Addressing Health Equity in Cost-Effectiveness Analysis: A Review of Distributional and Extended Cost-Effectiveness Analysis.

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Preamble

1. Copyright

2. Plagiarism declaration

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3. Acknowledgements

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Part A: Protocol

Addressing health equity in cost-effectiveness analysis:

a review of distributional and extended cost-effectiveness analysis.

1. Introduction

Health is universally valued (Marmot & Bell 2018) and almost all nations acknowledge the need for fairness in the provision of health care. For this reason, health interventions should aim to improve both overall health as well as the fair distribution of health in a population (WHO 2014). Researchers and policymakers are increasingly interested in finding ways to measure the impact of health policies on equity (Cookson *et al.* 2017).

Although there is no universally agreed definition of equity (Mooney 2003) Whitehead's definition of inequity will be used in this study:

> *'the term inequity […] refers to differences in health which are not only unnecessary and avoidable but, in addition, are considered unfair and unjust' (Whitehead 1991 p. 219).*

Implicit in this definition is that health inequity is not simply inequality in access to health resources, but also encompasses unfair inequalities which may violate human rights (WHO 2019). Despite the importance of health equity, equity is not adequately addressed in practice (Cookson *et al.* 2017).

1.1. Using cost-effectiveness analysis to address health equity concerns

Methods for assessing how best to maximise health, such as cost-effectiveness analysis, are well developed. While these techniques are able to assist decision-makers in making choices of how to most efficiently allocate health resources, they fail at informing healthcare priorities (Brousselle & Lessard 2011; Norheim *et al.* 2014). Economic evaluations which incorporate equity considerations are required for this task. However, equity is not given sufficient consideration when evaluating the impact of health policy decisions (Cookson *et al.* 2017)

After 2010, health economists became increasingly interested in finding ways to incorporate equity concerns within CEA and how best to measure the impact of health

policies on equity (Cookson & Mirelman 2017). In 2012 Culyer noted that, apart from a few exceptions, health economists had been 'quite extraordinarily bad' at providing practical decision-making tools to address equity considerations (Culyer 2012, p. E29) However, following this criticism, two new techniques, which are the focus of this study were developed in order to incorporate equity into cost-effectiveness analysis. First, Verguet, Laxminarayan and Jamison developed a method which they called 'extended cost-effectiveness analysis' (ECEA). They published their methodology in a study of universal public finance for rotavirus vaccination in India and Ethiopia (Verguet *et al.* 2013). In 2015, Asaria, Griffin, Cookson and colleagues at the University of York in the United Kingdom described a comparable technique they termed 'distributional cost-effectiveness analysis' (DCEA). They described the technique in a study based on a UK National Health Insurance programme for the screening of bowel cancer (Asaria *et al.* 2015). While CEA compares the relative costs and gains in health outcomes of an intervention, both DCEA and ECEA build on this capacity to also address equity (Griffin *et al.* 2014).

The development of DCEA and ECEA is a significant step forward in providing researchers and decision-makers with useful tools to incorporate equity considerations into cost-effectiveness analysis. DCEA and ECEA are unique in that they are the only two methods which specifically assist policymakers to decide which interventions will provide the best value for money when addressing concerns for health inequality (Love-Koh *et al.* 2019).

Prior to DCEA and ECEA, there were several techniques for incorporating health inequity concerns in economic evaluations. These typically involved 'equity weighting' of costs or outcomes (Nord *et al.* 1999) or using 'multicriteria decision analysis' (Baltussen & Niessen 2006). Both these approaches have subsequently been incorporated into DCEA, which provides a flexible framework rather than an alternative competing technique (Asaria *et al.* 2015).

1.2. Why focus on DCEA and ECEA?

This study will focus specifically on DCEA and ECEA for three reasons: firstly, DCEA and ECEA are likely to become the dominant methods of equity evaluation. This is because they are both flexible frameworks and so, as mentioned, they are able to incorporate existing techniques such as equity weighting or multi-criteria decision analysis into their

methodologies (Asaria *et al.* 2015). In this way, DCEA and ECEA have surpassed previous approaches to addressing equity in economic evaluations. Secondly, DCEA and ECEA allow for a number of different interventions to be compared for their health equity impacts (Lal *et al.* 2018), which is not possible with other equity evaluations. Thirdly, in the future, as new methods for evaluating and addressing equity are developed, they are likely to use and further develop either the DCEA or ECEA framework.

While DCEA and ECEA are excellent tools for simultaneously addressing both efficiency and equity, they also bring a significantly increased degree of complexity. Health inequality measures are complex to interpret and are easy to misinterpret (Lal *et al.* 2018). Decision makers who are concerned about reducing inequity in health would benefit from an overview and clearer understanding of DCEA and ECEA. This would enable them to recognise the limitations of the two techniques and to better interpret the outcomes of these economic evaluations.

For health economists and other experts conducting a DCEA or ECEA, the methodology of DCEA and ECEA has been described by Asaria *et al.* (2016) and Verguet *et al.* (2016), respectively. There are no equivalent resources available for decision makers. To date, there have been no studies which critically compare DCEA and ECEA, or which provide decision makers with an overview of the relative strengths, weaknesses or applicability of each approach. This study will go some way to filling this knowledge gap.

2. Aim and research questions

The overall aim of this study is to assist decision makers to better understand DCEA and ECEA and to recognise the strengths and weaknesses of each framework. This knowledge should help them to decide which, if any, of the two approaches is best able to address their particular equity concerns.

This study aims to address three research questions:

1. How do distributional cost-effectiveness analysis (DCEA) and extended cost-effectiveness analysis (ECEA) address equity concerns?

2. In what ways are DCEA and ECEA similar and/or different?

3. What are the relative strengths and limitations of DCEA and ECEA?

3. Justification for the study

A recurrent theme in the literature is that despite the importance of equity when considering policy decisions, equity evaluations are not given sufficient weight in decision-making (Cookson *et al.* 2017). One reason is that, up until the development of DCEA and ECEA, the available tools to incorporate equity into economic analyses were inadequate (Culyer 2012). While DCEA and ECEA are two effective techniques which address health equity, they are complex and are currently underused (Lal *et al.* 2018). This study aims to provide a clear overview of DCEA and ECEA to encourage researchers and decision makers to consider using these effective tools to address their equity concerns.

4. Methodology

4.1. Scoping literature review

This study will use a scoping literature review to address the three research questions listed in section 2 above. This methodology will be used to identify the key journal articles and important concepts in addressing equity in economic evaluations (Arksey & O'Malley 2005). A scoping literature review has been defined as:

> *'a form of knowledge synthesis that addresses an exploratory research question aimed at mapping key concepts, types of evidence, and gaps in research related to a defined area or field by systematically searching, selecting, and synthesising existing knowledge' (Colquhoun et al. 2014 p. 1292).*

A scoping review is a methodology well-suited for this study for two reasons: firstly, this study will focus on evaluating and synthesising information on two methods of economic evaluation, DCEA and ECEA, rather than a focus on, for example, the outcomes of a medical intervention from a range of comparable studies. Secondly, because DCEA and ECEA are relatively new techniques, and published literature on the techniques is limited, this review can be strengthened by incorporating unpublished (grey) literature. This

ability to incorporate both published and unpublished data is a strength of the scoping review (Levac *et al.* 2010).

The scoping review will be as comprehensive as possible in order to identify both published and unpublished articles, as well as reviews which will assist in addressing the research questions. The literature search will seek evidence from diverse sources, including electronic databases, reference lists, citation lists, and online documentation, discussion papers and presentations. These sources are discussed in more detail below.

4.1.1. Electronic databases

The literature search will be conducted using PubMed and Google Scholar. Duplicate references will be removed, and the remaining articles will be reviewed, and those which are relevant to addressing equity in economic evaluations will be retained.

4.1.2. Reference lists

The reference section of relevant articles will be searched to identify possible studies which would strengthen the review. In cases where these articles were not captured in the initial search, they will be appraised and where appropriate, will be included.

4.1.3. Citation lists

Additional searches will be conducted using the 'cited by' function of Scopus, Google Scholar and journal websites. This function lists other articles which have cited an earlier study.

4.1.4. Online documentation, discussion papers and presentations

Online documentation, discussion papers and presentations are important because they may assist the reviewer to better understand the development and characteristics of DCEA and ECEA. Of particular interest are the discussion papers from the Centre for Health Economics at the University of York, where DCEA was developed (see, for example, Love-Koh *et al.* (2019)). These documents, which form the basis of subsequent journal publications, provide an insight into the challenges faced in developing DCEA.

4.2. Two phases of the scoping review

The scoping review will be undertaken in two phases: the first phase will be a broad scoping review which will focus on the incorporation of equity concerns into economic

evaluations. The second phase will be a more focused review of DCEA and ECEA. These phases are discussed in more detail.

4.2.1. Phase 1: Scoping review of incorporating equity into economic evaluations

The aim of Phase 1 of the scoping review is to identify and synthesise the relevant literature and to identify gaps in the current knowledge on the incorporation of equity into economic evaluations (Levac *et al.* 2010). This stage is designed to be broad in order to include information relevant to quantitative economic analyses which explicitly address equity issues. The search strategy for PubMed is outlined in Table 1. The searches for Google Scholar will be similar.

Table 1. PubMed search strategy for Phase 1

Search #1	"Costs and Cost Analysis" [MeSH terms]
Search #2	“health equity” [MeSH Terms]
Search #3	(#1) AND (#2)

As discussed, results from this initial search will be supplemented by reviewing the reference lists of the selected articles.

4.2.2. Phase 2: Scoping review of DCEA and ECEA

Phase 2 will identify and synthesise the literature addressing DCEA or ECEA and will include studies that have used these two techniques. The search strategy for PubMed is outlined in Table 2.

Table 2. PubMed search strategy for Phase 2.

Search #1: "extended cost" [All Fields]
Search #2 "distributional cost" [All Fields]
Search #3: (#1) OR (#2)

The aim of this phase is to ensure that articles which refer to DCEA or ECEA are included. As with Phase 1, the references listed in these articles will be examined for other relevant references that may have been missed.

4.3. Selection of studies

There is limited information available on DCEA and ECEA. This is because the techniques were only developed from around 2014, and they fall within a specialised area of health economics. For this reason, the challenge will be to find as many as possible of those sources which address or mention DCEA or ECEA. References, abstracts and, where available, complete journal articles, will be stored using Endnote electronic reference manager (2013). Duplicate references will be removed, and the remaining references screened for relevance. Those articles which are not relevant to addressing equity using DCEA or ECEA will be excluded. There will be no language exclusion. The abstracts of articles in languages other than English will be translated into English using Google Translate and if they address DCEA or ECEA they will be fully translated and included in the review.

4.4. Data synthesis

This review will focus on describing and synthesising the important characteristics of DCEA and ECEA. In particular, the study aims to identify characteristics which would discriminate between DCEA and ECEA. In order to facilitate this process, the selected references from Endnote will be imported into NVivo qualitative data analysis software Version 12. This data analysis software will be used to assist in identifying the central themes and develop a structure to examine, evaluate and compare DCEA and ECEA.

5. Limitations of the study

As this study focuses on two equity evaluation techniques, rather than the outcomes of a variety of studies, this scoping review will not progress to a systematic review. Systematic reviews typically combine, analyse and draw conclusions from the results of a number of similar and comparable studies. Although there have been more than 20 studies using ECEA (Verguet 2017), these studies have included a wide range of different settings and a variety of health policy applications. Examples of ECEA studies include the treatment of schizophrenia in India (Raykar *et al.* 2016), the introduction of a tobacco tax in Lebanon (Salti *et al.* 2016), and the financial impact of a measles immunisation programme in Ethiopia (Driessen *et al.* 2015). A scoping literature review is the preferred approach to synthesise the literature from this wide variation in health policies and settings

6. Research ethics

6.1. Ethics approval

The proposed study is a structured review based on published or publicly reported literature. The information is in the public domain, and no human or animal participants will be used for the study. Privacy and confidentiality will not be issues of concern. For these reasons, this study will not require ethics approval from the Human Research Ethics Committee of the University of Cape Town, Faculty of Health Sciences.

6.2. Risks and benefits

There are no anticipated risks since the study has no human or animal participants. The potential benefit from the study is that a better understanding DCEA and ECEA can assist decision-makers to make rational and fair decisions for the equitable allocation of healthcare, In addition, the study can guide researchers to choose between using DCEA and ECEA, based on their context and data availability.

7. Time Frame

The proposed time frame for the dissertation is outlined in Table 3.

Table 3. Proposed time frame

Task Date	June 2020	July 2020	Feb 2021	July 2021
Proposal				
Literature review				
Write-up				
Prepare manuscript for publication				
Thesis submission				

8. Budget

This study is self-funded, with no major costs being foreseen and forms part of the dissertation for a Master of Public Health in Health Economics.

9. Dissemination of study findings

The results of this study form part of a dissertation for a Master of Public Health in Health Economics. On completion of the dissertation, the journal manuscript will be submitted to the Journal of Health Economics or another similar peer-reviewed journal for consideration for publication.

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Part B: Literature Review

Integrating equity into cost-effectiveness analysis.

1. Introduction

Both DCEA and ECEA extend the capabilities of cost effectiveness analysis in order to address equity concerns. To better understand DCEA and ECEA this literature review will:

1. Place DCEA and ECEA in the context of economic evaluations which address equity.
2. Review concepts central to the understanding of DCEA and ECEA, such as opportunity costs, the trade-off between equity and cost-effectiveness, and financial risk protection.
3. Compare DCEA and ECEA and highlight the strengths and weaknesses

This literature review first describes cost-effectiveness analysis and highlights the limitations to addressing equity issues within cost-effectiveness analysis. Next, approaches to defining health equity are discussed, and the most suitable definition is defined for the purposes of this study. The difference between equity and equality are explored as are the links between health inequity and poverty. Building on this background, methods for measuring health equity and the techniques to incorporate equity into cost effectiveness analysis are explored. Fourth, with this background, DCEA and ECEA are discussed in more detail. Examples of DCEA and ECEA studies are captured in order to compared DCEA and ECEA in terms of similarities and differences. The significant differences between DCEA and ECEA are highlighted by examining how they analyse trade-offs between improving health and reducing inequity, address financial impacts of health policies and incorporate health opportunity costs. Finally, the review summarises some of the future improvements recommended for DCEA and ECEA.

The search strategy is described in Part A: Protocol (above). In summary, the search strategy review was in two phases: Phase 1 was a broad scoping review which identified and synthesised the relevant literature and identified gaps in the current knowledge on

the incorporation of equity into economic evaluations (Levac *et al.* 2010). Phase 2 built on this and focused on the literature which specifically addressed DCEA or ECEA.

2. Review of the literature

2.1. Background

While health is universally valued, access to health care and health outcomes are unfairly distributed across and within countries (Love-Koh *et al.* 2019b; Marmot & Bell 2018) . These differences are apparent across global, regional, and local levels. At a global level, for example, a child born in the World Health Organisation (WHO) African region is six times more likely to die before completing their first year of life compared with a child born in the WHO European region (WHO 2020). At a regional level, within Southern Africa, the life expectancy at birth for a child born in 2018 in a relatively poor country such as Lesotho is 54 years, while a child born in South Africa has a life expectancy of 64 years and a child born in Mauritius has a life expectancy of 74 years (The World Bank 2020). This follows a trend of increasing life expectancy with increasing per capita gross domestic product as shown in Table 4.

