Eliciting Patient Perspective in Patient-Centered Outcomes Research: A Meta Narrative Systematic Review.

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A report prepared for the Patient-Centered Outcomes Research Institute

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2012

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I Plain language summary

Compelling reasons, both practical and ethical, support engaging patients in healthcare research. Patients are the ultimate user of healthcare research findings and the most important stakeholder. Questions remain, however, about how to engage patients or their representatives (e.g., family members, caregivers, community representatives).

Patient-centered outcomes research means research that is informed by the perspectives, interests and values of patients throughout the research process, from the selection of research questions to the dissemination of research results. To summarize what is known about how to engage patients in healthcare research, we performed a comprehensive search and review of all pertinent studies. Our goal was to describe the practical steps needed to better engage patients in research. We also asked a group of patients without medical or research training to provide their perspective on our results.

Many studies have successfully engaged patients in all research steps: preparation of research (agenda formulation, funding procurement), execution of research (study conduct, data analyses) and the translation of results into action. We did not find studies comparing different ways to engage patients. We found that patient engagement is often feasible – although barriers exist at every research step. Patient engagement appears to enhance the quality of research (e.g., improving enrollment rates in studies, making studies more consistent with patients' values, goals, and preferences).

Overall, the available research suggests that patients could successfully play an active and engaged role in research, from participant, to collaborator, to partner.

II Executive Summary

Patient-Centered Outcomes Research requires that the patient's voice and perspective drive every step of the research process, including prioritizing the research questions, designing and conducting the research, and implementing the results in practice.

However, the best approach to select patients (or their surrogates, or caregivers) and the best methods for obtaining their feedback and incorporating it in research, remains unclear.

We conducted a systematic review and environmental scan to summarize the available evidence about patient engagement in healthcare research. We aimed to identify who are the relevant patients for engagement and how to recruit them, how can they engage, and how can their engagement result in changes in research design, conduct, analysis and dissemination.

II.1 Methods:

We conducted a systematic review of the published biomedical literature. A comprehensive search included multiple electronic databases, from their inception until November 2011. We sought studies of any design in any language and of any size, in which patients or their surrogates engaged to any extent in shaping the research agenda, study design and conduct, or dissemination and implementation. An environmental scan sought to identify unpublished sources or reports outside the field of medicine. We looked for actors and stakeholders, key events, documentation (white papers, position papers, proceedings of meetings) and future trends. We collated recommendations made by individual studies. Results were synthesized qualitatively following a meta-narrative review framework.

II.2 Results:

The systematic review identified 194 relevant references including 7 RCTs and 11 systematic reviews. The environmental scan identified relevant websites, organizations, forums, blogs, videos, associations, workshops, presentations, governmental agencies, abstracts and other forms of unpublished resources that spanned across various topics of health care and non-health care consumers' engagement in research.

II.2.1 Framework of patient engagement:

We found 34 studies that proposed a complete or partial model or framework for patient engagement. These models converged into 4 common iterative steps: patient (or surrogate) selection, building a reciprocal relationship, co-learning and feedback and reassessment/feedback. The steps are continuous and repeated until saturation occurs. Patient engagement was described as a spectrum that starts from passive role to the most engaged role (from study subject, to collaborator, to a researcher). Engagement was described through all study phases (preparatory, execution and translation phases). Figures 1-2 describe these frameworks. Evidence supporting these proposed models is observational and non-comparative.

II.2.2 Methods of selecting patients/surrogates:

Most studies did not clearly report how they selected their patient informants. However, the choice seemed to be based on self selection in most cases. A convenience sample was invited and those interested in participation were included. The environmental scan highlighted two other possibilities: disease specific social networks that most frequently were non-for-profit and established by patients or surrogates; and few disease-specific and non-disease specific organizations (mainly in Europe and Canada) focused on patient engagement and offer education and training to patients and facilitate their recruitment by researchers.

We did not find comparative studies to determine the relative efficacy of a particular method of identifying patient representatives. Hence, all of the described methods can be useful ways of engaging patients, but depend on the research questions being asked and the aims of the study.

II.2.3 Methods of obtaining informant voice:

Studies have described various methods **(Table 1)**. We did not find comparative studies to determine the relative efficacy of a particular method of identifying patient representatives. Hence, all of the described methods can be useful ways of engaging patients, but depend on the research questions being asked and the aims of the study.

II.2.4 Potential benefits:

Although engaging patients is justified for ethical and moral reasons (considering that they are the ultimate user of research evidence); we found ample evidence suggesting that this engagement may also improve study design (by choosing outcomes more meaningful to patients or designs that are more culturally sensitive or consistent with patients' context), execution (improving subject recruitment and retention) and translation (better implementation, dissemination and uptake).

II.2.5 Potential Barriers:

Barriers and challenges to each step in the process of conducting research are summarized after the detailed description of patient engagement in that process. In brief, the most commonly cited challenges related to logistics (extra time needed to complete research, time constraints of patients or surrogates, incremental funding needed for patient engagement) and an overarching worry of researchers and patients that this engagement may become tokenistic (e.g., done to fulfill a mandate or a checkbox) and not real. Another potential challenge that was described is related to scope creep. A theoretical concern is that involving patients in the research may lead to including other community concerns and issues that may make the research unfeasible.

Few studies described potential solutions that were mainly based on spending adequate time to build a reciprocal relationship (between the patient or community and researcher), mutual respect and clear expectations that are explicitly described and documented.

II.3 Limitations:

The main limitation to this systematic review is the non comparative and observational nature of the available literature. Therefore, we present empirical evidence of the beneficial impact of patient and public involvement in research and describe the methods used to select informants and incorporate their feedback in various research stages. However, inference regarding the best appropriate and effective methods is limited.

Another limitation relates to the lack of specific indexing terms in bibliographic databases for patient engagement and the lack of standardized and explicit reporting in published studies of how this process took place and what were its outcomes. Heterogeneity of study populations, methods and outcomes, constitute further limitations to extrapolation of evidence across settings. Publication and reporting biases have also likely impacted the conclusions of this report and their impact could not be estimated.

II.4 Conclusions and recommendations

We propose an overarching recommendation for early and frequent patient engagement in all research phases (preparatory, execution and translation); a potential framework; and a recommendation for various methods of selecting representatives and obtaining their feedback that were described as feasible in various settings. We were unable to favor a particular method due to the lack of comparative evidence.

We recommend future research on this important topic that aims at validating the frameworks proposed in the literature and compares the various methods of selecting patient representatives and engaging them in research.

Recommendations

Engaging patients and surrogates in all research phases (preparatory, execution and translation) is suggested and is feasible in most cases.

Potential risks for engaged patients seem to be minimal and outweighed by benefits:

- Patients’ engagement as the ultimate user of research evidence is ethically and morally compelling
- Possible improvement in study design (outcome and intervention selection)
- Possible improvement in study execution (subject recruitment and retention)
- Possibly higher quality evidence (lower risk of bias)
- More applicable research

We recommend a framework for engaging informants that includes:

a. Proper and wide representation of the study population
b. Building a reciprocal partnership between researchers and informants that includes mutual respect and explicit expectations
c. A co-learning process (where researchers learn from the informants and vice versa) to be done throughout the whole engagement process and maintained during the study
d. The involvement process should be continually evaluated using predefined tools and possibly by external evaluators

We suggest selecting representatives that are as similar as possible to the community or population in which the study results are intended to be applied. This includes relevant ethnic minorities, elderly, young, disabled, incarcerated and any other special or vulnerable populations impacted by the research. Empiric evidence exists to suggest that the engagement of all these categories of patients or their surrogates is feasible in most cases.

We suggest that patient and surrogate engagement be initiated as early as possible in the research project and as frequent as feasible.

We suggest that the choice of methods for selecting patients or engaging them in research be made based on the research questions being asked and the overall aims of the research. We were unable to recommend a preferred strategy due to the lack of comparative data.

II.5 Proposed frameworks of engagement:

Figure 1.

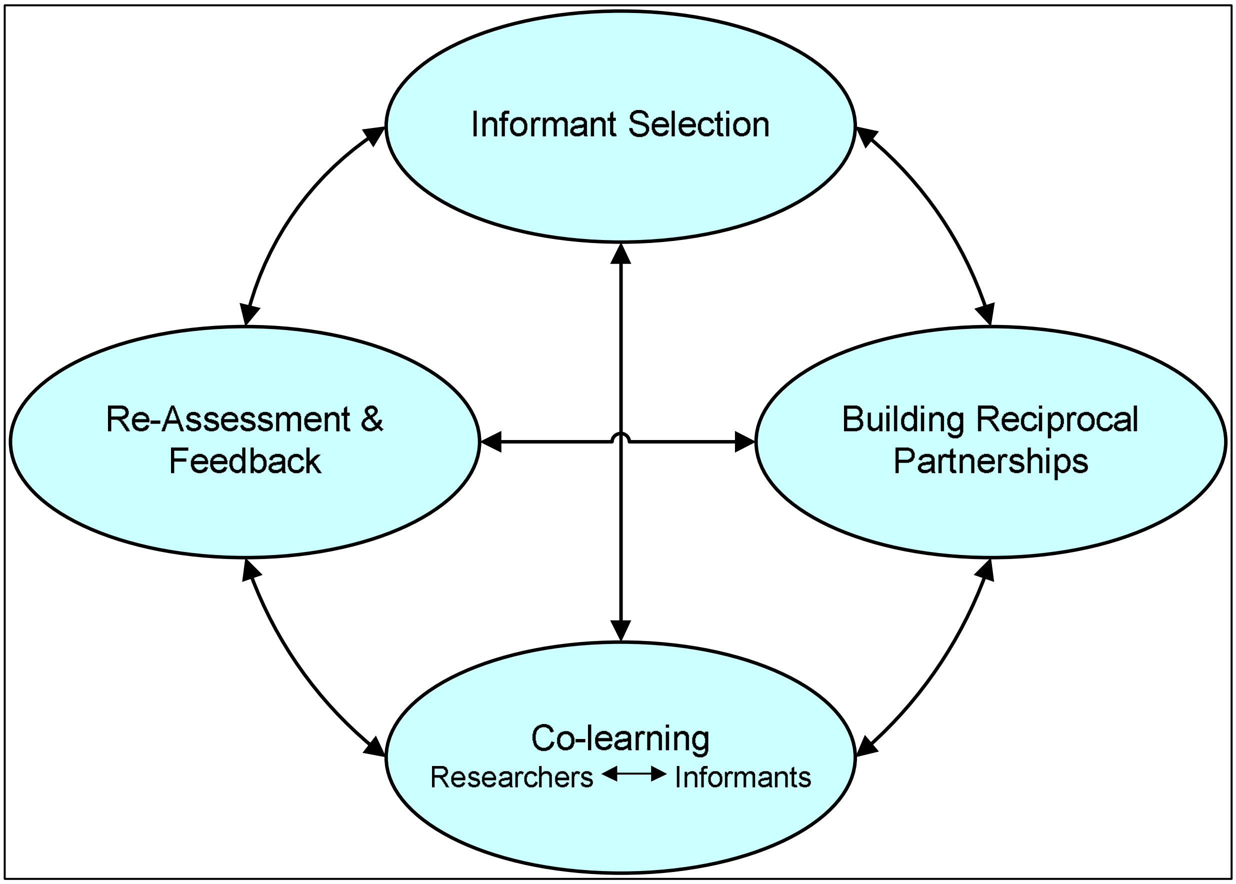
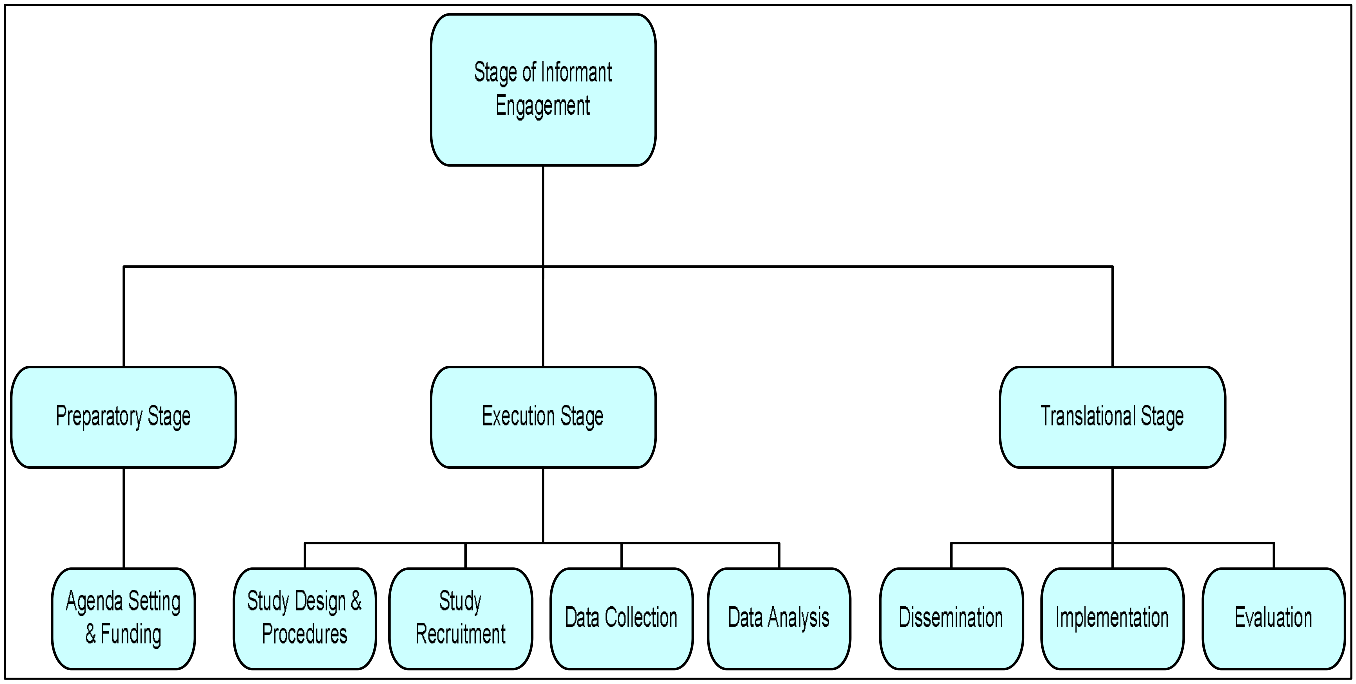
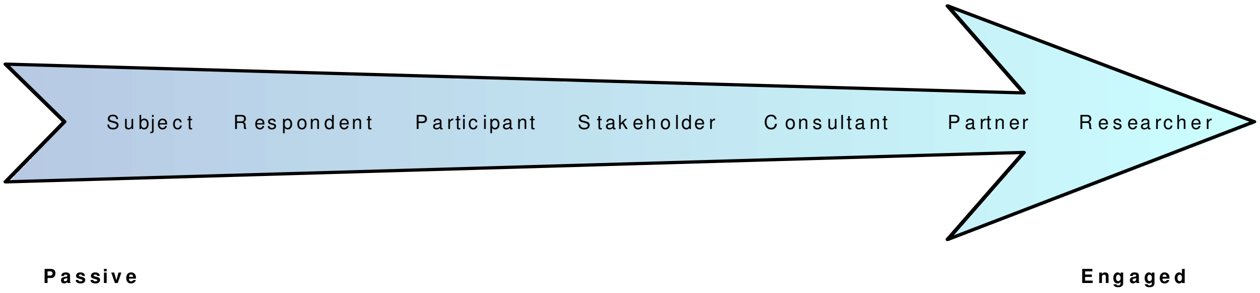


Figure 2 and Figure3





III Background

A defining principle of the Patient-Centered Outcomes Research Institute (PCORI) is ensuring that the patient's voice and perspective drive every step of the research process, including prioritizing the research questions, designing and conducting the research, and implementing the results in practice. Similarly, a growing drive is noted by patient movements, politicians, clinicians, scholars and researchers to increase the involvement of patients and other community members in the research process. Many rationales for such involvement are put forward including increased internal validity (by conducting better studies with higher rates of enrollment and retention), better outcome measurement that focuses on patient-important outcomes, increased external validity (more applicable and useful evidence) and an overarching, ethical mandate.

Nevertheless, there is no clear methodology to guide this process of patient engagement. Specifically,

- Whose opinion is relevant (categories of patients, surrogates, etc.)?
- How to find representatives of these categories of informants?
- How to obtain the information (the patient's voice or perspective)?
- How congruent is the perspective of other informants (clinicians and advocates) with the perspective of patients?
- What are the practical steps to transform the patient's voice into action (i.e., changes in research design, outcomes, interventions and comparators)?

Therefore, this systematic review and environmental scan aims at synthesizing the existing evidence about patient engagement in research. Proposed standards and recommendations from the existing evidence will constitute additional building blocks that would be added to research conducted by other groups under the auspices of PCORI to provide a framework for patient engagement processes.

IV Methods

IV.1 The systematic review:

A comprehensive systematic review of the published biomedical literature was conducted spanning across multiple data bases from their inception date to the present.

IV.2 Inclusion criteria:

We included studies of any design (randomized, observational, cross sectional, descriptive, qualitative, etc.), size or patient age or morbidity published in English language in which patients or their surrogates provided feedback regarding research agenda, outcomes or design. Non-original or summarized literature was also included (i.e., systematic or non-systematic literature reviews, opinion pieces such as commentaries, etc.).

IV.3 Literature search

The lead reference librarian at the Mayo Clinic, Rochester, with extensive expertise in conducting systematic reviews collaborated with study methodologists to develop a search strategy. Medical subject heading (MESH) terms and textwords are selected based on common indexing practices. Search terms are compiled and tested repeatedly to produce sensitive searches to capture potentially relevant publications.

The databases searched were:

1) PubMed/Ovid MEDLINE, Ovid EMBASE, Ovid PsycInfo, Ovid Cochrane - especially SysRev, Methods, and HTA, EBSCO CINAHL.
2) SCOPUS (indexes many potentially relevant journals in the social sciences, including sociology, economics, decision analysis, and communications).
3) Web of Science (multidisciplinary scientific content).
4) Business Search Premier, Academic Search Premier and Google Scholar (good resources for the communications, marketing, public opinion, and business literature that incorporate non-healthcare resources).

We also reviewed reference lists from eligible studies and conducted additional MEDLINE searches using the PubMed "related articles" feature for eligible studies and SciSearch for publications that cited eligible studies supplement the database search.

In **appendix 4**, we present the detailed search strategy. The strategy incorporates two approaches conducted simultaneously. The first approach focuses on the concept of incorporating the patient perspective in decision making processes (this was a more sensitive approach that may yield a larger number of studies that relate to decision making which is not necessarily the focus of this review but required for completeness of coverage) and the second approach is more specific (and less sensitive) and would directly address the issue at hand (patient engagement in research). We found the combination of both to be necessary to find the literature addressing the issue of eliciting and incorporating the patient's voice in research.

IV.4 Study selection

We collated initial references in citation files (using the software Endnote, Thompson ISI Research-soft), removed duplicates, and started a screening process for titles and abstracts against eligibility criteria. We performed this process using a Web-based reference management system (DistillerSR, Ottawa, Canada). This software allows instant monitoring of reviewers' progress and assessment of inter-reviewer agreement measures (real-time kappa). Studies were reviewed in duplicates until adequate agreement K>0.80 is achieved. Disagreements among reviewers in the initial abstract screening phase were automatically included. Potentially eligible studies were reviewed in full text following a similar procedure. We exclusively used electronic file formats (Portable Document Format, PDF) to maintain low cost and reduce paper usage. PDF files of full text articles were uploaded to the reference management system.

Disagreements among reviewers in the full text screening phase are reconciled by discussion, consensus or arbitration by a third reviewer.

IV.5 Data extraction

Data were extracted from included studies using standardized forms **(appendix 5)** developed based on the protocol and created in the web-based reference management system. Forms were tested on a sample of the included studies to assure better quality and performance.

Data extracted from studies included study description (e.g., demographics of participants and research setting), methods used to select informants and any measures for the validity or completeness of identifying informants, any measures of validity or accuracy of the information collected from participants (the patient's voice), description of methods used to implement/incorporate the patient's voice in research and any reported outcomes of patient engagement. We also captured authors' recommendations about the methods to be used for obtaining patient's voice and facilitating patient's engagement.

IV.6 Analysis

Meta-analysis was not conducted due to the observed heterogeneity in terms of study design (randomized, observational, cross sectional, etc.), informant type (healthy patients, patients with certain comorbidities related to study question, age, surrogate of patients, etc.), methods of eliciting the patient voice (survey, focus group, structured interview, etc.), methods of incorporating patient's voice (varying magnitudes of incorporation, outcome modification, study design change, changes in intervention or comparator, etc.). Therefore, data were presented qualitatively and we followed a meta-narrative approach suggested by

Greenhalgh et al [1]. As a first step, we reviewed studies that presented a framework or a conceptual model of the process and developed an inclusive model. The model describes the various steps in research (starting with agenda setting through execution of protocol and ends with implementation) and various levels of patient engagement (from passive engagement as a research subject to an active form of being a collaborator or researcher). From each study, we noted the conclusions and any potential recommendations about how to obtain and incorporate patient's voice. We also extracted any reported barriers or challenges to the process. Data elements extracted from each study are described in **appendix 5**. The overall approach of a meta-narrative review is summarized in Box.

IV.6.1 Phases in meta-narrative review*

(1) ***Planning phase***
(a) Assemble a multidisciplinary research team
(b) Outline the initial research question in a broad, open-ended format.
(c) Agree outputs with funder or client.
(d) Regular face-to-face review meetings including planned input from external peers the review.

(2) ***Search phase***
(a) Initial search led by intuition, informal networking and 'browsing', with a goal of mapping the diversity of perspectives and approaches.
(b) Search for seminal conceptual papers
(c) Search for empirical papers by electronic searching key databases, hand searching key journals and 'snowballing'

(3) ***Mapping phase***
Identify (separately for each research tradition):
(a) The key elements of the research paradigm (conceptual, theoretical, methodological and instrumental);
(b) The key actors and events
(c) The prevailing language and imagery

(4) ***Appraisal phase***
Using appropriate critical appraisal techniques:
(a) Evaluate each primary study for its validity and relevance to the review question
(b) Extract and collate the key results, grouping comparable studies

(5) ***Synthesis phase***
(a) Identify all the key dimensions of the problem
(b) Give a narrative account of the contribution of each dimension
(c) Explain conflicting findings and consider as the higher order data

(6) ***Recommendations phase***
Through reflection, multidisciplinary dialogue and consultation with the intended users of the review
(a) Summarize the overall messages from the research literature
(b) Distil and discuss recommendations for practice, policy and further research

*Adapted from Greenlagh et al [1]

IV.7 The environmental scan

The overarching goal of the environmental scan is to look for relevant information in sources not published in the biomedical bibliographic databases particularly outside the field of medicine. We looked for actors and stakeholders, key events, documentation (white papers, position papers, proceedings of meetings) and trends (what is coming up – what are we preparing for, what are people working on).

The project team conducted general and targeted Internet searches.

1. The general search utilized variations of the search terms that defined the two main concepts of the question at hand; which are:

- Concept 1: Patient perspective; patient voice; patient feedback; patient input; patient values and preferences.

- Concept 2: research agenda, research design, research outcomes; patient reported outcomes; patient important outcomes.

