Methods for Involving Patients in Topic Generation for Patient-Centered Comparative Effectiveness Research
–
An International Perspective

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ABBREVIATIONS

ACSQHC  Australian Commission on Safety and Quality in Health Care
AQUAMED  German Agency for Quality in Medicine (Ärztlches Zentrum für Qualität in der Medizin)
CAQDAS  computer-assisted quantitative data analysis
CER  comparative effectiveness research
CIHR  Canadian Institutes of Health Research
ECRIN  European Clinical Research Infrastructure Network
EFGCP  European Forum for Good Clinical Practice
EFSA  European Food Safety Authority
EGAN  European Genetic Alliances Network
EMA  European Medicines Agency
ENPCR  European Network of Patients Partnering in Clinical Research
EPF  European Patients’ Forum
EUNETHTA  European Network for Health Technology Assessment
EUPATI  European Patients Academy on Therapeutic Innovation
EURODIS  European Organization for Rare Diseases
IAPO  International Alliance of Patients’ Organizations
ICPHR  International Collaboration on Participatory Health Research
IOM  Institute of Medicine
NHS  National Health Service (United Kingdom)
NIHR  National Institute for Health Research
PCOR  patient-centered outcomes research
QUAL  research using qualitative methods
QUANT  research using quantitative, empirical methods
SCH  Scottish Health Services
VSOP  Dutch Genetic Alliance
WHO  World Health Organization

GLOSSARY

Comparative effectiveness research (CER): CER is used to describe clinical research and systematic CER reviews that compare the clinical effectiveness and safety of a treatment with at least one more alternative treatment, with the goal of determining which treatment provides the best clinical net benefit.

Culture: There are many different definitions of culture. For the purpose of this white paper, the definition of culture by the National Center for Cultural Competence (NCCC) is used, which defines culture as “an integrated pattern of human behavior, which includes but is not limited to—thought, communication, languages, beliefs, values, practices, customs, courtesies, rituals, manners of interacting, roles, relationships and expected behaviors of a racial, ethnic, religious, social or political group; the ability to transmit the above to succeeding generations; dynamic in nature” (NCCC, 2006).

Cultural and linguistic competence: This white paper uses the term cultural and linguistic competence as defined by the U.S. Department of Health and Human Services, Office of Minority Health (2001): “Cultural and linguistic competence is a set of congruent behaviors, attitudes, and policies that come together in a system, agency, or among professionals that enables effective work in cross-cultural situations. ‘Culture’ refers to integrated patterns of human behavior that include the language, thoughts, communications, actions, customs, beliefs, values, and institutions of racial, ethnic, religious, or social groups. ‘Competence’ implies having the capacity to function effectively as an individual and an organization within the context of the cultural beliefs,
behaviors, and needs presented by consumers and their communities” (OMH, 2001; based on Cross et al., 1989).

**Health disparities:** Health disparities refer to any population-specific differences in the incidence or prevalence of disease, health outcomes, or access to healthcare.

**Health experience research:** Health experience research is a type of social science research that uses primarily qualitative methods for eliciting in-depth experiential knowledge of what it is like to live with a health condition. This research aims to provide conceptual frameworks for the patient experience that can then also be used to define research areas or topics.

**Interpretivism (antipositivism):** Interpretivism is a philosophical approach in the social sciences. The premise of interpretivism is that people construct the social world and that the social world can only be understood through the analysis of people’s values and relationships. Because of this, interpretive sociology also recognizes that there may be different social realities and that people who live in different social constructs experience life differently. Interpretive sociology uses mostly qualitative methods.

**Method:** Method describes all instruments, tools, or processes that can be used to elicit the public’s voice in healthcare research.

**Nominal group technique (NGT):** NGT is a moderated group discussion using a format that encourages participation of all group members and, thus, prevents one person or one group from dominating the discussion (CDC, 2006). NGT was originally developed in the United States and has since diffused internationally (Delbecq and Van de Ven, 1971). This technique may be particularly useful if the discussion includes different stakeholders where one stakeholder may have (or be perceived as having) more power than others. NGT is divided into four phases:

- Phase 1: All individual group members write down their ideas silently
- Phase 2: The ideas from all group members are recorded
- Phase 3: All ideas are discussed
- Phase 4: The group votes and reaches consensus

**Patient-centered outcomes research (PCOR):** PCOR helps people in making informed healthcare decisions and allows their voices to be heard in assessing the value of healthcare options. This kind of research answers patient-focused questions such as the following:

- “Given my personal characteristics, conditions, and preferences, what should I expect will happen to me?”
- “What are my options, and what are the benefits and harms of those options?”
- “What can I do to improve the outcomes that are most important to me?”
- “How can the healthcare system improve my chances of achieving the outcomes I prefer?”

**Positivism:** Positivism is a science philosophical approach and system. The premise of positivism is that the social world, similar to the natural world, follows regular patterns that can be measured and understood through logical analysis. Positivism relies mostly on empirical (quantitative) methods.

**Public:** In this white paper, the term public is used to describe patients, their relatives and caregivers, patient advocates, and representatives of patient or consumer organizations, or any persons who are using or may be planning to use healthcare services and who are not healthcare researchers or healthcare service, policy, or industry professionals.
**Reflexivity:** The term *reflexivity* describes the belief that, in studying the social world, the researcher cannot maintain perfect objectivity as he or she is part of the social world and his or her own values and worldviews may impact the result of the engagement.

**Triangulation:** The term *triangulation* describes the combined use of alternate research approaches, methods, or data to address a specific research question. Triangulation is used to increase the validity and certainty about the elicited information in qualitative social science research in which the researcher’s own values and worldview could potentially impact the results (see *Reflexivity*).
ABSTRACT

Background: Engaging the public in healthcare research makes research more accountable and transparent, provides new insights that could lead to innovative discoveries, and generates research that is more relevant to patients. The international experience with engaging the public in research has shown that involving the public early in the design of studies, ideally as early as at the planning stage, leads to better results.

Objective: The objective of this white paper is to provide PCORI with an evidence-based review of best practices, processes, and methods for patient engagement that have been studied and implemented internationally, and that could be adapted and used to promote patient involvement in research topic generation in the United States.

Methods: Research for this white paper was based on a search of the peer-reviewed scientific literature using the MEDLINE, Embase, and PsycINFO databases. Additional information was obtained from the websites of international not-for-profit organizations, healthcare centers, and government bodies involved in patient-centered medicine.

Results: The search of the peer-reviewed literature identified several strategies, methods, and processes in which the public participates in the generation and selection of research topics. These can be broadly divided into four types of engagement: (1) minimal engagement, such as providing information to the public, or engagements requiring that the public members adapt to established researcher-controlled processes; (2) consultations, in which an individual or group provides experiential knowledge; (3) collaborations, in which public members work together with clinicians and researchers in active partnerships; and (4) publicly controlled research, in which the public is the driving force of research, either as active researchers or sponsors, promoting research that, as they think, otherwise would not be done. Patient advocate groups recommend higher levels of engagements and consider minimal levels of engagements to be “tokenism.”

Eliciting the public’s experiential knowledge, views, perspectives, and preferences employs primarily qualitative social science strategies and methods. The scientific strategy sets the framework of engagement and clarifies the researcher’s perspective regarding the type and extent of information that is elicited and the interpretation of the results. Thus, the use of an appropriate social science theoretical framework, and being aware of and transparent about its implications for the engagement and research, are as important as the specific research methods that are being used. The most frequently used methods for eliciting experiential knowledge are one-on-one interviews, focus group interviews, and observation, most commonly using phenomenological, ethnographic, and grounded therapy strategies. The best-researched public-clinician partnership approach for research topic generation is a process developed by the James Lind Alliance (JLA). This process is a collaboration in which the public and clinicians consult with their peers (using a method of their choice) to make sure the process covers diverse views; public members are empowered by the use of nominal group technique (NGT) to moderate the discussion. The process also incorporates scientific research data from systematic reviews to identify evidence gaps and avoid duplication of research.

Conclusions: In a process of engaging the public in generating PCOR research topics, researchers should actively reach out to the public. Public engagements should use a theory-based approach, researchers and moderators should be skilled in using and interpreting social science methods, and the engagement process should use more than one method to elicit sufficiently diverse views and appropriate detail. Combining methods for consultation with a process of collaboration has shown the best results. The use of data from health experience research is encouraged to provide sufficient detail of the patient experience, and should be combined with systematic CER reviews to identify research gaps and to avoid the duplication of research; a health disparities analysis should be conducted to ensure that the needs of underserved populations are
addressed. The process should also involve researchers who are skilled in translating the information received from the public into a research project.

**Suggested Process:** We propose a transparent and sustainable public-clinician partnership process for generating a concise list of research topics. The process is based on the JLA approach and has been adapted for use in the context of a PCORI methodology framework and the United States healthcare paradigm. The PCORI process will include these elements: (1) inviting members of the public and clinicians to create a research collaboration; (2) peer consultations, in which lead members of the public and the clinician group will consult with their peers to identify possible research topics (using validated consultation methods); (3) data analysis, using peer-reviewed articles from health experience research to provide more detailed information; employing health disparities analyses to strengthen the perspectives of underserved populations; and systematic CER to identify gaps in research and to avoid research duplication. The emerging research themes are then combined and sorted during: (4) a consensus-building workshop using a moderated discussion between the public and clinicians to translate the emerging research themes into research topics. These topics are then: (5) submitted to PCORI to inform the PCORI research prioritization and funding process. This is followed by: (6) program evaluation to measure the impact of this engagement process on research and health outcomes before a new engagement cycle is started.
INTRODUCTION

During the past 20 years, the United Kingdom, Europe, Australia/New Zealand, Canada, and the United States have been pioneers in developing patient-centered care by involving the public in improving the quality of their healthcare services (see Appendix B for a list of patient-centered outcomes research [PCOR] organizations around the world). The public has also increasingly been invited to collaborate at the meta- and meso-levels of healthcare, such as in health technology assessment, the development of clinical guidelines, drug approval and food safety, and clinical research (Donaldson and Sox, 1992; AQuMed, 2007; Mosconi et al., 2007; Boivin et al., 2010; Mack, 2010; ACSQHC, 2011; AQuMed, 2011a; BCBS, 2012; Demotes-Mainard and Kubiak, 2011; Gagnon et al., 2011; Légaré et al., 2011; NICE, 2011; EFSA, 2012; EMA, 2012). The National Institute for Health Research (NIHR) of the United Kingdom was one of the first organizations to support the active involvement of the public in healthcare services research, establishing INVOLVE, an initiative designed to promote research with and by the public (Oliver et al., 2008; INVOLVE, 2012a). At the same time, in the United States, an increasing number of community-based participatory research partnerships were developed to reach out to underserved populations and decrease health disparities. For example, in 1995, the Centers for Disease Control and Prevention (CDC) supported the establishment of three Urban Research Centers (URCs) in Detroit, New York City, and Seattle (Israel et al., 2010; URC, 2011). Experience with patient involvement in research has shown that engaging the public early in the design of studies, ideally at the planning stage, leads to better results (IAPO, 2005; INVOLVE, 2009; Viergever et al., 2010; Patient Partner, 2012a). The James Lind Alliance (JLA), established in 2004, was one of the first to involve the public already at the time of generating research topics by establishing a sustainable process for public-clinician partnerships. Since the beginning of 2011, the European Commission for Research and Development has been including patient involvement as an evaluation criterion for clinical research proposals in some disease areas (European Commission, 2010).

