Author’s response to reviews

Title: The Canadian Retinoblastoma Research Advisory Board: A Framework for Patient Engagement

Authors:

Maxwell Jesse Gelkopf (max.gelkopf@medportal.ca)
Iva Avramov (ivana.avramov@mail.utoronto.ca)
Richelle Baddeliyanage (richelleb@Dal.Ca)
Ivana Ristevski (ivana.ristevski@sickkids.ca)
Sarah Johnson (johnsonsarah@gmail.com)
Kaitlyn Flegg (kaitlynflegg@gmail.com)
Helen Dimaras (helen.dimaras@sickkids.ca)

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Author’s response to reviews:

Sophie Staniszewska, Warwick Research in Nursing, University of Warwick, UK
Richard Stephens, Involved and engaged patient, UK
Editors-in-Chief, Research Involvement & Engagement
27 December 2019

Dear Ms. Staniszewska and Mr. Stephens,


Thank you for reviewing our article submitted as an Original Article in Research Involvement and Engagement, our manuscript entitled, “The Canadian Retinoblastoma Research Advisory Board: A Framework for Patient Engagement”.

We respond to the comments in a point-by-point fashion below.

Reviewer #1:
This is an interesting reflection on the importance of public patient engagement and generally well written. The case for engagement to promote the management of this disease is justified. The development of the PPI group is presented as a constructive and informative way to support future research and policy development related to the support of families and individuals with retinoblastoma. The use of social media and alternative ways of communicating with the population could be valuable in terms of future engagement and as a supportive measure.

RESPONSE: Thank you for these positive comments on our manuscript.
The PPI group are volunteers and therefore more likely to be highly motivated and engaged with the topic, so it is interesting to discover that there is a passive subgroup, how did/does this affect their usefulness? This could be an area of future enquiry?

RESPONSE: The passive subgroup attended the general meetings, where their opinions and expertise contributed to CRRAB strategic directions and activity planning, along with those of the more actively involved subgroup (i.e. those participating in developing research proposals, engaging as research champions). In future, we plan to look at participation type and precise usefulness to patient engagement activities.

We revised the section on the passive subgroup as follows: “While the majority of CRRAB members reported being engaged in CRRAB activities, a significant proportion (35%) of the membership were passive participants, attending meetings but not participating in additional CRRAB activities (e.g. developing research proposals, becoming RB Champions). This might mean that CRRAB membership represents a natural distribution of involvement from passive to active, including individuals who are typically difficult to involve in research activities. Although an advisory board is considered one of the most active forms of patient engagement (19), CRRAB has been designed so that individuals can choose to participate each according to their interests, skills and availability. The involvement of both passive and active participants in CRRAB suggests that overall leadership and decision-making is shared and does not reflect only one type of participant. A future direction is to further understand the motivations and interests of this passive subgroup, and identify if there are other ways to increase their engagement, or if they desire to remain as they are.” (Pages 29-30, lines 674-688).

I'm unsure if the quantitative data, and data from the pre-test post-test questionnaires, adds much to the study. The qualitative findings are very interesting and could be further elaborated.

RESPONSE: We have further elaborated our qualitative findings (Pages 17-18, lines 381-413, and page 21, lines 453-470).

The limitations section needs more consideration in relation to the methods, recruitment and population in the study.

RESPONSE: We have expanded our limitations section to address issues related to methods, recruitment and population (Pages 31-32, lines 728-781).

The conclusion is rather bold. Suggesting that 'CRRAB is an effective vehicle…' needs rewording. Any declaration of effectiveness requires qualification with more data; however degree of engagement could be cited with possibilities for future evaluation of effectiveness.

RESPONSE: We have revised our lay summary (Page 2, lines 40-42), abstract (Page 4 lines 76-78) and conclusion (Page 33, lines 830-832) accordingly.

On the whole a nice neat little study which is promoting an important supportive mechanism for good clinical and research practice for retinoblastoma.

RESPONSE: Thank you, we hope you find our revisions suitable for publication.

Reviewer #2:
The concept of this paper is interesting, but this reviewer believes it to be premature. The perceived purpose of the paper is to document the efficacy of the Canadian Retinoblastoma Research Advisory
Board in patient engagement. The paper reports that there are approximately 2000 Retinoblastoma survivors in Canada. The paper also discusses that the number of patients that participated in the Canadian Retinoblastoma Research Advisory Board were 21 in 2016 and 35 in 2017. This reviewer did not feel that the numbers of patient participants was a meaningful amount to document high levels of patient engagement.

