

Article details: 2016-0030	
Title	The development of a conceptual framework for understanding financial barriers to care for patients with cardiovascular-related chronic disease: a protocol for a grounded theory (qualitative) study
Authors	David J.T. Campbell MD MSc, Braden J. Manns MD MSc, Brenda R. Hemmelgarn MD PhD, Claudia Sanmartin PhD, Kathryn King-Shier RN PhD
Reviewer 1	Dr. Mary Chiu
Institution	Mount Sinai Hospital, Psychiatry, Toronto, ON
General comments (author response in bold)	<p>Major comments:</p> <p>1. In presenting Grossman Theory and Brown Framework of SEP on Health in great detail, the authors highlighted the limitations of existing theories. There was a general statement that (p.8 line 23-27) a novel framework is required due to these limitations. However, it may not be clear, how the proposed framework would address these limitations. In other words, the rationale is a bit lacking. There was mentioning of a "preceding survey study" – was this published (p.10, line 39)? Assuming this is a completed study, one suggestion is for the authors to describe the findings of this "survey study", and how that guided the development of the interview guide and, in turn, how the guide was constructed to address the limitations of existing theories.</p> <p>Thank you for noticing this important omission – the description of our prior study, which was very important for informing the current research. This has now been rectified:</p> <p><i>"This study is informed by our previous research in the area of financial barriers. We previously conducted a survey of 1849 western Canadians with cardiovascular-related chronic diseases to understand the barriers that they face in self-managing their conditions. We found that financial barriers were common among Canadians with these conditions (12%) and that there were important associations between financial barriers and clinically meaningful outcomes (medication non-adherence, emergency department visits and hospitalizations). Despite these findings, in the absence of a framework for understanding how financial barriers are experienced by individual patients, we have been unable to conceptualize the mechanisms by which financial barriers may translate into adverse clinical outcomes. A qualitative approach is required to obtain this level of granular data."</i></p> <p>2. In the Methods section, again, there were several mentioning of a "preceding survey study", with no details attached. It was unclear how the "survey" was or will be administered and to how many people. What is the connection between the survey and the GT study? What is the content of the survey? Was one of the survey questions as presented on p.10, lines 4-7 – and is this question used to identify a sub-population of the survey participants to be approached or recruited for the GT study?</p> <p>We hope that the details now provided about the preceding survey will address this comment – and clarify for the reader that the survey was a completely separate project.</p> <p>3. Related to the comment immediately above, it was mentioned that "Recruitment will be via signage in FP offices and specialist clinics as well as via pre-existing research and clinical databases". This sounded like two different approaches: active targeted recruitment and self-identification - unclear if these approaches would be for the survey or for the interviews.</p> <p>This approach will be for the interviews only as the survey was completed some time ago. Indeed, these are different approaches.</p> <p>4. Sampling and data collection: Development of a "novel" theoretical framework typically requires a sample size of more than 40. In this case, with the anticipated number of strata listed on p.10, a higher level of heterogeneity in the sample is implied. It may be useful to provide a range (i.e. 40 to 70) and please provide justification of sample size (reference?)</p> <p>We are focusing on a relatively narrow group of individuals with considerable homogeneity in their chronic conditions. We appreciate that providing a target range of the number of projected participants along with a reference would be helpful and have included this in the manuscript:</p> <p><i>"We anticipate needing to complete between 30 to 50 interviews to achieve saturation, based on prior reports of grounded theory studies of this nature" (citation: Morse, 1994)</i></p> <p>5. It is unclear why cardiovascular-related chronic diseases were chosen as the focus of inquiry, but chronic diseases in general is the focus of the "Background" section. There was inconsistency throughout the manuscript in the mentioning of a focus on CV-</p>

