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DISTRIBUTIONAL COST-EFFECTIVENESS ANALYSIS OF HEALTH CARE PROGRAMMES – A METHODOLOGICAL CASE STUDY OF THE UK BOWEL CANCER SCREENING PROGRAMME

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ABSTRACT

This paper presents an application of a new methodological framework for undertaking distributional cost-effectiveness analysis to combine the objectives of maximising health and minimising unfair variation in health when evaluating population health interventions. The National Health Service bowel cancer screening programme introduced in 2006 is expected to improve population health on average and to worsen population health inequalities associated with deprivation and ethnicity – a classic case of ‘intervention-generated inequality’. We demonstrate the distributional cost-effectiveness analysis framework by examining two redesign options for the bowel cancer screening programme: (i) the introduction of an enhanced targeted reminder aimed at increasing screening uptake in deprived and ethnically diverse neighbourhoods and (ii) the introduction of a basic universal reminder aimed at increasing screening uptake across the whole population. Our analysis indicates that the universal reminder is the strategy that maximises population health, while the targeted reminder is the screening strategy that minimises unfair variation in health. The framework is used to demonstrate how these two objectives can be traded off against each other, and how alternative social value judgements influence the assessment of which strategy is best, including judgements about which dimensions of health variation are considered unfair and judgements about societal levels of inequality aversion. © 2014 The Authors. *Health Economics* published by John Wiley & Sons Ltd.

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KEY WORDS: health inequality; equity; cost-effectiveness analysis

1. INTRODUCTION

Cost-effectiveness analysis (CEA) is used to support health sector decisions about the allocation of limited resources with the objective of maximising health (Drummond *et al.*, 2005). When dealing with population health interventions, we often have the additional objective of minimising ‘unfair’ health inequality (Cookson *et al.*, 2009) and to this end are also interested in the social distribution of both health gains and health opportunity costs due to the intervention. In this paper, we propose a methodology for quantifying and combining these two objectives within an economic evaluation framework that highlights the social value judgements underpinning any particular conclusion. This ‘distributional CEA’ (DCEA) is demonstrated through a case study comparing potential redesign options to increase uptake of the National Health Service (NHS) bowel cancer screening programme (BCSP) in England.

Colorectal cancer (CRC) is the third most common cancer in the UK with approximately 40 000 new cases diagnosed annually resulting in almost 16 000 CRC-related deaths per year (ONS, 2012). Research has shown that using screening to diagnose and treat CRC earlier can significantly reduce the number of CRC deaths (Hewitson *et al.*, 2008). The Department of Health launched the BCSP in 2006 and currently offers biennial

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screening with guaiac fecal occult blood test (gFOBT) to persons aged 60–74 years. Variable uptake of screening has been observed among the first 2.6 million invitees to the national BCSP, with overall uptake averaging only 54% ranging from 61% in the least deprived areas to 35% in the most deprived areas, and showing a similar gradient in terms of small area-based ethnic diversity measures (Logan *et al.*, 2012; Von Wagner *et al.*, 2011). Furthermore, for those individuals with positive screening results, there is also evidence of inequality in the uptake of follow-up colonoscopy (Morris *et al.*, 2012). It is reasonable to expect that these inequalities in the uptake of the screening programme will exacerbate the already unequal distribution of health in the population, with the screening programme disproportionately benefiting more advantaged groups (for whom uptake is highest) – a classic case of ‘intervention-generated inequality’ (Lorenc *et al.*, 2012).

Prior to the introduction of the BCSP, a number of possible screening options were evaluated to help NHS decision makers determine whether a screening programme was worthwhile and if so the form that it should take. To that end, a model was developed to assess the total resource implications and health impacts of screening by simulating the natural history of CRC and the impact of screening on that natural history (Tappenden *et al.*, 2007). This model was later refined and updated to reflect data emerging from the BCSP (Whyte *et al.*, 2012; Whyte *et al.*, 2011). In this paper, we build on the latest version of this economic evaluation model and use it to estimate the distribution of health associated with alternative screening strategies. We then compare these health distributions using our DCEA framework to determine the strategy that best addresses the dual objectives of maximising health and minimising health inequality. The focus of our analysis is on the quantity and distribution of health gains related to increased participation in the screening programme; other potentially competing objectives of screening programmes such as the promotion of informed choice are not addressed in this paper.

