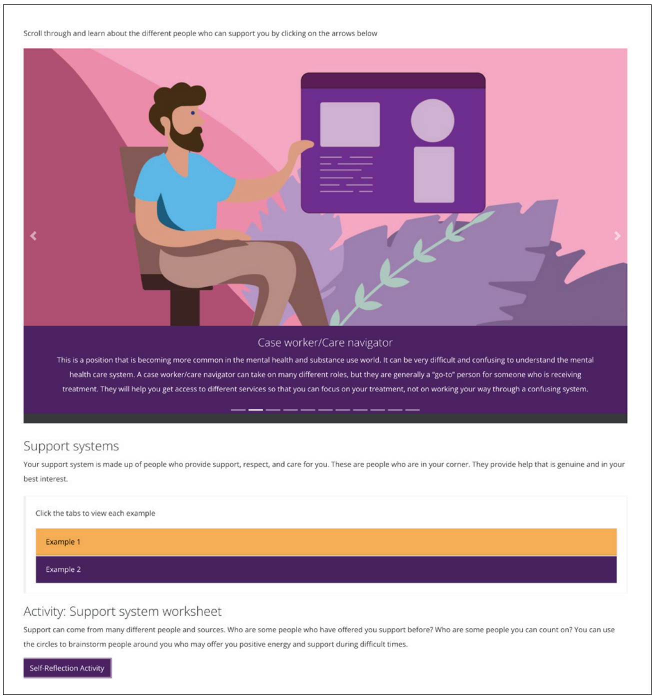
Once the web designers programmed the content, the same youth team pilot-tested the draft resource to identify optimizations. Revisions were made accordingly. Pilot testing and revision continued iteratively with the youth team until a launch-ready version was established. The final resource is freely accessible at www.youthwellnessquest.com. To complement the online version, the youth team developed a brief print version available for free download on the platform and for purchase in bulk for a cost-recovery fee at the CAMH store.

Dissemination and Knowledge Translation

The online YWQ resource was launched on May 13, 2021. According to the dissemination plan, the youth lead created multiple social media advertisements and shared them repeatedly through the NYAC mailing list and the NYAC and CAMH Twitter and Facebook accounts. As of August 13, 2021, three posts in the Facebook NYAC community group, four NYAC Twitter posts and five paid Instagram and Facebook advertisements targeting youth and other family members together garnered 1,626,707 views and generated 539 website hits.

Furthermore, the YWQ resource was circulated through existing networks of youth-serving organizations and community stakeholders, including healthcare professionals, through a newsletter and several presentations. The introductory video was uploaded onto YouTube (CAMH 2021), generating over 200 views. A landing page was created for the resource on the Centre for Innovation in Campus Mental Health website (Centre for Innovation in Campus Mental Health 2021). Through the sum of these dissemination activities, the online YWQ resource received 989 visits in its first three months. Future dissemination plans include the development of a French-language version and leveraging opportunities to insert the YWQ resource into the Canadian youth wellness landscape. The effectiveness of the tool and these dissemination practices will be evaluated over time.

FIGURE 3.
Sample excerpt of the YWQ guide website



Scroll through and learn about the different people who can support you by clicking on the arrows below

Case worker/Care navigator

This is a position that is becoming more common in the mental health and substance use world. It can be very difficult and confusing to understand the mental health care system. A case worker/care navigator can take on many different roles, but they are generally a "go-to" person for someone who is receiving treatment. They will help you get access to different services so that you can focus on your treatment, not on working your way through a confusing system.

Support systems

Your support system is made up of people who provide support, respect, and care for you. These are people who are in your corner. They provide help that is genuine and in your best interest.

Click the tabs to view each example

Activity: Support system worksheet

Support can come from many different people and sources. Who are some people who have offered you support before? Who are some people you can count on? You can use the circles to brainstorm people around you who may offer you positive energy and support during difficult times.

Challenges and Mitigating Strategies

With a strong youth engagement infrastructure in place, the team has previously worked through engagement challenges and developed effective mitigating strategies that were applied during this project (Hawke et al. 2018; Heffernan et al. 2017; Syan et al. 2021). However, this was a relatively long-term project, initially beginning in 2015 at the idea stage and achieving implementation in 2021. As a youth-led project, limited dedicated project hours and youth turnover slowed progress. In 2019, the team, therefore, hired a new dedicated project lead with sufficient hours to move the project forward to completion. Another challenge was regarding the inherent mobility of youth, who are at a life stage characterized by change in educational and occupational status. It was unclear whether the same young people would be available throughout the writing and publication process for the current manuscript or whether career progress would interfere with staffing continuity. This challenge was mitigated by ensuring that additional youth were engaged for flexibility and ongoing youth leadership despite individual mobility. No other major challenges regarding engagement were encountered. Although the COVID-19 pandemic has led to many organizations pivoting how their teams operate, this project already engaged with youth through online platforms. Thus, this project has continuously involved youth – albeit only those with Internet access – throughout its progression.

Some challenges were technical in nature – that is, creating an online resource with intuitive click navigation and engaging materials within the limitations of the online software. These challenges were discussed among the project team, including web designers, to develop solutions. However, at a draft development stage, the team realized that the resource was optimized for desktop usage, not the mobile usage that was expected to be the main means of access for youth. In team meetings, this challenge was discussed, and progressive refinements were identified until a version was established with acceptable mobile functionality.

Youth's Experience of Engagement

Youth led this project and were involved in every step. The co-lead authors of this paper are two of these youth (AS and JYL). One was engaged from the very early days of the conceptualization of the YWQ resource in 2015, and both have been engaged since the research stage. The project enabled them to contribute their knowledge and skills, develop their capabilities, enhance their understanding of the Canadian healthcare system and learn about online resource development. They consider the opportunity to collaborate with other youth to have been an "incredible experience." They emphasize that engaging youth with a range of experience in accessing care, advocating for themselves and navigating the healthcare system has made this resource more effective and tailored to its intended audience. Knowing this resource is now available for youth nationally, hopefully helping many, makes them "happy" and "proud."

Research Team's Experience of Youth Engagement

Given the youth leadership through the resource development and research project, the entire team felt that youth leadership during knowledge translation was essential. They, therefore, continued to support the YWQ youth team in leading the resource development. The researchers recognize that the engaged youth team was very familiar with the YWQ resource and with using online tools, making them the clear choice for design leadership. The researchers feel that the quality of the resource and its engaging, youth-centred content and design highlight the importance of ensuring youth leadership in all stages of a youth-oriented project like this one.

Youth led this project and were involved in every step.

Conclusion

The idea to develop the YWQ resource came from the lived experience of help-seeking youth, who described the mental health system as overwhelming and non-linear. Using a youth-led design, the team developed a unique Canadian health literacy and self-advocacy resource for help-seeking youth. The OSSU funding enabled us to pursue a youth-led knowledge-to-action process in which an online platform was developed, making the resource widely and freely accessible to youth.

In the past decade, there has been an increasing interest in the use of the Internet for mental health promotion and information dissemination (Clarke et al. 2015). Accessing mental health information online may offer youth increased privacy and anonymity (Clarke et al. 2015), which are major concerns for help-seeking youth (Williams and Chapman 2011). Online platforms may also provide minority and rural/remote populations with a cost-effective and accessible means of accessing services (Barak and Grohol 2011). Online platforms have been particularly important during the COVID-19 pandemic (WHO 2020), which has both negatively impacted the mental health of youth and led to services and resources moving online (Cielo et al. 2021; Hawke et al. 2020). Developing this online resource and complementary print materials allows us to reach a broad range of youth during this time of service disruption and help youth identify the types of services they wish to access.

Engaging youth service users in a project allows for a unique "insider" expertise that can lead to more effective and optimized designs of youth-focused services and resources (Gibbs et al. 2020). The value of youth engagement and leadership was experienced through all phases of the YWQ project, from defining the central need to disseminating the resource. Based on this positive experience, the authors call upon researchers, resource developers and service designers to engage youth for their expertise in developing youth-friendly resources that are developmentally appropriate and relevant to their intended audiences. HQ

References

Barak, A. and J.M. Grohol. 2011. Current and Future Trends in Internet-Supported Mental Health Interventions. *Journal of Technology in Human Services* 29(3): 155–96. doi:10.1080/15228835.2011.616939.

Bowers, H., I. Manion, D. Papadopoulos and E. Gauvreau. 2013. Stigma in School-Based Mental Health: Perceptions of Young People and Service Providers. *Child and Adolescent Mental Health* 18(3):165–70. doi:10.1111/j.1475-3588.2012.00673.x.

Centre for Addiction and Mental Health (CAMH). 2021, May 5. *The Youth Wellness Quest: Finding the Right Supports for You* [Video]. Youtube. Retrieved March 11, 2022. <https://www.youtube.com/watch?v=wYMelEfCE3Q>.

Centre for Innovation in Campus Mental Health. 2021. Youth Wellness Quest – Finding the Right Supports for You. Retrieved December 12, 2021. <https://campusmentalhealth.ca/resource/youth-wellness-quest-finding-the-right-supports-for-you/>.

Cielo, F., R. Ulberg and D. Di Giacomo. 2021. Psychological Impact of the COVID-19 Outbreak on Mental Health Outcomes among Youth: A Rapid Narrative Review. *International Journal of Environmental Research and Public Health* 18(11): 6067. doi:10.3390/ijerph18116067.

Clarke, A.M., T. Kuosmanen and M.M. Barry. 2015. A Systematic Review of Online Youth Mental Health Promotion and Prevention Interventions. *Journal of Youth and Adolescence* 44(1): 90–113. doi:10.1007/s10964-014-0165-0.

Garriguet, D. 2021, February 1. Chapter 1: Health of Youth in Canada. *Portrait of Youth in Canada: Data Report*. Statistics Canada. Retrieved March 18, 2022. <https://www150.statcan.gc.ca/n1/pub/42-28-0001/2021001/article/00001-eng.htm>.

Gibbs, L., M. Kornbluh, K. Marinkovic, S. Bell and E.J. Ozer. 2020. Using Technology to Scale Up Youth-Led Participatory Action Research: A Systematic Review. *Journal of Adolescent Health* 67(2): S14–23. doi:10.1016/j.jadohealth.2019.10.019.

Gulliver, A., K.M. Griffiths and H. Christensen. 2010. Perceived Barriers and Facilitators to Mental Health Help-Seeking in Young People: A Systematic Review. *BMC Psychiatry* 10(1): 113. doi:10.1186/1471-244X-10-113.

Hawke, L.D., S.P. Barbic, A. Voineskos, P. Szatmari, K. Cleverley, E. Hayes et al. 2020. Impacts of COVID-19 on Youth Mental Health, Substance Use, and Well-Being: A Rapid Survey of Clinical and Community Samples: Répercussions de la COVID-19 sur la santé mentale, l'utilisation de substances et le bien-être des adolescents: un sondage rapide d'échantillons cliniques et communautaires. *The Canadian Journal of Psychiatry* 65(10): 701–09. doi:10.1177/0706743720940562.

Hawke, L.D., J. Relihan, J. Miller, E. McCann, J. Rong, K. Darnay et al. 2018. Engaging Youth in Research Planning, Design and Execution: Practical Recommendations for Researchers. *Health Expectations* 21(6): 944–49. doi:10.1111/hex.12795.

Heffernan, O.S., T.M. Herzog, J.E. Schiralli, L.D. Hawke, G. Chaim and J.L. Henderson. 2017. Implementation of a Youth-Adult Partnership Model in Youth Mental Health Systems Research: Challenges and Successes. *Health Expectations* 20(6): 1183–88. doi:10.1111/hex.12554.

James, A.M. 2007. Principles of Youth Participation in Mental Health Services. *The Medical Journal of Australia* 187(S7): S57–60. doi:10.5694/j.1326-5377.2007.tb01339.x.

Jing, J. 2019, August. *Analysis of Youth Mental Wellness Survey*. Youth Mental Health Canada. Retrieved December 12, 2021. <https://ymhc.ngo/wp-content/uploads/2020/03/analysis-of-youth-mental-wellness-survey-v3.pdf>.

Kelly C.M., A.F. Jorm and A. Wright. 2007. Improving Mental Health Literacy as a Strategy to Facilitate Early Intervention for Mental Disorders. *The Medical Journal of Australia* 187(S7): S26–30. doi:10.5694/j.1326-5377.2007.tb01332.x.

McGorry, P., T. Bates and M. Birchwood. 2013. Designing Youth Mental Health Services for the 21st Century: Examples from Australia, Ireland and the UK. *The British Journal of Psychiatry* 202(S54): S30–35. doi:10.1192/bjp.bp.112.119214.

Mehra, K., L.D. Hawke, P. Watson, N.Y. Sheikhan, E. Leroux and J. Henderson. 2021. Youth Perspectives on Seeking Psychotherapy: A Concurrent Mixed Methods Study. *Journal of the Canadian Academy of Child and Adolescent Psychiatry* 30(3): 165–76.

Statistics Canada. 2019, May 8. A Portrait of Canadian Youth: March 2019 Updates. Retrieved November 18, 2021. <https://www150.statcan.gc.ca/n1/pub/11-631-x/11-631-x2019003-eng.htm>.

Syan, A., J.Y.Y. Lam, C.G.L. Huang, M.S.M. Smith, K. Darnay, L.D. Hawke et al. 2021. The Wellness Quest: A Health Literacy and Self-Advocacy Tool Developed by Youth for Youth Mental Health. *Health Expectations* 24(2): 659–69. doi:10.1111/hex.13214.

Williams, K. A. and M.V. Chapman. 2011. Comparing Health and Mental Health Needs, Service Use, and Barriers to Services among Sexual Minority Youths and Their Peers. *Health & Social Work* 36(3): 197–206. doi:10.1093/hsw/36.3.197.

World Health Organization (WHO). 2020, October 5. The Impact of COVID-19 on Mental, Neurological and Substance Use Services: Results of a Rapid Assessment. Retrieved November 23, 2021. <https://www.who.int/publications/i/item/978924012455>.

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> “The discussion-based human-centred design structure of the workshops helped bring to the forefront ideas from patients that are typically not discussed by researchers alone.”
>
> *– p. 67*

The Retinoblastoma Research Booklet: A Catalyst for Patient Involvement in Retinoblastoma Research

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Abstract

Peer-to-peer recruitment efforts are important in generating interest and participation of patients as partners in research but difficult to sustain when face-to-face interactions are limited. The *Retinoblastoma Research and You!* booklet, co-developed by patients, researchers and health professionals, serves as a guide for patient engagement in research while retaining an element of personalization. The *Retinoblastoma Research and You!* booklet was developed through two virtual workshops to iterate and finalize the booklet design and content. The booklet outlines how individual patients' lived experiences and skills can influence retinoblastoma research and highlights real-world examples of patient-partnered research activities at different stages of the research process.

Key Points

- The Canadian Retinoblastoma Research Advisory Board aims to identify and include a large and diverse group of patients in retinoblastoma research to create research that is meaningful and relevant to patients and improves outcomes.
- To facilitate recruitment, we developed a booklet with patient partners that outlines how individual patients' lived experiences and skills can influence retinoblastoma research. The booklet highlights real-world examples of patient-partnered research activities at different stages of the research process that is further illustrated through patient testimonials.
- The booklet was distributed through the retinoblastoma clinics and relevant cancer advocacy organizations across Canada in order to reach new patients and encourage them to get involved.

Introduction

Retinoblastoma is a childhood eye cancer with lifelong implications for survivors and their families. As such, patients (i.e., those with lived experience of the disease, including family and friends [CIHR 2014]) have expressed a desire to become involved in research as a means to improve care and impact on current and future patients. As described in our accompanying manuscript in this special issue (Ristevski et al. 2022), the Canadian Retinoblastoma Research Advisory Board (CRRAB) was developed with the ultimate goal to have patients work *alongside* researchers and health professionals to create meaningful retinoblastoma research that is relevant to patients and improves outcomes. Much of this work is led by a "parent in research" – an individual with lived experience of retinoblastoma who is employed as part of a research team. The CRRAB membership is drawn from those enrolled in the retinoblastoma research community (i.e., people in the retinoblastoma community who are interested in research and formally join by signing up online), which is situated within the broader retinoblastoma community (i.e., anyone who is

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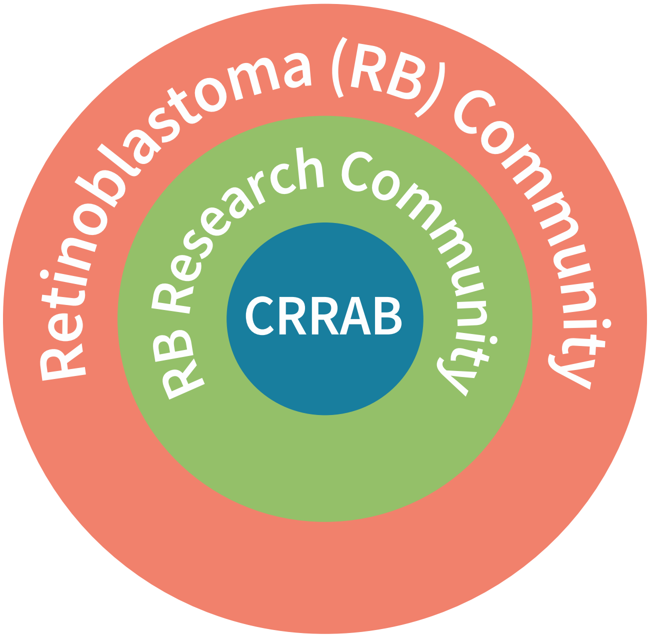
P = Patient with lived experience of retinoblastoma. R = Researcher with expertise in retinoblastoma. H = Health professional with expertise in retinoblastoma.

FIGURE 1.
CRRAB schematic



Navigating the Canadian retinoblastoma research landscape starts with the retinoblastoma community, including but not limited to patients, researchers and health professionals. The retinoblastoma research community is a subset of the broader community and includes those who express an interest in retinoblastoma research. CRRAB is a further subset of the retinoblastoma research community and includes those who actively participate in and contribute to patient-partnered research and related activities.

personally or professionally connected to retinoblastoma in Canada, including but not limited to patients, researchers and health professionals) (Figure 1).

A major CRRAB goal is to identify and include a large and diverse group of patients in research and have them join the retinoblastoma research community, an online database that endeavours to enroll a diverse and inclusive group of people who have already been affected by retinoblastoma, as well as newly affected individuals. Members are invited to specify their desired level of involvement, which may include (1) receiving information about retinoblastoma, including research results and updates, and (2) being contacted by The Hospital for Sick Children (SickKids) – where the majority of Canadian retinoblastoma patients are managed – retinoblastoma research team and/or external researchers for information about retinoblastoma and research opportunities (Gelkopf et al. 2020; White et al. 2019). To optimize benefit, involved patients should extend beyond those who most regularly participate, and incorporate a diversity of individuals representing different demographics (i.e., sex, geography, language, age, etc.) and lived experiences (i.e., relationship to retinoblastoma, treatment course, diagnosis, etc.).

To attract diverse participation, CRRAB generates awareness of, and interest in, patient engagement in research using a variety of methods, often relying on the strength of peer-to-peer networks, social media and the power of personal stories. Volunteer-led social media efforts share personal stories and create thematic content to inform followers about research results, upcoming events, research opportunities and the importance of participating in retinoblastoma research. Face-to-face discussions about retinoblastoma research during retinoblastoma clinic days at SickKids have proven successful, as have patient-led conferences and research symposia. However, since the onset of the COVID-19 pandemic, restrictions to in-person interactions temporarily halted recruitment and engagement efforts, requiring a new mode of recruitment that still maintains a semblance to peer-to-peer networking.

The purpose of this project was to improve patient recruitment and involvement in CRRAB through the development of (1) a novel recruitment and educational tool, the *Retinoblastoma Research and You!* booklet (https://www.rbcanadaresearch.com/introducing-the-retinoblastoma-research-and-you-booklet/) and (2) a distribution plan to reach retinoblastoma patients to introduce or re-engage them with CRRAB.

Patient Engagement Methods and Outputs

Co-creation of the *Retinoblastoma Research and You!* booklet

Prior to the start of this project, attendees at monthly CRRAB working group meetings (including patients, researchers and health professionals) had begun to brainstorm content for the proposed booklet, creating rough designs using Microsoft PowerPoint. The funding from the Ontario Strategy for

Patient-Oriented Research SUPPORT Unit (OSSU) Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Award facilitated further work to (i) design and implement two virtual workshops to iterate and finalize the booklet design and content and (ii) produce and distribute physical and electronic booklets to the patient community.

Design and leadership of workshops

The parent in research (IV) collaborated with the scientist (HD) to design the structure and content of the workshops, each contributing their lived or scientific expertise, respectively. In collaborative brainstorming meetings, they formulated goals for each workshop (e.g., desired feedback, tangible outputs) and considered effective methods to stimulate discussion and consensus among participants (e.g., Zoom polls, pre-readings, etc.). Equipped with information from these meetings, the parent in research developed workshop materials and used the structure of regular lab meetings to present and refine the materials with input from the scientist and the broader research team. The parent in research took overall responsibility for project implementation, drawing on her personal lived experience of retinoblastoma and work experience as a member of a health research team to lead the development of the booklet; this facilitated action as a liaison, or bridge, between scientific, health professional and patient contributors to the project.

Recruitment of workshop participants

An open invitation was sent to CRRAB members to attend the virtual workshops, which opened the possibility for participation across Canada, removing the barrier of travel. Thus, the travel restrictions of the pandemic did not affect recruitment, especially because CRRAB activities have routinely used virtual platforms for monthly and quarterly meetings since its start. Workshop dates and times were chosen to coincide with monthly working groups to facilitate availability of members. To ensure a broad representation of retinoblastoma experiences in the workshop, the parent in research additionally reached out to CRRAB members with unique lived experiences and professional expertise to round out the group.

The workshops were attended by one research student and 13 CRRAB members representing patients (three parents, six survivors, one survivor/parent), one researcher and two health professionals. The team included a variety of professional specialties, including an ophthalmologist, a retired genetic counsellor, an elementary school teacher and children's book author, a leader in digital health solutions and others with general business experience. Some participants also served as retinoblastoma advocates working with collaborating organizations, the Canadian Retinoblastoma Society (https://www.rbsociety.ca/) and Know the Glow Foundation (https://knowtheglow.org/). In terms of geography, attendees represented British Columbia, Ontario, Alberta and Newfoundland.

Implementation of workshops

In advance of the workshop, the parent in research prepared and sent the participants the draft content created using Microsoft PowerPoint, as well as draft designs and page layouts created by a hired graphic designer. Workshop One began with a brief icebreaker of introductions and sharing of their relationship to retinoblastoma and motivation for being involved in research.

Each workshop followed human-centred design methodology consisting of three phases: ideation, rapid prototyping and refinement. Workshop One focused on the look and feel of the booklet, including font, colour scheme and general page design as well as the front cover design. Workshop Two focused on finalizing specific graphics within the booklet and the design of the back page. Focused on these goals, all stakeholders discussed the merits of each design, suggested changes and voted to reach consensus. The graphic designer made modifications in real time and offline to produce new prototype designs. Online surveys were used between workshops to gather additional inputs, and revised designs were reviewed in the final workshop. Final suggestions for refinement were incorporated by the graphic designer after the workshops, in consultation with the parent in research.

Patients in particular were encouraged to draw on their personal experiences to contribute to booklet development. CRRAB member JK created a custom quick response (QR) code that opens to a list of various retinoblastoma-related links to inform the reader of the different ways they can be involved. He described his experience as follows:

> As restaurants reopened, I noticed that a large proportion of menus had disappeared; many establishments had gone paperless – making their menus accessible via scanning a QR code sticker on the tabletop. I thought to myself, "What if we could do the same thing for the retinoblastoma research booklet, so that every time an ophthalmologist was treating a patient or advising a parent, they would be able to scan something quickly to take with them?"