Table 4. Increasing life expectancy with increasing per capita income for three Southern African countries (The World Bank 2020)

Country	Life expectancy at birth (2018)	Per capita gross domestic product in US Dollars (2018)
Lesotho	54	1 299
South Africa	64	6 374
Mauritius	74	11 238

At a local level, within South Africa, in 2012 a five-year-old child is 2.8 times more likely to die in the relatively poor and largely rural province of KwaZulu-Natal than a five-year-old in the wealthier Western Cape (Nannan *et al.* 2019). For most nations, reducing health inequality and improving health equity is a key goal. For this reason, health policies should aim to improve overall health and reduce the unfair distribution of health in the population (WHO 2014).

Current methods for assessing how best to maximise health, such as cost-effectiveness analysis (CEA) and cost-benefit analysis (CBA) are well developed. While these techniques can assist decision makers in making choices on how to most efficiently allocate health resources, they fail at informing healthcare priorities (Brousselle & Lessard 2011; Norheim *et al.* 2014). Economic evaluations which incorporate equity considerations are required to address equity concerns. Two promising solutions which enhance CEA to address equity have been developed: distributional cost-effectiveness analysis (DCEA) and extended cost-effectiveness analysis (ECEA). However, these solutions are not yet widely applied or understood and so they will be the focus of this literature review.

2.2. Health economic evaluations and cost-effectiveness analysis

Economic evaluation is defined as the 'comparative analysis of alternative courses of action in terms of both their costs and consequences' (Rudmik & Drummond 2013 p. 1341). In other words, economic evaluations aim to 'identify, measure, value, and compare the costs and consequences of two or more alternative interventions' (Drummond *et al.* 2015 p. 9). This is shown diagrammatically in Figure 1.

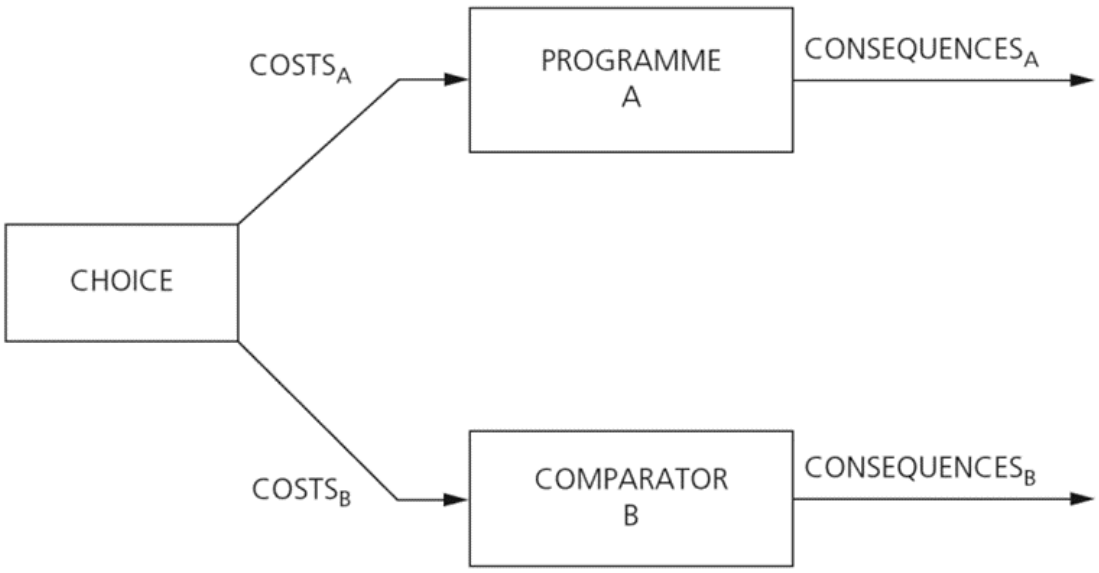

Figure 1. Economic evaluation involves a comparative analysis of alternative courses of action (Drummond *et al.* 2015)

Cost-effectiveness analysis (CEA) is an economic evaluation tool which identifies interventions which maximise health benefits and minimise health cost (Ashok *et al.* 2016). The technique provides a clear and straightforward method to establish the best value for money in health (Musgrove & Fox-Rushby 2006) and has become the '*de facto* normative standard to evaluate medical interventions' (Phelps & Madhavan 2019 p.505) with over 7300 published CEA studies and several specialised journals dedicated to CEA methodologies (Phelps & Madhavan 2019)

CEA enables comparisons across different health outcomes (Musgrove & Fox-Rushby 2006), which makes it highly suitable for comparing different health policies and interventions. CEA measures the outcomes of an intervention as relevant natural units such as 'quality-adjusted life-year (QALY) gained' or 'disability-adjusted life year (DALY) averted'. Typically, the final summary measure in CEA represents the benefits and costs of each intervention as an incremental cost-effectiveness ratio (ICER) (Rutstein *et al.* 2017).

A limitation of CEA is that it is not designed to address health inequality (Sassi *et al.* 2001). This is because CEA assigns equal value to each unit of health gain with no regard to the characteristics of the recipients. Maximising health, as measured by maximising the number of Quality-Adjusted Life Years (QALYs) irrespective of who receives those QALYs and how those QALYs are distributed has been encapsulated in the expression 'a QALY is a QALY'. This approach has been criticised, see for example Dolan *et al*. (2005) and Wailoo *et al.* (2009). One such criticism is that CEA cannot incorporate preferences shared by many people, such as giving priority to severely ill patients or to those who are disabled (Ubel *et al.* 2000).

CEAs generally do not evaluate non-health benefits of interventions, such as improvement in an individual's performance at work or school as a result of improved physical or mental health, unless these outcomes can be quantified as costs (Musgrove & Fox-Rushby 2006). Significant non-health benefits, including the impact on equity or the prevention of medical impoverishment through financial risk protection, require more sophisticated equity analyses (Ashok *et al.* 2016; Levin *et al.* 2015). The aim of many public health interventions is to reduce health inequities. For this reason, techniques

which can address equity issues are essential to evaluate public health interventions (Drummond *et al.* 2006). The focus of this literature review is on DCEA and ECEA, two methods which expand CEA to address equity issues.

2.3. Health equity

2.3.1. Defining health equity

Health is regarded as a 'special good' because it influences a person's well-being which allows them to pursue goals that they value. In this way, health determines a person's capacity to function (Anand 2002). The WHO constitution states that:

> *the enjoyment of the highest attainable standard of health is one of the fundamental rights of every human being without distinction of race, religion, political belief, economic or social condition (WHO 1946)*

This viewpoint is consistent with a human rights approach and includes the right to have access to effective healthcare that helps an individual. People are entitled to maintain their health, to diminish the chances of developing a disease, and, where this fails, to reduce the impact of a disease (Whitehead & Dahlgren 2006).

Health equity refers to the fairness in the differences in health between individuals or groups. While there is no universally agreed definition of equity (Mooney 2003), Margaret Whitehead's definition has been widely cited:

> *'inequity […] refers to differences in health which are not only unnecessary and avoidable but, in addition, are considered unfair and unjust' (Whitehead 1992 p. 430).*

Implicit in this perspective is that health inequity is not merely a difference in health, but the concept also encompasses unfair inequalities which may be unethical and may violate human rights (Braveman & Gruskin 2003; WHO 2019).

Within this ethics-based definition there are two broad approaches to defining equity which are based on the sources of health inequality. These overlapping approaches regard inequalities as unfair where they are, in the first approach, due to factors beyond the control of the individual or, in the second approach, reflect the degree of social disadvantage (Asada *et al.* 2015). The limitation of the first approach is that determining

which factors are beyond the control of an individual is not straightforward; it could be argued that cigarette smoking is within the control of the individual and so health differences between those who smoke and those who do not smoke should not be regarded as unfair. However, this view does not adequately consider the higher prevalence of smoking and lower smoking cessation rates in individuals from lower socioeconomic groups (Siahpush *et al.* 2010). The second approach, focuses on social advantage and is encapsulated in Braveman and Gruskin's definition of health equity as:

> *'the absence of systematic disparities in health [...] between social groups who have different levels of underlying social advantage/disadvantage' (Braveman & Gruskin 2003 p. 254),*

This definition recognises that individuals or groups of people who occupy lower positions in a social hierarchy, for example, those who are poor or the elderly, typically have limited access to healthcare and so their health is further compromised (Braveman & Gruskin 2003).

Whitehead's definition of inequity in which 'differences in health [...] are considered unfair and unjust' becomes difficult to delineate because what is considered fair and just in health differs across cultures (Starfield 2006). WHO has overcome this complexity by avoiding judgements of fairness and, instead, has defined equity as:

> *'the absence of avoidable or remediable differences among groups of people, whether those groups are defined socially, economically, demographically, or geographically'(WHO 2019).*

This definition will be used in this review because it incorporates Whitehead's idea of avoidable or remediable differences as well as Braveman and Gruskin's inclusion of the importance of different subgroups. Both concepts are central to evaluating equity using economic analyses.

2.3.2. Equity versus equality

The difference between equity and equality is well captured in Maguire's illustration (Figure 2).

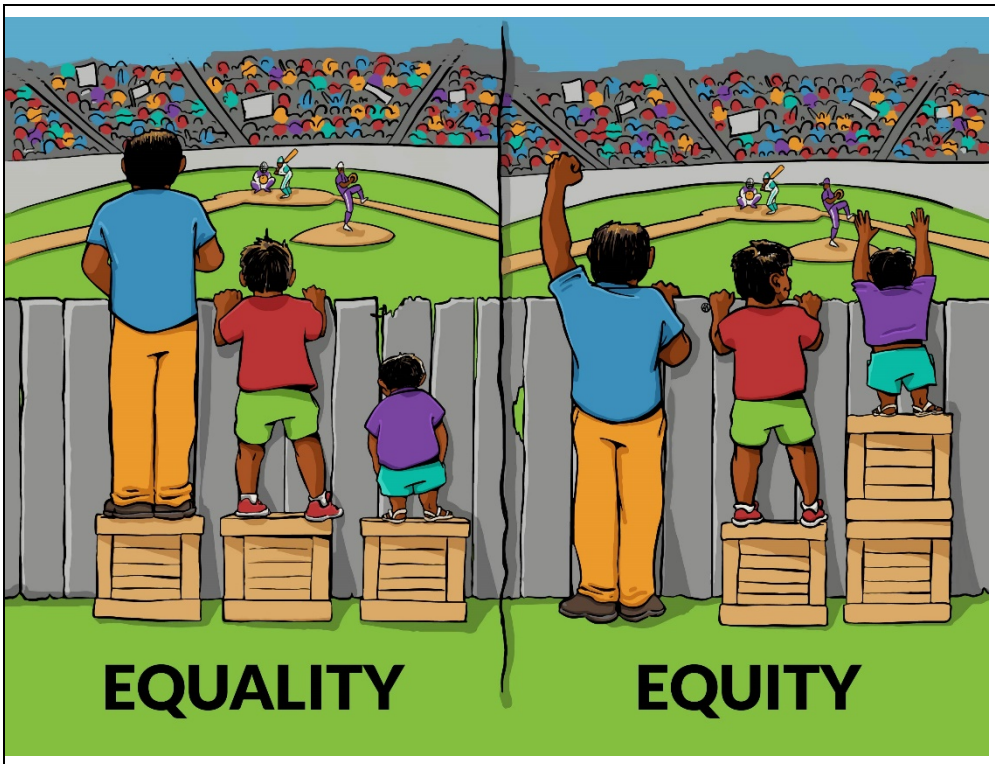

Figure 2 Illustration of the difference between equality and equity.

This image is free to use with the attribution: "Interaction Institute for Social Change, Artist: Angus Maguire."

'Health inequality' and 'health inequity' are often used interchangeably. However, they are materially different. Health inequality is the: 'systematic [difference] in the health of groups and communities occupying unequal positions in society' (Graham 2004 p. 101), while health inequity is a specific type of health inequality in which this systematic difference is judged to be avoidable or remediable by reasonable means (Arcaya *et al.* 2015). Both health inequality and health inequity exist across a wide range of social factors including gender, income, deprivation, and geography.

Inequality is a dimensional description of unequal quantities and reflects inherent differences between groups, for example, the differences in rates of myocardial infarction in men and in women. In contrast, differences in health in different social groups, such as those based on income or race, are likely to be health inequities because they usually reflect an unfair distribution of health risks and resources. In this way, there may be fair and unfair health inequalities. To illustrate this, the inequality in the incidence of

dementia across age groups is not regarded as being inequitable, although the funding for treating dementia compared with treating myocardial infarction may be.

There are occasions when some inequalities may be equitable, for example, treating someone who needs urgent medical care ahead of others who are in less urgent need (Culyer & Bombard 2012). Similarly, not all equalities are equitable, for example, providing the same level of physical access for patients to a community clinic is likely to unfairly disadvantage someone who is in a wheelchair. Equity has been categorised as *horizontal* when people of equal need receive equal resources and *vertical* where people who have unequal (higher) needs receive unequal (more) resources (Mooney 2003).

The focus of this study is on economic evaluations which address inequities which may be amenable to changes in health policy such as improving care to those who are socially disadvantaged, rather than innate differences in health outcomes. For this reason, this review will focus on equity and inequity rather than on equality and inequality, an approach which is consistent with that taken by Whitehead (1992), and Culyer and Bombard (2012).

2.3.3. Health inequity and poverty

Poverty and ill-health are linked. As discussed in the introduction, on average people living in poor countries have worse health outcomes than those living in wealthy countries. While these differences in health outcomes may be striking, the differences in health outcomes between a country's most and least privileged groups may be even more remarkable (WHO 2013). In all countries, including those which are well-resourced, illness follows an income gradient, where those lowest in the socioeconomic hierarchy carry the greatest burden of illness (WHO 2013). The relationship between poverty and ill-health is causal in both directions: 'poverty breeds ill-health, and ill-health keeps poor people poor' (Wagstaff 2002 p. 97).

Not only do the poor carry a higher burden of disease, but their access to healthcare is also typically limited. This imbalance has been encapsulated in the Inverse Care law, proposed by Hart in 1971, which states that the 'availability of good medical care tends to vary inversely with the need of the population served' (Hart 1971 p. 412). Hart based this law on his observations of limited health services in impoverished Welsh mining villages where the need for care was high, compared to the surrounding more prosperous

towns which had better facilities, and fewer health service requirements. Since Hart's description, this pattern has remained unchanged. A systematic review of equity in low- and middle-income countries (LMICs) found that individuals who are relatively well-off and carry a lower burden of disease, utilise healthcare services more than do those who are poor (Asante *et al.* 2016). This pro-rich pattern is particularly noticeable in the accessing of healthcare at hospitals, most of which are situated in urban areas (Asante *et al.* 2016).