RESPONSE: We have clarified this in the manuscript that “There are an estimated 2000 retinoblastoma survivors in Canada, each requiring extended clinical follow-up and long-term interaction with the healthcare community; however, it is not clear if these individuals remain aware of their childhood diagnosis or connected to a follow-up clinic.” (Pages 4-5, lines 91-101).

The numbers of individuals engaged with CRRAB are actually reflective of quite good coverage for recently affected families, for which we’d expect 20-25 new patients per year. Thus, our early results with CRRAB show promise for engaging the estimated 2000 survivors who have lost contact with the retinoblastoma clinics.

In the section entitled, "Lay Summary", the researchers make the statement, "the results suggest that CRRAB is effective in meaningfully engaging patients in retinoblastoma research and provides direction to sustain and enhance future patient engagement" (p. 2). Overall, the reviewer wonders how much engagement there is outside of a few interviews and focus groups at an annual conference. The results of those questionnaires/focus groups seemed biased since they are only reflective of attendees of an annual meeting and the overall numbers are low in comparison to the total population of Retinoblastoma survivors.

RESPONSE: We have revised the conclusion of the lay summary as indicated in above response to Reviewer 1.

We wish to clarify that the participation in CRRAB activities outside of the annual general meeting is shown in Figure 1; only 35% of meeting attendees reported not participating in any activity, and 65% reported participating in at least 1 activity. Engagement in these activities (i.e. working groups, RB champions) are currently being evaluated using the Public and Patient Engagement Evaluation Tool (PPEET) and not the focus of the current paper.

The chart board discussions identified on page four of the manuscript also imply a bias since these are people who already wanted to participate and not reflective of the overall patient population.

RESPONSE: We have indicated this as a potential limitation as follows, “The study also only reflects the population who was able to attend the annual general meeting, and may have excluded patients who participated in other CRRAB activities but missed the meetings” (page 31, lines 736-738).

The conclusion reached on page 4 states that, "CRRAB is an effective vehicle for engaging patients in retinoblastoma research" but does not really state why.

RESPONSE: We have revised the conclusion of the lay summary, abstract and full manuscript, as indicated in above response to Reviewer 1.

On page 6 of the manuscript, the researchers also specify that there were focus groups held in Toronto and Calgary but does not indicate how many or with what frequency.

RESPONSE: As mentioned in the manuscript, the results of the focus group study is reported in a pending publication.
Additionally, the researchers go on to say that "numerous patients were involved throughout the
development and implement of CRRAB" (p. 7), however, the term, numerous, is not quantifiable and
could be 10 or 100.

RESPONSE: We have revised this section for accuracy and clarity as follows: “Representatives of the
patient advocacy groups, World Eye Cancer Hope (www.wechope.org) and
The Canadian Retinoblastoma Society (https://www.rbsociety.ca/), along with 3 additional patient
partners, served collaborators on a grant to obtain seed funding for the initial activities of the strategy,
including the first CRRAB meeting. These patient partners were involved in the design, development
and initial implementation of CRRAB and continue to guide and direct CRRAB in leadership roles and
assist with research dissemination” (Page 7, lines 166-171).

In the section entitled, "Evaluation of CRRAB: Data Collection and Analysts Procedures, it indicates
that the data collected to evaluate the efficacy of the CRRAB patient engagement program was taken at
two annual meetings. This, in and of itself, would appear to be biased since the population of
Retinoblastoma patients would not be adequately represented in these surveys. It was noted by this
reviewer that the exclusion of questionnaires of people who did not completed both the pre-and post-
test was appropriate.

RESPONSE: There may have been patient partners who participated in CRRAB patient engagement
activities but who did not attend the annual general meeting to have their views counted in the survey;
we have indicated this is a limitation of the study (Page 31, lines 736-738). The engagement of
additional retinoblastoma patients is ongoing to ensure a representative group is achieved.

In the section entitled, "Chart Board Discussions", this reviewer feels that there was not sufficient
information to determine if this was meaningful since, again, the overall population of patients is not
well represented.