Proponents of involving the public in healthcare research argue that, not only is it the public’s moral and democratic right to participate in healthcare research, but that, even more importantly, lay members might also provide insights that could lead to new discoveries. For example, it was the mother of an affected patient who suggested that the drug diethylstilbestrol (DES) may be related to vaginal cancer (Chalmers, 1995), and it was the mother of a child with trisomy 18 who was the first to suggest that a low level of maternal serum A fetoprotein might be a prenatal marker for this chromosomal abnormality. Moreover, there is a general consensus that involving the public in healthcare research introduces a higher level of accountability and transparency to medical research, and generates research that is more relevant to patients (Entwistle et al., 1998; Oliver et al., 2008; Faulkner, 2010; Boote et al., 2011; Patient Partner, 2012a).

It has become apparent that one of the most important concerns of patients is the question of which treatment or treatments are best for their condition and characteristics. This information is best derived from clinical studies that directly compare different therapies within a single study. However, since most new drugs or therapies are compared with placebo, or sham treatment, there has traditionally been little information from high-quality studies as to how a new treatment compares with standard or alternative therapies. This gap in knowledge has given rise to the new paradigm of comparative effectiveness research (CER) to answer the most important question patients have: “Which treatment is the best treatment for my condition?” (Wu et al., 2010). To make sure that “...selected topics truly represent a potentially large impact on the clinical or other outcomes that matter most to patients,” the Agency for Healthcare Research and Quality (AHRQ), the Institute of Medicine (IOM), and the Center for Medical Technology Policy (CMPT) recognize the need for meaningful involvement of patients as stakeholders in the selection of topics for CERs (IOM, 2008; Tunis et al., 2010; Whitlock et al., 2010). To date, few head-to-head clinical trials have been conducted to compare the effectiveness and safety of different health technologies; most comparative information comes from systematic reviews that synthesize evidence from individual studies.
The Patient-Centered Outcomes Research Institute (PCORI) in the United States has made it its mission to close the evidence gap by promoting best practices in PCOR and sponsoring patient-centered CER studies. As PCORI focuses on developing and identifying appropriate methodology to support its mission, the international perspective is a valuable complementary resource for tools, instruments, and processes (thereafter referred to as “methods”) developed for various settings and diverse patient populations, and designed to make the patient’s voice heard. Therefore, this white paper reviews methods of patient involvement that have been developed abroad and might be used by PCORI to engage patients in the selection of topics for patient-centered CER.

**OBJECTIVE**

The objective of this white paper is to provide PCORI with an evidence-based review of best practices, processes, and methods for patient engagement that have been studied and implemented internationally, and that could be adapted and used to promote patient involvement in research topic generation in the United States.

**METHODOLOGY**

**LITERATURE SEARCH**

Research for this paper began with a search of the scientific literature using the MEDLINE, Embase, and PsycINFO databases. The search strategy is provided in Appendix A. Additional information was obtained from an environmental scan that included the websites of international not-for-profit organizations, healthcare centers, and government bodies involved in patient-centered medicine (see Appendix B).

We identified and retrieved published studies, articles, and other information about methods that elicit and integrate the patient’s perspective in PCOR and CER, patient-centered healthcare, or the development of guidelines and health policy. The method could include one or several approaches to patient involvement. The target population for these methods could be patients, caregivers, or patient advocate groups. The setting was international in scope, including governments, healthcare centers, hospitals, and not-for-profit patient or physician organizations in the United Kingdom, Europe, Canada, the United States, and Australia/New Zealand.

Documents were selected for detailed review if they were published in English, French, German, Spanish, or Dutch. These languages were selected based on the availability of bilingual speakers of these languages at Hayes. Priority was given to comprehensive documents providing insight into a variety of methods, especially those that provided information on best practices and methods with a proven positive impact on patient involvement.

**APPRAISAL AND SYNTHESIS OF PROCESSES OF PUBLIC INVOLVEMENT IN HEALTHCARE RESEARCH**

For this purpose, we created a 10-item quality checklist (see Appendix C). This quality checklist was created by Hayes using items adapted from the Method Report for Patient Involvement of the German Agency for Quality in Medicine (AQuMED) and the guideline for patient involvement of the International Alliance of Patients’ Organizations (IAPO). Additional items were added to address issues that are specific to this project, to PCORI, or the United States healthcare system. Only high-level engagement processes were to be scored, giving priority to engagement processes specifically designed to collaborate with the public to generate research topics. A team of three analysts independently used this checklist to rate the quality of the processes for
patient involvement; the final ranking of the methods was assigned by consensus. The results of this ranking are summarized and provided in Appendix E of this white paper as a matrix table.

We performed an informal gap analysis to determine what additional information and research is needed, with a focus on research that addresses differences in patient personalities, culture, learning style, and clinical settings.

RESULTS

PRINCIPLES OF ENGAGING THE PUBLIC IN HEALTHCARE RESEARCH

The environmental scan identified several guidance documents from international governmental bodies and patient organizations that recommend practices for engaging the public in healthcare research, the development of medical guidelines, or health technology assessment. This information, mainly from the International Alliance of Patients’ Organizations (IAPO), Patient Partner, Value+, German Agency for Quality in Medicine (AQuMed), and the European Patients’ Forum (EPF), reflects more than 10 years of experience with public engagement and is summarized as a list of Principles for Engaging the Public in Healthcare Research. A description of these organizations can be found in Appendix D. These practices take into account the barriers and facilitators of public engagement that have been reported in the peer-reviewed literature (see Facilitators and Barriers to Eliciting the Public’s Voice). In addition, they aim to address the three main quality criteria of participatory research (Wright et al., 2009): “the quality of the cooperative relationship and the forms of participation; the quality of the research and its scientific standards; and the quality of the influence and impact of the research on the community.”

Principles for Engaging the Public in Healthcare Research

- The development of the engagement process is transparent, includes patients and other stakeholders, and includes a conflict-of-interest statement.
- The process used to invite and select participants is inclusive and balanced in terms of ethnicity, gender, age, and socioeconomic status.
- The roles and relationships for researchers and lay participants are explained and clarified at the beginning of each project.
- Members of the public are empowered to become active, respected participants in the PCOR or CER project.
- The public is engaged using appropriate, validated, and diverse methods, and these are utilized by staff experienced in the social sciences and PCOR or patient-centered care.
- There is a process in which the public’s opinions, preferences, concerns, and expectations are prioritized and translated into a well-defined research topic; this process is feasible in a wide variety of settings and suitable for diverse conditions and populations.
- Practical aspects of diffusion and implementation are considered.
- The process is sustainable and establishes a culture of improvement. There are measures for quality control of patient participation to ensure that the integrity of the process of patient involvement is maintained over time and across different projects.
METHODS FOR ELICITING THE PUBLIC’S VOICE

CONSIDERATIONS BEFORE DECIDING ON A PROCESS OF PUBLIC INVOLVEMENT

Before implementing a process for involving the public in developing research topics, we recommend considering the following questions:

- Who of the public needs to be invited to participate in the project?
- What is the type and level of public engagement?
- What is the best strategy and framework for the engagement?
- What is the duration of engagement and where should it take place?

PUBLIC

Patients are at the center of the public engagement process in generating research topics for PCOR and CER. Lay people who currently are patients or who have been patients, relatives or friends and caregivers of patients, patient advocates, or any other lay person who anticipates using healthcare services in the near future can participate.

TYPE AND LEVEL OF ENGAGEMENT

The public can participate in the generation and selection of research topics in different ways: as an individual or group who provides experiential knowledge of what it is like to live with the disease either as a patient or caregiver, relative or friend; as active collaborators who work jointly with clinicians and researchers within a consensus-building process; or as the driving force, either as active researchers or sponsors. Different typologies have been suggested to group and categorize these types of engagement (provided in Appendix F) using either a hierarchical approach (Arnstein, 1969), a typology based on the direction of communication (Rowe and Frewer, 2005), or a matrix typology based on who initiates the engagement, the number of people who are engaged, and the social form of engagement (Oliver et al., 2008).

We developed a patient-centered typology of engagement in which we consider all forms of engagement in which researchers are in control of the project and patients (or other members of the public) are engaged on the terms of the researchers or the organization in charge of the project as minimal forms of engagement (see Figure 1). The premise of our typology is that patient-centered research organizations have to actively reach out to the public and choose several forms of engagement that encourage diverse engagement to obtain sufficient heterogeneity (consultation) and empower participatory decision making (collaboration). Patient-centered research organizations actively reach out to the public, inviting and encouraging public members to share their in-depth views, perceptions, preferences, experiential knowledge, and ideas, and empowering public members to become active and equal partners. When researchers restrict public engagement to minimal involvement, such as only providing information to the public or requiring that the public members adapt to established researcher-controlled processes, it is often perceived as “tokenism” and is, therefore, not useful when planning or performing patient-centered research.

Consultations are engagements, in which researchers and organizations actively solicit from the public information regarding their experiences, views, or preferences. In collaborations or partnerships, public members are empowered to actively participate in ongoing affiliations with researchers and healthcare professionals, and are formally engaged to complete a specific research project. Experiences with public involvement have shown that a combination of collaboration and consultation, with members of the public taking a leading role in consulting their peers, is the most effective method of involving the public in the design of large-scale research projects (Oliver et al., 2008).
In public-controlled research, members of the public design, perform, and disseminate the results of a research project (Faulkner, 2010). Researchers and healthcare professionals are only involved at the invitation of the public (Hanley et al., 2004; Oliver et al., 2004). An example for public control is AFM (Association Française pour la recherche sur la trisomie 21/French association for research on Trisomy 21) (Patient Partner, 2011). This organization is run solely by parents whose children are affected by Down syndrome. A few board members are medical professionals. Created in 1990, AFM promotes research in Down syndrome (Trisomy 21) by giving grants and fellowships.

![Figure 1. Typology of Engagement](image)

**SCIENTIFIC STRATEGY AND FRAMEWORK OF ENGAGEMENT**

The scientific philosophical paradigm that underlies public engagement is **interpretivism**. The premise of interpretivism is that people construct the social world and that the social world can only be understood through the analysis of people’s values and relationships. Because of this, interpretive sociology recognizes that there may be different social realities, and that people who live in different social constructs experience life differently. Interpretivism aims at understanding the social world by studying people’s values and relationships, and how they experience the world. This is in contrast to the premise of **positivism**, which presumes that the
social world, similar to the natural world, follows regular patterns that can be measured and explained through logical analysis (Alderson, 1998; Thomas, 2006; Denscombe, 2010).

Eliciting the public’s experiential knowledge, views, perspectives, and preferences employs mostly qualitative social science strategies and methods. The scientific strategy sets the framework of engagement and clarifies the researcher’s perspective regarding the type and extent of information that is elicited and the interpretation of the results. Thus, the use of an appropriate social science theoretical framework, and being aware of its implications for the engagement and research, are as important as the specific research methods that are being used. Common strategies for eliciting the public’s experiential knowledge and perspectives are described in the following sections (Denscombe, 2009; Denscombe, 2010):

- **Phenomenology**: Phenomenology is a research strategy that describes and interprets human experiences. As such, phenomenology is concerned with the public’s perceptions or meanings, attitudes and beliefs, and feelings and emotions.

  Phenomenological research does not look to explain or find the causes of an experience but to describe and interpret as accurately as possible how an individual or group experiences life—in other words, seeing it through the eyes of others. An example of a phenomenological study is the description of the parental experience of childhood cancer (Schweitzer et al., 2011). Another example is a study exploring the quality of life of family caregivers of Huntington’s disease patients (Aubeeluck et al., 2011).