2.3.4. Measuring health equity

Economic evaluations guide the allocation of limited resources. However, implementing the recommendations of standard economic evaluations, such as CEA, may often further disadvantage some social groups that the health system is meant to protect (Sassi *et al.* 2001). For this reason, it is essential to consider equity implications when allocating healthcare (Wailoo *et al.* 2009). However, despite the recognised importance of health equity, it is not adequately addressed in practice and only rarely included in economic evaluations (Cookson & Mirelman 2017; Johri & Norheim 2012). While methods to measure health inequality had been developed before 2013, they were used infrequently and did not assist with decision making (Cookson 2013). Culyer, writing in 2012, noted that, with a few exceptions, health economists had 'been quite extraordinarily bad' at providing practical tools to enable decision makers to address equity concerns (Culyer 2012 p. E27). At that time, there were no adequate available techniques which could assist health economists to incorporate equity considerations into efficiency analyses (Culyer 2012). However, following this criticism, this deficiency was addressed with the development of DCEA and ECEA. The two techniques used different approaches, and each was able to provide a significant advance in addressing equity in economic evaluations.

2.4. Incorporating equity into cost-effectiveness analysis

While there are no published reviews directly comparing DCEA and ECEA, there are four systematic reviews which address the incorporation of equity considerations into economic evaluations. Both the reviews, published prior to 2006, noted that despite the importance of health equity, equity is rarely included in studies using cost-effectiveness analysis. In the first of these, a systematic review by Sassi *et al.* (2001) of 424 economic evaluations published between 1987 and 1997, concluded that health economic studies seem to have 'completely neglected' distributional effects and did not provide a guide or

the information to enable decision makers to allocate resources in an equitable manner (Sassi *et al.* 2001 p. iv). The review concluded that formal approaches to integrate equity into CEA were inadequate and needed urgent development. In the second review, Drummond and other colleagues (including Cookson) described the methodological challenges faced in 154 economic evaluation studies published from 2000 to 2005 (Drummond *et al.* 2006). The authors identified four main challenges, one of which was how to include equity considerations in economic evaluations. Like Sassi and colleagues, they found that equity was rarely considered and never addressed directly in the studies they reviewed (Drummond *et al.* 2006). Both Sassi *et al.* and Drummond *et al.* highlighted the need for formal approaches to integrate equity into CEA.

In the third review, Johri and Norheim (2012) conducted a systematic review of formal methods used to incorporate equity in CEA. The review, which ended in January 2011, found that there were significant advances in incorporating equity into economic evaluations. They reviewed 51 eligible studies and delineated three broad approaches to integrating equity. These were 'equity weights', 'exploration of opportunity costs' and 'multi-criteria decision analysis'. Equity weights and exploration of opportunity costs will be discussed more fully in section 2.4.2 (below). Multi-criteria decision analysis is a systematic approach which can help resolve complex and multifaceted health decisions. The technique enables health economists to shift from a single focus (such as maximising population health) to simultaneously consider other relevant criteria such as the distribution of health and societal preferences (for example, for providing primary health care in rural areas) (Baltussen & Niessen 2006).

More recently, and following the development of DCEA and ECEA, Lal and colleagues (2018) conducted a systematic review of 29 studies of public health interventions which used 'socio-economic position' in CEA to address equity. Socio-economic position can be measured by income, place of residence, occupation, or education. Like Johri and Norheim they categorised three approaches, and also identified 'equity weights', to which they added the 'use of health inequality measures' and 'inclusion of financial impacts'.

While the reviews by (Johri & Norheim 2012) and (Lal *et al.* 2018) categorise equity evaluations differently, there is some overlap in that both reviews included 'equity weights' as a category. However, both reviews mix broad umbrella terms [such as 'multi-

criteria decision analysis' (Johri) and the 'use of health inequality measures' (Lal)] with highly specific approaches [such as the 'exploration of opportunity costs' (Johri) or the 'analysis of financial impacts' (Lal)].

A more consistent classification of equity evaluations was put forward by Cookson, who collaborated with other colleagues, including Norheim, a co-author of the 2012 review, to publish an overview and guide on using CEA to address health equity concerns (Cookson *et al.* 2017). They identified two broad approaches: equity impact analysis and equity trade-off analysis. There are two methods to equity trade-off analysis: equity constraint and equity weighting. To clarify, the authors propose the following classification:

1. Equity impact analysis
2. Equity trade-off analysis
    a. Equity constraint
    b. Equity weighting

These approaches to incorporating equity into economic evaluations are discussed in more detail below.

2.4.1. Equity impact analysis

Equity impact analysis disaggregates population data into different subgroups to determine how each subgroup stands to gain or lose from the implementation of a policy. These subgroups are determined based on 'equity-relevant' variables, such as age, sex, or socioeconomic status (Cookson *et al.* 2017 p. 206).

2.4.2. Equity trade-off analysis

Equity trade-off analysis is used in situations where the most cost-effective option is inequitable (Cookson *et al.* 2014). The analyst quantifies the opportunity costs of not selecting that option, which is a measure of the overall health forgone when selecting a more equitable but less cost-effective option. Economic evaluations of public health policies have two approaches to address this trade off: equity constraint analysis and equity weighting.

2.4.2.1. Equity trade-off analysis: equity constraint

In this approach, the incorporation of equity considerations 'constrains' the degree to which an intervention can be entirely cost-effective. In this way, equity constraint analysis determines the opportunity cost of choosing a policy which favours equity compared with one which maximises total health. The health forgone by selecting a more equitable option indicates the strength of the decision makers' concern for equity (Cookson 2016; Weatherly *et al.* 2009).

2.4.2.2. Equity trade-off analysis: equity weighting

Equity weighting was one of the first methods for incorporating equity concerns (Nord *et al.* 1999) and is discussed in all three reviews of equity evaluations by Cookson et al. (2009), Johri & Norheim (2012) and Sassi et al. (2001) . Equity weighting assigns different equity weights to provide extra weight to factors, such as socioeconomic deprivation, which are commonly associated with unfair health outcomes (Love-Koh *et al.* 2019b). In this way, interventions which reduce inequality are given greater weight and therefore more likely to be funded. The extent of the weighting reflects the reduction of health benefits decision makers are willing to accept in return for a given reduction in health inequality (Drummond *et al.* 2006).

The values of the weights are determined by sampling interested parties, such as policymakers or the public. Studies in equity weights indicate that, in general, people prefer health gains to accrue to the elderly, children, or individuals with severe illness (Round & Paulden 2018). Equity weighting is a method of reflecting this preference. While this approach is appealing, in practice deciding equity weighting has proved challenging (for review see Wailoo *et al.* 2009) and further research on how best to weight equity concerns in a public health context is required (Jamison *et al.* 2018). A significant limitation in the context of this study is that few of the studies using equity weighting have addressed social characteristics such as income or ethnicity (Cookson *et al.* 2017).

2.5. Introducing DCEA and ECEA

After 2010, health economists became increasingly interested in finding ways to incorporate equity concerns within CEA and how best to measure the impact of health policies on equity (Cookson & Mirelman 2017). In 2012 Culyer noted that, apart from a

few exceptions, health economists had been 'quite extraordinarily bad' at providing practical decision-making tools to address equity considerations (Culyer 2012, p. E29) However, following this criticism, two new techniques, which are the focus of this study were developed in order to incorporate equity into cost-effectiveness analysis. First, Verguet, Laxminarayan and Jamison developed a method which they called 'extended cost-effectiveness analysis' (ECEA). They published their methodology in a study of universal public finance for rotavirus vaccination in India and Ethiopia (Verguet *et al.* 2013). In 2015, Asaria, Griffin, Cookson and colleagues at the University of York in the United Kingdom described a comparable technique they termed 'distributional cost-effectiveness analysis' (DCEA). They described the technique in a study based on a UK National Health Insurance programme for the screening of bowel cancer (Asaria *et al.* 2015). While CEA compares the relative costs and gains in health outcomes of an intervention, both DCEA and ECEA build on this capacity to also address equity (Griffin *et al.* 2014).

While policies may be designed to improve the equitable allocation of health care, it cannot be guaranteed that a reduction of inequality will necessarily follow (Love-Koh *et al.* 2019b). For some diseases and settings, a relatively basic understanding of the distribution of the patient population may be sufficient to inform which group will benefit most (Love-Koh *et al.* 2019b). Where the impacts of a proposed health policy on inequity are more nuanced, analyses like DCEA and ECEA can provide a more sophisticated model of the likely outcomes. The approaches adopted by DCEA and ECEA to address these more complex impacts in health equity are discussed in more detail in the sections that follow.

2.6. ECEA

ECEA 'extends' CEA by addressing the distribution of health benefits, policy costs and financial protection by subgroups. ECEA allows decision makers to consider the impact of different health policies using multiple criteria, and to make explicit choices between competing demands. As illustrated in Figure 3, CEA can be visualised as a technique in which a range of policies are compared on the single criterion of cost-effectiveness

(Verguet *et al.* 2016). In ECEA, the policies are evaluated both in terms of cost-effectiveness and a second criterion such as financial risk protection (FRP).

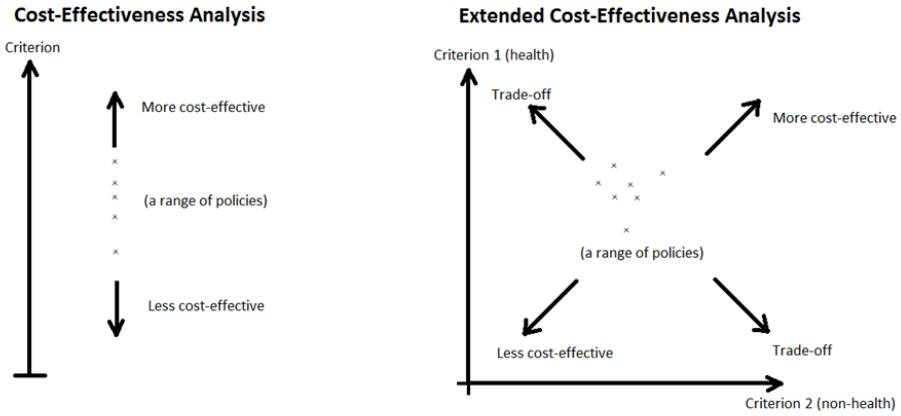

Figure 3: Cost-effectiveness analysis and extended cost-effectiveness analysis (Verguet *et al.* 2016)

ECEA is a technique which follows a sequence of steps outlined in Table 5. These steps include segmenting the population into subgroups (most commonly according to income quintiles) and assessing the net health gains and financial consequences for each subgroup following the implementation of a health policy.

Table 5. Steps to performing ECEA (Verguet et al. 2016).

1. Select the policy of interest.
2. Segment the population into subgroups (e.g. according to income, ethnicity, or sex).
3. Quantify the health gains per population group.
4. Estimate the distribution of health consequences.
5. Quantify the financial consequences for Individuals.
6. Estimate the financial risk protection.
7. Quantify the total costs of the policy.

2.7. DCEA

DCEA, like ECEA, expands the capabilities of CEA to incorporate equity concerns and estimates the health and non-health effects of health interventions across different

groups. DCEA evaluates the health distributions resulting from these interventions in terms of both health inequality and population health. Where necessary, the framework can then be used to guide the trade-off between these different dimensions in order to assist in selecting an option which both improves health and reduces inequity (Asaria 2018).

Asaria *et al.* (2016) have published a tutorial on how to conduct DCEA. The steps are summarised as Table 6.

Table 6. Overview of the steps to perform DCEA (Asaria *et al.* 2016)

1. Estimate the baseline health distribution.
2. Model changes to this baseline distribution due to the health interventions being compared.
3. Adjust the resulting modelled health distributions for alternative social value judgments about fair and unfair sources of health variation.
4. Use the estimated distributions to quantify the change in total population health and unfair health inequality due to each intervention.
5. Rank the interventions based on dominance criteria.
6. Analyse any trade-offs between improving population health and reducing unfair health inequality, allowing for alternative specifications of the underlying social welfare function.

2.8. The similarities between DCEA and ECEA

2.8.1. Equity impact analysis using DCEA and ECEA

Disparities in health are often a result of social factors, including income, ethnicity, or place of residence. These characteristics are essential to understanding the socioeconomic patterns of disease. Some of these health inequities can be reduced by targeted health policies (WHO 2019). Both DCEA and ECEA disaggregate health data by social groups in order to assess not only the total health gains, but also which social groups benefit from these gains.

Disaggregation according to equity-relevant criteria allows analysts to pay attention to the most vulnerable subgroups, including people with disabilities, the elderly, rural and migrant populations, and other marginalised groups. This is crucial, because while national statistics may, for example, show aggregate improvements in access to healthcare or poverty reduction, these aggregate statistics may fail to identify much slower progress for disadvantaged subpopulations (Karlow & O'Donnell 2017). Disaggregated data is essential not only for monitoring populations which are at risk but also for determining which policies may be most effective in redressing health inequities (Karlow & O'Donnell 2017).

Both DCEA and ECEA examine health benefits and costs by social groups. Studies using the two techniques have most commonly delineated social groups according to income (Dawkins *et al.* 2018) although social groups could be stratified on the basis of other equity relevant criteria such as age, sex or ethnicity (Love-Koh *et al.* 2019b).

An example of stratification of health outcomes according to income is shown in Figure 4. Driessen et al. (2015) used ECEA to examine three strategies for improving the coverage of measles immunisation in Ethiopia. The first strategy was routine immunisation, the second strategy was routine immunisation plus financial incentives to

families who were vaccinated, and the third strategy was a mass campaign known as supplemental immunisation activities.

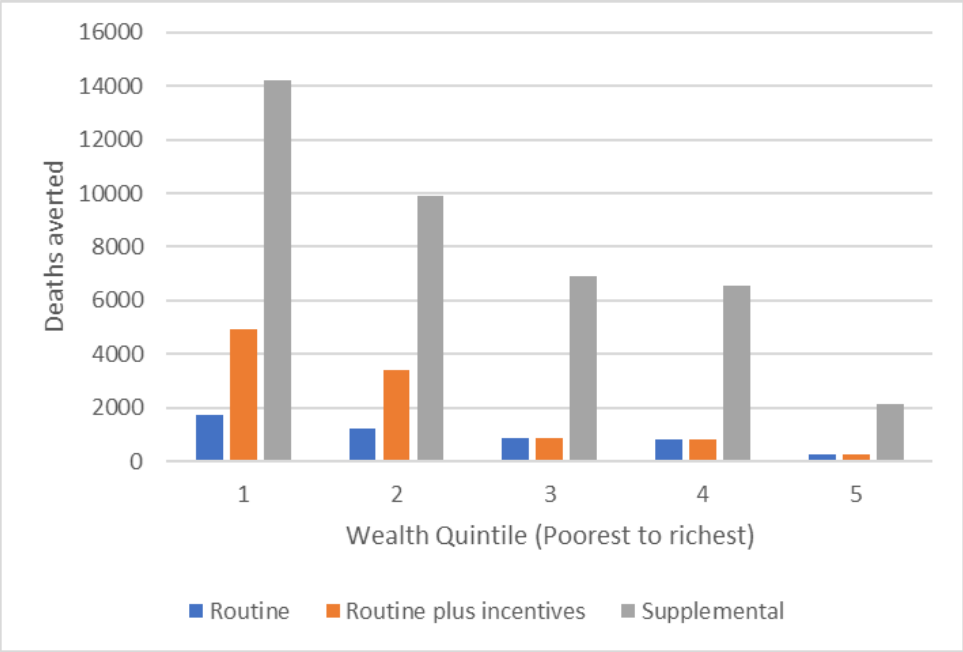

Figure 4. Disaggregation according to wealth quintile in an extended cost-effectiveness analysis of three measles immunisation strategies in Ethiopia (Driessen et al. 2015).

