

## PEER REVIEW HISTORY

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### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	Patient-driven innovations reported in peer-reviewed journals: a scoping review
<b>AUTHORS</b>	Reinius, Maria; Mazzocato, Pamela; Riggare, Sara; Bylund, Ami; Jansson, Hanna; Øvretveit, John; Savage, Carl; Wannheden, Carolina; Hasson, Henna

### VERSION 1 – REVIEW

<b>REVIEWER</b>	Ashcroft, Rachelle University of Toronto
<b>REVIEW RETURNED</b>	11-Jul-2021

<b>GENERAL COMMENTS</b>	<p>-Background is strong, good rationale. -Definition of patient innovation = patient initiated modification or development of treatment</p> <p>Methods -Performed a concept analysis within a workshop setting which is a strength for such a broad concept as innovation -What is the rationale for the years selected? 2008-2020 -Search strategy was developed in consultation with librarians which is an asset -Screening conducted by authors and research assistants. Specify how many &amp; if possible, include the initials of those who conducted screening -Selection of sources of evidence: Does not stipulate if abstract screen or if full-text review. -Full text review for those that had conflicts. Does not stipulate if all screened at abstract phase went to full text review prior to data charting. -Strength that there was patient involvement, and patient partner included as a co-author</p> <p>-Findings -Review reference list as a number of references missing publication year, and inconsistent formatting</p> <p>-Appendix 3.1, column Publication Type: There are inconsistencies in how the publication is reported. There are publications with “?” which suggests that this table is a draft and not the final version. This needs to be corrected.</p>
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<b>REVIEWER</b>	Lewis, Dana OpenAPS, Seattle
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	No financial competing or conflict of interest exists. However, the reviewer is an author on some of the research referenced or described within this paper.
<b>REVIEW RETURNED</b>	10-Sep-2021

<b>GENERAL COMMENTS</b>	<p>Thanks for writing this paper on the interesting but not well-understood topic of research on patient innovations. It's intriguing to quantify the amount of patient innovations in published research, which this paper sought to do.</p> <p>Suggestions for minor edits to improve reader understanding:</p> <ol style="list-style-type: none"> <li>1. In the abstract and in discussion, this sentence "Among the 20 innovations reported, "DIYAPS" and "PatientsLikeMe" made up half of the articles." assumes reader understanding of what DIYAPS and PatientsLikeMe is. It would be useful to articulate at least once what the acronym DIYAPS stands for (i.e. do-it-yourself artificial pancreas system) and which field it's attached to - diabetes, since that's quantified as one of the largest categories of innovations. Ditto for explaining PatientsLikeMe in context of the research and whether it's tied to any of the larger categories quantified. Do the authors have any insight into why these two categories dominated?</li> <li>2. There did not appear to be a section on limitations. I would suggest adding a subheading and a brief paragraph on study limitations which are briefly alluded to in the discussion but helpful to separate. One limitation worth addressing in greater detail might be that patient-driven innovations are not always labeled in the research as patient-driven innovations and as such, despite the methods used including snowball sampling, the result is likely an under-representation of research of patient driven innovation.</li> <li>3. I'm surprised to see Omnipod listed as an example under diabetes. While a parent of a person with diabetes co-founded the company Insulet, which commercialized Omnipod in later versions, it's storyline is very much a commercial product. By the argument that a parent co-founded the company or leads the company, many other diabetes commercial companies would be listed because they also have parents or people with diabetes in senior positions of the companies, but likely have less marketing story in the research publications about their technology. The two citations as explained in the appendix charts don't provide much confirmation on it being a patient-driven innovation. It might be worth revisiting this inclusion to determine whether it fits the categories or not. If authors do decide it still fits the criteria, this may be worth mentioning in the limitations as well that the methodology is therefore influenced by the amount of backstory narrative that is embedded in research publications, which may vary by innovation and not be universally used.</li> </ol> <p>Suggestion: it might be interesting to add a chart visualizing the yearly totals of the published research to show the increases over time and aggregating around the 2016-2020 timeframes described.</p> <p>Minor comments, typos, etc:</p> <p>In some cases the references have typos. Under references 42,44,45,46,52,53 one of the author's last names is somehow repeatedly truncated to "Leibr" in the citation - last name should be fully spelled out as "Leibrand". Reference 42 also has a "C" at the end of OpenAPS in the article title that is a typo. It might be worth</p>
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	<p>double-checking all of the references for additional typos - reference 85 might also have a typo.</p> <p>In Table 3, under diabetes, "Autosense" does not have an 'e'. It's shorthand is "autosens" which stands for "autosensitivity", and either of those would be fine to use. This should also be updated similarly in the appendix or anywhere else it's mentioned.</p>
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<b>REVIEWER</b>	Rana, Ruhina Douglas College, Nursing
<b>REVIEW RETURNED</b>	21-Sep-2021