- **Ethnography**: The term *ethnography* means, in its literal translation, the description (Greek: *graph*) of peoples or cultures (Greek: *ethnos*) (Hancock, 2002; Denscombe, 2009). Ethnographic research provides a holistic view of the relationships, processes, and interdependencies among the individuals of the studied community, taking into consideration all of its social, psychological, and cultural aspects. The premise of this approach is that a community can only be understood in its entirety. A recent example is an ethnographic study exploring the perceptions of treating and healing brain injury from a First Nations cultural perspective in Canada (Keightley et al., 2011).

- **Grounded theory**: Grounded theory is a research approach that aims at building theories based on empirical research. This approach opposes the process of generating purely abstract theories that are then subsequently confirmed with empirical research. Researchers start out with an open mind but do have knowledge of the area of study. The goal of grounded theory research is to develop theories that are useful in daily life, not only for social scientists but also for lay people. As such, the grounded theory approach is inherently based on the philosophical framework of pragmatism. The grounded theory approach is particularly useful for the study of human interactions researching practical activities and routine situations and the participant’s point of view. Recent examples of the use of grounded theory are studies evaluating how patients experience the management of rheumatoid arthritis in everyday life (Bergsten et al., 2011); the experience of living with fibromyalgia (Hallberg and Bergman, 2011); and the pain management of drug users in an acute care setting (McCreaddie et al., 2010).

- **Action research**: Action research unites the two fundamental components of research and implementation of research results (action) in one step. Action research has the following characteristics:
  - Action research addresses practical real-world problems or issues.
  - Change is an integral part of action research.
Action research is cyclical; the research findings create possibilities for change that are implemented and tested.

Action research is participatory; practitioners and stakeholders are actively, rather than passively, involved in the research project.

A recent example is the use of participatory action to integrate the suggestions of aboriginal peoples for conducting health studies in their own population (Maar et al., 2011).

Mixed methods (also, mixed methodology, multi-strategy research, integrated methods, multi-method research, combined methods): The mixed methods approach refers to studies that combine several research approaches in one research study. Its basis is the philosophical framework of pragmatism. Characteristics of mixed methods research include:

- The use of quantitative (QUAN) and qualitative (QUAL) data in one research project.
- An explicit link between the research approaches, justifying the use of the different methods, and explaining how these methods complement each other.
- A problem-driven research project that intentionally uses methods from different research paradigms that are conventionally regarded as incompatible, to solve a specific research question.

Mixed methods research is used to validate findings in terms of their accuracy, to check for biases in research methods, and to develop a research instrument. Key issues that have to be explicitly addressed in mixed methods research are: the order in which the methods are used, at what time point the methods are changed or combined, which method is considered the dominant method, comparing and contrasting methods (what are similarities and differences), and what is the purpose of using the approaches combined.

Recent examples are a study investigating the impact of systemic lupus erythematosus on health, family, and work from the patient’s perspective using a qualitative analysis of focus group data and a quantitative analysis of questionnaire responses (Robinson et al., 2010). A second example is a study investigating patient-reported barriers to colorectal cancer screening using a combination of qualitative analysis from focus group research and quantitative data from a questionnaire (Jones et al., 2010).

Survey: A survey is a research strategy that is used to obtain a broad overview rather than to elicit in-depth information. Another characteristic of surveys is that they provide information at a specific point in time, usually the present moment. Thirdly, surveys generally use empirical methods rather than qualitative data. A number of different methods can be used for a survey, including questionnaires, interviews, documents, and observation. A recent example pertaining to PCOR is a study investigating the importance of patient-centered care for different patient groups (de Boer et al., 2011).

When studying the subjective experience of a person or group, the researchers’ own worldview may impact the result of the engagement (referred to as reflexivity). Therefore, alternate methods, strategies, or data are combined to address a specific research question (or to engage the public) and at least two social science researchers elicit experiential knowledge—one person as the active researcher and the second as an observer—thus, increasing the validity and certainty about the elicited information. In the social science literature, this is referred to as triangulation (Denscombe, 2009; Denscombe, 2010). Therefore, while in the following sections we list the engagement methods separately, they are often combined to achieve more accurate results or a greater depth of information.
TIMING, DURATION, AND LOCATION OF ENGAGEMENT

The timing, duration, and location of involvement are also considerations. Interventions can take place at a physical location, over the phone, or using online media approaches. A combination of on-site and off-site approaches is also conceivable, and may be especially appropriate when there are scheduling problems or if there are participants who cannot travel to an on-site meeting location. Meeting planners should also be aware of cultural or religious holidays and events and should not schedule meetings on such dates (NCCC, 2000). Meeting venues should be accessible for all participants (NCCC, 2000; INVOLVE, 2009). An example of an Accessibility Checklist to evaluate the accessibility of a venue is available from the NCCC (NCCC, 2000) and the Scottish Health Service (SCH, 2010). Other considerations are dietary requirements, sign language interpretation and other translation requirements, and the economic impact of participation on the public members (NCCC, 2000).

Once these decisions have been made, methods of engagement are chosen that are appropriate for the condition and the lay members who are invited to participate.

METHODS FOR ELICITING THE PUBLIC’S VOICE IN HEALTHCARE RESEARCH

The following sections summarize specific methods for eliciting the public’s experiential knowledge or perspectives. These methods are organized based on the type of engagement. The first section describes methods that are used to elicit in-depth experiential knowledge, while the second describes processes that involve the public as active participants in public-clinician partnerships for the generation of research topics.

METHODS FOR ELICITING EXPERIENTIAL KNOWLEDGE

Several methods for eliciting experiential knowledge exist, including the use of (Denscombe, 2009):

- Interviews (one-on-one or group interviews)
- Observation
- Documents
- Questionnaires

The most frequently used methods are one-on-one interviews, focus group interviews, and observation, most commonly using phenomenological, ethnographic, and grounded therapy strategies (Hancock, 2002; Facey et al., 2010).

One-on-One Interviews: During one-on-one interviews, an interviewer elicits the perspective of the person interviewed. Ideally, an observer is also present. The interview can be unstructured, semi-structured, or structured. Unstructured in-depth interviews are preferred, as an unstructured format using open-ended questions draws more fully upon the complete experiential knowledge of the person being interviewed. Often, a one-on-one interview is combined with a focus group interview because a person may reveal information in a one-on-one interview that he or she would not share in a group setting, and as a form of triangulation. An example is the use of in-depth one-on-one interviews and focus groups for the identification of research topics for ulcerative colitis (see Case Study 1; Welfare et al., 2006).
Focus Group Interviews: During focus group interviews, a skilled moderator facilitates by setting the frame for the discussion, creating an atmosphere that encourages open dialogue, and making sure that all participants feel comfortable in sharing their thoughts, perceptions, views, feelings, and ideas about the given topic (Denscombe, 2009). Focus groups involve a small group of people, usually 6 to 9 participants. The group dynamics and interactions among group members permit the researcher to identify contrasting views and to understand the extent of agreement and disagreement regarding topics. Recent examples are the use of focus groups in developing research topics for chronic kidney disease (Tong et al., 2008); eliciting the individual experience of functioning and disability in Switzerland in spinal cord injury (Lüthi et al., 2011); in combination with in-depth one-on-one interviews for the identification of research topics for ulcerative colitis (see Case Study 1; Welfare et al., 2006); and as part of an expert consensus development process to develop research topics and priorities for pancreatic cancer (Robotin et al., 2010).

Advisory Panels: Advisory panels (also called people’s panel, community advisory committee/board/council, or citizen jury) are also moderated group interviews (Gooberman-Hill et al., 2008; Herbison et al., 2009). Citizen juries use a moderated process similar to focus groups and include an educational component. Citizen juries take more time than focus groups (focus groups last 1.5 to 2 hours, citizen juries may take 1 or several days) (Herbison et al., 2009). An example is the use of citizen juries to identify research topics and research priorities related to urinary incontinence in women (see Case Study 3; Herbison et al., 2009).

Interviews Using Visual Elicitation: Most interview techniques rely on verbal elicitation, in which an interviewer asks questions or stimulates discussion and the participants respond verbally. Interviews may also include a component of what is referred to as visual or graphic elicitation, using drawings or photographs. The use of photography is called photovoice or photo elicitation. Photovoice is a process in which lay persons take photographs that best represent their views, perspectives, and challenges of living with a health condition (Catalani and Minkler, 2009). Although photovoice has been extensively used in participatory action research projects, its use in PCOR is novel. Photovoice has been used to elicit the pain experience in patients with chronic pain (see Case Study 4; Baker and Wang, 2006). During an interview, the researcher elicits the concrete and symbolic meanings of the photographs. This method is also often combined with a narrative or diary. As with traditional interviewing methods, two researchers lead the study, and the photos, narratives, and interview responses are recorded. Photovoice has also been used to engage children (Nechele et al., 2007) and patients with learning or mental disabilities (Jurkowski, 2008; Fleming et al., 2009). Another example is the use of photovoice in a grounded theory strategy to elicit the quality-of-life concerns of African American breast cancer survivors in rural North Carolina (López et al., 2005). The use of digital images may make this method more accessible and could be combined with digital narrative formats such as blogs or online forums.

All types of interviews involve at least two researchers, with one researcher leading the interview and one researcher observing. The sessions are recorded with audio or video or both. The data are synthesized and analyzed using qualitative research methods (see section on TRANSLATING THE PUBLIC VOICE INTO A RESEARCH QUESTION). Interviews can be performed in person, on the telephone (Robotin et al., 2010), or via an Internet-based approach (Lüthi et al., 2011).
Observation: Observation is a method for eliciting the patient experience that is often used as a complement to interviews. The views and perspectives given in an interview may not always reflect all aspects of a person’s life with a health condition. In some cases, the patient may not be able to share his or her experience in a traditional interview setting. Observation could therefore be a useful method for mental health conditions, for example, in the case of a child with low-functioning autism. Although in this case, parents, caregivers, and teachers may provide insights into the child’s experience, a social scientist specifically trained in observation methods can provide more objective information. Observers gather data in the form of field notes, video and audio, or photography. An example is a study investigating children’s perspectives of home ventilation and how it affects their lives (Earle et al., 2006).

Documents: Another method for eliciting the public perspective is the use of documents, produced either by the patient or about the patient (Street et al., 2008). These may include written or video patient diaries, narratives, or blogs (Street et al., 2008; Denscombe, 2009). An example is an online database of videos, created by the charity DIPex, in which patients describe their experience of living with a health condition (DIPEx, 2011; DIPEx, 2012a; DIPEx, 2012b). Dr. Ann McPherson, CBE, and Dr. Andrew Herxheimer created DIPex, working closely with the Oxford Health Experiences Research Institute (HEXI) at the University of Oxford. DIPex uses validated social science methods to collect and analyze these interviews (Ziebland and McPherson, 2006; DIPex, 2012b). DIPex and HExi have established collaborations with research organizations in Japan, Germany, Spain, Korea, and Australia (GTC, 2012). An additional source of documental information is patient organizations. These organizations often keep records on the reasons that patients call them; through surveys, they may gather information on what it is like to live with a health condition (Facey et al., 2010).

Questionnaire: The questionnaire is a research method that is mostly used with the survey research strategy. Questionnaires provide a structured form of inquiry. They consist of a list of questions that is the same for all participants in a study. The collected information is then used as generally quantitative empirical data. Several types of questionnaires exist; the most common types are face-to-face, postal, and Internet-based questionnaires. Questionnaires are used when answers to relatively simple, straightforward questions are needed. A recent example pertaining to PCOR is a questionnaire study investigating the importance of patient-centered care for different patient groups (de Boer et al., 2011). Questionnaires may be less useful when eliciting experiential knowledge.

