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CURRENT OPINION

Patient and Public Involvement in Health Economics Modelling Raises the Need for Normative Guidance

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Abstract

Patient and public involvement in health economics research and health technology assessment has been increasing for some time; however, patient and public involvement in health economics modelling is a more recent development. One reason to advance this type of involvement is to help appropriately manage the social and ethical value judgements that are required throughout model development and interpretation. At the same time, patient and public involvement in health economics modelling raises numerous practical and philosophical issues that invite discussion and debate. Recently, we attended an engagement event which invited patients, members of the public, researchers and decision-makers to discuss some of these issues. One priority that emerged in the discussion was to develop normative guidance for patient and public involvement in health economics modelling. In this article, we reflect on this goal from our own perspective, focusing on why normative guidance is needed and what questions that guidance should answer.

1 Introduction

The field of health economics (HE) is seeing an increasing emphasis on patient and public involvement (PPI), as reflected in the new Consolidated Health Economic Evaluation Reporting Standards (CHEERS) 2022 statement, which includes the recommendation to "report on any difference patient/service recipient, general public, community, or stakeholder involvement made to the approach or findings of the study" [1, p. 605]. While PPI in HE research and health technology assessment (HTA) has been increasing for some time [2–8], PPI in HE *modelling* is a recent development [9–11, 33]. There is no single best definition of HE modelling, but it is useful to emphasize that HE models are "abstractions and representations of complex phenomena" [12, p. 2], "provide decision makers with quantitative information about the consequences of the options being considered" [13, p. 844] and constitute "normative decision-making aids" [14, p. 805]. In our previous work, we have underlined that numerous social and ethical value judgements arise throughout model development and interpretation [15–20], and so we regard efforts to advance PPI in HE modelling as being particularly significant. Social and ethical value judgements in modelling can be understood in

Key Points

While there are numerous frameworks to support patient and public involvement (PPI) in health research, there are special aspects of health economics (HE) modelling that suggest a need for tailored guidance

There is a need to continue the discussion about PPI in HE modelling, building on existing work and developing additional guidance informed by philosophical thinking, empirical research, and input from the wider community

In this Current Opinion article, we reflect on three key questions concerning PPI in HE modelling, including 'who should be involved in HE modelling?', 'what modelling decisions should patients and members of the public be involved in?', and 'how should patients and members of the public influence modelling decisions?

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different ways, including as decisions where there is flexibility from a scientific perspective, i.e. scientists agree there is more than one legitimate way of doing things, and there could be social or ethical consequences following the decision, whether these consequences are immediate or downstream [15]. Ideally, emerging PPI initiatives will give patients and members of the public the chance to directly inform value judgements in HE modelling, alongside other multi-stakeholders in healthcare systems.

In 2022, we helped design an online engagement event in Vancouver, Canada, titled "How Can Health Economic Models Best Reflect Patient and Public Values?", which welcomed patients, members of the public, researchers and decision-makers as participants [21]. Presentations and discussions covered current practices in HE, values and PPI in modelling, and the many value-laden dimensions of transparency in science [22, 23]. In discussions, one priority that emerged was to develop normative guidance for PPI in HE modelling, i.e. recommendations for best practice which are, by definition, value judgements. In this 'Current Opinion' article, we reflect on this goal from our own perspective, focusing on why normative guidance is needed and what questions that guidance should answer. Our perspective is informed by our engagement experience, our work in HE and the philosophy of modelling, and our knowledge and values as researchers and individuals.