2. METHODS

2.1. Cost-effectiveness analysis

While recognising that in reality there are an almost infinite number of screening strategies that could be designed, in order to demonstrate the framework, we simplify the comparison by considering four mutually exclusive options in our analysis:

- ‘No screening’.
- ‘Standard screening’ as implemented in the BCSP in 2006.
- ‘Targeted reminder’: screening plus a targeted enhanced reminder letter (personal GP-signed letter and tailored information package) sent only to those living in the most income deprived 40% of small areas (index of multiple deprivation (IMD) 4 and IMD5) and to those living in areas with the highest proportion of inhabitants from the Indian subcontinent (IS5). This targeted subgroup comprises of approximately half of the total population invited for screening. The costs of this strategy per person targeted are estimated to be £7 resulting in an estimated increase in average uptake of gFOBT among the targeted population of 12%.
- ‘Universal reminder’: screening plus a universal basic reminder letter (sending a GP-endorsed reminder letter to all eligible patients). The costs of this strategy per person are estimated to be £3.50 resulting in an estimated increase in average uptake of gFOBT of 6%.

We characterise the alternative reminder strategies in such a way as to ensure that both have approximately equal additional intervention costs and equal impact on the total screening uptake, while having very different distributional impacts. Although these reminder strategies are somewhat stylised constructed to highlight the trade-offs between health improvement and health inequality, the potential costs and increases in uptake because of the strategies are estimates based on studies of similar interventions (Shankaran *et al.*, 2007; Hewitson *et al.*, 2011).

The economic evaluation model follows a cohort of 1 million 30 year olds through their lifetimes (allowing it to simulate the adenoma–carcinoma sequence) with screening invitations being sent out biennially to individuals between the ages of 60 and 74 years. The model is run probabilistically to incorporate the uncertainty around the input parameters.

2.2. Inequality analysis

The CEA allows us to identify which of the strategies maximises total health. In order to extend this analysis to allow us to evaluate our other key objective, that of minimising unfair health inequality, we require descriptions of the estimated distributions of health produced by the interventions being compared. To produce these estimates, we condition the model input parameters on factors associated with inequalities in health and inequalities in the effect of screening. We then perform subgroup analyses according to these factors in order to estimate differential cost and health impacts. The health impact per person within each subgroup is scaled by the size of the subgroup in order to describe the total population distribution of health.

The distribution of changes in health attributed to an intervention are informed not only by the distribution of the health gains among recipients of the intervention but also by the distribution of health opportunity costs among those who would have received the displaced activities that the money spent on this intervention would otherwise have been spent on. These opportunity costs are unlikely to fall in proportion to the intervention costs or benefits for particular recipients, and those who would otherwise have benefited from the displaced activities may also include nonrecipients of the intervention.

2.2.1. Estimating a baseline population health distribution. We estimate baseline inequality in the population distribution of expected lifetime health by extending the economic model to incorporate differential all-cause mortality rates by level of socioeconomic deprivation in addition to age and gender. As estimates are based on the Office for National Statistics longitudinal study data (ONS, 2007), we map social class groupings to deprivation measures. We additionally include the differences in morbidity by using health-related quality of life data by age and gender based on UK norms for EQ-5D (Kind *et al.*, 1999) and further adjust for deprivation using the differences between life expectancy and disability-free life expectancy as observed in the Office for National Statistics general lifestyle survey (Smith *et al.*, 2010). Using this data in the model, we estimate a baseline population health distribution in terms of quality-adjusted life expectancy (QALE) at birth. As the ethnic diversity measure that we use in this study is not a variable that is routinely included in the administrative datasets used to estimate baseline QALE, we assume a baseline QALE adjusted for gender and deprivation that does not further vary by ethnic diversity.

2.2.2. Estimating the distribution of uptake of the bowel cancer screening programme. Analysis of the pilot study of the BCSP suggests that screening uptake (the proportion of those invited to screening who participate) varies by area level deprivation, area level ethnic diversity and gender (Weller, 2009). Area level deprivation is based on quintile groups of the IMD 2004, and ethnic diversity is derived from area-based quintiles measuring the proportion of people originating from IS. Significant differences in uptake are observed in the data between all IMD quintile groups and between the most ethnically diverse quintile group (IS5) and the four least ethnically diverse quintile groups (IS1-4). Area level variables are based on data at lower super output area level; these are small areas containing approximately 1500 individuals. Multivariate analysis of the pilot study results provides the independent effect of each characteristic on uptake (Weller, 2009), allowing us to calculate the average uptake of gFOBT and follow-up colonoscopy for each of our 20 distinct subgroups, comprising all possible combinations of the two genders, five deprivation levels and two ethnic diversity levels. We are unable to estimate the proportion of the population in each of the 20 groups from this data as correlation between characteristics was not reported. Therefore, for the base case analysis, we simply assume independence in the distribution of the characteristics. Data from the pilot are used to extrapolate to the population at large by further assuming that the population in the pilot study is representative of the population in general.