RESPONSE: Having clarified that the results are representative of CRRAB members who attended the
annual general meetings, we hope that this is now clear to the reader.
The CRRAB structure also does not provide sufficient information as to the makeup of the working
groups or the number of people to support a conclusion that CRRAB is effective in patient engagement.

RESPONSE: We now include information on the working groups: “WG are composed of 5-10
individuals, and led by patient and non-patient co-chairs” (Page 11, line 280).

Additionally, although the researchers do include information about their Twitter feed, it appears that
there is not much patient engagement by the amount of likes and retweets.

RESPONSE: Social media likes and reposts represent only one aspect of interaction with our target
population, and not reflective of the full spectrum of patient involvement in research (the focus of our
paper). We plan to look at social media analytics in future to better understand our recruitment and
awareness efforts (Page 33, lines 821-823).

Furthermore, with the questionnaires provided at the first and second annual CRRAB meetings, there is
still a concern that these results are not meaningful since they
represent a very small subset of patients as compared to the overall patient population in Canada.
Additionally, at the 2nd Annual CRRAB meeting, there was a small percentage of participants who were aware of the patient engagement website and other forms of social media which to this reviewer would infer that there was not efficacious patient engagement.

RESPONSE: The low awareness of CRRAB social media among some participants is an important and valid finding of this study. However, as we state above, this is not reflective of low or non-efficacious patient engagement in research, but indicative of a patient partner awareness and recruitment method that could be improved. We indicate in the discussion how we are attempting to improve awareness and reach of social media (Page 26, lines 601-604).

In the section entitled, "Impacts, Context of PPI", the researchers state, "our results showed robust patient awareness and strong involvement" (p. 21), but this reviewer, again, has concerns that this is based on data from a very small subset of patients as compared to the total population. Additionally, the information was taken based on people who attended the conference who are already considered to be highly engaged.

RESPONSE: We have revised the statement to more precisely reflect our findings: “Our results showed significant overall patient awareness of CRRAB activities (Figures 1 and 2) and 65% of participants reported participating in at least one CRRAB activity (Figure 1)” (Page 23, lines 509-511).

We remind the reviewer that the individuals who attended the annual general meeting consisted of both those who were highly engaged as well as a passive subgroup (Page 29, lines 675-678), i.e. the 35% who did not participate in any activity (Figure 1).

In the "Population Characteristics" section the researchers state that "for parents, those with more recent diagnoses might be less likely to engage with CRRAB as the diagnosis may be overwhelming and parents may struggle with maladaptive coping mechanisms" (p. 25). This reviewer would disagree with this statement. The inference of "maladaptive coping mechanisms" itself could turn away potential patients from engaging in CRRAB.

RESPONSE: The inference to maladaptive coping mechanisms refers to results from a prior study on parental coping with retinoblastoma (reference #2). We have altered the sentence for clarity: “Prior research indicates that parents may struggle with maladaptive coping mechanisms around the time of retinoblastoma diagnosis (2), thus these individuals may be less likely to participate in CRRAB and its activities” (Page 26, lines 599-601)

Additionally, it is mentioned that there is a push on Instagram, however, there was no Instagram account that could be easily attributable to CRRAB.

RESPONSE: We now reference the Instagram account (@rb_research) on page 26, line 603.

In the "Motivations and Benefits of Engagement" section, the researchers state that patients "are also motivated to be part of CRRAB to focus research on the patient and align research goals with their own" (p. 26), however, again the numbers of participants as compared to overall population would not support this conclusion.

RESPONSE: As per our previous response, we have revised the manuscript such that the reader can infer that the results are reflective of the participants in the study (i.e. CRRAB members), not the wider
retinoblastoma population.

Additionally, the researchers state that, "a significant number of participants identified as passive participants" might "represent a natural distribution of involvement from passive to active" (p. 28), however, this could also mean that there is not an efficacious patient engagement which would contradict this purpose of this paper. The conclusion appears to support the original position of the paper but is not based on data.

RESPONSE: We have edited this section for clarity: “While the majority of CRRAB members reported being engaged in CRRAB activities, a significant proportion (35%) of the membership were passive participants, attending meetings but not participating in additional CRRAB activities (e.g. developing research proposals, becoming RB Champions). This might mean that CRRAB membership represents a natural distribution of involvement from passive to active, including individuals who are typically difficult to involve in research activities. Although an advisory board is considered one of the most active forms of patient engagement (19), CRRAB has been designed so that individuals can choose to participate each according to their interests, skills and availability. The involvement of both passive and active participants in CRRAB suggests that overall leadership and decision-making is shared and does not reflect only one type of participant. A future direction is to further understand the motivations and interests of this passive subgroup, and identify if there are other ways to increase their engagement, or if they desire to remain as they are” (Pages 29-30, 675-689).