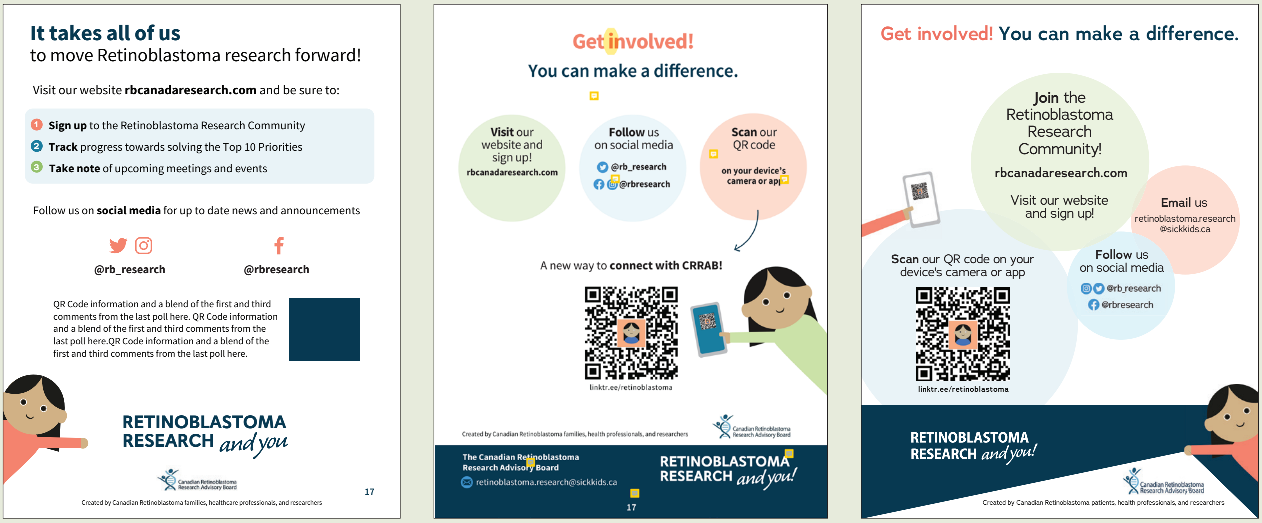
In this creative experience, Jay was able to use his professional skills as a leader in digital health solutions and create a simple but impactful contribution to the booklet.

Figure 2 illustrates how the human-centred design process was used to co-design the back cover of the booklet. The content in the back cover was initially agreed upon, with the main goal being to drive readers to visit the CRRAB website (Figure 2a). The ideation phase facilitated generation of the idea of including a QR code, which was subsequently rapidly prototyped and included in the design (Figure 2b). Through the process of refinement, the content and layout were finalized (Figure 2c). The iterative, discussion-based process facilitated crosstalk and opportunity for workshop participants to debate on the understandability and relevance of the booklet's content for the target patient audience. For example, because some workshop participants indicated that they were unfamiliar with how to use a QR code, instructions on "how to use" were incorporated into the final booklet.

Final booklet

The resultant *Retinoblastoma Research and You!* booklet (Figure 3) provided in a hard copy and digital version was created to be accessible to those with low and impaired vision by incorporating accessible design standards. The booklet outlines how individual patients' lived experiences and skills can influence retinoblastoma research with their participation in CRRAB activities. The booklet highlighted accessible routes for patients to get involved in research and showcased real-world examples of patient-partnered research activities at different stages of the research process. These real-world examples were further illustrated through testimonials written by Retinoblastoma Research Champions (https://www.rbcanadaresearch.com/join-crrab/champion-program/about_champion_program/), patients who actively promote patient engagement in research.

FIGURE 2.
Iteration of the booklet back cover
The human-centred design process was used to co-design the back cover of the booklet:



(a) The content of the back cover was initially agreed upon, with the main goal being to drive readers to visit the CRRAB website;

It takes all of us
to move Retinoblastoma research forward!

Visit our website **rbcanadaresearch.com** and be sure to:

1. **Sign up** to the Retinoblastoma Research Community
2. **Track** progress towards solving the Top 10 Priorities
3. **Take note** of upcoming meetings and events

Follow us on **social media** for up to date news and announcements

@rb_research **@rbresearch**

QR Code information and a blend of the first and third comments from the last poll here. QR Code information and a blend of the first and third comments from the last poll here.QR Code information and a blend of the first and third comments from the last poll here.

RETINOBLASTOMA RESEARCH *and you*

Canadian Retinoblastoma Research Advisory Board

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Created by Canadian Retinoblastoma families, healthcare professionals, and researchers

(b) the ideation phase facilitated the idea of including a QR code, which was subsequently rapidly prototyped and included in the design; and

Get involved!
You can make a difference.

Visit our website and sign up!
rbcanadaresearch.com

Follow us on social media
@rb_research
@rbresearch

Scan our QR code
on your device's camera or app

A new way to **connect with CRRAB!**

linktr.ee/retinoblastoma

Created by Canadian Retinoblastoma families, health professionals, and researchers

Canadian Retinoblastoma Research Advisory Board

The Canadian Retinoblastoma Research Advisory Board
retinoblastoma.research@sickkids.ca

RETINOBLASTOMA RESEARCH *and you!*

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(c) through the process of refinement, the content and layout were finalized.

Get involved! You can make a difference.

Join the Retinoblastoma Research Community!
rbcanadaresearch.com
Visit our website and sign up!

Email us
retinoblastoma.research@sickkids.ca

Scan our QR code on your device's camera or app
linktr.ee/retinoblastoma

Follow us on social media
@rb_research
@rbresearch

RETINOBLASTOMA RESEARCH *and you!*

Canadian Retinoblastoma Research Advisory Board

Created by Canadian Retinoblastoma patients, health professionals, and researchers

FIGURE 3.
The *Retinoblastoma Research and You!* booklet

It takes all of us
to move retinoblastoma
research forward!

Your guide to the Canadian
Retinoblastoma Research Community

Learn how your lived experience and skills
can be used to influence retinoblastoma research.

RETINOBLASTOMA
RESEARCH *and you!*

Canadian Retinoblastoma Research Advisory Board

Created by Canadian retinoblastoma patients, health professionals, and researchers

By Partnering in Res

✓ **Build relationships** with Cana
retinoblastama survivors, pare
health professionals and resea

✓ **Improve your knowledge** of
retinoblastoma and gain access to
the latest research findings

✓ **Apply your lived experience**
and skills to enhance
retinoblastoma research

research and related activities and events. CRRAB is led by a Steering Committee which is responsible for ensuring annual goals are set and progress is tracked towards patient partnership in research. The Steering Committee also manages CRRAB tools, such as the website, social media accounts, and the online sign-ups for the Canadian Retinoblastoma Research Community.

Members of CRRAB who draw from personal experience to promote patient partnership in research and encourage sign-up to the Canadian Retinoblastoma Research Community are designated **Retinoblastoma (RB) Champions** - read their stories on page 11.

The *Retinoblastoma Research and You!* booklet is a guide to patient engagement in retinoblastoma research in Canada developed by CRRAB members to encourage patients to join the Canadian retinoblastoma research community and participate in CRRAB by outlining how individual patients' lived experiences and skills can influence retinoblastoma research.

Development of a distribution plan for the booklet

For effective distribution of the booklet, it was critical to obtain buy-in from health professionals and patient stakeholders. At workshops and CRRAB regional working groups, CRRAB members discussed how patients access the latest information and research and how researchers and health professionals typically share research results. Patients revealed that a key mode for accessing information was through trusted stakeholders, such as health professionals and advocacy organizations. In turn, an exhaustive list of stakeholders and organizations was created by the research team and then prioritized by consensus at the workshops. Workshop participants reflected on potential organizations they themselves were members of, or had heard of in the past, which could help identify retinoblastoma patients beyond those who most regularly participate.

Distribution via health professionals

The parent in research presented the overview of the booklet and its purpose to the Canadian Retinoblastoma Tumor Board (attended by health professionals who manage retinoblastoma throughout Canada). The board members helped develop a distribution plan to target their clinics' patients, providing insights on logistics and requesting a standard operating procedure (SOP) to guide their involvement.

The final distribution plan included dissemination of the hard copy booklets in clinics and digital booklets via e-mail directly to patients. The SOP, developed by the parent in research in collaboration with the scientist (who reviewed the draft SOP for clarity, format and content), included suggested talking points to use with patients, an e-mail template and guidance on answering patient questions about the booklet. In addition, the QR code that was included on the booklet's back cover was printed in a sticker format to be adhered to professional ID badges to serve as a readily accessible method to connect patients with electronic resources – particularly important if print booklets ran out.

Distribution via media toolkit

It was decided that the booklet could be shared with identified professional and advocacy organizations using a media toolkit composed of a press release, social media posts and a poster along with instructions on how to use each media toolkit component.

CRRAB members drafted the media toolkit components at CRRAB monthly working groups, and an undergraduate research student helped refine them working alongside the parent in research and a scientist. The first iteration of the media toolkit was shared via e-mail with Jill Robert, a patient partner, who provided feedback from a patient perspective as well as a health network perspective due to her work. The patient partner was asked to assess language, style, visuals and the overall function and flow of the media toolkit.

Patient partners also played a key role in providing feedback to improve the distribution and uptake of the media toolkit and offered to also share it with their networks. For example, Mary Connolly-Wilson, a retired genetic counsellor, contributed to developing a plan on reaching genetic counsellors and also championed the booklet within her personal and professional network.

Key Messages

The *Retinoblastoma Research and You!* booklet, co-developed by patients, researchers and health professionals, serves as a guide for patient engagement in research while retaining an element of personalization that will help increase recruitment and participation in CRRAB. Patients co-created the booklet with researchers, health professionals and a graphic designer, incorporating their views and experiences on what is helpful to know about partnering in research and its benefits, which go beyond shaping research and include forming connections with other patients and professionals in the retinoblastoma community.

While we have yet to formally evaluate the impacts of patient involvement in this project, some patient partners explained the value of patient involvement in this project in their own words:

[Patient involvement in this project] helped to shape the content in a relevant way based on what we wished we had available to us. As a patient, it took me 30 years to obtain an awareness about any sort of retinoblastoma community. Moving forward, I hope that future patients will be connected to our community and obtain support much, much faster.

– Jay Kiew

At my daughter's diagnosis, I felt overwhelmed, but I didn't feel I was provided with many things that gave me hope. I believe this booklet not only provides valuable information; it also provides hope to those affected by retinoblastoma.

– Alena Vincent

My parents really struggled to come to grips with my diagnosis 20 years ago – what it meant for me, what it meant for our family, what I would and wouldn't be able to do – and had a lot of difficulty finding others to connect with who had been through it before. Knowing that I'm making it easier for another family is a great feeling!

– Mitch Hendry

The discussion-based human-centred design structure of the workshops helped bring to the forefront ideas from patients that are typically not discussed by researchers alone. For example, prior study has shown that motivations and expectations of patient involvement in research differs among patients and researchers (Schilling et al. 2019). In our project, during a discussion about the benefits of patient partnership in research, a parent shared that a personal benefit that they experienced by partnering in research was in connecting with other families affected by retinoblastoma. In addition, the feeling of making a difference for future affected families was important to them. These ideas helped enrich the discussion beyond just the potential patient impact on the research itself to include the impact of research partnership on patients themselves. Consequently, the discussion was incorporated into the content of the booklet.

Another important impact on the project was in exposing stakeholders at the periphery of the CRRAB network to patient engagement in research. For example, the recruitment of an undergraduate research student to this project resulted in them building research expertise in patient engagement and benefiting from mentorship from the parent in research. Similarly, the participation of the parent in research in the Canadian Retinoblastoma Tumor Board allowed the participation of health research stakeholders in this project, who – while members of CRRAB – would not be able to regularly attend meetings and workshops. This way, the health professional perspective was incorporated into the project and relayed to workshop participants via the parent in research.

The completion of the project was not without its challenges. Commonly with larger groups, ensuring all ideas are considered appropriately makes the process more complex and time consuming. The use of videoconferencing software came with its usual drawbacks: microphone/camera issues, inability to speak simultaneously, accessibility concerns regarding the chat function and scheduling across different Canadian time zones. We worked around this as best we could by scheduling meetings during weekends and lunch hours, with duplicate meetings to accommodate for different time zones and frequent e-mail reminders to avoid missed meetings. E-mails and one-on-one meetings were the most requested methods for participants who wanted to connect outside the group meetings. However, we believe that the early and routine engagement of patient partners through CRRAB helped ensure adequate recruitment of patient partners to this particular project, as shown for other projects (Vat et al. 2017). Still, it is possible that the virtual mode excluded participation by individuals who lacked a reliable Internet connection or suitable electronic device. Furthermore, we noticed that the virtual experience removed most of the casual conversations and networking common during past in-person research workshops, a feature that many CRRAB members value. We attempted to stimulate this by spending a little more time at the beginning of videoconferences with icebreaker activities. Another challenge we noticed during the pandemic was a decrease in participation by researchers and health professionals in the meetings; while we normally have a patient to non-patient ratio of 1:1 at our meetings, for this project it was 3:1. The decline in non-patient participation might be the result of the added strains and stressors induced by the pandemic and experienced by those working in the health field (Unadkat and Farquhar 2020).

The reach and uptake of the booklet, as well as the effect of the media toolkit, are currently being evaluated by standard knowledge translation metrics; results will be reported in a future publication. Booklets continue to be distributed virtually through a variety of means. CRRAB members are actively working on developing a French version to serve the francophone population in Canada.

In summary, peer-to-peer recruitment efforts are an important part of generating interest and participation of patients as partners in research. In the absence of in-person recruitment efforts, the *Retinoblastoma Research and You!* booklet – by nature of being co-created by patients and containing patient stories and impact on research – serves as a stopgap to fill this need.

References

Canadian Institutes for Health Research (CIHR). 2014. *Strategy for Patient-Oriented Research – Patient Engagement Framework*. Retrieved July 28, 2021. <https://cihr-irsc.gc.ca/e/documents/spor_framework-en.pdf>.

Gelkopf, M.J., I. Avramov, R. Baddeliyanage, I. Ristevski, S.A. Johnson, K. Flegg et al. 2020. The Canadian Retinoblastoma Research Advisory Board: A Framework for Patient Engagement. *Research Involvement and Engagement* 6(1): 7. doi:10.1186/s40900-020-0177-8.

Ristevski, I., J. Robert, R. Baddeliyanage, R. Noronha, M. Gelkopf, K. Flegg et al. 2022. EMPOWER Retinoblastoma: Engaging Patient Partners in Solving the Top 10 Priorities for Eye Cancer Research in Canada. *Healthcare Quarterly* 24(Special Issue): 11–17. doi:10.12927/hcq.2022.26780.

Schilling, I., H. Behrens, C. Hugenschmidt, J. Liedtke, G. Schmiemann and A. Gerhardus. 2019. Patient Involvement in Clinical Trials: Motivation and Expectations Differ between Patients and Researchers Involved in a Trial on Urinary Tract Infections. *Research Involvement and Engagement* 5(1): 15. doi:10.1186/s40900-019-0145-3.

Unadkat, S. and M. Farquhar. 2020. Doctors' Wellbeing: Self-Care during the COVID-19 Pandemic. *BMJ* 368: m1150. doi:10.1136/bmj.m1150.

Vat, L.E., D. Ryan and H. Etchegary. 2017. Recruiting Patients as Partners in Health Research: A Qualitative Descriptive Study. *Research Involvement and Engagement* 3(1): 15. doi:10.1186/s40900-017-0067-x.

White, E., R. Baddeliyanage, F. Shaikh and H. Dimaras. 2019. Meaningful Patient Engagement in Research: Lessons from Retinoblastoma. *Pediatrics* 143(6): e20182166. doi:10.1542/peds.2018-2166.

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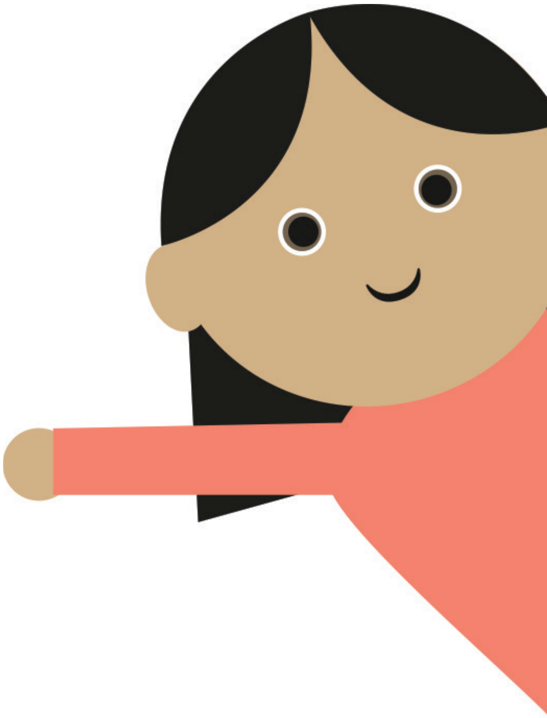


Partnering with Patients to Enhance Access to Kidney Transplantation and Living Kidney Donation

Kyla L. Naylor*, Susan Q. McKenzie*P, Amit X. Garg, Seychelle Yohanna and Jessica M. Sontrop

Abstract
Kidney transplantation gives many patients with kidney failure a longer and healthier life. Unfortunately, some transplant-eligible patients will never receive one. In this paper, we describe how patients and researchers collaborated on new strategies and programs to enhance access to kidney transplantation and living kidney donation. These efforts led to the creation of the Transplant Ambassador Program (TAP). TAP is a patient-led program that helps connect patients who have kidney failure to individuals who have successfully received a kidney transplant or donated a kidney. We also detail barriers, facilitators and lessons learned from engaging patients in research.

Key Points

- Patients and researchers collaborated to enhance access to kidney transplant. Efforts led to the creation of the Transplant Ambassador Program – a patient-led program that connects patients who have kidney failure with kidney transplant recipients.
- Several facilitators, barriers and lessons learned from engaging patients in research were identified. Some facilitators included biweekly meetings with patients and the research team to sustain engagement and keep an open line of communication.
- Patient partners were involved throughout the study, from study development to manuscript preparation, which enriched the research and ensured that the research is meaningful to patients and healthcare professionals.

Introduction

Patients with kidney failure have two main treatment options: dialysis or a kidney transplant. Compared with patients on dialysis, patients who receive a kidney transplant often live longer and experience an improved quality of life (Ortiz et al. 2014; Tonelli et al. 2011). Patients can receive a kidney from a deceased or living donor. Unfortunately, some transplant-eligible patients will never receive one. There are many barriers to receiving a kidney transplant, including a low number of kidneys available for transplant and a lack of knowledge about living kidney donation and transplantation among many patients and families (Garg 2018). Researchers and patients worked together to develop strategies to enhance access to kidney transplantation and living kidney donation. This paper describes our patient-oriented research (POR) project and the barriers, facilitators and lessons learned.

Development of Strategies to Enhance Access to Kidney Transplant

In 2015, a conference was held by Canadians Seeking Solutions and Innovations to Overcome Chronic Kidney Disease (Can-SOLVE CKD), a POR network that works to transform the care of people affected by kidney disease (https://cansolveckd.ca/). At the conference, patients with kidney disease, researchers, policy makers and healthcare professionals

* Lead co-authors.

P = Patient partner.

ranked improving access to living donor kidney transplantation as a top research priority. Then, in 2016, a workshop led by 19 patients was held in Ontario to understand patient-identified barriers and solutions to improve access to living donor kidney transplantation (living donor kidneys provide superior patient survival compared with deceased donor kidneys) (Axelrod et al. 2010; Getchell et al. 2017). A key solution suggested was to provide support to patients with kidney failure from kidney transplant recipients and living kidney donors. A subsequent collaboration between patients and researchers led to the development of the Transplant Ambassador Program (TAP) (transplantambassadors.ca), a patient-led, volunteer-driven support program for patients with kidney failure. With initial funding from Can-SOLVE CKD, we were able to make TAP a reality. Additional funding was received from the Ontario Strategy for Patient-Oriented Research (SPOR) SUPPORT Unit (OSSU) EMPOWER award to adapt and optimize TAP's online operation in response to the COVID-19 pandemic and to prepare a plan to implement and sustain TAP at CKD programs across Ontario.

TAP

> As a living donor, I see the importance of TAP every time I speak with a patient or a potential donor. When we share our lived experiences, we demystify the process of transplant and vividly show an example of life after the operation. People are eager to speak with someone with first-hand experience. It helps them feel less anxious and more confident in their decision to pursue kidney transplant. (Living kidney donor and transplant ambassador)

FIGURE 1.
Transplant ambassadors

Transplant ambassadors wear bright green vests that indicate whether they are a kidney transplant recipient or a living kidney donor. The back of the vest contains a print invitation to "Ask me about kidney transplantation." The vests make it easy for patients to identify transplant ambassadors.

TAP connects patients who have kidney disease to transplant ambassadors: individuals who have successfully received a kidney transplant or donated a kidney. Transplant ambassadors share their stories, offer practical advice and emotional support, share strategies on how to discuss living kidney donation with family and friends, connect patients with educational resources and help guide and motivate patients through the transplant evaluation process (Figure 1). Compared to healthcare professionals, they can often spend more time discussing transplantation and provide inspiration and hope to patients with kidney failure. TAP was initially developed by a kidney transplant recipient and by an individual whose mother received a living kidney donation from her father.

Enhance Access to Kidney Transplantation and Living Kidney Donation Strategy

Based on feedback from the patient-led workshop and evidence on the benefits of peer-support programs (Marlow et al. 2016; Sullivan et al. 2012), patients and researchers decided to include TAP as a key component in a province-wide, multi-component quality improvement intervention: The Enhance Access to Kidney Transplantation and Living Kidney Donation (EnAKT LKD) strategy. In brief, EnAKT LKD is a cluster-randomized clinical trial being tested in Ontario's 27 CKD programs, which care for ~28,000 patients with kidney failure. The four main intervention components include (1) quality improvement teams; (2) education for staff, patients and living kidney donor candidates; (3) TAP; and (4) CKD program-level performance reports on transplant metrics. The overall objective is to determine if the intervention enables more patients to complete key steps toward receiving a kidney transplant. The intervention (including TAP) began in November 2017. Further details on the EnAKT LKD trial can be found in the published protocol and in Figure 2 (Yohanna et al. 2021). The trial period ended on December 31, 2021.

Transplant Ambassador Program's Progress to Date

Since its inception, TAP has recruited over 100 transplant ambassadors in 13 of Ontario's 27 CKD programs (TAP was offered to all CKD programs starting in January 2022). Ambassadors have had thousands of interactions with patients and families. Although we do not yet know the impact of TAP on increasing access to transplant (results from the EnAKT LKD trial are expected in 2023), patients, healthcare professionals and transplant ambassadors have commented on the positive impact that they perceive TAP has had on patients. To improve interpretation of trial findings and future implementation, we are also conducting a mixed-methods process evaluation, which consists of surveys and interviews with healthcare providers at the CKD programs. Results from the process evaluation will provide further insights into the use of TAP.

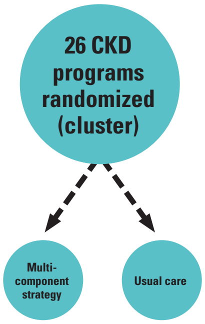
FIGURE 2.
Summary of the EnAKT LKD trial

EnAKT LKD

EnAKT LKD Summary:
A cluster randomized clinical trial designed to determine if a quality improvement intervention provided in Ontario's chronic kidney disease (CKD) programs (vs. usual care) enables more patients with CKD to complete more steps toward receiving a kidney transplant.

The EnAKT LKD trial comprises four main components:

1. Local quality improvement teams and administrative support
2. Education and resources for staff, patients and living kidney donor candidates
3. Program-level performance reports and oversight by program leaders
4. Support from kidney transplant recipients and living kidney donors through TAP



Source: Yohanna et al. 2021.

