| **Article details: 2019-0221** | |
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| Title | The top 10 retinoblastoma research priorities in Canada as determined by patients, clinicians and researchers: a patient oriented priority setting partnership |
| Authors | Kaitlyn Flegg MSc, Maxwell J. Gelkopf BSc, Sarah A. Johnson PhD, Helen Dimaras PhD; for the Canadian Retinoblastoma Research Advisory Board Priority Setting Steering Committee |
| **Reviewer 1** | Leon Bijlmakers |
| Institution | Health Evidence, Radboud University Medical Centre, Nijmegen, Netherlands |
| General comments (author response in bold) | Solid piece of research, well written, well referenced.<br>1. The only suggestion I have concerns the response rate of people who took part in Phase I of the study: how many people (patients, clinicians, researchers) were approached? Assuming the response rate was not 100%, have you examined the profile of non-respondents? Would non-response have affected the study results?<br>**We do not have a way to know how many people were reached by the multipronged recruitment strategy. We have addressed concerns over study sample in the editor's comments 6 and 9, above.** |
| **Reviewer 2** | Alexandre Grégoire |
| Institution | Patient reviewer, Centre of Excellence on Partnership with the Patients and the Public (CEPPP), Centre de recherche du CHUM, Montréal, Que. |
| General comments (author response in bold) | Hello to you, I am a patient partner with cystic fibrosis and diabetes. I am also a lung and liver transplant recipient for seven years now. I have been working in the field of patient-oriented research for almost five years. In my work, I am primarily interested in the methodology behind implementing strategies for partnership with patients and the public to reduce the risk of patients tokenism. I am very pleased to had the opportunity to participate in the review of your manuscript. Here are a few comments regarding the patient and public partnership strategy implemented in your project:<br><br>1. WHO?<br>- How did you identify and recruit the patient partners involved?<br>- Did you use any kind of competency framework to select them?<br>- What were the selection or exclusion criteria for recruitment?<br>- What was your method of recruiting patient partners?<br><br>2. HOW?<br>Once the patient partners were recruited:<br>- What measures did you put in place to ensure that patient partners were able to fulfill their roles (training, mentoring, pairing with a coach, etc.)?<br>- How did you support and mobilize patient partners throughout the research project?<br>- Concretely, how were patient partners involved in the study design, implementation and dissemination of results?<br><br>3. WHEN?<br>For each of the stages of the research in which the patient partners were involved:<br>- What was the number of encounters with patient partners?<br>- How long did they last?<br><br>4. ROLES AND RESPONSIBILITIES?<br>For each stage of the research in which patient partners were involved:<br>- What were their roles and responsibilities? |

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| | - How were these roles and responsibilities established with them?<br>Throughout your manuscript, I would liked to have answers to these questions. Not necessarily in the article itself, as the number of words is limited. But, to describe the whole process of involving patient partners in an appendix for example would, in my opinion, give great value to the reader in prioritizing research. Also, I would liked to know the issues encountered throughout the process. I found the article very interesting.<br>**The details of recruitment, engagement and involvement of patient partners in our work is described in detail in our previous publication entitled "The Canadian retinoblastoma research advisory board: a framework for patient engagement" (Citation #5 in the current manuscript). We describe how CRRAB was a source for patient involvement in Priority Setting in the Methods, and have revised the "Patient Oriented Research" section to provide further details.** |
| **Reviewer 3** | Stirling Bryan |
| Institution | School of Population and Public Health, University of British Columbia, Vancouver, BC |
| General comments (author response in bold) | Establishing priorities for health research with full engagement of patients and families is essential work and so I commend the research team for taking on this project.<br>I also commend the team for extending the scope of the JLA process to include clinical perspectives, in addition to patients/families. I have often reflected that we need all stakeholders with lived experience, and that points to including the clinical community too.<br><br>1. My primary concern with the research relates to the size of the samples included in the various phases. I would like a fuller discussion of the samples typically recruited to JLA and similar processes. Is some sample size calculation possible? I think the general consensus would be that the samples here are small and likely not sufficient for drawing the broad conclusions cited in the paper.<br>**We have addressed the sample size issues in the editor's comments 6 and 9, above.**<br><br>2. A second reservation relates to the omission of a literature review as part of the process. I appreciate that a systematic review might not be feasible or appropriate, but I do think research priorities have to be framed in relation to existing research evidence and activity. Relying on the knowledge and expertise in the research team feels limited and open to challenge.<br>**We have addressed the sample issues in the editor's comment 2d, above.**<br><br>3. The patient orientation in this work is important and I commend the team for including one patient as a core member of the research team. In general, good practice is to include more than a single patient but I see that connection to other patients was a critical piece in this work.<br>**Patient partners were critical members of the Priority Setting working group and steering committee. Lead authorship from the patient perspective was provided by Dr. Sarah Johnson.** |