As shown in Figure 4 above, the mass immunisation ('supplemental') strategy averted the most deaths but cost ten times that of the routine immunisation. The financial incentives program averted twice as many deaths overall when compared with routine immunisation. Significantly, there was a three times higher reduction in deaths in the lower two income quintiles, the target group for this financial incentive strategy (Driessen *et al.* 2015).

A similar approach to disaggregating health impacts according to equity related subgroups is seen in a DCEA study by Asaria and colleagues (Asaria *et al.* 2015) who compared two strategies aimed at increasing the uptake of a bowel cancer screening programme in England. A 'targeted' strategy focused on the most deprived social groups which have a low uptake, while a 'universal' strategy sent a standard reminder to everyone in the study. As can be seen in Figure 5, the targeted strategy reduced inequality but resulted in less total health when compared with the universal strategy.

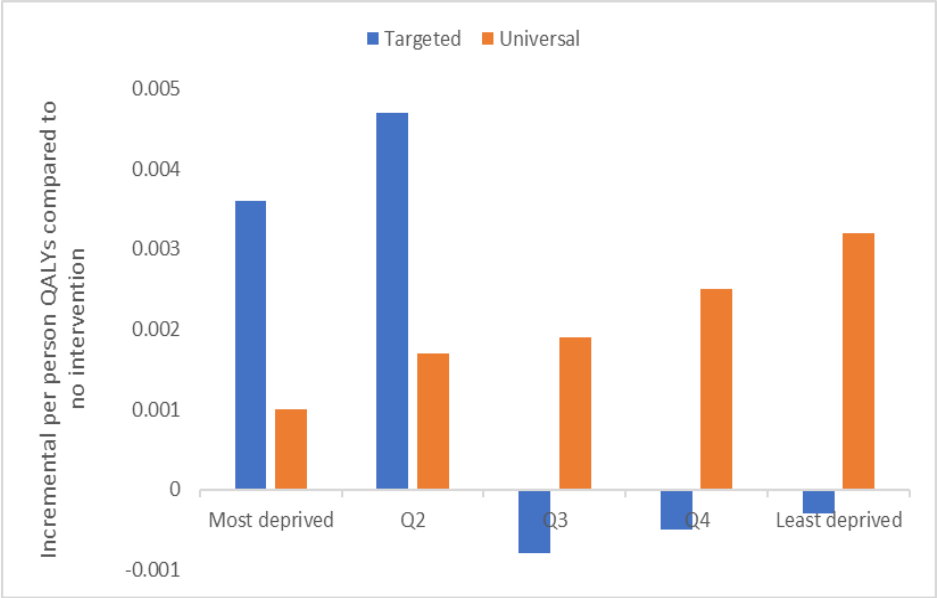

Figure 5. Example of a DCEA output disaggregated according to wealth quintiles in a study comparing strategies for increasing the uptake of bowel cancer screening (Asaria et al. 2015).

In summary, both DCEA and ECEA use similar approaches to disaggregating health benefits and health costs according to subgroups within the population. Both equity impact analyses allow decision makers to determine who gains and who loses from health interventions.

2.9. The differences between DCEA and ECEA

The significant differences between DCEA and ECEA are best illustrated in three areas:

1. Analysing trade-offs between improving health and reducing inequity.
2. Addressing financial impacts of health policies.
3. Incorporating health opportunity costs.

These three key areas of difference are discussed in more detail below.

2.9.1. Analysing trade-offs between improving health and reducing inequity

Decision makers often have to balance the conflicting demands of increasing cost-effectiveness and reducing inequity (Round & Paulden 2018). Efficiency equity trade-off occurs when a more cost-effective intervention may also result in a less equitable distribution of health benefits (Musgrove & Fox-Rushby 2006). These trade-offs can be conceptualised using the health equity impact plane.

2.9.1.1. Health equity impact plane

The relationship between two policy options can be illustrated on the health equity impact plane (Figure 6). This provides a useful method to conceptualise the potential

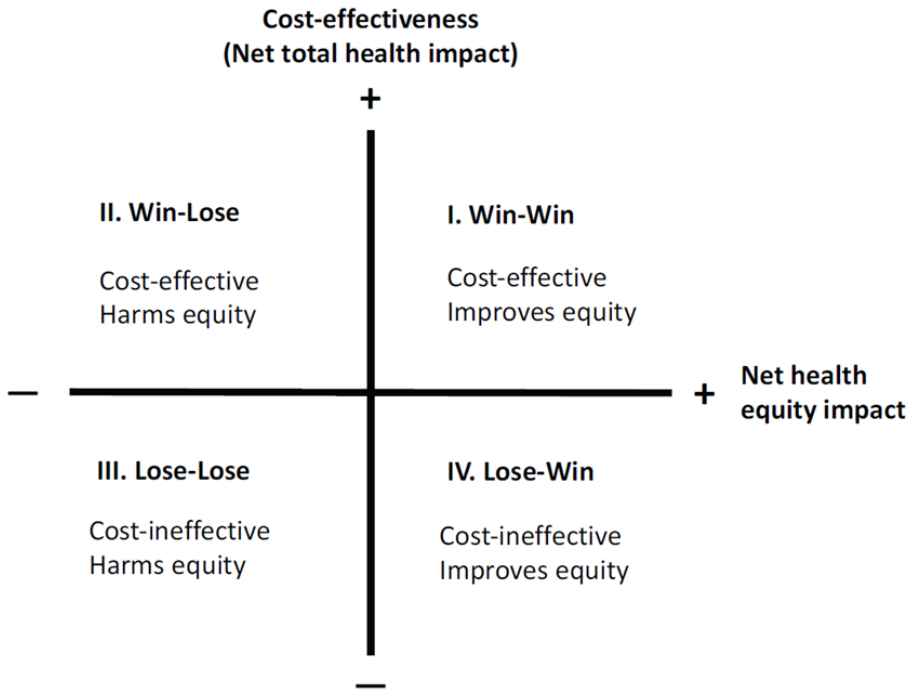

Figure 6. The health equity impact plane (Cookson *et al.* 2017)

trade-offs between a programme promoting cost-effectiveness and one which aims to improve equity.

Both ECEA (Verguet *et al.* 2013) and DCEA (Dawkins *et al.* 2018) have been used to examine a programme for vaccination against rotavirus in Ethiopia. Consider two policy options in this setting. The standard treatment policy aims to maximise health outcomes, while an alternative 'pro-poor' programme, is designed to improve health outcomes, but also reduce inequity by improving provision of vaccinations to disadvantaged rural communities. The predicted outcomes for health benefits and improvements in equity of the pro-poor policy (or other alternative policies) can be plotted on a health equity impact plane. Using the impact plane, the cost-effectiveness of a policy is represented on the vertical axis, while the net health equity impact is represented on the horizontal axis. The standard is the comparator which is represented at the origin, where the vertical and horizontal axes intersect (Cookson *et al.* 2017).

Cost-effectiveness can conveniently be thought of as the net health benefits. That is, the benefits derived from the selected pro-poor policy minus those that would have resulted if that money had been spent on the standard policy (equation 1).

Cost-effectiveness =

Health Benefit of pro-poor policy – Health Benefit of standard policy

Equation 1: Cost-effectiveness

Equity is represented on the horizontal axis. Again, this is the net improvement in equity, in other words, the improvement in equity of the pro-poor policy, less the equity that resulted from the standard policy (equation 2).

Net Equity = Equity pro-poor policy – Equity standard policy

Equation 2: Net equity

Where both cost-effectiveness and equity for a pro-poor policy are positive, the policy falls into Quadrant I and should usually be selected ahead of the standard policy. This is the ideal situation where equity and cost-effectiveness are compatible and may occur when a cost-effective intervention is targeted at a section of the population that carries the highest burden of disease. An example would be the provision of tuberculosis

treatment to impoverished communities in which there are significantly higher rates of tuberculosis than in the general population. On the other hand, if the pro-poor policy, which was expected to improve equity compared to the standard policy, fell into Quadrant III, reflecting a negative impact on both cost-effectiveness and equity, the programme should be rejected. Outcomes which fall into Quadrant I (accept) and Quadrant III (reject) are uncomplicated and do not require analysis beyond standard CEA (Cookson *et al.* 2017). A pro-rich vaccination policy which, compared with the standard policy, is more cost-effective but increases inequity (Quadrant II) would be rejected if improving equity was of primary importance to decision makers.

Where there is a trade-off between increasing health gains and reducing health inequity (Quadrant IV), both DCEA and ECEA can provide an explicit analysis of such trade-offs to assist decision makers in selecting the most suitable intervention (Asaria *et al.* 2016).

2.9.1.2. How DCEA analyses trade-offs

DCEA can assist in resolving the trade-off between reducing inequality and improving health. Where a trade-off exists, the analyst constructs an inequality measure, called the equally distributed equivalent (EDE) level of health, by combining the decision makers' level of aversion to inequality (high, medium, or low) with the distribution of health. The difference between the mean level of health and the EDE reflects the average amount of health per person that a decision maker is willing to sacrifice to achieve full equality in health (Asaria *et al.* 2016). Because the levels of aversion to inequality impact on the outcome of DCEA, the analyst presents a range of alternative levels of aversion to inequality which show decision makers how their selected social value judgements affect the outcome and recommendations from the DCEA.

Dawkins and colleagues (2018) used the health equity impact plane (Figure 7) to demonstrate that a theoretical pro-poor rotavirus vaccination programme in Ethiopia is

likely to promote equity but be less cost-effective compared with the standard program (Quadrant IV).

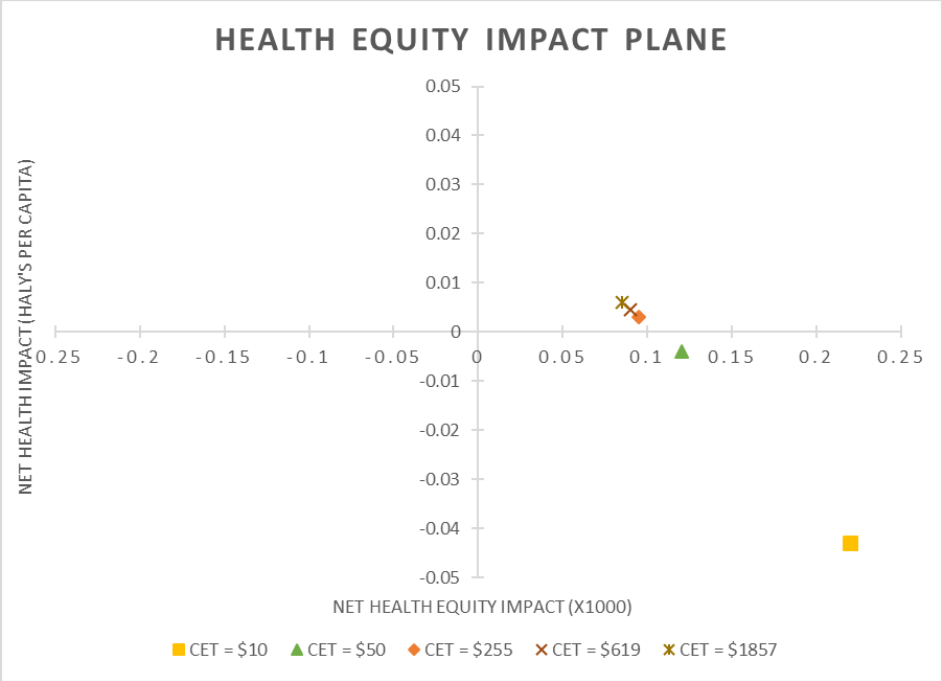

Figure 7. Health equity impact plane for pro-poor rotavirus vaccination programme at different cost-effectiveness thresholds (CET) (Dawkins *et al.* 2018).

At a cost-effectiveness threshold of $50, the pro-poor programme falls into Quadrant IV, reflecting a positive effect on improving equity but a negative impact on total health. The health equity plane in this study also illustrates the impacts of selecting from a range of feasible cost-effectiveness thresholds. The cost-effectiveness threshold in this context is a measure of the opportunity cost and reflects the 'amount of money that, if removed from the healthcare system, would result in one less unit of health being generated' (Woods *et al.* 2016 p. 960).

In some cases, where a policy falls into Quadrant I, as in the example of providing tuberculosis treatment in impoverished communities, there may not appear to be a trade-off as interventions may improve total health and, at the same time, reduce health inequality. However, on closer inspection, many such policies, which appear initially to be 'win-win' in terms of health gains and reducing inequality, have additional costs to reach disadvantaged groups, particularly in rural communities. DCEA can, during the

analysis stage, account for these additional costs, which will help to promote more equitable policies (Dawkins *et al.* 2018).

2.9.1.3. How ECEA analyses trade-offs

ECEA is helpful in comparing a number of potential policy options. For example, Verguet and colleagues (2015) used ECEA to assess health gains (measured as deaths averted) and FRP provided (measured as cases of poverty averted) for nine public health interventions in Ethiopia. They found that measles vaccination and caesarean section surgery averted the most deaths, while caesarean section surgery and tuberculosis treatment averted the most cases of poverty.

This relationship is depicted in a simplified graph of some of their outcomes (Figure 8). ECEA uses a 'dashboard' approach where outputs are presented separately rather than as an aggregated summary measure. This allows decision makers more freedom to choose those aspects of health, equity, or financial protection which they regard as most important for their situation (Cookson *et al.* 2016). This information can assist decision makers to better understand the trade-off between improving health or FRP and to select interventions which meet their specific policy goals.

