



How to develop a strategic research agenda for healthcare research

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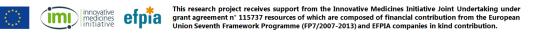
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Foreword

EPI-Net is a leading epidemiological research network established within the <u>COMBACTE-MAGNET</u> consortium with a strong objective to facilitate freely-accessible educational resources generated from its core research activities. EPI-Net has constructed a large publicly-accessible <u>central data repository</u> for epidemiology data on <u>epi-net.eu</u>, which combines multiple sources and allows greater insights into distribution of serious bacterial infections in the human and animal populations across the European Union and European Free Trade Association region and emergence of resistance to new antibiotics at global level. The EPI-Net surveillance platform also hosts the <u>AMR travel tool</u>, a free public service tool, to raise awareness on the antimicrobial resistance risks and prevention measures related to international travel. The interactive tool helps healthcare professionals evaluate travel history and travel-associated risk factors related to antimicrobial resistance as part of their clinical practice and informs travelers on preventive measures to reduce the acquisition and transmission of AMR across borders.

More recently, EPI-Net in collaboration with the <u>JPIAMR ARCH Network</u> published <u>four white papers</u> and a <u>strategic research agenda</u> on the topic of linking antimicrobial resistance and antimicrobial usage surveillance to antimicrobial stewardship. These documents are the product of a meticulous consensus process informed by the results of a state-of-the-art review conducted primarily to explore the tools and techniques available for priority setting exercises. Strategic research agenda help among others highlight critical areas in need of investment, future research, and policies. Systematic and reproducible approaches are essential crutches for priority setting exercises that are integral to developing such agenda. However without prior knowledge and experience, embarking on setting research priorities and roadmaps for healthcare or any research can be a daunting experience. In this latest educational document from EPI-Net, the results of the state-of-the-art review which supported the EPI-Net strategic research agenda are presented with the aim to guide any stakeholder looking for recommendations on how t o develop a research agenda.







A state-of-the art review to inform research priority setting exercises

1.1 Aim

Identify methodologies and tools for preparing research agenda and recommendations on priority setting exercises

1.2 Materials and methods

A rapid state-of-the-art review of published and grey literature

Source

- PubMed database
- Google search engine

Target

- Review or opinion articles with guidance on methods and tools for research prioritization
- Studies reporting research agenda for funding and steps involved in their creation

Search terms

PubMed database:

(research[ti] AND agend*[ti] AND exercise) OR (research[ti] AND priorit*[ti] AND exercise). Filter: 5 years

(research[ti] AND agend*[ti] AND setting) OR (research[ti] AND priorit*[ti] AND setting). Filter: 5 years

Google search engine:

"methods to identify priority areas for funding"
"research priority setting exercise"
"research priority setting exercise tools"
"research priority setting exercise strategies"

Screening

- Only the first page of the search results from Google was screened
- From the overall PubMed search results:
 - 1. Studies without full texts were excluded
 - 2. Title/abstract relevant to the topic of infectious diseases and review articles were screened as priority
 - 3. All other titles/abstracts published between 2019 and 2018 were screened

Data collection

From articles reporting research agenda, data for the following variables were extracted to understand the trend in priority setting exercises:





Reference
Publication year
Торіс
Objective
Participants
Use of established priority setting model (name)
Steps involved (when no priority setting model used)

Data synthesis and reporting

Qualitative data analysis and descriptive reporting was used to describe the results of the review

1.3 Results and inferences

1.3.1 Checklist

Consulting the nine elements of good practice proposed by Viergever *et al.*¹ prior to commencement

helps the execution of a well-structured priority setting exercise:

Preparat	tory work
	Context
	he focus, underlying principles/values, health, research and political environment in which the will take place
	Decision on a comprehensive approach
Decide if	f use of a comprehensive approach is appropriate, or if development of own methods is the
preferred	d choice
	Inclusiveness
Decide w	vho should be involved in the process
	Data collection
Literatur	e scoping for necessary evidence on research questions and gaps and technical data,
consultat	tion of experts, scoping of previously published research agenda, etc.
🗆 F	Planning for implementation
Establish	plans for translation of the priorities to actual research (via policies and funding. Who will
impleme	ent the research priorities (target audience) and how?
	g on priorities
	Criteria
	levant criteria to focus discussion around setting priorities
	Methods for deciding on priorities
	a method for deciding on priorities. Decide whether to use face-to-face discussions or a
	based approach (pooling individual rankings, scores, etc.), or a combination
After pri	orities have been set
	Evaluation
Define w	when and how evaluation of the established priorities and the priority setting process will take
place. He	ealth research priority setting should not be a one-time exercise!
	Transparency
Write a d	clear report that discusses the approach used: Who set the priorities? How exactly were the
priorities	s set?