We searched the scientific search engines Scirus and Sciverse, which contain scientific journals content, scientists' homepages, courseware, pre-print server material, patents and institutional repository and website information.

In Scopus, Scirus and Sciverse, we used the terms (patient OR patients OR consumer*) AND (involv* OR engage* OR participat* OR recruit*) AND ("research agenda" OR "trial design" OR perspective* OR partner* OR planning OR priorit*)). These terms were modified and used to search Google and Bing. The use of multiple search engines limits the impact of potential biased results due to filtered results (i.e., filters used to personalize searches). We examined the first 20 links resulting from each search and if relevant links were identified, we reviewed additional 10 until no relevant links were identified. ‘Snowballing’ (evaluating new links identified through included relevant links) was also used.

2. The targeted search consisted of examining Web sites recommended by topic experts including the project team, external advisors and PCORI methodology working group.

The content of each web link is reviewed and evaluated for relevance to the topic of interest (patient engagement in research). Data from relevant links are extracted including the title, source, author, URL, description of the content and main conclusions. The environmental scan is expected to identify some of the published literature already included in the systematic review; these overlapping references will be excluded from the environmental scan output. Searches were done by two reviewers independently and were subsequently combined.

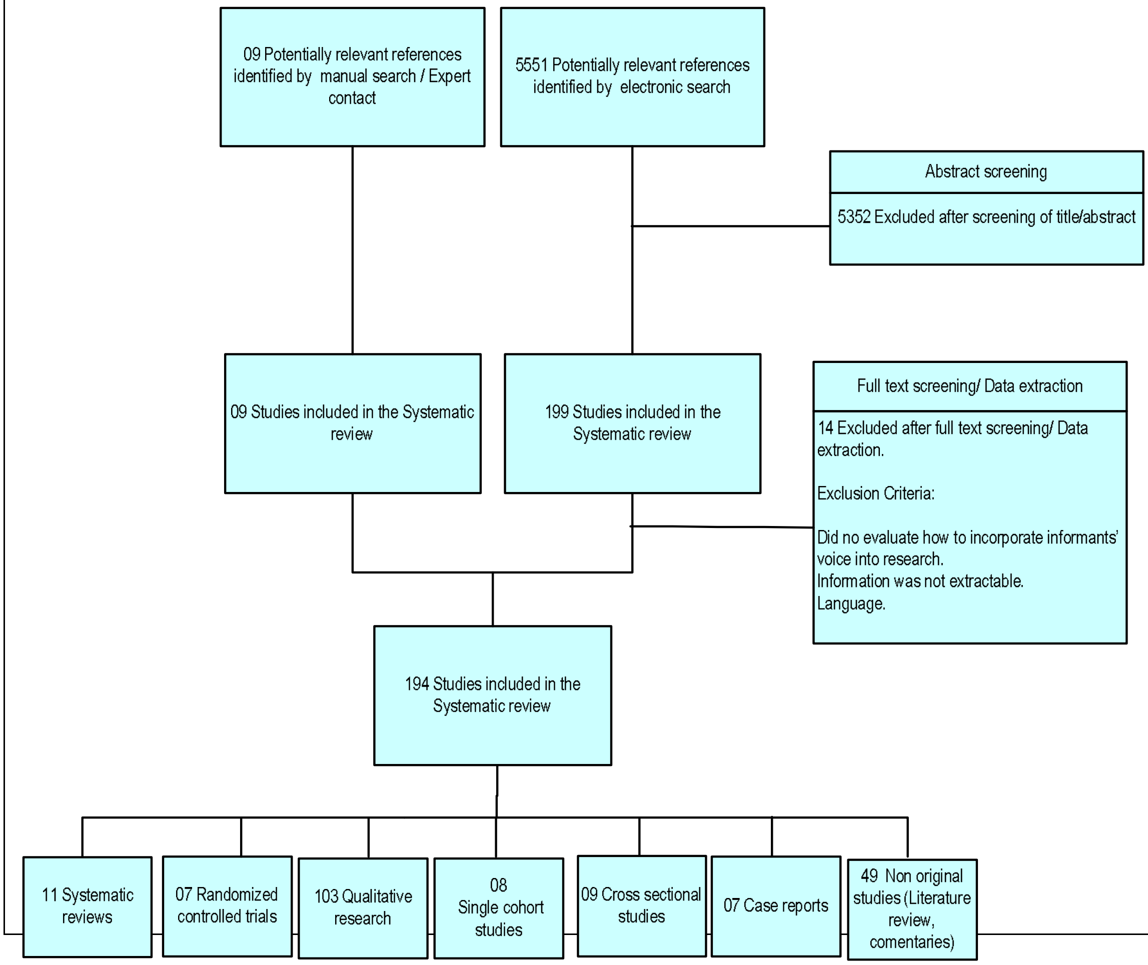
Data from the environmental scan were incorporated with the results of the systematic review following the same framework. **(appendix 3)**

V Results

V.1 Search and selection results

The literature search for the systematic review identified 5551 possibly relevant citations, of which 194 were finally included. Study selection process is described in **Fig. 1: Study selection process**

Figure 1. Study selection process



We summarized the most common methods used in the included studies in **table 1**. This table is showing the used method in each step of the research process by study design.

Table 1. Used Methods to obtain informant voice

		Preparation	**Execution**				**Translation**		
Methods	**Design**	**AS&F**	**SD&P**	**SR**	**DC**	**DA**	**Dis**	**Imp**	**Evl**
Focus group	SR	1	3	1	1	0	1	1	0
	QR	12	8	6	0	1	0	8	1
	RCT	0	1	0	0	0	0	0	0
	Cross	1	0	0	0	0	0	0	0
	Case Report	1	1	0	0	1	0	0	0
	Literature	0	0	0	0	0	0	0	0
	Commentary	0	1	0	0	0	0	0	0
	TOTAL	**15**	**14**	**7**	**1**	**2**	**1**	**9**	**1**
Interviews	SR	1	2	2	1	0	1	0	0
	QR	8	12	13	0	1	4	9	2
	RCT	1	1	1	0	0	0	0	0
	Cross	0	0	0	0	0	0	0	0
	Case Report	0	1	0	0	1	2	2	1
	Literature	0	0	0	0	0	0	0	0
	Commentary	0	0	1	1	3	0	0	0
	TOTAL	**10**	**16**	**17**	**2**	**5**	**7**	**11**	**3**
Survey	SR	1	1	0	0	0	0	0	0
	QR	1	6	6	2	1	1	4	1
	RCT	0	1	0	0	0	0	0	0
	Cross	2	1	4	1	2	0	0	0
	Case Report	0	0	0	0	1	1	1	0
	Literature	0	0	0	0	0	0	0	0
	Commentary	0	0	1	0	0	0	0	0
	TOTAL	**4**	**9**	**11**	**3**	**4**	**2**	**5**	**1**
Deliberation / Organizational participation	SR	0	1	1	0	0	0	1	0
	QR	3	2	1	0	1	2	2	1
	RCT	1	2	0	0	0	0	0	0
	Cross	0	0	0	0	0	0	0	0
	Case Report	0	0	0	0	2	1	1	0
	Literature	0	0	0	0	0	0	0	0
	Commentary	1	2	0	1	0	0	0	0
	TOTAL	**5**	**7**	**2**	**1**	**3**	**3**	**4**	**1**

*SR: Systematic Review, QR: Qualitative Research, RCT: Randomized Controlled Trials, LR: Literature Review, AS&F: Agenda Settings and Funding, SD&P: Study Design and Procedures, DC: Data Collection, DA: Data Analysis, Dis: Dissemination, Imp: Implementation, Eval: Evaluation. **Deliberation/Organizational participation** methods means that informants interact with researchers in several and unique ways such as be part of advisory groups, study design committee or informants have right to vote about the research design during the research process.

Table 2 Findings of randomized controlled trials

Name	Trial aim	N	Benefits of engagement	Barriers/challenges to engagement
Swartz, 2004[2]	To describe the implementation and baseline data of an inner-city community based participatory research clinical trial designed to test the effectiveness of a pollutant and allergen control strategy on children's asthma	100	To create a study design and procedures acceptable to an inner city community	The constant moving of the families was a limitation to their engagement. This forced researchers to develop strong bonds with the community in order to get information and trace the participants
			Maintained the enthusiasm of the participants and expanded their roles beyond their research duties and into the community Validated the project in the community and helped developing relationships with the families	Researchers were skeptical that a community based clinical trial could be conducted. Some were concerned that the purpose and goals of the study would become diffuse and the scientific validity would be lost if planning involved open discussion and incorporation of all community concerns
			Improved recruiting and retention strategies and writing of meaningful, understandable result reports for participants and for the community	Some members in the community felt that researchers had a history of doing studies in their community and leaving without giving back specific benefits or helpful information to the community
Shagi, 2008[3]	To investigate the feasibility of a participatory model of community liaison among an occupational cohort of women at high-risk of HIV and sexually-transmitted infections in Mwanza City, northwest Tanzania in preparation for a Phase III vaginal microbicide trial *Clusters	78*	Feedback during community advisory boards meetings suggested that the community liaison in itself is promoting the development of shared ideas and goals at least among community representatives, where there is an appreciation that women in this community can play a vital role in the development of female-controlled HIV prevention methods and that it is possible to work in partnership with researchers towards a shared vision	Contextual factors (occupational/labor related) may have affected the representativeness of engaged women (it is unlikely that all women in the cohort have an equal opportunity to participate)

			Clustering has allowed meetings to be held close to the workplace, resulting in minimal disruption to day-to-day work activities, helping to ensure on-going support from facility owners and managers	Participants may have been stigmatized (labeled as HIV positive)
			The model has facilitated direct interaction between researchers and study participants, which has helped ensure the validity of joint action plans designed to tackle key project-related concerns	
			The cluster-based model has developed into the foundation for ward-specific mobilization and recruitment strategies in the main trial and allowed targeted interventions to be implemented in several wards to tackle key issues.	External validity concerns: the model is for community liaison in HIV prevention trials among vulnerable, stigmatized high-risk groups in sub-Saharan Africa and other resource limited settings, but how it's extrapolation to other setting is limited
			Over time, community representatives have increasingly felt empowered to disclose their participation in the trial and to take on an advocacy role within the broader local community and working in order to promote debate about the trial	
Marsden, 2004[4]	To involve breast cancer patients in the design of RCT of hormone replacement therapy	83	Focus group methodology can be used to identify issues of relevance to breast cancer patients and help develop specific research questions and formulate priorities	Providing sufficient support to patients at all stages increases the time and resources needed for the development of the study Increased trial accrual does not necessarily reflect the complexity of patient decision making or the perceived quality of the trial

Daugherty, 1995[5]	To understand some of the complex issues related to the participation of cancer patients in phase I trials, and the perceptions of patients toward these trials	27	Better understanding of patients' motivation to participate in phase I RCTs that leads to developing better tools for informed consent	Involving patients in phase I RCTs raises important ethical concerns regarding possible lack of benefit and increased risks
Koops, 2002[6]	To determine whether consumer involvement would help solve some of the ethical concerns associated with research (thrombolysis for acute ischemic stroke, with its inherent risk of fatal intracranial hemorrhage)	54	Informants' involvement helped to refine trial consent procedures and led to an ethically acceptable trial design Better informational tools to educate trial enrollees	Obtaining informed consent for emergency stroke treatment is difficult and presents many ethical dilemmas Researcher could not select a variety of patients for the study that represented the actual population
Atkinson, 2011[7]	Patients with breast cancer participated in evaluating different versions of a website designed to help with decision making regarding enrollment in cancer trials	77	Better product/outcome (the revised online tool better supported patients' decisions to participate in clinical trials) Supported a sense of personal control and decisional autonomy	Patients involved were mainly highly educated, had medical insurance and few of them came from a racial or ethnic minority. This may lead to a selection bias and misrepresentation of the real population
Edwards, 2011[8]	To demonstrate how consulting parents about the design of a study led to the design and successful delivery of a RCT of osteopathy for children with cerebral palsy	20	Better choice of outcome measures consistent with parents' values Higher enrollment and retention rates Parents empowerment	potential conflict between scientific rigor and conducting a trial which is regarded as ethical and acceptable by parents and children Researchers are appropriately constrained by the concept of equipoise which dictates that is only acceptable to withhold treatment from some participants in a trial if the researchers genuinely do not know whether the treatment is beneficial. However families may find it hard to accept that their child may not receive treatment when taking part in a trial

Table 3 Main findings of included systematic reviews

Name	No. Included studies	Aims	Main conclusions	Barriers/challenges
Oliver, 2004[9]	286	To evaluate outcomes, approaches, benefits and barriers of consumer involvement in research development, agenda setting and prioritization	Methods of engagement depend on the question being asked, tasks needed and consumer characteristics More success might be expected if research programs embarking on collaborations approach well-networked consumers and provide them with information, resources and support to empower them in key roles for consulting Consultations should engage consumer groups directly and repeatedly in facilitated debate; when discussing health services research, more resources and time are required if consumers are drawn from groups whose main focus of interest is not health	Barriers to consumer involvement include: poor representation of consumers; consumers' unfamiliarity with research and research programs unfamiliarity with consumers; negative attitudes and poor working relationships'; difficulties in communication, training and support
Hussain-Gambels, 2004[10]	6	To investigate how South Asians conceptualize the notion of clinical trials and evaluate factors affecting their decisions about participation and their perceived risks and attitude regarding participation	Summary of patient motivations for clinical trial Participation: Altruistic factors • Health benefits • Effective follow-up • Clinician influence • Communication style of doctor/nurse • Satisfaction with previous experience	Barriers to clinical trial Participations (a range of practical reasons and moral objections).: trial burden, treatment preference, drug side-effects/fear of experimentation, randomization process and informed consent Strategies to improve South Asian under representation in clinical trials: Educational programs aimed at investigators, ethics committees and funding bodies to increase awareness

	of under-representation of ethnic minority people in clinical trials _ Additional resource allocation for language support _ Patient education and advance awareness of clinical trials _ Culturally sensitive training _ Recruiting more ethnic minority health professionals _ Need for guidelines and ethics committee policing _ Improved reporting of ethnic background in published trial findings and a greater sensitivity in reporting research findings _ Mandatory inclusion of ethnic minority people _ Culturally sensitive communication and culturally sensitive approaches to the consent process _ Promoting trust _ Developing innovative patient education materials _ Improving the informed consent process

Nilsen, 2010[11]	6	To assess the effects of consumer involvement and compare different methods of involvement in developing healthcare policy and research, clinical practice guidelines, and patient information material	information material is more relevant, readable and understandable to patients, without affecting their anxiety 'consumer-informed' material improve patients' knowledge There is low quality evidence that using consumer interviewers instead of staff interviewers in satisfaction surveys can have a small influence on the survey results There is low quality evidence that an informed consent document developed with consumer input (potential trial participants) may have little if any impact on understanding compared to a consent document developed by trial investigators only There is very low quality evidence that telephone discussions and face-to-face group meetings engage consumers better than mailed surveys in order to set priorities for community health goals	There is a huge gap in the evidence about desirable and adverse effects of consumer involvement in healthcare decisions at the population level, or how to achieve effective consumer involvement. The effects of involving consumers in developing healthcare policy and research, clinical practice guidelines and patient information material remain largely unevaluated
Boote, 2010[12]	7	To review published examples of public involvement in research design, to synthesize the contributions made by members of the public, as well as the identified barriers, tensions and facilitating strategies	Group meetings were the most common method of public involvement. Contributions that members of the public made to research design were: review of consent procedures and patient information sheets; outcome suggestions; review of acceptability of data collection procedures; and recommendations on the timing of potential participants into the study and the timing of follow-up	Involved patients could not recognize the experimental nature of medical research, and did not fully understand the intervention. Some patients could show discomfort towards specific aspects of research that could decreased their participation Low response rate The patient who accepts could misrepresent the study target population Researchers could influence the patient selection for their own

				benefits Identification of different values between different informants (patient vs surrogate) in the same topic The consultees wanted outcome measures included in the study for which there are no valid and reliable measures available Potential conflict identified between the views of the consultees and 'scientific rigor.' The cost and the times involved in adopting a more collaborative approach could be a real and important problem
van de Bovenkamp, 2009[13]	42	To identify the rationale and benefits for active patient participation in guideline development and related research	Active patient participation in guideline development is done by the assumption that it will enhance the quality of the guidelines and make guidelines more patient-centred. There was no empirical evidence to support this assumption.	Patients experienced several difficulties in the participation process, which cannot solely be traced back to flawed practices. Patient contributions were often not acted upon. They underscored the challenges of uncertainty about the goals and impact of participation and the possibility of tokenistic engagement. In one of their studies, patients in the development group were described as 'non-participating observers of technical discussion to which they could offer no input'. Van de Bovenkamp also reports that one of the qualifications on the job description for a prospective member in the guideline development group used by the National

				Institute for Health and Clinical Excellence (NICE), responsible for the development of guidelines, is the ability to understand scientific articles. However, this will automatically lead to highly educated patient representatives and possible bias. They suggested that the addition of a survey of patient preferences at large may complement the function of such highly selected sample of representatives.
Stewart, 2010[14]	250	To determine how patients and clinicians can be engaged to determine research priorities	A sizable literature is available to inform priorities for research and the methods of setting research agendas with patients and clinicians. Authors recommend that research funders and researchers draw on this literature to provide relevant research for health service decision-makers	
Diaz del Campo, 2011	NR	To present a strategy for patient involvement in the development of clinical practice guidelines	The study present a framework for patient involvement based on study phase (preparatory, elaboration and implementation. The study recommends conducting patient consultation in the preparation phase, information regarding patient perspectives, experiences with illness, social circumstances, habits, values and preferences can be obtained and incorporated in guidelines. This helps focus on patient important outcomes such as the quality of life.	Limited number of existing robust studies on the topic Non validated experts' assumptions and variability in patient participation level

Mockford, 2012[15]	28	To identify the impact of Patient Public Involvement on UK National Health Service healthcare services and to identify the economic cost. It also examined how PPI is being defined, theorized and conceptualized, and how the impact of PPI is captured or measured	Fifteen of the studies reported on the development of new and improved services attributed to user involvement. However, there was usually little description about how much influence service users had. An important area of service user activity was around information development and dissemination. User involvement was described in producing public and patient information, raising awareness of chronic conditions and the development of training sessions for both service users and health professionals. Many studies noted that working with service users contributed to changing health professionals' attitudes, values and beliefs about the value of user involvement, although in many studies this was described as a difficult task	There is little evidence of any economic analysis of the costs involved. A key limitation of the PPI evidence base is the poor quality of reporting impact. Few studies define PPI, there is little theoretical underpinning or conceptualization reported, there is an absence of robust measurement of impact and descriptive evidence lacked detail.
Butterfoss, 2006 [16]	NR	To synthesize the published public health literature to determine how process evaluation has been used to examine community participation and its intermediary role in health and social change outcomes	potential benefits that had been measured were: heightened public profile, increased utilization of expertise or services; enhanced ability to address an important issue; enhanced ability to affect public policy; development of valuable relationships with other groups; acquisition of useful knowledge about health issues, services, programs or people in the community; achievement of organizational goals; ability to make a contribution to the community; acquisition of additional financial support and enhanced	Diversion of time and resources away from other priorities or obligations

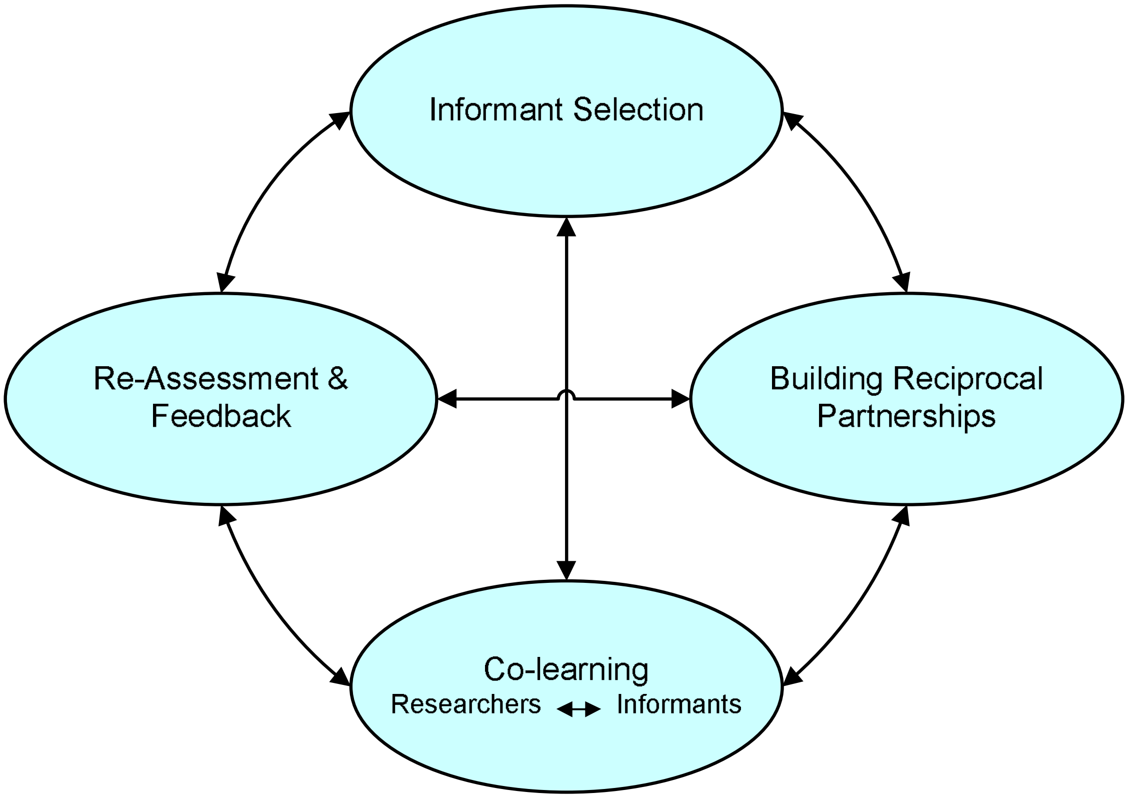
			access to priority populations	
Donovan, 2002[17]	352	To capture users' experiences of participating in cancer trials	The inclusion of users' views in research is consistently advocated. There are various research methods that can be used to capture aspects of users' views of participation in cancer trials. Qualitative research methods enable user views to be collected and used to drive changes to patient information and influence the design and conduct of cancer trials	Different methods have different focus and may produces different results

V.2 Engagement Process Framework

We found 34 studies that described a framework or a scheme that included steps for the process of patient engagement **(Figure 2)**. Out of these 34 studies 12 were qualitative research projects, 18 commentaries or literature reviews, 2 systematic reviews and 2 cross sectional surveys. Some studies focused on one step and other studies described several steps. Eight studies commented on informant's selection, 3 of them were qualitative research studies, 3 were commentaries and the last two were a case report and a cross sectional survey. Thirty studies commented on building reciprocal relationships between researchers and patients, 14 of them were qualitative research studies, 4 were randomized controlled trials, 9 commentaries and the last three were 2 case reports and one cross sectional survey.