Questionnaires can also be administered in the form of polling and televoting via smartphones (also referred to as “crowdsourcing”). Compared with traditional questionnaires, this approach is more interactive, requires minimal infrastructure, generates data more quickly, and does not require comprehensive design and planning before data are collected. It is simple to use. Furthermore, “crowdsourcing” is suitable for engaging many individuals from a broad range of settings and locations. Case Study 6 provides an (unpublished) feasibility study provided by InCrowd (www.incrowdnow.com) that used “crowdsourcing” to elicit information regarding the experience of arthritis pain.

The confidentiality of patient data has to be preserved; therefore, we recommend the use of platforms that protect the privacy of patients and the public. Thus, applications provided by search engines such as Google are not recommended.
Public-clinician partnerships engage patients in decision making.

A large number of public-clinician partnerships exist in the United States. These partnerships were specifically created to engage the public in healthcare services research with the goal to decrease health disparities. Examples are (not all inclusive):

- The Partnered Research Center for Quality Care (an infrastructure to support community-partnered participatory research in mental health) (Lizaola et al., 2011).
- Three Urban Research Centers (URCs) in Detroit, New York City, and Seattle, established in 1995 with core funding support from the Centers for Disease Control and Prevention (CDC) (Israel et al., 2010; URC, 2011).
- The University of California in Los Angeles (UCLA) family medicine community-based participatory research partnership (Moreno et al., 2010).

These centers provide many years of experience of high-quality collaborative engagements in research. However, we did not find specific frameworks of processes that were specific for the generation of topics.

Some Europe-wide healthcare organizations involve lay persons or patient organizations. The European Medicines Agency (EMA) established the Patients’ and Consumers’ Working Party (PCWP) that consists of up to 20 members of different patient organizations (EMA, 2012). The PCWP meets 4 times per year and provides recommendations to EMA’s Human Scientific Committees. The European Clinical Research Infrastructure Network (ECRIN) invites representatives of patient organizations to its advisory board (Demotes-Mainard and Kubiak, 2011). The European Food Safety Authority (EFSA) pursues different activities to engage the public (civic stakeholders). EFSA developed a stakeholder consultative platform, which meets 3 times per year, and also organizes an Annual Colloque (EFSA, 2012). In addition to these formal activities, EFSA also maintains constant informal relationships with the public and encourages public involvement through consultation, participation in data collection, or the submission of any data and information that could be relevant to the work of the EFSA. The disadvantage of these processes is that they still typically engage the public on the terms of the researcher, agency, or organization, and are thus not truly patient centered.

The James Lind Alliance (JLA) in the United Kingdom has created a sustainable process for public-clinician partnerships to identify and prioritize clinical uncertainties and translate these into research questions in which the public members are empowered to participate as equal partners, using the nominal group technique (NGT) to facilitate the consensus process between the public and clinicians (CDC, 2006; Lloyd and White, 2011; JLA, 2012). The NGT was developed in the United States in the 1970s and has since diffused internationally and is one of the most commonly used consensus-making techniques (Delbecq and Van de Ven, 1971). The process also includes a phase of public members and clinicians consulting with their peers; this process increases diversity and empowers the public members of the group. The JLA process consists of five phases: In phase 1, the members of the working partnerships are identified and invited; in phase 2, public members and clinicians identify research topics by consulting with their peers and by analyzing published evidence to identify gaps in the research (for example, using systematic reviews); in phase 3, the emerging research themes are categorized; and in phase 4, public representatives and clinicians meet during a workshop to define and prioritize the research questions. In phase 5, the research questions are published and added to an online database (NHS, 2012). Case Study 2 provides an example of this process used to identify and prioritize research topics for ulcerative colitis (Buckley et al., 2011). This process has been used to identify research topics for asthma (Elwyn et al., 2010), schizophrenia (Lloyd et al., 2006), and prostate cancer (Lophatananon et al., 2011), among others.
This was the only process described in the peer-reviewed medical literature that was specifically created for generating research topics. When we applied the predefined checklist to evaluate the quality of the engagement process (see Appendix E), the JLA process met all criteria and was, therefore, adapted to create a model for the PCORI process of public engagement for generating research topics (see SUGGESTED PROCESS OF ENGAGEMENT).

### TRANSLATING THE PUBLIC VOICE INTO A RESEARCH QUESTION

Methods for eliciting the public’s voice in healthcare research may involve a variety of different types of data, including audio and video material, transcripts of audio files, documents, or field notes. These are commonly stored and analyzed using computer-assisted quantitative data analysis (CAQDAS) professional software for qualitative data analysis (Ziebland and McPherson, 2006; Denscombe, 2009). Some of the most well-known software packages are:

- NVivo (QRS International, 2012)
- NUD*IST (Dartmouth College, 2012)

The material is synthesized using a process called content analysis. The data are coded, tagged, and classified into categories or themes (Hancock, 2002; Ziebland and McPherson, 2006; Denscombe, 2009). The use of content analysis software is controversial, and there are some researchers who believe that it violates the principles of qualitative methodology. Therefore, the software analysis is best used in combination with traditional content analysis by two experienced social scientists.

Once the data are grouped, the themes can be used to state research-specific questions. Research questions usually are phrased using these elements: Intervention, Comparison, Patient, and Outcome (also referred to as the ICPO format) (Daly, 2011). The following is an example of a translation of public voice into a research question:

**Original question:** Is there any long-term definitive data that a multiple, 4-a-day injection regime produces better control than 2 daily injections?

- Intervention: 4-a-day injection of insulin
- Comparison: 2-a-day injection of insulin
- Patient: People with type 1 diabetes
- Outcome: Better control

**Formatted question:** Does a 4-a-day injection of insulin or a 2-a-day injection provide better control of type 1 diabetes?

The research themes or questions can then enter a research prioritization process using validated methods, for example, using peer review, conjoint analysis, or discreet choice (Bridges et al., 2008; Viergever et al., 2010; Watson et al., 2011).

### CASE STUDIES OF ELICITING THE PUBLIC’S VOICE TO GENERATE OR REFINE RESEARCH TOPICS

We selected five case studies that illustrate methods for eliciting the public’s voice to generate research topics for different health conditions. These are presented in the following sections.

**CASE 1: THE IDENTIFICATION OF TOPICS FOR RESEARCH THAT ARE IMPORTANT TO PEOPLE WITH ULCERATIVE COLITIS (CONSULTATION/HEALTH EXPERIENCE RESEARCH)**

**Method:** Focus group and in-depth interview
The study had two aims: (1) to elicit the patient’s perspective to identify topics for research that are important to patients with ulcerative colitis; and (2) to develop a framework that would allow the classification of these data into research areas (Welfare et al., 2006). The study enrolled 40 people with ulcerative colitis (age range 19 to 71 years; 20 men and 20 women). A purposive randomized sampling approach was used to achieve a balance of people with different experiences and to represent a range of sex, age, and duration of ulcerative colitis. The setting was a general hospital in England that serves a mixed and mainly urban population of 190,000. An ethnographic approach using a combination of focus groups and in-depth interviews was used to explore the experiences and views of the local public. Each group had a moderator and an observer; both were social scientists with appropriate ethnographic research experience. All sessions were taped, transcribed, and entered into the software “Framework” (a software package for social science research, now NVivo) to store and analyze the gathered data (QRS International, 2012). Framework uses a content analysis process to classify and summarize data within a thematic framework; these data are then categorized into subject groups. The identified research topics were similar between the focus group and the in-depth interviews; however, only during the in-depth interview did people ask about prenatal genetic testing for ulcerative colitis for a possible termination of pregnancy if the fetus was affected. The research areas of interest were: (1) finding the cause of colitis; (2) cure of colitis; (3) prevention of colitis; (4) living with colitis; (5) treatment and its side effects; (6) control of particular symptoms; (7) information about ulcerative colitis; (8) communicating with health professionals; and (9) service delivery. Service users saw a link between finding the cause of colitis and discovering a cure and/or being able to prevent colitis. The participants identified many possible research topics in areas of their personal life.

CASE 2: METHODOLOGY FOR IDENTIFICATION AND PRIORITIZATION OF RESEARCH NEEDS FOR URINARY INCONTINENCE THROUGH PATIENT-CLINICIAN CONSENSUS (COLLABORATION COMBINED WITH CONSULTATION)

Method: Collaboration of members of the public and clinicians who, in turn, consulted with their respective peers

The aim of this project was to evaluate a methodology for identifying and prioritizing research topics for urinary incontinence through a process based on patient and clinician consensus (Buckley et al., 2011). The James Lind Alliance (JLA)—an initiative that promotes patient-clinician partnerships in health research—and the Cochrane Foundation supported this project. The process involved five phases:

- **Phase 1:** Patient and clinical organizations with an interest in urinary incontinence were identified through a web search and by consulting peers, and invited to participate in this study.

- **Phase 2:** Participating patient and clinician organizations asked their members to identify gaps in the scientific knowledge, defined as the absence of a recent systematic review, regarding the treatment of urinary incontinence, which regularly affected their ability to make decisions about treatment decisions.

- **Phase 3:** Representatives of each organization synthesized the information into research questions.

- **Phase 4:** Patient and clinician representatives worked together to identify and prioritize a “top 10” list of research topics related to urinary incontinence.

- **Phase 5:** The final list of research topics was published in the *Neurourology and Urodynamics* journal, presented at national and international research conferences, and sent to the National Institute for Health Research (NIHR) Evaluation, Trials and Studies Coordinating Center.
A total of 8 patient and 13 clinician groups participated in this study. In phase 2 417 individual submissions were received. In addition, a further 131 unanswered research questions were identified from the recommendations of systematic reviews and clinical guidelines. In phase 3, these unanswered research questions were synthesized into a final database containing 226 research questions: 79 from patients, 37 from clinicians, 6 from patients and clinicians, 2 from both patients and research recommendations, and 102 from research recommendations alone (Buckley et al., 2011). The top 10 questions were:

- What are the optimal pelvic floor muscle training (PFMT) protocols (frequency and duration of therapy) for the treatment of different patterns of urinary incontinence (UI)?
- Can guidance or training for general practitioners (GPs) on appropriate pathways of care improve the management of patients with UI?
- What is the best practice for the treatment of combined stress UI (SUI) and detrusor overactivity?
- What catheter regimens are most effective in preventing urinary tract infections in patients using intermittent self-catheterization for the management of a neurogenic bladder?
- Which treatment is most effective for the reduction of urinary frequency and urgency?
- Is urodynamic testing before surgery for UI associated with better continence rates and quality of life than surgery performed without such testing?
- What is best practice for the management of SUI following failed tension-free vaginal tape surgery?
- What are the most effective treatments of daytime UI in children?
- Are disposable catheters more or less acceptable than reusable catheters in terms of effective bladder management, patient experience, and urinary tract infections?
- In women with prolapse and SUI, should suburethral tapes be inserted at the same time as repairing the prolapse?

Since these prioritized research questions were published, five studies have been funded, of which three are in development; five new systematic reviews are in progress, one of which is being updated; and five questions are under consideration by a national research commissioning body.