2 Why is Normative Guidance Needed?

While there are numerous frameworks to support PPI in health research [24], there are special aspects of HE *modelling* that suggest a need for tailored guidance. One is that HE modelling is widely perceived as being particularly complex and technical [25], which raises practical questions about how to best involve non-experts in the process. Another is that HE modelling is often one step closer to informing policy-making, and has the potential to have a broad and significant impact on citizens. Perhaps most importantly, HE modelling decisions are often unconstrained by the available evidence – that is, highly flexible and discretionary – meaning modelling teams have a unique power over study results and downstream decision-making, giving them a particularly high level of social responsibility [17]. For these reasons, current frameworks for PPI in research may fall short in informing PPI initiatives in modelling. In the environmental sciences, there is a considerable literature on participatory modelling [26–29], which has informed early proposals to conceive the patient role in HE modelling and should continue to provide insight [30]. Among other things, this literature rightly identifies the *power* that models have: "first, in framing the problems, asking the questions, comparing alternatives, identifying the contexts and boundaries; and second, in determining the actual value sets that lead to action through successful management or governance" [31, p. 51], linking that power to the need for participatory modelling and to best practices in that field. However, we expect this literature will be of limited use as a step-by-step guide for PPI in the HE context, as others have found it to be [32]. Recently, in describing the co-production of a public health economic model with stakeholders, Gibbs et al. [33] remarked that, although stakeholder engagement is recommended in HE modelling, they "found little practical advice in precisely how a modeler might engage stakeholders in public health modeling".

To the best of our knowledge, the only existing framework concerning public involvement in HE modelling specifically is the Mathematical and Economic Modelling for Vaccination and Immunisation Evaluation (MEMVIE) Public Involvement Framework [9]. This framework comes in both short and long forms and describes how members of the public can participate in both the epidemiological and economic aspects of HE modelling. The framework's strengths are that it is highly detailed and instructive, it reflects the authors' experience working closely together over a 5-year period, and it outlines how members of the public can be involved in multiple stages of HE modelling – including evidence evaluation, which is seldom the locus of stakeholder involvement in modelling [27]. At the same time, the MEMVIE framework does not provide a philosophical justification for its recommendations, emphasizes considerations unique to vaccination and does not speak to all outstanding questions about PPI in HE modelling. The framework also focuses on *public* involvement in modelling, acknowledging that this may be different from patient involvement but without addressing whether or how that difference should be reflected in practice. For these reasons, there is a need to continue the discussion about PPI in HE modelling, building on the MEMVIE framework and developing additional guidance informed by philosophical thinking, empirical research and input from the wider community.

3 What Questions Should Normative Guidance Answer?

Frameworks for supporting PPI in health research serve numerous purposes, with emphases on setting research priorities, overcoming power imbalances within research teams, maximizing participant recruitment and retention, guiding the writing and appraisal of research reports, and assuring transparency and accountability in collaboration [24]. Most (if not all) of these purposes will apply in some way to HE modelling and other purposes exist too, such as establishing the optimal "modalities" for PPI in HE [32, p. 3] – so identifying every question that normative guidance should answer will take time. In this article, our objective is to point to just

three key questions concerning PPI in HE modelling, give our own preliminary/partial answers to them, and encourage them to be debated and studied in the field.

3.1 Question 1: Who Should Be Involved in HE Modelling?

A key question raised in our experience concerns who *exactly* should be involved in HE modelling. This is a question with many potential forms and meanings, which will be important to distinguish in the process of developing normative guidance. Just one form of the question concerns what individual characteristics people within groups of involved stakeholders should have: for example, in a qualitative study with HE modellers, participants questioned what training, capabilities and personal perspectives patients should have in order to be part of HE modelling [34]. In our view, such questions concerning individual-level characteristics will defy a standard answer, and thus normative guidance in HE modelling is likely to mirror general recommendations for PPI in research, including aiming for diversity, remembering that "no one person can ever fully represent a disease or group of people", and considering the purpose of the project [35, p. 14]. Even more challenging forms of the question '*who should be involved in HE modelling?*' draw on conceptual distinctions between groups of stakeholders: for example, in the qualitative study cited above, participants also questioned who besides patients should be involved in HE modelling, e.g. physicians or members of the public [34]. Indeed, given the goal of HE as a discipline, one particularly significant question is: should only patients be involved in HE modelling, should there be some balance of patients and members of the public, or should there, in fact, only be members of the public (who will, of course, sometimes include people who are, *inter alia*, patients)? Yet this particular question requires clarification, since there are many possible definitions of the terms 'patient' and 'member of the public', and in some HTA processes the terms 'patients' and 'public' have been used synonymously [36]. In developing normative guidance for PPI in HE modelling, one important task will be to better define the distinction between patients (roughly, individuals with lived experience of a health condition) and members of the public (roughly, individuals who do not necessarily have special knowledge of the health condition being modelled, but rather occupy the role of citizen or taxpayer) and to establish when it is useful to make this distinction. These clarifications will be necessary to address other challenging normative questions, such as whether and when patients should be involved in HE modelling *qua* patients (i.e. in a way that affords them a special role on the grounds of their lived experience) or *qua* members of the public (i.e. in a way that affords patients the same opportunities for involvement, the same influence, etc., as all citizens). In this short article, our goal is not to answer all of these questions, but to point to the need to address them carefully and to raise some ideas for further discussion.