Two reviewers coded the steps described in these studies into discrete categories and recorded the inter-relation of these steps. We found four essential steps: Informant's selection, building reciprocal relationships, co-learning and Re-assessment and feedback. These steps were described to have a circular and bidirectional relationship leading to consecutive feedback loops in which researchers and patients work together sharing the same goals. We were unable to identify one particular step as being the most important. The proposed framework describes a process to be performed until saturation (i.e., when no additional feedback or new information being shared and also both parties agree to progress with a particular step in the study).

Figure 2.


Informant Selection
Re-Assessment & Feedback
Building Reciprocal Partnerships
Co-learning
Researchers ↔ Informants

V.2.1 Selection of representatives for participation

Decker et al. reported that the selection of informants should be done thoroughly and this process can be lengthy although the time spent is quite justifiable. [18] Invited potential informants should be as similar as possible to the population being studied and to whom the results would be applied. However, those invited must also show some unique characteristics to ensure the success of the project (i.e. participatory behavior). The choice of representatives should be focused on individuals or communities that are directly impacted by the research (i.e., results will apply to this community or the outcomes of interest are of clear current or emergent concern to the community)[19,20]

Several studies [21-29] also demonstrated the importance of engaging patients and surrogates as early as possible in the process so that they can steer the agenda and outcome of research and provide the values context which will improve study design and applicability. Decker, 2010 [18] highlighted the value of this early engagement process with two examples. The first one described a study in which researchers were interested in cervical cancer screening. However, extensive consultations and interviews with a variety of stake holders in Cape Town, South Africa, led to a change in the study that incorporates a broader perspective toward cervical health. This revised research plan better reflected the communities' concerns and emphasized intersecting variables that were of unknown impact to researchers (poverty, sexual violence, and HIV). This change was described to improve applicability of interventions. The second example was when Sex workers in Brazil grew weary of being the subjects of epidemiological research after they have been identified to have increased prevalence of HIV infection. They approached national health authorities to collaborate on developing guidelines for future research engagements that will prevent their communities' stigmatization and lack of control.

Morrow et al. [30] described three elements to be considered in the selection of informants (the ability to engage, the agreement on the potential and expectations of the engagement, and the possibility of a sense of equality between participants and researchers).

In summary, factors to be considered in selection: selected participants should be as representative as possible of the community or population to which results would apply, they should exhibit the ability or characteristics to allow them to engage in meaningful and equal manner and should be able to engage as early as possible in the research process.

V.2.2 Reciprocal Relationship

The relation of the researchers and the informants should be non exploitative reciprocal and based in trust and continue communication.

Several studies [2,22,26,31-36] highlighted that from the very beginning, researchers should see informants as equal partners and consider them as a reliable component of team. They should not be considered as an additional isolated variable to deal and to invest time with. Both parties have to clearly know their task, roles and expectations and the importance and independence of their roles from each other. Also they have to be included in any other steps related with their main role. The partnership should be based on a mutual understanding of partners' needs, capacities and goals[37]. Conflicts should be solved promptly and explicitly. .

Oscos-Sanchez et al.[38] demonstrated that reciprocal cultural competence of researchers and informants made the project more feasible and efficient; and the lack of this element would have been an impenetrable barrier. Karmaliani et al.[39] proposed that prior suboptimal experiences with previous researchers (i.e. promises not kept and abandonment after data collection) could affect the new relationships with future researchers.

V.2.3 Co-learning process

Properly conducted, this most complicated step need to be conducted without losing the initial goals of the project, which would increase community acceptance and applicability. Danley and Ellison in their Handbook about participatory research[40] comment that this process must provide opportunities to all team members to acquire new knowledge and skills. Also they commented that researchers should begin by training themselves, so their project could be structured to adhere as nearly as possible to participatory principles.

As Harper et al.[41] pointed out; the informants need to have a level of expertise in research to prevent researchers from fully dictating their agenda and driving their opinion. The informants in most cases will require education or training about the content and methodology. White et al emphasized the utility of addressing learning needs of the researchers (including education and consciousness-raising about the realities that the consumers have faced and continue to face as and the social dynamics between the dominant and subordinate groups).

Other authors also advocated sufficient time investment to allow researchers understand the context and realities of their informants. This understanding is derived from sharing expertise, perspectives, and skills with them.

White also recommended that consumers should learn the basics of scientific methods and the mechanism of conducting well-designed studies so that they can carry on a dialogue with researchers and other professionals and provide a higher level of contribution (through the more advanced role of a researcher; i.e., they conduct research themselves)

Decker et al. [18] in 2010 provides the rational for co-learning in that it upholds the scientific soundness. By having a conversation with local counterparts and harnessing local knowledge, researchers can use better phrasing and fundamentally alter the protocol, results and applicability of their work. The example provided was about research into the prevalence of certain contraceptive methods and how different response rates were achieved when questions were asked differently (e.g., when asking about ‘‘tubal ligation’’ compared to ‘‘getting your tubes tied)

This dialogue if conducted properly can increase the ecologic validity (i.e., external validity) of research and the interaction between theory and practice (i.e., translation), and achieve flexibility in the choice of methods and professional inquiry. Effective communications based on collaboration with representatives of the community of the study can reduce the use of jargon considered to be a major impediment to the participatory process because it alienates participants.

All of these issues presented in the previous paragraphs could be addressed through proper co-learning processes in which the researchers become aware of their limitations, share and receive information and provide more externally valid inferences. Co-learning also may increase informants' confidence and drive them into a more engaged role and reduce the risk of passive and tokenistic engagement.

V.2.4 Reassessment and feedback process

This process aims at evaluating the whole engagement and can provide clarification of informants' and researchers' roles and expectations, and help modify the processes of informants' selection, relationship building and co-learning.

Doyle et al. [19] proposed that the whole engagement process should be continuously evaluated leading to process improvement and additional informant empowerment.

Two others studies[42,43] commented on the usefulness of the evaluation indicating that it improves participant empowerment and reveal potential facilitators and barriers. The execution of this step will improve the robustness of the upcoming research project.

V.3 Potential spectrum of patient engagement in research

Shippee et al.[44] and other studies [45-47] describe a spectrum of patient (or surrogates) engagement in research **(Table 4)**. This spectrum starts from the most passive role of being a participant in a study (a data point) to more engaged and advanced roles that include tighter collaboration with researchers all the way to becoming a partner researcher. In fact, patients have initiated and conducted studies themselves (several examples are mentioned in the section of patient participation in preparatory stages of research).

Table 4 Levels of consumer/patient involvement in research

	Shippee et al.[44]	Oliver et al.[45]	Hall et al.[46]	Happel et al.[47]
Passive	Subject	Minimal		Consumer advisory
	Respondent			
	Participant	Consultation	Consultation	Consumer consultation
	Stakeholder			
	Consultant	Collaboration	Collaboration	Consumer collaboration
	Partner			
Engaged	Researcher	Lay control	User control	Consumer led

V.4 Engagement as a function of research stage

The available literature of patient engagement describes successful instances of patients or surrogates participating in all phases of research. Shippee et al.[44] describes the stages of patient and public involvement (as with the stages of any research process) to *range from priority- or agenda-setting (including studies initiated by patient), to aspects of funding or other application processes, to study procedure development (e.g., determining consent procedures), to study design (including choice of outcomes[48]), to recruitment, data collection, analysis, and interpretation, and all the way to disseminating findings. Moreover, PPI does not necessarily occur at only one stage: rather, in many cases, it has occurred at several stages of the same project (e.g., agenda setting, study design, and recruitment, data collection, and analysis).* Our systematic review identified 5 other studies [49-53] that proposed similar frameworks or models that seems to revolve around the same three main stages of research described by Shippee et al (the first is preparatory phase of a study; the second is the execution phase of the study; and the last stage consists of post analysis procedures that include dissemination and implementation). All these frameworks converged into three main stages of research process with 8 steps in total where informant engagement can be conceptualized **(Table 5).**

Table 5. Stages of informants' involvement in the research process**

Stage	Step	Example
Preparation	Agenda setting & Funding	Steering committee, ensuring research is relevant, protocol preparation. Voting, review
Execution	Study design & Procedures	Review of consent procedures. Choice of guideline panel Choice of primary outcomes, development of outcome instruments
	Recruitment	Social networks
	Collection	Patient-administered interviews
	Analysis	Interpretation of findings, external review
Translation	Dissemination	Presentation, manuscript, pamphlet, social media, plan for future study
	Implementation	Developing decision aids tools, developing clinical practice guidelines
	Evaluation	Evaluation of process measures, adherence and uptake of interventions, plan for future research

** Based on: [44,49-53]

We found another relevant model Framework for Consumer and Community Participation in Health and Medical Research developed by the National Health and Medical Research Council, Commonwealth of Australia. The **table 6** describes the essential components of a patient engagement program.

Table 6

A Framework of Action: Constituents of a Program of Patient involvement [53]
◆ A process that is systematic and demonstrates the continuous involvement of users and patients/carers; ◆ A means of demonstrating to patients/carers and relevant agencies, how patients/carers have been involved; ◆ A mechanism for feeding back to users and patients/carers the outcome of their involvement; ◆ An inherent process of continuous evaluation of the system(s) employed to achieve the above.

V.5 Description of evidence of patient and/or surrogate engagement according to research stages)

In the following section, we describe the engagement process according to research phase (preparatory, execution and translation) starting by describing *the evidence* (i.e., the studies that described the process), *the methods of engagement* (how informants were selected and recruited and how was their feedback obtained and incorporated in research), potential *benefits or improved outcomes* due to engagement, potential *challenges* to the engagement process in that phase with any proposed solutions.

V.5.1 Stage 1: Preparation Process

The preparatory process of research includes setting research agenda, identifying key topics and questions and prioritizing them and securing funding and allocating resources. In this step researchers and informants would answer this question: What to research?[53] This step is extremely important because patients and other stakeholders exhibit different ideas and priorities about research.

V.5.1.1 Step 1: Agenda Setting and Funding

V.5.1.1.1 The evidence

Thirty three studies described patients' engagement in the preparatory stage of clinical research (agenda setting and funding). The most common conditions evaluated in these studies were maternal fetal medicine followed by mental health, then other chronic conditions like cancer, diabetes, arthritis and chronic lung disease.

There were studies initiated by patients [54,55]; which represents the most engaging form of patients in the phase of agenda setting. Several members of an international disease-specific support group on a social networking site approached researchers to initiate a study on their condition (spontaneous coronary artery dissection) and helped identified others with the condition. This study demonstrated the feasibility of a model for "virtual" multicenter disease registry through disease-specific social media networks to better characterize an uncommon condition. In the second patient-initiated study, patients with amyotrophic lateral sclerosis experimented with lithium carbonate treatment (a therapy that has not received regulatory approval for their condition), analyzed and reported their results on the website PatientsLikeMe.com

Engagement in the early stages of planning research (initiation and funding) is also demonstrated when parents in a pre-term birth support group contacted researchers and initiated a study (including a proposal for funding). The parents were motivated by dissatisfaction with several aspects of their children's' care[56]. Chenoweth and Kilstoff[57] described participatory action research in a study initiated by care givers of patients with dementia who designed, implemented and evaluated a new dementia therapy program.

In another example, sex workers in Brazil[58] grew weary of being the subjects of epidemiological research after they have been identified to have increased prevalence of HIV infection. They approached national health authorities to collaborate on developing guidelines for future research engagements that will prevent their communities' stigmatization and lack of control.

A included systematic review[59] that summarized studies published through 2008 in which patients provided input on research priorities. This systematic review demonstrated that a sizable literature is available to inform priorities for research (156 studies in which participants engaging with research rather than only considering their experiences or perception of health from which researchers inferred their priorities for research). Participants specifically identified important research topics or questions in 148 studies (full research questions, interventions, population and outcomes in 96, 11, 5 and 20 studies; respectively).

Cancer and mental health were the first and second most common areas; respectively.

Patients engagement in the funding aspects was limited (one study reported some involvement in developing a bid or a proposal and another reported patients' input leading to improved coverage of all trial expenses)[8,60]

V.5.1.1.2 Methods of obtaining patients' voice

The majority of the studies (66%) consisted of qualitative research in which focus groups, semi-structured interviews and surveys were used to elicit opinions of patients, parents, community board members, and occasionally, healthcare workers about the best ways to engage patients in this phase. Less commonly used were online forums. Patients in one study voiced that interviews and focused groups were the appropriate methods of engagement[60].

Studies summarized in a systematic review by Stuart et al[59] reported using formal methods for reaching decisions about research priorities such as Delphi exercises, individual ratings and applying criteria, voting, scoring or a consensus conference.

There were no data on the selection of informants in terms of the adequacy of their selection (representativeness) or the accuracy of the obtained feedback although four studies highlighted the importance of engaging/selecting ethnic minorities/indigenous people[61], the elderly[25], the young[62] and those with intellectual challenges[63,64]. Identifying informants was performed through inpatient and outpatient medical records, community venues (churches, schools, etc), professional contacts, support groups and websites; however, no comparative data are available to support the use of a particular approach and biases have been described in most methods [65]. Compensation of informants has been described in several studies. A set of core values that guide the interaction between community members and researchers have been identified early in the process of conducting one trial [2]. Written agreements that clarify expectations have been suggested by others [32,34].

Training of patients/community partners through workshops has been described. For example, a 2-day workshop was provided to community advisory board members on the condition (Tuberculosis) and the related research [66]. Community elders interested in becoming a “community researcher” received a training session that covered the topics of stages of the research process, recruiting participants and arranging research interviews, research ethics (consent, confidentiality, distress and disclosure), interviewing skills, and developing the interview schedule[34].

V.5.1.1.3 Potential Benefits

A randomized controlled trial[8] evaluated the effect of cranial osteopathy in children with cerebral palsy. Home interviews of parents were used to obtain their input in reported that involving parents in the early stages of study design, planning, choice of outcomes of most importance and the potential ethical challenges. This engagement led to a successful delivery of a trial with high rates of enrollment and retention (markedly higher than other studies in this condition). A second randomized controlled trial[2] was a community based environmental study that evaluated interventions to reduce inner city children with asthma to pollutants. A community advisory board was engaged from the very early stages and participated in drafting study protocol, selecting the control group, managing ethical challenges particularly in the control group, collection of data, and finally dissemination of results within the community. The method of obtaining informants' voice and feedback consisted of regular meetings in a neighborhood school. The study reported that the trust between trial participants and the community advisor board led to a successful trial with higher enrollment and retention rates. The immediate empowerment of patients is described as a major benefit of early engagement in research[57,65,67].

V.5.1.1.4 Potential Challenges

The included studies described several challenges and barriers to patient engagement in the agenda setting and funding phase of clinical research. The most commonly cited challenges were time constraints (from a researcher and patient perspective) and the worry that engagement may evolve to become a tokenistic endeavor designed to tick a required box. The term tokenistic has been used multiple times. Community perceptions of involvement in the research process were reportedly consistently lower than the degree to which researchers felt the community was involved. Community members can become over-consulted and become suspicious of the ultimate goal of their engagement[34,65,68] Barriers in the stage are summarized in **Box.**

Box.

1. Time constraints (researcher perspective): Patient engagement is time consuming and lengthens the duration required to complete the study
2. Engagement may evolve to become a tokenistic endeavor designed to tick a required box
3. Time constraints (patients' perspective)
4. Funding needed to engage patients
5. Undue amount of physical and emotional burden on seriously ill patients (may violate principles of autonomy and justice)
6. Inadequate representation of informants
7. Poor attendance when no compensation
8. Unclear expectations (differing researcher and patients goals of the study; consumers' unfamiliarity with research programs and research programs' unfamiliarity with consumers; negative attitudes and poor working relationships; difficulties in communication)
9. Lack of informant knowledge/skills in technology and communication methods
10. Inconsistent/intermittent contact with informants yields a group with lower expertise /skills
11. Scope creep (expansion into other unrelated community concerns and issues)
12. Perceived lower status of patients/informants compared to investigators
13. Tension between a commitment to process versus research products (sharing resources, responsibilities, and opportunities), balancing the interests of communities, researchers and public health partners.

V.5.2 Stage 2: Study Execution

This stage includes the development of study protocol (study design and procedures) and the execution of the protocol that includes subject enrollment, delivering the intervention and data collection and analysis.

V.5.2.1 Step 1: Study Design and Procedures

Study design includes the selection of the primary outcomes and methods of analysis. In this step, the question to be answered is: How to do the research project?[53]

V.5.2.1.1 The evidence

Thirty studies described patient engagement in the stage of study design and procedures. Seventeen of these were qualitative research studies, 2 systematic reviews, 4 randomized controlled trials and the rest of them were commentaries, literature reviews or cross sectional studies.

The most common health topics evaluated in these studies were: Neoplastic diseases, other non-transmissible chronic conditions and several general health topics. Three of studies addressed social conditions such as substance dependence and domestic violence.

Six of the included studies worked successfully with populations that are either underrepresented in research or maybe considered by some to be challenging to engage in research design, such as elderly people[25], people at risk of[22] or with HIV/AIDS[3,69], and inmates[70].

A systematic review by Donovan et al. done in 2002[17] concluded that even when the inclusion of users' views in research is consistently advocated there is not a clear guidance about how best this might be accomplished, also they commented that exist several methods to capture informants voice in cancer clinical trials and each of these represent different challenges and could led different results.

The involvement of patients in the decision making regarding ethical considerations in clinical trials could lead to discovering potential important harms, that otherwise would not have been recognized by the researchers. Morin et al. [22] performed a multicenter study to evaluate various aspects of HIV prevention. During this project, patients' representatives raised a specific ethical question about the main objective of the project: Why is the study enrolling Peruvian patients to test the drug Tenofovir if this drug is not available in our hospitals? This question started a whole discussion and concerns about the post study availability of the drug and led to the following changes. The authors suggested that community engagement can increase the research quality in developing countries decreasing the distrust associated with research and the stigma associated with HIV.

V.5.2.1.2 Methods of obtaining patients' voice

The majority of the included studies (55%) used the following interaction modalities: semi-structured interviews (23%), focus groups (13%), or surveys (19%). Twenty six percent of the included studies used more than one modality.

A systematic review by Stewart et al.[71] found that 9 out of the 27 studies they considered to have clear patient engagement used either a face to face method or a Delphi exercise as an interaction modality. They reported that Delphi design in general did not require patients to have debating or other specific skills although patients and researchers with emphatic skills would make this method more successful.

One additional narrative literature review [17] and 3 qualitative research studies suggested "*to ensure effectiveness and efficiency, the method to be used for patient participation must be selected according to the objective to be fulfilled, the necessary level of participation, the robustness of available methods, the characteristics and distribution of patients, and the availability of resources* [72]". Similarly, two other studies [17] [73] proposed the concept that every patient, even if they were selected by convenience, present individual preferences; hence, researchers may have to employ a variety of interaction methods to avoid obtaining feedback biased by the interactional preferences of the patient. For instance the study performed by Daly et al. [73] highlight this fact using as an example of one of the patient's view: "If I have a one on one interview I can give more ideas because I am not getting interrupted by everyone else and you are not worried that someone's gonna laugh at you (Darl, female, 16 years)"

V.5.2.1.3 Potential Benefits

Engaging patients and/or their surrogates in research conducted in communities with high prevalence of social inequities (poverty, unemployment and illiteracy) is particularly important. Their engagement is likely to improve internal validity and applicability of research and will have an empowering effect on participants. Martin et al. [70] worked with incarcerated women on 5 main health areas. The women's' involvement in research process led to drastic changes in the prison health policy and uncovered the needs for prison staff education on early heroin withdrawal symptoms that may lead to hypotension and volume depletion. Also this project led to change the daily menus (introducing salads and fruit instead of bread and high glycemic index carbohydrates) and was quite empowering to involved women.

Several qualitative research studies concluded that this approach is likely to increase patient satisfaction [69], the relevance, applicability, benefits, enrollment rate and overall success of research projects [3] It may also reduce misunderstanding, cultural insensitivity and the use of inappropriate methods [74]. Using an example of a study by Minkler et al. {Minkler, 2002 #6611 that aimed at involving patients in addressing a polarizing issue in the community: death with dignity and physician-assisted suicide legislation. The study demonstrated that patient engagement was empowering and may have actually improved their quality of life. One of involved patients said: *"When I come here to these meetings it lets me be on another plane. Out there I am in survival mode, you know, looking around and behind me, watching out for everything. But when I come in here, I am on another plane and can function differently. And I thank you for that. "*

Another qualitative study by Caron-Flinterman et al.{Caron-Flinterman, 2005 #7459} consulted patients with asthma and chronic obstructive pulmonary disease about their health research priorities. In a focus group, patients developed a prioritized list of problems identified as potential research targets. By identifying the primary outcome, patients may have driven the protocol of this study to generate more useful and relevant evidence.

A systematic review[11] involving 2123 health care consumers showed that patient-developed information material was more relevant, readable and understandable. Involved patients did not have adverse effects of their engagement (specifically, no increase in anxiety scores). The systematic review also demonstrated that when conducting surveys, using patients as interviewers rather than study personnel, led to a different and more accurate measurement of the outcome (less inflated estimates of a satisfaction score). Freysteinson et al.[75] described a study aimed to evaluate self-image (viewing body in the mirror after a mastectomy). Breast cancer survivors were consulted to assist in designing the study. They discovered that the time set up by researchers to enroll patients (3 months after surgery) was too short to ensure that patients were emotionally prepared to discuss their experiences (despite the fact that patients would consent to enrollment). Therefore, results would have been biased without this change in study protocol advocated by engaged patients.

V.5.2.1.4 Potential Challenges

Daly et al. [76] suggested offering more than one interaction method with informants and implied the need for researchers to invest time and resources to develop sufficient skills in each method offered. Donovan et al. [77] drew attention to potential heterogeneity caused by offering multiple methods.

Murad et al. [78] demonstrated that communicating complex concepts with patients via surveys is suboptimal. In a survey of over 2000 patients with diabetes, they demonstrated that about 1 in 4 patients reported hemoglobin A1C to be a more important outcome than death. They hypothesized that this is due to the complex nature of the surrogate endpoint concept and the fact that patients have been reminded by their treating providers for years about the importance of glycemic control.

Freysteinson et al. [79] commented that patients invited to participated in ethical advisory boards could be overprotecting and show over concern of the potential harms that the study subjects could be at risk, this behavior could lead to delaying the start of a trial, mainly due to the ethical opposition of patients serving on advisory boards.

Other authors[35,79,80] comment on the possibility to introduce selection bias when the informants' participation is determined by certain characteristics such as socio-economic status, job flexibility or available time. Hence, if researchers did no resolve these barriers (e.g., arrange transportation), the self-selected sample will not be representativeness of the study population. May et al[32] commented that patient invited to participate were focused mainly on creating and implementing programs and interventions; which was not the intention of the researchers or was the objective of the proposed research.

V.5.2.2 Step 2: Study Recruitment

V.5.2.2.1 The evidence

Forty three studies described patient engagement in the stage of patient/subjects recruitment of the research process. Thirty five of these were qualitative research studies, 3 systematic reviews, 3 cross sectional surveys and the other 2 were a randomized controlled trial and a commentary.

The most common health topics evaluated in these studies were: Neoplastic diseases, other non-transmissible chronic conditions, such as mental diseases, chronic heart failure and degenerative conditions.