**CASE 3: RESEARCH PRIORITIES IN URINARY INCONTINENCE IN WOMEN: RESULTS FROM PEOPLE PANELS (CONSULTATION)**

**Method:** Public panel

This is an example from New Zealand of public panels eliciting research ideas, prioritizing research, and developing outcome measures (Herbison et al., 2009). Purposive selection was used to invite women with self-reported urine leakage and no other important comorbidities such as diabetes or neurological conditions. The study involved two public panels: one for women with stress urinary incontinence and one for women with urge urinary incontinence. The participants had 1 day of education in incontinence, advantages and disadvantages of current treatments, and the outcomes that patients were likely to have. There was sufficient time for discussion after each session. The second day started with 1.5 hours of panel deliberation. Two researchers were available in the adjacent room to answer questions from panel members (jurors). During the final deliberation, panel members were asked to respond to two questions:

- What can researchers study to make your life better?
- What should we measure to see if your life is better?

The jurors noted ideas on flip charts. Then the researchers were invited into the room. During the discussion, which was led by one of the jurors, jurors could expand on the ideas, and all additional thoughts were added on additional flip charts. One researcher observed the discussion and the second researcher took notes on a new flip chart or added expanded notes to the flip charts already prepared by the jurors. The flip charts were
displayed in the room. The researchers encouraged everyone to participate and did not disregard any idea. This meeting was audio recorded. Towards the end of the discussion, the ideas were prioritized into three groups: (1) top priority; (2) still important but not top priority; and (3) lesser priority. One researcher categorized the ideas and a second researcher verified the categories. The emerging themes were given a descriptive title. This process was performed for each jury. At the end, the categories from both juries were combined. The researcher reported the results to the panels to confirm that the researchers had summarized their ideas correctly.

Five main research areas emerged: (1) research into interventions that make seeking help easier; (2) research into information giving and interventions designed to make day-to-day life more manageable; (3) research into the true costs of incontinence; (4) research to increase knowledge about causes (e.g., obesity/weight loss; ethnic/cultural factors; obstetric/gynecological history; role of the nervous system, hormones, chemicals); and (5) the effects of lifestyle modification on incontinence (e.g., complementary and alternative medicines, nutrition, exercise).

The participants considered quality of life the most important outcome. Quality of life had to include sex life, the quality of life of partners, and emotional stress. Quality of life was described as “what it stops them doing with their problem.” Both groups considered the commonly used clinical outcomes such as pad tests and bladder diaries as not valid and not reproducible—“Frequency and amount are really a subsidiary outcome and a little bit to one person is a lot to another.”

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**CASE 4: PHOTOVOICE: USE OF A PARTICIPATORY ACTION RESEARCH METHOD TO EXPLORE THE CHRONIC PAIN EXPERIENCE IN BLACK AND WHITE OLDER ADULTS (CONSULTATION/HEALTH EXPERIENCE RESEARCH)**

**Method:** Interview with visual elicitation (photovoice)

This study took place at the University of Michigan’s Multidisciplinary Pain Center (Baker and Wang, 2006). Baker and Wang used photovoice to explore the chronic pain experience in black and white adults older than 50 years of age who had experienced chronic pain for at least 3 months. The project had four phases:

- **Phase 1:** Nonclinic patients had an orientation session in the community. Clinic-based patients received written instructions. This was done for logistical reasons. Clinic-based patients were more impaired than nonclinic patients and it was, therefore, more difficult to schedule orientation sessions.

- **Phase 2:** All patients received a disposable camera. They were instructed to take photographs of objects that best represented their experience with chronic pain. The participants then returned the disposable cameras to the research center where they were developed, labeled, and returned to the participants. The participants were asked to choose four photos and to provide a title and narrative that described their experience of living with pain.

- **Phase 3:** Phase 3 used the same process as phase 2, but this time, all participants took photographs that best represented what they would like life to be without pain.

- **Phase 4:** Phase 4 consisted of a survey assessing patients’ residential and demographic information and an exit interview using open-ended questions about the patients’ participation in the study, their experience as a photographer, and their daily experiences with chronic pain.

The study did not attempt to generate research topics from the gathered information. Figures 2 and 3 provide examples of the elicited information.
Patient narrative for this photo: “We all walk by a rose and look at the beautiful flower, yet we fail to see the thorns. We walk by a person and can only see their face; not a true reflection of what hides inside. My whole life was spent outside, very active and really happy until the day my thorn (pain) pierced my life. Now that flower is my face and the thorns are my pain, my life.”

Figure 2. Photovoice example entitled “Rose in a Sea of Thorns”

Patient narrative for this photo: “Pain is an unwanted guest in your body. No one can see that it’s with you. After so many surgeries and doctor visits, it (pain) just won’t leave. The drugs are the only things that keep you at peace with the pain. Is this all modern medical science has to offer? Pills and patches...little electrical gadgets...a surgeon’s knife? Cover it up or cut it out? Aren’t there any other choices? I wonder what I will be like in the years ahead with my body’s exposure to all these drugs?”

Figure 3. Photovoice example entitled “The Unwanted Guest”

**CASE 5: PHOTOVOICE TO ELUCIDATE QUALITY-OF-LIFE ISSUES IN AFRICAN AMERICAN BREAST CANCER SURVIVORS IN RURAL NORTH CAROLINA (CONSULTATION/HEALTH EXPERIENCE RESEARCH)**

**Method:** Interview with visual elicitation (photovoice) using a grounded theory approach
African American women in rural communities have historically been discouraged from expressing quality-of-life concerns as breast cancer survivors (Lopéz et al., 2006). Older women, especially, still remember their experience of social inequality from a segregated healthcare system, making it difficult for them to express quality-of-life concerns, which may be different from those of white women. This study used photovoice as a method to facilitate discussions to understand the factors that impact the quality of life of African American women and their coping strategies.

The study took place in rural North Carolina in three counties that are ranked high as economically deprived and are 45% to 60% African American. Women who have breast cancer often travel 35 to 100 miles to receive specialized care or cancer care products. A purposive sampling strategy was used to invite 18 women who were diverse with respect to demographics and other characteristics that could influence the experience of cancer survivorship (e.g., education, age, time since diagnosis, insurance coverage, type of treatment) and who met the following selection criteria: (1) had completed initial treatment; (2) agreed to take photographs about their experience as a breast cancer survivor; and (3) agreed to share their experience with a small group of peers. Thirteen of the women, aged 44 to 82 years, agreed to participate in the study; of these, one was the mother of a woman who had died from breast cancer. The women received photography training but were free to choose their assignments. Each assignment took 1 month; the women then shared and discussed the photos during a 3-hour session. The following open-ended questions guided the discussion:

- What do you see in this photograph?
- What is happening in the photograph?
- How does this relate to our lives?
- Why do these issues exist?
- How can we become empowered by our new social understanding?
- What can we do to address these issues?

After five photo assignments, no new themes emerged. Figure 4 is an example of a photograph taken for an assignment that illustrates the positive and negative impact of religion on survivors.

The analysis of the data showed that three social forces (stigmatizing beliefs about cancer, racial discrimination, and cultural beliefs about African American women) affected concerns about quality of life. These concerns were: needing safe sources of support, adjusting to the role of being a breast cancer survivor, and feeling comfortable with the future. All women agreed that reaching out to community members and serving as a role model for other women who suffer from breast cancer was a positive result of her experience.

The women used various strategies to address their quality-of-life concerns. They identified faith and spirituality as a source of strength (see Figure 4). The women also found strategies to maintain their social standing. Some achieved this by continuing to serve the community and keeping health concerns to themselves. The women also avoided situations in which they felt there was racial discrimination, and they turned away from people, support groups, or service providers that exhibited signs of racism. An example was a support group of mostly white women who decided to change the meeting location to a site that black women would usually not frequent. This was perceived as a sign that the group (of white women) wanted to exclude the black women from participating in the support group.
CASE 6: FEASIBILITY STUDY FOR THE USE OF SMARTPHONE POLLING (“CROWDSOURCING”) TO ELICIT THE PUBLIC VOICE

Method: Smartphone polling (“crowdsourcing”)

The objective of this study was to test whether “crowdsourcing” is feasible for eliciting the pain experience of patients with arthritis and to elicit the patient’s voice with regard to whether their physician outlined clear expectations of the improvement they could expect.

This technology can be used to access a “crowd” of more than 6 million healthcare consumers encompassing different racial and ethnic minorities, women and men across the adult (≥ 18 years of age) age range, and those with a wide variety of different health concerns. These patients have opted into a community, agreed to be surveyed regarding healthcare-related questions and preferences, and promised to answer candidly and promptly. The participants receive rewards for each question they answer. Questions are sent out to this “crowd” via text message and e-mail; answers are collected, collated, analyzed, and summarized.

The web-based InCrowd survey platform works in the following manner:

• For each question, 1 of 10 different question types is permitted, including yes/no, multiple choice (select one or more), open-ended, multiple short answer, rating scale, forced ranking, numerical allocation, numerical response, or a matrix question that allows categorical responses for several items.
• The question and response options are entered; response options can include free text.
• The target audience is selected (can select specific groups based on demographic characteristics or disease condition). Demographic characteristics include age, ethnicity, sex, income, educational level, and geographic location. Patients can be identified according to a comprehensive range of health conditions, including chronic problems such as arthritis, Alzheimer’s disease, asthma, back problems, diabetes, cancer (by type), chronic obstructive pulmonary disease (COPD), kidney disease, and various aspects of chronic musculoskeletal pain.
• The number of responses and/or time frame is specified for each question.
• The question(s) is/are sent out via e-mail and text message.
• Responses come directly to the questioner within hours; participants have agreed to answer questions promptly.
• The results are tabulated automatically and stratified according to designated demographic or disease characteristics.
• Follow-up questions can be posed to clarify or amplify respondents’ answers and particular aspects of the results can be replicated quickly with more focused samples if necessary.

Using the InCrowd platform, two questions were sent out to patients with arthritis:

**Question 1:** Please list below the top five (5) items that describe how your arthritis pain and difficulties impact your daily life, items that you think your doctor should ask you about when evaluating the effectiveness of your arthritis medication or treatment. For example, pain at night or difficulties getting out of the car. Please list them in order starting with the most important.

The response time for this question was under 10 minutes. The sample size was 20 respondents. The responses confirmed many items that are already included in the WOMAC (hip/knee) and AUSCAN© Questionnaire pain and disability scales but also identified additional items that may reflect more contemporary concerns. The responses were:

- Walking on flat surface
- Going up/down stairs
- Lying in bed
- Sitting/lying
- Standing upright
- Getting out of chair
- Bending
- Getting in/out of car
- Shopping
- Putting socks/stockings on/off
- Getting out of bed
- Getting in/out of bath
- Toileting
- Heavy domestic duties
- Light domestic duties

The respondents mentioned several additional items that do not appear on the WOMAC or AUSCAN scale:

- Texting
- Working/typing on the computer
- Pain in hands/lack of finger strength
- Difficulties at work
- Challenges with sexual relations
- Achiness with changing weather
- Playing with grandkids
- Negative impact on spouse/partner
- Drowsiness from pain medication
- Problem driving

**Question 2:** Think back to when your current treatment was initiated for your disease. How clearly did your physician describe the degree of improvement you could expect from the treatment you were given?
The response time for this question was 20 minutes. The sample size was 25 respondents. In response to this question, fewer than half of the sample respondents reported that their physician outlined clear expectations of the improvement they could expect. The responses were:

- 48% of respondents answered that the physician gave very clear expectations
- 36% of respondents answered that the physician gave some general expectations
- 16% of respondents answered that physician gave unclear/vague or no information

DEVELOPING CULTURAL AND LINGUISTIC COMPETENCE

A primary concern of patient-centered CER is to eliminate health disparities and provide equitable high-quality healthcare to all patients. Research has shown that minority Americans, compared with white Americans, experience poorer health outcomes from preventable and treatable conditions (IOM, 2002). Some of the reasons are socioeconomic factors and lack of access (e.g., no insurance coverage), which affect minority Americans more often than white Americans. Health disparities also exist at the level of healthcare, with minority Americans receiving a lesser quality of care, which is exemplified by research showing that minority Americans are less frequently referred to specialist treatments for diagnostic and therapeutic procedures and the use of preventative services (e.g., immunization, mammography) (Betancourt, 2006).