In the literature, there is considerable support for the view that what patients and members of the public contribute is a 'lived experience' perspective, suggesting that an important criterion for being involved in HE modelling is lived experience relevant to the project at hand [9, 11, 25, 33]. To add to this conversation, one idea we would raise is that lived experience of a particular kind is not a necessary criterion for involvement in HE modelling – in fact, there are reasons to involve members of the public *qua* citizens in any given HE modelling project. Although HE models are often particularly relevant to specific patient groups (e.g. individuals with lived experience of depression in British Columbia [11]) or particular knowledgeable stakeholders (e.g. individuals with lived experience of informal alcohol outlets in South Africa [33]), HE models have the potential to affect everyone who uses and supports the target health system, within its wider social and economic context. Moreover, HE models are a product of health science as an institution. This means that HE model developers are accountable in some sense to *everyone*, insofar as everyone has a stake in local health systems, the wider infrastructure in which they operate, and the health sciences. Thus, in principle, there is a role for members of the public in *all* HE modelling projects, even those focused on evaluating interventions for health conditions or public health risks of which they have no special knowledge. After all, members of the public *qua* citizens have an interest in ensuring that models incorporate the highest quality evidence and carefully explore the role of uncertainty: members of the public, at least in the abstract, will bear the opportunity cost of funding a given treatment and/or make other sacrifices imposed by population-level interventions. Furthermore, members of the public *qua* citizens represent the general interest in safeguarding health science as an institution, e.g., in resisting forces that would see HE models developed for the purpose of advancing pre-determined agendas. At least in principle, involving individuals with no obvious special interests could help ease concerns that patient involvement (or the involvement of any other group of interested stakeholders) may introduce bias into HE modelling and HTA [34, 37].

In some public health modelling projects, effectively all members of the public will have a direct interest in the model results, and it may not be useful to think in terms of involving individuals with 'no special interests'. In these contexts, the more appropriate goal may be to involve a heterogeneous group of members of the public, taking into account not only what different types of knowledge are relevant to the project but what public *values* are likely

to be in tension in the context of the project. Ultimately, a heterogeneous group of members of the public would include people who are and are not at higher risk of a given health problem, who have and have not experienced adverse effects from public health interventions, who work and do not work in industries likely to be impacted by the intervention at hand, and so on – i.e. people who will likely have different knowledge and different values. For example, in the context of COVID, parents whose children have been differently affected by the disease and public health measures, respectively, will likely have different knowledge and values that would influence future model development differently. In developing public health models, involving members of the public from salient groups whose knowledge and values are expected to differ may help to ensure that models are adequate to inform decision-making downstream.

3.2 Question 2: What Modelling Decisions Should Patients and Members of the Public Be Involved in?

Although the HE modelling process includes many activities, we focus our attention in this section specifically on modelling *decisions*. Previously, we summarized modelling decisions in terms of *representational* and *inferential* decisions: respectively, decisions about *what to represent* and *how to represent it*, which take place during model development, and decisions about *what is true or likely,* which take place during model interpretation [16, 38]. Decisions about 'what to represent' concern what to include in the model and what to exclude from it, decisions which may often be thought of in terms of defining the research question. Examples include decisions about what health interventions to model in the first place, what interventions to compare them with, what costs and outcomes to represent, and what other aspects of the target system to consider, from among infinite characteristics of disease states, patients, providers, environments, etc. Decisions about 'how to represent' concern the treatment of entities already chosen for inclusion in the model, which may often be thought of as technical decisions. Some examples are decisions about what data sources are appropriate to use as model inputs, how the model should be calibrated and to what standard of predictive accuracy, and to what extent the model must explore the impact of uncertainty. Finally, decisions about 'what is true or likely' concern what *facts* to infer – and publicly endorse – on the basis of model results. For example, decisions about what estimates to report for the incremental cost-effectiveness ratio and the probabilities that a new treatment is cost-effective over a range of willingness to pay values.