2.2.3. Estimating the distribution of opportunity cost. Additional costs of screening and related downstream diagnostic and treatment costs come out of a fixed health budget, and the health opportunity cost due to the disinvestment of these funds from other uses within NHS is assumed to be one quality-adjusted life year (QALY) per £20 000, in line with current practice in NHS. Owing to the absence of further information on how these opportunity costs are distributed, in the base case analysis, we assume that they are distributed equally across all population subgroups. We then perform sensitivity analyses by exploring two extreme assumptions around the distribution of opportunity cost: first, where the entire opportunity cost is borne by the healthiest of our 20 subgroups (women living in the least deprived and ethnically diverse areas), and second, where the entire opportunity cost is borne by the least healthy of our 20 subgroups (men living in the most deprived and ethnically diverse areas).

2.2.4. Assuming all other factors equally distributed. We are able to estimate a modelled distribution of health net of opportunity costs by incorporating the three sets of adjustments to the model that we have described previously, namely, the following: (i) the distribution of factors impacting baseline health; (ii) the distribution of factors impacting screening uptake; and (iii) the distribution of opportunity cost. In so doing, however, we assume that all other factors in the model remain constant between the different subgroups of interest. In particular, a key assumption is that CRC incidence and severity levels are equal across the relevant subgroups. This assumption was made because of data limitations and is supported by limited evidence suggesting that variation in CRC incidence by social class is small (National Cancer Intelligence Network, 2004).¹

2.2.5. Measuring inequality in the resulting health distributions. Inequality in health distributions can be quantified in a variety of ways, and we present a battery of measures in order to be able to inform different inequality concerns from different stakeholders. We start with relative measures of inequality – those that measure the proportional changes in health across the distribution. These range from simple measures focusing only on the extremes of the distribution, such as the relative gap index, to more sophisticated measures assessing the entire distribution and allowing for different levels of relative inequality aversion. An example of the latter is the Atkinson index, shown in the succeeding texts for a population of n individuals with h_i representing the health of individual i , \bar{h} representing mean health in the population and ε representing the level of constant relative inequality aversion (Atkinson, 1970).

$$A_\varepsilon = 1 - \left[\frac{1}{n} \sum_{i=1}^n \left[\frac{h_i}{\bar{h}} \right]^{1-\varepsilon} \right]^{\frac{1}{1-\varepsilon}}$$

We also look at absolute measures of inequality – those that measure the absolute changes in health across the distribution. These also range from simple extreme group measures, such as the absolute gap index, to more sophisticated measures assessing the entire distribution and allowing for different levels of absolute inequality aversion. An example of the latter is the Kolm index shown in the succeeding texts with α representing the level of constant absolute inequality aversion (Kolm, 1976).

$$K_\alpha = \left(\frac{1}{\alpha} \right) \log \left(\frac{1}{n} \sum_{i=1}^n e^{\alpha[\bar{h}-h_i]} \right)$$

2.3. Social welfare analysis

Having separately quantified average population health and the level of health inequality resulting from each of our four screening strategies, we next combine concerns for maximising population health and concerns for

¹Note that the stage of detection will be on average later in those groups with lower uptake, and so modelled cancer-related mortality does differ between subgroups.

minimising health inequality using social welfare analysis. We first check for distributional dominance in a very general sense using the idea of generalised Lorenz dominance (Shorrocks, 1983) to compare the estimated health distributions and eliminate dominated strategies. Health distribution A can be said to generalised Lorenz dominate health distribution B if the Lorenz curve for health in A multiplied by the mean level of health in A dominates the Lorenz curve for health in B multiplied by the mean level of health in B. To compare the remaining, nondominated strategies, we turn to more restricted social welfare indices that explicitly trade-off increases in the mean health against greater equality in the distribution of health (Wagstaff, 2002). These indices are calibrated on the same scale by calculating an ‘equally distributed equivalent’ (EDE) level of health for the health distribution: the level of health each person in the population would receive in a hypothetically perfectly equal health distribution such that society would be indifferent between that equal distribution of health and the actual unequal distribution of health. We focus on two such social welfare indices constructed by combining the mean level of health with the Atkinson and Kolm inequality indices, respectively.