The three included systematic reviews aimed to examine and analyze trial participants' experiences to address trial participation in different populations. Donovan et al. highlighted that the current cancer trial enrollment rate is as low as 1-2% of the possible rates expected in these types of trials [81-83]. They analyzed user's experiences of participating in cancer trials to determine factors in trial design that can increase enrollment.

Hussain-Gambles et al. [10] in their systematic review evaluated how the South Asian patients conceptualize clinical trials. They identified influential factors in the participation decision making of patients and health workers. This systematic review and an accompanying qualitative project were driven by the low enrollment of South Asian participants in trials.

V.5.2.2.2 Methods of obtaining patient voice

Forty four percent of the included studies in this step used structured one on one interviews, 30% used surveys and only 2% used focus groups as an interaction methods to obtain patient voice.

Carey et al. [69] performed a qualitative research study to evaluate the psychosocial environment surrounding people living with HIV infection. They concluded from focus groups that the only way of gathering the information was through anonymous self-report questionnaire. This fact added to our findings that the 74% of the included studies used individual interaction methods emphasizing that the topic of research dictates the method of interaction (e.g., sensitive issues require a personal and private communication method).

The systematic review by Donovan et al. [77] found evidence from qualitative and quantitative studies that a critical reason for the current low trial enrollment rate could be due to confusion and difficulties with the concept of randomization. Hence, engaging patient's representatives in this process of a study may facilitate communication of the concept to other patients.

Snowdon et al. in 1996 [84] performed structured one on one interviews of parents of critically ill babies and concluded that without an understanding of the structure and aim of a trial (mainly the rationale for randomization and the concept of equipoise), the decision to participate will be extremely difficult to make. The parents is this study showed up mainly because they were expecting that the intervention that have being tested increased their babies survival changes and they remarked these behavior using the phrase: "It is a baby…they are going to and must to do whatever is necessary for the baby… and the intervention going to be tested is a chance of life for my baby." Snowdon et al in 2006 [85] performed a study aimed to explore the decision-making process of clinicians and parents in one or more of four perinatal trials. They were focused on parental accounts of their decisions and used one on one interviews. They concluded that there is a lack of distinction between the goals of research and medical care and they suggested that all parties (parents and clinicians) need assistance to ensure that decisions were consistent with parents' values. This would be achieved by having the participants fully embedded in the research activity.

V.5.2.2.3 Potential Benefits

A systematic review and the posterior qualitative study done by Hussain-Gambles et al. [10] conducted face to face interviews with 75 concluded that in order to increase South Asian people recruitment in clinical trails the researchers should use:

- Muti-recruitment strategies (mass media, community health workers, etc)
- Defined their population
- Use focus group and advisory groups to identify any potential barriers.
- Develop educational and recruitment approaches to attract ethnic minority health professionals and ensure health professional are adequately trained in culturally and ethnically oriented service provision.

V.5.2.2.4 Potential Challenges

Hussain-Gambles et al. [10] reported sampling bias as the main limitations of their qualitative analysis; which was due to a limited number of south Asian participants. This group of participants has successfully been enrolled in clinical trials suggesting that they may already be sympathetic to clinical trials or have a different understanding of science than that of the general South Asian population.

V.5.2.3 Step 3: Data Collection

V.5.2.3.1 The evidence

Seven of the included studies addressed this topic; of these 3 were commentaries, 2 were qualitative research studies and one was a systematic review. The studies more commonly discussed chronic health conditions.

A survey taken to several Cochrane research groups [86] showed that one in three respondents did not include consumer's perspective in the evaluation of the data to be collected in their systematic reviews and that patient-reported outcomes were infrequently used. There was also no apparent consensus across respondents on the importance attached to identifying and collecting information on patient-defined outcomes or on integrating such information into their activities.

The involvement of patients in the development of a questionnaire [87] or a patient-specific index (to assess the outcome of total hip arthroplasty)[88] was found to be quite helpful to researchers and was hypothesized to produced evidence more consistent with patients/consumers concerns. This approach minimized bias toward the provider's perspective, which has been identified as a potential disadvantage of traditional questionnaire research. Informants by using their insight suggested suitable modes of data collection that were successfully implemented [87] and the process was described as reliable, valid and responsive[88]. For this purpose focus groups were used, they were well attended and well liked because they allowed clients who were uncomfortable with the questionnaire to participate. [87]

Whitley and Goldman conducted a collaborative evaluation with disabled clients to inform future planning. Clients made significant contributions to the successful development of the evaluation by assisting in the development of a questionnaire ensuring that it covered clients' concerns; and using their insight and experiences to suggest suitable modes of data collection that were successfully implemented. Training key clients as research assistants to assist with data collection and analysis was described as “methodologically fruitful”. Whitney described: *I noted that some clients, even some with serious mental illnesses, were quite capable of quickly understanding the scientific basis of research.* [87]

In another study, the community and academic partners developed a cross-sectional instrument to assess the mental health status, beliefs, and knowledge of resources among rural and urban Latinos residing in a Midwestern state. [89]

V.5.2.3.2 Methods of obtaining patients’ voice

The most commonly used method in this step was surveys (3 studies). One study used combined methods: focus groups followed by one on one interview.

Surveys were useful to assess the current situation and to clarify the aspects that should be improved [90]. Focus groups and one on one interviews were useful to assess patients’ preferences to a deeper level and to address their preferences [50]. Surveys were also described to have less than a clear feedback in one study. {Wright, 1997 #3762

V.5.2.3.3 Potential Benefits

The following benefits were mentioned in the included studies:

- The involvement of patients in the development of a questionnaire resulted in one that was more consistent with clients' concerns. {Whitley, 2005 #6090} Consumer input can help ensure that reviewers not only consider outcomes identified by clinicians and researchers but also consider the extent to which primary research has addressed outcomes identified by patients as being important indicators of quality and effectiveness of treatment and care. [90]
- Receiving input from a population with specific characteristics is a useful method for collecting data taking into account their needs; improves the quality of the data and makes the process of data collection more efficient. [91]
- Training key clients as research assistants to assist with data collection and analysis was described to have methodological benefits. [87,91]

V.5.2.3.4 Potential Challenge

The following challenges were mentioned in the included studies:

- Many researchers felt that receiving input from patients about this subject may lead to deviation from the main objective of the study. [91]
- Researchers may consider that most of the patients do not have the sufficient knowledge and skills for this engagement.

- Patients may become concerned and question the honesty and intentions of researchers limiting their participation. [92].
- Much greater resources in terms of time, labor, and finances are needed to develop a tool for data collection following this approach. [87]
- Generalization of the results (i.e., success of patient's engagement in a particular setting) is limited and has direct correlation with the population involved. [88]

V.5.2.4 Step 4: Data Analysis

V.5.2.4.1 The evidence

Twelve of the included studies presented relevant data regarding this step. These studies had several designs: 4 were qualitative research studies, 3 commentaries, 2 cross sectional studies, 2 case reports and 1 systematic review.

The included studies reported that the engagement of patients in data analysis resulted in higher ability to contextualize conclusion to patients' environment and believes. It also can highlight patients priorities for a more focused analysis[22,29,93,94 95,96]. McCuley conducted a community participatory research about exposure to pesticides and potential health effects in migrant farmworkers and their children. Study data and conclusions were given to the community members to receive feedback prior to publication. Half of the community was involved in the process. Researchers modified conclusion accordingly to incorporate the socio-cultural environment and community's believes. The involved individuals added language and cultural insight and expanded the academic researchers understanding of the issues. [22,29,93,94]. Chalmers et al. showed the effect that patients can have over researchers' conclusion: *Lay people have helped researchers to think through the implications of the results of research. For example, I was pleased when a large randomized trial showed that intensive monitoring of babies during labour reduced their likelihood of having seizures after delivery, because this hypothesis had been derived from my first attempt to prepare a systematic review of controlled trials. Women's comments on the trial helped me to put the confirmed hypothesis into perspective. For many of them, increasing the chances of a baby not having seizures from 996 per 1000 to 998 per 1000 (with no evidence that this would be reflected in any more substantive beneficial effect in the longer term) was simply not an adequate incentive to accept the encumbrance of being connected to intensive fetal monitoring equipment during labour.*

In this step, patients engagement requires sufficient information, education and training. [22,29,87] Whitley concluded that patient involvement in this step may increase the validity of the findings, as the different parties must reach consensus on emerging conclusions and their significance with staff, clients, and independent academics acting as a check and balance on one another's biases. [87].

Patients abilities in this step should not be undervalued, even those with severe mental diseases[87] or low educational level [22]can be capable of understanding certain research basics and give adequate feedback to the process. The information should be presented in a suitable manner for patients to understand but avoiding over-simplification because it may lead to obscuring complex relationships that engaged individuals need to know. [93]

V.5.2.4.2 Methods of obtaining patients' voice

The included studies used several methods for obtaining patients' voice: Three of used surveys, 1 used structured one on one interview, 1 used focus-group design and 2 used more than one method. The studies with combined methods (focus group or survey followed by one on one interview) were able to explore deeper and elicit the details of certain concepts that would have otherwise been ignored. Combined methods seemed to also be useful for working with communities or larger groups of individuals [22,97]

V.5.2.4.3 Potential Benefits

The following benefits were mentioned in the included studies:

- The inclusion of patient opinion in data analysis may highlight patients priorities for a more focused analysis [22,29,93-96]
- Involving all members of the partnership in data analysis and interpretation strengthened community capacity building and increases community members' ability to understand complex issues that affected their health [93]

- The roles and skills of community partners were found to be different from but complementary to those of the academic partners. The process was thought to decrease certain elements of bias [93,95]

- Face validity and construct validity were strengthened by the inclusion of informants in the process [93,95]

V.5.2.4.4 Potential Challenge

- Some traditional researchers may feel that including patients in the analysis phase may decrease the scientific sense of the data and is difficult to accomplish. [93,95]
- Much greater resources in terms of time, labor, and finances are needed for this collaboration. [29,87,93]
- It may be challenging to maintain the interest and commitment of patients during the process.[93]

V.5.3 Stage 3: Translation

The translation stage consists of the post-analysis activities and includes three steps: dissemination, implementation and evaluation of the new knowledge generated by the study.

V.5.3.1 Step 1 Dissemination

V.5.3.1.1 The evidence

Twelve studies were found (one systematic review, 7 qualitative research studies, 2 case-reports and 2 commentaries). Studies addressed several health topics: 4 evaluated general health topics, 8 evaluated chronic conditions (HIV, neoplastic diseases, diabetes, etc.) and the remaining two evaluated social topics

Although the scientific peer reviewed literature is the target of dissemination for academic researchers; it is clearly not the way of disseminating research findings to patients, communities or consumers [41,98]. A final approach of dissemination decided jointly by researchers and the involved community or individuals was described to be critical for the success of this partnership [41,50,99]

Several studies concluded that the process should be personalized and suitable for the patient's characteristics and abilities, even when the group may share certain characteristics. [34,99] The study developed by Evans et al. which evaluated how to provide the research project findings to elderly population living in assisted living settings illustrated the following: *"Our experiences demonstrate that individual information and communication needs to vary considerably and requires a personalized approach. For example, some older people are very comfortable communicating via e-mail, while others much prefer to talk on the telephone and in this population a written document should always be considered"* **[34].** The included studies showed that the language is one of the aspects that should be personalized according to the target population and the purpose of the publication. In most scenarios, technical jargon and non-applied information should be avoided [34,41,50,100]. The involvement of some members of the population in the development of the material was found to be very helpful to solve language, cultural and acceptance issues. [22,41,50,79,100,101]

A study conducted in Thailand demonstrated that the involvement of the community gave researchers a media to disseminate study findings that was not available previously: *"The coordinator and her assistant disseminated information on upcoming studies, trial progress and results through a monthly community radio program and bi-monthly newsletter".* This method and the information it provided gave the community more confidence in the researchers and simplify the development of future studies [22]. The same effect was evident in a study developed in the US that focused on the agricultural Hispanic community ("Proyecto Bienestar") were the results were presented through a Spanish radio by young community members and letters with the results were send to the community members that left their addresses. This last point gave community members a sense of importance and that their opinion was being considered. [29]

The involvement of patients can also help to develop new creative methods that may even be more efficient and may not have been considered by researchers [50,102,103]: *"The results of the overall project were disseminated in a national conference that started with a theatre play created by adolescents worked with a professional drama teacher to create the play, using role-play and* *improvisation… Their self-created rap urged the hospital board to listen to young people and adjust care to their needs”.* [102]

The adequate dissemination of the gathered information has proved to be helpful for future implementation, making the results of the projects known within several stratums may facilitate obtaining resources and funding for the implementation. [41]

V.5.3.1.2 Methods of obtaining patient voice

Of the eleven studies that evaluated the dissemination step, two studies used surveys as their method for obtaining patient’s voice. The combination of focus group followed by structured/ semi-structured one on one interviews was reported in 2 studies, the remaining two used only one on one interviews.

The combination of more than one method allowed better exploration of multiple dimensions of patient’s opinion [50]

V.5.3.1.3 Potential Benefits

The involvement of patients in this steps allowed researchers to personalized the dissemination method, including language and cultural issues [41,50]

Inclusion of informants/patients in this step may reveal new and innovative methods that are more creative and effective than what researchers had originally considered [41,98] researchers [50,102,103].

A successful dissemination of a project in the community has proved to be helpful for future research, future funding and subsequent implementation. [41]

V.5.3.1.4 Potential Challenges

The included studies hypothesized but did not demonstrate several social, cultural and ethical concerns. [22,29,41,50]

A particular method of dissemination may satisfy some, but not all, community members. [41,50,99].

V.5.3.2 Step 2 Implementation

V.5.3.2.1 The evidence

Twenty-eight studies described patient engagement in the implementation step of the research process. Three of them were systematic reviews followed by a qualitative research, another one was a conceptual model of community based org partnerships 15 were qualitative research studies, 7 were commentaries, 3 were literature reviews and 2 case reports

Most of the included studies (11 studies) evaluated general health concerns such as the involvement of patients in the development of guidelines, mental health and disabilities, etc. Six studies evaluated social topics such as occupational and environmental health in a Hispanic agricultural community or the situation of elderly patients in nursing homes. The remaining studies addressed patients with chronic conditions (diabetes mellitus, neoplastic diseases HIV infection and others).

Several studies advocated for involving patients throughout the implementation steps[29,41,50,100,104]. They emphasized the need for adequate patient education and support [22,50,100] to maintain their interest and enthusiasm in the project. [16,105]

Patient engagement was most helpful in setting priorities and developing the most suitable sequence of steps and methodology for implementation [34,66,99]. Researchers' dissemination plans changed accordingly in a study by Roe et al.: "Despite the researchers' grand plans for immediate policy advocacy and program development`, our new grandparent colleagues reminded us that the radical shift from isolation to activism begins with smaller steps closer to home" [99]*.* The studies demonstrated that flexible and creative plans have to be developed in order to adapt to the changes and problems as they arise. Several studies highlighted that problems may arise unexpectedly during the implementation process and that they should be considered as learning opportunities that set a milestone for the development of new and better strategies. [16,22,104] The involvement of figures of authorities in the community in implementation was also a helpful strategy for better buy in.[22,106]

It is worth noting that patients may create a force to implement knowledge by themselves, without the interaction with researchers*. I have been astonished by* *the extent to which the results of systematic reviews of research assessing the effects of care during pregnancy and childbirth have been taken up by lay people, both at an individual level (ranging from women using the maternity services to the undersecretary of state for health in the House of Lords) and as groups (ranging from local branches of the National Childbirth Trust to the health committee of the House of Commons).* [96]

V.5.3.2.2 Clinical practice guidelines:

Patient and public involvement in developing and implementing clinical practice guidelines (CPG) is a form of patient engagement in the step of research implementation and may have the greatest impact on how patients' care is delivered and experienced. A systematic review by Légaréet et a.l [107] searched bibliographic databases and contacted relevant organizations. Study selection to determine the key components of this process. Seventy one references were evaluated over half were published after 2002. The most frequently cited objective for patient and public involvement in developing guidelines was to incorporate patients' values or perspectives in CPG recommendations. Patients and their families and caregivers were the parties most often involved. Methods used to recruit participants included soliciting through patient/public organizations, sending invitations, and receiving referrals and recruits from clinicians. Patients and the public most often participated by taking part in a CPG working group, workshop, meeting, seminar, literature review, or consultation such as a focus group, individual interview, or survey. They helped formulate recommendations and revise drafts.

Most modern guideline development schemes (e.g., the Grading Recommendations, Assessment, Development and Evaluation, or GRADE), guideline rigor evaluation tools (e.g., APPRAISAL OF **G**UIDELINES For **RE**SEARCH & **E**VALUATION INSTRUMENT, or AGREE II) or guideline Implementability evaluation tools (Guideline Implementability Appraisal, or GLIA); all emphasize patient involvement to express values and preferences. Boivin et al. [108] surveyed 56 guideline developers and highlighted the role of patient and public involvement in agenda setting stage and the need for their training and education. Gracia et al. [50] described a framework to involve patients through the preparatory, elaboration and implementation step of the guideline development process. However, there remains no clear guidance in the guideline development field on how to best do this, or comparative evidence showing that this engagement improves process measures (e.g., guideline uptake) or outcome measures (i.e., patient important outcomes). [50]

Lastly, despite the compelling argument for engaging patients in guideline development, the impact of this process remains unclear and not necessarily substantiated by empiric evidence. A systematic review by van de Bovenkamp et al. [109] attempted to identify normative or empirical studies on this subject. They identified two strands of thought in the literature: One argues that it is important for patients to participate actively in the guideline development process; and the second argues that guidelines should accommodate individual patient preferences without seeking active patient participation in the guideline development process.

In terms of methods of participation, they described co-opting a patient into the guideline development group, organizing a one-time focus group, holding a workshop where patients came together multiple times, and co-opting a professional patient advocate into the development group. They did not identify studies showing an incremental benefit of patient participation and reported that patients contributed the most on the subjects of patient education, self-management and making guidelines more accessible to laypersons. However, patient contributions were frequently not acted upon. The review reports the challenges of uncertainty about the goals and impact of participation and the possibility of tokenistic engagement.

A qualitative study published this year [110], aimed to explore current approaches to involve consumers in the development of systematic reviews, after performing semi-structured interviews with key informants concluded that consumers (including patients) are currently involved in several ways in the process to conduct systematic reviews. These systematic reviews will provide the evidence for the upcoming clinical practice guidelines, therefore assessing which approaches are most effective in achieving different aims of consumer involvement is required to inform future recommendations on consumer involvement.

V.5.3.2.3 Methods of obtaining patient voice

Studies used focus groups (3 studies), surveys (3 studies) and community advisory boards/group meeting mechanisms (4 studies). Nine of the included studies used more than one method which allowed exploring several dimensions of patient's opinion [50]. The combinations most commonly used (8 studies) was focus group and structured/semi structured one on one interviews (in one study this was preceded by a systematic review), followed by survey and structured/semi structured one on one interviews (2 studies, one of them after this two steps evaluated the process by a the population in a town hall meeting [29]). The combination of focus groups followed by interviews seemed to be superior to focus groups in certain settings (clearer responses) particularly in sensitive topics (self image in breast cancer survivors). [79]

V.5.3.2.4 Potential Benefits

The involvement of patients in the implementation step is considered to likely be beneficial in the development of appropriate measures of diffusion of ideas and processes that a community or population needs.[50,96,99,104,106,111].

The involvement of informants was helpful in setting priorities, in developing the most suitable sequence of steps for the implementation process and choosing an appropriate methodology [34,66,99]. Roe et al. [99] reported: "One of the most important community activities was the support we were able to provide to a grandparents "warmline" – the innovative idea of a member of our community advisory committee who was staff volunteer in a local country supervisor's office. She imagined a hotline that would provide easy and anonymous informational, emotional and practical support for local grandparents raising children."

V.5.3.2.5 Potential Challenges

Lack of the necessary knowledge by informants/participants requires resources and time. [29,47,50]

There is a potential of imbalanced representation of the community. [16,75,99,104]

There is a potential of being unable to satisfy the expectations of the involved population. [102]

There is a potential of being unable to keep patients' interest and enthusiasm for pursuing the goals of the project. [16,105] The systematic review by van de Bovenkamp [109] about patient engagement in guidelines reported that patient contributions were often not acted upon. They underscored the challenges of uncertainty about the goals and impact of participation and the possibility of tokenistic engagement. In one of their studies, patients in the development group were described as 'non-participating observers of technical discussion to which they could offer no input'. Van de Bovenkamp also reports that one of the qualifications on the job description for a prospective member in the guideline development group used by the National Institute for Health and Clinical Excellence (NICE), responsible for the development of guidelines, is the ability to understand scientific articles. However, this will automatically lead to highly educated patient representatives and possible bias. They suggested that the addition of a survey of patient preferences at large may complement the function of such highly selected sample of representatives.

V.5.3.3 Step 3 Evaluation

V.5.3.3.1 The evidence

Very few of the included studies (only 5) addressed this step and mostly did not provide sufficient details. Four of them were qualitative research studies and one was a case report. These studies were conducted in populations with chronic health conditions (HIV, Neoplastic diseases, etc.)

Authors of these studies advised that the evaluation process should be constant, it's implementation should not wait to the end of the process since solving problems at this point will be more difficult and may require more human and material resources [22,104] A continuous flow of information from the informants demonstrated being of great value in this step[79,102]. Also, having clear predefined assessment tools during this process was also found to be helpful. [106]

Studies showed that evaluating the relationship between informants and the research team constantly resulted in great benefits; however, the extent of informants' participation should be clarified in order to avoid conflicts and to favor the development of future projects [41].

V.5.3.3.2 Methods of obtaining patients' voice

These studies used mainly a survey or focus group followed but structure/semi-structured one on one interviews.

V.5.3.3.3 Potential Benefits

A continuous flow of information from the informants was described to have great value during the evaluation process (unclear description).[79,102].

V.5.3.3.4 Potential Challenge

There may be difficulty in achieving adequate representation of the population of interest. [75,99,104]

V.6 Environmental scan

The environmental scan revealed numerous links that included relevant websites, organizations, forums, blogs, videos, associations, workshops, presentations, governmental agencies, abstracts and other forms of unpublished resources that spanned across various topics of health care and non-health care consumers' engagement in research.

Some of the resources were focused on shared decision making in the clinical context and were less relevant to the topic of this review. Others discussed methods of participatory action research and community based participatory research; which are more pertinent. The most relevant resources can be categorized into several observed types:

1) Disease specific social networks that most frequently were non-for-profit and established by patients or surrogates. These networks provided patients with disease specific education and focused on improve their wellness. Many offered insight on participation in research and guided patients to ongoing trials and access to investigational treatment. Few provided education to patients (e.g., The Association of Cancer Online Resources, that empowered patients to participate in research by providing clinical trial FAQ; guide to trial terminology, access to investigational drugs and a guide to find a clinical trial). We did not identify specific methods used to obtain feedback from patients (other than patients' opinions as expressed in the blogs) or select patients or surrogates for research engagement.