We make the above distinction to emphasize that, while inferential decisions concern what factual claims to make, representational decisions concern the extent to which it is acceptable to simplify and/or distort the system being modelled and are informed by the goals and priorities of the model users [39]. Both types of representational decisions have considerable flexibility from a scientific perspective and together will determine the model's *adequacy for purpose* – giving them strong potential for downstream social impact, especially when models used for healthcare decision-making.

We have argued that representational and inferential decisions always require making social and ethical value judgements, though in distinct ways [16, 38]. This can be understood in terms of two philosophical insights: first, inferential decisions carry *inductive risk*, the risk of endorsing a 'fact' whose objective truth value is false. Because we can never be 100% sure that a factual claim is true, inferential decisions depend in part on our ethical assessment of how serious it would be to endorse a false claim. Second, representational decisions carry *representational risk*, the risk that a representational decision will be *inadequate for purpose*. Because representational decisions determine what information will be highlighted and what information will be obscured by a scientific model [38, p. 16], they depend in part on our ethical assessments of what purposes models should aim to serve and the significance of failing to meet those purposes. While inductive risk links to the downstream harms of endorsing a false claim, representational risk links to this and more: to the harms of a scientific institution that studies what are perceived to be irrelevant or even harmful questions, that systematically conceals variables relevant to specific groups, that fails to do its due diligence in characterizing uncertainty, and so on.

To be clear, while representational and inferential decisions always require social and ethical value judgements, they also always require scientific or 'epistemic' value judgements too. For example, making representational decisions requires considering epistemic features of potential data sources, such as sampling method and other possible sources of bias, while making inferential decisions involves considering the many epistemic features of a model as a whole. However, in practice, it is extraordinarily difficult to separate epistemic and ethical considerations and to recognize, let alone communicate, what values are driving a particular modelling decision [18]. This is what justifies involving patients and members of the public in all modelling *decisions*. During model development, this includes deciding which health interventions are socially and ethically acceptable and worthy of the significant attention a model can bring. It also includes deciding which aspects of the target system will be necessary to represent in order to trust the results of the model, deciding what evidence will be adequate to represent those phenomena, and deciding

what constitutes a sufficient exploration of uncertainty in the context at hand. At the stage of model interpretation, it includes deciding what factual claims should be endorsed on the basis of model results. Because it clarifies these points, we argue that the above philosophical framework is a good starting point for developing normative guidance for PPI in HE modelling.

In reality, it may not always be feasible or maximally beneficial to involve patients and members of the public in every modelling decision. For this reason, to develop normative guidance for PPI in HE modelling, future work is needed to understand the practical implications of PPI in different modelling decisions, what factors have influenced modelling teams when planning how to work together [9–11, 33], and what goals are most important to achieve through PPI. In parallel, we argue, the process to develop normative guidance should include defining and understanding *pivotal* representational decisions. We take pivotal representational decisions to be at least those where there is a high degree of uncertainty or indeterminacy regarding the best representational choice, where the choice that is made will have a significant impact on model results, or where it is expected that different patient and public groups will have conflicting values that would steer modellers in different directions. At a minimum, we think normative guidance should help model building teams recognize pivotal representational decisions and emphasize that these are a priority for PPI.

3.3 Question 3: How Should Patients and Members of the Public Influence Modelling Decisions?

A key question that normative guidance should address is what type of *influence* patient and public contributors should have on HE modelling decisions. In the HTA context, there is a reasonable expectation from patients to know *how* their contributions will be used and in what way submitting their input will benefit them [7] – yet it remains difficult to characterize the influence that PPI has on HTA [40–42]. We anticipate a similar expectation in HE modelling. Furthermore, as the 2022 CHEERS statement [1] invites researchers to report the difference that PPI makes to a study's approach or findings, we think it is important to reflect on what sort of difference PPI *should* make to HE modelling decisions.

To start, we should distinguish between four different questions one might ask about influence: (1) what information should be sought from patients and members of the public when they participate in modelling decisions?; (2) how should that information be used?; (3) how should researchers report the difference that PPI made to modelling decisions?; (4) how much power should patients and members of the public have over HE modelling decisions? Ultimately, normative guidance for PPI in HE modelling should give detailed answers to *at least* these four questions, and a number of contextual factors are likely to figure in them. Our aim here is not to cover everything, but to raise key points that are relevant to the development of that detailed guidance.

