Reviewer’s report

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

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Reviewer: Maria Carr

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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.

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. 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. 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.

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. 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.

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.

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.

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. 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.

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.

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.

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

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.

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.

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. 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. 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.

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