Patient Partnership

In addition to patients developing and leading TAP, patients helped formulate the EnAKT LKD research questions, select and design the intervention components and deliver intervention components. Briefly, the patient partners (1) are members of the quality improvement teams at each CKD program in the study; (2) helped review, create and select educational materials and connect patients to these resources; (3) review quarterly performance reports on transplant metrics to monitor their CKD program's progress; and (4) collect the data on process measures to include in these performance reports. Two of the patient partners are members of the provincial priority panel to improve access to kidney transplant. Patients continue to co-author all related manuscripts. Once results are available, patients will play a key role in interpreting the results and disseminating the results through patient and family advisory councils and through TAP.

To foster engagement with this work, patients meet regularly. For example, transplant ambassadors who have taken on a leadership role at their CKD program meet monthly, all transplant ambassadors meet twice a year, three transplant ambassadors attend monthly provincial calls with the 13 CKD-program quality improvement teams and one patient representative attends monthly calls with the core research team for the EnAKT LKD trial. Including patients on the core research team has been integral to ensuring that patients recognize themselves as equal members of the project and ensuring that patient input is incorporated into the project.

Researchers have commented that having patients involved in all parts of the research process and leading a key component of the intervention has generated unique insights about improving access to transplant. For example, through conversations between patients and transplant ambassadors, TAP has improved the patient experience by bringing forward patient-identified barriers to transplant. For example, some potential donors had commented that they had to wait a long time before receiving a call from the living kidney donor coordinator; transplant ambassadors brought this issue forward, which resulted in a solution to reduce wait times. Patients have commented that having a strong leadership role on the research team has provided them with a sense of empowerment and responsibility. Patients who have experienced kidney failure are often the most passionate to drive change in the transplant process and, therefore, it makes sense to have a group of motivated people lead TAP. Clinicians on the quality improvement teams have felt that having patients on the team has kept them accountable to improve access to kidney transplants and challenged them to change processes. Patients with kidney failure have commented that transplant ambassadors have been invaluable with helping them through the difficult journey to receiving a transplant, and the educational events hosted and attended by transplant ambassadors have been well received by patients.

Impact of the COVID-19 Pandemic

The COVID-19 pandemic forced all interactions between transplant ambassadors and patients to be virtual (i.e., via telephone or Zoom for Healthcare). There have been challenges with the switch to virtual, including the virtual environment being less engaging for patients and their families. The inability of ambassadors to be present in the clinic has impacted patient referrals to TAP. Patients are often less likely to engage with transplant ambassadors outside the clinic setting. For patients who engage virtually, transplant ambassadors have been given additional training on strategies to build rapport with patients in this new setting. For the broader EnAKT LKD strategy, we had to change all in-person meetings to a virtual format, which reduced opportunities for informal discussions and networking between healthcare professionals and patient partners across CKD programs.

Despite the challenges presented by the pandemic, the patients leading TAP continued to provide support to patients and even made improvements to the existing program. The new virtual environment provided an opportunity to connect ambassadors with patients in different locations. For example, a patient in London, ON, can now connect with an ambassador in Sudbury, ON. The pandemic also generated the idea to gather profile information on all the ambassadors (e.g., age, gender) so patients can connect with ambassadors who may have similar life experiences. During this time, TAP also expanded its recruitment using social media. One of the goals of this expansion was to create a more diverse group of transplant ambassadors and increase TAP's visibility.

Barriers and Facilitators to Patient Engagement

Despite our overall success with engaging patients in this research, there have been some barriers. First, it has been difficult to embed a patient-led program (TAP) into the workflow of the CKD programs. One reason for this difficulty is that during the pandemic, healthcare providers who were champions of TAP were often redeployed to other departments. To help embed TAP into the workflow, we gave presentations to the CKD program staff on TAP and encouraged healthcare professionals on the quality improvement teams to champion TAP to their colleagues. Second, ensuring full cultural, gender and kidney failure/transplant experience diversity across transplant ambassadors and patient partners on the research team has been difficult. To help overcome this barrier, TAP has increased its online presence to expand recruitment efforts and has recruited volunteers to help develop a diversity strategy for TAP, which will include diversity training for all ambassadors. Finally, it has been difficult to ensure that the CKD programs recognize patients as key members of the quality improvement teams. Patients sometimes struggle to describe barriers identified by other patients and donors in a way that can be acted upon by healthcare professionals. This has improved over time as healthcare professionals became more comfortable with having patients make these suggestions.

Despite some barriers, there have been multiple facilitators to engaging patients in this work. First, several of the patients had worked with the researchers on prior projects, establishing trust and a rapport. This trust allowed for an open line of communication between the researchers and patients to discuss any issues that might arise. This trust also allowed patients to develop, lead and run an entire component of the intervention (i.e., TAP). This contrasts with many other POR projects where researchers lead the research and patients support it. An assumption some researchers have is that patients should only be required to put in minimal effort; this may be because of concerns about the burden of disease in patients, misconceptions that POR is simply having a patient tell their story or because many patients are volunteers. While we must be cognisant of all members of the research team's time, TAP is proof that many patients are willing to put in hours of effort with patients having a vested interest in improving the system. Second, including patients in all components of the intervention has increased the quality and relevance of the intervention. Specifically, having patients lead an entire component of an intervention has allowed patients to be involved with identifying barriers and solutions and then actively break down those barriers. Third, having patients lead a component of the intervention has allowed for additional barriers and solutions to transplant be identified through ambassador conversations with patients. Finally, biweekly meetings with the core research team, including patients, has been key to sustaining patient engagement throughout this multi-year trial.

Conclusion

Patients have successfully developed and led a key component of a quality improvement intervention to enhance access to kidney transplant. Patient partners were involved throughout the study, from quality improvement intervention development to manuscript preparation, which has undoubtedly enriched the research and has ensured that the research is meaningful to patients and healthcare professionals. HQ

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References

Axelrod, D.A., K.P. McCullough, E.D. Brewer, B.N. Becker, D.L. Segev and P. Rao. 2010. Kidney and Pancreas Transplantation in the United States, 1999–2008: The Changing Face of Living Donation. *American Journal of Transplantation* 10(4 Pt 2): 987–1002. doi:10.1111/j.1600-6143.2010.03022.x.

Garg, A.X. 2018. Helping More Patients Receive a Living Donor Kidney Transplant. *Clinical Journal of The American Society of Nephrology* 13(12): 1918–23. doi:10.2215/CJN.00760118.

Getchell, L.E., S.Q. McKenzie, J.M. Sontrop, J.S. Hayward, M.K. McCallum and A.X. Garg. 2017. Increasing the Rate of Living Donor Kidney Transplantation in Ontario: Donor- and Recipient-Identified Barriers and Solutions. Canadian *Journal of Kidney Health and Disease* 4: doi:10.1177/2054358117698666.

Marlow, N.M., A.S. Kazley, K.D. Chavin, K.N. Simpson, W.Balliet and P.K. Baliga. 2016. A Patient Navigator and Education Program for Increasing Potential Living Donors: A Comparative Observational Study. *Clinical Transplantation* 30(5): 619–27. doi:10.1111/ctr.12728.

Ortiz, F., P. Aronen, P.K. Koskinen, R.K. Malmström, P. Finne, E.O. Honkanen et al. 2014. Health-Related Quality of Life after Kidney Transplantation: Who Benefits the Most? *Transplant International* 27(11): 1143–51. doi:10.1111/tri.12394.

Sullivan, C., J.B. Leon, S.S. Sayre, M. Marbury, M. Ivers, J.A. Pencak et al. 2012. Impact of Navigators on Completion of Steps in The Kidney Transplant Process: A Randomized, Controlled Trial. *Clinical Journal of the American Society of Nephrology* 7(10): 1639–45. doi:10.2215/CJN.11731111.

Tonelli, M., N. Wiebe, G. Knoll, A. Bello, S. Browne, D. Jadhav et al. 2011. Systematic Review: Kidney Transplantation Compared with Dialysis in Clinically Relevant Outcomes. *American Journal of Transplantation* 11(10): 2093–109. doi:10.1111/j.1600-6143.2011.03686.x.

Yohanna, S., K.L. Naylor, I. Mucsi, S. McKenzie, D. Belenko, P.G. Blake et al. 2021. A Quality Improvement Intervention to Enhance Access to Kidney Transplantation and Living Kidney Donation (EnAKT LKD) in Patients with Chronic Kidney Disease: Clinical Research Protocol of a Cluster-Randomized Clinical Trial. *Canadian Journal of Kidney Health and Disease* 8. doi:10.1177/2054358121997266.

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Building a Platform for Meaningful Patient Partnership to Accelerate "Bench-to-Bedside" Translation of Promising New Therapies

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Abstract

Engaging patients as partners in the design and execution of early-phase clinical trials offers a unique opportunity to ensure patient perspectives are considered. Here we describe our experience partnering with four individuals with lived experience of blood cancer to co-develop documents and services to support participants of an early-phase trial. Through regular team meetings, patient partners co-developed a visual informed consent document and a non-technical summary of the informed consent document to facilitate participant understanding of trial procedures. Overall, patient partners highlighted important trial components that would not have been identified without their input.

Key Points

- Patient partners identified areas of a clinical trial that needed more attention and informed the development of patient-facing informed consent documents and a policy brief.
- Co-development of the project was facilitated through regular virtual meetings and a final face-to-face meeting.
- All team members described working together as a positive experience.

Patient Engagement Approach and Structure

Background and aim of patient engagement

Canadian-Led Immunotherapies in Cancer trial (CLIC-01) is an early-phase clinical trial assessing chimeric antigen receptor T (CAR-T) cell therapy. CAR-T cell therapy is a promising immunotherapy to treat hematologic malignancies where a patient's own T cells are genetically engineered to identify and kill cancer cells (Lee et al. 2012). CAR-T cell therapy has demonstrated efficacy as a novel treatment (Grigor et al. 2019; Jackson et al. 2016; Lee et al. 2015). In order to create a patient-centred trial, our team had previously collaborated with two patient partners on various projects to compile evidence for the co-development of the CLIC-01 trial protocol (NCT #03765177). This research program was titled "Getting Better Outcomes with Chimeric Antigen Receptor T-Cell Therapy" (GO-CART) and has been summarized elsewhere (Foster et al. 2020). When the GO-CART program came to a close, we wanted to extend engagement beyond protocol development to further improve trial processes and support clinical trial participants. To achieve this, we initiated a project funded by the Ontario Strategy for Patient-Oriented Research (SPOR) SUPPORT Unit (OSSU) Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Award. We worked with patient partners to enhance patient-facing informed consent documents, plan a peer support panel and co-develop a policy brief.

P = Patient partner.

Recruitment of patient partners

Four patient partners were recruited to our research group. One patient partner continued their involvement from the GO-CART program into the OSSU EMPOWER Award initiative and, as a result, had a detailed understanding of the CLIC-01 trial. Another patient partner reached out to our research group after hearing about the GO-CART program at a BioCanRx Summit for Cancer Immunotherapy conference and expressed interest in joining our team (BioCanRx is a federal government–funded Network of Centers of Excellence that supports partnerships among industries, academic institutions and patient organizations) (https://biocanrx.com/). She brought a completely novel perspective as she had participated in a similar trial in the US when the immunotherapy was under investigation there. Two additional patient partners were identified and recruited through the circulation of an advertisement by the Leukemia and Lymphoma Society of Canada (https://www.llscanada.org/).

The onboarding process and team structure

All patient partners were formally onboarded to the research team. This process included a meeting (in-person or virtual) with research assistants, where we provided details of the previous GO-CART project, an overview of the CLIC-01 trial and how the OSSU EMPOWER project would support previous work. Team members' headshots and biographies were presented to provide patient partners with information on team members' interests and expertise. We also allocated a portion of the meeting to discuss patient partners' expectations, availability and what they hoped to gain from the experience. To ensure that engagement was supported throughout the research project, we co-developed a Terms of Reference document outlining the agreed-upon roles and responsibilities of each team member (Alberta SPOR SUPPORT Unit 2018).

Planned engagement

From inception, the aim of patient engagement was to obtain the patients' perspectives on how to improve the experience for CLIC-01 trial participants. Though potential areas for collaboration were identified in advance (e.g., informed consent documents, peer support panel, policy brief), specific details were left open for discussion as we wanted patient partners to play a role in the planning process. We scheduled bimonthly team meetings, where we worked together to (1) identify areas and elements of the CLIC-01 trial that required support and (2) co-develop services and documents to meet patient needs. Additionally, we aimed to develop a policy brief on patient engagement in early-phase clinical trials to support researchers and institutions.

Meetings and engagement activities

To finalize project deliverables, we organized a full-day face-to-face meeting (November 2019), where all team members could meet in person as well as virtually to discuss the project's progress and the next steps.

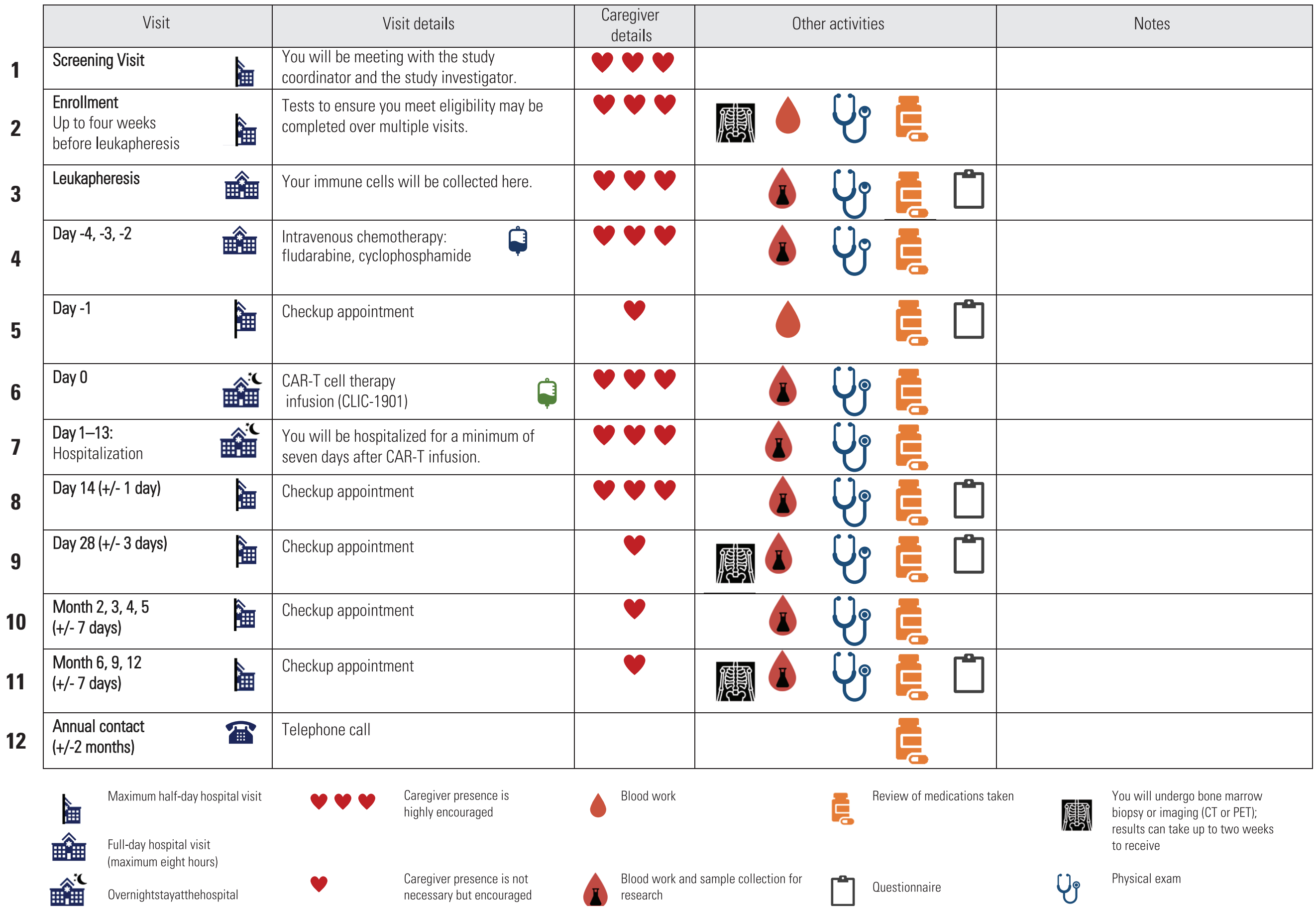
Our first engagement activity focused on supporting the informed consent process. Because CLIC-01 is an early-phase clinical trial, the informed consent documents can be lengthy and technical (Brehaut et al. 2012, 2015; Somers et al. 2017). Furthermore, potential trial participants are unlikely to have other available treatment options. These two trial characteristics further highlighted the informed consent process as an important trial component wherein participants may need additional support in understanding what participation in the CLIC-01 trial entails. Patient partners suggested that more user-friendly documents could help support participants' understanding of the trial and allow them to easily share information about the trial with friends and family. To address this, we co-developed a visual informed consent document and a one-page non-technical summary. As a group, we went through the informed consent document to identify sections that could be simplified and presented graphically. The schedule of assessments was highlighted as an important component because it outlines involvement (i.e., hospital visits, procedures). We formatted the schedule of assessments as a one-page graphical timeline where procedures were represented by colourful icons (Figure 1).

One patient partner highlighted the supportive role played by caregivers because they are usually present for procedures and hospital visits. In order to address this issue, we added an icon to denote the length of hospital visits, which would allow caregivers to anticipate visit length and provide them with the opportunity to make scheduling arrangements if necessary. At the close of the project, visual informed consent documents were finalized and sent for review by the Research Ethics Board (REB).

We also co-developed a policy brief outlining ways that funding agencies can encourage uptake of patient engagement in the development and conduct of early-phase clinical trials. We co-designed three approaches that funding agencies can adopt to encourage researchers to engage patient partners in the development and conduct of early-phase clinical trials, including (1) making web-based educational resources on patient engagement available, (2) implementing a checkbox on grant applications to indicate the intent to engage patients and requesting a written report from successful applicants as an interim analysis and (3) incorporating patient engagement within the funding agency. Further details will be outlined in our policy brief (registered on Open Science Framework: https://osf.io/6jequ/).

FIGURE 1.
Visual informed consent document

CLIC-01
Canadian-Led
Immunotherapies in Cancer



	Visit		Visit details		Caregiver details	Other activities	Notes
1	Screening Visit	🏢	You will be meeting with the study coordinator and the study investigator.		♥ ♥ ♥		
2	Enrollment Up to four weeks before leukapheresis	🏢	Tests to ensure you meet eligibility may be completed over multiple visits.		♥ ♥ ♥	🩻 💧 🩺 💊	
3	Leukapheresis	🏥	Your immune cells will be collected here.		♥ ♥ ♥	🩸 🩺 💊 📋	
4	Day -4, -3, -2	🏥	Intravenous chemotherapy: fludarabine, cyclophosphamide	💉	♥ ♥ ♥	🩸 🩺 💊	
5	Day -1	🏢	Checkup appointment		♥	💧 💊 📋	
6	Day 0	🏥🌙	CAR-T cell therapy infusion (CLIC-1901)	💉	♥ ♥ ♥	🩸 🩺 💊	
7	Day 1–13: Hospitalization	🏥🌙	You will be hospitalized for a minimum of seven days after CAR-T infusion.		♥ ♥ ♥	🩸 🩺 💊	
8	Day 14 (+/- 1 day)	🏢	Checkup appointment		♥ ♥ ♥	🩸 🩺 💊 📋	
9	Day 28 (+/- 3 days)	🏢	Checkup appointment		♥	🩻 🩸 🩺 💊 📋	
10	Month 2, 3, 4, 5 (+/- 7 days)	🏢	Checkup appointment		♥	🩸 🩺 💊	
11	Month 6, 9, 12 (+/- 7 days)	🏢	Checkup appointment		♥	🩻 🩸 🩺 💊 📋	
12	Annual contact (+/-2 months)	☎	Telephone call			💊	

🏢 Maximum half-day hospital visit
🏥 Full-day hospital visit (maximum eight hours)
🏥🌙 Overnightstayatthehospital
♥ ♥ ♥ Caregiver presence is highly encouraged
♥ Caregiver presence is not necessary but encouraged
💧 Blood work
🩸 Blood work and sample collection for research
💊 Review of medications taken
📋 Questionnaire
🩻 You will undergo bone marrow biopsy or imaging (CT or PET); results can take up to two weeks to receive
🩺 Physical exam

A third engagement activity was to organize a peer support panel consisting of individuals with lived experience of hematologic malignancies or having experience participating in a clinical trial. In fact, one patient partner had previous experience as a peer support mentor, and their perspective informed the overall direction of the peer support plan. For example, an online platform was identified as the preferred format over mentor–mentee conference calls to avoid emotionally burdening mentors.

Unfortunately, we encountered several hurdles to developing an online peer support panel, including preserving anonymity and ensuring that the platform would be accessible outside of the hospital setting while maintaining confidentiality. As a result, we decided that partnering with a patient organization that has an established peer support infrastructure might help us overcome these hurdles. We plan to continue exploring this approach in future work. Further details of engagement can be found in Appendix 1: Table A1, available online at www.longwoods.com/content/26770.

Assessment of engagement

In order to improve patient-partner engagement in future initiatives, we assessed engagement methods at the face-to-face meeting by disseminating a survey to patient partners. The final evaluation consisted of nine questions (a combination of surveys developed by Patients Canada [Maybee et al. 2016] and the Patient and Family Advisory Council at The Ottawa Hospital [https://www.ottawahospital.on.ca/en/clinical-services/deptpgrmcs/programs/cancer-program/patient-and-family-advisory-council/]). Through the questionnaire, our patient partners expressed some issues with the combined virtual and in-person attendance to our last meeting. We recruited two additional patient partners after the project started, but the research team did not have the funding to provide travel reimbursement for the additional patient partners to attend the face-to-face meeting. This caused an unintended divide between local and non-local patient partners; non-local patient partners expressed feeling disconnected from the rest of the team at the face-to-face meeting.

> Although challenging, all recruitment should take place as early as possible in the program. It would have been more effective if all four patient partners were able to attend the full day face-to-face meeting. It was challenging for those who had to attend by teleconference. (Terry Hawrysh)

Obstacles

Despite the successes of our project, we encountered several obstacles to engagement that should be noted. First, it was challenging to identify patient partners within the timeline of the program. Two patient partners were involved from the onset of the study; however, it was difficult to identify and onboard additional patient partners. With that said, partnering with an established organization (e.g., The Leukemia & Lymphoma Society of Canada in our case [https://www.bloodcancers.ca/]) to circulate an advertisement was an effective strategy for patient-partner recruitment.

Second, patient partners expressed disappointment with the delay in the implementation of the informed consent resources. Due to the COVID-19 pandemic, the REB shifted focus to approving COVID-related research materials and projects. As a result, it took several months to gain approval to include the documents as a part of the informed consent process.

> [It was disappointing] seeing that the initiatives that we tried to start didn't come to fruition because of various hurdles. This prevented [us from] seeing a direct impact of our involvement in the program. (Stefany Dupont)

In retrospect, this disappointment could have been mitigated by providing regular updates (e.g., monthly) on the project's progress and including the visual informed consent form with the initial clinical trial submission to the REB to maximize the use of these documents. The documents have now been approved for use and will be implemented for future patients recruited to the clinical trial.

Equity, diversity and inclusion

Given the obstacles faced when identifying and recruiting patient partners to the research team, equity, diversity and inclusion were not at the forefront. With that said, it is clear that inclusivity of diverse perspectives is of the utmost importance in patient engagement and cancer research (i.e., cancer does not discriminate). From our experience with recruitment and our improved understanding of organizational roles in identifying interested patient partners, we may be able to prioritize these issues in future engagement efforts.