First, in some PPI contexts, we might judge that patients and members of the public, respectively, should play different roles and have different types of influence – but not always. For example, in HTA, patients specifically are often asked to provide factual information about disease manifestations and treatment effects [7, 8, 43]. In this context, patient involvement functions, at least in part, as a method complementary to the systematic collection of clinical data. The justification for this is that patients may know empirical facts about a disease, treatment or technology, which the systematic collection of clinical data has missed. In this process of *evidence gathering*, patients' experiential, factual knowledge is a reason for them to play a different role and have a different type of influence than members of the public. However, later in the HTA process, when decisions are being made concerning the cost-effectiveness of health technologies, to argue that patients should have a distinct influence would be controversial. Relatedly, some current challenges in PPI in HTA are to clearly define the respective roles of patients and the public [6, 44], and to resolve conflicting views around how to use experiential knowledge versus systematically collected data in decision-making [37, 42, 45].

In HE modelling, there is a similar process of evidence gathering, and one future task will be to define the role or roles patients and members of the public should play in that process. A separate question concerns how patients and members of the public should influence modelling *decisions*, which is our focus here. On this specific question, we argue that the philosophical framework we outlined in Sect. 3.2 can provide guidance. What it suggests is that, in the context of inferential decisions, patients and members of the public should be invited to help answer the following *type* of ethical question: *is it better to err on the side of claiming that a particular treatment is likely to be cost-effective when it is not, or vice versa, in a given context?* This is the question of how to manage inductive risk. In the context of representational decisions, patients and members of the public should be invited to help answer the following *types* of ethical questions: *is it important that a model represent the effectiveness of a new treatment in different subpopulations? Is it acceptable for a model to compare the effectiveness of a new treatment to placebo? Is modelling the effect of treatment on quality adjusted life years sufficient, or is another outcome relevant? Is the value, or range of values, being used for a model parameter adequately supported by existing empirical work? Among the known sources of uncertainty in the model, how many should be explored in sensitivity analyses?* This is the question of how to manage representational risk.

In summary, when it comes to modelling decisions, the purpose of PPI is not to add to the body of known facts, but to *manage value judgements*. This suggests, foremost, that patients and members of the public should play a similar role in HE modelling: there is no prima facie reason that HE models should better serve patients' purposes than the general public's, or vice versa, or that factual conclusions should reflect a particular group's values. This means that researchers should seek the same information from all members of the public, be they patients or not, and use it in the same way. That is, researchers should seek not factual knowledge through PPI, but *values*, and use that information to help resolve unconstrained modelling decisions. When reporting the difference that PPI made on a modelling project, research teams should collaboratively describe the impact that working together had on those value-laden decisions.

This leaves us to address the question of how much *power* patients and members of the public, as a unified group, should have on HE modelling decisions. This is not an easy question to answer, in part because of how difficult it is in modelling practice to separate epistemic and non-epistemic considerations [18]. In principle, we might want to say that, where a modelling decision is to be driven by epistemic values, the final deciding power should be granted to researchers who are trained in the sciences and thus are likely to have better intuitions – whereas if a modelling decision is to be driven by social and ethical values, the final deciding power should be granted to patients and members of the public, as HE modelling should serve *their* interests, not researchers'. Regrettably, this simplistic principle stands to be difficult to follow in practice, because as we noted above, many modelling decisions reflect a mixture of social and ethical value judgements and epistemic judgements. This, we argue, is a core complexity we will face in developing normative guidance for PPI in HE modelling.

4 Conclusions

To help appropriately manage value judgements in HE modelling, PPI is a key strategy, but PPI itself raises value-laden questions that invite discussion. We have argued that there is need to develop normative guidance that is informed by philosophical thinking, empirical research and input from the wider community. Such guidance should speak to a number of complex questions, just some of which we have addressed here, and will take significant initiative to develop. For the time being, there is reason to reflect on existing guidance for best modelling practices [14, 46, 47], recent experience [9–11, 33] and the need for transparency in the context of PPI [22, 23].

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Declarations

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