<b>GENERAL COMMENTS</b>	<p>Excellent use of Arksey and O'Malley framework.</p> <p>Suggestions for minor revisions:</p> <p>While your objective is clear, you may want to frame it as a research question so that your results and discussion serve to answer the question.</p> <p>Pg 6, line 40 - Please clarify what you mean by "significant activity" as this is a bit vague.</p> <p>Pg 6, line 52 - you state there is limited knowledge regarding patient driven innovations. Perhaps you could make a statement as to why this might be so. eg: lack of resources and support, lack of opportunities, lack of skills in writing for publication, lack of access to academic journals as non-academics, etc???</p> <p>Page 7, line 11 - you state this is the most appropriate method but do not explain why until Pg 13, line 14. Consider including this rationale earlier in the paper.</p> <p>Under definition of key concepts, your definitions could be more detailed and clear. State exactly the definition of each key term.</p> <p>Pg 7, line 59 - you state this is the first review of its kind but limit your date range to 12 years. Was this term not used before 2008? If it was, consider expanding your range to be more complete.</p> <p>Pg 9, line 16 - please describe more specifically how disagreements were resolved. By consensus, consent, majority?</p> <p>Discussion section feels repetitive from results. Include more about why these results may have been found. In this framework, results are collated and summarized however information about barriers or challenges to either innovating or publishing would have better informed the review. You mention that these were not systematically explored, however narrative accounts would have been helpful to inform this question.</p> <p>Thank you for the opportunity to review your manuscript.</p>
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## VERSION 1 – AUTHOR RESPONSE

Reviewer 1 comments:

What is the rationale for the years selected? 2008-2020

Authors response:

One of our earlier searches found few reports or publications about patient innovations before 2008, and we assumed any key publications would be referenced in reviews and primary studies since 2007. However we noted this as one of the limitations of our review in the discussion. We added in the methods the following justification for limiting the systematic search and selection to after 2007

"We limited the review to reports and publications made from 2008 because our initial searches found few reports or publications about patient innovations before 2008, and we assumed any key publications would be referenced in reviews and primary studies since 2007"

Reviewer 1 comments:

-Screening conducted by authors and research assistants. Specify how many & if possible, include the initials of those who conducted screening

Authors response: we added the following specification with initials of researchers who conducted screening as follows:

"Records were screened by six authors (MR, AB, HJ, SR, HH, CW) and two research assistants (see acknowledgements) in the open-source software Rayyan (16), according to eligibility criteria."

Reviewer 1 comments:

-Selection of sources of evidence: Does not stipulate if abstract screen or if full-text review.

Authors responses we clarified this section as follows:

"To screen the large number identified at this stage we first collected and applied the selection criteria to titles and abstracts of the papers discovered in the search. If abstracts were not available we retrieved the full paper to decide if the selection criteria were met or not met to carry forward to the next stage of review."

Reviewer 1 comments:

-Full text review for those that had conflicts. Does not stipulate if all screened at abstract phase went to full text review prior to data charting.

Authors response: In cases where reviewers agreed on exclusion in the abstract phase articles were not moved forward to fulltext screening.

Reviewer 1 comments:

-Review reference list as a number of references missing publication year, and inconsistent formatting

Authors response: we undertook a thorough checking of citations and corrected formatting where necessary to follow the journals citation guidance.

Reviewer 1 comments:

-Appendix 3.1, column Publication Type: There are inconsistencies in how the publication is reported. There are publications with "?" which suggests that this table is a draft and not the final version. This needs to be corrected.

