

# Meaningful Patient Engagement in Research: Lessons From Retinoblastoma

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## **RICHELLE'S STORY: MY RETINOBLASTOMA JOURNEY**

At only 20 months old, I was diagnosed with bilateral, heritable retinoblastoma: an aggressive childhood eye cancer. The news was unexpected, because I was the first person in my family to be diagnosed with this cancer. With approximately 8000 new cases around the world each year and just 24 new cases annually in Canada,<sup>1</sup> it is unsurprising that my family had never heard about retinoblastoma. My right eye was surgically removed (enucleated), and I received chemotherapy and cryotherapy over the course of about 1 year. That was when my family heard the good news: the treatment succeeded in saving my left eye with good vision.

Growing up in Ontario, I received exceptional care and returned to the eye clinic and oncology clinic at The Hospital for Sick Children for annual appointments. However, as young adulthood emerged, I felt disheartened and overwhelmed in trying to understand the complexities behind retinoblastoma as a rare, heritable cancer. I had assumed it to be a childhood cancer that was behind me. I instead learned of the lifelong implications ahead of me: I could pass on a disease-causing gene to my future children and I carry a higher risk of developing second cancers.<sup>2</sup> Through oncology appointments, genetic counseling, and my own research, I was able to gain a rudimentary understanding of retinoblastoma.

However, I still felt that I lacked access to new, ongoing, and relevant research in the field, something I now know to be a commonly felt experience among retinoblastoma patients.<sup>2</sup>

In my search for new research, I was connected with the retinoblastoma research team at The Hospital for Sick Children. I initially presumed that, as a patient, I would not have a primary role in research. Historically, and from my own understanding, patients participating in research are treated as subjects in clinical trials and are not commonly engaged in the research process. To my surprise, this was not the case. I was asked to codirect, design, and implement a patient-engagement strategy for retinoblastoma research in Canada.

## **RESEARCHER AND CLINICIAN PERSPECTIVE: ENGAGING PATIENTS IN RETINOBLASTOMA RESEARCH**

People affected by retinoblastoma have indicated that they are deeply incentivized to keep abreast of research and help co-create it.<sup>2</sup> Parents want better outcomes for their children as they grow older and start families of their own, and survivors want research focused on preventing retinoblastoma, given that it can be predicted in familial cases.<sup>2</sup> As the retinoblastoma team at The Hospital for Sick Children, we noticed patients' interest in becoming more deeply involved in research but recognized that there was little opportunity for such involvement.

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Ms Baddeliyanage and Dr Shaikh wrote sections of the manuscript and reviewed the finalized manuscript; Dr Dimaras conceptualized the manuscript, wrote a section of the manuscript, and reviewed the manuscript; Ms White conceptualized the manuscript, synthesized the patient, medical, and research perspectives to write the initial manuscript, and revised the manuscript; and all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

**DOI:** <https://doi.org/10.1542/peds.2018-2166>

Accepted for publication Mar 27, 2019

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PEDIATRICS (ISSN Numbers: Print, 0031-4005; Online, 1098-4275).

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**FINANCIAL DISCLOSURE:** The authors have indicated they have no financial relationships relevant to this article to disclose.

**FUNDING:** No external funding.

**POTENTIAL CONFLICT OF INTEREST:** The authors have indicated they have no potential conflicts of interest to disclose.

**To cite:** White E, Baddeliyanage R, Shaikh F, et al. Meaningful Patient Engagement in Research: Lessons From Retinoblastoma. *Pediatrics*. 2019; 143(6):e20182166

More broadly, the importance of better engaging patients in research is also recognized by health researchers and health care professionals, with more granting agencies and research institutions making it a priority. The Canadian Institutes of Health Research (CIHR) has developed a Strategy for Patient-Oriented Research (SPOR), which defines patient-oriented research as the process by which patients (including survivors, their families, and informal caregivers) are engaged throughout the research and knowledge translation process.<sup>3</sup> SPOR follows in the footsteps of international efforts to center research and care around the patient, including the United States' Patient-Centered Outcomes Research Institute<sup>4</sup> and the United Kingdom's INVOLVE.<sup>5</sup> Including patients in research may be 1 way to more rapidly translate research results from bench to bedside.<sup>6</sup> Patients can contribute by creating novel research questions or sharing exciting research results with their peers. Knowledge created from such patient involvement in research is more likely to be relevant, adopted, and implemented.<sup>7</sup>