Compensation and acknowledgement

All travel expenses (transportation, accommodations, parking, meals, etc.) were reimbursed for patient partners who attended the face-to-face meeting in person (according to institutional policies at The Ottawa Hospital). Two local patient partners were offered compensation for attending the full day face-to-face meeting in person. The method of compensation and amount were informed by the SPOR Evidence Alliance Patient Partner Appreciation Policy and Protocol (SPOR Networks in Chronic Diseases and the PICHI Network 2018). All patient partners were acknowledged as co-authors on manuscripts.

Next Steps

The OSSU EMPOWER Award project inspired the development of a new program, Making Patient Partnerships A Reality in Very Early Phase Clinical TriaLs (MARVEL) funded by BioCanRx (https://biocanrx.com/csei7-lalu). The MARVEL program will aim to develop a patient engagement platform to facilitate engagement throughout the development and conduct of four unique early-phase research initiatives (including the ongoing CLIC-01 trial).

Patient partners identified patient needs that we could not have identified without their perspectives ...

Our Experience with Patient Engagement

Overall, our experience was positive. Patient partners identified patient needs that we could not have identified without their perspectives (e.g., importance of emotional support). Patient partners stated that their involvement in the OSSU EMPOWER project was positive, gratifying and educational. They noted that they believed their contributions were valued and had great influence on the course of the project and the final products.

> As a patient living with blood cancer, I was especially encouraged with the work done on simplifying the informed consent process through the creation of lay summaries and a visual rendition of what to expect during the trial. Undergoing a clinical trial can be an overwhelming experience for patients and their families; the support methods developed by the team will go a long way to help them. This should greatly benefit potential trial participants in their understanding and evaluation of the research approach, time commitment, costs and risks associated with trial participation. (Terry Hawrysh)

> It was an opportunity for me to "give back" and ensure that people facing the same situation I faced in 2017 [cancer diagnosis] had a smooth experience. (Stefany Dupont)

We have compiled the following lessons learned that would help guide future initiatives:

- Patient engagement activities were identified organically. At the onset of the project, specific details were left open for team discussion, which generated most of the project deliverables.
- It is important to have an open discussion about patient partners' availability, interest and acknowledgement (compensation, reimbursement, co-authorship, etc.) while providing sufficient background on the project.
- Terms of reference documents are helpful in setting expectations.
- Virtual peer support panels require extensive legal, technological and administrative support, which were prohibitive in our project.
- Maintaining an open line of communication is important (e.g., circulating regular project updates, and communicating how patient-partner feedback was implemented).

Key Messages and Implications

Early-phase clinical trials offer a unique opportunity for patient partners to provide input early in the "translational" pipeline of therapy development. Engagement at this phase of research is particularly impactful because it allows for the streamlining of researchers' priorities and those of the ultimate end-users of the technology. Incorporating the patient's perspective may help improve chances of a successful trial (reducing obstacles, providing necessary supports, clear and understandable information, etc.) and overall translation of the therapy to practice, which is essential for early-phase clinical trials (Crocker et al. 2018; Gasson et al. 2015). Here, patient partners were able to identify areas of the clinical trial that needed more attention. This would not have been accomplished without their input. HQ

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References

Alberta SPOR SUPPORT Unit. 2018. Terms of Reference Template. Retrieved September 7, 2021. <https://absporu.ca/resource/terms-of-reference-template/>.

Brehaut, J.C., K. Carroll, G. Elwyn, R. Saginur, J. Kimmelman, K. Shojania et al. 2012. Informed Consent Documents Do Not Encourage Good-Quality Decision Making. *Journal of Clinical Epidemiology* 65(7): 708–24. doi:10.1016/j.jclinepi.2012.01.004.

Brehaut, J.C., K. Carroll, G. Elwyn, R. Saginur, J. Kimmelman, K. Shojania et al. 2015. Elements of Informed Consent and Decision Quality Were Poorly Correlated in Informed Consent Documents. *Journal of Clinical Epidemiology* 68(12): 1472–80. doi:10.1016/j.jclinepi.2015.03.002.

Crocker, J.C., I. Ricci-Cabello, A. Parker, J.A. Hirst, A. Chant, S. Petit-Zeman et al. 2018. Impact of Patient and Public Involvement on Enrolment and Retention in Clinical Trials: Systematic Review and Meta-Analysis. *BMJ* 363: k4738. doi:10.1136/bmj.k4738.

Foster, M., D.A. Fergusson, T. Hawrysh, J. Presseau, N. Kekre, S. Schwartz et al. 2020. Partnering with Patients to Get Better Outcomes with Chimeric Antigen Receptor T-Cell Therapy: Towards Engagement of Patients in Early Phase Trials. *Research Involvement and Engagement* 6: 61. doi:10.1186/s40900-020-00230-5.

Gasson, S., J. Bliss, M. Jamal-Hanjani, M. Krebs, C. Swanton and M. Wilcox. 2015. The Value of Patient and Public Involvement in Trial Design and Development. *Clinical Oncology: A Journal of the Royal College of Radiologists* 27(12): 747–49. doi:10.1016/j.clon.2015.06.020.

Grigor, E.J.M., D. Fergusson, N. Kekre, J. Montroy, H. Atkins, M.D. Seftel et al. 2019. Risks and Benefits of Chimeric Antigen Receptor T-Cell (CAR-T) Therapy in Cancer: A Systematic Review and Meta-Analysis. *Transfusion Medicine Reviews* 33(2): 98–110. doi:10.1016/j.tmrv.2019.01.005.

Jackson, H.J., S. Rafiq and R.J. Brentjens. 2016. Driving CAR T-Cells Forward. *Nature Reviews. Clinical Oncology* 13(6): 370–83. doi:10.1038/nrclinonc.2016.36.

Lee, D.W., D.M. Barrett, C. Mackall, R. Orentas and S.A. Grupp. 2012. The Future Is Now: Chimeric Antigen Receptors as New Targeted Therapies for Childhood Cancer. *Clinical Cancer Research* 18(10): 2780–90. doi:10.1158/1078-0432.CCR-11-1920.

Lee, D.W., J.N. Kochenderfer, M. Stetler-Stevenson, Y.K. Cui, C. Delbrook, S.A. Feldman et al. 2015. T Cells Expressing CD19 Chimeric Antigen Receptors for Acute Lymphoblastic Leukaemia in Children and Young Adults: A Phase 1 Dose-Escalation Trial. *The Lancet* 385(9967): 517–28. doi:10.1016/S0140-6736(14)61403-3.

Maybee, A., B. Clark, A. McKinnon, and E.N. Angl. 2016. *Patients as Partners in Research: Patient/Caregiver Surveys*. Patients Canada. Retrieved August 23, 2021. <https://ossu.ca/wp-content/uploads/EvaluationSurveysPatient_2016.pdf>.

Somers, R., C. Van Staden and F. Steffens. 2017. Views of Clinical Trial Participants on the Readability and Their Understanding of Informed Consent Documents. *American Journal of Empirical bioethics* 8(4): 277–84. doi:10.1080/23294515.2017.1401563.

SPOR Networks in Chronic Diseases and the PICHI Network. 2018. Recommendations on Patient Engagement Compensation. Retrieved August 23, 2021. <https://diabetesaction.ca/wp-content/uploads/2018/07/TASK-FORCE-IN-PATIENT-ENGAGEMENT-COMPENSATION-REPORT_fiNAL-1.pdf>.

Staniszewska, S., J. Brett, I. Simera, K. Seers, C. Mockford, S. Goodlad et al. 2017. GRIPP2 Reporting Checklists: Tools to Improve Reporting of Patient and Public Involvement in Research. *BMJ* 358: j3453. doi:10.1136/bmj.j3453.

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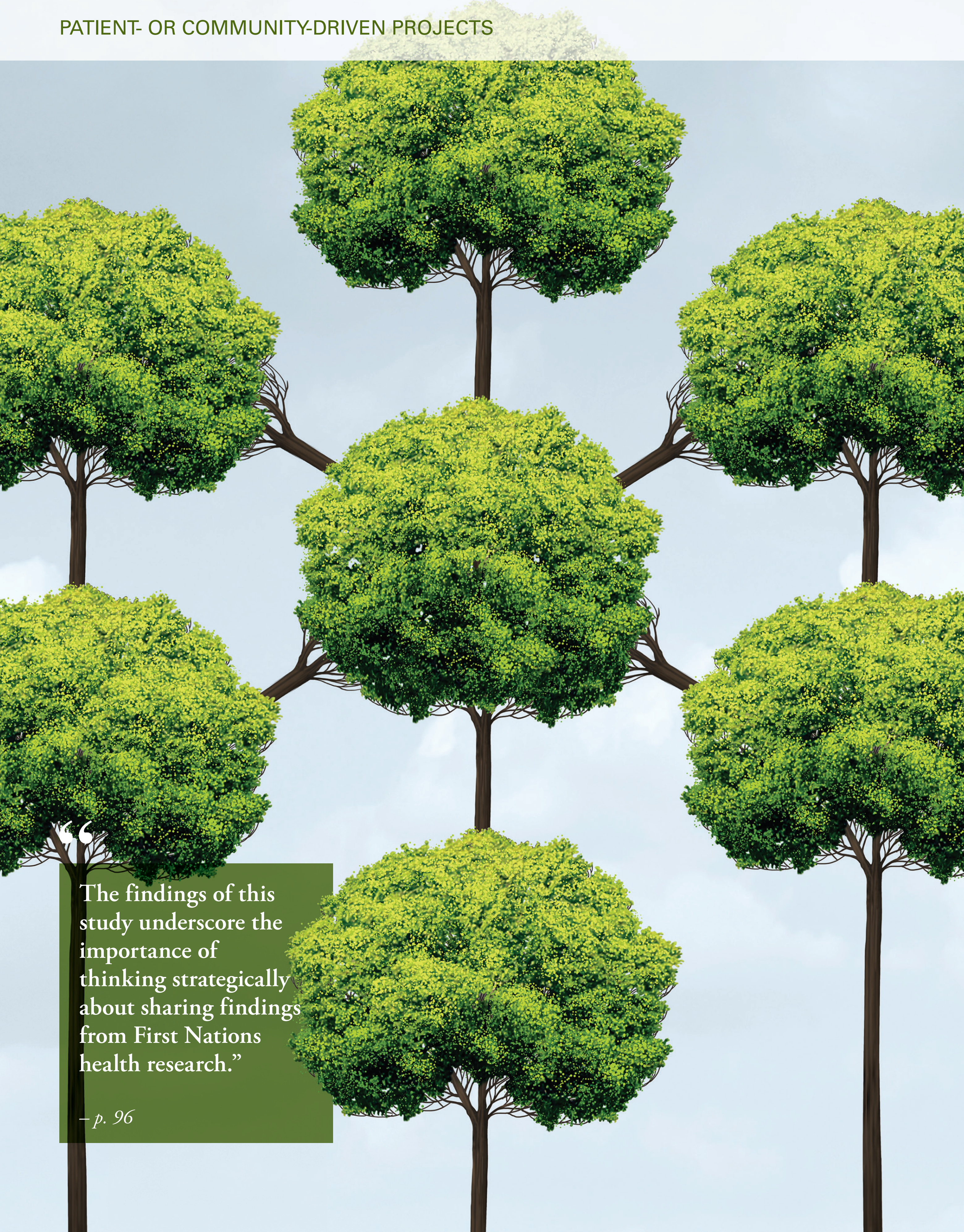

> “The findings of this study underscore the importance of thinking strategically about sharing findings from First Nations health research.”
>
> *– p. 96*

Patient Engagement in a Multi-Stakeholder Workshop to Plan the Collection of Patient-Oriented Outcomes for Children with Inherited Metabolic Diseases

Kylie Tingley, Maureen Smith[P], Nicole Pallone[P], Pranesh Chakraborty and Beth K. Potter

Abstract

Building on a study to develop core outcome sets for children with rare inherited metabolic diseases, the purpose of this workshop was to inform the design of longitudinal pediatric registries that support registry-based clinical trials. This workshop was co-designed by two patient/family partner investigators and attended by two family advisors who received preparatory training. Patient partners and advisors recommended integrating the collection of registry data into everyday life and highlighted the importance of transparent communication and attention to the issue of integration of patient-reported data into clinical care. We propose a need to explore strategies for engaging patients in post-project knowledge translation.

Key Points

- Building on previous work, we successfully co-designed a workshop with patient/family partner investigators to inform the design of longitudinal pediatric registries that support registry-based clinical trials.
- Patient partners and advisors recommended integrating the collection of registry data into everyday life and highlighted the importance of transparent communication and attention to the integration of patient-reported data into clinical care.
- We attribute much of our success and sustainability of our partnership to co-developing a comprehensive patient engagement strategy that included regular feedback to patient partners/advisors about the positive impact of their contributions.

Introduction

Core outcome sets (COSs) are an agreed list of the minimum standardized outcomes that should be measured and described for all clinical trials in a specific disease area (Williamson et al. 2017). Ideally, COSs should be developed by a multi-stakeholder team that includes patients and family members, healthcare providers, health policy decision makers and methodologists (Williamson et al. 2017). Incorporating patients and family members as partners in COS development is key so that outcomes included in a COS are reflective of what is important to those affected by the disease and those who stand to benefit most from clinical research (Young and Bagley 2016). Our team established the first COSs for two inherited metabolic diseases (IMDs) in children: phenylketonuria (PKU) and medium chain acyl-CoA dehydrogenase (MCAD) deficiency (Pugliese et al. 2021).

Overview of the COS study

Methods and findings from our COS development study are described elsewhere (Potter et al. 2017; Pugliese et al. 2020, 2021). Briefly, following guidance from the Core Outcomes Measures in Effectiveness Trials (COMET) Initiative (Williamson et al. 2017), we systematically reviewed published studies for each condition to identify candidate outcomes

P = Patient partner.

(Pugliese et al. 2020). Subsequently, parents of children with PKU or MCAD deficiency, clinicians and policy advisors participated in a multi-round Delphi consensus survey (Pugliese et al. 2021). Final COSs were selected by discussion and voting at an in-person multi-stakeholder meeting, where more than 30% of the attendees were patients and caregivers (Pugliese et al. 2021).

Our patient engagement strategy for the COS study has also been described in another study (Vanderhout et al. 2021). Briefly, two patient or family member partners were engaged as co-investigators throughout the study and a Family Advisory Forum (FAF), which included seven parents of children with IMDs, were engaged at key stages. Patient partner investigators contributed to protocol development, co-designed and co-led all patient engagement activities, identified challenges and solutions to incorporating patient perspectives and communicated with FAF members. FAF members advised and provided feedback at several points during the study, including reviewing patient/family member–facing materials and contributing to outcome selection. Our team adapted existing resources from the COMET Initiative on patient and public engagement to support this work (COMET Initiative 2021). In addition, the principal investigator (BKP) and one patient partner investigator (MS) attended training on patient-oriented research from the Ontario Strategy for Patient-Oriented Research (SPOR) SUPPORT Unit (OSSU).

Planning the implementation of COSs to support registry-based clinical trials

The next phase of our work involved identifying barriers and facilitators to the implementation of our COSs. We were specifically interested in the potential for the outcomes to be collected across Canadian centres in new disease registries designed to support the development and implementation of registry-based randomized trials (Li et al. 2016). Registry-based randomized trials use patient registries as the platform for recruiting clinical trial participants and to optimize trial data collection (Li et al. 2016; Mathes et al. 2018). To integrate our COSs in registries to support trials, we needed to better understand preferences of stakeholders, including patients and families, regarding their collection and use. One of our patient partners suggested that we co-develop an OSSU Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Award application, which was successful and helped fund a knowledge translation workshop to gain this multi-stakeholder perspective. Here, we report the methods and results from the workshop, emphasizing the contributions of patient partners, who were integral in its design and conduct, and of the family member participants. We have used the Guidance for Reporting Involvement of Patients and the Public, Version 2 (GRIPP2)–Short Form reporting checklist to guide our reporting (Staniszewska et al. 2017).

Aim

This project is built on our previously funded research to develop COSs for children with rare IMDs. The workshop's purpose was to solicit ideas and preferences from multiple stakeholders to inform the design of a registry focused on longitudinal collection of the COSs for PKU and other IMDs and to outline what would be required to successfully implement such a registry across Canada.

Method

Two patient/family partner investigators (MS and NP) from the original study were involved as patient partner co-lead investigators. These patient partners co-designed the knowledge translation workshop and led our patient engagement strategy. They were involved in all the stages from the writing of the funding application to the final workshop report. Specifically, the patient partner investigators provided feedback and refined the scope of the grant proposal, co-designed preparatory training materials for parent workshop participants, contributed their own perspectives at the workshop, led a breakout session with parent participants about the selection of outcome measurement instruments and conducted an informal evaluation among parent workshop participants about their experience. In addition, one patient partner (MS) co-led the opening workshop session and presented the patient engagement strategy from the original COS study. Finally, the patient partner investigators reviewed and revised the final workshop report and continued their collaboration with the research team based on a grant for subsequent successful research, which has made use of the findings from the workshop for designing the disease registries.

Alongside these patient/family partner co-lead investigators, all seven FAF members from the original COS study were invited to attend the workshop as participants; two FAF members attended. These advisors were provided with preparatory training co-designed by the patient partner investigators, including a list of possible questions to consider for each workshop discussion and a document explaining the process of selecting outcome measurement instruments. Workshop topics that emphasized patient partner and advisor contributions included the following: selection of specific outcome measurement instruments for patient- or family-reported outcomes in the disease registries and methods for the regular collection of such outcomes, and sharing and integration of patient-/family-reported research registry data for use in clinical care.

Honoraria and travel expenses for patient partners and advisors were included in the grant proposal, in line with the Canadian Institutes of Health Research (CIHR) SPOR guidelines for compensation (CIHR 2019).

Results

Collecting patient- and family-reported outcomes

Workshop participants recognized the importance of engaging with patients and family members throughout the registry design process to encourage participation and ensure that the registries contribute meaningful data. Regarding patient- or family-reported outcomes, family advisors commented on the relevance and acceptability of specific outcome measurement instruments for measuring the outcomes from the COS study. We also discussed strategies for collecting patient- and family-reported data. Participants identified a need to carefully consider the frequency of data collection to minimize respondent burden – for example, requesting data quarterly or less frequently. They also noted that incentives may facilitate registry participation. Several patient or family member participants recommended using approaches that integrate the collection of registry data into "everyday life" – for example, using mobile apps with functions such as appointment reminders. Access to technology was raised as a potential concern, which could be mitigated with an option to answer questionnaires when visiting a care clinic.

With respect to the degree of integration of patient- or family-reported outcomes data from a disease registry into clinical care, both patient or family member participants and clinician participants strongly emphasized a need for clear and transparent communication. This is particularly important if patients or parents are answering questionnaires for research purposes at the time of a clinic visit (e.g., in the waiting room). This could lead to misunderstandings about whether data are incorporated into the medical chart or otherwise shared with and considered by clinicians. Patient and family member partners and advisors also discussed that while completing a survey may help to organize one's thoughts before engaging with clinicians, parents may worry about compromising their child's care if the research data were shared, depending on the sensitivity of the information. They also expressed concern that research data, particularly data focused on parental (vs. a child's) experiences, may distract from priority clinical discussions during children's appointments. Parent participants also felt that data specific to parental well-being should not be invariably integrated into the child's medical chart.

Communication and consent

Workshop participants agreed that regular and effective communication with registry participants would be critical for the success of the registry. Concerning consent and privacy, they emphasized a need for transparent communication to inform decision making and to build trust. Consent from children or their family members should be viewed as a process rather than a one-time event. For example, ongoing opportunities for consent should be incorporated at the stage when a child reaches the designated age to give assent or their own informed consent, or if new information becomes available that may change the decision to consent.

Partners and advisors also raised questions that the team has taken on in further research.

Discussion: Reflection on Strengths and Limitations

This workshop followed the completion of a COS study that incorporated a comprehensive patient engagement strategy (Vanderhout et al. 2021). This facilitated the co-designing of the workshop with the patient partners who had been investigators on that study and contributions from some of the same family advisors. Our established continuity of patient and family investigators and advisors also enabled all the members of our team to benefit from mutual learning, strong team cohesiveness and collaboration toward a shared goal.

From the patient-partner perspective, we attribute much of our success to the co-development of a comprehensive patient engagement strategy that included regular feedback to patient partners and advisors about how their contributions positively impacted the project, and we feel this contributed to the sustainability of our partnership. In addition, patient-partner co-investigators felt empowered in their participation as research team members and appreciated the study team's openness to new ideas and responsiveness to their suggestions. Taking into account the perspective of other research team members, we recognize the importance of the insights brought forward by patients and family members, which have been critical to our team's ongoing design of patient registries. Partners and advisors also raised questions that the team has taken on in further research. For example, the discussion about whether and how to integrate patient-/family-reported outcomes data collected for research purposes into the clinical chart requires further investigation from multiple perspectives.

The patient-partner co-investigators have continued to co-design the patient engagement strategy for our program of work, including our ongoing design of registries to support registry-based trials. Recruiting a group of family advisors who received training and support to contribute to the COS study and the knowledge translation workshop has also allowed us to build capacity in the field, including engaging new patient and family advisors.

Our patient engagement strategy for this workshop was not without limitations. Inviting only the patient-partner co-investigators and members of the FAF from the COS study rather than recruiting additional advisors meant that we had a relatively small number of patients and family members contributing to the knowledge translation workshop (participation in this workshop was not part of the original commitment of the partners and advisors to the COS study). In addition, there was limited diversity among advisors who contributed to our study. The need to increase diversity among patient and public partners in health research is an established priority (Reynolds et al. 2021). To address this, we have increased our reach to include a more diverse group of patient partners in our current work to implement registries. Furthermore, we did not engage children or youth themselves in the original COS development study or in the knowledge translation workshop. Although the COSs we developed were targeted toward children aged 12 years and younger, our ongoing work has specifically engaged a youth advisory group so that the registries we develop can meaningfully include older children. Finally, we did not formally evaluate the patient engagement strategy for the workshop. The patient-partner co-investigators and advisors informally reported that they were supported and able to fully participate, but, in the future, a more formal approach would be beneficial.

Conclusion

This co-designed workshop confirmed the importance of understanding and incorporating the preferences of patients and families as key stakeholders in the collection and use of patient- and family-reported data and in the design of disease registries. Their perspectives on issues such as consent, frequency of data collection, tools that are patient friendly and expectations about how data translate to clinical visits are all fundamental to developing registries that meet the needs of all stakeholders. We propose a need to further explore knowledge translation strategies for patient engagement in post-COS activities, including those that inform registry design. HQ

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References

Canadian Institutes of Health Research (CIHR). 2019. Considerations When Paying Patient Partners in Research. Strategy for Patient-Oriented Research. Retrieved September 9, 2021. <https://cihr-irsc.gc.ca/e/51466.html>.

COMET Initiative. 2021. Patients and the Public. Retrieved October 28, 2021. <https://www.comet-initiative.org/Patients>.

Li, G., T.T Sajobi, B.K. Menon, L. Korngut, M. Lowerison, M. James et al. 2016. Registry-Based Randomized Controlled Trials – What Are the Advantages, Challenges, and Areas for Future Research? *Journal of Clinical Epidemiology* 80: 16–24. doi:10.1016/j.jclinepi.2016.08.003.

Mathes, T., S. Buehn, P. Prengel and D. Pieper. 2018. Registry-Based Randomized Controlled Trials Merged the Strength of Randomized Controlled Trails and Observational Studies and Give Rise to More Pragmatic Trials. *Journal of Clinical Epidemiology* 93: 120–27. doi:10.1016/j.jclinepi.2017.09.017.

Potter, B.K., B. Hutton, T.J. Clifford, N. Pallone, M. Smith, S. Stockler et al. 2017. Establishing Core Outcome Sets for Phenylketonuria (PKU) and Medium-Chain Acyl-CoA Dehydrogenase (MCAD) Deficiency in Children: Study Protocol for Systematic Reviews and Delphi Surveys. *Trials* 18: 603. doi:10.1186/s13063-017-2327-3.

