

Ethics and equity in research priority-setting: stakeholder engagement and the needs of disadvantaged groups

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Introduction

A transparent and evidence-based priority-setting process promotes the optimal use of resources to improve health outcomes. Decision-makers and funders have begun to increasingly engage representatives of patients and healthcare consumers to ensure that research becomes more relevant. However, disadvantaged groups and their needs may not be integrated into the priority-setting process since they do not have a "political voice" or are unable to organise into interest groups. Equitable priority-setting methods need to balance patient needs, values, experiences with population-level issues and issues related to the health system.

The Cochrane Collaboration approved the establishment of the Cochrane Agenda and Priority Setting Methods Group (CAPSMG) in 2011. The CAPSMG examines empirical methods for setting research agendas and prioritising topics for systematic review. The Cochrane Collaboration has previously evaluated how Cochrane entities set research priorities (1). Few strategies address the question of engaging and meeting the needs of disadvantaged groups, despite the fact that previous studies have shown that disadvantaged groups value health problems differently (2).

Objective

To develop recommendations on the engagement of different stakeholders and disadvantaged populations in priority-setting exercises.

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Methods

The CAPSMG conducted a workshop and a special session at the 22nd Cochrane Colloquium held in Hyderabad, India, on September 25 and 26, 2014. Different tables were assigned to the participants at the workshop, and each table took the perspective of a policy-maker, clinician or member of the public. The participants at each table then discussed one of two topics: Ebola prevention or the implementation of "sin taxes" on sugary beverages. They reported their discussions, together with the keynote presentations. These were used to develop recommendations for future priority-setting exercises. In the special session, the participants discussed strategies for the improvement of priority-setting.

Summary of presentations and discussions

Overview of the CAPSMG priority-setting process and use in the Cochrane Public Health Group (CPHG)

The CPHG was one of the first review groups to develop and implement a structured priority-setting method to improve the focus of systematic reviews on public health, and more specifically, on topics of interest to end users, particularly in developing countries (3,4). Rebecca Armstrong described the challenges faced in the CPHG prioritisation process. These included difficulty in identifying a representative sample of "experts"; in reaching an agreement on a generalisable list of priorities since the experts were likely to identify priorities selectively, on the basis of their own interests or expertise; and in converting topics to research questions in population, intervention, control and outcomes (PICO) format. Retaining the involvement of experts throughout the process of prioritising topics for systematic review was another challenge. Methods of prioritisation must be flexible and diverse to allow its application in different research areas.

Case study of national health research priority-setting in an LMIC: challenges and opportunities in Nepal

Purushottam Dhakal and Sangeeta Rana described how the Nepal Health Research Council (NHRC) led four national research prioritisation exercises. For this purpose, the Council organised workshops for the stakeholders to arrive at a consensus. These involved NHRC representatives, policy-makers, planners, external development partners, national and international non-governmental organisations, hospitals and academic institutions. The challenges encountered in this process included the scarcity of epidemiological and baseline health data. In addition, it was difficult to narrow down the extensive and comprehensive list of priorities. The NHRC plans

to approach future priority-setting exercises in a step-wise fashion to develop political will, engage a broader group of stakeholders, and see to it that funding for health research is matched with priorities.

Using the burden of disease to set research priorities

The Global Burden of Disease (GBD) study provides publicly accessible data using disability-adjusted life year (DALY) which is used to quantify the burden of disease. It also enables one to compare conditions (5–7). Chante Karimkhani and Robert Dellavalle described the empirical methods used to assess the representation of the Cochrane Database of Systematic Reviews (CDSR) in comparison to GBD metrics for dermatological, ophthalmological and otolaryngological diseases (8–10).

In the course of discussion, it became clear that the use of data-driven methods may have limitations in low- and middle-income countries (LMICs), which have inadequate or poor-quality epidemiological data. For diseases which affect disadvantaged populations, greater emphasis is required on equity and economic consequences.

Priority-setting for guidelines and health equity impact assessments

Kevin Pottie explained that it is essential to take planned and coordinated action to ensure security, healthcare and health equity, particularly in LMICs. His presentation highlighted new methods of promoting health equity, such as the PRISMA-E. It also underlined the work that is necessary to support health equity approaches, such as overcoming the difficulty in acquiring sufficient data to make equity interpretations, integrating community members into the equity approach, and ensuring buy-in from decision-makers. The work of the World Health Organization (WHO), the development of the Refugee Health Guidelines, and the Health Equity Impact Assessments (11–13) were cited as examples of the current use of health equity approaches.