The three most common barriers to equitable healthcare are: poor communication between the provider and the patient; biases and stereotypes that affect clinical decision making; and patient mistrust (Betancourt et al., 2003). These factors will need to be considered in the design of an engagement process for the generation of research topics. A culturally and linguistically competent engagement process:

- Develops culturally and linguistically competent organizational structures.
- Involves culturally and linguistically diverse populations by actively reaching out to underserved populations.
- Invites and empowers public participants to participate as active partners using culturally and linguistically appropriate methods of engagement.
- Includes health disparities research data to address unmet research needs.

CULTURAL AND LINGUISTIC COMPETENCE AT THE INSTITUTIONAL LEVEL

There are many definitions of cultural competence at the organizational and institutional level. The National Center for Cultural Competence (NCCC) in the United States defines cultural and linguistic competence for organizations as those that (NCCC, 2012; based on Cross et al., 1989):

- “Have a defined set of values and principles, and demonstrate behaviors, attitude, policies, and structures that enable them work effectively cross-culturally.
- Have the capacity to (1) value diversity, (2) conduct self-assessment, (3) manage the dynamics of difference, (4) acquire and institutionalize cultural knowledge, and (5) adapt to the diversity and cultural contexts of the individuals, families, and communities they serve.
- Incorporate the above in all aspects of policy making, administration, practice, service delivery, and systematically involve consumers, families, and communities.”

In March 2001, the U.S. Department of Health and Human Services Office of Minority Health (OMH) developed national standards for culturally and linguistically appropriate services (CLAS) with the goal that this guideline
will improve access to healthcare, improve the quality of care, and ultimately improve health outcomes (OMH, 2001). The CLAS standards include views from a broad range of stakeholders, including patient advocates, advocacy groups, consumers, educators, healthcare organizations, healthcare providers, professional associations, policy makers, purchasers of healthcare, and accreditation and credentialing agencies (OMH, 2001). This guideline contains 14 standards for culturally competent care, language access services, and organizational supports. An enhanced set of standards is in preparation and will be published in early 2012.

Several states now require cultural and linguistic competence training (MCCG, 2012a; MCCG, 2012b) and the law requires that federal agencies use plain language that is consistent and easy to understand for the public (Plain Language.gov, 2012a). To fulfill these national and state requirements, it is essential that patient-centered research organizations base their organizational structures and services on the principles of cultural and linguistic competency.

RESOURCES TO EVALUATE AND DEVELOP CULTURAL AND LINGUISTIC COMPETENCE

A number of resources exist to help organizations make their processes compliant with national standards and to continuously improve the cultural and linguistic competence of their leadership and staff. The Office of Minority Health website features resources on cultural and linguistic competence (OMH, 2012).

Other resources are:

- The Cultural and Linguistic Competence Policy Assessment Instrument, which is a self-assessment instrument for organizations (NCCC, 2006).
- A checklist to conduct culturally and linguistically competent meetings and conferences (NCCC, 2000).
- Plain language training resources are available on the Plain Language Action and Information Network website (Plain Language.gov, 2012b).
- Guidance documents on cultural competence and patient-centeredness in healthcare published by The Commonwealth Fund (Beach et al., 2006).
- A series of online e-learning solutions on cross-cultural healthcare developed by the Manhattan Cross Cultural Group (MCCG) (MCCG, 2012c).
- Yearly conferences and an online peer-learning and peer-networking platform offered by the Diversity Rx organization (Diversity Rx, 2012).

INTERNATIONAL RESOURCES FOR INVOLVING THE PUBLIC IN HEALTHCARE RESEARCH

International resources for engaging diverse populations in healthcare research are available from the Australian Commission on Safety and Quality in Health Care (ACSQHC), the Scottish Health Council (SCH), and the European Patients’ Forum (EPF). ACSQHC published a discussion paper on consumer participation and culturally and linguistically diverse communities (Romios et al., 2007). SCH created an equality and diversity tool kit, which is available on their website (SCH, 2012a; SCH, 2012b). EPF issued a guide on the young patient perspective (EPF, 2011).

HEALTH DISPARITIES RESEARCH

An engagement process for generating topics for PCOR CER should include data from health disparities research as an additional resource to identify gaps in research. Health disparities research data are available
from the peer-reviewed medical literature and, among others, the Centers for Disease Control and Prevention (CDC, 2011) and the National Cancer Institute, which also offers a health disparities calculator (SEER, 2011).

USE OF CULTURALLY AND LINGUISTICALLY APPROPRIATE METHODS OF ENGAGEMENT

The methods covered in this white paper have been used in the United States for more than 40 years. Some, such as focus groups, have been developed in the United States and have been successfully employed in diverse populations. These methods are used worldwide and are an integral part of social science research. We propose that PCORI actively reaches out to involve heterogeneous populations in these methods and processes. We also encourage the use of various methods, as the public may have different preferences for engaging in the process of topic generation and should be allowed to choose methods that are meaningful to them. Furthermore, innovative methods such as photovoice and “crowdsourcing” can increase diversity and demographic reach. We also recommend using online approaches and virtual communication techniques. In addition, we recommend reaching out to already existing community-based participatory research organizations.

The patient-clinician partnership, as described by the James Lind Alliance includes a phase of peer-consultation that serves to: (1) empower the public members of the group; (2) increase heterogeneity by reaching out to the community and using diverse consultation techniques; (3) provide room for innovation, using novel techniques of engagement. The peer-consultation phase makes this model of engagement particularly suitable for the United States because it permits engaging many individuals to choose methods that are meaningful to them. The moderating technique used in the decision-making workshop (the nominal group technique) was developed in the United States in the 1970s and has since diffused internationally and is one of the most commonly used consensus-making techniques (Delbecq and Van de Ven, 1971). This method is therefore fully transferable to the United States, as it actually originated here for the specific purpose of giving an equal voice to each member of a diverse group. The consultation phase provides an opportunity to engage diverse groups throughout the United States using different types of methods, permitting the public to choose a method that is meaningful to them.

Regardless of the method that is being employed “it is the quality of the relationships that underpins the quality of participatory research and any techniques that are used” (Springett et al., 2011). Therefore, it is essential that the individuals facilitating public engagement processes are skilled in using social science research strategies and methods, and that the engagement team itself is trained in these methods.

QUALITATIVE RESEARCH RESOURCES IN THE UNITED STATES

Qualitative research organization provides moderating services throughout the United States. Of special interest are individuals and organizations providing cross-cultural moderation services. Moderators need to be self aware, provide a comfortable ambiance conducive to engaging diverse groups, have respect and empathy, and keep the discussion moving toward the common goal.

One example is

- Asher Consulting ([www.asherconsult.com](http://www.asherconsult.com))

This agency specializes in healthcare and provides social science services to universities.

Some consultants also provide moderating services to the pharmaceutical and device industries, which already elicits the public voice for research and marketing purposes, and should therefore reveal any potential conflict of interest.
FACILITATORS AND BARRIERS TO ELICITING THE PUBLIC’S VOICE

Recent systematic reviews identified barriers and facilitators of involving the public as active partners in healthcare research (Faulkner, 2010; Saunders and Girgis, 2010; Gagnon et al., 2011; Légaré et al., 2011; Patient Partner, 2012b). The most common barriers were:

- Discrepancy between perspectives of experts and patients/public members
- Dilemmas surrounding identity and power
- Lack of adequate organizational resources
- Difficulty in recruiting patients or representatives of the public
- Patients and members of the public not sufficiently diverse
- Public participants’ lack of familiarity with scientific and medical terminology

To overcome these barriers, the authors recommended these facilitators to public participation (Faulkner, 2010; Saunders and Girgis, 2010; Légaré et al., 2011; Patient Partner, 2012b):

- Creating an organizational structure and providing resources that facilitate patient involvement (appropriate funding, coordinators, patient-centered process, cultural competence).
- Providing participant training (training days and seminars to familiarize the public with technical matters and appraisal skills).
- Supporting participants before, during, and after the involvement (welcome pack, mentoring, telephone and e-mail assistance, analysis grid for knowledge synthesis).
- Communicating clear expectations of the process at the beginning of the partnership (e.g., who is involved and what role they are expected to fill, disclosure of the funds available, and specification of the time commitment expected).
- Involving a group rather than single participants.
- Empowering patients by using processes that give an equal voice to professional and lay participants (e.g., nominal group technique for moderating discussions).
- Providing networking opportunities between members of the public currently involved and those who had previously been involved.

FUTURE RESEARCH

There is a need to evaluate novel methods of engagement, especially Internet-based approaches and visual methods such as photovoice, to generate topics for PCOR and CER, and to measure their impact on healthcare research. The results of the impact of public involvement on research, health outcomes, and patient satisfaction should be evaluated on a regular basis to ensure the integrity and validity of the process, which a recent study suggests is feasible (Barber et al., 2011). The recently established Collaboration on Participatory Health Research (ICPHR) is developing guidelines for conducting and evaluating participatory health research, similar to the Cochrane Collaboration, which could accelerate the development and evaluation of engagement methods and processes and health experience research in the near future (Wright et al., 2010; IIICPJR, 2012).

CONCLUSIONS

Various methods and processes exist to elicit the public’s voice in the generation of PCOR research topics. The most common formats are one-on-one and focus group interviews, where unstructured interviews using open-ended questions provide the most in-depth information. The best-researched approach of a collaboration of the public with clinicians and researchers is a patient-clinician partnership process developed by the James Lind Alliance (JLA). This process is a sustainable collaboration in which the public and clinicians consult with their
peers to make sure the process covers diverse views. The process also uses scientific research data from systematic reviews to generate topics and to avoid duplication of research. The public members are empowered by the use of nominal group technique to moderate the discussion.

Based on the review of the literature and best practices, public engagements should use a theory-based approach, researchers and moderators should be skilled in using and interpreting social science methods, and the engagement process should use more than one method to elicit sufficiently diverse viewpoints and appropriate detail. The process should also involve researchers who are skilled in translating the information received from the public into a research project. The use of data from health experience research is encouraged to provide sufficient detail of the patient experience, combined with systematic CER reviews to avoid the duplication of research, and a health disparities analysis to ensure that the needs of underserved populations are addressed.

**SUGGESTED PROCESS OF ENGAGEMENT**

Based on the best practices and case studies found in the peer-reviewed literature, we propose an engagement process that uses public-clinician partnerships to generate a concise list of specific research questions, research topics, or both, that can then enter the PCORI research prioritization and funding process (see Figure 5). This engagement strategy is based on the JLA approach but has been modified to be used in the context of a PCORI methodology framework and the United States healthcare paradigm. The strategy expands on the JLA approach to elicit a broad and diverse engagement with sufficient depth of information, introduces data from health experience research to provide more detailed information, and employs a health disparities analysis to strengthen the perspective of underserved populations.