2) Non-disease specific websites focused on patient engagement (most commonly found in Europe, specifically in the United Kingdom, and also in Canada). An example is National Institutes for Health Research whose vision is that Involving patients and members of the public in research can lead to better research, clearer outcomes, and faster uptake of new evidence. The goals of this organization are: Set research priorities, Identify the important questions that health and social care research needs to answer, Give their views on research proposals alongside

clinicians, methodologists, scientists, and public health and other professionals, Help assess proposals for funding, Take part in clinical trials and other health and social care research studies, not just as subjects but as active partners in the research process, and publicize the results. Another example (also from the UK) is the James Lind Alliance that facilitates the collaboration between an organization and the patients and helps create a steering group, which comprises both clinical and patient representation who sign a protocol setting out their aims for the partnership, and their agreed commitments. They define the key components of a priority setting process are: engaging with patients, carers and clinicians to participate in the partnership, collecting patients', carers' and clinicians' treatment uncertainties, reviewing existing research recommendations to identify uncertainty and working with patients, carers and clinicians to prioritize the uncertainties.

3) Models for patients' engagements: these were fairly uncommon; two examples are:
    - A Model Framework for Consumer and Community Participation in Health and Medical Research. National Health and Medical Research Council. Commonwealth of Australia 2005)
    - The National Health Service - Patient Involvement Toolkit. These resources provided rationale for patient engagement in every step

of research and their findings are incorporated in the standards and frameworks presented in this report.

- The PatientPartner project; which is a three year project funded by the European Commission and ended in 2011 and aimed to promote the role of patient organizations in the clinical trials context PatientPartner was based on the belief that involving patient organizations as equal partners at all stages of clinical trials contributes to research that is better adjusted to the real needs of patients. The project looked closely at the part that patient organizations play and are willing to play in clinical trials and also focussed it's attention on clinical trials with children, the use of biobanks and ethical issues. The main objectives of the project were to identify best practices of patient organization's active involvement in clinical research in Europe; to facilitate the dialogue between all involved stakeholders in clinical research as to how to establish this involvement and integral part of clinical research, and to develop guidance material to be used by all stakeholders in order to facilitate the future partnerships in clinical research between patient organizations, sponsors and investigators.

VI Patient Advisory Group Feedback

We presented our findings and recommendations to a group of community members with long history and experience of research engagement (http://shareddecisions.mayoclinic.org/stakeholders/diabetes-advisory-group/).

We asked for feedback on terminology, usefulness and applicability. We presented a brief introduction to the aims and methods of this project. Community members (patients) provided the following feedback:

1. The terms informant/patient/surrogate/consumer/customer/representative were fairly confusing and none seemed satisfactory although patient or informant seemed most intuitive. Associated explanatory text attached to any terms used was deemed necessary.
2. Patients understood the purpose of the frameworks presented and valued the need for such frameworks.
3. Patients equally rated the importance of the 4 components of the framework (participant selection, building reciprocal provided, co-learning and evaluation and feedback).
4. Patients rated as most important the first recommendation (which was an overarching recommendation to engage patients in all three phases of research as results of benefits that would likely outweigh barriers and harms).
5. In general, patients found the extent of possible engagement in research to be surprising.

6. Patients provided some suggestions for wording, graphics and presentation.

VII Limitations and Strengths

The main limitation to this systematic review is the non comparative and observational nature of the available literature. Therefore, we present empirical evidence of the beneficial impact of patient and public involvement in research and describe the methods used to select informants and incorporate their feedback in various research stages. However, inference regarding the best appropriate and effective methods is limited.

Another limitation relates to the lack of specific indexing terms in bibliographic databases for patient engagement. Therefore, some studies in which patient engagement process were performed, may have been missed. Furthermore, there is also a lack of standardized and explicit reporting of how this process took place and what were its outcomes. Standard reporting guidelines for different study designs (e.g., the CONSORT statement for randomized trials) can be enhanced by including a template for reporting whether a particular study has engaged patients, how and what were the outcomes of such engagement. To overcome these challenges in indexing and reporting, we attempted an environmental scan to supplement literature search.

Heterogeneity of study populations, methods and outcomes, constitute further limitations to extrapolation of evidence across settings. Publication and reporting biases have also likely impacted the conclusions of this report and their impact could not be estimated.

The strengths of this report include a comprehensive and sensitive search strategy that spanned across multiple databases and augmented by an environmental scan of unpublished relevant sources and contact with experts in the field to further capture related studies, web sites and interest groups. A priori established protocol for selecting and appraising evidence were also implemented to reduce biased selection of studies.

Lastly, a systematic review by Mockford et al. [15] published in 2012 evaluated the impact of patient and public involvement on the UK National Health Service (NHS) healthcare services and to identify the economic cost. The review summarized 42 studies and concluded that there is little evidence of any economic analysis of the costs involved , poor quality of reporting, little theoretical or conceptual underpinning, lack of measurement and evaluation; and overall weak supporting evidence base for patient and public involvement. Therefore, there is a clear knowledge gap in this area and need for future research.

VIII Knowledge gaps and recommendations for research:

- Studies of patient engagement in acute care settings research (i.e., emergency department, prehospital care, etc) are needed. The available literature is mainly focused on chronic conditions.
- Studies of patient engagement in the phase of study execution and specifically in the areas of data collection and analysis are needed. The available literature is focused on the earlier stages of research (agenda setting and patient enrollment) and latter stages (translation).
- In general, studies that measure the impact of patient engagement on research execution, validity and applicability are lacking. Such studies will provide a rationale for engagement beyond the ethical and political one, and may increase the update of engagement in research.
- Research in the area of selecting participants (patients) for engagement in various research topics and setting is virtually nonexistent.
- Research that compares the different methods of engagement and obtaining patient voice is needed. Comparative research of different methods will provide evidence about the relative efficacy of the different methods and facilitate the choice of method that is consistent with the goal of each particular research question.
- Standards for reporting the methods and outcomes of engagement are also needed. This report highlights difficulty in finding these data in published original studies.

- Proper indexing methods of patient engagement in research will facilitate future retrieval and synthesis of the available evidence and would lead to advancement of the methods and outcomes of engagement.

IX Appendixes

IX.1 Appendix 1. Study Characteristics

Study Name Study Design	Goal	Method of obtaining patient voice	Study settings	Informants	Informants' role Informants ‘ action	Sample selection
Abma, 2005[112] Qualitative Research	To describes a responsive-constructivist approach to evaluate the aims and features of patient participation and learning experience.	Structured one on one interview, online patient forum, workshops	Spinal cord injury	Patients	Partner Collaborate	Convenience
Abma, 2010[113] Opinion/ Commentary	To describe a methodology for patient participation in a trial	Structured one on one interview	Burns	Members of patient organizations	NA	NA
Acquadro, 2003[114] Opinion/ Commentary	To addresses key issues of patient reported outcome within drug evaluation	NA	Health research in general	NA	NA	NA
Ågård, 2001[115] Qualitative Research	To investigate how patients included in trials on treatment in the early phase of acute myocardial infarction experience the consent procedure	Semi structured interviews	Myocardial infarction	Patients	Participant Input, Dialogue	Convenience
Ahmed, 2010[37] Opinion/ Commentary	To explore approaches that funders can use to develop community engagement in research training programs	NA	community engagement research	NA	NA	NA
Andejeski, 2002[116] Single cohort	To evaluate the impact of having breast cancer survivors with advocacy experience (consumers) participate as voting members of scientific review panels for proposals on breast cancer research.	Survey	breast cancer	Patients	Participant Review	Convenience
Ard, 2005[117] Qualitative Research	To identify unique variables for African Americans that might limit the effectiveness of behavioral interventions in clinical trials	Focus group	African Americans	Patients	Participant Dialogue	NR
Asai, 2004[118] Qualitative Research	To explores laypersons' attitudes towards and experiences of medical research	Focus group	Health research in general	Patients, physicians	Respondent Undergo	Convenience
Atkinson, 2011[7] Randomized controlled trial	To investigate the effects of changes in an informatics application on patients' participation	Survey, Structured one on one interview	Breast cancer	Patients	Subject Consent, Undergo	Convenience
Australian	To present a Model Framework for Consumer	Participatory	Participation in	community	Partner	NR

Government,2005[53] Qualitative Research	and community Participation in Health and Medical Research	methods	Health and Medical Research	members	Generate	
Avins, 2007[119] Opinion/ Commentary	To outline proposals to help create the necessary recognition and engage participation by patients, clinicians, health-care delivery systems, and the research community to establish the long-lasting growth needed for achieving the full potential of clinical research.	NA	Health research in general	NA	NA	NA
Balcazar, 1998[120] Opinion/ Commentary	To explore 4 general principles of participatory action research and discusses some of its challenges	NA	disabilities	NA	NA	NA
Bigrigg, 1999[121] Cross sectional study	To assess patient's views of commercial clinical trials	Survey	contraceptive methods	Patients	Subject Consent	Convenience
Boote, 2010[12] Systematic Review	To review published case examples of public involvement in primary research design,	Secondary data analysis	Public involvement in research design	Researchers	Researcher NA	NA
Bradburn, 2005[122] Opinion/ Commentary	To involve terminally ill cancer patients receiving palliative therapy in research	NA	cancer patients	Patients	NA	NA
Brody, 2009[123] Qualitative Research	To examine the impact of physician-investigator relationships and clinical research-participation recommendations on family decisions to enroll adolescents in a asthma RCT	Self-reported scale	Adolescent asthma	Patients, Relative	Stakeholder Dialogue	Volunteer
Brown, 2010[124] Opinion/ Commentary	To discuss the insider-outsider distinction in the context of people with disabilities.	NA	disabilities	Patients	NA	NA
Buchanan, 2007[125] Opinion/ Commentary	To identify potential sources of tension between the values of scientific rigor and community participation in CBPR	NA	Social topic	NA	NA	NA
Buckley, 2005[126] Opinion/ Commentary	To present an overview of some of the issues and problems associated with involving patient perspectives in healthcare research	NA	health research in general	NA	NA	NA
Butterfoss, 2006[127]	to review published public health and social science literature to determine how process	NA	community participation	NA	NA	NA

Systematic Review	evaluation has been used to examine community participation					
Caldwell[128] Opinion/ Commentary	To address problems associated with research conducted in Indian Country by investigators who lack understanding of the circumstances of the communities	NA	Indian tribe	Patients	NA	NA
Campbell, 1998[129] Qualitative Research	to learn collaboratively about health care service provision from the standpoint of people with disabilities	NR	Care of patients with disabilities	Patients, health providers, research staff and university faculty	Researcher Generate	NR
Carey, 1992[130] Qualitative Research	To describe the author's use of qualitative data in the refinement of research in a medical setting	Focus group	HIV infected patients	Patients	Participant Collaborate	NR
Carey, 2001[131] Qualitative Research	To describe the experience of 45 outpatients who recently completed their participation in a randomized clinical trial	Structured one on one interview	Severe and persistent mental illness	Patients	Participant Input	Convenience
Caron-Flinterman, 2005[132] Qualitative Research	To discuss the validity of patients' experiential knowledge in the context of biomedical research processes.	Structured one on one interview	Health research in general	Patients, scientists and patients' organizations	Participant, Researcher Input	Convenience
Caron-Flinterman, 2005[133] Cross sectional study	To assess the ability of patients to prioritize research in a well-argued way.	Survey, Focus group, feedback meeting	Asthma and COPD patients	Patients	Respondent Input, Dialogue	Random
Carr, 2003[134] Qualitative Research	To explore the patient's perspective of outcomes in rheumatoid arthritis to identify which outcomes are important to patients	Focus group	Rheumatoid arthritis	Patients	Respondent Dialogue	Convenience
Casarett, 2001[135] Qualitative Research	To define the endpoints of pain research that are important to patients with chronic pain and to identify clinical and demographic variables that are associated with patients' choices of endpoints.	Structured one on one interview	pain	Patients	Participant Dialogue	Convenience
Cashman, 2008[136] Case report	Describes 4 cases where community members participated in data analysis, interpretation, or both,	Focus group, advisory meetings	Community involvement	Community members, health professionals	Partner Dialogue	NA
Chalmers,	To highlight the importance of involving people	NA	Health	NA	NA	NA

1995[137] Opinion/ Commentary	into research		research in general			
Chenoweth, 1998[57] Qualitative Research	To describe the way in which family careers participated with day-care staff to design a therapeutic program for people attending a dementia day care program.	NA	Dementia	Patients, Relative, day-care staffs	Subject, Researcher Input, Dialogue	Convenience
Cohen, 2007[138] Literature Review	Patient's perspective in Neuroscience research	NA	Neuroscience	Patients	NA	NA
Corneli, 2007[139] Qualitative Research	To learn the attitudes and concerns of the local community on participating in research	Focus group	HIV	Patients	Respondent Undergo	Convenience
Cotterell, 2008[140] Qualitative Research	To evaluate the process and outcomes of service user involvement in the analysis of data	NA	Life limiting conditions, including cancer, COPD	Patients	Consultant Input	Convenience
Cox, 1996[141] Qualitative Research	To describe the findings of a study that explored the psychosocial aspects of participation in early anticancer drug trials from the perspective of the patients.	Structured one on one interview	cancer	Patients	Participant Input	Convenience
Cox, 2000[142] Qualitative Research	To examine patient's perceptions of participating in early phase anti-cancer drug trials	Survey, Structured one on one interview	Advanced cancer	Patients	Respondent , Participant Input, Dialogue	Volunteer
Crowe, 2008[36] Case report	To demonstrate concrete examples of ways in which community members can be involved in all stages of research	Survey, Community meetings	Occupational and environmental health in the Hispanic agricultural community	Patients, Community organizations	Consultant Collaborate	Volunteer
Curry, 2006[143] Qualitative Research	To allow key staff and patients to be involved at all levels of the new nurse-led urgent care team (UCT). The project aimed to evaluate the impact of the UCT	Structured one on one interview	COPD patients	Patients	Respondent Dialogue	Volunteer
Hall, 2009[46] Opinion/ Commentary	To describe the growth of service user or public involvement in health research	NA	Engaging participatory research	NA	NA	NA
Daly, 2009[76] Qualitative Research	Explore ways in which foster care children's values and preferences regarding research in which they would be involved.	Focus group	Foster care	Patients	Stakeholder Dialogue	Convenience

Daugherty, 1995[5] Randomized controlled trial	To describe a survey of cancer patients and their physicians to understand some of the complex issues related to the participation of cancer patients in phase I trials	Structured one on one interview	cancer	Patients	Participant Input	Convenience
Daugherty, 1999[144] Qualitative Research	To understand some of the complex issues related to the participation of cancer patients in phase I trials, and the perceptions of patients toward these trials	NA	cancer	Patients	Subject Consent	NA
Davison, 2008[145] Cross sectional study	To identify factors that patients with prostate cancer believe to be important determinants in their decisions about future enrollment in clinical trials.	Survey	Cancer	Patients	Subject, Participant Consent, Undergo	Random
Decker, 2010[18] Opinion/ Commentary	This article draws on the professional experiences of the authors as well as published examples of international participatory health research with women	NA	Participatory health research	NA	NA	NA
Dellson, 2011[146] Qualitative Research	To describe patients' opinions about the written information used in 3 clinical trials for breast cancer	Survey Focus group	breast cancer	Patients	Participant Undergo, Input	Convenience
Dencker, 1986[147] Cross sectional study	To determine if research related values and priorities differed between patients and controls	Survey	Mental health	Patients	Participant Review	Convenience
Dixon-Woods, 2006[148] Qualitative Research	To explore trial participants’ responses to receiving a summary of the results of a trial in pregnancy.	Structured one on one interview	ORACLE trial of antibiotics for preterm labor and preterm rupture of the membranes	Patients	Respondent Dialogue	Volunteer
Donovan, 2002[77] Systematic Review	To assess how users’ experiences of participating in cancer trials can be gathered and whether they can and should be used to improve the design of trials	Secondary data analysis	Cancer	NA	NA	NA
Doyle, 2010[19] Case report	To learn from a CBPR project.	Survey, Focus group	Elderly people	Patients	Respondent Undergo	Convenience, Volunteer
Dunn, 2001[149] Systematic Review	A systematic review of the effectiveness of interventions to improve informed consent	NA	NA	Patients	Subject Consent	NA
Edwards, 2011[8] Randomized controlled trial	To demonstrate how consulting parents about the design of a study led to the design and successful delivery of a Randomized controlled	Structured one on one interview	Cerebral palsy	Surrogate	Participant Input	Convenience

	trial of osteopathy for children with cerebral palsy					
Ellen, 2010[150] **Opinion/ Commentary**	To present the challenges of community engagement in the development of biomedical HIV prevention clinical trials	NA	HIV	NA	NA	NA
Eng, 2005[151] **Qualitative Research**	To examine the difference between the reasons for accepting and declining participation in a two-arm active treatment Randomized controlled trial comparing external beam radiation therapy vs. cryapy	Structured one on one interview	prostate cancer	Patients	Participant Input	Convenience
Evans, 2011[34] **Opinion/ Commentary**	To explore the role of 'community researcher' and the experiences of those involved.	NA	Older people in care homes	Patients	NA	NA
Featherstone, 1998[152] **Qualitative Research**	To explore trial participants' understandings of randomization.	Structured one on one interview	Benign prostatic disease.	Patients	Participant Dialogue	Convenience
Fern, 2011[97] **Qualitative Research**	To involve young people in research. the report was focused on how the young people were involved to inform each stage of the action research	Participatory group methods	Involvement of young people in research	Young people	Participant Dialogue	NR
Forbes, 2010[153] **Qualitative Research**	To assess women's views of the design of randomized controlled trial	Focus group	breast cancer	Patients	Respondent Undergo	Convenience
Freysteinson, 2010[79] **Case report**	To describe the use of a community consultation ethical framework in the pre-research stage of investigating the experience of viewing oneself in a mirror after mastectomy	Structured one on one interview, telephone and e mail communication, group forums	Breast cancer	Patients, nurses and health care providers	Respondent Input	NR
Garber, 2007[154] **Qualitative Research**	To develop a questionnaire that measures attitudes and concerns about HIV treatment trials among HIV-infected African Americans. To determine actual participation rates and willingness to participate in future HIV treatment trials among HIV-infected African Americans	Survey	HIV-infected African-American adults	Patients	Respondent Input	Volunteer
Gracia, 2010[50] **Systematic Review**	To develop a strategy to include patients opinion in the development of clinical guidelines	Focus group, Structured one on one interview	Anxiety, insomnia, autism and stroke	Patients, Surrogate	Partner Collaborate	Convenience
Gittelsohn,	To develop a community-based chronic disease	Structured one on	Community	Patients,	Consultant	Convenience

2010[155] Qualitative Research	prevention program for Inuit in Nunavut, Canada	one interview	involvement	community leaders and members, health and social service staff	Collaborate	
Gooberman, 2008[156] Qualitative Research	To describe challenges and benefits of using citizen's juries as member of the public	Focus group	primary health and social care	Citizens	Consultant Dialogue	Convenience
Hanley, 2001[157] Single cohort	To assess the extent to which consumers are involved in the work of clinical trial in the United Kingdom	Survey	Health research in general	Clinical trial coordinating centers	Participant Input	NR
Happell, 2007[47] Literature Review	To examine the literature relating to consumer involvement in mental health research	Secondary data analysis	Psychiatric diseases	Researchers	Researcher NA	NA
Harper, 2000[41] Qualitative Research	To present a model to address the interactional process that occurs while developing a CBO.	CBO collaborative partnership model	AIDS/HIV	Patients	Partner, Researcher Collaborate, Generate	NA
Higgins, 2001[158] Qualitative Research	To describe the process of participatory research	NR	urban health issues	Patients	Partner Collaborate	NA
Hsu, 2010[159] Qualitative Research	To provide insight into the full range of meaningful outcomes experienced by patients who participate in clinical trials of complementary and alternative medicine therapies.	Structured one on one interview	Patients who participate in clinical trials	Patients	Respondent Dialogue	Volunteer
Hussain-Gambles, 2004[10] Systematic Review	To Investigate how South Asian patients conceptualize the notion of clinical trials	Structured one on one interview, Secondary data analysis	Health research in general	Patients	Respondent Dialogue	NR
Hutchison, 1998[160] Qualitative Research	To determine how cancer patients perceive phase I clinical trials in reference to trial participation and trial information received.	Structured one on one interview	Cancer	Patients	Participant Input	Convenience
Irani, 2010[161] Qualitative Research	To describe the development of a research protocol on secondhand tobacco smoke exposure and chronic rhino sinusitis for a future population-based case control study using a participatory research model.	Focus group	Secondhand tobacco smoke exposure and chronic rhino sinusitis	Community members, health practioners, and researchers	Partner Dialogue	Convenience

Jenkins, 2002[162] **Cross sectional study**	To examine whether there is a preferred way to describe the randomization process that may facilitate discussions about clinical trials of cancer therapy.	Survey	Cancer	Patients	Subject, Researcher Review, Input	Convenience
Jenkins, 2005[163] **Qualitative Research**	To identify the preferred and most disliked descriptions of randomization found in current cancer patients	Survey	patients from cancer centers throughout the UK	Patients	Respondent Input	Random
Jinks, 2009[26] **Qualitative Research**	To establish a community knee pain forum aimed at engaging stakeholders in design, dissemination and prioritization of knee pain research	Group meeting	knee pain	Patients, health professional, community members, researchers	Stakeholder Dialogue	Convenience
Johnson, 2009[63] **Opinion/ Commentary**	To explore links between the inclusive research aspect of project and the rights of people with intellectual disabilities to participate in their communities	NA	intellectual disabilities	Patients	NA	NA
Johnston, 2008[164] **Case report**	To discuss engaging patients in cancer and palliative care research	Workshop	Cancer	Patients	Subject, Participant, Partner Consent, Review, Input, Collaborate	NA
Johnston, 2008[165] **Opinion/ Commentary**	To elicit the importance of patient involvement in care an research, and to look for innovative ways that nurses can involve patients internationally	NA	Palliative care	Nurse	NA	NA
Jones,2006[166] **Cross sectional study**	To assess cancer patients' knowledge and attitudes towards clinical trials.	Survey	cancer	Patients	Respondent Input	Random
Kamps, 1987[167] **Single cohort**	To discuss parental attitude and perceptions of a child's responsibilities on experimental therapy	Survey	pediatric cancer	Surrogate	Respondent Undergo	Convenience
Karlawish, 2008[168] **Qualitative Research**	To examine the views of Alzheimer disease patients and their study partners on the ethics of proxy consent for clinical research	Structured one on one interview	Alzheimer's disease	Patients, Surrogate	Respondent Dialogue	Volunteer
Karmaliani,2009[39] **Qualitative Research**	To share concerns about maternal depression, partner violence, and child functioning and the goal of offering an intervention (i.e., program) to the community to improve maternal mental health and child functioning	Focus group	Maternal depression and child health	Community leaders, agency directors	Partner Collaborate	NA