In response to patient demand and need for inclusive, high-quality, and translatable retinoblastoma research, we developed the Canadian Retinoblastoma Patient Engagement Strategy in 2016. The strategy is governed by the Canadian Retinoblastoma Research Advisory Board (CRRAB), which includes patients with retinoblastoma, health care professionals (eg, medical doctors, nurses, genetic counselors, and social workers), researchers, and patient-engagement experts (eg, individuals whose professional or research role involves patient engagement) who all volunteer their time.<sup>8</sup> The overall aim for CRRAB is to facilitate the development of meaningful,

codirected retinoblastoma research that is relevant to patients and improves health outcomes. Seed funding for the development of CRRAB was provided by a CIHR-SPOR collaboration grant, with additional funding and in-kind support from 2 advocacy groups: the Canadian Retinoblastoma Society and World Eye Cancer Hope. To recruit CRRAB members, clinicians treating retinoblastoma across Canada were asked to send invitation letters to their current and past patients. Similarly, advocacy organization partners promoted CRRAB via social media. Interested members self-identified and attended the first annual general meeting. Members selected the leadership and developed patient-engagement activities at the meeting.

CRRAB meets as a whole on an annual basis, and smaller focused working groups push forward its mandate throughout the year. Since its inception, CRRAB has (1) developed a research registry,<sup>9</sup> which is a sustainable vehicle to engage a diverse group of patients in research; (2) completed a priority-setting exercise, which set the top 10 retinoblastoma research priorities in Canada (manuscript in preparation); and (3) developed a Web site and newsletter to communicate important information with the retinoblastoma community.<sup>8</sup> The impact of these patient engagement initiatives is currently being evaluated and will be reported in future publications.

### **RICHELLE'S STORY: REWARDING ENGAGEMENT**

As an undergraduate student studying biology, I was unfamiliar with the concept of patient engagement in research. I did not entirely understand its importance until I was fully immersed in it as a survivor of retinoblastoma. Now having worked with CRRAB

for the last 2 years, it is clear to me that patient collaboration in setting research priorities is vital to ensure that results produced are relevant, adopted, and implemented into the health care system. Authentic collaborations between patients, health care professionals, and researchers are essential to the success of research and, in turn, its applicability to improve health outcomes. In my experience, patient engagement not only improves research findings but also empowers patients to be advocates for this new information.

As a patient, my voice, experiences, and knowledge were greatly valued in the creation and implementation of the Retinoblastoma Champion Program. The purpose of this program is to bring together a diverse group of individuals affected by retinoblastoma to promote patient engagement in research. As the program lead and champion myself, I train interested patients with retinoblastoma to promote the research registry (through social media and personal networks) among the Canadian retinoblastoma community. As a member of the retinoblastoma research team, I provide an important perspective to researchers, especially to those who rarely interact with patients.

My mother and I were also involved in a research priority-setting exercise to identify the top 10 research priorities in Canada. Throughout this process, a highlight for me was the mutual respect among researchers, clinicians, and patients, which drove inspiring and meaningful dialogue. Patient participants were recognized for their diverse life experiences rather than seen as 1 homogenous group. Although this made the process of narrowing down the list to just 10 research priorities a difficult and arduous process, the results are now much richer and may lead to more meaningful research.

## RESEARCHER AND CLINICIAN PERSPECTIVE: CHALLENGES WITH PATIENT ENGAGEMENT IN RESEARCH

CRRAB seeks to embody meaningful, authentic patient engagement; however, doing so is not without its challenges. The first challenge is in eliminating confusion about what “patient engagement in research” really means. Even among health care professionals and researchers, patient engagement in research is often misconstrued as equivalent to patient participation in focus groups or the tokenistic involvement of patients on advisory panels.<sup>6</sup>

Beyond confusion, the “engagement” piece of patient engagement is difficult to achieve. Despite good intentions, CRRAB working group meetings suffer from inconsistent participation of members. It is not necessarily for lack of buy-in but for practical reasons of limited time and resources. Falling attendance is a common problem according to a systematic review of 66 studies that engaged patients in the research process; even after patients agreed to engage in research, low attendance rates lengthened the time (and therefore the cost) of the study.<sup>10</sup>

