

Appendix 1

We conducted an initial exploratory search to see if there were any existing systematic reviews of patient-oriented research and we uncovered eight reviews including four that specifically related to impact^{9,13-15}. Using a snowball sampling technique, we searched references of the eight reviews then all relevant papers and located a further 36 papers and reports, which were read in detail. To ensure we did not miss any significant papers, we also searched all EBSCO databases for journal articles in the most recent 5-year period of January 2014 to December 2018 for relevant studies, with particular attention to the CINAHL, MEDLINE, PubMed databases. The database searches used the following terms in the text: "patient-oriented research" OR "patient involvement in research" OR "public involvement in research" combined with the term "impact" in the title or as a subject term (keyword), and resulted in six (7) new papers that were read in detail. We concluded that saturation was reached as the results from the new searches did not identify any new papers, or the articles we read in detail duplicated the main messages. The searches were not limited by study design or language of publication. For the narrative analysis, we used a process that involved identifying key points, amalgamating these into a central list, then thematically analysing and synthesizing the evidence.

See Supplemental Table 1 for a description of the articles.

Supplemental Table 1: Description of articles related to the impact of patient-oriented research

Title	Author(s)	Year	Description	Findings about impact
Can the impact of public involvement on research be evaluated?	Barber R, Boote JD, Parry GD, et al.	2012	Delphi study with UK and international panelists investigating whether it is feasible to evaluate the impact of public involvement on health and social research	<ul style="list-style-type: none"> • Consensus from panelists that impact of public involvement on the following is feasible: <ul style="list-style-type: none"> ○ Public involved ○ Research team ○ Disseminating research ○ Identifying and prioritizing topics for research • Most panelists did not consider it feasible to evaluate impact of public involvement on research process and outcomes, especially because of its complexity and varying conceptual frameworks, terminology and practice • Some panelists questioned appropriateness of applying scientific enquiry to a social partnership
User involvement in research (in <i>Critical perspectives on user involvement</i>)	Barnes M, Cotterell P	2012	Book chapter focused on challenges of involving users in research and of measuring the impact	<ul style="list-style-type: none"> • Researchers resist user involvement because there is a perception there is no proof that it makes a positive difference to research, that involvement is unrepresentative, costly, time consuming or too difficult. • Key challenges to measuring impact include: <ul style="list-style-type: none"> ○ Involvement is a complex process ○ Difficult to predict where involvement will have greatest impact • Difficult to identify the added value of involvement
From craft to reflective art and science	Boivin A	2018	Commentary on how we should be evaluating patient engagement	<ul style="list-style-type: none"> • Cautions that patient engagement should not be treated as a technology where effectiveness, benefits and harms can be tested in the same way as other health technologies (such as drugs or dialysis) • Evaluation of patient engagement should move from causal analysis to a contribution analysis, and use a reflective approach

<p>Patient and public engagement in research and health system decision making: a systematic review of evaluation tools</p>	<p>Boivin A, L'Espérance A, Gauvin F-P, et al.</p>	<p>2018</p>	<p>Review and assessment of evaluation tools for patient and public engagement in research and health system decision-making</p>	<ul style="list-style-type: none"> • Without adequate evaluation tools, it is difficult to ensure the integrity of engagement principles and practices, assess the outcomes of engagement and demonstrate accountability to the public. • Most evaluation tools evaluate the context and process of patient engagement, and perceived self-reported impacts • The included tools mostly assess whether patients were effectively engaged and how (and not on whether patient engagement had an impact on policy decisions or health outcomes)
<p>A systematic review of the impact of patient and public involvement on service users, researchers and communities</p>	<p>Brett J, Staniszewska S, Mockford C, et al.</p>	<p>2014</p>	<p>Review explores the impact of patient and public involvement (PPI) on service users, researchers and communities involved in health and social care research</p>	<ul style="list-style-type: none"> • Benefits to service users included feeling empowered and valued, improved confidence and self-worth, better knowledge of research, more direct access to information • Impact of PPI on researchers included gaining fresh insights into issues, better understanding of patient needs, preconceived assumptions challenged, greater respect for community being studied • Impact on community included increasing acceptability of research, increase in awareness of disease or condition, better KT, made science more accountable to community, better targeted services • Note that primary studies do not report impact well • Recommend better conceptualization of impact and development of reliable tools to measure impact
<p>Mapping the impact of patient and public involvement on health and social care research: a systematic review</p>	<p>Brett J, Staniszewska S, Mockford C, et al.</p>	<p>2012</p>	<p>Systematic review (resulting in 66 studies) to assess the impact of patient and public involvement (PPI) on health and social care research</p>	<ul style="list-style-type: none"> • Found evidence that patient and public involvement can have a positive impact on research, for example by: <ul style="list-style-type: none"> ○ Grounding research in reality of user experience ○ Adapting language to lay audience ○ Assessing appropriateness of research instruments

