PEER REVIEW HISTORY

ARTICLE DETAILS

TITLE (PROVISIONAL)	Quantification and visualization methods of data-driven chronic care delivery pathways: protocol for a systematic review and content analysis
AUTHORS	Siqueira do Prado, Luiza; Allemann, Samuel; Viprey, Marie; Schott, Anne-Marie; Dediu, Dan; Dima, Alexandra

VERSION 1 - REVIEW

REVIEWER	Aslak Steinsbekk Norwegian University of Science and Technology, Norway
REVIEW RETURNED	29-Nov-2019

GENERAL COMMENTS	It was a pleasure to read the protocol. It was very well written, systematically presented and followed the guidelines and thus ensuring that all relevant information was included. Suggestion for cosmetic changes the author can consider are: Remove the methods bits under Aims and objectives Given the type of review, do not separate primary and secondary research question The third paragraph in methods explaining CDP could be moved to Type of publ... Parts of the text in the Deductive-inductive content analysis could be moved to Data analysis

REVIEWER	Victoria Tiase University of Utah, USA
REVIEW RETURNED	25-Dec-2019

GENERAL COMMENTS	I believe this is a worthy topic for review, but I question the type of review that is suggested. 1. Given this is a systematic review and is contingent on previous work to inform the question, could the introduction section include comments on previous work in this area and how the research question was out of this previous research? 2. In a systematic review, the research question is generally more precise - the current questions feel like a scoping review to me. Could you add more detail to the research question(s) that includes feasibility, appropriateness, meaningfulness and/or effectiveness? 3. Same with the inclusion/exclusion criteria - this should be more precise in a systematic review. Could you define what you mean

	by "visualization or quantification of data-driven chronic CDP"? This will also be important for replication. 4. The data analysis and synthesis could use more detail. Content analysis is generally used in scoping reviews. Will you examine the study population? Types of analysis? What statistical methods will be used? 5. I would like to see the PROSPERO registration before reviewing again. I think the feedback received from the PROSPERO registration and will answer many of my questions.

VERSION 1 – AUTHOR RESPONSE

Reviewer: 1

1. Remove the methods bits under Aims and objectives

Modification: Removed from Aims and Objectives, which became “We aim to identify and describe the methods that have been proposed to quantify and/or visualize data-driven CPDs of people living with chronic conditions. Given the complexity of their context of use, more than only reviewing technical methods, we aim to investigate how these tools have considered the three domains described above.”

2. Given the type of review, do not separate primary and secondary research question

Modification: Changed in the research questions, which became “For this end, we propose the following research questions: 1. What clinical information does the method use and how was it considered relevant? 2. What are the method’s development and implementation characteristics? 3. Which behaviours and interactions does the method aim to promote among users and how?”

3. The third paragraph in methods explaining CDP could be moved to Type of publ...

Modification: The paragraph was moved to Type of Publications.

4. Parts of the text in the Deductive-inductive content analysis could be moved to Data analysis

Modification: The item Data extraction became Data extraction and analysis, as we agree with the reviewer that the content analysis we propose is, in the same time, a process of extracting information about these methods from the source articles and an analysis method. Data analysis and synthesis became Data synthesis.

Reviewer: 2

1. Given this is a systematic review and is contingent on previous work to inform the question, could the introduction section include comments on previous work in this area and how the research question was out of this previous research?

Modification: Indeed, our research questions have been informed by an examination of previous research in this area, some of which we cite in the introductory paragraph. We appreciate that some examples of prior work would help illustrate this point better. Therefore, in order to clarify this point, we have added the following sentences in the first paragraph of the introduction “For example, Zhang et al. have produced longitudinal trajectories using electronic health records (EHR) and cost pathways of people living with chronic kidney disease to inform patient engagement and to detect common pathways. Bettencourt-Silva et al. have reported on the development of a patient-centric database from multiple Hospital Information Systems (HIS)18 and on building data-driven pathways from routine hospital data on people living with prostate cancer to explore their potential use in biomedical

research. However, generating these informative trajectories from disparate and often incompatible data sources proves challenging".

2. In a systematic review, the research question is generally more precise - the current questions feel like a scoping review to me. Could you add more detail to the research question(s) that includes feasibility, appropriateness, meaningfulness and/or effectiveness?

Response: We thank the reviewer for this comment. We agree that our research aim is broad as it targets all chronic conditions and therefore may appear as less precise. To improve precision, we have specified three research questions each referring to one of the three domains we consider relevant for a comprehensive description of these methods: clinical, technological, behavioral. However, it is important to note that this is a methodology review and in this type of reviews the formulation of the research question(s) can be broader depending on the type of method targeted. In writing the content of this protocol, we followed the recommendations of the Cochrane Collaboration (Appendix A of the Cochrane Handbook Version 5.1.0 https://handbook-5-1.cochrane.org/appendix_a/a_5_main_text.htm), and several methodological systematic reviews published recently with comparable broad objectives, including Moreno-Conde et al. who proposed the coding scheme we are using in our review. Following to these sources, we believe the term 'systematic review' is suitable for this work, given the methodology we employ to search and select articles, extract information relevant to the research questions, and synthesize and interpret this information. However, given the broad aim and extensive content analysis we intend to perform, if it is in line with BMJ Open policy/criteria, we would propose to describe this review as "systematic review and content analysis", following the example of Moreno-Conde et al. (as we use their inductive work to base our content analysis on).