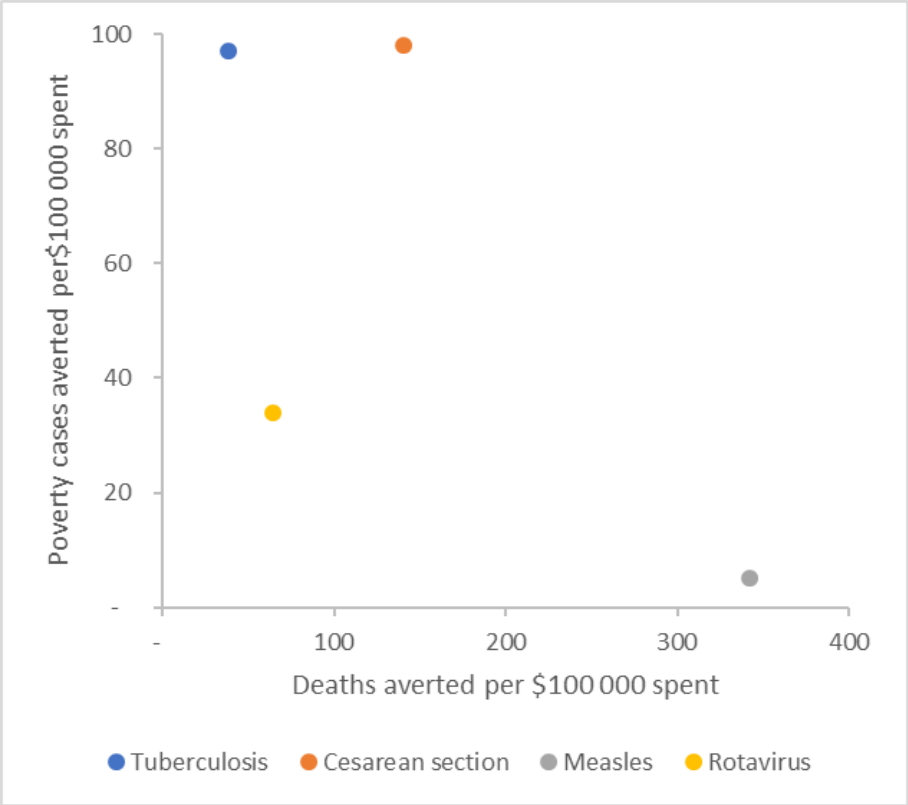

Figure 8. Financial risk protection versus health gains for ECEA study on providing universal public finance for health conditions in Ethiopia (Verguet *et al.* 2015b)

2.9.2. Addressing financial impacts of health policies

Access to health services should be based on need and not on the ability to pay (WHO 2014). Direct payments for health impose a significant barrier to accessing healthcare and exerts a heavy financial burden for millions of households around the world. Financial risk protection is defined as "access to all needed quality health services without financial hardship" (Saksena *et al.* 2014). It is a system designed to safeguard people against financial adversity arising from paying directly for health services (Nugent 2014; Saksena *et al.* 2014). This safeguard is particularly valuable in low- and middle-income settings (Love-Koh *et al.* 2019b) where financial safety nets in the form of unemployment benefits or disability payments are typically not available.

FRP is a target of the Sustainable Development Goals and is a critical aspect of achieving universal health coverage (WHO 2016). To this end, reducing out-of-pocket spending is

an essential step towards preventing extreme poverty (Chang *et al.* 2018). In order to achieve universal health coverage countries must expand priority services, provide health care to more people, and reduce out-of-pocket payments (WHO 2014 p. 4). The challenge is how best to reduce out-of-pocket payments. The WHO report on equity and universal health coverage (2014) suggests the initial step is to reduce out-of-pocket payments for high priority services and for disadvantaged groups. While implementing this step, services should be accessible to those in need, irrespective of their ability to pay.

In a setting with proportionally high out-of-pocket payments, medical costs may lead to impoverishment, financial catastrophe, or forgoing the health service (Ngcamphalala & Ataguba 2018). The lack of financial protection may result in catastrophic health expenditure or impoverishment for the household. Health expenditure is regarded as catastrophic when a household's out-of-pocket medical expenses exceed a defined threshold of their resources or their ability to pay (Ataguba 2012). Although there is no consensus on where this threshold is set, most definitions set the threshold within a range of 10 – 40% of total household expenditures (Levin *et al.* 2015). WHO has recommended that health expenditure of 40% or more of a household's income after paying for basic needs may be regarded as catastrophic (WHO 2005 p. 2). Such catastrophic health expenditure may lead to impoverishment, in which, as a result of paying for health, non-poor families are pushed into poverty or already poor families sink deeper into debt (Ngcamphalala & Ataguba 2018). WHO (2010) estimates that each year 150 million people experience financial catastrophe due to direct payments such as user fees, while 100 million are pushed below the poverty line.

Families faced with the burden of out-of-pocket payments typically attempt to cope by using savings, selling assets, borrowing money, withdrawing children from school to save on school fees or reducing expenditure on food and clothing (Dyer *et al.* 2013; Jamison *et al.* 2018; Whitehead *et al.* 2001). These coping strategies are usually not enough to overcome the financial burden on the household (Ngcamphalala & Ataguba 2018). The outcomes, shown in Figure 9, may include impoverishment, inadequate health care and anxiety from bearing financial risk (Jamison *et al.* 2018).

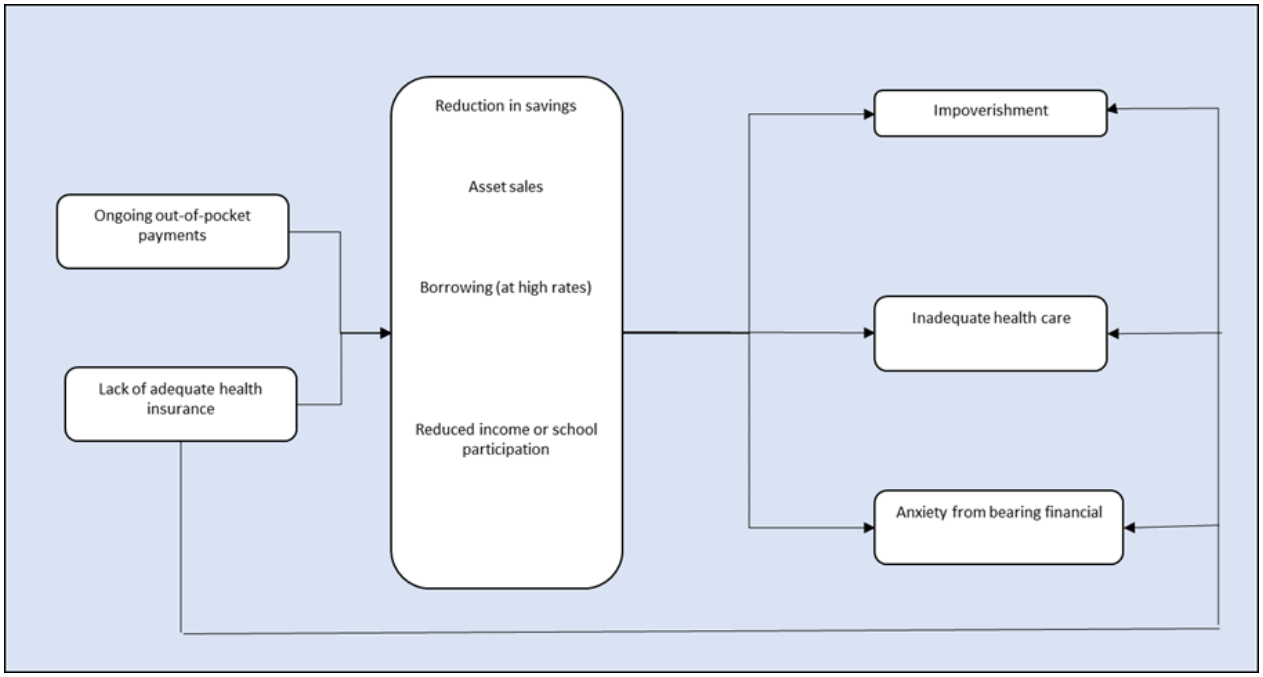

Figure 9. Financial risk protection aspects of health insurance (Jamison *et al.* 2018)

Paying for medical expenses does not necessarily mean that someone can pay (Whitehead *et al.* 2001). Many poor people pay for healthcare despite not being able to afford to do so and suffer financial hardship as a result. Being pushed into debt as a result of paying out-of-pocket expenses has been termed the 'medical poverty trap' (Whitehead *et al.* 2006 p. 10).

Most nations have developed national health policies which are designed to protect their citizens and provide at least some FRP against debilitating healthcare expenses (Verguet *et al.* 2016). A degree of FRP is usually achieved through the mechanism of health insurance. This allows a shift from a system of out-of-pocket payments to one which pools resources and facilitates prepayment mechanisms. In this way, the risk of unexpected medical costs can be mitigated (Skinner *et al.* 2019; Watkins *et al.* 2017). Pooling is the combining of accumulated funds in order to spread the financial costs to all members of the pool, rather than just to those who fall ill. The purpose is to reduce the financial risk of paying for health services. The pooled funds are typically paid, through taxes or health insurance, before the illness occurs (WHO 2010).

Publicly funded health interventions can save households from future expenses. For example, a pneumococcal vaccination programme can reduce the risk of subsequent illness or death from pneumonia which may otherwise have led to financial hardship (Johansson *et al.* 2015; Levin *et al.* 2015). A critical step in improving FRP is the ability to evaluate the potential impacts that health policies may have on out-of-pocket payments. FRP is not accounted for in conventional economic analyses.

2.9.2.1. The ECEA approach to financial risk protection

ECEA is designed to analyse FRP benefits (Verguet *et al.* 2016) whereas DCEA cannot (Dawkins *et al.* 2018). ECEA models the financial protection against catastrophic payments as well as the private expenditures averted from providing a health intervention on the public budget (Verguet *et al.* 2016). The ECEA framework has two specific strengths which make it ideally suited for analysing FRP. First, ECEA can estimate the relative importance of FRP by comparing the amount of out-of-pocket payments relative to a household's disposable income. This approach recognises that direct and indirect costs arising from an illness are likely to be more easily absorbed in a household with sufficient disposable income compared with a family which is living close to the poverty line. Second, ECEA can help decision makers to determine how much FRP to purchase, and to select the best combination of benefits of health and FRP according to their preference. In other words, they can determine how much health and FRP they can purchase and vary their choices to meet their needs.

The study of three immunisation strategies in Ethiopia discussed earlier (Driessen *et al.* 2015) illustrates the analysis of FRP in a clinical study, which analysed 'expenditure averted' according to wealth quintiles (Figure 10).

ECEA studies have used three different approaches to estimate FRP: first, the number of

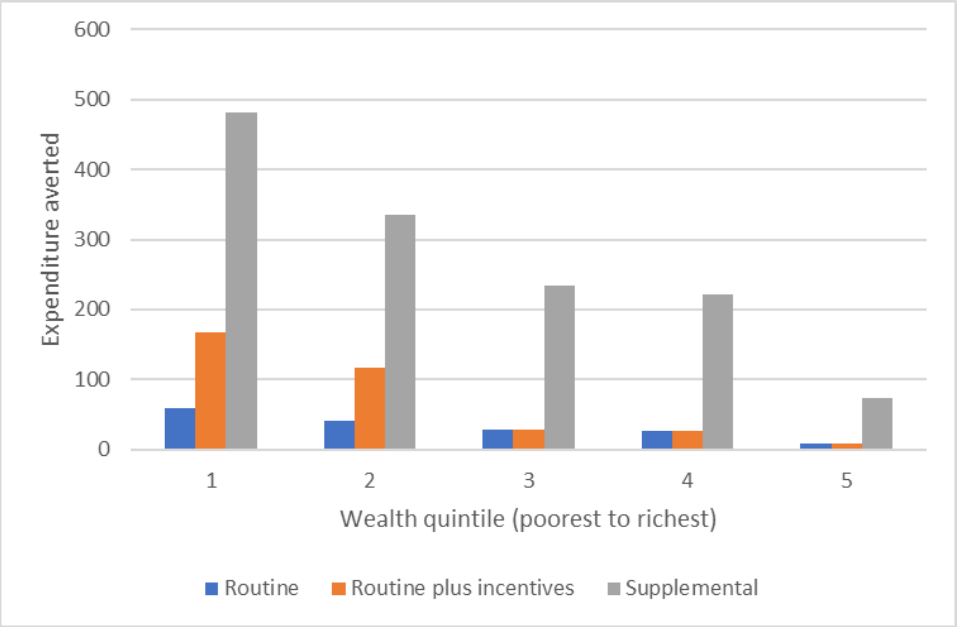

Figure 10. Financial risk protection (expenditure averted in thousands of dollars) according to wealth quintile in an extended cost-effectiveness analysis of three measles immunisation strategies in Ethiopia. Based on data from Driessen *et al.* (2015); Love-Koh *et al.* (2019).

cases of catastrophic health expenditure averted [see, for example, Liu *et al.* (2019); Saxena *et al.* (2019)], second, the number of cases of poverty averted [see, for example, Shrime *et al.* (2016); Watkins *et al.* (2016)], and third, the financial value of insurance against an illness (Verguet *et al.* 2016) [see, for example, Ashok *et al.* (2016); Chisholm *et al.* (2016)]. This measure estimates the amount of money a person would be willing to pay to 'avoid an ailment' (Ashok *et al.* 2016 p. 340).

Health policies may be used to purchase both health benefits, for example, quality-adjusted life years gained and non-health benefits, for example, equity or FRP afforded. While CEA uses the incremental cost-effectiveness ratio (ICER) to quantify health benefits, ECEA takes this a step further by using ICER to quantify FRP. In this way, ECEA can quantify the efficient purchase of both FRP and health. More significantly ECEA can highlight and quantify potential trade-offs among the partially competing aims of improving health, providing FRP and reducing health inequality (Verguet & Jamison 2017).

In countries with universal health insurance where the individuals already have FRP, determining FRP becomes less important. However, even in these situations, ECEA can still provide helpful information on the financial protection offered by specific interventions when compared with others (Verguet *et al.* 2016). Addressing FRP is the unrivalled domain of ECEA. DCEA is not currently able to address FRP (Dawkins *et al.* 2018). This is an important limitation of DCEA particularly in LMICs, where many households are at risk of financial hardship due to out-of-pocket payments for health care (Cookson 2016).

2.9.3. Incorporating health opportunity costs

Because health budgets are finite and limited, expenditure in one programme will reduce available resources in another. Opportunity costs refer to the benefits which are forgone by not selecting the next best alternative. In order to make difficult choices about where best to allocate scarce resources, decision makers need to have a measure of the net health, in other words, the health gain minus the opportunity costs (Cookson 2013). Knowing the net health inequity is necessary because reducing health inequity often incurs additional costs which could have been used to deliver other health gains elsewhere (Cookson 2013).

These opportunity costs do not usually follow the same pattern of costs or benefits of the new intervention. In other words the costs and benefits of the new intervention are not offset in similar proportion by the costs and health distributions of the previous intervention (Asaria *et al.* 2016). An individual in one subgroup who was benefiting from the discontinued intervention may not benefit from the new intervention. For example, a new programme for rotavirus vaccination may take resources away from an existing malaria treatment programme.

While ECEA does not address opportunity costs, DCEA analyses and incorporates health opportunity costs using the health equity impact plane described in section 2.9.1.1 (above). DECA has the advantage of presenting the impact of health opportunity costs in a way which is transparent to the user (Dawkins *et al.* 2018). In so doing, DCEA addresses opportunity cost and accounts for resources used in one programme that could have reduced morbidity or mortality through a different programme. Because ECEA does not account for health opportunity costs of displaced expenditure within the health sector

budget, DCEA is better suited to analysing interventions in health care systems which have a fixed health budget, such as the United Kingdom's National Health Service (NHS).

2.10. Future improvements of DCEA and ECEA

There are three areas where DCEA and ECEA could be improved in the future. The first area is to reduce the requirement for complex data. For example, a DCEA study of rotavirus vaccine Ethiopia, required data on adult mortality which was not available for that country (Dawkins *et al.* 2018). The researchers used an alternative data proxy, in this case available child health data, to substitute for the unavailable data (Dawkins *et al.* 2018; Karamagi & Dovlo 2015). An alternative approach to dealing with this data constraint is a simplified version of DCEA called 'aggregate DCEA' in which the average health gains from a CEA is scaled up in proportion to health utilisation per population subgroup (Love-Koh *et al.* 2019a). The second area for future improvement is the development of a clear method to assist decision-makers to clarify their equity concerns (Asaria *et al.* 2016). The third area for future development is the improvement of the presentation of outcomes to make them accessible to non-specialists (Asaria *et al.* 2016).