An equity lens for priority-setting approaches

Vivian Welch discussed the equity lens developed for priority-setting by the CAPSMG (14). It takes into account all steps of priority-setting, starting from the inclusion of relevant stakeholders in decision-making to the consideration of differences in the burden of disease, assessment of differences in effects, and consideration of potential differences in values, preferences, or the acceptability of the interventions. An acronym, PROGRESS-Plus, was coined to describe the characteristics of people and places associated with health inequity. "PROGRESS" stands for the place of residence, race/ethnicity/culture/language, occupation, gender/sex, religion, education, socioeconomic status, and social capital. "Plus" refers to other characteristics such as age, disability and personal relationships that might also be associated with inequity. The use of PROGRESS-Plus identifies potential gaps in the prioritisation process, such as poor comprehensibility

of material for those with low literacy or different language backgrounds, and differences in values or preferences across people and settings. This lens also describes eight steps for the design, monitoring, and evaluation of research prioritisation process and its outcomes.

Stakeholder engagement in priority-setting

There were two presentations on this theme.

Soumyadeep Bhaumik raised the issue of varying priorities among the stakeholders, citing the example of snakebite, which kills thousands in India (15). Interventions and the diagnostics available for snakebites have a poor evidence base and yet, are not being researched (16). He said that while public health professionals are concerned about issues pertaining to the health system and adherence to public health standards, clinicians focus on dosing and management guidelines. He underlined the need to reach the "real stakeholders". He identified fear, tokenism and a lack of confidence as the barriers to the effective participation of stakeholders.

Sandy Oliver spoke on the priority-setting process used to identify research topics in the area of pre-term births in the UK (17). Difficulties have been faced in reaching out to individuals not aligned with organisations. Internet-based questionnaires, though easier to administer, are less likely to reach disadvantaged families; yet, these are the people who are more likely to face the problem of pre-term birth.

In the discussion, it was suggested that stakeholder engagement is more effective when priority-setting begins at the micro level, before expanding nationally or globally. It was recommended that communication strategies be developed over time so as to be able to win the trust of the relevant populations, build their capacity to participate, take into account their concerns and provide feedback on the results.

Focus group discussion: topical public health scenarios

The methods section of the manuscript has a detailed description of the process utilised for the workshop.

The key messages that emerged from the focus group discussion were as follows.

1. It is important to learn from other similar interventions/problems (for example, learning from tobacco control when planning for sin tax for sugary beverages and using information on influenza outbreaks for Ebola control.)
2. Consult/include (collaborate with) people who will be affected, including the most disadvantaged groups.
3. There were substantial differences in the perspectives and priorities of various stakeholders: the consumers valued transparency of science and the effect of interventions on themselves; the policy-makers prioritised the implications of equity, cost and feasibility; and the clinicians saw treatment and diagnosis as the foremost considerations.

Use of the evidence sandwich model in equity-focused research priority-setting

Soumyadeep Bhaumik presented the evidence sandwich model, a conceptual framework for any research priority-setting process. The model broadly consists of three steps: (i). defining objectives, marking domains, and elucidating criteria for prioritising research by epistemic groups, (ii). rapid review of evidence and systematic mapping of research gaps, and (iii) setting priorities through a deliberative process that includes group discussions.

Research agenda for the CAPSMG

The participants identified the following as key contributions that the CAPSMG could make towards priority-setting within the Cochrane Collaboration.

- a. Collecting global priority-setting exercises, results and methods
- b. Identifying strategies to engage with different stakeholders and consider differences in values in the priority-setting process
- c. Adopting methods to balance equity and ethics, keeping effectiveness in mind
- d. Adopting methods to ensure that the voices of all stakeholders are given equal consideration

Recommendations for equity-focused priority-setting for the disadvantaged

Research priority-setting exercises need to meet organisational objectives and be aligned with the values of the stakeholders. This workshop suggested the following strategies to develop an ethical and equity-oriented framework for setting research priorities.

1. Involve diverse stakeholders in a transparent priority-setting process while systematically collecting, analysing and reporting the conflicting interests of all participants.
2. Create strategies to engage stakeholders from different socioeconomic groups to examine research values, expectations and context.
3. Synthesise research agenda-setting publications from different regions of the world in a publicly available database.
4. Cross-link groups working on priority-setting in similar domains to empower low- and middle-income nations with limited research capacity.
5. Identify common themes (meta-priorities) across these research agendas.
6. Identify funding for necessary research on disadvantaged populations and devise strategies to win the trust of these “disadvantaged groups” and build their capacity. To build their capacity and maximise their participation, information could be provided in the appropriate languages and easy-to-understand formats.

7. Guard against tokenism or over-dominance by opinion-movers in the process of engaging stakeholders.

Substantial resources and time will be required to produce a transparent priority-setting process, that is to say, one which will account for the evidence available, will be characterised by optimal stakeholder engagement, will address equity concerns to improve health outcomes, and will remove disparities.

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