	<p>related chronic conditions until on p.9, line 21. Are other chronic illnesses excluded in the development of this framework? How does that affect the subsequent utility of the proposed framework? If indeed CV-related chronic condition is the focus, at least the title of the article should reflect that, and a rationale for this focus to be provided.</p> <p>Thank you for this important point of clarification. We have indeed changed the title to reflect the narrow focus of chronic diseases included in our proposed study and made it clearer throughout the background section that these are our target conditions.</p> <p>Minor comments:</p> <ol style="list-style-type: none"> 1. Inconsistency in statistics presented: In the Abstract, authors wrote "As many as 12% of Cdns with chronic diseases experience a financial barrier to carer". p.4, line 44, authors wrote "Up to 10%. Please clarify. Thank you for pointing this out, this has been rectified. 2. p.13, line18: repeated word - policymakers was mentioned twice in the sentence Thanks - corrected 3. p.17, line 11: contraction - What are the challenges you HAVE faced? Corrected 4. p.17, line 44: missing word - "What kinds of things do you do TO ensure..." Corrected
Reviewer 2	Dr. Luis Gabriel Cuervo, Ms. Pilar Garcia Del Vello Moreno, Mr. Christopher Khanoyan, Ms. Maria C. Medina, Ms. Louisa T. Stuwe
Institution	Pan American Health Organization, Research Promotion & Development, Washington, DC
General comments (author response in bold)	<p>General structure</p> <ol style="list-style-type: none"> 1. Although there is a structure for the article, it wasn't easy to follow the differences between objectives of the framework and the objectives of the study. It cannot be clearly seen if expected results of the study will be to design and apply a new instrument or finding new insights on different barriers of access to healthcare. Also elements such as inclusion and exclusion criteria need to be properly marked. Thank you. We have significantly reformatted the manuscript and hope that it reads much more clearly. In response to this comment we have also clearly marked the inclusion and exclusion criteria for study participants. 2. Chronic diseases seem to be an extremely wide group of conditions to be grouped under a unique framework that is later described as focusing on some very specific conditions, i.e. some cardiovascular diseases. The title is hence misleading. We have changed the title of the article to specify that we are dealing with cardiovascular-related chronic conditions. 3. It is usually helpful to provide a box with three bullets summarizing (1) what is known about the topic; (2) what the study adds; and (3) what needs to be done with the new knowledge (in terms of implementation and new research questions; this). Even if this is relevant to completed studies, it would be good as from the protocol to plan ahead on what is to be done with the findings so that any results have an impact. While some journals utilize this format, we have not seen this used in previous articles published in CMAJ Open. If the editors would like us to provide such a box, we would be happy to do so. <p>Background</p> <ol style="list-style-type: none"> 4. Stating that this kind of framework is novel needs to be backed by a proper review of the literature. The methodology described to assess this is excessively brief and superficial. A quick search using filters as "access and chronic diseases" or "access and cardiovascular disorders" in www.healthsystemsevidence.org and http://global.evipnet.org; the search drew some relevant studies, suggesting that there may be helpful studies and benchmarks. We invite the authors to check and provide a detailed description for the search of evidence to back their statements (filters, inclusion and exclusion criteria, period of time studied, dates of the study, how the evidence was summarized, databases used, etc.). We respectfully point out that the purpose of the current article is not to provide a detailed review of the literature, but rather to describe our future study. The kind of review described by the reviewer would add significantly to the word count, limiting our ability to adequately describe our study. We have focused the background section on the point that frameworks which describe access to care in the general sense may be inadequate to describe the detailed process of patients who experience financial barriers specifically. 5. The article needs to recognize that public health decisions are not always based

just on effectiveness, and that lack of access to all prescribed treatments is not per se a proof that the system is "not functioning properly. After all, the focus should be on providing safe cost-effective interventions. If a healthcare provider is prescribing an unsafe or ineffective treatment, or one that has poor cost-benefit ratio, there may be sound reasons for not providing it; this is similar with other health technologies. Therefore, the definition of "adequate health care" needs to be clarified (Page 4, Line 5-8). Clarification is needed about what is not universally included within the Canadian public health system, what the direct and indirect financial barriers are such as access to medicines for example (is there a copayment? How much does it cost? Is there evidence that this is a barrier?).

We have simply proposed a study to explore the experiences of individual Canadian patients who experience financial barriers. We do not wish to make any particular policy claims now. Certainly, the results of this study may have policy relevance, but since we are not presenting detailed results in this manuscript, we are somewhat unclear about this comment.

We have provided further clarification around our assertion about adequate healthcare, and provided greater details on the state of access to goods and services in Canada:

"For example, public insurance plans for outpatient prescription medications vary by province, but no province offers universal public medication insurance. Those fortunate enough to qualify for public medication insurance are still faced with considerable copayments and/or deductibles."