1.3.2 Priority setting models

Creation of a research agenda in general could be envisioned as a process involving (non-sequentially) idea generation, idea analysis, idea socialization and idea selection.² Over the years, some priority setting models that outline frameworks for this process have emerged as common techniques for designing research agenda.^{3, 4} These include:

•	James Lind Alliance (JLA) method
•	Child Health Nutrition Research Initiative (CHNRI) method
•	Three dimensional (3D) Combined Approach Matrix (CAM) method
•	Essential National Health Research (EHNR) method

The elements of a priority setting model guide users on the choice of stakeholders, steps in identifying research ideas, steps in filtering the right ideas, steps in finalizing, and dissemination. An outline of the workflow described in each of these 4 models is provided in tables 1-4. Table 5 provides a quick comparison of the main aspects of the 4 models.







	Table 1: JLA method ⁵		
	Developed to help set research priorities	in areas of treatment uncertainty	
	Step 1: Identify stakeholders to contribute	A priority setting partnership (PSP) is established composed with equal representation of patients, carers and clinicians. This group agrees the plan of action or 'protocol'	
ldea generation	Step 2: Identify uncertainties	Stakeholders surveyed for questions they have regarding diseases, treatment, etc. and by searching existing literature to find evidence gaps	
ger	Step 3: Summarize responses	All responses sorted to create a list of questions	
ldea analysis	Step 4: Evidence checking	The list checked is against literature evidence to ensure they are true uncertainties. Any questions that have already been answered by research are removed	
ion and n	Step 5: Interim Priority setting	Stakeholders vote on the most important questions in an interim priority setting survey. This is usually via an online ranking survey	
ldea socialization and selection	Step 6: Workshop	The highest ranked 25-30 questions from the interim priority setting survey are discussed in a workshop to agree (adapted nominal group technique; chaired by JLA adviser) on the 'Top 10' list of priorities	
	Step 7: Dissemination	Top 10 is announced and published on the JLA website and promoted to researchers and funders. Possible publication of full report or articles about PSP findings. Continued long- term promotion of research priorities. Long-term tracking of impact of PSP	



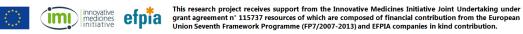




	Table 2: CHNRI method ^{6,7}				
	Recommends that research agenda should target international agencies, large research funding donors, national governments and policy makers. Should aim at reducing the burden of disease within the context of the investments being made				
.s	Step 1: Define context and criteria	Process managers define context and criteria (and provide evidence if necessary) to assess competing research priority options. Examples of criteria, i) answerability ii) attractiveness iii) novelty iv) potential for translation v) effectiveness vi) affordability vii) deliverability viii) sustainability ix) public opinion x) ethical issues xi) potential impact on disease burden xii) equity xiii) community involvement xiv) cost and feasibility xv) enterprise generation. Longer the criteria the greater the possibility of overlap reducing their usefulness as independent criteria			
ldea analysis	Step 2: Refine criteria	A limited set of the most useful and important criteria selected. CHNRI recommends i) answerability, (ii) effectiveness, (iii) deliverability, (iv) maximum potential for disease burden reduction, and (v) the effect on equity			
	Step 3: Involve technical experts	Technical experts (TEs) to be invited to carry out steps 4-6			
	Step 4: Develop means to assess the likelihood that proposed health research options will satisfy selected criteria	Stakeholders develop 3 supporting questions (simple yes or no) addressing selected criteria and are used for judging potential research options. Example under effectiveness, "Based on the best existing evidence and knowledge, would the intervention which would be developed/improved through proposed research be efficacious?"			
ldea generation	Step 5: Identify research options	A systematic list of competing research options prepared by stakeholders. These can be categorized by research domain and avenue			
ldea analysis	Step 6: Scoring, ranking and prioritization	Research options are scored against the three questions under each criterion: 0= I disagree/1=I agree/0.5 neither agree nor disagree. The scores of the TEs are calculated for each research option and divided by the number of received answers per criterion and converted to percentage, thus assigning each research option a value 0% to 100%. The research options can thus be ranked under each criterion and prioritized			
ldea socialization	Step 7: Involve stakeholders for further input	To obtain the views of a wider audience a heterogeneous group of stakeholders can be involved (can include researchers, policy makers, programme implementers, research beneficiaries) to i) determine the minimal score (threshold) for each criterion that needs to be achieved in order to consider any research option a funding priority ii) allocate different weights to each criteria			
ldea selection	Step 8: Finalization of priorities	Those research options that do not match the threshold determined by stakeholders are discarded. An overall research priority score (weighted arithmetic mean) is calculated for each research option. For transparency, assess level of agreement between technical experts for each research option/question using Kappa calculation.			



