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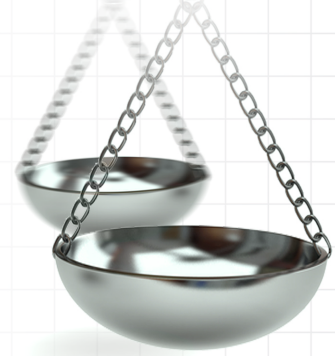

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Querying stakeholders to inform comparative effectiveness research

Yoon Duk Hong*[1], Daisuke Goto[1] & C Daniel Mullins[1]
[1]Pharmaceutical Health Services Research Department, University of Maryland School of Pharmacy, Baltimore, MD, USA
*Author for correspondence: yhon039@umaryland.edu

Despite the growing recognition of the value of stakeholder engagement in research, there is limited guidance on effectively eliciting stakeholder views during the comparative effectiveness research (CER) process. This article outlines the potential role of each stakeholder (patient, provider, policymaker and payer) throughout the CER process and provides examples of practical questions that researchers can ask the four primary stakeholder groups at each step of the CER process. This guide aims to assist in the development of meaningful stakeholder–researcher shared decision-making to incorporate stakeholder views in the design, conduct and dissemination of patient-centered CER.

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The primary goal of comparative effectiveness research (CER) is to provide stakeholders with the evidence needed to make informed healthcare decisions [1–5]. Previously, research was designed and conducted primarily by researchers, with limited stakeholder involvement. However, stakeholder engagement in CER has gained increased attention in recent years as an effective way of aligning research efforts with the needs of real-world decision-makers [6–10]. By generating CER findings with practical value, stakeholder engagement is believed to increase the acceptance of study results, and accelerate the dissemination and adoption of evidence into practice [6,9,11–14].

The creation of the Patient-Centered Outcomes Research Institute by Congress in 2010 fueled the shift in the role of patients in research, from that of mere receivers of information to active contributors to the generation of evidence [15–18]. In addition to patients, engaging other key stakeholders, such as policymakers, payers and providers has also been increasingly recognized in the literature [6,7,19–21]. However, despite the growing interest in engaging stakeholders in CER as research partners, literature has remained largely conceptual, with little practical guidance for eliciting stakeholder views.

The 10-step process for engaging patients in CER proposed by Mullins *et al.* provides suggestions for continuous patient engagement [22]. In this article, we build on this 10-step process and offer suggestions for eliciting patient and other key stakeholders' views throughout the research process. The intent of this article is to provide examples of questions that researchers may want to use to query stakeholders so that meaningful input may be obtained from each stakeholder throughout the 10-step process and to provide references for related work (see Table 1). Our proposal and sample questions are based on our experiences putting together over 40 Patient-Centered Outcomes Research proposals and our work on a dozen Patient-Centered Outcomes

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Table 1. Stakeholder roles and sample questions researchers can ask stakeholders across ten steps of comparative effectiveness research.

Stakeholder	Stakeholder roles and sample questions for stakeholders	Ref.
Topic solicitation		
Patients	Propose topics relevant to their experiences living with the illness What are your biggest concerns about treatment options? What decisions related to your medical condition would you like to have addressed through research?	[23,33,34]
Providers	Propose clinical topics where there is an evidence gap, or topics related to quality improvement Which topics have the biggest gaps in evidence that impact your clinical practice and/or the quality of care you provide to patients? Which topics, if studied, could we potentially see improvements in patient-centered outcomes?	[23,33,35]
Payers	Propose topics pertaining to financing treatment or assessing the value of treatment Which disease states or treatments are placing the biggest burden on constrained budgets? Which research topics do you think would help inform more efficient use of resources?	[23,35]
Policymakers	Propose topics of public health significance What are the biggest public health concerns locally/nationally?	[23,35]
Prioritization		
Patients	Prioritize topics that may improve patients' quality of life and/or reduce the burden of disease Which topics do you think will have the biggest impact on your/other patients' decisions to improve health-related quality of life or minimize disease-related burden?	[36–39]
Providers	Prioritize topics that impact how they treat patients and/or those that can lead to improved care delivery Can you rank each of the following topics in the order they should be addressed, taking into consideration urgency in terms of the need for evidence, improved clinical decision-making and/or improved care delivery?	[35,36,39,40]
Payers	Prioritize topics that may affect reimbursement decisions, and/or lead to formulary change Which of the following research topics do you think are most likely to have an impact on reimbursement decisions and/or lead to formulary change?	[35,38,39]
Policymakers	Prioritize topics that are more likely to have an impact on priority populations and lead to policy change Which topics would you say are most timely and relevant, most likely to have an impact on priority populations and potentially lead to policy change?	[35,38,39]
Framing the question		
Patients	Evaluate if the question can be applied to improve their health and their quality of life Do you think answers to this research question will help you and other patients manage or treat your conditions better and ultimately lead to better quality of life? If you were to change components of the research question, how would you change them?	[41]
Providers	Evaluate if the question is relevant to current practice Is this question relevant to current treatment patterns and clinical issues of importance? Is this question clinically appropriate?	[35]
Payers	Evaluate if the question can be used to provide a meaningful solution to an issue of priority Would this question provide a meaningful solution to an issue of priority within your organization or to other payers? Is this research question framed in a manner that would help you identify and target factors contributing to poor beneficiary health outcomes/high costs?	[35]
Policymakers	Ensure that the research question is relevant to current issues of importance, and useful for making health policy recommendations Is this question relevant to current public health issues of importance and would it be useful for making policy recommendations? If not, how should it be framed differently?	[35]