As to the reviewer's question about whether the costs represent a barrier, we feel that we answer this question in the next paragraph:

"A previous survey conducted by our group found that 12% of Canadians with cardiovascular-related chronic diseases experience financial barriers, and these people are 70% more likely to have hospitalizations or emergency department visits related to their ambulatory care-sensitive condition"

6. The diseases targeted by the study change progressively as the paper develops. Initially, the paper talks about chronic diseases in general. Then, in page 4, they will be cardiovascular. And finally, in page 9 line 54, the authors narrowed it to diabetes, hypertension, coronary artery disease, and stroke. It is not a study of chronic diseases; it is a study of a subset of NCDs listed above. What is meant by "they" in page 4, line 16?

We have now clarified from the beginning of the article (and the title page) that we are only dealing with cardiovascular-related chronic conditions. The ambiguous language in line 16 has been changed to "these" indicating the treatments that were described.

7. Lastly, the background is excessively long and the writing is ambiguous. The paper seems a mix of an educational article and a protocol for a research proposal. We suggest the authors to visit EQUATOR Network and use tools such as the checklist and guidance on PRISMA equity extension;www.equator-network.org because it may provide some insights into variables to consider in a study to later assess equity issues; and to check the guidelines for protocols and for public health studies.

We have restructured the article and moved the majority of the previous background information to the discussion section. While we appreciate the reviewer's suggestion to consult a standardized checklist, we do not feel that PRISMA is the appropriate checklist to inform our qualitative study as this tool is to be used for systematic reviews and meta-analyses. We have, however, ensured to be inclusive of the major reporting categories described in the COREQ checklist for qualitative research.

Need for a novel framework

8. The specific objectives we believe may have been covered by some existing research, including some cited.

While the research we cite certainly contributes to an understanding of access to care, we maintain that no framework exists for describing the experience of encountering financial barriers from the patient's perspective.

We agree that the sub-objective to describe what goods and services patients may have difficulty accessing has been described in the literature, so we have removed this from our listed objectives.

Methods

9. The study is only targeting the English speaking population. There should be a remark about how it affects populations in situation of vulnerability, migrants, indigenous people, and the bias this may introduce, or whether this will be taken into account as a limitation of the study.

We have added a limitation to this effect.

10. Interview patients with hearing and speech impairments. For example, patients with stroke, deafness, or another communicative limitation. This problem should be anticipated and explained in the methodology.

Thank you. We have clarified this in the exclusion criteria and included it in our limitations section.

11. Clarification needed about the types of population included: Migrants, aboriginal populations, etc.

We specify in our sampling strategy the types of individuals that we are explicitly seeking to include in this study, including Aboriginals. No specific inclusion or exclusion was made on the basis of immigration status.

12. Definition of a minimum sample size is needed before stopping, even if three consecutive cases give the same information by play of chance this could happen with the first participants, which would be a misleading way of terminating the study.

While this is a theoretical possibility, no experienced qualitative researcher would stop data collection after only a handful of interviews. Please see our response to Reviewer #1, comment #4 for a more thorough explanation regarding the sample size estimation.

13. Page 10, inclusion and exclusion criteria need to be listed, including issues such as language, communication skills, etc. For example, suppose that you are dealing with stroke patients.

We have clarified our inclusion and exclusion criteria as specified.

14. Expansion of sampling framework and data collection is needed. Clarify what information will be collected to assess equity (check equity extension of the PRISMA guideline for ideas www.equator-network.org). For example, you may want to add education status, employment, and financial situation.

The goal of this study was not necessarily to explore the issue of equity, but rather to explore the experience of having financial barriers, from the patient's perspective. Again, we do not feel that PRISMA is an appropriate tool to inform this work. We have included a comment specifying that our predetermined sampling strata were informed by our previous survey.

15. The description in page 11 line 46 of how coders will reach a decision is missing. The rules need to be presented upfront.