Table 2: CHNRI method^{6,7}



	Table 3: 3D CAM method ⁸				
	Involves consideration of factors along three axes: the public health, the institutional and the equity dimensions. Supports priorities based on evidence. Does not provide recommendations on stakeholder selection				
	Step 1	Best available information presented to participants to start discussion on public health and institutional aspects of the health problem. Information can be collected from global reports and international, peer-reviewed literature, and expert opinions. Participants can be split into 2-3 groups to work on specific dimensions or aspects			
		Dimension 1: Public health			
	Define health problem	The magnitude of the health problem under investigation (appropriate measures are chosen depending on the problem) is defined and the factors that are responsible for the persistence of the health problem are analyzed			
l socialization	Evidence/knowledge of interventions	Present knowledge available to help solve the health problem is assessed. Applicability of solutions, including the costs and the effectiveness of existing interventions are evaluated. <u>If information</u> is not available this becomes a priority area for research			
ldea generation, analysis, and socialization	Cost factor	To know whether the desired intervention is, or is expected to be, cost-effective, the promise of the R&D effort against other potential interventions is assessed with the help expert opinions. Present level of investments in research for the health problem that is being considered is calculated to reveal the sources and amount of research funds that are being allocated to the specific problem and give a clear sense of whether the problem is a high priority on the country's research agenda. Information may not always be available			
Ide		Dimension 2: Institutional			
	Individual, family/household or community	Available information/evidence relating to interventions for identified problems are assessed to define implementation at different levels, for example by the individual, family/household or community			
	Role of governmental and public institutions	The contribution of the health ministry and health institutions to the control of the specific health problem/ condition being explored are assessed. In addition other the roles of other public sectors (not related to health directly) are also assessed			
	Governance	Structures and institutions at central government level and international decisions or initiatives that may increase or decrease the burden of disease are assessed			
Idea analysis, socialization, and selection	Step 2	Following consensus on the discussions (public health and institutional), a 2-D matrix is filled and the data is reviewed from the perspective of an equity stratifier such as gender, poverty, religion or race to add the 3rd dimension to the data			
Idea analysis, and se	Step 3	Following completion of the 3D matrix, a report is prepared on the results of the discussions and consensus achieved			



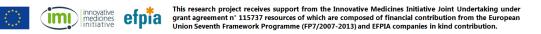




	Table 4: EHNR method ⁹		
	An integrated and systematic approach for organizing and managing country specific and global health research in order to promote health and development on the basis of equity and social justice		
Step 1: Identify stakeholders to contributeBroadened spectrum of stakeholders selected: researche decision-makers at different levels, health service provid communities, private sector representatives, parliament donors and international agencies. Although participants from different groups, selection should be narrow enoug 			
dea generation	Step 2: Gather evidence	Quantitative and qualitative data along the domains of the health problem, health systems and health research systems is collected from multiple sources as evidence	
ldea ge	Step 3: Identify research ideas	Through consultative group process (focus groups, interviews) and consensus, gaps and important research areas are identified with the help of the evidence	
Idea analysis and selection	Step 4: Define criteria	To ensure transparency, criteria against which priorities can be set are defined. Criteria could be along the lines of magnitude and urgency, extent of previous research, feasibility and impact. A manageable core of criteria should be no more than six or seven and final criteria will depend on the purpose and level of action (global, national, etc.). Criteria finalized through consensus	
ldea an	Step 5: Evaluate research ideas and finalize	Criteria are applied to potential research ideas and selection among these ideas can be based on scores (simple scales to complex matrices)	