CER: Comparative effectiveness research.

Table 1. Stakeholder roles and sample questions researchers can ask stakeholders across ten steps of comparative effectiveness research (cont.).

Stakeholder	Stakeholder roles and sample questions for stakeholders	Ref.
Selection of comparators and outcomes		
Patients	Propose outcomes that are of highest priority to them and their quality of life, and suggest the most appropriate way of measuring those outcomes Which outcomes are most important to you? Which outcomes have the biggest impact on your quality of life?	[41–46]
Providers	Propose comparators based on current treatment patterns, and propose the most appropriate outcomes for decision-making What is the most appropriate comparator for drug X in today's treatment landscape? Which outcomes would best measure the effectiveness of treatment X? Which outcomes will influence clinical decision-making?	[9,43,47]
Payers	Propose comparators that could lead to cost savings and alleviate budgetary burden Which outcomes should be evaluated in the study in order to influence reimbursement decisions? Which drugs used for the treatment of disease X do you think should be compared?	[9,43]
Policymakers	Propose comparators and outcomes that evaluate current health policy and direct future policy Which comparators and outcomes do you think would best assess existing policy and direct future policy?	[43]
Creation of conceptual framework		
Patients	Supplement the conceptual framework with additional factors not documented in the literature based on their personal experiences From your experience of disease X and how it has impacted your life, are there any additional factors that should be included in this conceptual framework?	[41]
Providers	Verify logic of the conceptual framework with the clinical knowledge and experience providers have Based on your clinical knowledge and experience, does the conceptual framework make sense? Does it make sense that the management of disease X is affected by the management of comorbid conditions? Does the conceptual framework provide clinical utility and value?	[35]
Payers	Provide a 'reality check' of the conceptual framework from the payers' perspective Would this conceptual framework make sense in the real-world setting (e.g., does it make sense to say that prescribing patterns are influenced by clinical evidence and reimbursement only)?	[35]
Policymakers	Take a broad, societal view of the conceptual framework Are there any laws or socioeconomic factors that may have an impact on the variables and outcomes included in the conceptual framework that you believe are important?	[35]
Analysis plan		
Patients	Assist in the selection of variables, identification of confounders and utility measurement Other than the use of medications, can you list any other factors that may affect symptoms of disease X?	[43,48]
Providers	Contribute to the process of defining the study population, exposures and outcomes How should we define the study population? If we were to evaluate outcome X, when should it be measured? What would be the best way to measure outcome X?	[9,43]
Payers	Identify subgroups of interest and ensure that these subgroups are adequately accounted for within the analysis plan Which subgroups do you think should be analyzed and how?	[9,43,49]
Policymakers	Make sure the proposed analysis plan does not compromise the generalizability of the study results to the overall population Looking at this analysis plan, do you think the results of this study could be generalized to the overall population?	[43]

CER: Comparative effectiveness research.

Table 1. Stakeholder roles and sample questions researchers can ask stakeholders across ten steps of comparative effectiveness research (cont.).