3. Conclusions

Determining the impact on health equity is critical in situations where a policy may negatively impact health in already disadvantaged groups (Lal *et al.* 2018). While CEA evaluates health benefits and costs, both DCEA and ECEA expand this capacity and establish how these benefits and costs are distributed across social groups within a population. The frameworks help decision makers understand how a policy may affect health inequity and focus on how best to reduce health inequity for disadvantaged groups. A key strength of ECEA is that it analyses FRP, an important step towards universal health care (Verguet *et al.* 2016), while the key strength of DCEA is that it provides an explicit analysis of trade-offs between reducing health inequity and increasing overall health (Dawkins *et al.* 2018).

4. Abbreviations

CEA	Cost-effectiveness analysis
DCEA	Distributional cost-effectiveness analysis
ECEA	Extended cost-effectiveness analysis
EDE	Equally distributed equivalent level of health
FRP	Financial risk protection
HALY	Health-adjusted life years
ICER	Incremental cost-effectiveness ratio
LMIC	Low- and middle-income countries
NHS	National Health Service
QALY	Quality-adjusted life years
UK	United Kingdom
WHO	World Health Organisation

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Part C: Journal Manuscript

Incorporating health equity concerns into cost-effectiveness analysis: A review of Distributional and Extended Cost-Effectiveness Analysis.

Proposed Journal: Journal of health economics

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Abstract

Background

Equity is rarely included in health economic evaluations, partly because the techniques for addressing equity have been inadequate. Since 2013 health economists have developed two competing health economic technologies: distributional cost-effectiveness analysis (DCEA) and extended cost-effectiveness analysis (ECEA). Both technologies represent a significant advance, and each provides a framework to address equity considerations in cost-effectiveness analysis.

Methods

A scoping literature review was used to identify and synthesise the relevant literature on incorporating equity concerns into economic evaluations. A second focused review identified literature which discussed or applied DCEA and ECEA. Key themes in the literature were identified using NVivo qualitative data analysis software.

Results

The review revealed three key areas of difference between DCEA and ECEA: First, the analysis of trade-offs between improving health and reducing inequity; second, the analysis of financial impacts of health policies; and third, the incorporation of opportunity costs. ECEA can analyse financial risk protection while DCEA can analyse opportunity costs and trade-offs between improving equity and reducing health. ECEA is designed for low- and middle-income countries, whereas DCEA is better suited to developed health systems such as the National Health Service in the United Kingdom. To date, there have been 27 studies using ECEA and five studies using DCEA.

Future developments for DCEA and ECEA include incorporating alternative methods to simplify the data requirements for the techniques, providing methods to assist decision makers to clarify their equity concerns, and improving the presentation of outcomes to make them accessible to non-specialists.

Conclusions

DCEA and ECEA are both economic frameworks which address equity considerations in cost-effectiveness analysis. This study examines and compares these two techniques in

order to assist policymakers and decision makers to determine which of the two methods is best able to address their specific needs for their particular circumstances.

JEL classification

I14: Health and Inequality

Keywords

Health equity; inequality; cost-effectiveness analysis; distributional cost-effectiveness analysis (DCEA); extended cost-effectiveness analysis (ECEA).

1. Background

Current methods for assessing how best to maximise health, such as cost-effectiveness analysis (CEA) and cost-benefit analysis (CBA), are well developed. While these techniques can assist decision makers in making choices on how to allocate health resources most efficiently, they fail at informing healthcare priorities (Brousselle & Lessard 2011; Norheim *et al.* 2014). For example, while CEA can be used to determine whether a tuberculosis treatment program would be more or less cost-effective when compared with a measles immunisation programme, CEA cannot provide information on how either intervention may impact the most deprived 20% of a community. Economic evaluations which specifically evaluate the impact of policies on equity are required to address this type of equity concern.

1.1. Health equity

Although there is no universally agreed definition of equity (Mooney 2003), the World Health Organisation (WHO) has defined equity as:

> 'the absence of avoidable or remediable differences among groups of people, whether those groups are defined socially, economically, demographically, or geographically' (WHO 2019).

Equity differs from equality; health inequality is the systematic difference in the health of groups which occupy 'unequal positions in society' (Graham 2004 p. 101), while health inequity is a specific type of health inequality in which this systematic difference is judged to be avoidable or remediable by reasonable means (Arcaya *et al.* 2015). To illustrate this, the inequality in the incidence of dementia across age groups is not regarded as being

inequitable, although the funding of treatment for dementia compared with that for myocardial infarction may be.

While health is universally valued, access to health care and health outcomes are unfairly distributed across and within countries (Marmot & Bell 2018). Measures of health, such as child mortality rate and life expectancy, typically reflect a pattern of worse health outcomes in more impoverished regions. For example, the risk of a child dying before the age of one is six times higher for a child born in the WHO African region than for a child born in the WHO European region (WHO 2020).

Poverty and ill-health are linked. In all countries, including those which are well-resourced, illness follows an income gradient, where those lowest in the socio-economic hierarchy carry the highest burden of illness. The relationship between poverty and ill-health is causal in both directions: 'poverty breeds ill-health, and ill-health keeps poor people poor' (Wagstaff 2002 p. 97). To correct this unfair distribution, health policies should aim to improve overall health and reduce the inequitable provision of health in the population. However, despite the recognised importance of health equity, equity is not satisfactorily addressed in practice and only rarely included in economic evaluations (Cookson *et al.* 2017).

1.2. Addressing equity in cost-effectiveness analysis

CEA is a quantitative economic analysis which identifies interventions which maximise health benefits and minimise health costs (Ashok *et al.* 2016). The technique relates improvements in health to the cost of an intervention and focuses on the relative costs per unit of health gain (Raykar *et al.* 2016). Health gains are measured in meaningful units such as quality-adjusted life-years (QALY) gained, or premature deaths averted (Musgrove & Fox-Rushby 2006). While CEA has become the standard technique to evaluate medical interventions (Phelps & Madhavan 2019), it is not designed to address health inequity (Cookson 2013; Sassi *et al.* 2001). This is because CEA assigns equal value to each unit of health gain with no regard to the characteristics of the recipients. Maximising health, as measured by maximising the number of QALYs irrespective of who receives those QALYs and how those QALYs are distributed, has been encapsulated in the expression 'a QALY is a QALY'. This approach has been criticised, see, for example, Dolan *et al.* (2005) and Wailoo *et al.* (2009). One such criticism is that CEA cannot incorporate

preferences shared by many people, such as giving priority to severely ill patients or to those who are disabled (Ubel *et al.* 2000).

Until 2013 there were no adequate tools to incorporate equity into economic evaluations. As a consequence, health economists did not know how to address equity, nor did they know how to integrate equity considerations into efficiency analyses (Culyer 2012).

In 2013 and 2015 two promising solutions which enhance CEA to address equity were developed: distributional cost-effectiveness analysis (DCEA) and extended cost-effectiveness analysis (ECEA). In 2013, Verguet, Laxminarayan and Jamison developed a method which they called 'extended cost-effectiveness analysis' (ECEA). They published their methodology in a study of universal public finance for rotavirus vaccination in India and Ethiopia (Verguet *et al.* 2013). In 2015, Asaria, Griffin, Cookson and colleagues at the University of York in the United Kingdom described a comparable technique they termed 'distributional cost-effectiveness analysis' (DCEA). They described the methodology in a study based on a UK National Health Insurance programme for the screening of bowel cancer (Asaria *et al.* 2015).

However, DCEA and ECEA are not yet widely applied or understood. This paper examines and compares these two techniques with the aim of assisting policymakers and decision makers to determine which of the two methods is best able to address their specific needs for their particular circumstances.

2. Methods

2.1. Scoping literature review

A scoping literature review was conducted to determine the range and nature of information on economic evaluations which can address equity issues. A scoping review is defined as:

> 'a form of knowledge synthesis […] aimed at mapping key concepts […] related to a defined area or field by systematically searching, selecting, and synthesising existing knowledge' (Colquhoun et al. 2014 p. 1292).

The scoping review was undertaken in two phases: the first phase was be a broad scoping review using PubMed and Google Scholar which focus on the incorporation of equity

concerns into economic evaluations. The second phase was a focused review of DCEA and ECEA.

2.1.1. Scoping review phase 1

For phase 1, the PubMed database was searched using the as used the search strategy for PubMed as outlined in Table 7. Additional articles were sought using Google Scholar as well as searching for additional reference listed in the retrieved articles.

Table 7. PubMed search strategy for Phase 1.

Search #1	"Costs and Cost Analysis" [MeSH terms]
Search #2	“health equity” [MeSH Terms]
Search #3	(#1) AND (#2)

2.1.2. Scoping review Phase 2

The second phase was a focused review of DCEA and ECEA using the search strategy for PubMed is outlined in Table 8.

Table 8. PubMed search strategy for Phase 2.

Search #1:	"extended cost" [All Fields]
Search #2	"distributional cost" [All Fields]
Search #3:	(#1) OR (#2)

This second phase identified 47 articles which provided information specific to DCEA and ECEA. Of these articles, 27 were ECEA studies, and four were DCEA studies. The remaining 16 articles either discussed the methodology of DCEA or ECEA, or mentioned the two techniques in the broader context of economic evaluations.

A scoping review is the methodology best suited for this study for two reasons: firstly, this study focuses on evaluating and synthesising information on two methods of

economic evaluation, DCEA and ECEA, rather than a review which, for example, examines the outcomes of a medical intervention from a range of similar studies. Secondly, because DCEA and ECEA are relatively newly developed, there is limited published literature available. Unpublished information, for example, two articles on blog sites written by the developers of DCEA and ECEA, provided additional insights on the frameworks (Asaria 2018; Verguet 2017). An essential strength of a scoping review is that it can include both published and unpublished data (Levac *et al.* 2010).

The focussed review addressed three research questions:

1. How do distributional cost-effectiveness analysis (DCEA) and extended cost-effectiveness analysis (ECEA) address equity concerns?

2. In what ways are DCEA and ECEA similar and/or different?

3. What are the relative strengths and limitations of DCEA and ECEA?

2.2. Extraction and analysis

The information from phase 2 of the scoping review was analysed using NVivo data analysis software to identify and synthesise key themes to examine, evaluate and compare DCEA and ECEA. Concepts which were relevant to the three research questions were assigned to one or more topic heading (called 'nodes' in NVivo). The initial nodes used for coding are shown as Table 9.

Table 9. Initial NVivo nodes to address research questions.

NVivo Node	Explanation
01 DCEA Address equity	How DCEA addresses equity concerns
02 ECEA Address equity	How ECEA addresses equity concerns
03 Similar	How DCEA and ECEA are similar
04 Different	How DCEA and ECEA are different
05 DCEA Strength	Strengths of DCEA
06 ECEA Strength	Strengths of ECEA
07 DCEA Limits	Limitations of DCEA
08 ECEA Limits	Limitations of ECEA
09 DCEA Other	Other DCEA topics e.g. future improvements for DCEA
10 ECEA Other	Other ECEA topics e.g. future improvements for ECEA

Concepts relevant to DCEA and ECEA which emerged during the review process, such as financial risk protection (FRP), opportunity costs and equity trade-offs, were added as additional nodes.

2.3. Scope and limitations of the study

This study compares DCEA and ECEA based on the most important similarities and differences. This comparison is not exhaustive, and differences which were considered to be of minor importance were excluded. For example, the similar approach that DCEA and ECEA use to examine health benefits across social groups is discussed, while the minor differences in how DCEA and ECEA estimate these health benefits are excluded.

The comparison is aimed at assisting decision makers who assess healthcare policies to differentiate between the capabilities of DCEA and ECEA. The focus is specifically on which challenges the economic evaluations can address, rather than the steps undertaken in an analysis. For health economists and other experts undertaking DCEA or ECEA, the methodology of the frameworks has been described by Asaria *et al.* (2016) and Verguet *et al.* (2016), respectively.

3. Discussion

DCEA and ECEA are significant advances in health economics. They are robust frameworks which expand cost-effectiveness analysis (CEA) to address equity concerns (Love-Koh *et al.* 2019b). They can be used to explicitly determine which policy or intervention will provide the best reduction in health inequity for the resources invested (Love-Koh *et al.* 2019b). ECEA has been used in more studies and has covered a much broader spectrum than has DCEA. To date, 27 ECEA studies have been published. In contrast, DCEA has been applied in five studies.

ECEA has been used for a variety of health policy assessments across a range of settings and health interventions (Love-Koh *et al.* 2019b). These studies have included evaluating the financial consequences of increasing tobacco tax in Lebanon (Salti *et al.* 2016), examining the effects on health equity and medical impoverishment resulting from motorcycle helmet regulation in Vietnam (Olson *et al.* 2016) and investigating the health and economic implications of three different measles vaccination strategies in Ethiopia (Driessen *et al.* 2015). DCEA studies include a National Health Service bowel cancer

screening programme in England (Asaria *et al.* 2015) and a rotavirus vaccination program in Ethiopia (Dawkins *et al.* 2018).

Although DCEA and ECEA are excellent tools for simultaneously addressing both efficiency and equity, they have been recently developed and are not yet widely known or used in practice (Cookson *et al.* 2017). There are limited resources available to aid decision makers in understanding whether DCEA or ECEA best addresses their particular equity concern. This study provides a guide to differentiate DCEA and ECEA. The similarities and differences between DCEA and ECEA are discussed in greater detail below.

3.1. Similarities of DCEA and ECEA

DCEA and ECEA have both been categorised as 'equity-informative CEA' (Cookson *et al.* 2017), and each builds on CEA to provide an explicit framework for incorporating equity concerns into the standard method of CEA (Asaria *et al.* 2014). Both frameworks aim to identify health policies which reduce inequity and preferentially benefit disadvantaged subgroups. These subgroups are disaggregated based on equity-relevant criteria such as age, income or education level (Love-Koh *et al.* 2019b).

An example of the approach to disaggregating health impacts according to equity-related subgroups, is seen in a DCEA study by Asaria and colleagues (2015). The authors compared two strategies aimed at increasing the uptake of a bowel cancer screening programme in England: a 'universal' strategy in which everyone in the study received a standard reminder, and a 'targeted' strategy which focused on the most deprived social groups. Figure 1 shows the predicted changes in health, in which the targeted strategy reduces inequality but results in less total health when compared with the universal strategy. Although this is a DCEA study, ECEA studies use a similar disaggregated approach.