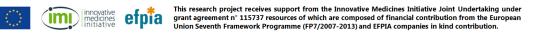




Table 5: Framework comparison of well-established models for research agenda creation

	JLA	CHNRI	3D CAM	EHNR		
Context	Patient-centered research prioritization	Funding agency/organizatio n, government, and policymakers - targeted research prioritization	Organization, economy and equity-centered research prioritization	National and global level health research prioritization		
	Research idea generation					
	Stakeholders surveyed for research ideas	Technical experts propose research ideas based on evidence	Stakeholders propose research ideas based on evidence	Stakeholders propose research ideas based on evidence which are refined through consensus		
		Research ide	a evaluation			
Priority setting exercise	Research ideas are analyzed against literature evidence to narrow down the choices	Criteria are defined and research ideas are scored against these criteria by technical experts	Research ideas encompassing the public health and institutional aspects are developed with the help of literature evidence and group consensus	Criteria are defined and research ideas are scored against these criteria		
		Research priori	ties finalization			
	Stakeholders vote to define an interim list of research priorities. Finalization through group consensus at a workshop	Wide-spectrum stakeholders determine thresholds and weights for each criteria, based on which research priorities (determined by Technical experts) are filtered and finalized	The research ideas are further analyzed using the equity dimension and the priorities finalized	Research priorities finalized based on scores		

In all these models there is an emphasis on evidence-based research priority setting. Moreover, two types of priority setting exercises can be inferred from these models: 1, by using existing literature as a starting point to identify gaps and limitations new research ideas can be developed. These can then be scored against criteria and ranked to finalize the research agenda; 2, by using the knowledge and opinions of participants, research ideas can be developed. These can then be analyzed against existing literature to eliminate topics. Through group deliberation a research agenda can then be established.





1.3.3 Trends in priority setting exercises

Screening of 202 PubMed hits in this rapid review revealed that a total of 69 research agenda, with priorities concerning health and healthcare research, were published in the years 2018 and 2019.

1.3.3.1 Utilization of models to set priorities

Of the 69 research agenda, 44% (30) reported the use of a known priority setting model (Figure 1a). Amongst these 30 articles, 22 (73%) employed the JLA method and the remaining 8 CHNRI method (27%). Considering that the method was developed for research priorities with patients' perspectives, the JLA method was used, not surprisingly, in those studies engaging patients in their priority setting exercises. CHNRI method was utilized in exercises involving (research and/or non-research) experts in the field only.

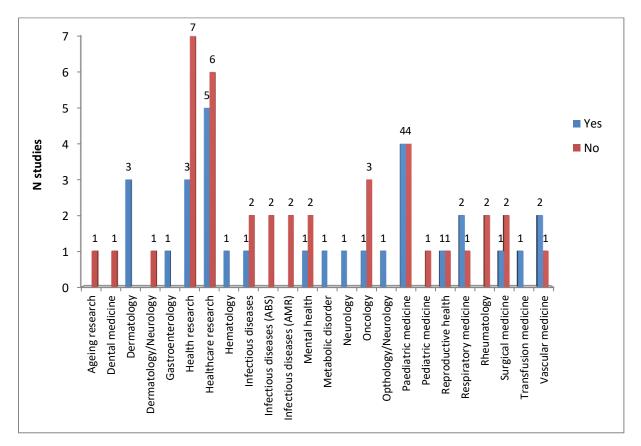


Figure 1a: Tendencies to use comprehensive priority setting models to develop research agenda



Note: Those research agenda topics that did not fit under a specific scientific discipline are reported as health and healthcare research in general.



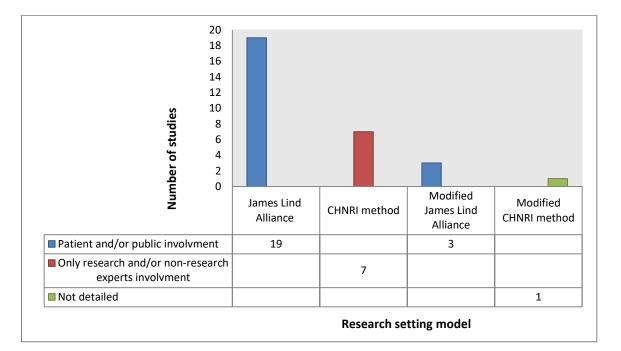


Figure 1b: Models used to create research agenda in health and healthcare research in 2018 and 2019