Pugliese, M., K. Tingley, A. Chow, N. Pallone, M. Smith, P. Chakraborty et al. 2021. Core Outcome Sets for Medium Chain Acyl-CoA Dehydrogenase Deficiency and Phenylketonuria. *Pediatrics* 148(2): 1–13. doi:10.1542/peds.2020-037747.

Pugliese, M., K. Tingley, A. Chow, N. Pallone, M. Smith, A. Rahman et al. 2020. Outcomes in Pediatric Studies of Medium-Chain Acyl-CoA Dehydrogenase (MCAD) Deficiency and Phenylketonuria (PKU): A Review. *Orphanet Journal of Rare Diseases* 15(1): 12. doi:10.1186/s13023-019-1276-1.

Reynolds, J., M. Ogden and R. Beresford. 2021. Conceptualising and Constructing 'Diversity' through Experiences of Public and Patient Involvement in Health Research. *Research Involvement and Engagement* 7(1): 1–11. doi:10.1186/s40900-021-00296-9.

Staniszewska, S., J. Brett, I. Simera, K. Seers, C. Mockford, S. Goodlad et al. 2017. GRIPP2 Reporting Checklists: Tools to Improve Reporting of Patient and Public Involvement in Research. *The BMJ* 358. doi:10.1136/bmj.j3453.

Vanderhout, S.M., M. Smith, N. Pallone, K. Tingley, M. Pugliese, P. Chakraborty et al. 2021. Patient and Family Engagement in the Development of Core Outcome Sets for Two Rare Chronic Diseases in Children. *Research Involvement and Engagement* 7(1): 66. doi:10.1186/s40900-021-00304-y.

Williamson, P.R., D.G. Altman, H. Bagley, K.L. Barnes, J.M. Blazeby, S.T. Brookes et al. 2017. The COMET Handbook: Version 1.0. *Trials* 18: 280. doi:10.1186/s13063-017-1978-4.

Young, B. and H. Bagley. 2016. Including Patients in Core Outcome Set Development: Issues to Consider Based on Three Workshops with Around 100 International Delegates. *Research Involvement and Engagement* 2(1): 1–13. doi:10.1186/s40900-016-0039-6.

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> "… there was limited diversity among advisors …"
>
> *– p.84*

Equity-Mobilizing Partnerships in Community (EMPaCT): Co-Designing Patient Engagement to Promote Health Equity

Ambreen Sayani, Alies Maybee[P], Jackie Manthorne[P], Erika Nicholson, Gary Bloch, Janet A. Parsons, Stephen W. Hwang, James A. Shaw, Aisha Lofters and the Members of EMPaCT

Abstract

Equity-Mobilizing Partnerships in Community (EMPaCT) is a novel approach to patient engagement that centres diverse lived experiences and promotes equity-oriented and inclusive partnerships. As an independent community table, EMPaCT is made up primarily of patients/diverse members of community. Researchers and other decision makers come to this table with their projects to learn how to make their project more inclusive and equitable. In this paper, we detail how we used participatory co-design to define, build and grow EMPaCT as an innovative and scalable patient partnership model that promotes bottom-up action for health equity.

Key Points

- Equity-oriented patient partnerships can be co-designed together with members of community so that the needs and priorities of community drive the process and outcomes of engagement.
- Community-led and community-driven patient engagement tables, such as Equity-Mobilizing Partnerships in Community (EMPaCT), can be a useful resource in a learning health system for decision makers who currently have few ways to engage with diverse patients.
- EMPaCT uses tools such as Health Equity Assessments and consultations to help decision makers make their projects more inclusive and equitable.

Introduction

With the maturing of patient engagement, the demand to include a more diverse set of perspectives has grown. Equity-Mobilizing Partnerships in Community (EMPaCT) is a novel approach to engaging with diverse and structurally seldom-heard population groups by partnering on terms and projects that are prioritized by members of community themselves. Through this process of co-design, EMPaCT has grown to become an independent community-led and community-driven table made up primarily of patients/diverse members of community. Members of EMPaCT meet regularly every month to provide Health Equity Assessments (HEAs) through the lens of their collective lived experience to project decision makers.

EMPaCT exists as an expert advisory group independent of any specific project. We have co-produced our engagement structures and policies. Since our establishment in January 2021, we have consulted on several projects brought to us by decision makers, policy influencers and research teams seeking to learn from diverse lived experiences, enhance the inclusivity of their work and reduce health inequities. By centring the diverse lived experiences of community members, we are building capacity for impactful outcomes by mobilizing community knowledge on issues related to health equity and translating them into tangible recommendations for projects.

EMPaCT is housed at the Women's College Hospital (WCH) Institute for Health System Solutions and Virtual Care (WIHV). Designed as a scalable model of equitable patient

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P = Patient partner.

engagement, it has received the Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Award from the Ontario Strategy for Patient-Oriented Research (SPOR) SUPPORT Unit (OSSU). In this paper, we share the participatory co-design journey that led to EMPaCT. Participatory co-design refers to partnership with members of community who have relevant lived experiences and the involvement of all members in the co-production of processes and collaborative decision making toward a unified goal (Palmer et al. 2019). Documenting the co-design process is increasingly recognized as an important way to understand the linkages among participatory action, project outcomes and associated impacts (O'Brien et al. 2021; Palmer et al. 2019).

Conceptual Approach to Co-Design

We have applied an equity-oriented approach (engaging with those who are least likely to be included with the greatest amount of outreach; Sayani et al. 2021) to the co-design of EMPaCT. As such, we recognize that health inequities have been created by the historical and systematic disempowerment of communities through interlocking structures of sexism, colonialism and racism resulting in the unjust and unfair distribution of power, privilege and prestige, which determine opportunities to be healthy and health outcomes (Hankivsky and Christoffersen 2008; McGibbon and McPherson 2011; Raphael and Bryant 2015; Sayani 2019). As a consequence, groups experiencing the most marginalization by society (based on the intersections of gender, race, income, etc.) are the most likely to be left out of decision making unless proactive outreach is done and relationships rooted in trust are built (Sayani et al. 2021).

We have grown as a community by applying an equity-oriented and trauma-informed (nurturing relationships of trust that recognize structures and systems of oppression) approach to engagement (Sayani et al. 2021). We have taken a stepwise approach to enhance diversity, equity and inclusivity using the ConNECT Framework (Alcaraz et al. 2017) in the following ways:

1. *Integrating* CON*text:* recognizing the intersections among structural oppression, the COVID-19 pandemic and the heightened need to integrate structurally seldom-heard diverse voices into policies that can shape action for health equity;
2. *Fostering a* N*orm of inclusion:* co-designing our process of engagement, ensuring compensation for members unless they choose to decline it and not using predefined labels to describe people;
3. *Ensuring* E*quitable diffusion of innovations:* facilitating real-world benefit for all by leveraging lived experiences and integrating community priorities into the design and implementation of research, policies and plans;
4. *Harnessing* C*ommunication technology:* prioritizing digital equity and deliberately supporting digital participation for all members of EMPaCT; and
5. *Prioritizing specialized* T*raining:* maximizing participation through mutual co-learning, in which each member of EMPaCT is both a learner and a teacher, and identifying future learning needs that may be serviced by outside providers.

Co-Designing a Sustainable and Scalable Model of Equitable Patient Partnerships

Three key issues have hindered effective action to improve health inequities across sectors:

1. Absence of a conceptual framework that makes explicit the linkages among social, political and economic inequalities and health outcomes (Ndumbe-Eyoh and Moffatt 2013);
2. Lack of social participation to the degree that communities experiencing the most inequities have little influence in decision making (WHO 2010); and
3. A failure to take considerations of scalability and sustainability into program design (WHO 2010).

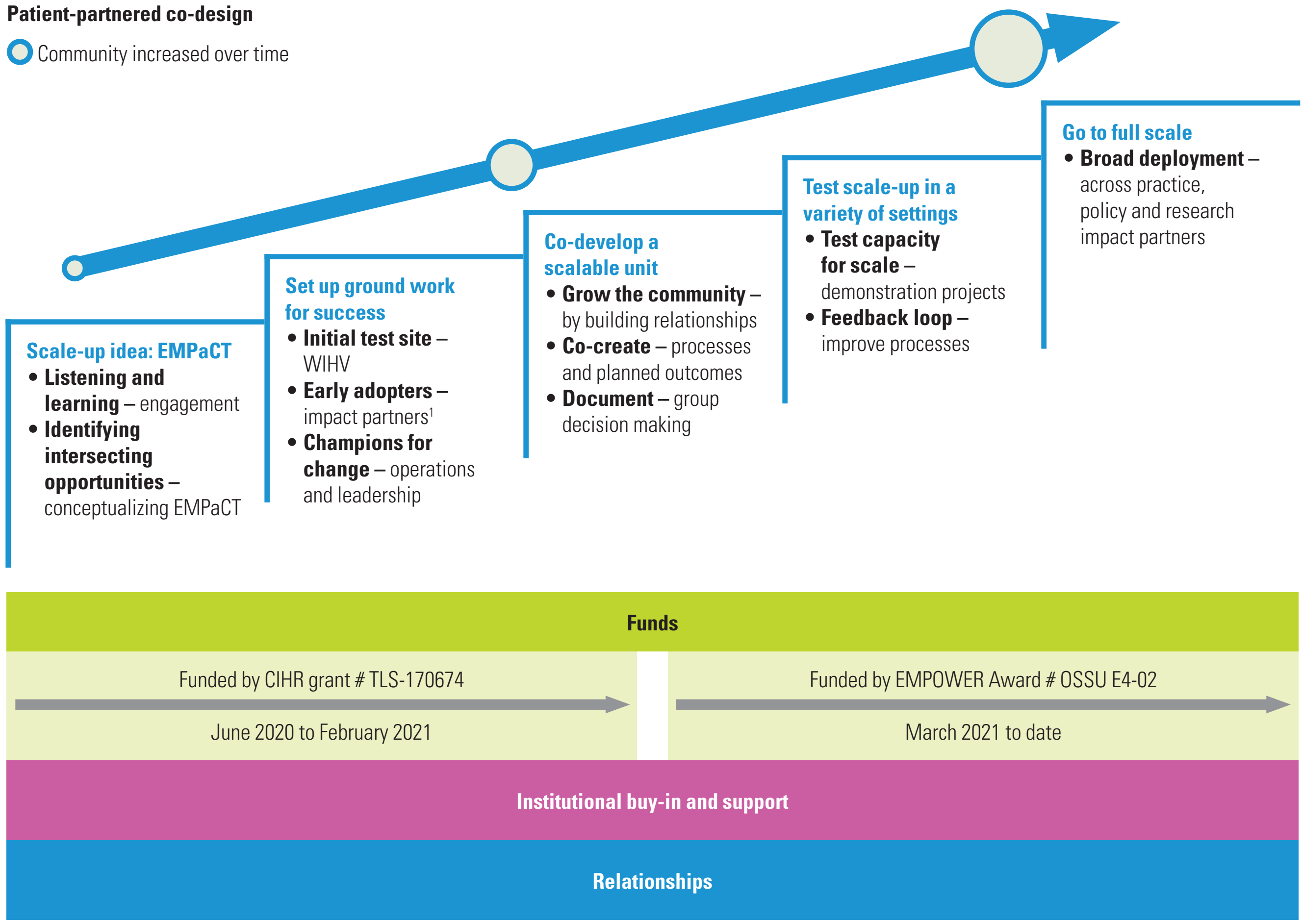
We have described our conceptual underpinning earlier; now in this section, we describe how we have applied the Institute for Healthcare Improvement (IHI) Framework of Going to Full Scale (Barker et al. 2016) to co-design EMPaCT as a scalable and sustainable model of diverse patient engagement (Figure 1).

It is important to note that the foundation of our co-design work rests on relationships (Figure 1). Some relationships existed prior to beginning this project, and many more have been built along the way. As a fundamental value, relationships are core to our existence, and relationships take priority over any processes and outcomes we may wish to achieve as a group. EMPaCT's foundational blocks include institutional buy-in and supportive structures (legal, finance, strategic direction and senior scientists) and the availability of resources such as time and grant funds.

Scale-up idea

We have expanded on the IHI Framework of Going to Full Scale (Barker et al. 2016) by including an additional section on the scale-up idea at the start. We define a scale-up idea as a learning health system innovation that results from engaging in dialogue about the possibilities for change that can be packaged as a solution and implemented across settings. As a patient-partnered project, it is important to detail how we co-designed the idea for EMPaCT.

FIGURE 1.
Framework of going to full scale for EMPaCT, based on the IHI Framework for Going to Full Scale and the underlying foundations for success

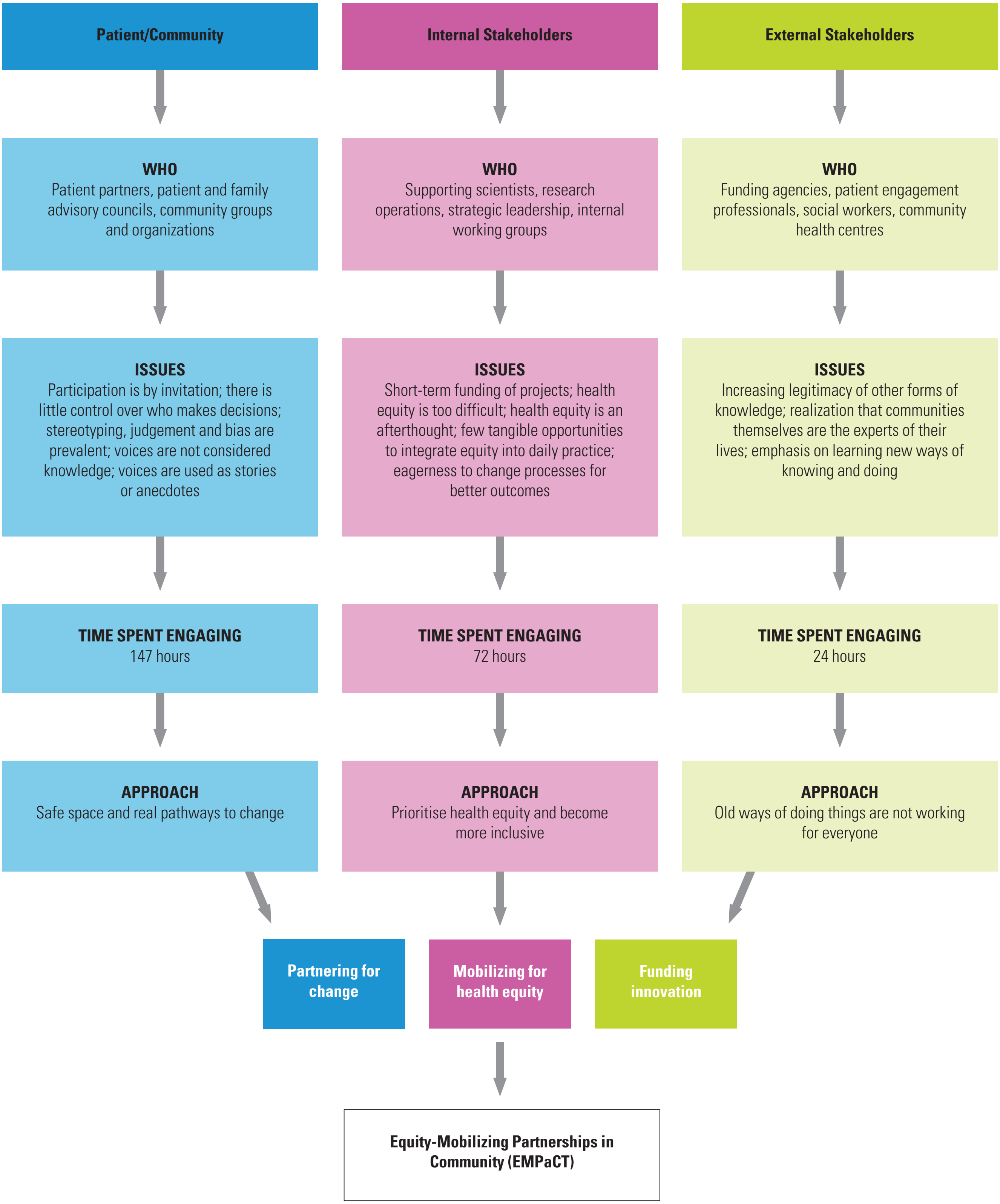


[1] Impact partners = policy makers, health/social service administrators and/or research collaborators interested in engaging with EMPaCT.

Source: Barker et al. 2016

In May 2020, members of a patient-oriented research team (AS, JM, EN, JP, SH, GB and AL) realized that their work lacked the diversity of perspectives needed to appropriately inform their study on lung cancer screening. Participation in lung cancer screening is influenced by barriers such as discrimination, historical injustice and stigma, particularly for Indigenous populations, recent immigrants and those living in conditions of poverty and precarious housing, for whom smoking and access to care are shaped by social and structural inequality. To appropriately proceed with the lung cancer screening study, it was first important to partner with patients with the relevant lived experiences. Engaging patients who bring diverse perspectives particularly from structurally underserved communities is a recognized challenge in patient engagement. This sparked a broader engagement with patients, community members and institutional stakeholders and ultimately the co-initiation of EMPaCT. AS and AM began by *listening and learning* about the barriers to diverse patient engagement through formal and informal conversations with a variety of different stakeholders (June 2020–December 2020). Using integrated knowledge translation (iKT; Jull et al. 2017), we specifically looked for intersecting opportunities that could be channelled into *actionable steps*. The details of this step, including the hours of engagement per stakeholder group, key themes and opportunities, are outlined in Figure 2.

FIGURE 2.
Co-designing the scale-up idea by listening and learning, and looking for intersecting opportunities



Patient/Community

WHO
Patient partners, patient and family advisory councils, community groups and organizations

ISSUES
Participation is by invitation; there is little control over who makes decisions; stereotyping, judgement and bias are prevalent; voices are not considered knowledge; voices are used as stories or anecdotes

TIME SPENT ENGAGING
147 hours

APPROACH
Safe space and real pathways to change

Internal Stakeholders

WHO
Supporting scientists, research operations, strategic leadership, internal working groups

ISSUES
Short-term funding of projects; health equity is too difficult; health equity is an afterthought; few tangible opportunities to integrate equity into daily practice; eagerness to change processes for better outcomes

TIME SPENT ENGAGING
72 hours

APPROACH
Prioritise health equity and become more inclusive

External Stakeholders

WHO
Funding agencies, patient engagement professionals, social workers, community health centres

ISSUES
Increasing legitimacy of other forms of knowledge; realization that communities themselves are the experts of their lives; emphasis on learning new ways of knowing and doing

TIME SPENT ENGAGING
24 hours

APPROACH
Old ways of doing things are not working for everyone

Partnering for change

Mobilizing for health equity

Funding innovation

Equity-Mobilizing Partnerships in Community (EMPaCT)

Set up groundwork for success

Set up refers to the preparation needed to seed a scale-up idea (Barker et al. 2016). This includes an *initial test site* to nurture and promote an idea and give it space to grow. WIHV is an innovation lab where team members collaborate to generate solutions to emerging health issues. In an organization committed to equity, we *identified early adopters* (supporting scientists, JS and AL) who were ready to fund and engage with members of EMPaCT on the terms set by members of EMPaCT. The institutional *champions for change* (peers, the WIHV Equity-Advisory Committee, research operations and strategic leadership for WIHV) all collaborated to meet the needs and priorities of EMPaCT as it formed with early members that included AS and AM.

Co-develop a scalable unit

A "scalable unit" is defined as a microsystem of key infrastructure and relationships needed for full scale (Barker et al. 2016). For EMPaCT, we have grown as a community one relationship at a time (from two members in December 2020 to 15 members in March 2022, 12 of whom are patients/diverse members of community). We devote half our monthly meeting time to nurturing our relationships and co-designing the next steps. Meetings are steered by AS and AM, and members take turns to lead and reflect on different activities. This process has built trust as a group, sustained engagement over time and refined our collective vision. We see ourselves as advocates for change unified by our desire to push for health system changes to promote health equity. We are disseminating knowledge concepts related to health equity, patient partnerships and pathways to change in the form of a digital library containing co-presented videos and webinars. We have co-designed a consultation process whereby we conduct HEAs. Traditionally, most HEAs are conducted by scientific experts, academics and policy administrators. The EMPaCT HEAs are based on the expertise of the members' diverse lived experiences brought to the process as a collective analytical lens. To our understanding, we are the first community of those with diverse lived experiences to offer and conduct HEAs. The strategic location of EMPaCT in an innovation hub has allowed us to navigate exclusionary institutional policies that create barriers to diverse patient engagement, including *modifications to financial and legal agreements* between institutions and community members.

As a scalable unit, EMPaCT is an expert advisory group existing independent of any specific project. Project decision makers (policy makers, health/social service administrators and research collaborators) are referred to as "impact partners". They engage with EMPaCT to learn and co-create knowledge/policy/practice solutions that advance health equity. The impact partners request to meet with EMPaCT. As part of the intake process, AS and AM coach impact partners for engagement with EMPaCT. This includes refining scope and communication skills so that the consultation results in authentic dialogue for co-learning. During EMPaCT consultations, members engage with project teams to help them understand how different communities might be impacted by a decision, what unintended outcomes may occur and how equity in health can be better addressed for the communities involved. Impact partners receive a written report validated by all members containing recommendations from the consultation process. Impact partners provide feedback on how they modified their project based on the findings of the report. They also provide feedback on the process of engagement. The community uses this to continuously improve how we do things. This relationship is the "impact partnership" between EMPaCT and decision makers.

Test of scale-up and potential for impact

To date, EMPaCT has engaged in a variety of impact partnerships, including research and hospital policy for four hospitals in Ontario, Canada. We have disseminated knowledge through a variety of platforms and received a provincial award as a scalable model of equitable patient engagement. Table 1 shows the impact categories for EMPaCT based on the Canadian Academy of Health Sciences Framework (CAHS) (CAHS 2009).

TABLE 1.
Impact from EMPaCT based on the CAHS Framework for measuring impact from research investments

Building capacity		Advancing knowledge		Informing decisions	
Members of EMPaCT	14	Publications	3	Impact partnerships	7
Sources of funding	3	Presentations	6	Impact partners engaged	20
Awards	1	Workshops	2	Decision makers engaged*	9
		Digital videos	3	Hospitals served	4
		Technical reports	8		

Source: CAHS 2009

*One senior vice president, two hospital vice presidents, one hospital medical director and five research principal investigators.

Key Learnings

We have learned from our projects that there is much demand for EMPaCT as an expert group to guide health system stakeholders to improve health equity in their projects. We are hopeful that many small actionable steps taken across the system are the start of a bottom-up community movement promoting health equity.

We have also learned that our health systems are structured to be exclusionary and discriminatory. The terms of patient engagement should be directed by members of community, safe spaces need to be built on relationships of trust and power hierarchies must be blurred so that each person occupies the role of both teacher and learner. This includes impact partners who receive preliminary coaching to scope and prepare for their consultation with EMPaCT.

Finally, a key learning has been that a willing host organization committed to adapting institutional policies and procedures to support a community is essential. EMPaCT has been able to grow and sustain itself because of the change makers at WCH who have believed in its value and supported its development.

In parallel with our scale-up, we have initiated a project evaluation loop where we are collecting anonymous feedback from the members of EMPaCT and impact partners to understand their experiences and perspectives on EMPaCT. We will analyze these findings together as a community to co-design better processes in the future.

Conclusion

The current processes of patient engagement within an already deeply hierarchical healthcare system reinforce the structures and policies that perpetuate inequalities and create barriers. Transformative change striving to improve health outcomes across the population requires innovative models of partnership centred around public participation and citizen empowerment. The involvement of patients and community members in decision making can contribute to the design of a patient-responsive healthcare system. We have described how we have co-designed EMPaCT as a patient partnership model that promotes bottom-up action for health equity. This community-driven model has potential for impact across multiple sectors and is rooted in a framework to promote social justice and health equity. HQ

References

Alcaraz, K.I., J. Sly, K. Ashing, L. Fleisher, V. Gil-Rivas, S. Ford et al. 2017. The ConNECT Framework: A Model for Advancing Behavioral Medicine Science and Practice to Foster Health Equity. *Journal of Behavioral Medicine* 40(1): 23–38. doi:10.1007/s10865-016-9780-4.