Authors response: we made corrections to the appendix table, which is now available as supplementary data in the Zenode repository.

Reviewer 2 comments:

Reviewer 2 comments:

In the abstract and in discussion, this sentence "Among the 20 innovations reported, "DIYAPS" and "PatientsLikeMe" made up half of the articles." assumes reader understanding of what DIYAPS and PatientsLikeMe is.

It would be useful to articulate at least once what the acronym DIYAPS stands for (i.e. do-it-yourself artificial pancreas system) and which field it's attached to - diabetes, since that's quantified as one of the largest categories of innovations. Ditto for explaining PatientsLikeMe in context of the research and whether it's tied to any of the larger categories quantified. Do the authors have any insight into why these two categories dominated?

Authors response: Thank you for good suggestions. We added a short sentence in the results about both these innovations as follows,

Among the 20 innovations reported, the Do-It-Yourself Artificial Pancreas System (DIYAPS) and PatientsLikeMe.com, an online health-related social network made up half of the articles.

The review did not give any insight into why these two innovations dominated. We recommend further studies that may explore patient innovators who publish in peer-reviewed journals reason around research collaboration and publishing.

Reviewer 2 comments:

2. There did not appear to be a section on limitations. I would suggest adding a subheading and a brief paragraph on study limitations which are briefly alluded to in the discussion but helpful to separate. One limitation worth addressing in greater detail might be that patient-driven innovations are not always labeled in the research as patient-driven innovations and as such, despite the methods used including snowball sampling, the result is likely an under-representation of research of patient driven innovation.

Authors response: We agree that the earlier version of the manuscript lacked a section on limitations. We elaborated the section on the strengths of the study to also include limitations, as suggested, and this section now reads:

"The strengths of the study include the broad scope of the review. We followed the process outlined in Arksey and O'Malley (13) and the review was guided by a predetermined strategy for data collection and analysis. Methodological strengths lie in this systematic approach to searching the four large databases, complemented by snowball sampling. Earlier research has reviewed specific patient-driven innovations, e.g., a review of "Do-It-Yourself Artificial Pancreas Systems, DIYAPS" by Kesavadev, et.al. (115), or investigated characteristics of patient innovators (7). This is the first review undertaken of patient-driven innovations, according to a broad and comprehensive definition, and one of the very few undertaken of innovations in which patients have played a significant role in development of the innovation. This builds upon Oliveira and colleagues' definition of patient innovations where innovation is limited to "a treatment, a technical aid product, or a medical device" (7). We co-created a broader definition together with patient innovators in order to include social innovations such as collaborative or social networks. Thus, this broader scope and definition of patient-driven innovations was able to capture more innovations in which patients have played a significant role in the development.

The limitations of the study include the choice to only select peer-reviewed articles, but this was motivated by our aim to explore the proliferation of patient-driven innovations within the scientific literature. If the purpose had been to create an inventory of patient-driven innovations, the inclusion of grey literature would have yielded more results. Furthermore, the source of information was restricted to the included articles and in some cases the webpage [www.patient-innovation.com](http://www.patient-innovation.com). Patient-driven innovations are not always labeled as such in the publications. Therefore, despite the broader definition and the use of snowball sampling and online searches to identify the drivers of innovations, our results are likely an under-representation of research on patient-driven innovations. Also, as patient-driven innovations may initially go through commercialization processes with a shift of "drivers", it is possible that we would relabel some innovations as not being patient-driven if we had access to more information."

Reviewer 2 comments:

3. I'm surprised to see Omnipod listed as an example under diabetes. While a parent of a person with diabetes co-founded the company Insulet, which commercialized Omnipod in later versions, it's storyline is very much a commercial product. By the argument that a parent co-founded the company or leads the company, many other diabetes commercial companies would be listed because they also have parents or people with diabetes in senior positions of the companies, but likely have less marketing story in the research publications about their technology. The two citations as explained in the appendix charts don't provide much confirmation on it being a patient-driven innovation. It might be worth revisiting this inclusion to determine whether it fits the categories or not. If authors do decide it still fits the criteria, this may be worth mentioning in the limitations as well that the methodology is therefore influenced by the amount of backstory narrative that is embedded in research publications, which may vary by innovation and not be universally used.