1.3.3.2 Other methods used to set priorities

Heterogeneous techniques were utilized by those studies (N=39) that did not use any of the wellknown models. Two research agenda were created by writing committees using systematic review and scoping review, respectively. Additionally, two research agenda did not detail all the steps of their priority setting exercises. These were excluded for further analysis since they did not provide information relevant to stakeholder driven research priority setting. Majority of the research agenda (30; 86%) employed a consensus-based approach¹⁰ (group decision making with consent or support from all members) while only a minority (5; 14%) used consultation¹⁰ (information gathering activity where decisions are not shared). To achieve consensus, 8 studies (27%) adapted the Delphi method and 5 (17%) Nominal Group Techniques; the remaining 17 (57%) did not state the use of any such formal methods.

Delphi Method

The Delphi approach, a data gathering and analysis technique, in simple terms involves iterative survey of participants to reach consensus. The results of each survey are shared with the participants, creating opportunities to review scores, modify votes, and propose new statements/comments in each round.¹¹ All the research agenda created with the Delphi method (table 6) used iterative surveys based on Likert scale (excepting the survey conducted with *Codigital*) to evaluate research ideas which were identified through literature scoping (3) or inputs from experts, individual opinions based on previous experiences, etc. (5).

For prioritizing research ideas:





- Three studies used the scores from the final surveys to select top ranking ideas, while 3 other studies carried out face-to-face group discussions for a final consensus.
- Two studies employed Hanlon Process of Prioritisation¹² in which participants score research ideas by criteria (A,B,C), the results are then filtered by feasibility factor (when necessary) and assigned priority scores with formula (A+2B)×C. The research ideas are finally ranked according to the priority scores.

N studies	Participants	Research idea generation	Research idea evaluation	Research priority finalization
1	Only research and/or non-research experts involvement	Published research agenda scoping	Iterative online survey	Hanlon Process of Prioritisation
1	Patient and/or public involvement	Consultation (online survey)	Iterative online survey and group discussion (telephone conference)	Group discussion
1	Patient and/or public involvement	Consultation (online survey)	Iterative online survey	Ranking
1	Patient and/or public involvement	Iterative consultation (Codigital*)	Iterative online survey (Codigital*)	Group discussion
1	Patient and/or public involvement	Literature scoping	Iterative online survey (eDelphi*)	Ranking
1	Only research and/or non-research experts involvement	Consultation (interviews)	Iterative online survey	Ranking
1	Only research and/or non-research experts involvement	Consultation (online survey) and group discussion	Online survey (REDCAP*)	Hanlon Process of Prioritisation
1	Patient and/or public involvement	Technical data and published research agenda scoping	Iterative online survey	Group discussion and online survey

 Table 6: Delphi method in priority setting exercises

*<u>Codigital</u>: cloud-based idea management solution; facilitates pair-wise ranking. <u>eDelphi</u>: Delphi survey software. <u>REDCAP</u>: online or offline data capture software

Nominal Group Technique

Nominal Group Technique (NGT) used in face-to-face meetings involves individual (silent) brainstorming to generate ideas, sharing the ideas (round robin) and deliberation as a group (without evaluation), followed by ranking to prioritize ideas¹³. According to the five research agenda identified in this review using NGT (table 7):

- Research ideas can be obtained from opinions and experiences of people (4) and/or from literature evidence (1).
- Through iterative group deliberation (1), voting [approval, ranking or scoring] (2) or a combination of both (2), the significance of these ideas can be assessed and then prioritized





N studies	Participants	Research idea generation	Research idea evaluation	Research priority finalization
1	Patient and/or public involvement	A priori	Group discussion	Voting
1	Patient and/or public involvement	Consultation (online free text survey)	Group discussion	Group discussion
1	Only research and/or non-research experts involvement	Silent brainstorming and group discussion	Ranking	Group discussion
1	Patient and/or public involvement	Literature scoping and group discussion	Ranking	Ranking
1	Only research and/or non-research experts involvement	Silent brainstorming and group discussion	Iterative survey*	Ranking

Table 7: Nominal Group Technique in priority setting exercises

*1st 3-point scale scoring; 2nd 5-point scale scoring and editing

General consensus approaches

Similar to research agenda based on Delphi method and NGT, literature evidence (4), consultation/group discussions (7) or both (6) served as starting points for research agenda development. Mixed methods were used by other studies to set priorities:

- In 2 studies thematic analysis was performed to review research ideas for descriptive reporting
 of research agenda. In another study thematic analysis was followed by survey to prioritize
 research ideas.
- One study used a combination of consultation and group discussion for evaluation and prioritization, respectively.
- Iterative group discussions helped evaluate and prioritize research ideas in case of 7 studies.
- Six studies used iterative survey or voting to assess research ideas; one study among these used a "concept mapping" technique additionally for cluster analysis. Finalization of research ideas in these 6 studies ensued ranking (3), group discussion (2) or comparison against literature evidence (1).