Barker, P.M., A. Reid and M.W. Schall. 2016. A Framework for Scaling Up Health Interventions: Lessons from Large-Scale Improvement Initiatives in Africa. *Implementation Science* 11(1): 12. doi:10.1186/s13012-016-0374-x.

Canadian Academy of Health Sciences (CAHS). 2009. Making an Impact: *A Preferred Framework and Indicators to Measure Returns on Investment in Health Research*. Retrieved July 11, 2016. <https://www.cahs-acss.ca/wp-content/uploads/2011/09/ROI_FullReport.pdf>.

Hankivsky, O. and A. Christoffersen. 2008. Intersectionality and the Determinants of Health: A Canadian Perspective. *Critical Public Health* 18(3): 271–83. doi:10.1080/09581590802294296.

Jull, J., A. Giles and I.D. Graham. 2017. Community-Based Participatory Research and Integrated Knowledge Translation: Advancing the Co-Creation of Knowledge. *Implementation Science* 12(1): 150. doi:10.1186/s13012-017-0696-3.

McGibbon, E. and C. McPherson. 2011. Applying Intersectionality & Complexity Theory to Address the Social Determinants of Women's Health. *Women's Health and Urban Life* 10(1): 59–86.

Ndumbe-Eyoh, S. and H. Moffatt. 2013. Intersectoral Action for Health Equity: A Rapid Systematic Review. *BMC Public Health* 13(1): 1056. doi:10.1186/1471-2458-13-1056.

O'Brien, J., E. Fossey and V.J. Palmer. 2021. A Scoping Review of the Use of Co-Design Methods with Culturally and Linguistically Diverse Communities to Improve or Adapt Mental Health Services. *Health and Social Care in the Community* 29(1): 1–17. doi:10.1111/hsc.13105.

Palmer, V.J., W. Weavell, R. Callander, D. Piper, L. Richard, L. Maher et al. 2019. The Participatory Zeitgeist: An Explanatory Theoretical Model of Change in an Era of Coproduction and Codesign in Healthcare Improvement. *Medical Humanities* 45(3): 247–57. doi:10.1136/medhum-2017-011398.

Raphael, D. and T. Bryant. 2015. Power, Intersectionality and the Life-Course: Identifying the Political and Economic Structures of Welfare States that Support or Threaten Health. *Social Theory & Health* 13(3–4): 245–66. doi:10.1057/sth.2015.18.

Sayani, A. 2019. Social Class and Health Inequalities. In D. Raphael, T. Bryant and M.H. Rioux, Eds., *Staying Alive: Critical Perspectives on Health, Illness and Healthcare* (3rd edition) (pp. 171–98). Canadian Scholars' Press Inc.

Sayani, A., A. Maybee, J. Manthorne, E. Nicholson, G. Bloch, J.A. Parsons et al. 2021. Building Equitable Patient Partnerships during the COVID-19 Pandemic: Challenges and Key Considerations for Research and Policy. *Healthcare Policy* 17(1): 17–24. doi:10.12927/hcpol.2021.26582.

World Health Organization (WHO). 2010. *Intersectoral Action to Tackle the Social Determinants of Health and the Role of Evaluation*. Retrieved June 30, 2021. <https://apps.who.int/iris/bitstream/handle/10665/70579/WHO_ETH_10.1_eng.pdf>.

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Research, Sovereignty and Action: Lessons from a First Nations–Led Study on Aging in Ontario

Carol Mulder, Derek Debassige, Maureen Gustafson, Morgan Slater, Eugenia Eshkawkogan and Jennifer D. Walker

Abstract

First Nations in Ontario are building capacity to leverage health services data in Ontario to provide robust, First Nations–driven health evidence. Beyond providing evidence, population health research processes must involve diverse First Nations' perspectives, collective capacity building and translation of research findings into action through integrated and community engaged knowledge translation and exchange (KTE) approaches. Suggested ways include integrating stories and traditional knowledge, prioritizing gatherings and establishing an enduring commitment to action. To effectively support First Nations' self-determination and sovereignty, First Nations' principles of ownership, control, access and possession (OCAP®) in research could be expanded to include "action" (OCAPA).

Key Points

- While vital to the realization of data sovereignty and the generation of First Nations–centred knowledge, research that is OCAP®-aligned does not necessarily lead to community action and uptake.
- It is important to actively share findings from First Nations health research in ways that align with communities' preferred formats, venues and information sources.
- There is a need to reframe conversations around knowledge translation and exchange (KTE) for First Nations health research. Effective KTE should support self-determination and sovereignty.

Introduction

Over the past 10 years, Chiefs of Ontario (http://chiefs-of-ontario.org/) has been working with ICES to leverage the extensive, routinely collected health services data in Ontario to answer research questions that are relevant to First Nations. These initiatives – including reports on cancer (Chiefs of Ontario et al. 2017), diabetes (Green et al. 2019; Slater et al. 2019; Walker et al. 2020), opioid prescriptions (ICES 2017), COVID-19, aging (Walker et al. 2019) and mental health (ongoing) – provide robust, First Nations–driven health evidence. The research teams – which include representatives from Chiefs of Ontario, First Nations community members and health practitioners – focus on knowledge translation and exchange (KTE) strategies that meet the needs of First Nations, their epistemologies and their inherent understanding of well-being. Yet, for many reasons, research findings may not be taken up by First Nations. Beyond providing evidence, research processes must facilitate the translation of findings into action through integrated and community-engaged KTE (Graham et al. 2006). This shift is critical to support First Nations' sovereignty and self-determination through research. In response to these issues, we evaluated the impact of KTE products from the First Nations Aging Study (Walker et al. 2019). Our findings highlight the importance of sharing research findings in ways that support First Nations' sovereignty, self-determination and action.

First Nations' Data Governance and Sovereignty

In Canada, First Nations have been explicitly asserting data sovereignty rights since the early 1990s with the articulation of a set of core principles that establish First Nations' ownership, control, access and possession (OCAP®) over First Nations data (First Nations Information Governance Centre 2014). In response to direction from the elected First Nations leadership in Ontario in 2009, Chiefs of Ontario established a strong relationship with ICES, leading to a formal *Data Governance Agreement* in 2012 and linkage with the federal Indian Register (Pyper et al. 2018; Walker et al. 2017, 2018a).

Research conducted using First Nations data at ICES is different from other projects, in that Chiefs of Ontario must provide permission before any First Nations data is accessed and used. Research applications are considered by a First Nations Data Governance Committee appointed by the Ontario Chiefs Committee on Health. Approaches to data governance are highly community-engaged and unique to each project and partner. Engagement usually includes the involvement of First Nations health directors or coordinators, Elders and people with lived experience (see, for example, Walker et al. 2018b). Research questions come from communities, analysis is guided by communities and results are collaboratively interpreted by communities. The resulting information is shared in ways that are accessible and policy-relevant for communities. These processes are critical to First Nations' self-determination and sovereignty.

What We Did: First Nations Aging Study (2015–2019)

Using the above-mentioned approach, we recently completed the First Nations Aging Study (FNAS) funded by the Canadian Institutes of Health Research. The project was co-created and co-led by a First Nations university-based researcher and Chiefs of Ontario, with other researchers with expertise in qualitative methods, aging and frailty and health services. The initial grant strengthened research capacity at Chiefs of Ontario by funding a partial short-term position at Chiefs of Ontario, which transitioned to an ongoing position when the funding ended. This is an example of how project-specific funding can seed and build sustained capacity.

The project included ongoing engagement with First Nations policy makers and brought together a Knowledge Circle that included First Nations people who were older adults, front-line health workers serving older First Nations adults, First Nations language speakers and Elders. These individuals were not employed by a university or by Chiefs of Ontario, so we provided honoraria for their participation. This was not a closed circle; some individuals came and went, while others stayed for the whole project. Mindful of shifting community priorities, we were flexible in our interactions, which included individual visits and phone calls before and after larger group meetings, integration in group meetings and enabling knowledge holders to join for parts of the project. This flexibility allowed us to build one-on-one relationships, as well as a comfortable space for Elders to share their teachings and thoughts on their own terms. When Elders could not attend large group meetings due to health challenges or shifting priorities, others stepped in to open and close the meetings and offer their guidance and perspectives on the research.

Many findings from the FNAS were policy relevant and shared at five of the Chiefs of Ontario's First Nations Health Forums from 2016 to 2021 and with the Standing Committee on Indigenous and Northern Affairs (https://www.ourcommons.ca/Committees/en/INAN). We completed the final report in fall 2019, integrating qualitative findings from conversations with Anishinaabemowin language speakers and older Anishinaabeg on Manitoulin Island, ON, with quantitative findings from ICES data and the First Nations Regional Health Survey (Walker et al. 2019). Once reviewed by Chiefs of Ontario and health directors from First Nations organizations across Ontario, we shared the report at the February 2020 First Nations Health Forum just weeks before the COVID-19 pandemic began to affect Ontario. These findings informed models prepared for First Nations leadership in the early days of the pandemic. We found that First Nations people had multiple chronic conditions and frailty with a higher prevalence and at younger ages than other people in Ontario, placing them at risk for severe COVID-19 outcomes. However, from a strengths-based perspective, these individuals also tended to report feeling a sense of emotional, physical, spiritual and mental balance.

What We Did: Evaluating, Learning and Building Capacity

Our team built and strengthened relationships with First Nations communities throughout the project. Nonetheless, we were left with several questions: What was the impact at a community level? Did the report support First Nations in decision making, prioritization and sovereignty?

In 2020, members of the research team, Chiefs of Ontario and the Knowledge Circle reconvened to answer these questions. As a starting point, we implemented a survey at the 2021 First Nations (virtual) Health Forum (Chief of Ontario Assembly Center 2021). Our intent was, firstly, to continue raising awareness about the FNAS, as the initial release coincided with the onset of the COVID-19 pandemic, when communities were necessarily shifting their focus to pandemic preparedness and prevention. Secondly, we wanted to understand and acknowledge opportunities for improvement in the way we reported the findings. This would help us understand if the results reached the intended audience and were useful to them. It would also provide constructive feedback for future projects.

All 341 registrants for the 2021 First Nations Health Forum were invited to participate in the survey, which asked about uptake of the FNAS report and preferences for sharing knowledge gained from research. As an incentive, survey respondents were offered the opportunity to win $300 credits for training in research, KTE or aging. This capacity-building element is central to the principle of reciprocity that is embedded in First Nations' research methodologies. Participants were also asked if they would be willing to be interviewed to provide more in-depth information.

The semi-structured follow-up interviews explored themes of research sovereignty, processes for decision making and research communication to inform a framework for putting First Nations health research into action. A First Nations undergraduate student conducted, recorded, transcribed and analyzed data from the interviews under the mentorship of members of the broader research network. This mentorship was another way to strengthen First Nations' research capacity and sovereignty. Analysis of interview data was done using NVivo and is ongoing. It will be presented elsewhere.

In First Nations research, both individual and collective consent from the community are critical. To establish the collective consent that is embodied in OCAP®, we followed the terms of the *Data Governance Agreement* between ICES and Chiefs of Ontario. In addition, all survey and follow-up interview participants were asked to provide informed individual consent, which they agreed to.

Our team met monthly for nine months. The reflections in our meetings influenced our interpretation of the findings. According to the Indigenous research paradigm described by Shawn Wilson (2008): "If research doesn't change you as a person, then you haven't done it right" (p. 135). In First Nations' research methodologies, researchers and community members reflect on, change and influence research while doing it. Notably, in this project, our discussions made us active influencers of our collective interpretation of the findings regarding impact and action. This was key to our relational approach to patient and community engagement and our relational accountability to First Nations people across Ontario.

What We Learned: The Importance of Stories and Sharing

In all, 29 of the 341 attendees at the 2021 Health Forum responded to our survey. This response rate reflects the challenges of engaging participants at virtual conferences. The respondents were mostly clinicians and health directors and were almost exclusively employed by a First Nations band council. Despite persistent efforts to engage with First Nations community members throughout the FNAS, only three of the 29 respondents reported having seen the report at the previous First Nations Health Forum, and only one had read it. This is a strong indication that the report did not receive wide community uptake. It also made it challenging to directly evaluate how helpful the report was.

We also asked respondents how they like to receive information and how to make research findings useful in their work. A strong majority indicated that stories (97%), traditional teachings or knowledge (90%) and research evidence (86%) were key sources of information. Community gatherings and social media were the preferred venues for finding and receiving information. While we assumed that infographic formats were important KTE tools, only 41% preferred infographics, 69% preferred reports and 62% wanted PowerPoint slides. This suggests that people are thinking of ways to share the findings from the start. Unsurprisingly, academic journals were the least preferred medium.

In First Nations research, both individual and collective consent from the community are critical.

What We Learned: Challenges of Engagement and the Importance of Action

One of the biggest challenges we faced was difficulty in engaging Elders in this follow-up project. We discussed this extensively and attempted to problem-solve this gap. This challenge was heightened by the additional demands and stresses that people faced through the COVID-19 pandemic, as well as difficulties associated with virtual meetings (e.g., interest, familiarity and bandwidth or hardware limitations). Engagement was also limited due to the uncovering of gravesites at former residential school sites and the resulting collective grief in First Nations communities. We had to respect that Elders had many other responsibilities and commitments beyond our project.

The involvement of the Chiefs of Ontario and front-line First Nations health professionals has transformed our learning in this follow-up to the initial research project. We prioritized their questions about transferring knowledge to change agents in communities and facilitating the translation of knowledge to action. We saw that OCAP® principles are effective guides to data governance and the generation of First Nations–centred knowledge. But, for research to support sovereignty and self-determination, we need a new principle of action. In our discussions, we have begun to change our language and to refer to OCAPA – collective First Nations ownership, control, access, possession *and* action in research. From a First Nations' perspective, knowledge is intended to be shared. And we have a collective responsibility to act on that knowledge.

Conclusion

Much of the current research and associated KTE with the First Nations data at ICES have been OCAP®-aligned. However, that has not necessarily led to action and community uptake, as we found with the FNAS report. The findings of this study underscore the importance of thinking strategically about sharing findings from First Nations health research. Supportive ways may include integrating stories and traditional knowledge, prioritizing gatherings and social media and establishing an enduring commitment to change, improvement and action based on findings. We need to shift our thinking to include a reciprocal responsibility for research that builds capacity, is strongly community engaged and supports First Nations' sovereignty and self-determination. This is a shift from OCAP® to OCAPA. Findings from the quantitative and qualitative phase of this project will be combined in a research-to-action framework that will facilitate these processes. In this way, research can embody sovereignty over research and data, as well as action based on that data. HQ

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References

Chief of Ontario Assembly Center. 2021. Chiefs of Ontario 15th Annual Health Forum. Retrieved March 1, 2022. <https://www.chiefsmeeting.com/coo-health-forum>.

Chiefs of Ontario, Cancer Care Ontario and ICES. 2017, October. *Cancer in First Nations People in Ontario: Incidence, Mortality, Survival and Prevalence*. Retrieved September 15, 2021. <https://www.cancercareontario.ca/en/statistical-reports/cancer-first-nations-people-ontario-incidence-mortality-survival-and-prevalence>.

First Nations Information Governance Centre. 2014, May 23. *Ownership, Control, Access and Possession (OCAP™): The Path to First Nations Information Governance*. Retrieved September 15, 2021. <https://achh.ca/wp-content/uploads/2018/07/OCAP_FNIGC.pdf>.

Graham, I.D., J. Logan, M.B. Harrison, S.E. Straus, J. Tetroe, W. Caswell et al. 2006. Lost in Knowledge Translation: Time for a Map? *Journal of Continuing Education in the Health Professions* 26(1): 13–24. doi:10.1002/chp.47.

Green, M.E., C.R. Jones, J.D. Walker, B.R. Shah, K. Jacklin, M. Slater et al. (Eds.). 2019, November. *First Nations and Diabetes in Ontario*. Chiefs of Ontario and ICES. Retrieved September 15, 2021. <http://chiefs-of-ontario.org/wp-content/uploads/2020/10/COO_firstNationsDiabetesOntario_Report.pdf>.

Institute of Clinical Evaluative Sciences (ICES). 2017. *Opioid Use among First Nations in Ontario: A Report of Current Findings*. Retrieved September 27, 2021. <http://chiefs-of-ontario.org/wp-content/uploads/2020/10/COO_OPIOIDS_Opioid-Use-Among-first-Nations-in-Ontario-2017-10-05.pdf>.

Pyper, E., D. Henry, E.A. Yates, G. Mecredy, S. Ratnasingham, B. Slegers et al. 2018. Walking the Path Together: Indigenous Health Data at ICES. *Healthcare Quarterly* 20(4): 6–9. doi:10.12927/hcq.2018.25431.

Slater, M., M.E. Green, B. Shah, S. Khan, C.R. Jones, R. Sutherland et al. 2019. First Nations People with Diabetes in Ontario: Methods for a Longitudinal Population-Based Cohort Study. *CMAJ Open* 7(4): E680–88. doi:10.9778/cmajo.20190096.

Walker, J.D., K. Jacklin, M. Andrew, S. Bronskill, J. Smylie, W. Warry et al. 2019, November. *Ontario First Nations Aging Study: Overview & Report*. Chiefs of Ontario, Laurentian University, Northern Ontario School of Medicine and ICES. Retrieved September 15, 2021. <http://chiefs-of-ontario.org/wp-content/uploads/2020/10/COO_ONfirstNationsAgingStudy.pdf>.

Walker, J., R. Lovett, T. Kukutai, C. Jones and D. Henry. 2017. Indigenous Health Data and the Path to Healing. *The Lancet* 390(10107): P2022–23. doi:10.1016/S0140-6736(17)32755-1.

Walker, J.D., E. Pyper, C.R. Jones, S. Khan, N. Chong, D. Legge et al. 2018a. Unlocking First Nations Health Information through Data Linkage. *International Journal of Population Data Science* 3(1). doi:10.23889/ijpds.v3i1.450.

Walker, J.D., R. Rowe and C.R. Jones. 2018b. Describing the Process of Ethical Conduct of Research in an Ontario-Wide First Nations Diabetes Research Project. *CMAJ* 190(Suppl): S19–20. doi:10.1503/cmaj.180479.

Walker, J.D., M. Slater, C.R. Jones, B.R. Shah, E. Frymire, S. Khan et al. 2020. Diabetes Prevalence, Incidence and Mortality in First Nations and Other People in Ontario, 1995–2014: A Population-Based Study Using Linked Administrative Data. *CMAJ* 192(6): E128–35. doi:10.1503/cmaj.190836.

Wilson, S. 2008. *Research Is Ceremony: Indigenous Research Methods*. Fernwood Publishing.

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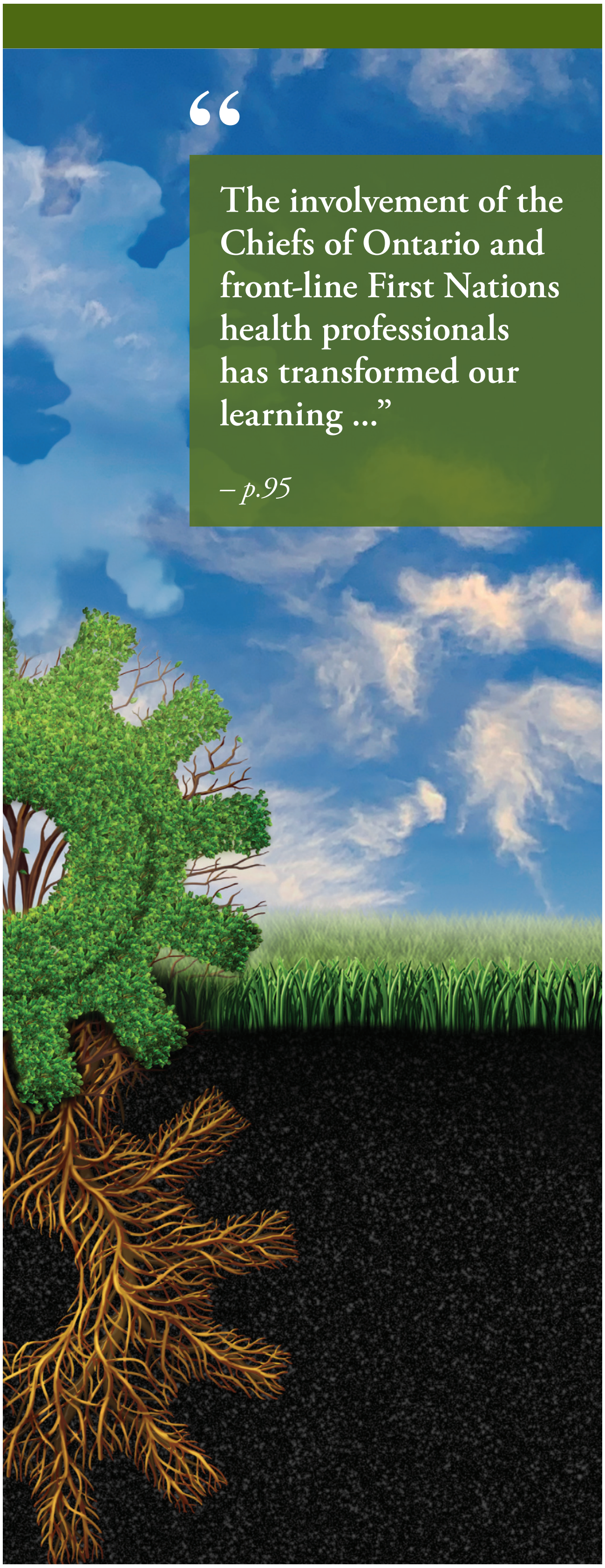

> "The involvement of the Chiefs of Ontario and front-line First Nations health professionals has transformed our learning …"
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> *– p.95*

"This Is about My Health": Partnering with Patients and Families to Share Knowledge and Tools about Healthcare Communication for Adults with Intellectual and Developmental Disabilities

Muhammad Irfan Jiwa, Janet Durbin, Victor Pereira[P], Erica Streisslberger[P], Lee Steel[P] and Yona Lunsky on Behalf of the H-CARDD Healthcare Communication Group

Abstract

This project involved patients with intellectual and developmental disabilities and a family caregiver as advisors on a knowledge translation (KT) effort on healthcare communication. The project demonstrated that with the right supports, patient and family advisors can effectively share their experiences and add a powerful voice to KT activities. Lessons learned included the importance of being creative, responsive and flexible to support the advisors, of recognizing their expertise and of building capacity in multiple advisors to allow for diverse voices and greater flexibility. This work requires adequate time and funding, which needs to be factored into planning.

Key Points

- People with intellectual and developmental disabilities (IDD) and their families are not always included in patient engagement and knowledge translation initiatives. However, this project demonstrates how they can be involved in an authentic, meaningful and collaborative way.
- Supporting the inclusion of IDD patients and family advisors requires creativity, responsivity and flexibility to support them; recognition of their expertise in the power of sharing their lived experience; and expanding capacity by involving multiple advisors.
- With the right supports, patient and family advisors can effectively leverage and communicate their experience to their peers and others and share their expertise.