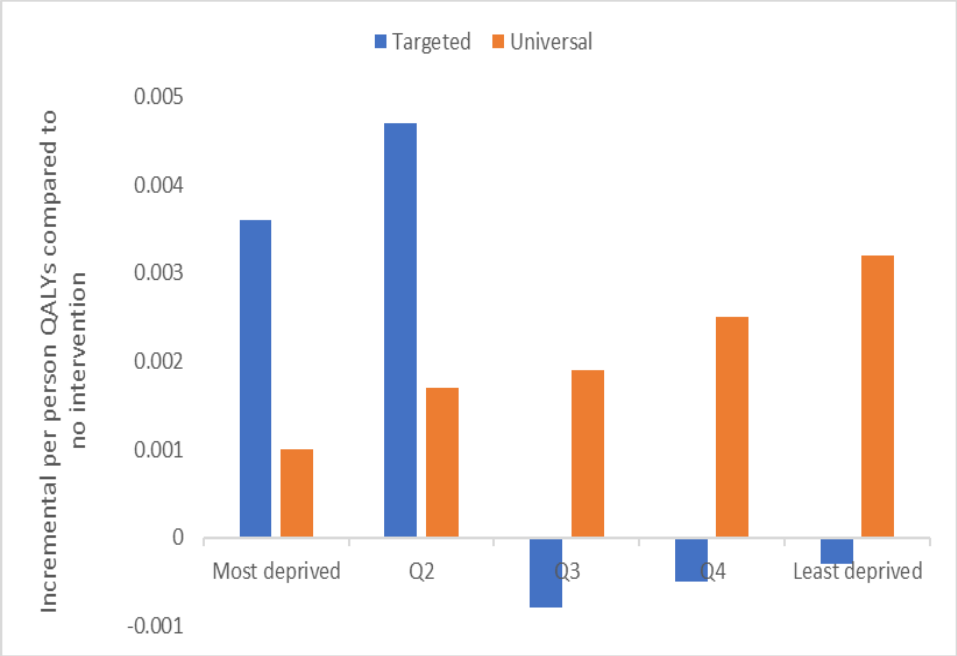

Figure 1. Example of disaggregation according to wealth quintiles in a DCEA study comparing two strategies for increasing the uptake of bowel cancer screening (Asaria *et al.* 2015).

DCEA and ECEA face a common challenge, in that they both require health data which may not be readily available. While the limited availability of health data may be an obstacle for most health equity analyses (Love-Koh *et al.* 2019b), this is particularly so for complex analyses like DCEA and ECEA. For example, in order to assess the change in lifetime health due to a new intervention, the health economist requires data on baseline distribution of morbidity and mortality which may vary by gender, age, socio-economic status and a host of other variables (Dawkins *et al.* 2018). This requirement for data poses a problem, notably in developing countries, where the collection of reliable data within the health system may be prohibitively expensive (Love-Koh *et al.* 2019b).

3.2. Differences between DCEA and ECEA

The significant differences between DCEA and ECEA are best illustrated in three areas:

1. Analysing trade-offs between improving health and reducing inequity.
2. Addressing the financial impacts of health policies.
3. Incorporating health opportunity costs.

These three key areas of difference are discussed in more detail below.

3.2.1. Analysing trade-offs between improving health and reducing inequity

Decision makers often have to balance the conflicting demands of increasing cost-effectiveness and reducing inequity (Round & Paulden 2018). An efficiency-equity trade-off occurs when a more cost-effective intervention may result in a less equitable distribution of health benefits (Musgrove & Fox-Rushby 2006). These trade-offs can be conceptualised using the health equity impact plane (Figure 2), which plots the potential trade-offs between promoting cost-effectiveness and improving equity. Policies under consideration can be plotted according to their health equity impact (on the horizontal axis) and cost-effectiveness (on the vertical axis). The standard is the comparator which is represented at the origin, where the vertical and horizontal axes intersect (Cookson *et al.* 2017).

A policy which falls into Quadrant I should usually be selected ahead of the standard policy. In this case, equity and cost-effectiveness are compatible. This may occur when a

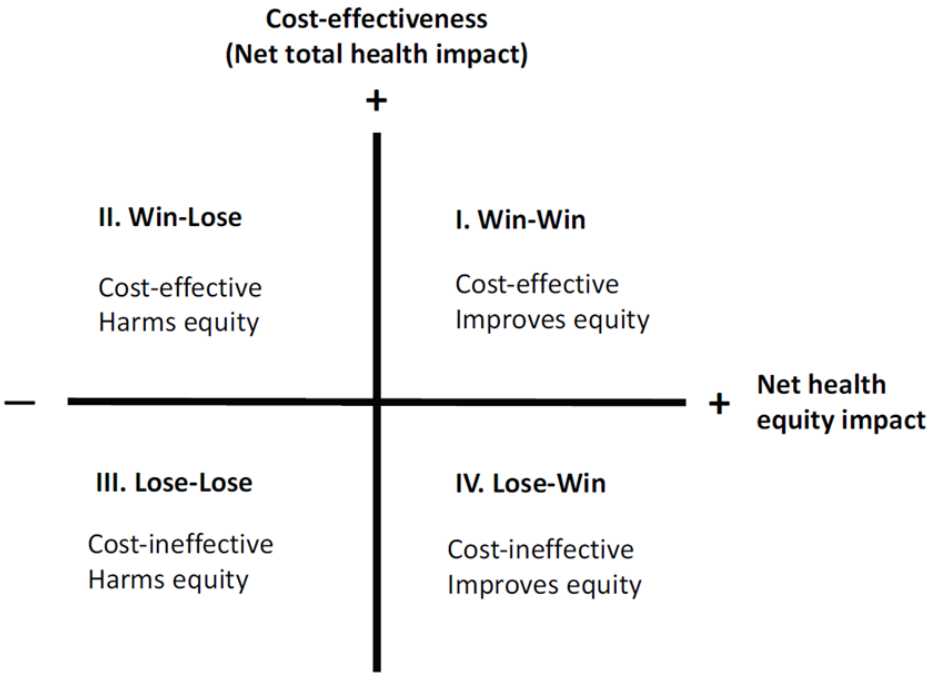

Figure 2 The health equity impact plane (Cookson *et al.* 2017)

cost-effective intervention is targeted at a section of the population that carries the highest burden of disease, for example, the provision of tuberculosis treatment for an impoverished community with high rates of tuberculosis. On the other hand, an

intervention such as a hospital-based renal dialysis programme in a poor country is likely to fall into Quadrant III and should be rejected as it indicates a negative impact on both cost-effectiveness and equity. Outcomes which fall into Quadrant I (accept) and Quadrant III (reject) are uncomplicated and do not require analysis beyond standard CEA (Cookson *et al.* 2017). A proposed policy, which, compared with the standard policy, is more cost-effective but increases inequity (Quadrant II) would be rejected if improving equity was important to decision makers. Where there is a trade-off between increasing health gains and reducing health inequity (Quadrant IV), DCEA can provide an explicit analysis of such trade-offs to assist decision makers in selecting the most suitable intervention (Asaria *et al.* 2016).

The analysis of a trade-off between health and equity concerns is evident in a DCEA study of a rotavirus vaccination program in Ethiopia in which the proposed intervention fell in Quadrant IV (Dawkins *et al.* 2018). The researchers used a range of feasible cost-effectiveness thresholds which is a measure of the opportunity cost and has been defined as the 'amount of money that, if removed from the healthcare system, would result in one less unit of health being generated' (Woods *et al.* 2016 p. 960). They found that, at a cost-effectiveness threshold of $50, a pro-poor rotavirus vaccination programme is likely to promote equity but be less cost-effective compared with the standard program (Quadrant IV). This outcome is shown in the health equity impact plane (Figure 3).

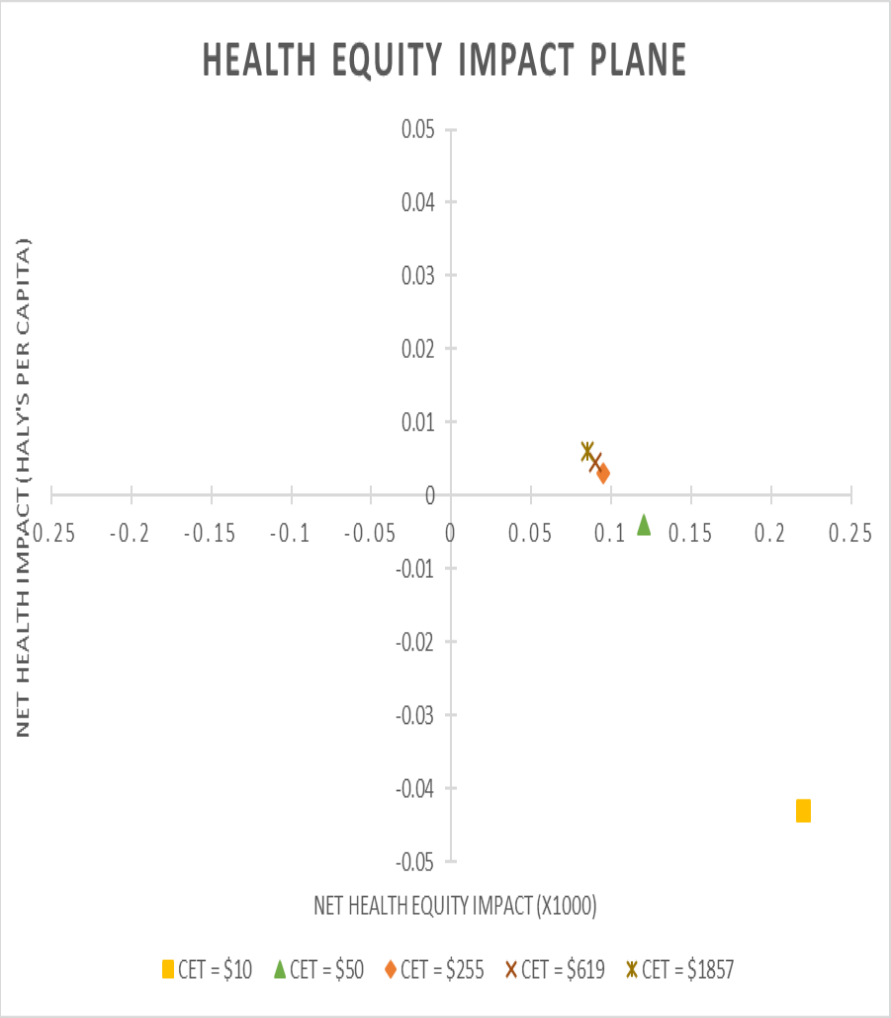

Figure 3. Health equity impact plane for a rotavirus vaccination programme in Ethiopia (Dawkins *et al.* 2018).

To help decision makers resolve the trade-off between equity and health, Dawkins et al. calculated an inequality measure, called the equally distributed equivalent (EDE) level of health, by combining the decision maker's level of aversion to inequality (high, medium, or low) with the distribution of health. The difference between the mean level of health and the EDE reflects the average amount of health per person that a decision maker is willing to sacrifice to achieve full health equity (Asaria *et al.* 2016). This allows decision makers to assess the impact that their selected level of aversion to inequality has on the recommendations of DCEA. ECEA does not analyse trade-offs between health and equity, although it does address the trade-off between health and financial risk protection (FRP), which is discussed in the next section.

3.3. Addressing the financial impacts of health policies

Access to health services should be based on need and not on the ability to pay (WHO 2014). Direct payments for health impose a significant barrier to accessing healthcare and exert a heavy financial burden for millions of households around the world. Financial risk protection (FRP) is a system designed to safeguard people against financial adversity arising from paying directly for health services (Nugent 2014; Saksena *et al.* 2014) and is a critical step towards achieving universal health coverage (WHO 2016). This safeguard is particularly valuable in low- and middle-income countries (Love-Koh *et al.* 2019b) where financial safety nets in the form of unemployment benefits or disability payments are typically not available.

While FRP is not accounted for in CEA or DCEA (Dawkins *et al.* 2018), ECEA is designed to analyse FRP benefits (Verguet *et al.* 2016). ECEA models financial protection against catastrophic payments as well as the 'private expenditures averted' when a health intervention is paid for using public funds (Verguet *et al.* 2016). ECEA can guide decision makers in three areas: First, the framework can help decision makers to determine how much FRP to purchase and to select their preferred combination of health benefits and FRP. In other words, they can determine how much health and FRP they can purchase and vary their choices to meet their needs. Second, ECEA can estimate the relative importance of FRP by comparing the amount of out-of-pocket payments relative to a household's disposable income. This approach recognises that costs arising from an illness are likely to be more easily absorbed in a household with sufficient disposable income compared with a family which is living close to the poverty line. Third, ECEA can highlight and quantify potential trade-offs among the partially competing aims of improving health and providing FRP (Verguet & Jamison 2017).

ECEA helps compare the FRP provided by several potential policy options. For example, Verguet and colleagues (2015) used ECEA to assess health gains (measured as deaths averted) and FRP provided (measured as cases of poverty averted) for nine public health interventions in Ethiopia. They found that measles immunisation and caesarean section surgery averted the most deaths, while caesarean section surgery and tuberculosis treatment averted the most cases of poverty. This relationship is depicted in a simplified graph of some of their outcomes (Figure 4).

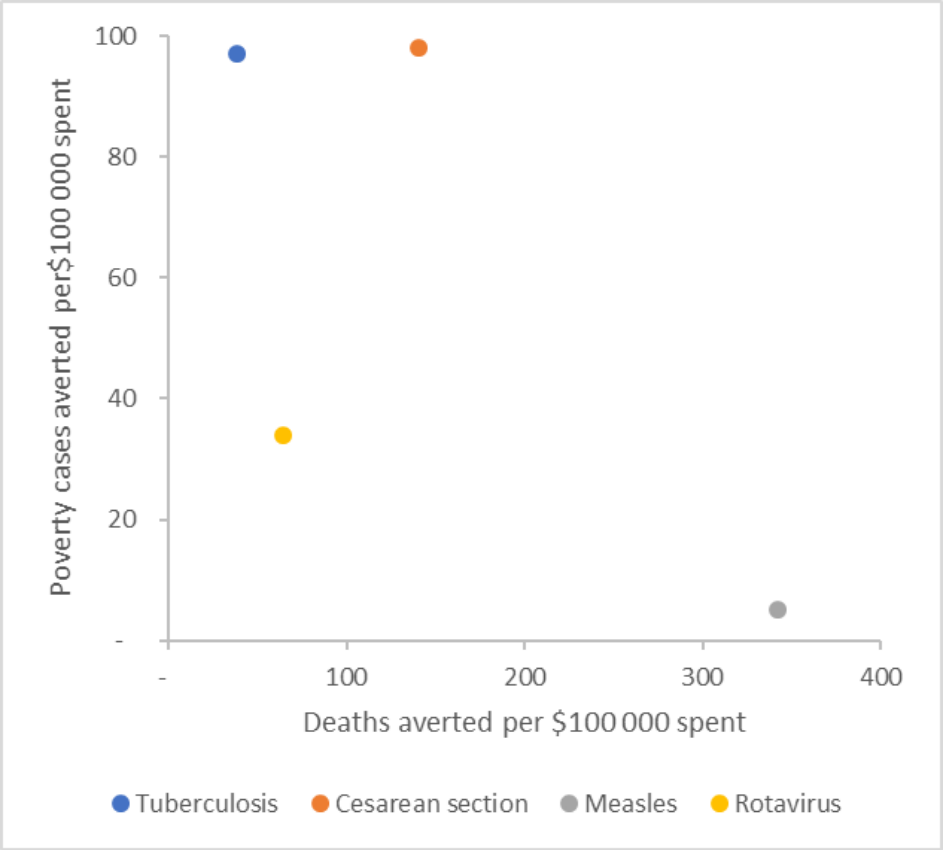

Figure 4. Illustration of financial risk protection versus health gains in an ECEA study of universal public finance in Ethiopia (Verguet *et al.* 2015b).