				<ul style="list-style-type: none"> ○ Providing a wider and more relevant viewpoint ● Data about impacts in these studies are often brief and lack detail ● Need greater consistency in reporting full range of impacts ● Lack of robust tools to measure impact ● Evidence of PPI impact is weak and needs significant enhancement
Questioning patient engagement: research scientists' perceptions of the challenges of patient engagement in a cardiovascular research network	Carroll SL, Embuldeniya G, Abelson J, et al.	2017	Assessment of researchers' perceptions of the meaning and value of patient engagement in cardiovascular research	<ul style="list-style-type: none"> ● Participants suggested that patient engagement lends research legitimacy, relevance and meaning while balancing power and leading to democratization of science ● Participants concerned about the paradox: need to retain authenticity of patients' unique experiences while also representing a global disease experience represented by population-level data ● Many participants concerned about lack of evidence of impact, which remains difficult as long as the outcomes of patient engagement were not clear ● In order to measure impact of patient engagement, there needs to be a shift away from traditionally positivistic approaches towards qualitatively reported outcomes
Is it worth it? Patient and public views on the impact of their involvement in health research and its assessment: a UK-based qualitative interview study	Crocker JC, Boylan AM, Bostock J, Locock, L	2017	A qualitative interview study with patient and public involvement (PPI) contributors to get their perspectives on the impact of their contributions on health research	<ul style="list-style-type: none"> ● Participants describe positive impacts of their own roles as persons with lived experience ● Participants describe potential impacts on research processes ● Participants questioned whether it was possible to do an impact assessment, given the complex nature of PPI and how difficult it is to isolate the impact of the PPI ● Prospective research (eg. ethnography) may uncover how PPI roles lead to impact on research
Successful stepwise development of patient research partnership: 14 Years' experience of actions and consequences in	de Wit M, Kirwan JR, Tugwell P, et al.	2017	Explored potential benefits and challenges of involving patient research partners in conferences and working group activities	<ul style="list-style-type: none"> ● Describe benefits of patient engagement to the OMERACT consensus project, including providing face validity to the process, identification of new domains, changing the culture of OMERACT, providing a "reality

outcome measures in rheumatology (OMERACT)				<p>check” of the relevance and quality of the project</p> <ul style="list-style-type: none"> • Patients were less certain about the added value of their experiential knowledge to the research • Conclude that engaging patients is “infinitely better than not involving patients at all” • Acknowledge that developing metrics to demonstrate value of patient engagement is challenging
Patient engagement in research: a systematic review	Domecq JP, Prutsky G, Elraiyah T, et al.	2014	Meta-narrative systematic review aimed to determine how best to conduct the process of patient engagement, including benefits, harms and barriers of patient engagement	<ul style="list-style-type: none"> • Although most studies claimed some benefits to patient engagement, there were no comparative data to suggest best practices • Found several studies that reported engaging patients improves patient enrollment and retention • Future research needed to demonstrate the value of patient engagement to researchers and funders • Recommend that bibliographic databases use indexing terms that identify patient engagement in research
Evaluation of public involvement in research: time for a major re-think?	Edelman N, Barron D	2015	Argument to avoid evaluating public involvement (PI) in the same way as you would therapeutic interventions	<ul style="list-style-type: none"> • Evaluating PI as a therapeutic intervention is the reason why evaluation has proved difficult and derailed the development of an evidence base • Recommend using a deontological perspective and evaluate PI as a contribution of expertise and advocacy, no different than we would for the contribution of clinicians, statisticians for example • Suggest adopting a consequentialist rationale, viewing PI as a methodology that improves research quality
Evaluating patient and stakeholder engagement in research: moving from theory to practice	Esmail L, Moore E, Rein A.	2015	Synthesis of literature on impacts of patient and stakeholder engagement and identifies gap between expectations and evidence	<ul style="list-style-type: none"> • Hypothesized impacts include better quality research, empowering patients, increased KT, democracy and accountability and moral obligation • While many papers theorize intended impacts, very few empirical studies have tested them – most often retrospective and self-reported