Regarding the question of feasibility, appropriateness, meaningfulness and effectiveness, we believe these terms correspond to the 'Validation' category in the Data extraction and analysis section. In this category we intend to describe how the methods were validated and if they were considered feasible in the context they were designed to be applied. To clarify this, we have added the following details to each category proposed by Moreno-Conde et al., which will be used for describing these selected methods:

"Scope definition leading to selection of the domain and selecting relevant experts: identifying the domain and expected uses of the method through the creation of a group of experts.

Analysis of the information covered in the specific domain: creation of definitions, identification of clinical scenarios, workflows, users, guidelines, literature, etc., so the method meet the requirements of clinical practice or other intended usages.

Design of the tool: detailing the set of attributes associated with the method, domain terminologies, ensuring compatibility across domains.

Definition of implementable tool specifications: description of implementable technical specification.

Validation: use of techniques to validate the method, such as peer-review validation or creation of prototype screens.

Publishing and maintenance: availability in public repositories.

Governance: description of the organization responsible for developing and maintaining the tool."

3. Same with the inclusion/exclusion criteria - this should be more precise in a systematic review. Could you define what you mean by "visualization or quantification of data-driven chronic CDP"? This will also be important for replication.

Modification: To make the exclusion criteria more precise, we added the sentence “We will exclude studies that aim only to assess healthcare utilization over a specific period as part of a single research study, for example as an outcome to evaluate health-related interventions, to describe populations or disease prevalence, or as a proxy measure of disease aggravation risk” to Types of publications/studies and eligibility criteria. In addition, the sentence “To describe the methods proposed for synthetically displaying objective measures or assessments of health status or healthcare utilization (e.g., quantifying) and graphically showing the temporal elements of these trajectories (e.g., visualizing) chronic CDP, we will assess how they addressed three domains” was included to clarify the terms ‘quantification’ and ‘visualization’. A reference was added to support the definition of chronic condition.

4. The data analysis and synthesis could use more detail. Content analysis is generally used in scoping reviews. Will you examine the study population? Types of analysis? What statistical methods will be used?

Response: We modified the text to provide more detail of the information extracted following Moreno-Conde et al, as described above. Indeed, we also extract information about the study population. However, as our aim is to describe the development and use of the methods more broadly and not applied to specific populations, we chose to not exclude studies based on study design. Thus, the selected papers could include data about a clinical population or could describe only the methods without practical application to a clinical area, therefore we might not be able to examine study population for all the methods. To be able to synthesize the findings considering the multiple formats, we will present the information extracted/coded in descriptive tables, also describing outcomes if studies measure and report them. We will summarize these results via descriptive statistics where appropriate, and provide a narrative summary and interpretation in light of current developments in health technology for chronic care.

5. I would like to see the PROSPERO registration before reviewing again. I think the feedback received from the PROSPERO registration and will answer many of my questions.

Response: The PROSPERO registration has been concluded on November 14th 2019 (CRD42019140494) without feedback, which we believe supports the fact that our review meets their criteria for systematic reviews (their policy not to pre-register scoping reviews). Minor modifications have been made to the protocol since then as the study advances, and new ones will be made to align the PROSPERO record with this manuscript following the comments and suggestions of the reviewers.

VERSION 2 – REVIEW

REVIEWER	Victoria Tiase United States
REVIEW RETURNED	30-Jan-2020

GENERAL COMMENTS	This is a worthwhile topic of study. However, I wonder if this topic lends itself to scoping vs. systematic. Regardless, I have a few minor comments:

	1. When discussing the use of Kappa for agreement, it would be helpful to include the criteria that you will use to measure adequate agreement. 2. In the study screening process, if you plan to take action when no full text is available, it should be included in the methods. 3. I could not find the timeframe in which this will be conducted. It will also be important to note when the search strategy was created and consequently implemented (the latter for the final review paper).

VERSION 2 – AUTHOR RESPONSE

Reviewer: 2

1. When discussing the use of Kappa for agreement, it would be helpful to include the criteria that you will use to measure adequate agreement.

Response: We thank the reviewer for the comment. We believe reporting inter-rater reliability during the selection process is important and we consider values greater than 0.80 (almost perfect to perfect agreement) to be adequate values. We also have decided to report Kappa during the data extraction development process, when a subset of 10% of selected full texts will be assessed by two investigators.

Modification: The sentence “Inter-rater reliability (Cohen’s Kappa) between primary and secondary reviewers will be computed and reported, values greater than 0.80 will be considered adequate” was modified in the Screening section in Methods. Also, the sentence “Inter-rater reliability (Cohen’s Kappa) will be computed, and values greater than 0.80 will be considered adequate.” was added in the last paragraph of the Deductive-Inductive content analysis subsection.

2. In the study screening process, if you plan to take action when no full text is available, it should be included in the methods.

Response: For the missing full texts, we will contact the corresponding authors by email. In the absence of a reply in a week, we plan to re-send the request. If after two weeks full text is still unavailable, the record will be excluded. We did not consider necessary to go into detail about this procedure in the manuscript, but we specified the full text non-availability as an exclusion criterion.

Modification: Changed in Methods “We will also exclude studies that do not mention population or data characteristics or do not state they analyze data from people living with chronic conditions, unavailable full texts, papers not written in English, conference abstracts or abstract-only papers, systematic or narrative reviews, meta-analyses and grey literature..”

3. I could not find the timeframe in which this will be conducted. It will also be important to note when the search strategy was created and consequently implemented (the latter for the final review paper).

Modification: We now added this information in Ethics and dissemination: “The search strategy was developed in collaboration with health sciences librarian services in early 2019. Database searches will be initiated in May 2019. The review is expected to be completed by February 2020.”