Kelly, 2005[169] Qualitative Research	To describe background steps that researchers can use when conceptualizing and initiating a research project with community partners in participatory action research	NA	health research in general	NA	NA	NR
Kelson, 1999[86] Cross sectional study	To identify the extent to which the Cochrane Collaboration involves consumers as members of Cochrane Review Groups	Survey	NA	Cochrane Review Groups	NR	NR
King, 2009[170] Qualitative Research	describes the development of a 33-item, survey questionnaire measuring community members' perceptions of the impact of research partnerships addressing health or social issues	Focus group	Health research in general	Researchers and community members	Consultant Review, Dialogue	Convenience
Kirwan, 2003[171] Qualitative Research	To assess the outcomes of intervention in rheumatoid arthritis (RA) from the perspective of those who experience the disease themselves.	Focus group	rheumatoid arthritis	Patients	Consultant Generate	Convenience
Kirwan, 2005 Qualitative Research	To evaluate outcomes of intervention in rheumatoid arthritis (RA) from the perspective of those with RA	Focus group	Rheumatoid arthritis	Patients	Respondent Undergo	Convenience
Koops, 2002[6] Randomized controlled trial	To determine whether consumer involvement would help to solve some of the ethical problems associated with research into thrombolysis for acute ischemic stroke, with its inherent risk of fatal intracranial hemorrhage	Survey, Focus group	acute ischemic stroke	Patients	Respondent, Participant Review, Input, Dialogue	Convenience
Lammers, 2004[172] Qualitative Research	To describe the establishment and function of a reference group established to guide the conduct of a research project examining the experiences of consumers and careers with psychiatric disability support services.	Focus group	Consumers and carers with psychiatric disability support services	Patients	Subject Collaborate	NR
Langston, 2005[173] Case report	To outline the experiences of an integrated relationship between the organizers of a clinical trial and a consumer organization.	NA	Paget Disease	Patients	Consultant Input	Volunteer
Lavender, 2009[174] Qualitative Research	To explore women's views of participation in a trial of planned cesarean birth vs. planned virginal birth.	Structured one on one interview	Delivery	Patients	Participant Input	NA
Leidy, 2006[175] Opinion/ Commentary	To highlight the importance of patient reported outcomes in device studies	NA	medical devices and technology	NA	NA	NA
Leinisch-Dahlke, 2004[176] Single cohort	To discuss patient preference compared with the expert preference regarding clinic trials and drug therapy	Survey	headache	Patients	Respondent Input	Convenience

Lindenmeyer, 2007[31] Qualitative Research	To determine what makes user involvement successful, effective and meaningful from the researcher standpoint	Semi-structured interviews	Diabetes	Researchers	Researcher Generate	Extensive interaction with advisory group
Llewellyn-Thomas, 1989[177] Qualitative Research	To describe a method to test patients' attitudes towards the possible risks and benefits associated with any new treatment which has been incorporated into a clinical trial protocol.	Structured one on one interview	breast cancer	Patients	Participant Input	Convenience
MacKinnon, 2010[178] Qualitative Research	To describe the methodology and lessons learned from the collaboration with community-based organizations to explore how to measure the difficult-to-measure outcomes of participation in community-based programs	Structured one on one interview	Aboriginal	Patients	Participant Input	Convenience
Madsen, 2000[179] Single cohort	To investigate the preferred extent of written information in clinical trials among potential and actual trial participants	Survey	patients in an outpatient clinic and clinical trial participants	Patients	Subject Undergo	Random and convenience sample
Manson, 2004[180] Qualitative Research	To illustrate successful strategies in working with American and Alaska Native communities in aging and health research	Secondary data analysis	Health research in general	NA	NA	Convenience
Manson, 2007[181] Opinion/ Commentary	To describe recent advances in research on American Indian and Alaska Native	NA	American Indian and Alaska Native	Researchers and communities	NA	NA
Marsden, 2004[4] Randomized controlled trial	To show breast cancer patient involvement in the design of a study	Focus group	Breast cancer	Patients, stakeholder and health professionals	Consultant Input	Convenience
Martin, 2009[80] Qualitative Research	To determine the feasibility of engaging incarcerated women in CBPR and to identify by and with the women the health concerns that needed to be addressed.	Structured one on one interview and group interviews	Incarcerated women	Patients	Researcher Generate	Volunteer
Maslin-Pro, 2003[182] Qualitative Research	To describe the experience of user involvement in health care research drawing on current UK health policy documents and an examination of the factors affecting recruitment to breast cancer clinical trials	Focus group	breast cancer	Patients	Participant Dialogue	NA
Mastwyk,2002[183] Qualitative Research	To assess why the carers of people with Alzheimer's disease seek the participation of their relatives in clinical trials	Survey	Alzheimer's disease	Relative	Subject Consent	Volunteer
May, 2008[32]	To integrate research knowledge and skills into	NA	Social topic	Members of a	NA	NA

Opinion/ Commentary	each CBO's organizational structure and programmatic design			community based organization		
McCauley, 2001[68] Opinion/ Commentary	To describe a community participatory model to characterize the degree of exposure to pesticides and potential health effects in migrant farm workers and their children.	NA	exposure to pesticides and potential health effects	Patients, community organization members	NA	NA
Mc Comber, 1998[184] Opinion/ Commentary	To describe a community participation project to prevent diabetes in Canadian aboriginal school children	NA	diabetes prevention	Community members	NA	NA
McLaughlin,2006[62] Literature Review	to contribute to the debate concerning the benefits and costs of involving young service users in research	NA	Health research in general	NA	NA	NR
McQuiston, 2005[185] Qualitative Research	To generate preliminary data to be used to write a grant proposal	Focus group	HIV	Community leaders	Consultant Collaborate	Convenience
Mease,2007[186] Qualitative Research	To assess the core domains assessed in fibromyalgia studies	Focus group	Fibromyalgia	Patients	Respondent Undergo	Convenience
Medd, 2005[187] Qualitative Research	To assess men's experience of prostate biopsy	Survey, Semi-Structured interview shortly after their prostate biopsy	Men for needle biopsy of the prostate.	Patients	Respondent Input, Dialogue	Volunteer
Meropol, 2003[188] Cross sectional study	To describe and compares the perceptions of cancer patients and their physicians regarding phase I clinical trials.	Survey	cancer patients	Patients,	Respondent Undergo	Convenience
Milewa, 2008[189] Qualitative Research	To describe the issues of an advisory group established to provide lay perspectives on the work of the UK's Medical Research Council	Structured one on one interview	Health research in general	Health professionals (researchers, clinicians, and members from health professional organizations)	Respondent Dialogue	Convenience
Millat, 2005[190] Opinion/	To look at alternatives and the potential advantages of adopting more flexible and	NA	Randomized controlled	NA	NA	NA

Commentary	clinically relevant approaches to the design of surgical trials.		design in clinical surgical research			
Mills, 2003[191] Qualitative Research	To explore patients' perceptions of randomization and reasons for consent or refusal to participate in a clinical trial	Structured one on one interview	prostate cancer	Patients	Participant Input	Convenience
Minkler, 2002[192] Qualitative Research	To explore the use of participatory action research by and with a community of people with disabilities in addressing a polarizing issue in that community: death with dignity or physician-assisted suicide legislation.	Structured one on one interview	patients with substantial physical disabilities	Patients, community members	Participant Input	NR
Minkler, 2010[193] Opinion/ Commentary	To explore a successful community-based participatory research and organizing effort and its contributions to both local policy outcomes and changes in the broader policy environment	Focus group, Structured one on one interview	Community involvement	Community leader, community health promoters	NA	NA
Minogue, 2010[194] Case report	Describe current state of patient involvement in research in UK	NA	All research in UK	Patients, Researcher	Consultant, Researcher Collaborate	NA
Mirand, 2004[195] Literature Review	To describe the factors that influence partial Randomized controlled trial participants' preferences with regard to the allocation procedure and the treatment options.	Structured one on one interview	insomnia	Patients	Respondent Undergo	Convenience
Mockford, 2012[15] Systematic Review	To identify the impact of patient and public involvement on UK National Health Service healthcare services and to identify the economic cost	NA	Patient and public involvement on UK NHS health care	Online databases	NA	NR
Moreno-Black, 2004[196] Qualitative Research	To describe the results of a study that was designed to examine some of the ways in which participants in a randomized double blind clinical trial perceived their participation in the clinical trial	Structured one on one interview	HIV	Patients	Participant Input	Convenience
Morin, 2008[22] Qualitative Research	To understand the evolution of community advisory boards and community partnerships at international research sites conducting HIV prevention trials	Focus group	HIV prevention	Community advisory board members	Consultant Collaborate	Volunteer
Morrow, 2010[30] Opinion/ Commentary	To describe a model of quality involvement that could be used by research teams	NA	Health research in general	NA	NA	NA
Murad, 2010[78]	To examine patients' preferences regarding the	Survey	Diabetes	Patients	Respondent	Random

Qualitative Research	design of diabetes trials.				Input	
Nair, 2004[197] Qualitative Research	To explore the consent preferences of patients whose health data are currently being used for research purposes.	Structured one on one interview	Health research in general	Patients	Participant Input	Convenience
Nilsen, 2010[11] Systematic Review	To assesses the effects of consumer involvement and compare different methods of involvement in developing healthcare policy and research, clinical practice guidelines, and patient information material.	Secondary data analysis	Health research in general	NA	NA	NA
Noe, 2007[198] Single cohort	To evaluate whether community-based participatory research principles might influence an individual's decision to participate in research	Survey, Focus group	American Indian	Patients	Respondent Undergo, Dialogue	Convenience
Nolan, 2005[199] Opinion/ Commentary	Reform nursing research to include patients as informants	NA	Research involving nurses	Nurse-researcher	NA	NA
Ntshanga, 2010[66] Qualitative Research	To present a framework which describes the establishment and benefits of the community advisory board in the Inanda, Ntuzuma and KwaMashu area	Focus group	Tuberculosis	Patients	Stakeholder Review, Generate	Convenience
Oliver, 2004[9] Systematic Review	To look at the process and outcomes of identification and prioritization and to identify the barriers and facilitators to meaningful participation by consumers	Secondary data analysis	Health research in general	Reports	Participant Researcher Review, Input	NA
Oliver, 2008[45] Literature Review	To describe the development of a multidimensional conceptual framework capable of drawing out the implications for policy and practice of what is known about public involvement in research agenda setting	NA	Health research in general	Patients	NA	NA
Ong, 2003[200] Qualitative Research	To involve users in the design of a research project that aims at describing a 12-month course of low back pain in an adult population sample, and to determine how patient and professional perceptions of low back pain and its treatment relate to the use of health-care and to subsequent outcome	Focus group	back pain	Patients	Consultant Dialogue	Convenience
Oppenheim, 2005[201] Opinion/ Commentary	To present and discuss the complex relational, psychological and ethical issues pediatric phase I/II trials based on an interview with the mom of a child enrolled in such a trial	Structured one on one interview	Pediatric oncology	Surrogate	NA	NA

Oscos-Sanchez, 2008[38] Opinion/ Commentary	Opinion/ Commentary on coalition designed to conduct and evaluate school-based violence prevention program	NA	Elementary school violence prevention program	Researchers	NA	NA
Paul, 2011[202] Qualitative Research	To identify the perceptions of professionals, patients and carers regarding prioritizing psychosocial research efforts about hematological cancers	Survey	NM	Patients, carers and health professional	Input Collaborate	NR
Plumb, 2008[203] Qualitative Research	To determine the relationship between the collaborative process of conducting the Community Research Collaboration projects and reported outcomes	Semi-structured interviews	Projects with underserved populations	Surrogate	Researcher Collaborate, Generate	All significant members
Potvin, 2003[204] Opinion/ Commentary	To describe 4 principles as basic components for an implementation model of Aboriginal community programs.	NA	diabetes	Community members	NA	NA
Read, 2009[205] Qualitative Research	To address the factors that influence young adults decision to participate in health care research	Survey	cancer	Patients, Surrogate	Respondent Undergo	Random
Read, 2011[206] Opinion/ Commentary	To explore the challenges to researchers intending to involve vulnerable populations in health and social care research	Focus group	vulnerable population	Patients	NA	NA
Reddy, 2010[207] Qualitative Research	To examine within the South African HIV vaccine clinical trial environment: the purpose of the CABs; the structure and representivity of the CABs; the scope of power and authority of the CABs; and the level of independence of the CABs.	Focus group	HIV patients	Principal investigators, counselors, community liaison officers, recruiters and CAB members.	Participant, Stakeholder, Consultant Input, Dialogue	Volunteer
Redwood, 2010[33] Qualitative Research	To evaluate the use of community-based participatory research principles led to more effective study design and implementation in a study in Alaska	Survey	health disparities	Surrogate	Consultant, Partner Collaborate	Random
Repper, 2006[208] Opinion/ Commentary	To Explore the User-Focused Monitoring approach	NA	Mental health	Experts in the topic	NA	NA
Rhodes, 2001[65] Opinion/ Commentary	To explore some of the issues raised by patient involvement	NA	Diabetes	Patients	NA	NA

Richards, 2002[209] Qualitative Research	To place community involvement at a high priority in the development of health intervention research through a NIH funding initiative in the Washington DC area	Focus group	Studies focused on infant mortality and low birth weight outcomes	Researcher	Researcher Generate	NA
Roberts, 2004[210] Qualitative Research	To describe how individuals with serious mental illness and psychiatrists view ethically important aspects of biomedical research participation	Structured one on one interview	schizophrenia	Patients, psychiatrists	Participant Input	Convenience
Roberts, 2002[211] Qualitative Research	To examine the perspectives of schizophrenia patients and psychiatrists regarding medication washouts and placebo treatment conditions in clinical researches.	Structured one on one interview	schizophrenia	Patients	Respondent Input	Convenience
Roberts, 2000[212] Qualitative Research	To examine the perspectives and preferences regarding ethically important aspects of recruitment, consent, and debriefing of people with schizophrenia who volunteered for research protocols	Structured one on one interview	schizophrenia	Patients	Participant Input	Convenience
Roe, 1995[213] Qualitative Research	Document experience of grandmas as primary caregivers to grandchildren	Semi-structured interviews	Grandmothers serving as primary caregivers	Patients, Researchers and community advisory board	Partner Dialogue	Convenience
Rogers, 1994[214] Qualitative Research	To determine the residential needs and preferences of two important groups: primary consumers of mental health services and family members of consumers.	Survey	Mental health	Patients and family members	Input Dialogue	NR
Rosen, 2007[215] Qualitative Research	To explore whether overall satisfaction with clinical care was a function of participation in research.	Survey	Persons with mental illness	Patients	Respondent Input	Convenience
Ross, 2005[216] Qualitative Research	To evaluate the perceptions of risk of falls of older people	Focus group	Risk of falls	Patients	Respondent Undergo	Convenience
Salcido, 1996[217] Opinion/ Commentary	To describe the empower model to involve the disabled to health care decision making.	NA	Patient with disabilities	Patients	NA	NA
Saurbrey, 1984[218] Qualitative Research	To evaluate patient's attitudes towards medical trials	Structured one on one interview	Research involving humans	Patients	Subject Consent	Random

Savage, 2006[219] Qualitative Research	to explore the culture of pregnancy and infant care among African American women	Focus group	Social topic	Patients, Nurses in the community	Partner Dialogue, Collaborate	Random
Sayers, 2008[101] Qualitative Research	To understand patient groups' perceptions and experiences of the Roche clinical trials registry	Survey	Oncology, osteoporosis, virology	Patients	Respondent Review, Input, Dialogue	NA
Seifer, 2006[220] Opinion/ Commentary	To discuss the challenges of CBPR and how to mitigate them in an urban health setting.	NA	public health research in urban settings	NA	NA	NA
Serrano-Aguilar, 2009[72] Qualitative Research	To describe a study of incorporating patients in developing the early stages of a systematic review process	Delphi methods	degenerative ataxias	Patients	Participant Dialogue	Convenience
Shagi, 2008[3] Randomized controlled trial	To investigate the feasibility of a participatory model of community liaison among an occupational cohort of women at high-risk of HIV and sexually-transmitted infections in Mwanza City, northwest Tanzania in preparation for a Phase III vaginal microbicide trial	Meetings with community members	HIV high risk population	Patients	Stakeholder Input	NR
Shilling, 2011[221] Qualitative Research	to optimize recruitment of children to clinical trials	Survey, Structured one on one interview	clinical trials in general	Relative, practitioners	Respondent Undergo	Convenience
Slomka, 2008[222] Qualitative Research	To explore motivations of underserved African American drug users to participate in research	Structured one on one interview	HIV/AIDS	Patients	Subject, Participant Consent	Convenience
Smith, 2007[223] Qualitative Research	To examine African American women's thoughts and perceptions about the clinical research process and about participation in the University of Michigan Women's Health Registry research database	Focus group	African American women	Patients	Participant Dialogue	Convenience
Snowdon, 2006[85] Qualitative Research	To explore the pace of decision-making for 78 parents associated with clinical trials in the UK	Structured one on one interview	recruitment of children in trials	Surrogate	Participant Input	Convenience
Snowdown, 1997[84] Qualitative Research	To describe the views of parents who consented that their critically ill newborn baby should be enrolled in a neonatal trial		Randomized controlled trials involving neonates	Parents	Respondent Input, dialogue	NR
Sood, 2009[224] Single cohort	To assess attitudes of patients about participation in clinical trials	Survey	Health research in	Patients	Respondent Undergo	Convenience

			general			
Staniszewska, 2007[67] Qualitative Research	To involve users in the development of a research bid to examine parents’ experiences of having a pre-term baby	Meetings with patients' parents and researchers	pre-term child	Surrogate	Partner Collaborate	Convenience
Staniszewska, 2011[60] Qualitative Research	to develop the Guidance for Reporting Involvement of Patients and Public checklist to enhance the quality of patient and public involvement reporting	Thematic analysis	Patient and public involvement in research	NA	NA	NA
Stewart, 2006[61] Qualitative Research	To raise the importance of Indigenous participation in the ethical review process	Survey	Research involving humans	Indigenous human research ethics committees	Partner Review	All the committees
Stewart, 2009[35] Qualitative Research	To discuss the participatory development and implementation of a community research workshop, the community and organizational contexts, the content of the workshop, and lessons learned.	Focus group	health research in general	Community members	Partner Collaborate	Convenience
Stewart,2011[59] Systematic Review	To identify important areas of research, question for research and tools for assessment using patients' and clinicians' input	Survey, Focus group, Structured one on one interview	health research in general	Patients, Clinicians	Subject, Participant, Stakeholder, Researcher Review, Input, Collaborate, Generate	NR
Stirman, 2010[225] Qualitative Research	To describe the collaborative process, key challenges, and strategies employed to meet the goals of the first phase of the IP-RISP grant.	Focus group	Depression	Patients	Stakeholder, Partner, Researcher Review, Dialogue	NA
Sugarman,1998[226] Qualitative Research	to determine patients' attitudes about medical research	Focus group	cancer and heart disease	Patients	Respondent Consent, Undergo	Convenience
Sullivan, 2005[227] Qualitative Research	To describe specific ways in which a participatory action research operationalized and how community participation shaped various stages of the research	Meetings with Project Advisory Group	domestic violence	Patients, members from community organizations, community advise	Partner Dialogue	Convenience
Swartz, 2004[2] Randomized	To describe the implementation and baseline data of an inner-city community based	NA	asthma	Surrogate, community	Stakeholder Input	Convenience

controlled trial	participatory research clinical trial designed to test the effectiveness of a pollutant and allergen control strategy on children's asthma morbidity			organization members, school principals, pastor, nun, health professionals		
Thomas, 1999[228] Cross sectional study	To examine the reasons for women's participation in breast screening	Survey	breast cancer	Patients	Respondent Undergo	Convenience
Thornton, 2001[229] Opinion/ Commentary	To discuss the beliefs that provision of good quality information is the key to involve lay people in the research process.	NA	Health research in general	NA	NA	NA
Thurston, 2005[230] Qualitative Research	To describe the theoretical framework developed to evaluate public participation in the context of regionalized health governance.	Survey, Structured one on one interview, case study	Public involvement in health care	Patients	Consultant Collaborate	NR
Timotijevic, 2007[231] Qualitative Research	The objectives of the current research are to evaluate two deliberative methods—citizens' jury and citizens' workshop for both their process and outcome	Survey	Health research in general	Patients	Participant Input, Dialogue	NR
Tischler, 2010[232] Qualitative Research	To explore the experience of both professionals and patients taking part in a research project that strove to be collaborative and patient-centered	Survey, Focus group	patient centered outcome	Patients	Respondent Undergo	Convenience
Tobin, 2002[233] Qualitative Research	To evaluate the level, extent and quality of consumer participation, and to examine differences between services with different resource commitments	Structured one on one interview	Mental health	Patients	Participant Input	Volunteer
van de Bovenkamp, 2009[13] Systematic Review	To present a review of the literature search we performed on patient participation in guideline development	NA	Patient participation in the development of guidelines	Patients	NA	NR
Van Olphen, 2009[234] Qualitative Research	To evaluate the participatory approach in translating scientific findings from two key projects to the public.	Focus group	breast cancer	Researchers, community members, and the hosting organization	Researcher Generate	NR
van Staa,	To evaluate feasibility, benefits and limitations	Structured one on	chronically ill	Patients	Participant,	Convenience

2010[235] **Qualitative Research**	of a participatory research project involving chronically ill adolescents as co-researchers	one interview	adolescents		Researcher Input, Generate	
Walmsley, 2004[64] **Opinion/ Commentary**	To describe the lessons drawn from what is termed 'inclusive' learning disability research where people with learning difficulties (intellectual disability) are involved as active participants	NA	learning disability	Patients	NA	NA
Washington, 2004[236] **Opinion/ Commentary**	To describe the barriers, approaches to overcoming these barriers, and principles for the maintenance of good collaborative research relations.	NA	NA	Members from underserved communities	NA	NA
Wells, 2009[237] **Opinion/ Commentary**	To review evaluation principles for a community-based project	NA	Social topic	Community members	NA	NA
Wersch, 2001[238] **Qualitative Research**	To evaluate consumer's involvement in clinical guidelines development.	NA	Health research in general	Patients	Stakeholder Dialogue	NR
White, 2001[239] **Literature Review**	To present and discusses a model for participatory action research	NA	Two case illustrations of disabling conditions	NA	NA	NA
White, 2002[240] **Opinion/ Commentary**	To explores the value of the disability research process and outcomes, as viewed by the research participants and their peers.	NA	Patients with disabilities	Patients	NA	NA
White, 2004[241] **Literature Review**	To define Participatory Action Research and provide an overview of the characteristics of this approach	NA	patients with disability		NA	NA
White, 2005[242] **Single cohort**	To increase the understanding of decision making about cancer management by men with prostate cancer who have decided to forgo conventional treatment	Survey	prostate cancer	Patients	Stakeholder Dialogue	Convenience
White, 2008[243] **Qualitative Research**	To determine if patients with advanced cancer are interested in participation in palliative care research and the importance of demographic factors in decision making	Survey	Patients with an advanced disease and limited prognosis is	Patients, Relative	Respondent Dialogue	Random
Whitley, 2005 **Opinion/ Commentary**	An academic mental health consultancy reports on key lessons learned from his experience on collaborative with clients in research	Structured one on one interview	Mental health	Patients	NA	NA
Wilson, 2011[244]	To identify suggestions that could enable	Secondary data	Elderly care	Researchers	Researcher	NA

Literature Review	researchers to consider how quality may be evidenced using constructivist principles including the perspectives of older people and their caregivers	analysis			Generate	
Wright, 1997[245] Qualitative Research	To determine the reliability, validity, and responsiveness of the Patient-Specific Index and to compare different methods of combining patients' ratings of the severity and importance of their complaints	Survey	Patients with total hip arthroplasty	Patients	Respondent Input	Convenience
Yuval, 2001[246] Opinion/ Commentary	To examine patients' motivation for participating in clinical trials, their comprehension of the trial's purpose, their perceived clinical benefit, and their interest in participating in future clinical trials.	Survey	Heart failure trials	Patients	NA	NA
Zullino, 2003[247] Qualitative Research	To evaluate the general readiness of psychiatric inpatients to give their consent to different forms of studies and to assess their reasons for accepting or refusing a hypothetical participation.	Structured one on one interview	Psychiatric diseases	Patients	Consultant Dialogue	Convenience