				<ul style="list-style-type: none"> • Need to establish an evaluative framework or set of criteria for engagement prior to execution of research • Lack of clarity or consensus on the major purpose of engagement • The field needs to develop more conceptual guidance and consensus to drive evaluation efforts
The prevalence of patient engagement in published trials: a systematic review	Fergusson D, Monfaredi Z, Pussegoda K, et al	2018	Review aims to assess benefits of meaningful patient engagement to research and clinical practice (included randomized controlled trials and non-randomized comparative trials)	<ul style="list-style-type: none"> • Found limited evidence of benefits and difficulties of patient engagement (and only 1 study included from Canada) • Answers to basic questions about patient engagement and patient-oriented research largely unanswered • Inconsistent and vague reporting of patient engagement research
Power to the people: to what extent has public involvement in applied health research achieved this?	Green G.	2016	Despite the efforts to promote public involvement in the UK, there is a concern that it has limited impact on research decision-making. The paper explores whether the power has indeed shifted from the scientific research community to the public	<ul style="list-style-type: none"> • Even though there is recognition of the importance of involving the public, the public voice is often not given the same weight as that of professionals • The authors are concerned that there exists tokenism and “box ticking” and that the same members of the public are being overused • There is still a way to go towards establishing a more democratic approach in health research
Searching for the impact of participation in health and health research: challenges and methods	Harris J, Cook T, Gibbs L, et al.	2018	Aims to advance methods for systematically reviewing the impact of participation in health research, and provide recommendations for improving primary research on participation in health	<ul style="list-style-type: none"> • As yet no guiding conceptual frameworks for establishing a relationship between participation and impact in health research • Primary research often contains inadequate descriptions of participation • Suggest the development of a theoretical or conceptual framework, or a logic model to illustrate relationships between participation, research design, implementation and outcomes for specific populations in a given context
Assessing the extent to which current clinical research is consistent with	Jun M, Manns B, Laupacis A, et al	2015	Scoping review (using case study) assessing whether recently completed and ongoing clinical research was	<ul style="list-style-type: none"> • Found there is a critical gap between current research in dialysis and what dialysis patients consider a high priority

patient priorities: a scoping review using a case study in patients on or nearing dialysis			consistent with priorities identified by patients, caregivers and clinicians	<ul style="list-style-type: none"> Limited involvement of patients in identification of research priorities as research proposals are still largely driven by researchers based on their own interests, the burden of the disease and commercial interests Review does not negate importance of investigator-initiated research, but rather reinforces need for patient input
Beware zombies and unicorns: toward critical patient and public involvement in health research in a neoliberal context	Madden M, Speed E	2017	Commentary about patient and public involvement (PPI) that provides a critical view of the value of PPI	<ul style="list-style-type: none"> Lack of consensus about what effective PPI should look like Many localized case studies are examining PPI but little conclusive evidence on how best to implement No guarantee that participation improves outcomes PPI remains conceptually and theoretically vague “Formal PPI can be seen as a ghastly composite of a zombie policy that continually pops up, offering (but never providing) a solution to purported deficits in democratic engagement”
Patient and public involvement and research impact: a reciprocal relationship	McKenna H	2015	Author’s perspective on how patient and public involvement (PPI) can lead to better research impact. Includes descriptions of four studies that illustrate how involvement of patients has led to greater research impact (no assessment methods are described)	<ul style="list-style-type: none"> The impacts described include: <ul style="list-style-type: none"> Influence on the shape of the project, on development of new intervention and on identification of how to overcome barriers to implementation Provided momentum for the study, and helped shape the project plan and develop intervention Helped design the project, resolve practical issues, and recruit participants
The impact of patient and public involvement on UK NHS health care: a systematic review	Mockford C, Staniszewska S, Griffiths F, et al.	2012	Study aimed to identify impact of patient and public involvement (PPI) on UK National Health Service (NHS) healthcare services, and to examine how the impact of PPI is captured or measured	<ul style="list-style-type: none"> Dearth of research about impact of PPI No validated measurements for capturing impact of PPI Studies did not provide robust evidence of PPI impact Lack of consistency of definition of public and patient involvement No reliable measurement tool Impact is dependent on context, policy, people, resources, purpose