Stakeholder	Stakeholder roles and sample questions for stakeholders	Ref.
Data collection		
Patients	Suggest how data should be collected to produce believable results Do you think the questions in this survey are adequate? How could the questions be improved?	[25,41]
Providers	Assist in assessing and addressing data source adequacy, and in creating definitions of data elements Is it clinically valid to obtain information on variable X from data source Y? Which diagnosis codes should be used?	[43]
Payers	Propose the most suitable data source for the question concerned and its ability to robustly capture covariates, or suggest linkage of data from more than one source To capture variables X, Y and Z, and outcomes A and B, which data source(s) should be used?	[43]
Policymakers	Assist with the selection of data sources that best represent populations of interest To evaluate the outcomes in population X, which data source would be most appropriate?	[43]
Reviewing and interpreting results		
Patients	Assess if the results reflect their experiences living with a certain disease and suggest the implications of the results for patients Do the results reflect your experience with disease X? Do you think these study results provide value to patients with disease X? If so, how?	[41,42,50,51]
Providers	Assess believability of results based on their experiences and clinical expertise From your clinical knowledge and experience, are the results believable?	[42]
Payers	Suggest possible explanations for the results seen, considering economic factors From a business perspective/taking economic factors into consideration, can you offer additional explanations of the results?	[52]
Policymakers	Suggest potential policy-related explanations for the findings Could you suggest any policy-related explanations for the results seen?	[53]
Translation		
Patients	Make sure that the results can be easily understood by the target population Are the results easy to understand? How can the results be presented so that they are easily understood by the target patient population?	[41,42]
Providers	Assess which research finding will most likely influence decision-making Which finding do you think will most likely influence clinical decision-making? Based on these results, what conclusions would you draw, and what recommendations do you think should be made?	[42,54]
Payers	Provide input on which research finding will most likely affect reimbursement decisions Which finding stands out to you the most? Why? Which finding will most likely influence formulary decisions?	[55]
Policymakers	Help interpret results in a manner that is meaningful in terms of public policy-making Which finding do you find most meaningful in terms of addressing current public health issues? Which finding do you think can be applied to policy-making?	[55]
Dissemination		
Patients	Help tailor communication strategies to increase awareness and understanding of study findings by patients What would be the most effective communication strategies to increase awareness and understanding of study findings by patients?	[41,42,56,57]
Providers	Help formulate key messages targeted at healthcare providers Do these messages adequately highlight key clinical outcomes in a manner appropriate for healthcare providers?	[54,58]

CER: Comparative effectiveness research.

Table 1. Stakeholder roles and sample questions researchers can ask stakeholders across ten steps of comparative effectiveness research (cont.).

Stakeholder	Stakeholder roles and sample questions for stakeholders	Ref.
Payers	Suggest ways of engaging other payers, increasing awareness and assisting payers make informed decisions How would you suggest that we raise awareness? How can we help payers make informed decisions?	[55]
Policymakers	Facilitate dissemination of research findings and suggest ways of advocating for improved policies What would be the best ways of disseminating research findings, raising awareness and advocating for improved policies?	[55,58]

CER: Comparative effectiveness research.

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Planning the study

In the initial stages of the CER process, stakeholders can identify CER topics that are important to them. Patients can propose topics important to them, based on their personal experiences living with the illness as well as what they have heard from other patients [22]. Providers can help identify key topics related to their clinical practice, where there is a gap in evidence, or topics related to improving the quality of healthcare services they provide. Payers may bring a health technology assessment perspective to the table and propose topics pertaining to value assessments or the financial burden posed by costly diseases and treatments. Policymakers can help identify topics of public health significance as well as CER questions that impact priority populations. Through stakeholder engagement, all stakeholders, including patients, clinicians, payers and policymakers have a greater chance of having their questions answered, and together, the team of researchers and stakeholders can determine the most relevant and useful topic to be studied [7,23].

Once potential research topics are identified, stakeholders can engage in the process of determining which topic is most relevant and urgent, and help researchers decide on the topics that should be given priority.

In Step 3 of the research process, framing the question, stakeholders can participate in the generation of specific aims of the study and specific questions that the study will answer, while researchers can determine which information is already available and assess what new information this study will add to the field. The key role that stakeholders play in this step is ensuring that the research question is framed in a manner which will provide useful information that each stakeholder can use [22].

Once the research question is appropriately framed, stakeholders can engage in the process of determining patient-centered outcomes and comparators. Researchers can inquire about outcomes that are of greatest interest to stakeholders, and together, select the most relevant comparators which will have an impact on decision-making. Patients can bring a unique perspective and suggest comparators and outcomes stemming from their personal experiences that will likely have the biggest impact on patients' quality of lives. Providers may suggest comparators based on current treatment patterns, whereas payers may propose comparators that could potentially lead to cost savings while optimizing patient outcomes. Policymakers may provide input by proposing comparators and outcomes that can be used to evaluate current policy and direct future policy. The range of perspectives provided by stakeholders can enrich the comparator and outcome selection process.

Stakeholders can also play an important role in determining and refining the conceptual framework, by providing input on the formulation of hypotheses and supplementing the framework with additional factors based on their own personal, clinical, regulatory and payer experiences [24].

Conducting the study

In the development of the analysis plan, stakeholders can provide valuable input on aspects such as the most appropriate outcome measures and time frame that should be used. Stakeholders can help ensure that the analysis plan adequately accounts for subgroups of interest, while ensuring that the proposed analysis plan does not compromise the generalizability of the study results to the overall population.

At the stage of data collection, stakeholders can suggest the most appropriate data source(s) for capturing variables and populations of interest and propose ways of collecting data that can produce believable results. This may involve participating in defining data elements, or reviewing questionnaires that will be used for data collection [25].