Furthermore, teaching patients about the research process, from writing grants to understanding study design and dissemination, is currently a large focus in the patient-engagement community and an area that is actively being funded.<sup>11</sup> Although this is necessary and important, in our experience, this can only go so far without providing patients with the logistic support and resources they need to effectively contribute. Support tasks, such as setting working group meetings, following-up with patients, maintaining registries, or establishing a social media presence, are essential to facilitating patient engagement in research. Yet, these items are often not covered by funders because they are considered to be tasks that can be done by patient volunteers. We argue

that it is not always possible, nor fair, to rely on volunteers from the patient community to fill these roles. To achieve sustainable patient engagement, new research and administrative staff roles need to be considered, with the express purpose of facilitating patient-oriented research. Funding agencies (such as CIHR-SPOR and Patient-Centered Outcomes Research Institute, which already support the development of patient-engagement methods) could be 1 source of support for such roles. At The Hospital for Sick Children, we have recently embedded a Parent in Research role within our retinoblastoma research team, an attempt to more effectively engage patients in research. As evidence mounts regarding the benefits of patient engagement in research, it is possible that these roles may become common in research institutes across the world.

## JOINT PERSPECTIVE

### Achieving Meaningful Patient Engagement in Research

Authentic collaboration among patients, health care professionals, and researchers is essential to meaningful patient engagement. For Ms Baddeliyanage, being engaged in research that directly affects her is an exceptionally empowering experience and is related to her personal goal of pursuing a career as a genetic counselor. Her tasks as part of the Canadian Retinoblastoma Patient Engagement Strategy were tailored to match her experience, interests, and skills. Meaningful patient engagement, in our experience, necessitates collaboration with patients to identify roles that can add value to the research. This does not preclude seeking out training to develop additional skills and expand patient contributions to research (only recognizing that this requires additional time, funding, and support).

On the other side of the partnership, clinicians and researchers want to learn more about the tangible, evidence-based benefits of patient engagement in research. The often-cited benefits are the following: aligning research funding to patients’ priorities and daily experiences, identifying relevant outcome measures, improving recruitment and retention in studies, and disseminating results in patient-centric language such that it reaches beyond the confines of academia.<sup>6,7,12</sup> Yet, the incentive to pursue patient engagement must be demonstrated as greater than the added time, money, effort, and complexity that it involves. Changing the criteria by which health researchers are evaluated to include patient-engagement activities may provide added incentive in this regard.

### A Vision for the Future: Sustaining Meaningful Patient Engagement in Research

Ms Baddeliyanage now has a greater understanding how patients can meaningfully engage in research. She envisions a future in which patients can be empowered to participate from an early age and in which patient-oriented research becomes common practice. Although there has been great progress in the last decade, there is still a way to go to achieve Ms Baddeliyanage’s vision. From our experiences of engaging patients in retinoblastoma research, we have identified 3 main areas of change to support sustainable patient engagement in research.

First, we call on funders to recognize and respond to the need to support nonresearch activities that build capacity and facilitate patient engagement in research, taking into consideration the additional time and resources sustainable engagement requires. Funding such activities will help to create an environment conducive to meaningful patient engagement by making it accessible

to researchers daunted by the time and resources required. Second, the value of patient engagement should be made clear to health care professionals and researchers, and mechanisms should be in place to reward such work. Tokenistic patient engagement will only lead to inauthentic results and patients feeling that their time is being devalued. By communicating the evidence-based, tangible benefits and rewarding quality engagement, all parties are more likely to benefit from the experience. Third, meaningful patient engagement evolves from authentic partnership: all parties should consider capacity, skills, and interests of both patients and researchers to develop patient-research roles that are mutually beneficial.

These lessons learned from engaging patients with retinoblastoma in research are translatable to other fields. In pediatrics, several other such partnerships have seen clinicians and patients co-identify research priorities for childhood disability, mental health in children and young people, and neurodevelopmental disorders.<sup>13</sup> Similar to CRRAB, such partnerships systematically place researchers, clinicians, and patients on equal footing and provide each party with the opportunity to shape the research agenda.<sup>13</sup> As patient-oriented

research becomes more common, it becomes ever more important to support and reflect on the best practices of patient partnership in research, so that we can continue to learn from and advance the field.

#### ABBREVIATIONS

CIHR: Canadian Institutes of Health Research  
 CRRAB: Canadian Retinoblastoma Research Advisory Board  
 SPOR: Strategy for Patient-Oriented Research

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*Pediatrics* 2019;143;

DOI: 10.1542/peds.2018-2166 originally published online May 23, 2019;

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