Authors response: Thank you for highlighting this. We totally agree that commercial companies' marketing stories can impact whether an innovation is perceived as patient-driven. We address the difficulty in judging if an innovation is patient-driven or not in the limitation section. The articles related to Omnipod (Lebental, 2011 and Zisser 2011) were included since Omnipod was listed on [www.patient-innovation.com](http://www.patient-innovation.com) and we assumed from the description on that webpage and from this webpage: Expert Opinion: John L. Brooks III, Healthcare Capital LLC ([transcendit.health](http://transcendit.health)) that John Brooks had been, to at least some extent, involved in initiating and driving development of the Omnipod.

Reviewer 2 comments:

Suggestion: it might be interesting to add a chart visualizing the yearly totals of the published research to show the increases over time and aggregating around the 2016-2020 timeframes described.

Authors response: We struggled regarding how to present the data and at the end chose not to follow this suggestion. This information is visualized in Figure 1., which shows number of publications per year.

Reviewer 2 comments:

Minor comments, typos, etc:

In some cases the references have typos. Under references 42,44,45,46,52,53 one of the author's last names is somehow repeatedly truncated to "Leibr" in the citation - last name should be fully spelled out as "Leibrand".

Reference 42 also has a "C" at the end of OpenAPS in the article title that is a typo. It might be worth double-checking all of the references for additional typos

- reference 85 might also have a typo.

Authors response: Thank you for noticing and pointing out these typos and errors. We made the corrections to these references as shown in the new manuscript and track changes manuscript.

Reviewer 2 comments:

In Table 3, under diabetes, "Autosense" does not have an 'e'. It's shorthand is "autosens" which stands for "autosensitivity", and either of those would be fine to use. This should also be updated similarly in the appendix or anywhere else it's mentioned.

Authors response: we followed the reviewers suggestion and changed "autosense" to "Autosens" in table 3 and the Appendix.



Reviewer 3 comments:

Reviewer 3 comments: While your objective is clear, you may want to frame it as a research question so that your results and discussion serve to answer the question.

Authors response: following the reviewer's suggestion we changed the objective to a question as follows.

"The objective of the review was to answer the question: what is the nature and extent of patient-driven innovations published in in peer-reviewed scientific journals"

Reviewer 3 comments:

Pg 6, line 40 - Please clarify what you mean by "significant activity" as this is a bit vague.

Authors response: We revised the text to more clearly present the number of patient-driven innovations detected and listed by patients-innovation.com.

"The patient innovation website, created to collect innovations by patients and/or family caregivers ([www.patient-innovation.com](http://www.patient-innovation.com)), lists over 1.200 innovations which gives one indication of significant activity of patients and their family caregivers driving health innovations, often independently of the health system."

Reviewer 3 comments:

Pg 6, line 52 - you state there is limited knowledge regarding patient driven innovations. Perhaps you could make a statement as to why this might be so. eg: lack of resources and support, lack of opportunities, lack of skills in writing for publication, lack of access to academic journals as non-academics, etc???

Authors response: we changed the statement in the background section to following "Although the awareness of patients' innovation capacity is increasing, there is still limited knowledge regarding the extent and nature of patient-driven innovations in the peer-reviewed literature (7, 12). In the discussion section of this paper we consider possible explanations for this".

In the discussion section we added:

Although there are over 1.200 posts about patient-driven on [www.patienter-innovation.com](http://www.patienter-innovation.com) we found a limited number of publications up to 2020 in peer-reviewed scientific journals that met our broad definition and selection criteria and most related to two innovations. Possible explanations for these findings are that patient innovators lack resources and support related to research and academic writing. Further on, there are journals considering patient engagement (for example Patient engagement and involvement), but articles in this review were almost exclusively published in medical, healthcare processes or digital health journals. It would be valuable to explore how patient innovators reason about research and publishing in general and when choosing the journal they wish to submit their research to.

Reviewer 3 comments:

Page 7, line 11 - you state this is the most appropriate method but do not explain why until Pg 13, line 14. Consider including this rationale earlier in the paper.

