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.

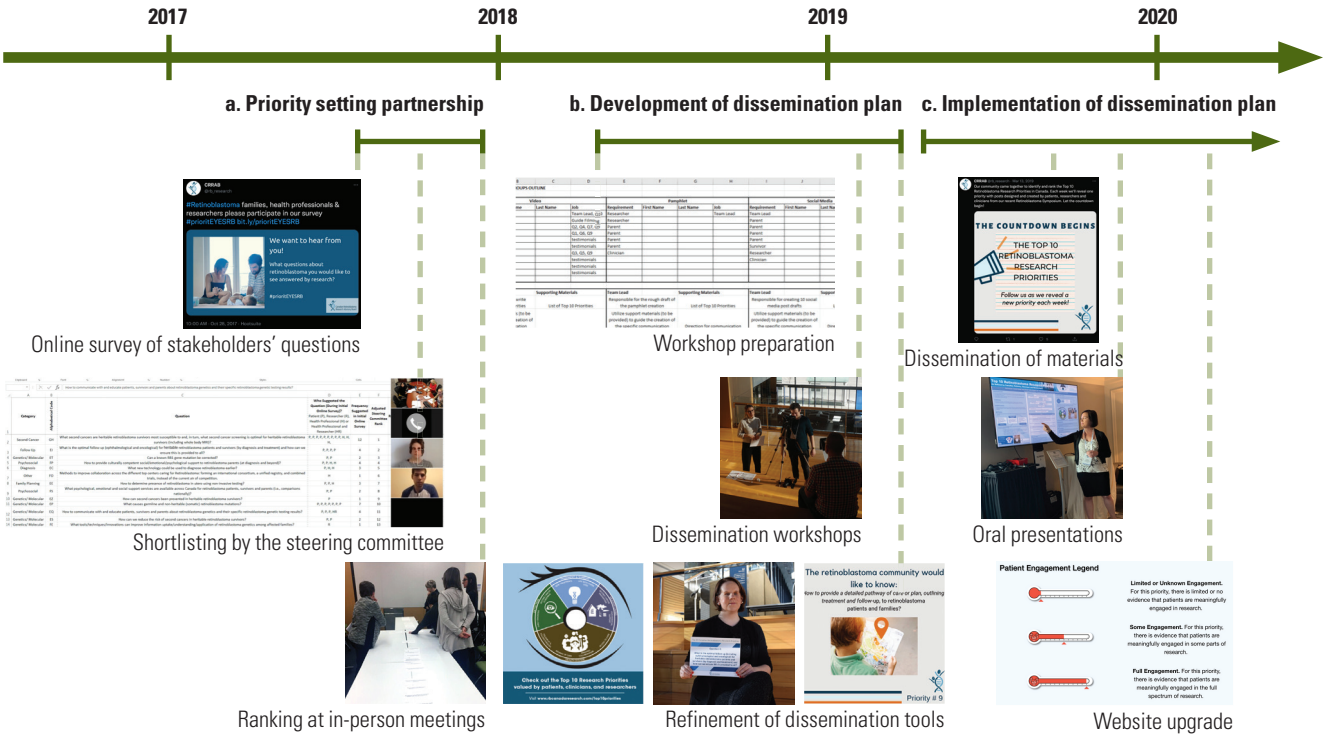
At its inception, the CRRAB structure was composed of a steering committee and three working groups, each led by a patient and non-patient co-chair. CRRAB members attended an annual general meeting to set goals and evaluate annual progress. Throughout the year, the working groups met online every four 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).

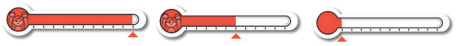
Presentation at an international conference

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.

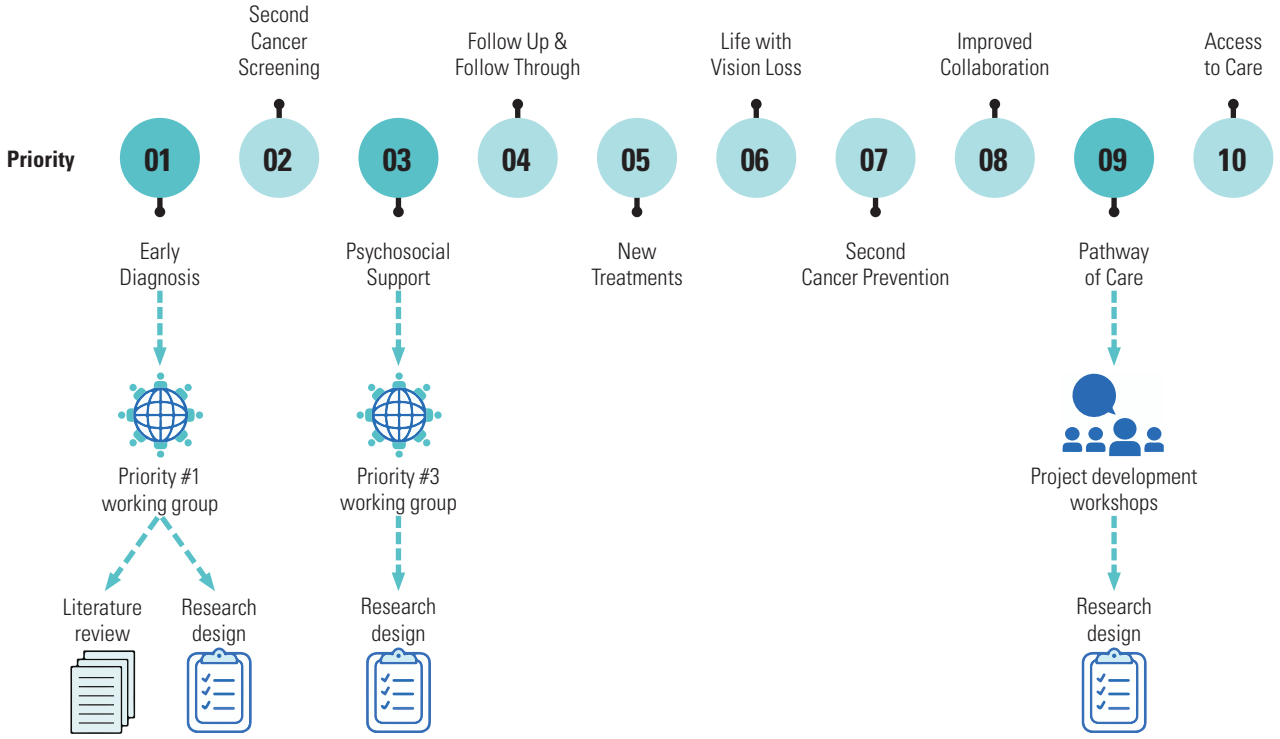
Discussion

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.

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