				of PPI and culture of organizations and individuals
Public involvement in research: making sense of the diversity	Oliver S, Liabo K, Stewart R, et al.	2015	Presents a framework for planning, designing and evaluating public involvement in research, including impact of involvement	<ul style="list-style-type: none"> Based on what authors maintain is the ultimate aim of public involvement – to have outsiders influence research – they suggest measuring impact of public involvement on people (patients, public, researchers), on research and on policy, practice and personal decisions
Should we? Could we? Measuring involvement (in <i>Critical perspectives on user involvement</i>)	Purtell r, Rickard W, Wyatt K	2012	Book chapter focused on merit of involving users/patients/public within health and social care (not specific to research)	<ul style="list-style-type: none"> Questions why we are concerned with determining the impact of involving users in research when we do not ask for measures of impact of the statistician or the health economist (because there is an underlying assumption that statistician and health economist expertise is valuable)
Patient-centred medicine and patient-oriented research: improving health outcomes for individual patients	Sacristán J	2013	Commentary comparing patient-centered medicine and patient-oriented research	<ul style="list-style-type: none"> Patient-oriented research should be based on identifying the best intervention for individual patients and should assign great value to observations and exceptions Patient-oriented research can help close the gap between clinical research and clinical practice
Patient and service user engagement in research: a systematic review and synthesized framework	Shippee N, Domecq Garces JP, Prutsky G, et al.	2015	Aimed to develop a framework that provides a standard structure and language for reporting and indexing patient and service user engagement (PSUE) research	<ul style="list-style-type: none"> Developed a framework that describes the 3 phases (preparatory, execution and translational) and 8 stages of PSUE in research. Note that their framework is built from disconnected and insufficiently tested or reported literature. Proper indexing will facilitate future synthesis of evidence. <ul style="list-style-type: none"> The need for a standard framework and language is evident.
Exploring perceived barriers, drivers, impacts and the need for evaluation of public involvement in health and social care research: a	Snape D, Kirkham J, Britten N, et al.	2014	Used Delphi technique to explore perceived barriers, drivers and impacts of public involvement (PI)	<ul style="list-style-type: none"> Note that there is very little high-quality research effort around assessing the impact of PI Participants acknowledged the difficulty of assessing impact with scientific methods Despite high endorsement from participants of Delphi study of the potential positive impacts of PI, there was no consensus that it

modified Delphi study				improves the quality and relevance of the research
'Is it worth doing?' Measuring the impact of patient and public involvement in research	Staley K	2015	Review article arguing that we are taking the wrong approach in trying to answer "what difference does patient and public involvement (PPI) make" and suggest an alternative approach	<ul style="list-style-type: none"> • Much of evidence of impact is anecdotal and weak • There is an expectation that empirical methods will provide a definitive answer to "is PPI in research worth doing?" but this approach does not work for PPI because impact is highly contextual and experiential • Researchers' accounts of PPI are not detailed enough, and should include: <ul style="list-style-type: none"> ○ What recommendations were made by the public patients ○ What changes were made in response ○ What outcomes were observed
Patient and public involvement in patient-reported outcome measures: evolution not revolution	Staniszewska S, Haywood KL, Brett J, Tutton L	2012	Explores potential for patient and public involvement (PPI) in development, application, evaluation and interpretation of patient-reported outcome measures (PROMs)	<ul style="list-style-type: none"> • Argue that PPI is important to ensure that all outcomes important to patients are included in models such as PROMs
Unresolved tensions in consumer engagement in CER: a US research perspective	Workman T, Maurer M, Carman K	2013	Identifies the tensions surrounding consumer engagement and makes recommendations for resolving them	<ul style="list-style-type: none"> • Tensions include: <ul style="list-style-type: none"> ○ Definition the term patient, and establishing whether they can represent larger population of patients ○ Culture of research where there is no incentive to partner with patients ○ Patient centred methods viewed as non-traditional and are less likely to receive support from scientific community • Recommendations include: <ul style="list-style-type: none"> ○ Increase training and capacity development for consumer engagement in CER ○ Build evidence base for consumer engagement in CER