Authors response: we followed the suggestion to elaborate on why we chose scoping review as the most appropriate answer the research question at an earlier point and under the methods design subheading as follows,

“A scoping review method was chosen as the most appropriate for the objective of the study because our initial investigations revealed a diverse range of types of studies and publications, and the method is recommended as useful when examining emerging areas of research (114)”.

Reviewer 3 comments:

Under definition of key concepts, your definitions could be more detailed and clear. State exactly the definition of each key term.

Authors response: we made changes and rewrote this section to be clearer as follows:

“Using the concept analysis method, the team agreed to define “innovation” based on the WHO definition of health innovation as “an innovation that identifies new or improved health policies, systems, products and technologies, or services and delivery methods that improve people’s health and wellbeing. The innovation aims to add value in the form of improved efficiency, effectiveness, quality, sustainability, safety and/or affordability. The innovation can be preventive, promotive, curative, rehabilitative, assistive and/or palliative care.” The other part of the concept was “patient-driven” that we agreed consisted of two parts and was defined by 1. The innovation is user-driven, meaning that it is both initiated and driven (in development, implementation etc.) by patients and/or family caregivers and 2. The innovation responds to one or more unmet needs of the innovator. Unmet needs are defined by the innovator. This provided a definition broad enough to allow for a comprehensive review of the nature of patient-driven innovations but limited “patient-driven” to focus on the role of patients and/or family caregivers.” (see Table 1).

Reviewer 3 comments:

Pg 7, line 59 - you state this is the first review of its kind but limit your date range to 12 years. Was this term not used before 2008? If it was, consider expanding your range to be more complete.

Authors response: We clarified our statement as follows, “This is the first review undertaken of patient driven innovations, according to our definition, and one the very few undertaken of innovations in which patients have played a significant role in development of the innovation.”

Reviewer 3 comments:

Pg 9, line 16 - please describe more specifically how disagreements were resolved. By consensus, consent, majority?

Authors response: we clarified how disagreements were resolved as follows, “In 7% of the cases, researchers disagreed about inclusion/exclusion, and these conflicts were resolved by consensus through discussion among authors based on a full-text screening”

Reviewer 3 comments:

Discussion section feels repetitive from results. Include more about why these results may have been found. In this framework, results are collated and summarized however information about barriers or challenges to either innovating or publishing would have better informed the review. You mention that these were not systematically explored, however narrative accounts would have been helpful to inform this question.



# Authors response:

We agree with the reviewer that the discussion felt repetitive from results and reduced possible repetition by rewriting the discussion section. Thank you for pointing this out. We agree that barriers or challenges to innovating or publishing is important areas to explore. Information regarding this was not mentioned in the included articles and we therefor recommend further studies to explore how patient innovators reason regarding research collaboration and collaboration. In the discussion (section Unanswered questions and further research) we write:

“Considering the potential benefits that patient-driven innovations can have if they become widely used, it will be important to understand factors that may facilitate or hinder implementation, spread and scale-up of patient-driven innovations; none of the included articles in this review systematically examined these questions. It may also be important to gain deeper understanding of patient-driven innovations in general, what unmet user need they address, how they are used and by whom and what outcomes they have for patients and health care systems. A further unanswered question is what determines whether patient innovators decide to publish their results and if so, in which journal(s). Patient innovators were often listed as co-authors in publications related to their innovations, there was a broad variation in type of publication, and it was common for patient innovators to co-author articles together with established researchers and/or clinicians. Some patient innovators presented their innovations in single authored papers and others were not listed as authors. A suggestion for future studies is to interview or survey patient innovators who publish in peer-reviewed journals and explore how their reasoning around research collaboration and publishing. “

## VERSION 2 – REVIEW

<b>REVIEWER</b>	Ashcroft, Rachelle University of Toronto
<b>REVIEW RETURNED</b>	27-Nov-2021
<b>GENERAL COMMENTS</b>	Authors have responded appropriately to reviewers' comments. The Prisma Flow Chart appears to be missing, and needs to be added for publication.
<b>REVIEWER</b>	Lewis, Dana OpenAPS, Seattle  No financial competing interests, although some of my published work is referenced within the article.
<b>REVIEW RETURNED</b>	02-Dec-2021
<b>GENERAL COMMENTS</b>	Well done addressing all of the reviewer comments; I have no additional feedback as it was all addressed in the revision.