At the beginning of the process (phase 1), a PCORI research advisory board that also includes public representatives invites lay and clinician members to participate in the partnership. During an opening meeting, a moderator, skilled in moderating diverse groups, uses various activities to introduce participants to each other and create cohesiveness. The goal of the partnerships is explained and the roles of the participants are clearly defined.

During phase 2, the partnership generates research ideas. During this phase, each group consults with their peers to include broader and more diverse viewpoints. Each group may choose their own method(s) as long as these are valid and observe good research practice (for example, one-on-one interviews or focus groups). The use of a diverse set of strategies, methods, and tools is encouraged to engage a population that reflects the heterogeneity of the U.S. demographics. We encourage involving already existing community-based participatory research partnerships. Ideally, social scientists support this process. The groups also use health experience research to add sufficient depth to the public perspective, and employ health disparities analysis to address the health concerns of underserved populations. Evidence from the peer-reviewed literature (systematic CER reviews) can also be used by the groups to identify research gaps.

During phase 3, the partnership combines the ideas from each group, the systematic reviews, and the health experience research and health disparities analysis, and organizes them into research themes. During this process, questions that have already been addressed in the literature are taken off the list, and duplicates are merged into one theme.

During phase 4, the partnership meets at a workshop and translates the research themes into research questions. The group then uses a simple process for prioritizing topics. For example, the research topics are listed on a pin board and each participant places one to three adhesive labels (“sticky dots”) on their priorities. The process should use moderators skilled in engaging diverse groups, and social science researchers skilled in
interpretative methods should assist the public and clinicians in organizing, synthesizing, and interpreting the results and translating the research themes into precise research topics and questions.

In phase 5, the partnership publishes the pre-prioritized research questions/topics and submits these to PCORI with all of the documentation. Feedback is obtained from all participants and is used to improve the process of engagement. The research themes or questions can then enter the PCORI research prioritization process using validated methods such as peer review, conjoint analysis, or discreet choice (Bridges et al., 2008; Mühlbacher and Nübling, 2010; Viergever et al., 2010; Watson et al., 2011).

In phase 6, the partnership (or PCORI) evaluates the impact of this process on PCOR and CER before a new cycle of engagement is started.

Figure 5. Suggested PCORI Process for Engaging the Public to Generate and Select Topics for PCOR and CER


Hancock B. Trent Focus for Research and Development in Primary Health Care: An Introduction to Qualitative Research. Trent Focus, 1998 (updated 2002).


APPENDIX A. QUALITY CRITERIA FOR METHODS OF PATIENT INVOLVEMENT

1. Does the methodology include a process for inviting and selecting patients or patient advocate groups to participate in the PCOR or CER project?
2. Can the methodology be used in a wide variety of settings and for diverse conditions and populations?
3. Does the methodology empower patients to become active, respected participants in the PCOR or CER project?
4. Is there a process in which the patient’s opinions, preferences, concerns, and expectations are prioritized and translated into a well-defined research topic?
5. Has the methodology been validated?
6. Are there measures for quality control of patient participation to ensure that the integrity of the process of patient involvement is maintained over time and across different projects?
7. Does the methodology consider practical aspects of diffusion and implementation?
8. Was the development of the methodology transparent, did it include patients and other stakeholders, and did it include a conflict-of-interest statement?
9. Can the methodology be adapted to the requirements of the United States healthcare system?
10. Can the methodology be integrated into a larger framework of informing the setting of priorities in research at PCORI?

This quality checklist was created using items that specifically apply to PCORI and the United States healthcare system combined with selected items adapted from the Method Report for Patient Involvement of the German Agency for Quality in Medicine (AQuMED) and the guideline for patient involvement of the International Alliance of Patients’ Organizations (IAPO).
APPENDIX B. DESCRIPTION OF INTERNATIONAL ORGANIZATIONS INVOLVED IN PCOR

The organizations that are listed in the following sections provide comprehensive documents on principles of patient engagement in healthcare research and patient-centered care. Because these were the most comprehensive documents found during the environmental scan, these were used to provide a concise synthesis of principles of patient engagement. The organizations are presented in alphabetical order.

European League Against Rheumatism (EULAR)

EULAR is a pan-European organization in Zurich, Switzerland, representing the patient, scientific, and health professional rheumatology societies of all European nations (EULAR, 2012). EULAR recently published recommendations for patient involvement in scientific projects (de Wit et al., 2011).

European Patients’ Forum (EPF)

EPF is the umbrella organization for 51 European patient organizations across the 27 European Union Member States. Founded in 2003, EPF represents the views of an estimated 150 million European patients and their families and caregivers. EPF advocates the meaningful involvement of patients in all health-related projects, including clinical research and health policy design, to ensure that the public’s voice is included in all healthcare developments. EPF recently issued a response to the EU directive on clinical trials, providing specific recommendations regarding the involvement of the public in clinical trials (EPF, 2011).

German Agency for Quality in Medicine (AQuMed) (Ärztliche Zentrum für Qualität in der Medizin)

AQuMed is a not-for-profit organization established in 1995 by the German Medical Association and the National Association of Statutory Health Insurance Physicians. AQuMed coordinates healthcare quality programs, including patient empowerment, medical guidelines, evidence-based medicine, patient safety programs, and quality management (AQuMed, 2011b).

International Alliance of Patients’ Organizations (IAPO)

IAPO is an international not-for-profit organization that represents patients of all nationalities across all disease areas and has been promoting patient-centered healthcare around the world for more than 10 years. The members of IAPO are patients’ organizations working at the international, regional, national, and local levels to represent and support patients, their families, and caregivers. IAPO’s vision is that patients throughout the world are at the center of healthcare.

INVOLVE (United Kingdom)

INVOLVE is a national advisory group in the UK that was established in 1996 to promote public involvement in research with the goal “to improve the way that research is prioritized, commissioned, undertaken, communicated, and used” (INVOLVE, 2012a). INVOLVE is part of the National Institute for Health Research, which also funds the initiative. This group was established in 1996 under name of “Consumers in NHS Research.” In July 2003, the group changed its name to “INVOLVE – promoting public involvement in NHS, public health and social care research” to better reflect its growing mission. INVOLVE maintains an online database (People in Research) listing opportunities for the public to be involved in research (INVOLVE, 2012b).

INVOLVE has issued a large number of guidance documents and case studies. Examples include:

- Good practice in active public involvement in research (INVOLVE, 2009)
- Changing our Worlds: Examples of user-controlled research in action (Faulkner, 2010)
**Patient Partner**

Patient Partner, also called Patients Partnering in Clinical Research, is a European project aimed at identifying the needs for patients as partners in clinical research. Patient Partner is a joint effort of four organizations: the European Forum for Good Clinical Practice (EFGCP), European Genetic Alliances Network (EGAN), Genetic Alliance UK (formerly, Genetic Interest Group), and the Dutch Genetic Alliance (VSOP).

The European Commission funded Patient Partner. The project began in May 2008 and was completed in May 2011. At the present time, Patient Partner is developing an online forum, European Network of Patients Partnering in Clinical Research (ENPCR), which will serve as a platform for exchange. Patient Partner has issued policy recommendations for patient organizations and sponsors of clinical research (Patient Partner, 2012a; Patient Partner, 2012b).

**Value+**

Value+ is a collaboration of various partner organizations of which the European Patients’ Forum (EPF) is the leading coordinator. The partner organizations are European patients’ organizations, national patients’ platforms, and a research firm with expertise in patients’ advocacy and health-related awareness-raising, research and policy. Value+ has developed a guidance document on the ethical principles of research partnerships with the public. It has issued a number of guidance documents such as “Ethical principles of partnership” (EPF, 2011).
APPENDIX C. LIST OF INTERNATIONAL ORGANIZATIONS INVOLVED IN PCOR

The following is a small selection of international organization involved in PCOR that does not intend to be all-inclusive. The countries/continents are presented in alphabetical order.

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<th>INTERNATIONAL</th>
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<td>International Alliance of Patients’ Organizations (IAPO)</td>
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<td><a href="http://www.patientsorganizations.org">http://www.patientsorganizations.org</a></td>
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<td>World Health Organization (WHO)</td>
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<td><a href="http://www.who.int/en/">http://www.who.int/en/</a></td>
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<td>International Collaboration on Participatory Health Research (ICPHR)</td>
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<td>Australian Commission on Safety and Quality in Health Care (ACSQHC)</td>
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<td>Health Issues Centre</td>
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<td>Canadian Institutes of Health Research (CIHR) – Strategy for Patient-Oriented Research</td>
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<td>Health Canada – Office of Consumer and Public Involvement</td>
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<td>European Food Safety Authority (EFSA)</td>
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<td><a href="http://www.efsa.europa.eu/">http://www.efsa.europa.eu/</a></td>
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<tr>
<td>European League Against Rheumatism (EULAR)</td>
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<td><a href="http://www.eular.org/">http://www.eular.org/</a></td>
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<tr>
<td>European Medicines Agency (EMA) – Patients’ and Consumers’ Working Party</td>
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<td><a href="http://www.ema.europa.eu">www.ema.europa.eu</a></td>
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<tr>
<td>European Network for Health Technology Assessment (EUnetHTA)</td>
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<td><a href="http://www.eunethta.eu/">http://www.eunethta.eu/</a></td>
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<tr>
<td>The European Network of Rare Diseases (EURORDIS)</td>
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http://www.eurordis.org/
European Patients’ Forum
http://www.eu-patient.eu/
Patient Partner
http://www.patientpartner-europe.eu
Picker Institute Europe
http://www.investinengagement.info/

FRANCE
Collectif inter associatif Sur la Santé (CISS)
http://www.leciss.org/
National French Health Authority (Haute Autorité de Santé, HAS)
http://www.has-sante.fr

GERMANY
German Agency for Quality in Medicine (AQUMED) (Ärztliche Zentrum für Qualität in der Medizin, ÄZQ)
http://www.aezq.de/
German Network for Evidence-based Medicine (Deutsches Netzwerk Evidenzbasierte Medizin)
http://www.ebm-netzwerk.de
Patient als Partner
http://www.patient-als-partner.de/

NETHERLANDS
Federation of Patients and Consumer Organizations in the Netherlands
http://www.npcf.nl/

SPAIN
Foro Español de Pacientes
http://www.webpacientes.org/fep/

SWITZERLAND
Patient Platform
http://patienten.ch
Swiss Clinical Trial Organization (SCTO)
http://www.scto.ch/de/Portrait.html

UNITED KINGDOM
British Medical Association
http://www.bma.org.uk/patients_public/ppgintro.jsp?page=1
CETL4Health North East
http://www.cetl4healthne.ac.uk/view
Diabetes Research Network

INVOLVE
www.invo.org.uk

James Lind Alliance
http://www.lindalliance.org/Introduction.asp

National Children’s Bureau (NCB) Research Centre
www.ncb.org.uk/research

National Health Service (NHS)
http://www.institute.nhs.uk

National Institute for Health and Clinical Excellence (NICE)
http://www.nice.org.uk/

Participation Works (children and young people)
www.participationworks.org.uk

The PEAR project
www.ncb.org.uk/PEAR

UNITED STATES OF AMERICA

Agency for Healthcare Research and Quality (AHRQ)
http://www.effectivehealthcare.ahrq.gov/

Center for Medical Technology and Policy (CMPT)
http://www.cmtpnet.org/

The Commonwealth Fund
http://www.commonwealthfund.org/

The Detroit Community-Academic Urban Research Center
http://www.detroiturc.org/about-cbpr/cbpr-principles.html

Harvard Medical School Center of Excellence in Minority Health and Health Disparities
http://www.mfdp.med.harvard.edu/coe/

Institute of Medicine (IOM)
http://www.iom.edu/

National Center for Cultural Competence (NCCC)
http://www.clcpa.info/

National Institutes of Health (NIH) – Patient Reported Outcomes Measurement Information System (PROMIS)
http://www.nihpromis.org/default.aspx

Patient-Centered Outcomes Research Institute (PCORI)
http://www.pcori.org

Picker Institute USA
http://pickerinstitute.org/

The Urban Institute
http://www.urban.org/
APPENDIX B. SEARCH STRATEGIES

SEARCH OF THE PEER-REVIEWED LITERATURE

The time frame for the search of the peer-reviewed medical and psychology literature ranged from January 2006 to December 2011. However, earlier articles were also included if they provided a specific example of a method or provided historical context for processes, methods, and developments. The search focused primarily on systematic reviews and meta-analyses, and articles investigating a specific method or process of public engagement were also included. The primary focus was on evidence for public engagement for generating topics in healthcare research. The secondary focus was to retrieve guidelines for principles of patient engagement in patient-centered outcomes and comparative effectiveness research, development of clinical guidelines and health technology assessments that could be applied to processes of public engagement in generating and prioritizing healthcare research topics.