N studies	Participants	Research idea generation	Research idea evaluation	Research priority finalization
1	Patient and/or public involvement	Consultation (focus groups/interviews) and group discussion	Multivoting technique	Literature evidence
1	Patient and/or public involvement	Consultation (online survey)	Multicriteria decision analysis (1000minds*)	Group discussion
1	Only research and/or non-research experts involvement	Literature scoping	Group discussion	Group discussion

Table 8: Consensus approach in priority setting exercises





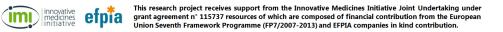
1	Only research and/or non-research experts involvement	Literature scoping and consultation (focus groups/interviews)	Iterative online survey (Codigital*)	Ranking
1	Only research and/or non-research experts involvement	Literature scoping and consultation	Consultation	Group discussion
1	Patient and/or public involvement	Literature scoping and consultation	Iterative survey and group discussion	Ranking
1	Only research and/or non-research experts involvement	Brainstorming	Survey and concept mapping (Provalis*)	Group discussion
1	Only research and/or non-research experts involvement	Literature scoping and consultation	Iterative online survey (Codigital*)	Ranking
1	Not detailed	Literature scoping	Group discussion	Group discussion
1	Patient and/or public involvement	Group discussion	Thematic analysis	
1	Patient and/or public involvement	Literature scoping, consultation and group discussion	Group discussion	Group discussion
1	Only research and/or non-research experts involvement	Literature scoping	Group discussion	Group discussion
1	Patient and/or public involvement	Literature scoping	Group discussion	Group discussion
1	Only research and/or non-research experts involvement	Group discussion (telephone conference)	Group discussion (telephone conference)	Group discussion
1	Patient and/or public involvement	Group discussion	Thematic analysis	Online survey (for final ranking)
1	Only research and/or non-research experts involvement	Group discussion	Group discussion	Group discussion
1	Only research and/or non-research experts involvement	Literature scoping, consultation (online survey, paper survey, interviews) and group discussion	Thematic analysis	

*<u>1000minds</u>: decision-making software for prioritization, group decision-making, conjoint analysis. <u>Codigital</u>: cloud-based idea management solution; facilitates pair-wise ranking. <u>Provalis</u>: qualitative, quantitative and mixed methods analysis software

Consultation

Due to the nature of the approach 3 research agenda involved descriptive reporting of the results of content analysis of consultations. In one research agenda, the results of iterative voting by stakeholders on research ideas developed through literature review was used for preparing the research agenda. In another research agenda experts were surveyed additionally to generate a list of research ideas which were voted against criteria and finally ranked by preferences.







N studies	Participants	Research idea generation	Research idea evaluation	Research priority finalization
1	Only research and/or non-research experts involvement	Literature scoping	Voting	Voting
1 (AMR research agenda)	Only research and/or non-research experts involvement	Literature scoping and consultation	Multicriteria decision analysis (1000minds)	Ranking
1	Patient and/or public involvement	Consultation (draw/write and tell techniques) and group discussion	Qualitative content analysis (Nvivo 11)	Not applicable
1	Patient and/or public involvement	Consultation (focus groups/interviews)	Qualitative content analysis (Nvivo 11)	Not applicable
1	Patient and/or public involvement	Consultation (focus groups/interviews)	Qualitative content analysis	Not applicable

Table 9: Consultation in priority setting exercises

1.3.4 Outlook and implication

Due to varying objectives and use of diverse techniques in research priority setting exercises, there is no one approach or a golden standard to formulate a research agenda. However, use of a systematic approach which offers transparency, replicability, and results in outcomes that match end User needs is strongly recommended across literature. Importantly, research priorities should not only reflect opinions but also be supported by evidence. ^{3, 14-17}

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