Once the data have been collected and analyzed, and the results become available, these results can be presented to the stakeholders for their review and interpretation. Stakeholders' roles may include, but not be limited to assessing the believability of results and providing possible explanations for the results seen, which may be from a cost perspective in the case of payers, or based on clinical expertise with providers. Patients can assess if the results reflect their experiences living with the medical condition and describe the implications of the results for patient quality of life [24]. Researchers may help stakeholders as necessary in their review of the study findings; however, it is important for researchers to allow stakeholders to develop their own interpretations freely.

Stakeholders can assess if the conclusions drawn make sense, and work together with researchers to translate the results into a comprehensible message for dissemination that is appropriate for the target population. For instance, the use of medical jargon would be appropriate if healthcare providers were the target audience, but not appropriate for the patient population.

Disseminating the study results

Stakeholders can suggest venues and means for disseminating results and recommendations for various audiences. Stakeholders can help tailor communication strategies to increase awareness and understanding of study findings by target audiences [22,24]. For example, key messages may be conveyed via local organizations, news outlets or professional, or lay publications depending on the target audience. Providers can help formulate messages for healthcare providers, whereas patients can help develop messages for the lay public and suggest potential avenues for dissemination. Also, patients, providers, payers, and policymakers can all provide input on how to encourage the uptake of the evidence generated, which is our ultimate goal.

Discussion

Stakeholder engagement in CER enhances the applicability of research in providing practical information that improves patients' decision-making and maneuver through healthcare delivery systems.

Each stakeholder has unique views to contribute to the research process. Patients are the end receivers of healthcare. The 10-step approach identifies and incorporates patients' views that are based on their personal experiences of the illnesses and healthcare interventions. Furthermore, engaging diverse patient groups would help produce CER results that are applicable to specific patient subgroups, and generate findings that go beyond 'average treatment effects [26]'. Providers are also encouraged at each step of engagement to bring their unique perspectives based on their professional expertise and experience as practitioners. The 10-step approach further affords payers an opportunity to bring their financial concerns and interests to the table so that they can be equipped with the ability to make sound financial decisions that are mutually beneficial to all stakeholders. The richness of the perspectives and ideas that stakeholders bring to the table in each of the ten steps proves the importance of partnering with stakeholders in CER so that researchers may incorporate diverse views and ultimately provide value to all stakeholders involved.

Previous research has shown that providers, policymakers, and payers are not as frequently engaged in research relative to patients, and that stakeholder engagement occurs much less frequently at later stages of the research process [6]. It may be challenging for researchers to effectively engage and elicit all stakeholder groups' views throughout the entire research process. However, there are meaningful roles for all stakeholder groups to play throughout a research study and value they can add. With a clearer understanding of the value and perspectives each stakeholder is able to bring, researchers may be able to more easily create questions they can ask stakeholders that ultimately lead to more meaningful engagement.

Conclusion

The value of incorporating stakeholder views in CER is well described, but there is limited practical guidance on how specifically to elicit stakeholder views. We have provided practical suggestions on how to elicit stakeholder views throughout the research process by outlining potential stakeholder roles and providing examples of questions that researchers may ask in order to enable impactful collaboration between stakeholders and researchers. It is our hope that the guidance we have provided will assist researchers initiate meaningful engagement of stakeholders in CER.

Future perspective

With the growing interest in research that engages stakeholders as research partners [27], it is our hope that the next 10 years will see a shift toward stakeholders being readily engaged throughout the entire CER process. The need for evaluation of stakeholder engagement in research has been identified in the literature [28–32]. Moving forward, there will likely be a growth in research efforts aimed at evaluating the outcomes associated with stakeholder engagement in CER, which will further inform and support stakeholder engagement by identifying the most effective modes of engagement.

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Executive summary

Background

- The value of engaging stakeholders in comparative effectiveness research has been increasingly recognized in the literature but there is limited practical guidance on how specifically to elicit stakeholder views during the research process.
- This article outlines the potential role of each key stakeholder (patient, provider, payer and policymaker) and offers suggestions for eliciting stakeholders' views throughout the entire research process.

Planning the study

- Stakeholders provide input on topic solicitation, prioritization, framing the question, selection of comparators and outcomes, and creation of the conceptual framework.
- Questions for stakeholders should be geared toward understanding each of their needs and how this research project can be designed to fulfill those needs.

Conducting the study

- Stakeholders assist with the development of the analysis plan, data collection as well as the review and interpretation of results.

Disseminating the study results

- Stakeholders participate in the process of translating and disseminating the study results, collaborating with researchers to translate the study findings into a comprehensible message for dissemination that is appropriate for the target population.

Discussion

- Stakeholder engagement in comparative effectiveness research enhances the applicability and utility of research findings.
- Each stakeholder has unique qualities to contribute to the research process and should be incorporated into research.
- This guide provides practical suggestions for eliciting stakeholder views and a clear framework outlining the role of each stakeholder at each step of the research process.

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