IX.2 Appendix 2: Recommendations about informant engagement made by individual publications/studies

Studies	Recommendation
Engagement process Framework	
Informant selection	
Decker,2010 [18]Dencker, 1986[147] Fawcett, 1994[20] Morrow, 2010[30]	Factors to be considered when selecting informants: 1. Adequate representation of the population or community in which results of research would be applied 2. Informant should be able to engage as early as possible in the process of research 3. Informants need to have certain ability and sufficient background to be able to engage in research 4. Researchers should consider and solve barriers that could decrease the enrollment rate of the informants into the project particularly those that can lead to a sample of participants with biased view
Building reciprocal Relationships	
Evans, 2011[25] Jinks, 2009[26] Lindenmeyer, 2007[31] Morin, 2008[22] May, 2008[32] Redwood, 2010[33] Stewart, 2009[35] Swartz, 2004 [2]	The informants should be engaged as soon as possible into the research process. The engagement time will be regulated/governed/dictated by: Project resources (time, funding, informant's and researcher's skills), project complexity/difficulty degree, project goals and also in which steps informants will be engaged.
Evans, 2011[34] Jinks, 2009[26] Lindenmeyer, 2007[31] Morin, 2008[22] May, 2008[32] Redwood, 2010[33] Stewart, 2009[35] Swartz, 2004[2]	Researchers should see informants as an equal and reliable partner of their team and not as an extra isolated variable to deal and to invest time with. Both parties have to clearly know their tasks and roles, the importance of their efforts and their independence from each other. Also they have to be included in any other steps related with their main role. The partnership should be based on a mutual understanding of partners' needs, capacities and goals
Co-learning	

White, 2001[239] White, 2004[241] White, 2005[242]	1. Researchers should learn about the population needs and informants realities 2. Informant should learn about the research process and methodology 3. The co-learning process should be done during the whole engagement process and should be maintained during the research project
Re-Assessment and Feedback	
Doyle, 2010[19]	The involvement process should be continually evaluated using predefined tools designated for this purpose. This evaluation process should be preferably done by an impartial/unbiased/external evaluator
Preparatory stage	
Agenda Setting and funding	
Karmaliani,2009[39] Richards, 2002 [209] Savage, 2006[219]	Engage community members to inform research agenda to ensure community concerns are met and research does not interfere with community concerns, as well as provide unique insight on groups within the community. Use focus groups to inform protocol development cultivating trusting partnerships, sustaining collaboration, gaining access to key informants, and scheduling focus groups is appreciable.
May, 2008[32]	Investigators should demonstrate utmost respect for every member of each research team and approach every stage. The academic PI must never talk down to or personalize any criticism of the work of CBO participants. The nonacademic PI and academic PI maintain a positive collaborative relationship with one another. Clearly laying out the structure and budget, being transparent about the work that is expected and how the structure addresses the time issues; highlighting the potential benefits of participation; providing some no-strings-attached financial incentives. The use of logic modeling has proven a major asset. PIs take and create opportunities to talk about how research can be integrated as one part of CBO interventions and programming; can enhance and complement an existing culture of systematic inquiry in a CBO; and can enhance the writing of funding proposals, the development of community advocacy efforts, and the development and revision of CBO programs and policies.
Stirman, 2010[225]	Stable consistent and frequent contact with informants creates more experienced informants with higher skill/knowledge
Evans, 2011[34]	• Recruit via a range of settings • Develop written agreement clearly detailing expectations of engagement • Take time to develop relationships built on trust and empathy • Have a single contact person on the research team

	• Provide training to informants • Harness and value informant’s life skills and experience • Provide compensation
Johnston, 2008[164]	In critical health conditions where patients cannot be involved for a long period (limited survival or limited function), consider a cross-sectional design in which consecutive patients are involved at consecutive phases of research
Rhodes, 2001[65] Staniszewska, 2007[67] Chenoweth, 1998[57]	Members of the community/patients should be involved early (drawing up the initial protocol or even the proposal) so that they achieve immediate empowerment; which should be one of the primary aims of research
Jinks, 2009[26]	Establish a disease specific community (forum) aimed at engaging stakeholders in design, dissemination and prioritization of research
Repper, 2006[208]	• Individual service users should be involved at different levels in the research process: question development, data collection and analysis, writing the report and recommendations for change, dissemination and implementation • There should be a commitment to addressing equalities issues • There should be independence (researchers having control over certain areas such as timeline and budgets, with the freedom to write and publish their findings without interference). • There should be a commitment from commissioners and service providers to implement the recommendations.

Swartz, 2004[2]	Core values governing the collaboration between members of the community advisory board and researchers/study community staff: 1. Cultural competence and inclusiveness: investigators and community members recognize, accepts, and celebrate their differences and value and include different community perspectives 2. First do no harm: studies should be drafted so that they are safe and ethical 3. Honesty: conversations between the community and investigators should be frank and honest 4. Confidentiality: private information should be kept confidential 5. Productive use of resource 6. Effective communication 7. Commitment to advocacy 8. Education/co-learning/sustainability 9. Sound science. Core values were established as the framework for all discussions, saved time, and were the basis for continuing cooperation.
Dencker, 1986[147]	Since research related values and priorities differ between patients and non-patients, choosing real patients (as opposed to using hypothetical scenarios/healthy volunteers) is preferred
Curry, 2006[143]	User contributions to a research proposal result in a bid that is more firmly rooted in the reality of parental experience, has more relevant research questions, uses appropriate and sensitive methods and has a strong dissemination strategy to reach out to health-care professionals.
Freysteinson, 2010[79]	Incorporating a community consultation experience in the planning phase of research creates a richer environment that enables better understanding of the ethical principles surrounding the protection of human participants and the field of interest
Decker, 2010[18]	Invest necessary time in upfront planning. Initial field work to determine partners, establish roles and responsibilities, and discuss expectations takes place over months, not days. Involve community from the beginning to determine scope of research to better reflect the communities' concerns Include all stake holders to build trust and buy-in, increase community involvement and response rates. Ensure conceptual clarity to reduce misunderstandings and the possibility of raising false expectations. Uphold scientific soundness and be flexible to adapt to new perspectives Define benefits to community Facilitate practical conditions that can enable participation Anticipate the probability of conflict (e.g., participatory research may reveal underlying divisions within a community or differences in opinion)
Edwards, 2011[8]	Consulting parents about the design and outcomes of a study leads to higher enrollment and retention

Execution stage	
Study Design	
Carey, 1992[130] Donovan, 2002[77] Edwards, 2011[8] Irani, 2010[161] Jenkins, 2002[162] May, 2008[32] Saurbrey, 1984[218] Shagi, 2008[3] Stewart, 2006[61] Swartz, 2004[2]	The more efficient way to involve patient in study design is doing it from the very beginning and also considering that the patient have to be involved in any other step if this is related with the study design in some way.
Freysteinson, 2010[79] Stewart, 2006[61]	The patients involved can find extra important potential harms or pitfalls linked with the study design.
Daly, 2009[76] Evans, 2011[34] Martin, 2009[80]	Populations considered in the past as a problematic or unable to be involved in study design could be very helpful and contribute in this step equally effective as other ones.
Daly, 2009[76] Donovan, 2002[77]	The researchers should consider interaction methods other than the willingness to participate

Ågård, 2001[115] Carr, 2003[134] Chenoweth, 1998[57] Forbes, 2010[153] Jenkins, 2005[163] Johnston, 2008[165] Kirwan, 2005[248] Koops, 2002[6] Leidy, 2006[175] Marsden, 2004[4] Murad, 2011[78] Nilsen, 2006[11] Richards, 2002[209] Serrano-Aguilar, 2009[72] Stewart,2011[59] Sullivan, 2005[227] Whitley, 2005[249] Yuval, 2001[246]	The involvement of patients in research will allow the researchers to identify and modify the trial protocol. This may lead to a change in enrollment process and outcome selection.
Subject's Recruitment	
Carey, 1992[130] Snowdon, 1997[84]	If the study involves sensitive issues that need to be address in a personal and intimal way, we suggest one on one interviews or personal surveys as a interaction methods with the informants.
Donovan, 2002[77] Daugherty, 1999[144] Karlawish, 2008[168]	Researchers should invest more time to explain concept such as randomization, use more than one manner to explain the informed consent (written vs. oral explanation, detailed vs. sketchy). This could increase trials enrollment rate.
Data collection	
Kelson, 1999[86]	The Collaboration should develop a more consistent approach to the issue of patient-defined outcomes, including consideration of whether or not: (a) to attempt to identify patient-defined outcomes to inform the review process; (b) to point out, where relevant, the lack of data on patient-defined outcomes in completed trials; and (c) to encourage future trials to collect information on patient-defined outcomes. These points were obtained via a survey

Gracia, 2010[50] White, 2002[240] Whitley, 2005[249] Wright, 1997[245]	Addressing specific needs during the process of data collection will lead to a better data quality and a more efficient process. To obtain this, the authors used focus groups, one on one interviews and survey
Acquadro, 2003[114]	The selection of the included techniques for collecting the outcomes should be justified, based on the domains of interest for the patients, the drug profile, and the hypotheses being tested. This should be based on patient interviews and it should provide adequate assessment of the domains of interest. Psychometric evidence of the questionnaire's reliability and validity must be provided. This information came from a commentary
Data analysis	
Cotterell, 2008[140] Cashman,2008[136] McCauley,2001[68]	Academic and community partners' respective roles in data analysis or interpretation of findings should be determined collaboratively. This will increase the commitment of patients in the process and make it more efficient and feasible
Cashman,2008[136] Crowe, 2008[36] Morin, 2008[22] Whitley,2005[249]	Patients should receive as much information and training as possible. Patients' abilities should not be undervalued and we should avoid over-simplification because it may obscure complex relationships patients need to know
Translation Stage	
Dissemination	
Gracia, 2011[50] Harper, 2000[49] Roe, 1995[99]	The researchers and the informants need to agree on how and where the results will be disseminated. This can include non-traditional forums. Studies used Focus groups and one on one semi-structured interviews
Evans, 2011[25] Gracia, 2011[50] Harper, 2000[49] Morin, 2008[22] Roe, 1995[99] Sayers, 2009[250] vanWersch, 2001[100]	The dissemination method should be personalized and suitable for the patient's characteristics and abilities. Language should avoid technical jargon and non-applied information. It is also helpful to include members of the community to address language and cultural issues. For this purpose, patients' voice was obtained through focus groups followed by one on one structured/semi-structured interviews and community consultation

Harper, 2000[49]	Adequate authorship credit should be given to those individuals intimately involved in the process, regardless of their background or affiliation. In this study, Community consultation was used and acknowledged
Dixon Woods, 2006[251]	Research participants should be routinely provided with the results of studies to show the potential impact of findings. More research is needed to evaluate the process of feedback and assessment of the associated risks, benefits, and costs. In this study one on one interviews were used
Implementation	
Butterfos, 2006[16] Crowe, 2008[29] Gracia, 2011[50] Potvin, 2003[104] Thurston, 2005[106] vanWersch, 2001[100]	The implementation process should involve patients in every step of the way and for this to be successful they need to receive as much information as possible. This will help in maintaining their interest and enthusiasm. In these studies surveys, one on one interviews and community consultations were used. A systematic review by Gracia et al. evaluated the adequate information that patients should receive.
Evans, 2011[25] Ntshanga, 2010[252] Roe 1995[99]	The implementation should have clear priorities and should start with the easiest and more achievable steps according to a methodology decided by the informants and research team as a unit. The fact that several small goals are achieved during a certain period of time will result in better acceptance of the project by the involved community. One on one interviews were used to obtain patient's opinion about this topic.
Gracia, 2010[50] vanWersh,2001[100] Potvin, 2003[104] Harper, 2000[49] Morin, 2008[22]	Appropriate support is critical to facilitate effective patient engagement in the implementation phase, overall providing clear guidance on their roles and responsibilities within the group and ensuring the opportunities to attend training events for all guideline/project development group members. Community consultation and focus groups followed by one on one interviews were used to obtain patients' voice about this topic
Butterfos, 2006[16] Morin, 2008[22] Potvin, 2003[104]	Flexible and creative plans need to be developed to adapt to the changes and problems as they arise. These plans should be tailored by researchers and informants. Studies used focus groups to get a general idea followed by one on one interviews where more specific points were clarified

Thurston, 2005[106]	Involvement of local authorities, who are respected and recognized by the community, may be helpful during the implementation process. This may increase the sense of representation and involvement of the informants leading to a bigger interest and enthusiasm about the project. Community consultation and one on one interviews were used to achieve this point.
Evaluation	
Freyteinson, 2010[75] Morin, 2008[22] Potvin,2003[104] Thuston, 2005[106]	The evaluation process should be constant from the beginning of the project in order to troubleshoot problems and resolve barriers. Evaluation can improve resources use and increase efficiency. Community consultation, group meetings and one on one interviews were used
Harper, 2000[49]	The evaluation of the relationship between informants and researchers diminishes the current conflicts and is beneficial for future projects. This seems to be even more important when researchers are dealing with a community

IX.3 Appendix 3. Relevant sources identified through the environmental scan

Title/source/ author/origin	URL	Content description/conclusions
Forum: Bringing The Patient Voice Into Research Youtube.com PatientsLikeMe	http://www.youtube.com/watch?v=pn5jb-elf2E	The Website encourages disease specific forums in which patients share their symptoms and responses to treatment, encouraging patient initiated research which sets the agenda and provides the pilot data for more rigorous research.
Organization: The Association of Cancer Online Resources, Inc. (ACOR) USA	http://www.acor.org/index.html	ACOR is a non-profit organization incorporated in New York that offers information and support through its integrated system of online discussion groups. It offers access to 159 mailing lists that provide support, information, and community to everyone affected by cancer and related disorders. ACOR creates specific websites and also hosts a growing number of websites, created by: • Patients for patients and caregivers. • Cancer Advocacy Organizations, many of which were created directly from the membership of an ACOR mailing list. • Professional Organizations. The site educate patients and empower them to participate in research (clinical trial FAQ; guide to trial terminology, access to investigational drugs and a guide to find a clinical trial)
Organization: The Patients Voice by Healthcare Landscape UK	http://www.thepatientsvoice.org/	The Patients Voice is run by company that invites patients, their friends, family and carers to participate in market research. Members can interact with each other through a social network forum called the Icare Café.
White Paper: Patient and Public Involvement (PPI) in the Research Process Nathan Shippee, Anna Johnson, Victor Montori (Mayo Clinic, USA)		The paper describes a framework for involving patients in research and defines the potential areas and roles for this involvement
Blog: New Voices for Research	http://newvoicesforresearch.blogspot.com/	A blog that allows direct communication between early career researchers so that they become advocates for communities. No clear description of methods for patient engagement
Organization:The Lupus Research Institute	http://www.lupusresearchinstitute.org/news/discoveries/09_advocacy	A patient was nominated to serve as a “consumer reviewer” for scientific lupus proposals submitted to the Department of Defense “Peer Reviewed Medical Research Program.” That year, the research program received $50 million in appropriations to fund research in 21 research topic areas—including lupus.

Network: Community of practice in ecohealth Canada	http://www.copeh-canada.org/index_en.php	A network made up of individuals and organizations that share common concerns or interest in ecohealth. Their methods could be a model extrapolated to biomedical research. Their vision: - to build the community's foundation with the support of the three initiating university's expertise on the Ecohealth approach; an expertise that hinges on research projects, education, and development work with Latin American, African and Asian partners; -to support the progressive development of the community of practice through the participation of colleagues, partners and collaborators interested in the ecosystem approach to health; -to support the development of regional groups and sustainable studies that aim to inform research, education and public policy about the contributions and ideas that result from ecosystem approaches to health
Organization: National Institutes for Health Research (NIHR) UK	http://www.nihr.ac.uk/awareness/Pages/default.aspx	The organization vision is that Involving patients and members of the public in research can lead to better research, clearer outcomes, and faster uptake of new evidence. Goals: • Set research priorities • Identify the important questions that health and social care research needs to answer • Give their views on research proposals alongside clinicians, methodologists, scientists, and public health and other professionals • Help assess proposals for funding • Take part in clinical trials and other health and social care research studies, not just as subjects but as active partners in the research process • Publicize the results. • Available structures to fulfill goals: • All the NIHR's Research Programmes actively engage patients and the public in all stages of research. The NIHR coordinating centres recruit members of the public to help in the commissioning and reviewing of NIHR research proposals. • The funding of INVOLVE which promotes active public participation in NHS, public health and social care research to improve the way that research is prioritised, commissioned, undertaken, communicated and used. • The establishment of the Clinical Research Network across England to increase the number of people recruited onto clinical trials and to the validity of their results.

Organization: INVOLVE UK	http://www.invo.org.uk/	A national advisory group created and funded by the National Institute of Health Research and supports greater public involvement in public health and social care research. Thirty members help to identify issues that need to be addressed by INVOLVE and also to disseminate new ideas on policy and practice. Vision: -maximize the opportunities for public involvement -learn and share knowledge and experiences of public involvement -support inclusion, diversity and equity and the active involvement in research of groups and individuals who are often excluded from research Rich site with a database of published and unpublished research projects in the field of health, public health and social care that have or plan to actively involve members of the public as partners in the research process.
Organization: James Lind Alliance UK	http://www.lindalliance.org	The alliance facilitates Priority Setting Partnerships by bringing patients, carers and clinicians together to identify and prioritize for research the treatment uncertainties which they agree are the most important. Vision: -addressing uncertainties about the effects of treatments should become accepted as a much more routine part of clinical practice -patients, carers and clinicians should work together to agree which, among those uncertainties, matter most and thus deserve priority attention Workflow: The alliance facilitates the collaboration between an organization and the patients and helps create a Steering Group, which comprises both clinical and patient representation who sign a Protocol setting out their aims for the Partnership, and their agreed commitments. The key components of a priority setting process are: ○ engaging with patients, carers and clinicians to participate in the Partnership ○ collecting patients', carers' and clinicians' treatment uncertainties, then checking them ○ reviewing existing research recommendations to identify uncertainty ○ working with patients, carers and clinicians to prioritise the uncertainties, and agreeing a ranked top 10 questions for research to address Includes bibliography of research reports about patients', clinicians' and researchers' priorities for new research (identified through a fairly rigorous scoping study)

Book: Patients, the public and priorities in healthcare, **Edited by Peter Littlejohns and Michael Rawlins.** Radcliffe, 2009.	http://www.lindalliance.org/Publications.asp	Focus on public involvement in agenda setting agenda setting
Organization: Patient Voices Network	http://www.patientvoices.ca/	At the core of the Patient Voices Network is the belief that patients affected by a health care decision should be involved in the process of reaching that decision.
Organization: International Association of Public Participation	http://www.iap2.org/	An association that advocates for public participation as means to involve those who are affected by a decision in the decision-making process and it communicates to participants how their input affects the decision. The association provides training and professional development (certificate in public participation). The focus is not research and not healthcare.
Organization: National Cancer Institute USA	http://outcomes.cancer.gov/areas/pcc/communication/monograph.html	The focus is on Patient-Centered Communication in Cancer Care. No clear descriptions of methods of engagement in research
Organization: the Community Impacts of Research Oriented Partnerships (CIROP) Measure Canada	http://impactmeasure.org/about.htm	Researchers from five community-university research partnerships from Ontario, Canada, joined together to develop a reliable and valid survey to measure the community impacts of research partnerships between universities and community agencies. **The CIROP Measure is a 33-item, generic measure of community members' perceptions of the impact of research partnerships. It** can be used to: 1. assess the effectiveness of knowledge sharing approaches, 2. determine the most influential activities of partnerships, 3. determine structural characteristics of partnerships associated with various types of impact, and 4. demonstrate accountability to funding bodies.
Organization: Civic Change (Pew Partnership for Change) USA	http://www.pew-partnership.org/whoweare.html	An example of community-university Partnership. This civic research organization provides consulting and program support to communities, governments, foundations, and nonprofit agencies to help clients identify and implement solutions and strategies crucial to making communities stronger.
Organization: Health Issues Centre Australia	http://www.healthissuescentre.org.au/subjects/list-library-subject.chtml?subject=44	An independent, not-for-profit organization that promotes equity and consumer perspectives in the Australian health system. Its mission is to improve the health outcomes especially the disadvantaged. Activities: -policy analysis and advocacy from consumer and equity perspectives -consumer-focused research

		-promoting and supporting consumer participation -disseminating information. Model: The organizations is approached by systems/clients in need of consumer perspective and link them to database of available consumers
A Model Framework for Consumer and Community Participation in Health and Medical Research. National Health and Medical Research Council. Commonwealth of Australia 2005. Australia	http://www.healthissuescentre.org.au/documents/items/2011/01/360783-upload-00001.pdf	A detailed model is described in 57 page document providing a framework for consumer and community participation in health and medical research The model describes the current practice and the proposed one (involving the public) in every step of the research process.
Workshop: Consumer and Community Involvement in Research, a course for researchers. University of Western Australia workshop	http://www.sph.uwa.edu.au/courses/winter-spring-summer-school/research-involvement	This workshop aimed at increasing awareness of the contribution consumers and community members can make to research; identifying and addressing the barriers to consumer and community participation; developing understanding and skills on the 'how and why' for implementing consumer and community participation; providing information about resources and other sources of help to support consumer and community participation). December 2011
Abstract: Typology of Public Participation Mechanisms. Lynn J. Frewer	http://sth.sagepub.com/content/30/2/251.	An attempt to define the key concepts in the domain: public communication, public consultation, and public participation, according to the nature and flow of information between exercise sponsors and participants.
Systematic review: Patient and Public Involvement in Clinical Practice Guidelines: A Knowledge Synthesis of Existing Programs Medical Decision Making November 1, 2011 31: E45-E74	http://mdm.sagepub.com/content/31/6/E45.full.pdf+html	Systematic review that aims at defining the key component of engaging patients in guideline development.
Website: Community campus partnership for health	http://ccph.info	Useful resources for community based participatory research with multiple toolkits and resources
Curriculum: Developing & sustaining community based participatory research University of Washington, USA	http://depts.washington.edu/ccph/cbpr/u1/u13.php	A curriculum for developing and sustaining community based participatory research focus on building these skills including how to get started, building partnership, financial perspectives and dissemination ideas.
Book/online: Community Engagement: Guide from the Centers for Disease Control and	http://atsdr.cdc.gov/communityengagement	Manual with multiple chapters on community engagement, including principals