Introduction

Context and original research

Adults with intellectual and developmental disabilities (IDDs) receive problematic healthcare in Ontario, across Canada and internationally. Population-based research in Ontario and elsewhere shows that poor health combined with poor healthcare puts adults with IDDs at greater risk for repeated emergency department visits and hospitalizations, delayed discharges and premature mortality (Lin et al. 2019). Among contributors to inadequate healthcare are providers feeling ill-equipped to adapt care to the needs of these individuals (Selick et al. 2018) and patients with IDD and caregivers (e.g.,

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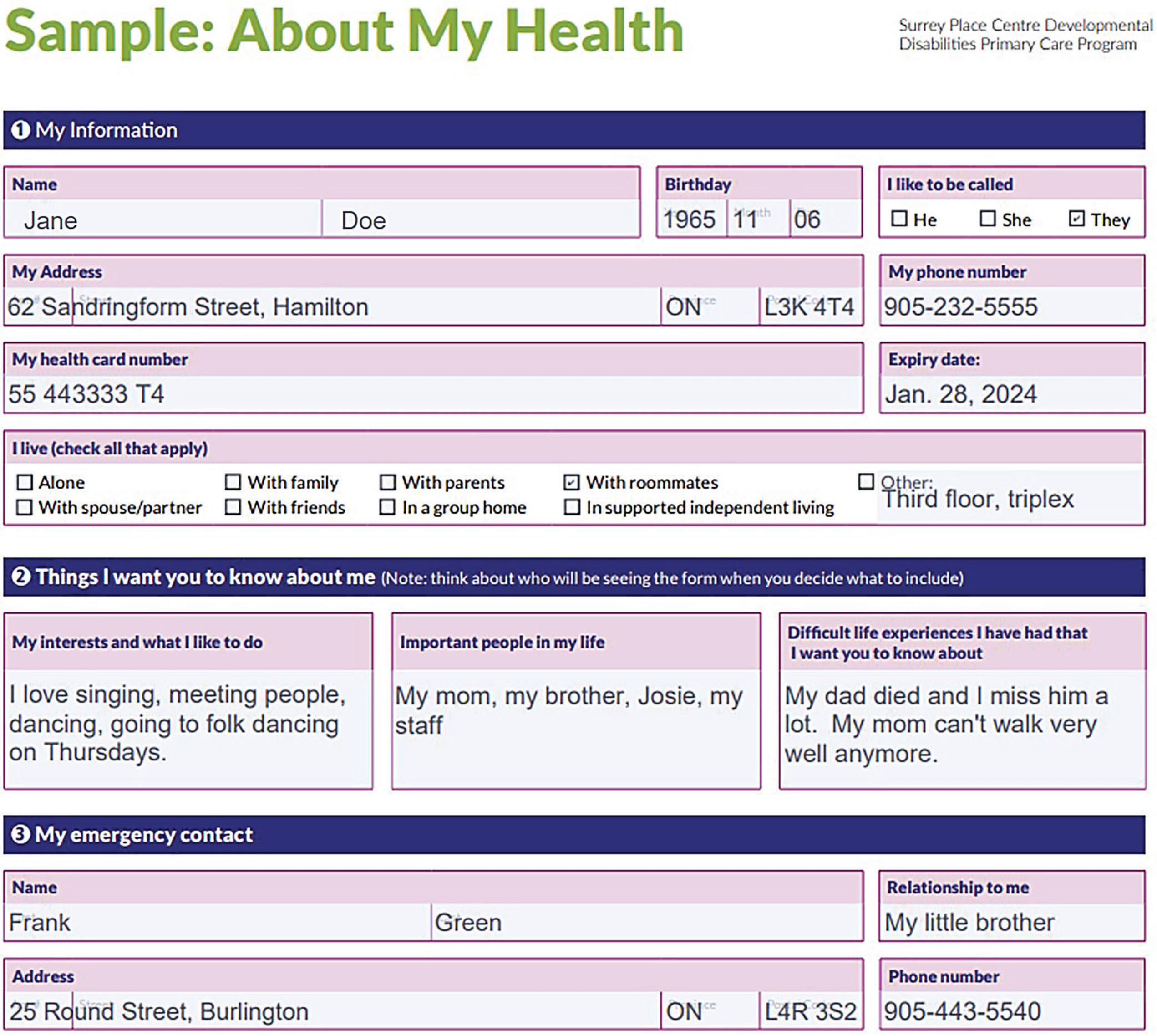
P = Patient or family advisor.

family or paid support workers) not feeling empowered or prepared to effectively manage healthcare interactions (Boyd et al. 2018; Spassiani et al. 2017). The COVID-19 pandemic and related restrictions have made it even more difficult for people with IDD to obtain needed healthcare – for example, attending in-person appointments, managing virtual appointments and including caregivers in the visits (Grier et al. 2020; Lunsky et al. 2021).

The mandate of the Health Care Access Research and Developmental Disabilities Program (H-CARDD; www.hcardd.ca) is to work with healthcare and social service providers, adults with IDD and caregivers to identify healthcare gaps and develop strategies to improve healthcare. In 2017, H-CARDD partnered with Vita Community Living Services and the Developmental Disabilities Primary Care Program on the "About My Health" study of patient-oriented healthcare communication tools (https://nutsandbolts.ddtoolkits.com/). This project was conducted together with people with IDD, caregivers and health and social care providers. Two tools were created as a result and were made available online: one tool introduces the patient generally to the provider (About My Health [https://ddprimarycare.surreyplace.ca/tools-2/general-health/about-my-health/]) (Figure 1) and the second one helps the patient (and caregiver) in preparing for and documenting what occurs during an appointment (My Healthcare Visit [https://ddprimarycare.surreyplace.ca/tools-2/general-health/todays-visit/]) (Figure 2). The tools are intended to be completed by or with extensive input from the person

FIGURE 1.
A screenshot showing part of the "About My Health" tool, filled out with details of a hypothetical patient



Sample: About My Health

Surrey Place Centre Developmental
Disabilities Primary Care Program

❶ My Information

Name: Jane | Doe

Birthday: 1965 | 11 | 06

I like to be called: ☐ He ☐ She ☑ They

My Address: 62 Sandringform Street, Hamilton | ON | L3K 4T4

My phone number: 905-232-5555

My health card number: 55 443333 T4

Expiry date: Jan. 28, 2024

I live (check all that apply)

☐ Alone
☐ With spouse/partner
☐ With family
☐ With friends
☐ With parents
☐ In a group home
☑ With roommates
☐ In supported independent living
☐ Other: Third floor, triplex

❷ Things I want you to know about me (Note: think about who will be seeing the form when you decide what to include)

My interests and what I like to do: I love singing, meeting people, dancing, going to folk dancing on Thursdays.

Important people in my life: My mom, my brother, Josie, my staff

Difficult life experiences I have had that I want you to know about: My dad died and I miss him a lot. My mom can't walk very well anymore.

❸ My emergency contact

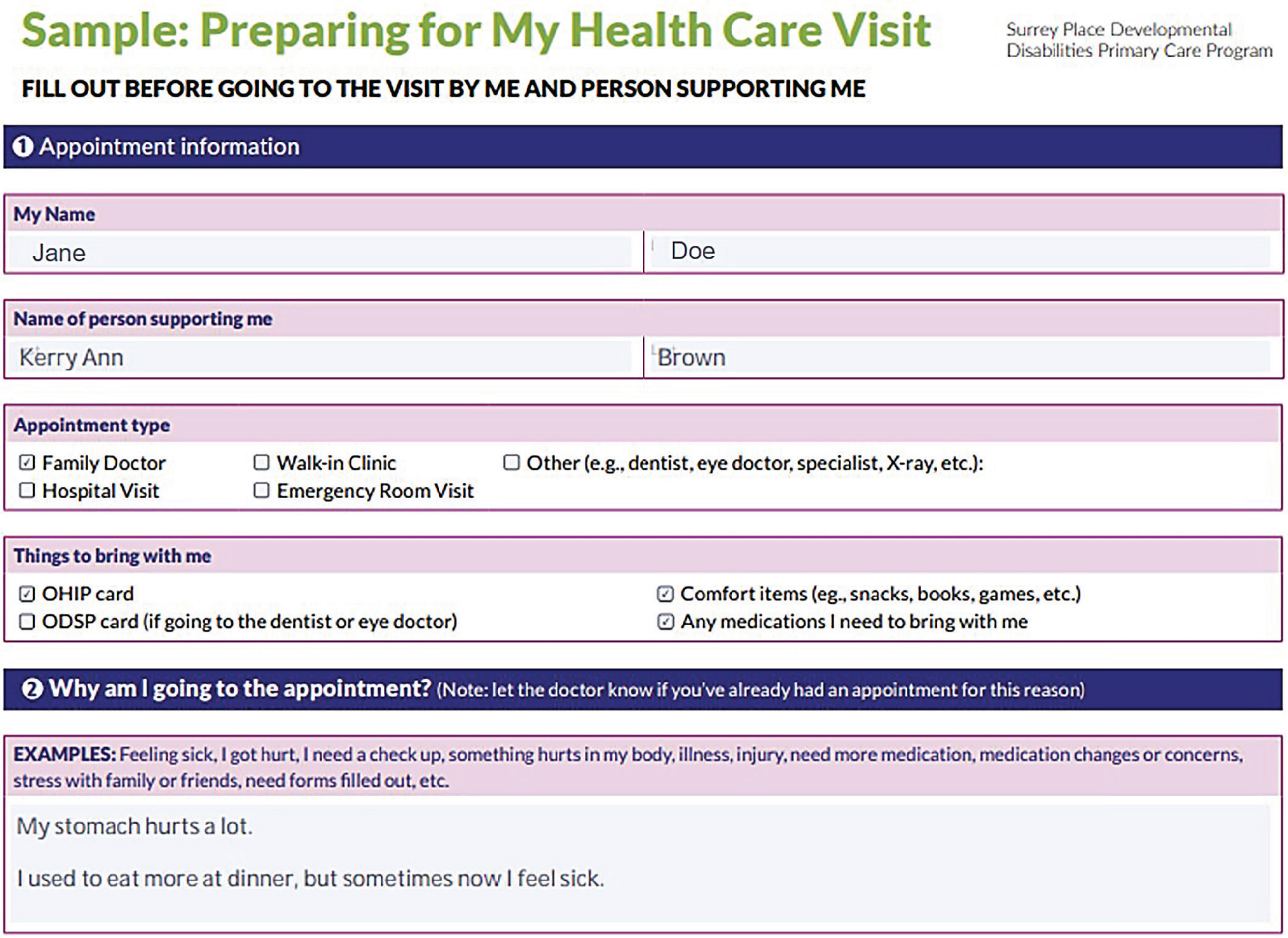
Name: Frank | Green

Relationship to me: My little brother

Address: 25 Round Street, Burlington | ON | L4R 3S2

Phone number: 905-443-5540

FIGURE 2.
A screenshot showing part of the "My Health Care Visit" tool, filled out with details of a hypothetical patient



Sample: Preparing for My Health Care Visit

Surrey Place Developmental
Disabilities Primary Care Program

FILL OUT BEFORE GOING TO THE VISIT BY ME AND PERSON SUPPORTING ME

❶ Appointment information

My Name

Jane

Doe

Name of person supporting me

Kerry Ann

Brown

Appointment type

☑ Family Doctor
☐ Hospital Visit
☐ Walk-in Clinic
☐ Emergency Room Visit
☐ Other (e.g., dentist, eye doctor, specialist, X-ray, etc.):

Things to bring with me

☑ OHIP card
☐ ODSP card (if going to the dentist or eye doctor)
☑ Comfort items (eg., snacks, books, games, etc.)
☑ Any medications I need to bring with me

❷ Why am I going to the appointment? (Note: let the doctor know if you've already had an appointment for this reason)

EXAMPLES: Feeling sick, I got hurt, I need a check up, something hurts in my body, illness, injury, need more medication, medication changes or concerns, stress with family or friends, need forms filled out, etc.

My stomach hurts a lot.

I used to eat more at dinner, but sometimes now I feel sick.

with IDD and provide a shared communication resource (Selick et al. 2022).

The Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) grant funding allowed for a scale-up of the availability, awareness and use of these communication tools across Ontario and other parts of Canada. All knowledge translation (KT) activities were performed in partnership with persons with IDD and caregivers. The core EMPOWER team included two patient advisors and one family advisor, with other patient and family advisors joining the team for specific KT activities. These individuals had been working on other H-CARDD projects in an advisory capacity prior to joining the EMPOWER team. The project originally ran from October 2019 to March 2020, but was extended to August 2020 due to the pandemic.

Approach

The initial KT plan included in-person and virtual sharing of the About My Health and My Health Care Visit communication tools at conferences and H-CARDD–hosted events, targeting health and social service professionals, persons with IDD and families. Patient and family advisors actively participated at these events by presenting the tools to persons with IDD and their families, staffing exhibition booths at professional conferences and teaching clinical providers and support workers about healthcare communication. They also contributed to developing and delivering a telemedicine interprofessional continuing education program and a family education series.

At the onset of the COVID-19 pandemic in March 2019, the project pivoted to virtual-only KT activities and, with additional funding from the Canadian Institutes of Health

Research, later expanded the virtual education programs to reach families, people with IDD and providers across Canada. The courses covered general and COVID-19–specific topics that resonated with patients and families (e.g., mental health during the COVID-19 pandemic), and the communication tools were embedded as part of the curriculum. Patient and family advisors participated in virtual teaching to share their perspectives on the value of the tools. They were also involved in the development of a video (https://www.youtube.com/watch?v=AOKJKBSAFWA) explaining the use of the tools in a virtual healthcare environment.

Through these various KT strategies, the tools reached broad audiences. As of January 2022, the healthcare communication tools have had a total of over 6,400 views.

What the Team Learned along the Way

Be creative, responsive and flexible while supporting patient and family advisors

During this EMPOWER-funded KT project, the strategies used to support the patient and family advisors evolved. Coaching was always a key strategy. This included in-person and later online coaching on how to use the communication tools and how to talk about them with others. We also instituted weekly small-group coaching/support sessions to co-develop the presentations/resources and discuss delivery for different audiences, including other people with IDD, as well as healthcare providers. A key (and unanticipated) issue was helping the patient and family advisors learn how to integrate their personal experiences into their presentations while maintaining privacy and boundaries on how much information to share.

> I think having a family member share their personal experience is impactful to other caregivers. I was able to give real-life examples of the benefits and challenges we encountered. Dr. Lunsky helped me refine my wording as in my first presentation I realized I was disclosing too much information that my son may not have appreciated me sharing publicly. I was grateful for the sensitivity she provided in helping me craft a useful but less personal sharing for future presentations. (Family advisor)

When our team shifted to remote-only working, we adopted several additional strategies. During presentations, we provided "off-camera" coaching through text, direct chat messaging or phone calls for instances where patient advisors had questions, needed prompts to recall speaking notes or reminders when they veered off topic. We found that we needed to have a variety of communication methods to accommodate their individual abilities and skill sets, as well as backup options in the event of technical difficulties such as Internet and software troubles. We also learned that it was critical to debrief after events as a team to learn, reflect and provide feedback. However, this preparation and debriefing could add two to three hours to each hour of active presentation/meeting, and required prior planning.

The power of sharing personal experiences

By sharing their personal experience using the communication tools, the patient and family advisors demonstrated their expertise both about how to apply the tools in healthcare settings and their personal value.

> I use the tools before the healthcare visit. I fill it out and turn it in to the doctor. How I use the tool [is that] I would use it as a prompt [for] what I need to say. A lot of [the] times what I'm saying doesn't come out too clearly, so having it written down works for me. A lot of [the] times we tend to forget what we need to say. (Patient advisor)

> When I completed these forms with my own son, the tools empowered us to think about essential health information ahead of time, while we were both calm. We discussed information and strategies that should be shared with healthcare providers. Capturing this information in one place helped to record a lot of the information that I hold in my mind as my son's primary caregiver. The information was recorded in a succinct format in one document that could be located at his doctor's office, [in] the emergency department, or [at] new healthcare providers. In addition, the forms provided a place where recommendations from the healthcare provider could be captured accurately, avoiding confusion or misunderstanding of [the] next steps. (Family advisor)

Although it took time and hard work for the advisors to be able to understand the tools and craft their story, this approach was incredibly powerful. One example of this was when one of the patient advisors was promoting the tools in an exhibition booth at a physician conference. About 60 physicians stopped by, and the patient advisor engaged with nearly every visitor, sharing his personal experience about going to the doctor.

> This doctor said, "Why would I need the tools?" But when I explained, from my point of view – the patient's point of view – I explained it to him, and it really changed his perspective on what the tools actually

> mean, like what the tools actually are good for. They are good for people with disabilities who are scared of going to the doctor, or even people who are mute and going to the doctor for the first time.

The staff facilitator working with the patient advisor at the booth noted a visible shift in the physician's attitude from skeptical to understanding. In speaking about this experience afterward, the patient advisor described feeling empowered to make a difference:

> To be able to change a person's perspective was really amazing for me because I can never change a person's mind.

Respect and amplify the expertise of the patient and family advisors

It took some practice for the academic members of the team to appreciate the unique expertise of all the members. This included respecting all voices and amplifying the lived experience of patient and family advisors instead of taking the lead or instructing them on what to do. This meant being aware of pre-existing power dynamics and being open to the advisors' styles of speaking about their experience, even if it did not always match preconceived ideas of how things should go.

> I realized rather quickly that we would have to be very flexible with how we did our KT activities to amplify their expertise and not over-direct our patient advisors. For example, while preparing for a webinar, they expressed how instead of having prepared speeches to read out, they would much rather have me ask them questions on a panel as a way of sharing their experiences and understanding of the tools. (Academic team member)

Expanding capacity by involving multiple patient and family advisors

A small group of patient and family advisors were active participants on the research team, and they were not always available for all activities, particularly when personal circumstances, health concerns or family issues arose. While this can be an issue for any team member, without a broader roster of patients and families to draw on, some activities had to proceed without their participation or had to be rescheduled. Additionally, they described in different ways the pressure or burden of representing the patient or family voice when they were the only patient/family advisor involved in a specific activity. This is not unique to projects involving patients with IDD but it does need to be addressed. Our team respected and supported everyone to participate as best they could, given the circumstances. Having an expanded roster of patient and family advisors would provide a richer sharing of experiences and include individuals with a variety of lived experiences of not only disability but also other intersectional identities, such as race, gender, age, sexual orientation, etc. A larger team of advisors would also provide greater flexibility so that the work could be maintained while accommodating the various needs and availability within the team. This requires funding to support multiple people in similar roles and investment in ongoing capacity building. See Figure 3 for key lessons learned.

FIGURE 3.
Key lessons learned

It is important to be creative and flexible in how patient and family advisors are supported, and be open to allowing strategies to evolve as needs emerge.

When patient and family advisors share their personal experiences, knowledge translation is more powerful and impactful.

Key lessons learned

Researchers should aim to respect and amplify the expertise of patient and family advisors instead of over-directing them.

Involving multiple patient and family advisors makes for expanded capacity – allows for diverse voices and greater flexibility.

Discussion

This project demonstrated the feasibility and value of meaningful collaboration with people with IDD and family members. The EMPOWER grant has allowed us to not only disseminate communication tools to address an important gap in healthcare for adults with IDD and reach stakeholders nationally, but also to build capacity and develop processes to integrate patients and families into the project team. The lessons learned from this project, summarized in Figure 3, are informing our current work and can help others engage people with IDD and families in research and KT initiatives. Among these were the importance of being flexible and adapting in real time to advisor needs. For example, we incorporated additional coaching and debriefing time to our support approach (e.g., an extra two hours to help advisors prepare for presentations and to debrief), and we took direction from the patient advisors on their preferred presentation formats (e.g., delivering information through a structured panel question and answer session over a more open didactic format). We also invested effort to increase the number of patient advisors on the team to reduce the demands on any one individual and to bring more perspectives forward.

This work also demonstrated the relevance of being flexible and of individualizing the coaching and support we provide to patient advisors with IDD and family advisors. Beginning projects with an exploration of learning styles, communication preferences and individual strengths and then planning KT activities around this was key. Still, we needed to be iterative and quickly evolve our KT plans based on how situations and needs unfolded. The advisors learned in different ways and at different paces. It is important to know individual abilities, strengths and skills and match them to tasks accordingly. It is also crucial to be able to provide extra time and support to accommodate each advisor's unique needs and provide the right support to build their capacity and confidence to take on new roles. We found that by using a collaborative, strengths-based approach to designing KT events, we were able to develop more accessible products and deliver targeted inclusive presentations.

Additionally, this project showed that "experts" should include people with IDD, and not just the families or paid workers supporting them. This is an important message for academic researchers who may not think to include this group because of traditional research hierarchies and/or perceived barriers, including overcoming difficulties comprehending verbal and written communication and challenges related to expressing their thoughts verbally and in writing. Accommodations – such as simplifying information to be used by patient advisors, providing it in a range of formats (written, with pictures, recordings, etc.) and offering more time for individual coaching debriefs – might help multiple groups but are particularly important for including people with IDD.

We are continuing to expand our roster of advisors, drawing on the lessons learned in this project. We have achieved some success, and our learning on how to effectively engage and support continues to evolve. However, this work does require having adequate time and sufficient funding (to pay advisors for their time, including preparation and debrief; and provide dedicated staff support). This cost and time need to be factored into planning. It also requires flexibility to authentically engage and support people with IDD to fully participate in the initiative. When such strategies and accommodations are made, we all benefit from the inclusion of diverse voices and perspectives.

Conclusion

The efforts undertaken in this initial smaller-scale end-of-grant KT project demonstrate several lessons learned to facilitate authentic, non-tokenistic, patient engagement for people with IDD and their families. With the right supports, patient and family advisors can effectively leverage and communicate their experience to their peers and others and share their expertise as part of end-of-grant KT activities. Although this project only included a small group of patient and family partners, with more funding, resources and staff support, the strategies developed could be scaled to involve larger groups of adults with IDD and family caregivers in KT and research, more broadly. Based on the lessons learned in this project, we have started working with additional patient and family advisors to prepare them for future KT efforts, while allotting the necessary time and staffing to properly support their roles as teachers and champions. HQ

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References

Boyd, K., H. Diepstra, K. Elbard, Y. Hamdani and Y. Lunsky. 2018. Consumer Inclusion: Experience of Patients with Intellectual and Developmental Disabilities Informs Primary Care. *Canadian Family Physician* 64(Suppl 2): S8–11.

Grier, E., Y. Lunsky, W.F. Sullivan and I. Casson. 2020, April 9. Health Care of Adults with Intellectual and Developmental Disabilities in a Time of COVID-19 [Blog post]. *Canadian Family Physician*. Retrieved February 24, 2022. https://www.cfp.ca/news/2020/04/09/04-09-02.

Lin, E., R.S. Balogh, A. Durbin, L. Holder, N. Gupta, T. Volpe et al. 2019. Addressing Gaps in the Health Care Services Used by Adults with Developmental Disabilities in Ontario. Institute for Clinical Evaluative Sciences. Retrieved February 24, 2022. <https://www.ices.on.ca/Publications/Atlases-and-Reports/2019/Addressing-Gaps-in-the-Health-Care-Services-Used-by-Adults-with-Developmental-Disabilities>.

Lunsky, Y., N. Bobbette, A. Selick and M.I. Jiwa. 2021. "The Doctor Will See You Now": Direct Support Professionals' Perspectives on Supporting Adults with Intellectual and Developmental Disabilities Accessing Health Care during COVID-19. *Disability and Health Journal* 14(3): 101066. doi:10.1016/j.dhjo.2021.101066.

Selick, A., J. Durbin, I. Casson, J. Lee and Y. Lunsky. 2018. Barriers and Facilitators to Improving Health Care for Adults with Intellectual and Developmental Disabilities: What Do Staff Tell Us? *Health Promotion and Chronic Disease Prevention in Canada* 38(10): 349–57. doi:10.24095/hpcdp.38.10.01.

Selick, A., J. Durbin, C. Salonia, T. Volpe, E. Orr, H. Hermans et al. 2022. The Nuts and Bolts of Health Care: Evaluating an Initiative to Build Staff Capacity to Support the Health Care of Individuals with Intellectual Disabilities. *Journal of Applied Research in Intellectual Disabilities* 35(2): 623–32. doi:10.1111/jar.12975.

Spassiani, N., M.S. Abou Chacra and Y. Lunsky. 2017. "Why Are You Here? Can't You Cope at Home?" The Psychiatric Crisis of People with Intellectual Disabilities and the Community's Response. *Journal of Mental Health Research in Intellectual Disabilities* 10(2): 74–92. doi:10.1080/19315864.2016.1278290.