This is not typically done in qualitative research as it is not in keeping with the fundamental interpretive paradigm that underpins qualitative inquiry. We specify in our manuscript that the purpose of coder meetings is to 'allow for consideration of various perspectives' – due to the subjective and inherently interpretive nature of qualitative data, rules should not be imposed, but rather an iterative discussion between coders with the objective of expanding discussion as opposed to reaching consensus. Further detail on this has been provided in the manuscript:

"The process of initial coding will be done individually in triplicate, meaning that three experienced analysts will individually code the data. All coders will meet weekly to discuss their interpretations of the data to allow for consideration of various perspectives. Given that this research is informed by the interpretive paradigm, exact agreement is not the goal of these sessions, but rather to gather a multitude of viewpoints on the various incidents and themes derived from the data. However, consensus about how to code a given piece of data will be achieved after thorough discussion of each point. In cases where consensus is not reached allowances will be made such that a passage may retain multiple codes to enable future discussion."

16. The authors may consider the inclusion of a control group. For example, include in the study patients with chronic diseases but without known economic barriers as a control group. This may help to determine which barriers are specific to the vulnerable populations, which are affecting all.

Qualitative research is often used as a supplement or adjunct to experimental studies. It would be highly atypical to utilize a control group in a grounded theory study and would not necessarily help us achieve our objectives of developing a framework.

Ethics approval

17. Information about the registration status is required. Consider registering this research proposal in a database compliant with WHO's ICTRP (www.who.int/ictip), and please consider adding the registration number in the title and/or ethics section.

We note that this is the International Clinical Trials Registry Platform. Given that this is a proposed qualitative study, it would seem inappropriate to do so. As far as we are aware, observational studies are not required to be registered with any database.

Discussion

18. The social determinants of health are a key framework that is absent and should

be considered in the discussion.

Our background section contained a very detailed description of Arlene Brown's model for understanding the impact of socioeconomic position (which includes consideration of each of the social determinants of health) on health outcomes. While we have retained cursory mention of these frameworks in the introduction to set up the need for a novel framework, we provide some more detail regarding the critiques of these frameworks in the interpretation section.

19. The practicability, usefulness and operationality of the instrument are not entirely clear. The practical reasons for developing this tool should be clearly presented together with an explanation of the applicability of the framework inside Canada and beyond Canada. The authors should elaborate on what needs to be done once the results are out, to make a difference; to what extent expected results could be translated into policy recommendations. The objective of the study should be to have an impact, and if implementation is not considered, this would be missed.

The rationale for our study is more clearly outlined in the concluding paragraph of the manuscript:

"We are optimistic that the development of a comprehensive framework to understand the experience of financial barriers for patients with chronic diseases will be educational and highly relevant for policymakers, clinicians and health services researchers. Through our framework, we hope to generate an understanding of how and why some patients come to experience financial barriers which may be useful for informing future health policy around healthcare accessibility. Furthermore, the development of a framework to understand this particularly vexing problem would also be of great value to individual clinicians who care for these patients and may not fully understand the barriers that prevent their patients from being fully adherent to their recommended medical and lifestyle therapies. Finally, we plan to utilize the findings from this study in our future research on financial barriers to help design and test interventions to minimize the impact of these barriers on patient-relevant outcomes."

20. Have the authors considered insights from consumers or patient representatives to ensure their study is grounded applicable experience? This could be something to explore and add in the methods section.

The entire proposed study will be grounded in data obtained from individual patients. As we describe, we also plan to use member checking to verify the face validity of our framework with the contributors of the framework.

Final comments

21. Taking advantage that this is a protocol, the authors should offer readers a straightforward and clear manner of submitting comments to improve their proposal. **Should this paper be published, CMAJ Open will publish the email address of the corresponding author with the article for interested parties. If the editors wish for us to include further contact details, please let us know.**

22. A deliberation on what to do and how to specifically address sticking questions once this study is done should be considered. It is a suggestion to elaborate on how the results can potentially be used to enhance health care delivery and guide policy and prevention. Please elaborate on the dissemination plans and the added value this study offers.

We find it difficult to speculate about the results of the study given that this is simply a protocol. We agree that these will be important points to consider in the ultimate presentation of our final results, but at this point we cannot describe the detailed implications of our findings given that we do not know what the final framework will entail. Again, we refer back to our concluding paragraph where we describe how we hope this study will be able to contribute to policy and clinical practice.