Prevention.		
Article: Power relations in qualitative research. Karnieli-Miller O, Strier R, Pessach L. Qual H Research 2009: 19(2); 279-89.	Expert contact	The study describes research participants roles and the corresponding spectrum of power relations (high egalitarian, equal partnership, low/high hierarchical).
Presentation: Margo Michaels. Agency for Health Care Research USA	http://www.ahrq.gov/about/annualmtg08/091008slides/Michaels.htm	Using EPC Reports to Promote Changes in Research, Practice and Policy: A Perspective from Two Non-governmental Organizations. 2008.
Unpublished article: Stakeholder participation in comparative effectiveness research: defining a framework for effective engagement. In Press. Journal of Comparative Effectiveness Research. Patricia A Deverka et al.	Expert contact	Development of a stakeholder definition and a conceptual framework.
Unpublished article: Facilitating Comparative Effectiveness Research in Cancer Genomics: Evaluating Stakeholder Perceptions of the Engagement Process. Patricia A Deverka et al.	Expert contact	Questionnaire and subsequent interviews to evaluate engagement.
Unpublished article: Stakeholder Engagement in CER: How will we measure success? Danielle C. Lavallee and Patricia A Deverka et al.	Expert contact	This is a review article that presents principals for the evaluation of the stakeholder experience in engagement in comparative effectiveness research (how effective and meaningful engagement practices were?). Authors recommend the inclusion of 6 meta-criteria that represent normative goals of multiple studies: respect, trust, legitimacy, fairness, competence and accountability

Database: PubMed Health USA	http://www.ncbi.nlm.nih.gov/pubmedhealth/	PubMed Health specializes in reviews of clinical effectiveness research, with easy-to-read summaries for consumers as well as full technical reports. PubMed Health is a service provided by the National Center for Biotechnology Information (NCBI) at the U.S. National Library of Medicine (NLM). This represents a good tool for patients to become more informed. No recommended methods of engagement; however.
Center: Center for Shared Decision Making USA	http://patients.dartmouth-hitchcock.org/shared_decision_making.html	Indirectly relevant resource. The focus is on shared decision making in clinical context. One-on-one counseling sessions for any medical condition are provided along with a Decision Aid Library of helpful videotapes, audiotapes, booklets, CD-ROMs, and websites and a Health Care Decision Guide
Center: Mayo Clinic Shared Decision Making National Resource Center USA	http://shareddecisions.mayoclinic.org/	Potentially relevant resource. The focus is on shared decision making in clinical context. However, trials of decision aids are frequently conducted with heavy input from patients and designers, as well as an advisory patient group. Mission: Developing and evaluating patient decision aids Contributing to defining international decision aid standards Defining high performance organizations using shared decision making Educating and training care providers in communication techniques Adopting and using patient decision aids at the point of care Contributing to statewide implementation efforts Certifying patient decision aids through collaboration with external partners
Center: Society for Participator Medicine USA	http://participatorymedicine.org/	Potentially relevant resource. The focus is on shared decision making in clinical context. It is a cooperative model of health care that encourages and expects active involvement by all connected parties (patients, caregivers, healthcare professionals, etc.) as integral to the full continuum of care. The 'participatory' concept may also be applied to fitness, nutrition, mental health, end-of-life care, and all issues broadly related to an individual's health.
Center: CARE: Community Alliance for Research and Engagement. Yale Center for Clinical Investigation. USA	http://care.yale.edu/index.aspx	The alliance fosters rigorous community-based research and to translate scientific breakthroughs into practical benefits for residents of New Haven. In partnership with local health centers and hospitals, the city of New Haven, community organizations, businesses, faith communities, and other parts of Yale University, this School of Public Health program brings together diverse people and organizations to improve health in the city of New Haven. Its cornerstone project, Community Interventions for Health (CIH), is an international collaborative of the Oxford Health Alliance that addresses chronic disease risk factors—unhealthy diet, physical inactivity and tobacco use— through policy and structural initiatives and health promotion programs.
Organization: Healthcare Financial Management Association	http://www.hfma.org	The focus is mostly from management/financial point of view

Report: Community-based Participatory Research: Assessing the Evidence. Agency for Healthcare Research and Quality USA	http://www.ahrq.gov/downloads/pub/evidence/pdf/cbpr/cbpr.pdf	AHRQ Evidence Report 99: The reports defines community-based participatory research (CBPR) as a collaborative approach to research that combines methods of inquiry with community capacity-building strategies to bridge the gap between knowledge produced through research and what is practiced in communities to improve health. Key questions: What defines community-based participatory research? How has CBPR been implemented to date, What is the evidence that CBPR efforts have resulted in the intended outcomes? What criteria and processes should be used for review of CBPR in grant proposals?
Center: Institute for Participatory Action Research & Design	http://web.gc.cuny.edu/che/start.htm	The website describes several projects with clear patient engagement. The projects seek to reveal theoretically and empirically the contours of injustice and resistance and challenge the traditional conceptions of "expert knowledge. The focus is on youth.
Center: Center for Participatory Action Research	http://cadres.pepperdine.edu/ccar/define.html	(a) The website describes the goals of Action Research as: The improvement of professional practice through continual learning and progressive problem solving; A deep understanding of practice and the development of a well specified theory of action; An improvement in the community in which one's practice is embedded through participatory research. A model for learning cycles and feedback is described.
Website: Learning for sustainability	http://learningforsustainability.net/research/action_research.php	Numerous Participatory Action Research projects and methodologies are described. A relevant publication (Stakeholder Participation for Environmental Management: A Literature Review) points to the need to focus on participation as a process. It then identifies a number of best practice features from the literature. Finally, it argues that to overcome many of its limitations, stakeholder participation must be institutionalized, creating organizational cultures that can facilitate processes where goals are negotiated and outcomes are necessarily uncertain. The paper acknowledges that seen in this light, participatory processes may seem very risky, but there is growing evidence that if well designed, these perceived risks may be well worth taking.
National Resource Centre for Consumer Participation	http://www.healthissuescentre.org.au/subjects/list-library-subject.chtml?subject=44	This website includes major policy documents and history of consumer participation. A Clearinghouse for information on consumer feedback and participation methodologies
Book: Avard D etal. Research Ethics Boards and Challenges for Public Participation. Health Law Review, 2009;17(2-3) 66-72	E-book: http://www.amazon.com/Research-ethics-boards-challenges-	Ethical focus.

	participation/dp/B002F1RB2W	
Organization: Health Quality Improvement Partnership UK	http://www.hqip.org.uk/patient-and-public-engagement-2/	The organization was established in 2008 to promote quality in healthcare. They consider effective patient and public involvement/engagement in clinical audit to be fundamental to good quality improvement practice. The website contains templates and case studies
Project: Patient Partner Europe	www.patientpartner-europe.eu	A three year project funded by the European Commission and ended in 2011 and aimed to promote the role of patient organizations in the clinical trials context PatientPartner was based on the belief that involving patient organizations as equal partners at all stages of clinical trials contributes to research that is better adjusted to the real needs of patients. The study looked closely at the part that patient organizations play and are willing to play in clinical trials and also focused its attention on clinical trials with children, the use of biobanks and ethical issues. The main objectives of the project were; • To identify best practices of patient organization's active involvement in clinical research in Europe. • To facilitate the dialogue between all involved stakeholders in clinical research as to how to establish this involvement and integral part of clinical research • Develop guidance material to be used by all stakeholders in order to facilitate the future partnerships in clinical research between patient organizations, sponsors and investigators.
NHS - Patient Involvement Toolkit	www.rcn.org.uk/?a=56801	Very relevant guide (toolkit) that describes the principles of patient engagements—mostly based on hypothesis and not empirical evidence
Health Canada Policy Toolkit for Public Involvement in Decision Making	http://www.hc-sc.gc.ca/ahc-asc/pubs/_public-consult/2000decision/index-eng.php	Case studies are available describing various projects that included patient participation, less focused on research
Center for Patient Partnerships. University of Wisconsin-Madison USA	http://www.patientpartnerships.org/research/	The Center for Patient Partnerships directly documents the impact of advocacy services on patients through evaluation, field research, and synthesis of existing empirical studies. Ongoing research activities: State Consumer Assistance/Patient Support Policy: A Comparative Analysis Comparative Analysis of Advocacy and Other Support Services The Social Compact for Advancing Team-Based Care Survivorship Care Planning Consumer Engagement in Health Systems Quality Improvement

CAPOIRA: Increasing patient involvement in research activities Europe	http://www.eurordis.org/content/capoira-increasing-patient-involvement-research-activities	The CAPOIRA project, funded by the European Commission, facilitates the creation of structural links to bring civil society and the scientific community closer together. The main idea is to foster the participation of patient organizations in research activities by increasing their knowledge, skills and capabilities in the two areas of clinical trials (private or public; at national or European level) and EU-funded health research projects. Training is offered to strengthen the capacity of rare disease patients' representatives and empower patients' representatives to advocate effectively for rare diseases at both the local and EU level. Disease specific forums are available. The focus is on rare diseases and orphan drugs.
Organization: National Association for Patient Participation	http://www.napp.org.uk/	The association promotes the proactive engagement of patients through 'Patient Reference Groups' focusing mainly on community engagement and health. Research engagement is limited to policy and agenda setting. Tools used are mainly surveys.
Presentation: Jo Brett, Sophie Staniszewska, Sandy Herron-Marx, Kate Seers, Helen Bayliss, Carole Mockford School of Health and Social Studies, Warwick University	www.rcn.org.uk/__data/assets/pdf_file/0010/380665/2011_RCN	Presentation of framework for patient and public involvement (PPI). Highlights challenges and the need for better reporting of PPI in published journal articles and study reports.
Report: An evaluation of the process and impact of patient and public involvement in the advisory groups of the UK Clinical Research Collaboration	www.ukcrc.org/index.aspx?o=1540	A detailed report describing the collaboration experience with PPI, challenges and recommendations. Clearly, the report identifies the difficulty of assessing the impact of PPI.
Article: Measuring the impact of patient and public involvement: the need for an evidence base. Sophie Staniszewska. Int J Qual Health Care (2008) 20 (6): 373-374.	http://intqhc.oxfordjournals.org/content/20/6/373.full	A call for better reporting of PPI.

Presentation: The Impact of Patient Public Involvement on Clinical Research. Sue Pavitt. Leeds Institute of Health Sciences. University of Leeds	ec.europa.eu/research/health/pdf/event05/sue-pavitt_en.pdf	Description of benefits of PPI using 4 case studies. Domains of benefit presented: Trial designed to take account of patients needs; Trial operations/ logistics made patient friendly; Trial literature simplified ensuring informed consent
Toolkit: Working With Practices and Communitie. Washington's Institute for Translational Health Sciences Clinical and Translational Sciences.	http://www.researchtoolkit.org/home/developing-proposals/working-with-practices-and-communities.html	Partnership-driven Resources to IMprove and Enhance Research (PRIMER). The PRIMER project was funded through an administrative supplement to the University of Washington's Institute for Translational Health Sciences Clinical and Translational Sciences Award (CTSA) UL1 RR025014 from the NIH National Center for Research Resources.
Partnership Self-Assessment tool from the Center for the Advancement of Collaborative Strategies in Health. From the Center for the Advancement of Collaborative Strategies in Health, Division of Public Health, New York Academy of Medicine.	http://partnershiptool.net/	The Center for the Advancement of Collaborative Strategies in Health has developed ideas and tools to enhance partnerships, including an adaptable tool to measure strength and cohesion in a given group. The Partnership Self-Assessment tool, scoring instructions and a coordinator's guide are all freely available for download, along with a published article on the concepts underlying synergy.
Curriculum: Developing and Sustaining CBPR Partnerships: A Skill-Building Curriculum. The Examining Community-Institutional Partnerships for Prevention Research Group. 2006.	http://www.cbprcurriculum.info/	From the Examining Community-Institutional Partnerships for Prevention Research Group. The curriculum is intended as a tool for use by community-academic partnerships that are using or planning to use a Community-based Participatory Research (CBPR) approach to improving health. It can be used by partnerships that are just forming as well as existing partnerships. It is intended for use by health professions faculty and researchers, students and post-doctoral fellows, staff of community-based organizations, and staff of public health agencies at all skill levels.

Patient group: Diabetes Advisory Group. Mayo Clinic, Rochester. USA	http://shareddecisions.mayoclinic.org/stakeholders/diabetes-advisory-group/	A group of community members with diabetes in the Rochester, Minnesota, and surrounding area. This advisory group has been meeting with Mayo Clinic researchers on a monthly basis for the past 7 years to provide feedback on research proposals, participant recruitment materials, surveys, and all areas of proposed and existing research. The members view research through a patient's perspective, which assists in detecting potential barriers and contributes to effective and meaningful research. Members contribute their personal time and practical experiences in living with diabetes, and through their involvement, researchers remain connected to the real world of the patients that will be impacted
Patient group: One Voice Patient & Family Advisory Council. Mayo Clinic, Rochester. USA	http://shareddecisions.mayoclinic.org/stakeholders/one-voice-advisory-council/	This advisory council is created by patients and their families to contribute to the design and operations of cardiovascular clinical services and research. They publish a quarterly newsletter written for and by patients and families.
Organization: The Parkinson Pipeline Project	http://www.pdpipeline.org/	A grassroots group of advocates whose goal is to provide the patient perspective in the treatment development process of Parkinson disease. The group develops a cadre of well informed PD patient consultants. -Promotes policies that accelerate the evaluation, approval of safe, effective, and timely new treatments by providing the unique patient perspectives to industry sponsors and investors, clinical scientists, and government regulatory agencies. -Maintains a comprehensive database of information Tracks the progress of new therapies and diagnostics -Recruits, trains, provides peer supervision, and up-to-date information on the views of grass roots opinion leaders and clinical trial participants to represent authentic patient interests in the regulatory processes of the FDA in cooperation with the FDA's Office of Special Health Issues. -Increases medical provider and patient awareness -Advocates for the rights of clinical trial participants in relation to sponsors as well as researchers and offers participants the confidence of peer support to address grievances with researchers or sponsors
Engaging Stakeholders To Identify and Prioritize Future Research Needs	www.effectivehealthcare.ahrq.gov/reports/final.cfm.	Important considerations regarding stakeholder engagement to prioritize research include use of: 1. Consistent terminology and definitions throughout the process. 2. In-person methods for brainstorming, identifying topics, clarifying issues, and eliciting a deeper understanding. 3. Quantitative methods for prioritizing research.

Evidence summary: The Quality of Patient engagement and involvement in primary care. The King's Fund. UK	http://www.pickereurope.org/Filestore/PIE_reports/project_reports/Quality_Patient_Engagement_Primary_Care_Kings_Fund_July_2010.pdf	The report is excellent in describing patient engagement in their own health care system (clinical focus)

IX.4 Appendix 4. Search strategies

MEDLINE /EMBASE

1. patient participation/ or consumer participation/ or patient advocacy/ or consumer advocacy/

2. patient centered care/ or ((patient* or consumer* or stakeholder* or user* or lay* or client* or citizen* or communit* or public or advoca* or carer* or caregiver* or surrogate* or parent* or relative) adj2 (important or perspective or centered or centred or participa* or collaborat* or partner* or voice* or unvoiced)).mp. [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

3. ((patient* or consumer* or stakeholder* or user* or lay* or client* or citizen* or communit* or public or advoca* or carer* or caregiver* or surrogate* or parent* or relative) adj2 (involv* or represent* or consult* or contribut* or engage* or activat* or opinion* or dialog* or partner* or input*)).mp. [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

4. *patient participation/ or *consumer participation/ or *patient advocacy/ or *consumer advocacy/ or (patient centered care or ((patient* or consumer* or stakeholder* or user* or lay* or client* or citizen* or communit* or public or advoca* or carer* or caregiver* or surrogate* or parent* or relative) adj2 (important or perspective or centered or centred or participa* or collaborat* or partner* or voice* or unvoiced))).ti,ab. or ((patient* or consumer* or stakeholder* or user* or lay* or client* or citizen* or communit* or public or advoca* or carer* or caregiver* or surrogate* or parent* or relative) adj2 (involv* or represent* or consult* or contribut* or engage* or activat* or opinion* or dialog* or partner* or input*)).ti,ab. or patient-centered care/

5. exp evaluation studies as topic/ or exp methods/

6. 4 and 5

7. 6 and (outcome*.mp. or quality of life/ or patient preferences/ or risk assessment/ or patient satisfaction/) [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

8. 6 and ((utilities or values or empiric* or feedback* or communication*).mp. or health priorities/ or research priorities/) [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

9. 6 and exp clinical trials as topic/

10. 6 and (physician-patient relations/ or patient acceptance of health care/)

11. Health Services Research/ or Needs Assessment/

12. 6 and 11

13. 6 and agenda*.mp. [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

14. 4 and *research design/ and (choice behavior/ or cooperative behavior/)

15. 6 and (choice behavior/ or cooperative behavior/)

16. 7 or 8 or 10 or 12 or 13 or 15

17. 16 and ((panel* or jury or juries or forum).mp. or qualitative research/ or interview*.mp.) [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

18. 16 and (recruit* or participat* or "focus group*" or instrument* or scale* or questionnaire* or consultant* or questionnaire* or survey* or interview* or "nominal group" or delphi*).mp. [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

19. 17 or 18

20. 16 and observation*.mp. [mp=protocol supplementary concept, rare disease supplementary concept, title, original title, abstract, name of substance word, subject heading word, unique identifier]

21. (19 or 20) and exp clinical trial as topic/

22. 19 or 20

23. limit 22 to (consensus development conference or consensus development conference, nih or multicenter study or "research support, american recovery and reinvestment act" or research support, nih, extramural or research support, nih, intramural or research support, non us gov't or research support, us gov't, non phs or research support, us gov't, phs)

24. 22 and (*patient satisfaction/ or *consumer satisfaction/ or *patient-center care/ or *patient preferences/)

25. 22 and (technology assessment, biomedical/ or community-based participatory research/ or px.fs.)

26. 21 or 24 or 25

27. 23 and 2

28. 26 or 27

29. limit 28 to (comment or editorial or interview or introductory journal article or legislation or letter or news or newspaper article or patient education handout or retracted publication or "retraction of publication")

30. 28 not 29

31. limit 30 to humans

32. 31 not animals/

33. ((patient* or consumer* or stakeholder* or user* or lay* or client* or citizen* or communit* or public or advoca* or carer* or caregiver* or surrogate* or parent* or relative) adj2 (important or perspective or centered or centred or participa* or collaborat* or partner* or voice* or unvoiced)).ti,ab.

34. ((patient* or consumer* or stakeholder* or user* or lay* or client* or citizen* or communit* or public or advoca* or carer* or caregiver* or surrogate* or parent* or relative) adj2 (involv* or represent* or consult* or contribut* or engage* or activat* or opinion* or dialog* or partner* or input*)).ti,ab.

35. (24 or 33 or 34) and 32

EBM Reviews - Cochrane Methodology Register 4th Quarter 2011 # Searches Results Search Type

1 (participat* adj2 research).mp. [mp=title, abstract, subject heading word] 141 Advanced

2 "CMR: Evaluation methodology - patient involvement".kw. 521 Advanced

3 "CMR: Other methodology - patient based outcome measures".kw. 252 Advanced

4 (design* or planning or priorit* or agenda* or participat*or decision*).mp. and (2 or 3) [mp=title, abstract, subject heading word] 244 Advanced

5 (2 or 3) and (perspective* or preference*).mp. [mp=title, abstract, subject heading word] 128 Advanced

6 5 not 4 78

PsycINFO 1967 to November Week 3 2011 # Searches Results Search Type

1 client participation/ 1087 Advanced

2 experimentation/ or exp consumer research/ or exp interdisciplinary research/ or exp qualitative research/ or exp experimental design/ or exp methodology/ 147651 Advanced

3 1 and 2 128 Advanced

4 1 and (priorit* or participatory or engage* or planning or design* or perspective* or preference*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] 594 Advanced

5 3 or 4 655 Advanced

6 limit 5 to (all journals and human) 545 Advanced

7 1 and (agenda* or involve*).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] 413 Advanced

8 limit 7 to (all journals and human) 336 Advanced

9 6 or 8 646 Advanced

10 9 and outcome*.mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] 155 Advanced

11 9 and (2 or methodol*.mp.) [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures] 124 Advanced

12 10 or 11 252 Advanced

13 *client participation/ and 12 202

S11 S8 and S9 Limiters - Exclude MEDLINE records

Search modes - Boolean/Phrase

View Results (224) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S10 S8 and S9 Search modes - Boolean/Phrase

View Results (1120) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S9 method* Search modes - Boolean/Phrase

View Results (502921) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S8 S2 AND (S6 OR S7) Search modes - Boolean/Phrase

View Results (2571) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S7   (MH "Study Design+")   Search modes - Boolean/Phrase

View Results (407274) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S6   (MH "Research+") OR (MH "Behavioral Research") OR (MH "Medical Practice, Research-Based")   Search modes - Boolean/Phrase

View Results (803685) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S5   S2 and S4   Search modes - Boolean/Phrase

View Results (91) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S4   (MH "Action Research")   Search modes - Boolean/Phrase

View Results (2501) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S3   (MH "Patient Centered Care")   Search modes - Boolean/Phrase

View Results (8236) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S2 (MM "consumer participation" OR "consumer advocacy") Search modes - Boolean/Phrase

View Results (6819) .View Details .Edit .Interface - EBSCOhost

Search Screen - Advanced Search

Database - CINAHL .

S1 input* OR perspective* OR involve* OR planning OR design* OR outcome* Search modes - Boolean/Phrase

View Results (529181)

PubMed

(patient participation/methods[majr] OR consumer participation/methods[majr]) AND (clinical trials as topic[mesh] OR practice guidelines as topic[mesh] or health services research[mesh])

IX.5 Appendix 5. Data extraction form

Study Name

Primary goal of the study (text box)

Design
Demographic of the informants

- Age
- Sex
- Socio-economic status

Whose voice is being incorporated?

- Patients
- Relative
- Surrogate
- Other

Response rate

- No. Patients whose opinion was considered
- No. Patients invited
- Other

How were the informants selected?

- Convenience
- Random
- Volunteer
- Other

Does this study use a patient reported measure (patient reported outcome measure)?

- Yes

    - Survey
    - Focus group
    - Self-reported scale
    - Structured one-on-one interview
    - Internet
    - Other
        - Have this measure been previously validated?
            - Yes
            - No
            - Don’t know
- No
- Don’t know

Procedures for obtaining these voices:

- Training personnel involved
    - Who was trained? (text box)
- How was this training developed
    - Rules for the interaction
    - Pre-design interactions
    - Pre-design context/setting
    - Other

Challenges and questions for patients in research

- Ethics
- Capacity

- Power
- Trust/ Bias/ Representativeness
- Epistemology
- Terminology
- Interaction format
- Evaluation
- Reporting
- Ownership

How was the information used? (All these questions need a free text box to include the results)

- Agenda setting
- Funding
- Procedures
- Study design
- Recruitment
- Analysis
- Dissemination
- Others

Outcomes of incorporating patient voice (effect of the information):

  - Author’s conclusions about the incorporation process and whether it led to a change in the conduct of research
  - Actual data of the effect of the incorporation process

Barriers and Obstacles:

  - Author conclusions
  - Actual data

Congruence of patient opinion/voice

- With whom and how:

Involvement of the subject

- Definitions of the person:
  - Subject
  - Respondent
  - Participant
  - Stakeholder
  - Consultant
  - Partner
  - Researcher
- Person role:
  - Consent
  - Undergo
  - Review
  - Input
  - Dialogue
  - Collaborate
  - Generate

Recommendations:
-Authors recommendations /conclusions of How to incorporate patients' voice into research? –--Actual

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