The paper does indicate that there was a third annual CRRAB meeting, but there is no data from that event to support the position that CRRAB's model is effective for patient engagement.

RESPONSE: The purpose of this study was to report on the initial implementation and outcomes of CRRAB. Results from subsequent meetings will be reported in future.

This reviewer does not find that the data provided would support a conclusion that "CRRAB is an effective vehicle for engaging patients in retinoblastoma research" (p. 31). Specifically, this reviewer does not feel that the number of participants would be able to support this conclusion. A suggestion for a longer longitudinal study to include the third annual CRRAB data and future years would provide more data.

RESPONSE: We have revised the conclusion to more accurately reflect our findings, as stated above.

It would also be suggested that the researchers consider reaching out to physicians across the country as a means of outreach instead of solely relying on the few participants who engage at an annual meeting.

RESPONSE: Recruitment efforts have focused on physicians, researchers and patients from across the country (Page 7, lines 179-180), many of whom have attended the annual meeting and participated in additional activities.

Reviewer #3:  
This article is of interest in the ever developing field of public and patient involvement research, which has become a discrete area of research. Of particular interest is the methodological approaches to measuring the engagement of the retinoblastoma community in Canada. The need for future studies to use predefined, validated tools to evaluate patient engagement at regular intervals is a point well made. There is heavy emphasis on the role of engagement (research done to) in this study and I do think that
there should be further focus on involvement (research done with), which is not the same as engagement. It would have been useful to establish this from the outset rather than conflate the two.

RESPONSE: We acknowledge that there are different terms and definitions for engagement and involvement. We have revised our introduction for clarity, where we indicate that we use Canadian definitions throughout the manuscript: “Patient and Public Involvement (PPI) in research is that which is carried out ‘with’ or ‘by’ patients and the public, as opposed to research conducted ‘on’ or ‘about’ these individuals (3). In Canada, PPI goes by the name of ‘Patient Engagement’, a concept that similarly shifts the idea of a patient from a passive data-point to an active participant who is encouraged to be involved in any or all aspects of the research process (4). The Canadian Strategy for Patient-Oriented Research (SPOR) promotes patient engagement as a meaningful and active partnership between patients and healthcare professionals in governance, priority setting, conducting research, and knowledge translation (5). In line with this notion, the SPOR definition of the term ‘patient’ to includes individuals with lived experience of disease, including family members and informal caregivers (6).” (Page 5, lines 102-111).

FROM THE EDITORS:
The reviewers have produced a wide range of responses, which is why we feel that Major Revisions are needed. There are perhaps two recurring themes. One is to be careful in the use of language to describe the significance of the work and the findings, and linked to that is the second theme, which is to recognise more prominently what the study does not show and what are the limitations of the work. We feel that this is a good study and look forward to sharing it with the wider community, but we want to ensure that it is evaluated and valued for what it does show rather than criticised for (perceived) over-claiming or for (unacknowledged) missed opportunities. It has the potential to be an important (if niche) addition to the evidence base, and that is what we hope the authors can achieve with a further revision. We are allowing up to 50 days for this work instead of the usual 30, because we do not want you to feel under pressure to meet artificial deadlines in responding to all the reviewers' comments and our suggestions too.

RESPONSE: Thank you for your consideration. We have made major revisions to clarify the language, and to more clearly point to our results and limitations. WE hope you now find this study suitable for publication.

Sincerely (on behalf of all co-authors),

Helen Dimaras, PhD
Assistant Professor,
Department of Ophthalmology & Vision Sciences, Faculty of Medicine, &
Division of Clinical Public Health, Dalla Lana School of Public Health, University of Toronto
Scientist
Child Health Evaluative Sciences & Center for Global Child Health, SickKids Research Institute
Director of Global Eye Health Research,
Department of Ophthalmology & Vision Sciences, The Hospital for Sick Children
555 University Ave, Toronto, ON, Canada M5G 1X8 | T: 416-813-7654  x201876 | E: helen.dimaras@utoronto.ca