In countries with universal health insurance, where the individuals already have FRP, determining FRP becomes less critical. However, even in these situations, ECEA can provide helpful information on the levels of financial protection offered by competing policy options (Verguet *et al.* 2016).

In summary, ECEA can quantify the efficient purchase of both FRP and health benefits. More significantly, ECEA can highlight and quantify potential trade-offs among the partially competing aims of improving health, providing FRP and reducing health inequity (Verguet 2017). Addressing FRP is the unrivalled domain of ECEA. DCEA is not currently able to address FRP (Dawkins *et al.* 2018). This is an essential limitation of DCEA, particularly in LMICs, where many households are at risk of financial hardship due to out-of-pocket payments for health care (Cookson 2016).

3.4. Incorporating health opportunity costs.

Because health budgets are finite and limited, expenditure in one programme will reduce available resources in another. Opportunity costs refer to the benefits which are forgone by not selecting the next best alternative. In order to make difficult choices about where best to allocate scarce resources, decision makers need to have a measure of the net health, in other words, the health gain minus the opportunity costs (Cookson 2013). Knowing the net health benefit is necessary because reducing health inequity often incurs additional costs which could have been used to deliver health gains elsewhere (Cookson 2013).

These opportunity costs do not usually follow the same pattern of costs or benefits of the new intervention. In other words, the costs and benefits of the new intervention are not offset in similar proportion by the costs and health distributions of the previous intervention (Asaria *et al.* 2016). An individual in one subgroup who was benefiting from the discontinued intervention may not benefit from the new intervention. For example, a new programme for rotavirus vaccination may take resources away from an existing malaria treatment programme.

DCEA analyses and incorporates health opportunity costs in a way which is transparent to the user (Dawkins *et al.* 2018). In contrast, ECEA does not address health opportunity costs (Dawkins *et al.* 2018) and assumes that an intervention is funded through the tax system (Asaria *et al.* 2015). This represents a significant weakness in ECEA because, as discussed, in order to assess the health benefits of a new programme accurately decision makers need to understand the health benefits, such as lives saved, that may have accrued if the money had been spent on an alternative health intervention (Love-Koh *et al.* 2019b). In addition, the lack of analysis of opportunity costs means that ECEA is less well suited to analysing interventions in health care systems, like the National Health Service (NHS) in the United Kingdom, which have a fixed health budget.

Figure 6 summarises the key similarities and differences between DCEA and ECEA.

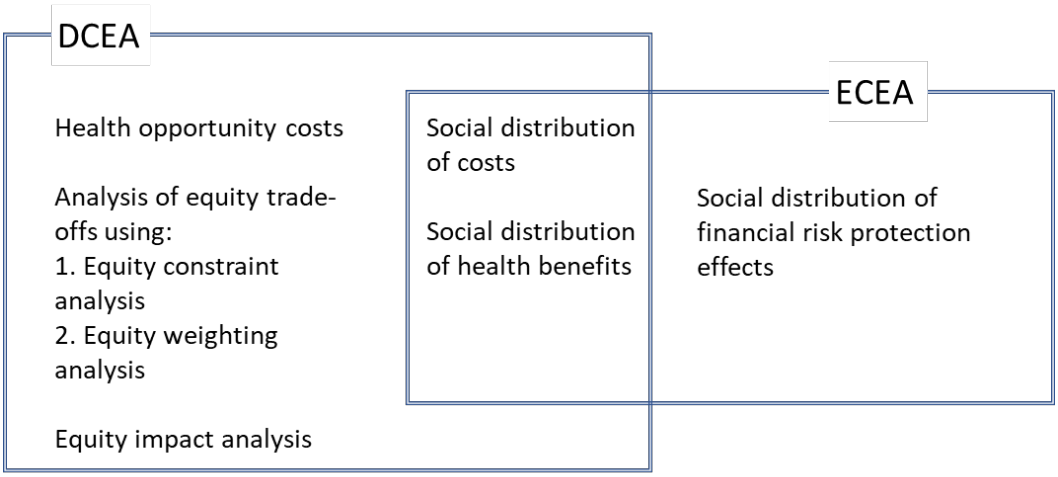

Figure 5. The key similarities and differences between DCEA and ECEA

3.5. Future improvements in DCEA and ECEA

There are three areas where DCEA and ECEA could be improved in the future. The first area relates to the need for complex health data, which, as mentioned, is a feature of sophisticated economic analyses, particularly DCEA and ECEA. One proposed method is to use alternative 'data proxies' to substitute for unavailable data (Karamagi & Dovlo 2015). An alternative approach to dealing with this data constraint is a simplified version of DCEA called 'aggregate DCEA' in which the average health gains from a CEA is scaled up in proportion to health utilisation per population subgroup (Love-Koh *et al.* 2019a). Methods which reduce the need for complex health would be of particular benefit in DCEA which typically requires more complex data than does ECEA. The second area for future improvement is the development of a guide to assist decision-makers to clarify their equity concerns (Asaria *et al.* 2016). Clarifying equity concerns would help to guide decision makers and researchers in selecting between DCEA and ECEA. For example, a focus on assessing health opportunity costs or analysis or equity trade-offs would favour the use of DCEA, whereas concerns for financial risk protection would favour using ECEA. The third area for future development is the improvement of the presentation of outcomes to make them accessible to non-specialists (Asaria *et al.* 2016). Good examples of clear and understandable presentation of outcomes are illustrated in the bar chart in DCEA study comparing two strategies for increasing the uptake of bowel cancer

screening (Figure 1, above) and in the illustration of financial risk protection versus health gains in an ECEA study of universal public finance in Ethiopia (Figure 5, above).

4. Conclusions

Determining the impact on health equity is critical in situations where a policy may further disadvantage already vulnerable groups (Lal *et al.* 2018). While CEA evaluates health benefits and costs, both DCEA and ECEA expand this capacity and establish how these benefits and costs are distributed across social groups within a population. The frameworks help decision makers understand how a policy may affect health inequity and focus on how best to improve health provision for disadvantaged groups. A key strength of ECEA is that it analyses FRP, an essential step towards universal health care (Verguet *et al.* 2016), while a key strength of DCEA is that it provides a precise analysis of trade-offs between reducing health inequity and increasing overall health (Dawkins *et al.* 2018).

Author's contribution

Competing interests

No conflicts of interests.

Declarations

The author funded publication of this article in his personal capacity.

5. Abbreviations

CEA	Cost-effectiveness analysis
DCEA	Distributional cost-effectiveness analysis
ECEA	Extended cost-effectiveness analysis
EDE	Equally distributed equivalent (level of health)
FRP	Financial risk protection
HALY	Health-adjusted life year
LMIC	Low- and middle-income countries
NHS	National Health Service
QALY	Quality-adjusted life year

UK	United Kingdom
WHO	World Health Organisation

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Part D: Policy Briefing

Addressing health equity in economic evaluations

Learners and teachers circle up in Zithulele, Eastern Cape. Photo courtesy of Storm Lewis.

Policy Brief

Addressing Health Equity in Economic Evaluations

Addressing health equity in economic evaluations

Health equity

Health is universally valued and almost all nations acknowledge the need for fairness in the provision of health care. For this reason, health interventions should aim to improve both overall health as well as the fair distribution of health in a population (WHO 2014). The goal is to move towards health equity in which there is no 'avoidable or differences among groups of people, whether those groups are defined socially, economically, demographically, or geographically'(WHO 2019)

Key Points

Health equity is the absence of avoidable or remediable differences among groups of people.

Two newly described techniques, DCEA and ECEA, can analyse the health equity impact of new policies.

These techniques differ:

- DCEA addresses opportunity costs, and the trade-off between cost-effectiveness and equity.
- ECEA assesses financial risk protection.

Cost-effectiveness analysis (CEA)

The standard technique to evaluate medical interventions is cost-effectiveness analysis (CEA). This is a quantitative economic analysis which identifies interventions which maximise health benefits and minimise health costs. However, CEA is not designed to address health inequity. This is because CEA assigns equal value to each unit of health gain with no regard to the characteristics of the recipients.

DCEA and ECEA

Two techniques have recently been developed which overcome the limitations of CEA and can address equity concerns. These techniques are 'distributional cost effectiveness analysis' (DCEA) and 'extended cost-effectiveness analysis (ECEA).

DCEA and ECEA are robust are frameworks which are significant advances in health economics. However, although DCEA and ECEA are excellent tools for simultaneously addressing both efficiency and equity, they were developed recently and are not yet widely known or used in practice.

DCEA and ECEA differ in 3 key areas:

1. The analysis of trade-offs between improving health and reducing inequity,
2. The analysis of financial risk protection, and,
3. The analysis of opportunity costs.

These are differences are discussed in more detail below.

1. Trade-offs between improving health and reducing inequity

Decision makers often have to balance the conflicting demands of increasing cost-effectiveness and reducing inequity (Round & Paulden 2018). An efficiency-equity trade-off occurs when a more cost-effective intervention may also result in a less equitable distribution of health benefits (Musgrove & Fox-Rushby 2006). These trade-offs can be conceptualised using the health equity impact plane (Figure 1), which plots the potential trade-offs between promoting cost-effectiveness and improving equity. Policies under consideration can be plotted according to their health equity impact.

A policy which falls into the top-right (green) quadrant should usually be selected ahead of the standard policy. In this case, equity and cost-effectiveness are compatible. This may occur when a cost-effective intervention is targeted at a section of the population that carries the highest burden of disease, for example, the provision of tuberculosis treatment to an impoverished community with high rates of tuberculosis. On the other hand, an intervention such as a hospital-based renal dialysis programme is likely to fall into the bottom-left (red) quadrant should be rejected as it indicates a negative impact on both cost-effectiveness and equity.

Outcomes which fall into the green quadrant (accept) and the red quadrant (reject) are uncomplicated and do not require analysis beyond standard CEA. A proposed policy, which, compared with the standard policy, is more cost-effective but increases inequity (top-left, amber quadrant) would be rejected where improving equity is important to decision makers. Where there is a trade-off between increasing health gains and reducing health

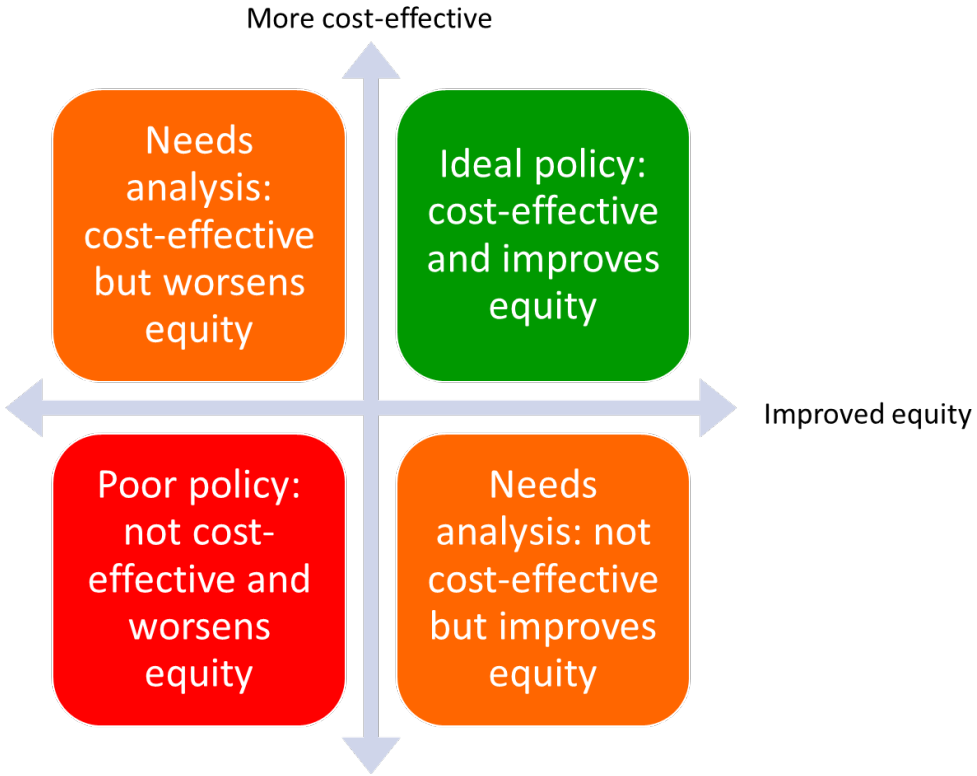

Figure 1 The health equity impact plane

inequity (bottom-right amber quadrant), DCEA and can provide an explicit analysis of such trade-offs to assist decision makers in selecting the most suitable intervention(Asaria *et al.* 2016). In contrast, while ECEA does address trade-offs between financial risk protection and total health, it does not guide decision makers on how to resolve these trade-offs (Dawkins *et al.* 2018).

2. Addressing the financial impacts of health policies

Access to health services should be based on need and not on the ability to pay (World Health Organization 2014). Direct payments for health impose a significant barrier to accessing healthcare and exerts a heavy financial burden for millions of households around the world. Financial risk protection is a system designed to safeguard people against financial adversity arising from paying directly for health services (Nugent 2014; Saksena *et al.* 2014) and is a critical step towards achieving universal health coverage (World Health Organization 2016). While financial risk protection is not accounted for in CEA or DCEA (Dawkins *et al.* 2018), ECEA is designed to analyse FRP benefits (Verguet *et al.* 2016).

3. Incorporating health opportunity costs.

Because health budgets are finite and limited, expenditure in one programme will reduce available resources in another. Opportunity costs refer to the benefits which are forgone by not selecting the next best alternative. In order to make difficult choices about where best to allocate scarce resources, decision makers need to have a measure of the net health, in other words, the health gain minus the opportunity costs (Cookson 2013). Knowing the net health benefit is necessary because reducing health inequity often incurs additional costs which could have been used to deliver health gains elsewhere (Cookson 2013).

DCEA analyses and incorporates health opportunity costs in a way which is transparent to the user (Dawkins *et al.* 2018). In contrast, ECEA does not address health opportunity costs (Dawkins *et al.* 2018) and assumes that an intervention is funded through the tax system (Asaria *et al.* 2015).

Future improvements in DCEA and ECEA

There are three areas where DCEA and ECEA could be improved in the future. The first area

relates to the need for complex health data, which is a feature of sophisticated economic analyses, particularly DCEA and ECEA. One potential solution to this challenge alternative approach to dealing with this data constraint is a simplified version of DCEA called 'aggregate DCEA' in which the average health gains from a CEA is scaled up in proportion to health utilisation per population subgroup (Love-Koh *et al.* 2019). The second area for future improvement is the development of a guide to assist decision-makers to clarify their equity concerns (Asaria *et al.* 2016). The third area for future development is the improvement of the presentation of outcomes to make them accessible to non-specialists (Asaria *et al.* 2016).

Conclusion

Although the provision of health equity is an essential step towards universal health care, equity is often neglected in economic evaluations.

CEA is not designed to incorporate of equity concerns. Two new techniques, DCEA and ECEA, have been developed to specifically address health equity.

Selection of DCEA or ECEA depends on the which equity concern is the main priority. This is because DCEA and ECEA differ in three important areas: While DCEA analyses trade-offs between improving health and reducing inequity, as well as addressing opportunity costs, ECEA analyses financial risk protection.

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Part E: Appendices

Journal of Health Economics: Author information pack

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