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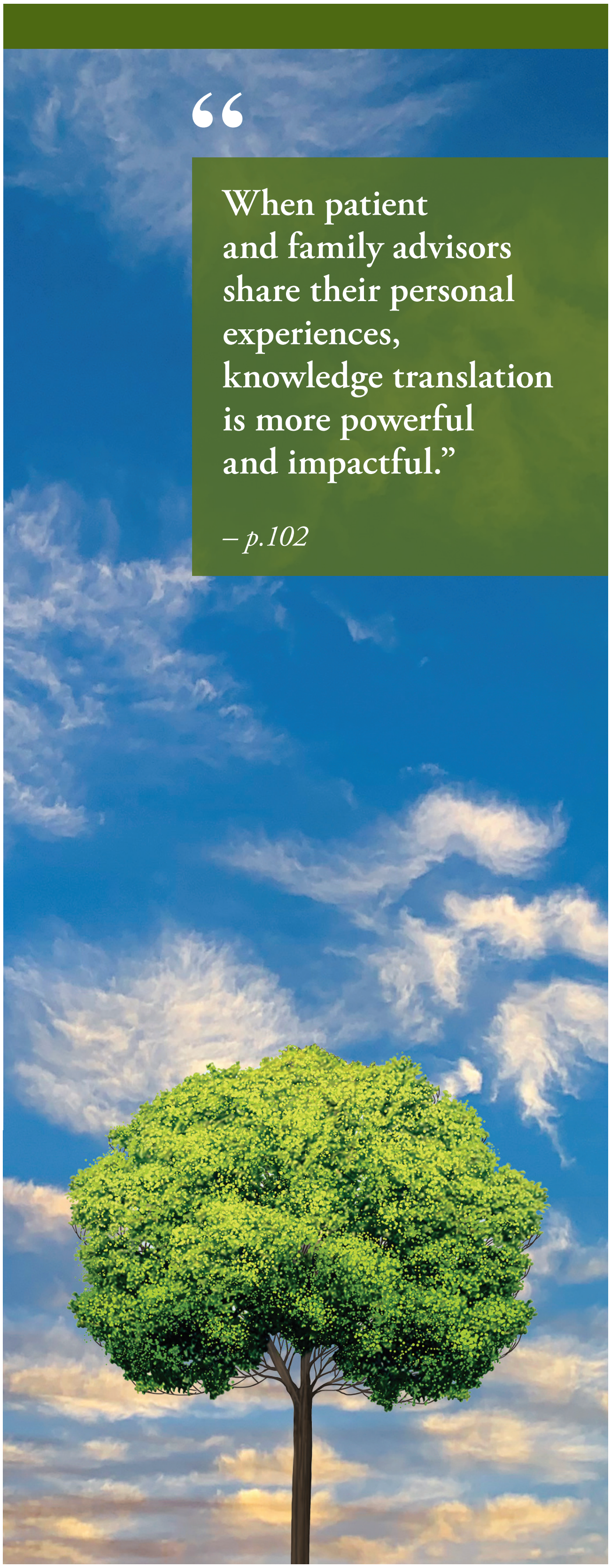

> "When patient and family advisors share their personal experiences, knowledge translation is more powerful and impactful."
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> *– p.102*

Advancing Patient-Partnered Research: Empowerment, Innovation and Evolution

Annette McKinnon and Maureen Smith

Abstract

From the perspective of patient partners, the Ontario SPOR SUPPORT Unit Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach Awards have facilitated successful patient-partnered research projects, which, in turn, have led to an evolution in patient partnerships and engagement strategies. The 15 projects profiled in this special issue point to the beneficial impacts of patient-partnered research.

Introduction

As experienced patient partners, we welcome the opportunity to reflect on the Ontario Strategy for Patient-Oriented Research (SPOR) SUPPORT Unit's (OSSU) Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Awards, now in their fourth round of funding. Can we say that they are aptly named? And more importantly, has patient-partnered research in Ontario truly been empowered? Was this empowerment manifested in research that responded to the untapped opportunity to encourage and support knowledge translation activities that would build relationships and "push the results to those who can use them to effect real impact and change in Ontario" (OSSU n.d.)?

Discussion

With the SPOR ideals and vision for patient-partnered research, this special edition of the EMPOWER Award studies demonstrates the evolution in patient partnerships and innovative engagement strategies in diverse healthcare research. The EMPOWER Awards were meant to motivate teams to take their patient engagement to a new level. The breadth and depth of the 15 projects reveal the growing appetite to further develop patient engagement strategies and, more significantly, the extent and reach of patient-partnered research. This special edition represents the following topics: priority setting and best practices, equity in patient partnerships, co-designing interventions and tools, tools for patient engagement and patient-driven or community-driven projects.

These studies bring to light three fundamental impacts on health research:

1. In partnering with patients, research benefits from identifying unanswered questions, patient-relevant topics and new areas of research.
2. Patient-partnered research influences and shapes knowledge translation strategies and activities.
3. Established patient–researcher relationships provide impetus to continue collaborating.

In this special edition, we witness researchers successfully partnering with patients using co-creation, co-design and co-leadership, and we get a glimpse of the evolution of patient-partnered research. In one project, an empowered patient brought the grant opportunity forward; in another, the community saw the need. We note novel approaches to address equity, diversity and inclusion through strategic equity lenses and by partnering with more diverse populations. There is also the acknowledgement that much work remains to be done in this area. One thing that resonates is that these partnerships have a good dose of generosity, commitment and originality, resulting in innovative strategies for patient partnership that will serve Canadian society.

Since 2012, when a literature search on patient engagement yielded only 34 results, to the present, where a plethora of more than 12,000 articles is available, the growth of patient partnership in research has exploded and is charting new territory (Marlett 2022). When researchers' passion for discovery and innovation is combined with the power of committed and enthusiastic patients and caregivers with the unique expertise of lived experience, both the transformative aspect and the hope for the future as we democratize and revolutionize health research together takes root. The EMPOWER Awards have enabled research teams to push their patient-oriented research further and build on these essential patient partnerships.

We believe that the EMPOWER Awards are aptly named. This special issue highlights proof of empowerment on multiple levels: stronger and more diverse patient–researcher partnerships, knowledge translation that has a greater reach and impact and innovative strategies for patient-partnered research. Empowerment is realized through meaningful collaboration and co-production. As patient partners ourselves, we look forward to more patient partner–initiated applications as ideas for end-of-grant translation can be generated by any of the team members.

Conclusion

It has been inspirational to see the optimism of people working hard to attain the ultimate results from the SPOR partnership. This special edition also emphasizes that we need to share our successes and challenges with one another and learn from our missteps as we develop patient partnerships and strategies that truly enable patients and caregivers to be an integral part of the research that has an incredible impact on our lives. HQ

References

Marlett, N. 2022. The Science of Engagement in Health Research and Innovation [Preprint]. University of Calgary Press.

Ontario SPOR SUPPORT Unit (OSSU). n.d. OSSU EMPOWER Awards. Retrieved November 30, 2021. <https://ossu.ca/for-researchers/work-we-support/ossu-empower-awards/>.

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Patient-Oriented Research: Enhancing Partnership-Engaged Knowledge Mobilization for Impact

Diana Urajnik*, Rebecca Ganann* and Peter J. Gill

Abstract

Patient-engaged research requires strong patient and applied partnerships to realize innovative knowledge mobilization. Demonstrating impact and sustainability, advancing health equity, evaluating engagement and incorporating flexibility are key ingredients to advance engagement science.

Demonstrating Impact and Sustainability

Over the span of a decade, Canada's Strategy for Patient-Oriented Research (SPOR) has provided the impetus for core developments in patient-engaged research in the country (CIHR 2011). SPOR SUPPORT Units across the provinces and territories have built key infrastructure and capacity for patient-oriented research (POR), thereby strengthening the science and practice of POR. Ontario's SPOR SUPPORT Unit (OSSU), specifically, has been a major catalyst for patient-engaged research within the province. Since the launch of OSSU in 2015, the field of patient engagement and involvement in Ontario has shifted from foundational levels of involvement to increasingly advanced engagement (Hamilton et al. 2021). Core to advanced engagement is equal partnership and shared decision making, and the reciprocity of shared decision making and balanced partnerships illustrates the necessity of impact for sustainability. Perhaps the epitome of this evolution is that of meaningful partnership and integrated knowledge translation.

In 2019, OSSU initiated the Engaging Multi-stakeholders for Patient Oriented-research Wider Effects and Reach (EMPOWER) Awards to mobilize knowledge and enhance patient-partnered relationships arising from research seeking to integrate patient and community voices. Awardees addressed core criteria of partnership and demonstration of impacts for patients, policy and practice. Across the compendium of the 15 EMPOWER projects are valuable lessons learned, which can inform future directions to advance POR for improved health outcomes.

Of note, the supplement outlines the impacts achieved through patient partnership. Perhaps, a key descriptive – and foundational – word is "collective"; without collective efforts, the translational gains achieved would certainly be more modest. EMPOWER Award recipients emphasized project co-creation and co-design from inception to impact and partnerships often outlasting end-of-project funding. Common to all projects are strong and diverse engagement strategies that help involve end-users in knowledge mobilization. Some strategies are familiar, such as involving patients and caregivers on Advisory Councils, and patient participation in research team meetings. Other strategies have sought to "push the boundaries" of innovation (e.g., Syan et al.'s [2022] youth-led grant application preparation) and address engagement in virtual technology (e.g., Birnie et al.'s [2022] engagement with youth and families to develop best practices for virtual care). The

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projects also aptly illustrate that engaged partnership can begin anew or that enhanced partnerships can emerge from existing relationships. At the core are values of trust, humility, respect and shared vision, with an emphasis on communication, community, shared leadership, active and meaningful engagement and shared decision making (Haywood et al. 2017).

Widespread use of virtual technology has also amplified concerns around digital equity ...

Advancing Health Equity

A predominant and cross-cutting theme is the increasing recognition of and emphasis on health equity across projects. Health equity involves fairness in the distribution of resources needed for health, access to available opportunities and supports and health outcomes (CIHR 2019). Rigorous and diffuse integration of equity considerations is essential for meaningful research impact (Browne et al. 2012), and EMPOWER awardees appreciated the importance of equity, diversity and inclusion (EDI) in this manner. Some projects included diverse populations or communities reflective of end-users, with the goal of addressing priorities or translating best practices that would meaningfully meet the needs of stakeholders. Many authors embodied equity in shared design and decision making since project inception via patient- and community-driven projects; some projects specifically emphasized the inclusion of marginalized voices (e.g., Black, Indigenous and People of Colour communities) to address stigma, discrimination and bias; others had patients/caregivers directly affected by health issues as project leads in dissemination efforts. One project that highlighted advanced engagement was Mulder and colleagues' (2022) First Nations–led study on aging, which describes ongoing work with First Nations people, makes unique contributions by integrating traditional First Nations' knowledge and methodologies with health services data and calls attention to the need for action based on knowledge gained through research.

As we move forward in a learning health system that prioritizes EDI and Indigenous sovereignty, health and wellness, OSSU's Fairness is Excellence EDI Framework (Ontario SPOR SUPPORT Unit 2022) can serve as an exemplar for POR researchers, trainees and health system decision makers. In particular, the framework outlines concrete and specific steps that individuals can take to address health equity, from selecting a research topic, recruiting and retaining staff and students to analysis and knowledge mobilization.

Incorporating Flexibility to Address New Challenges

While these EMPOWER Awards outline a number of success stories of partnering with patients to conduct research and knowledge dissemination – much as with research in general – unanticipated challenges emerge. The most notable challenge was the emergence of the COVID-19 pandemic and the unanticipated health system impacts due to both direct and indirect impacts of the pandemic. The shift to virtual platforms and remote care as the default has forced teams to think and adapt. For projects that were already underway, teams had to rapidly pivot to incorporate virtual models. For example, the Transplant Ambassador Program (TAP), which traditionally situated TAP ambassadors in dialysis units and in hospitals, had to shift to virtual platforms. Yet by doing so, TAP ambassadors expanded their geographical reach to engage patients from across Ontario. Virtual tools have suddenly increased the scale of projects by removing previous geographical limitations and yet introducing challenges in terms of scope and feasibility.

Widespread use of virtual technology has also amplified concerns around digital equity: digital and virtual care may compound existing barriers if these options are inaccessible to some groups or for those who are vulnerable because of existing health inequities (Asmundson et al. 2020). Birnie al.'s (2022) project sought to address this challenge by specifically engaging a diverse sample of youth to ensure that recommendations for best practices for virtual care are inclusive and equitable. Furthermore, the perspectives of patients in Tingley et al.'s (2022) project shed light on another potential challenge: that of data sharing and privacy concerns. This team engaged patients as partners to better understand how core outcome sets can be implemented in research.

A recurring challenge that continues to emerge is sustainability: How do we continue to recruit and retain patient partners? Certain projects are particularly susceptible to this challenge, particularly those working with certain populations or diseases. For example, Syan et al. (2022) working with the Youth Wellness Quest resource, a youth-led resource for mental health, encountered multiple challenges with turnover, given the multi-year duration of the project combined with high mobility of youth. The landscape of patient partner researchers, while growing, remains small. Therefore, it can also be difficult to identify individuals to take on leadership roles in projects that require substantial time commitments for various reasons, including family commitments, employment or interest.

Strengthening Evaluation of Patient Engagement

There is growing recognition of the need to evaluate patient engagement, including its impact not only on patient outcomes but also on patient and researcher experiences and outcomes and research quality. There is also a need for transparency around potential biases/limitations that may occur, such as implicit biases surrounding diversity of experiences or capabilities for engagement. Impact evaluation remains an emerging field, with no established standard for measurement; however, it is vital to engage patient partners in the planning and conduct of any evaluation. Several of the EMPOWER awardees discuss tools or instruments used to evaluate engagement. Vanderhout et al. (2022) required members of the Parent and Clinician Team to complete the Patient Engagement in Research Scale (PEIRS), a 37-question instrument developed by Clayon Hamilton and Arthritis Research Canada (Hamilton et al. 2018). Other projects used the Patient and Public Engagement and Evaluation Tool (PPEET), which was developed by Julia Abelson and the PPEET Research-Practice Collaborative at McMaster University (Abelson et al. 2016). Both tools were developed in partnership with patient partners, adding relevant lived experience content expertise to the tool development process. Few projects evaluated the impact of engagement on health or health system outcomes.

There are other important questions to ask about evaluating patient engagement that require further research. Beyond the psychometric or performance properties of evaluation tools, what about the evaluation of co-design in patient partnership? The effectiveness of research co-design has seldom been evaluated, despite its widespread use and descriptions of the benefits and challenges of the design-led process (Slattery et al. 2020). Nor do we know what engagement methods work for whom and in what context. Furthermore, what about the impact on patient care and outcomes? Certainly, applying tools to evaluate patient engagement that have been co-designed with partners should be the minimum expectation to measure and yield critical input regarding how patients are involved in research, including adherence to the values of respect, collaboration and teamwork. But as outlined in Rahimi and colleagues' (2019) study, tools that predict the results of research that engages patients are also required, as well as how those results may vary by level of engagement or change over time. We further suggest evaluation of the impact of patient partnership on the research team and quality of research and movement beyond patient-specific measurement to assessment that includes multiple stakeholders. Moreover, study teams should outline how they have responded to patient engagement evaluation findings – evaluation and tools are only useful if they stimulate action.

Ingredients for Impactful Research

Grounded in values and shared decision making, EMPOWER recipients are clear on successful ingredients for impactful research: first and foremost is the central role of patients and communities. If POR is intended to yield impactful change, matching patient/community priorities and engagement approaches is essential for success in dissemination, uptake and wider impacts. Awardees emphasized the need to create engagement-capable environments and the strategies they used to achieve them: mechanisms to promote genuine inclusion, joint decision making about the research, intended impacts and mechanisms to achieve these impacts. Together teams co-created alternate methods of engagement, taking a multi-faceted approach – for example, including multiple patient partner perspectives, involving clinicians and policy makers, patient and researcher role-sharing, centralizing lived experience, providing resources and supports to increase awareness of roles, sharing collaboration along a pathway forward and active engagement in research priority setting.

Positive impacts exemplify the value-add of the projects: increased engagement and motivation to effect change on the part of both patients and researchers, mutual learning, strengthening team cohesiveness and mobilizing findings into policy recommendations. That all of the articles are either led or co-authored by patient partners speaks to the prioritization of patient and community perspectives and experiences.

The continuation of SPOR signals the ongoing importance of POR in Canada and a shift toward a rapid learning health systems approach. Learning health systems approaches leverage advancements in data science, experience and best practices for health system improvement (Menear et al. 2019). These systems are characterized by engaged patients, relevant and timely generation of data-driven evidence, appropriate decision-making supports and flexibility in governance, finances and service delivery to support rapid learning and performance improvement that fosters collaboration, continuous learning, adaptation and overall system improvement (Lavis et al. 2018). Patient and community engagement, experience and partnership are core to health system transformation at a time when building comprehensive and performance-effective systems are key to the best possible health outcomes. Moving forward in this context, it behooves us to consider how to embed patient engagement infrastructure and policies within our health institutions and organizations for meaningful impact.

Embedding POR within learning health systems will ensure that patient perspectives and experiences are centralized in health systems and infused throughout rapid-cycle learning and performance improvement efforts. How can we maintain or advance timely, meaningful research–practice–policy partnerships with patients and communities and avoid losing POR gains in the context of rapid-cycle health system improvements? Instead, how do we apply what we have learned in POR to maximize patient engagement in health system design and delivery? How do we shift the narrative such that involvement becomes part of everyday conversations? How do we support organizations and health systems to incorporate patient engagement within their governance and policies and evaluate their engagement strategies to foster rapid-cycle learning in this realm? And importantly, how do we create spaces to equitably include patient and community perspectives, avoid further marginalization and address intersectionality?

As patient partners expressed at the recent OSSU Training and Capacity Building Event, we need to "meet people at the intersections" to best understand how to address the determinants of health and achieve health equity for all. The messages assert a call to action to mobilize the knowledge we have gained as we continue to advance the science and practice of patient and community engagement. It is about enhancing *partnership*-engaged knowledge mobilization for impact. HQ

References

Abelson, J., K. Li, G. Wilson, K. Shields, C. Schneider and S. Boesveld. 2016. Supporting Quality Public and Patient Engagement in Health System Organizations: Development and Usability Testing of the Public and Patient Engagement Evaluation Tool. *Health Expectations* 19(4): 817–27. doi:10.1111/hex.12378.

Asmundson, G.J.G., C. Blackstock, M.C. Bourque, G. Brimacombe, A. Crawford, S.H. Deacon et al. 2020. Easing the Disruption of COVID-19: Supporting the Mental Health of the People of Canada—October 2020—An RSC Policy Briefing. *FACETS* 5(1): 1071–98. doi:10.1139/facets-2020-0082.

Birnie, K.A., T. Killackey, G. Backlin, F. Gavin, C. Harris, I. Jordan et al. 2022. Ensuring Equity and Inclusion in Virtual Care Best Practices for Diverse Populations of Youth with Chronic Pain. *Healthcare Quarterly* 24(Special Issue): 25–30. doi:10.12927/hcq.2022.26778.

Browne, A.J., C.M. Varcoe, S.T. Wong, V.L. Smye, J. Lavoie, D. Littlejohn et al. 2012. Closing the Health Equity Gap: Evidence-Based Strategies for Primary Health Care Organizations. *International Journal for Equity in Health* 11: 59. doi:10.1186/1475-9276-11-59.

Canadian Institutes of Health Research (CIHR). 2011, August. *Canada's Strategy for Patient-Oriented Research: Improving Health Outcomes through Evidence-Informed Care*. Retrieved November 15, 2021. <https://cihr-irsc.gc.ca/e/documents/P-O_Research_Strategy-eng.pdf>.

Canadian Institutes of Health Research (CIHR). 2019, May. Strategy for Patient-Oriented Research – Patient Engagement Framework. Retrieved August 20, 2021. <https://cihr-irsc.gc.ca/e/48413.html>.

Hamilton, C.B., A.M. Hoens, S. McQuitty, A.M. McKinnon, K. English, C.L. Backman et al. 2018. Development and Pre-Testing of the Patient Engagement in Research Scale (PEIRS) to Assess the Quality of Engagement from a Patient Perspective. *PLoS ONE* 13(11): e0206588. doi:10.1371/journal.pone.0206588.

Hamilton, C.B., A.M. Hoens, A.M. McKinnon, S. McQuitty, K. English, L.D. Hawke et al. 2021. Shortening and Validation of the Patient Engagement in Research Scale (PEIRS) for Measuring Meaningful Patient and Family Caregiver Engagement. *Health Expectations* 24(3): 863–79. doi:10.1111/hex.13227.

Haywood, K., A. Lyddiatt, S.J. Brace-McDonnell, S. Staniszewska and S. Salek. 2017. Establishing the Values for Patient Engagement (PE) in Health-Related Quality of Life (HRQoL) Research: An International, Multiple-Stakeholder Perspective. *Quality of Life Research* 26(6): 1393–404. doi:10.1007/s11136-016-1465-5.

Lavis, J.N., F.-P. Gauvin, C.A. Mattison, K.A. Moat, K. Waddell, M.G. Wilson et al. 2018, December 10. *Rapid Synthesis: Creating Rapid-Learning Health Systems in Canada*. McMaster Health Forum. Retrieved November 28, 2021. <https://www.mcmasterforum.org/docs/default-source/product-documents/rapid-responses/creating-rapid-learning-health-systems-in-canada.pdf?sfvrsn=4>.

Menear, M., M.A. Blanchette, O. Demers-Payette and D. Roy. 2019. A Framework for Value-Creating Learning Health Systems. *Health Research Policy and Systems* 17: 79. doi:10.1186/s12961-019-0477-3.

Mulder, C., D. Debassige, M. Gustafson, M. Slater, E. Eshkawkogan and J.D. Walker. 2022. Research, Sovereignty and Action: Lessons from a First Nations–Led Study on Aging in Ontario. *Healthcare Quarterly* 24(Special Issue): 93–97. doi:10.12927/hcq.2022.26767 .

Ontario SPOR SUPPORT Unit. 2022. Fairness is Excellence: The Ontario SPOR SUPPORT Unit's Equity Framework. Retrieved March 11, 2022. <https://ossu.ca/resources/equity-framework/>.

Rahimi, S.A., H.T.V. Zomahoun and F. Légaré. 2019. Patient Engagement and Its Evaluation Tools – Current Challenges and Future Directions. *International Journal of Health Policy and Management* 8: 378–80. doi:10.15171/ijhpm.2019.16.

Slattery, P., A.K. Saeri and P. Bragge. 2020. Research Co-Design in Health: A Rapid Overview of Reviews. *Health Research Policy and Systems* 18: 17. doi:10.1186/s12961-020-0528-9.

Syan, A., J.Y. Lam, L.D. Hawke, K. Darnay and J. Henderson. 2022. The Youth Wellness Quest: A Comprehensive Online Mental Health Literacy and Self-Advocacy Resource Developed by Youth for Youth. *Healthcare Quarterly* 24(Special Issue): 55–59. doi:10.12927/hcq.2022.26773.

Tingley, K., M. Smith, N. Pallone, P. Chakraborty and B.K. Potter. 2022. Patient Engagement in a Multi-Stakeholder Workshop to Plan the Collection of Patient-Oriented Outcomes for Children with Inherited Metabolic Diseases. *Healthcare Quarterly* 24(Special Issue): 81–85. doi:10.12927/hcq.2022.26769.

Vanderhout, S.M., C.S. Birken, P.D. Wong, S. Weir, J. Batten and J.L. Maguire. 2022. Parent Engagement in a COVID-19 Cohort Study of Children and Families: Successes, Challenges and Next Steps. *Healthcare Quarterly* 24(Special Issue): 31–34. doi:10.12927/hcq.2022.26777.

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