The search of the peer-reviewed literature used three databases:

- MEDLINE
- Embase
- PsycINFO

The specific search terms are detailed in the following sections.

MEDLINE

The search of the MEDLINE database used the following MeSH terms:

- (consumer participation OR patient participation) AND (research design OR advisory committees OR decision making, organizational)
- comparative effectiveness research

In addition to these MeSH terms, the following key words were used. Each line was combined with “OR”, separate lines were combined with “AND”:

- caregiver, carer, client, community, consumer, lay member, lay person, patient, public, service-user, user
- collabor*, engage*, eliciting, involve*, participat*
- generating topics, topic generation, prioritiz*, selecting topics, topic selection, voice

In order to search for specific methods of engagement, individual searches were performed using the following key words:

- advisory board, advisory committee, citizen board, citizen council, citizen jury, focus group, graphic elicitation, grounded theory, ethnography, health experience research, mixed methods research, interview, participatory action research, participatory health research, observation, phenomenology, photovoice, photo elicitation, patient board, patient council, patient panel, people board, people council, people panel, public board, public council, public panel, questionnaire, social science methods, survey, verbal elicitation, visual elicitation

OVID EMBASE
Subject headings:

- decision making
- health care management
- health care planning
- health care policy
- health care quality
- participatory management

In addition to these MeSH terms, the following key words were used. Each line was combined with “OR”, separate lines were combined with “AND”:

- caregiver, carer, client, community, consumer, lay member, lay person, patient, public, service-user, user
- collabor*, engage*, eliciting, involve*, participat*
- generating topics, topic generation, prioritiz*, selecting topics, topic selection, voice

In order to search for specific methods of engagement, individual searchers were performed using the following key words:

- advisory board, advisory committee, citizen board, citizen council, citizen jury, focus group, graphic elicitation, grounded theory, ethnography, health experience research, mixed methods research, interview, participatory action research, participatory health research, observation, phenomenology, photovoice, photo elicitation, patient board, patient council, patient panel, people board, people council, people panel, public board, public council, public panel, questionnaire, social science methods, survey, verbal elicitation, visual elicitation

OVID PSYCINFO

The search of the OVID PsycINFO database used the following terms as abstract, title, and keywords. Each line was combined using “OR,” separate lines were combined using “AND.”

- carer, caregiver, client, consumer, community, patient, public, service-user, user
- collaborat*, engage*, inclus*, involve*, participat*
- comparative effectiveness research, guideline development, healthcare research, health policy, health technology assessment, relative effectiveness research, patient-centered healthcare research

In addition, the following keywords were used in separate searches:

- advisory board, advisory committee, citizen board, citizen council, citizen jury, focus group, graphic elicitation, grounded theory, ethnography, health experience research, mixed methods research, interview, participatory action research, participatory health research, observation, phenomenology, photovoice, photo elicitation, patient board, patient council, patient panel, people board, people council, people panel, public board, public council, public panel, questionnaire, social science methods, survey, verbal elicitation, visual elicitation

ENVIRONMENTAL SCAN

In addition, an environmental search of the Internet was performed to identify organizations dedicated to patient-centered healthcare and patient-centered healthcare research. The following search engines were used:
ENGLISH LANGUAGE KEYWORDS

The following English-language keywords were used combined with “OR” for each line, and combined with “AND” to combine lines:

- carer, caregiver, client, community, consumer, patient, public, service-user, user
- consulting, consultation, collaborating, collaboration, eliciting, engaging, engagement, including, inclusion, involving, involvement, participating, participation
- opinion, perception, perspective, view, voice
- comparative effectiveness research, guideline development, healthcare research, health technology assessment, health policy, research design, relative effectiveness research, research topics

Individual searches were performed for each of the following search terms:

- advisory board, advisory committee, citizen board, citizen council, citizen jury, focus group, graphic elicitation, grounded theory, ethnography, health experience research, mixed methods research, interview, participatory action research, participatory health research, observation, phenomenology, photovoice, photo elicitation, patient board, patient council, patient panel, people board, people council, people panel, public board, public council, public panel, questionnaire, social science methods, survey, verbal elicitation, visual elicitation

FOREIGN LANGUAGE KEYWORDS

The following foreign language terms were also used, each line was combined with “OR,” different lines were combined with “AND.”

**German:**

- Patienten, Pfleger, Konsumenten, Öffentlichkeit
- Auswahl, auswählen, Prioritäten setzen, Teilhabe, teilhaben
- Versorgungsrichtlinien, Forschungsthemen, Patientenorientierte Forschung, patientenorientierte Medizin, Patientenkompetenz, Personalisierte Medizin, Partizipative Entscheidungsfindung

**French:**

- Consommateurs, patients, publique
- choisir, choix, consultation, consulter, sélection, sélectionner, participation, participer
- la décision médicale partagée, recherche clinique orientée patient, soins orientée patient, sujets de recherche
### APPENDIX E. TABULAR SUMMARY OF PROCESS APPRAISAL

<table>
<thead>
<tr>
<th>Process for generating research topics</th>
<th>Q1</th>
<th>Q2</th>
<th>Q3</th>
<th>Q4</th>
<th>Q5</th>
<th>Q6</th>
<th>Q7</th>
<th>Q8</th>
<th>Q9</th>
<th>Q10</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Suggested PCORI process</strong></td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>partially</td>
<td>yes</td>
<td>yes</td>
</tr>
<tr>
<td><strong>JLA process</strong></td>
<td>yes</td>
<td>Some limitations</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>yes</td>
<td>Yes, it is a process designed for diverse groups and includes a process for reaching out into the community</td>
<td>Yes, with some modifications</td>
<td></td>
</tr>
<tr>
<td><strong>Effective Health Care (EHC) Program (AHRQ)</strong></td>
<td>no</td>
<td>no</td>
<td>no</td>
<td>?</td>
<td>no</td>
<td>no</td>
<td>partially</td>
<td>?</td>
<td>Does not reach out into the community, may exclude underserved populations</td>
<td>As part of a consultation process</td>
</tr>
</tbody>
</table>

The JLA process can serve as a model for PCORI to develop a validated process for public engagement in the generation and selection of CER topics that can be integrated into a larger framework of informing the setting of research priorities at PCORI. PCORI aims to further improve on this process and to include elements required by the U.S. healthcare system. These additional elements are:

1. The use of health experience research data to increase the depth of experiential knowledge
2. Health disparities analyses to address the needs of underserved populations
3. The use of systematic CER to generate additional topics by identifying gaps in research and to ensure that research is not duplicated

In addition, we recommend involving already existing community-based participatory research partnerships in both, the collaborative and the consultation phases, to ensure that members of underserved populations are empowered to actively participate in the process.
APPENDIX F. TYPOLOGIES OF PUBLIC ENGAGEMENT

Hierarchical typology indicating increasing levels of public control by Arnstein (1969)

Typology of public engagement based on the flow of information by Rowe and Frewer (2005)

Public Communication

Sponsor → Public Representatives

Public Consultation

Sponsor ← Public Representatives

Public Participation
Typology matrix based on the degree of researcher involvement versus the degree of public involvement by Oliver et al. (2008)

This typology was created to describe and categorize different possible scenarios of consumer involvement. It describes the following hypothetical scenarios of public engagement:

- **Type H**: The public is in control of research. Researchers or clinicians only participate when invited by the public.
- **Type G**: The public does not participate in the research. Investigators respond to lay action by providing information.
- **Types A and B**: Investigators invite groups of lay people for collaboration (Type A) or consultation (Type B)
- **Types C and D**: Investigators invite individual lay people for collaboration (Type C) or consultation (Type D)
- **Types E and F**: Investigators respond to the public request for collaboration (Type E) or consultation (Type F)

<table>
<thead>
<tr>
<th>Researcher degree of involvement</th>
<th>Degree of public involvement</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Control</td>
</tr>
<tr>
<td>Inviting lay groups</td>
<td>A</td>
</tr>
<tr>
<td>Inviting individual lay people</td>
<td>C</td>
</tr>
<tr>
<td>Responding to lay action</td>
<td>E</td>
</tr>
<tr>
<td>Minor partner or absent</td>
<td>H</td>
</tr>
</tbody>
</table>
### APPENDIX G. ANALYSIS OF THE EFFECTIVE HEALTH CARE PROGRAM FOR THE GENERATION OF RESEARCH TOPICS

<table>
<thead>
<tr>
<th>Agency/Organization</th>
<th>Description of process</th>
<th>Analysis of patient-centeredness</th>
</tr>
</thead>
</table>
| **Effective Health Care (EHC) Program (AHRQ, 2011)** | Stakeholder involvement program | • The program does not use an evidence-based structured process for engaging the public in the generation of research topics.  
• The program is not patient centered. Patients and lay members are only one of several stakeholders.  
• The stakeholder engagement guide is long and complex and there is no plain language summary.  
• The public is not actively engaged in decision making  
• EHC is a new program that does not yet have a proven, validated, peer-reviewed track record. |
|                     | Topic nomination sheet. The public may submit questions via the Internet or mail a topic nomination sheet. | • The topic nomination sheet is complex. A high level of functioning is required to fill in the form. This process excludes individuals with lower education, children, those with mental disabilities, immigrants, people who are unable to define a research topic, those without Internet access.  
• The topic nomination sheet asks for additional information that may deter some members of the public, especially underserved or vulnerable populations, to contribute because they may feel that they lack the level of expertise.  
• The role of the public is not sufficiently explained.  
• There is no active elicitation process to help people access issues that are important to them. |
|                     | In person-meeting by invitation for group discussion | • Group discussions involving diverse groups require moderating techniques that give each person an equal voice. The process for topic generation and moderating the discussion is not described. There is the potential that vulnerable populations are excluded, especially those who have difficulties expressing themselves verbally.  
• The public has to travel to the AHRQ location, which excludes individuals from rural areas and individuals who may be intimidated by government organizations, and people with disabilities who may not be able to leave the home. The process also may not be suitable for children or adolescents. |
| **Conclusion**      |                        | • The process does not use an evidence-based structured process for engaging the public in the generation of research topics. There is a strong possibility that underserved and vulnerable populations are not effectively engaged.  
• The process could, however, be a resource for research topics in the consultation phase of the PCORI process of engagement that we suggest in this white paper, provided that the topics submitted by the public are separated from those submitted by other stakeholders, |