$$h_{ede} = (1 - A_\epsilon)\bar{h}$$

$$h_{ede} = \bar{h} - K_\alpha$$

In the case of no concern for inequality ($\alpha = \epsilon = 0$), the social welfare indices just collapse to the mean level of health. The difference between mean health and EDE health for a given level of inequality aversion indicates the average decrement in health per person society is willing to sacrifice in order to achieve a perfectly equal distribution of health conditional on the level of inequality in the current health distribution. Calculating and comparing the EDEs for the predicted health distributions allows us to rank these strategies over a range of possible inequality aversion levels.

2.4. Adjustment for alternative social value judgements

If inequality concern does not apply to all sources of variation in health – for example, if some determinants of individual ill health are deemed to be a matter of unavoidable bad luck or individual responsibility – then further analysis is required in order to isolate just the variation in health deemed to be unfair.

We can isolate this health distribution of interest by undertaking multivariate analysis on our raw health distribution, to control for ‘fair’ variation in health in order to leave a distribution of health reflecting only the ‘unfair’ variation. The adjustment process we use here has been referred to as ‘direct unfairness’ in the literature (Fleurbaey and Schokkaert, 2009). This ‘fairness-adjusted’ distribution of health is then evaluated in place of the unadjusted distribution, using the same inequality and social welfare index approaches. Alternative judgements about which variation in health is considered fair or unfair can lead to different conclusions as to which intervention strategy is preferred, and so the sensitivity of the decision to alternative sets of reasonable social value judgements regarding fairness should be assessed. In the current case study, the social variables of interest are gender, area-based level of deprivation and area-based ethnic diversity. There are eight possible permutations of social value judgements we can make on whether or not each of these three social variables represents a fair or unfair source of variation in health, ranging from all three being deemed unfair to all three being deemed fair (resulting in the trivial case where there is no variation in health in the adjusted distribution).

For our base case analysis, we characterise all variation in health as unfair. We then check the sensitivity of the ranking of strategies to each of the other seven possible social value judgements that can be made in this example. To apply these alternative social value judgements, we adjust our health distribution to only reflect unfair variation in health by using reference values for the fair variables while preserving the actual values for the unfair variables.

In cases where dominance rules, such as generalised Lorenz dominance, do not provide a complete ordering of strategies, additional social value judgements are required to assess trade-offs between improving total population health and reducing unfair health inequality. The key additional social value judgements that need to be made relate to the choice of inequality measures underpinning social welfare and the level of inequality

aversion. We calculate our results for relative (Atkinson) and absolute (Kolm) inequality indices at both high and low levels of inequality aversion and check the sensitivity of our decision across a range of inequality aversion levels in order to identify the thresholds at which each strategy would be preferred.

3. RESULTS

3.1. Cost-effectiveness results

Table I shows the population level cost-effectiveness results for the four different strategies. The results are based on a lifetime model of a cohort of 1 million 30 year olds, and net health benefits are calculated at a cost-effectiveness threshold of £20 000 per QALY.

The screening programme in any form improves population health and has positive net health benefits as compared with no screening. On the basis of these cost-effectiveness results, if the objective is solely to maximise population health, we should choose screening with the addition of the universal reminder.

3.2. Inequality results

The baseline distribution of health measured in QALE is shown in Figure 1. We can see from this distribution that in the absence of any screening programme, there are substantial inequalities in the population health distribution.

We next look at the impact of the three screening options on this baseline health distribution. Figure 2 shows the impact of each option in terms of screening uptake by baseline population health. Figure 3 shows how uptake translates into changes in the health distribution.

It is evident from Figure 2 that there is a positive monotonic relationship between baseline health and gFOBT uptake, with uptake being higher for those who are already more healthy, regardless of the specific form of the screening programme under consideration. The universal reminder results in a parallel shift in gFOBT uptake as compared with the standard screening programme, with uptake increasing by the same amount (6%) in each health quintile. The targeted reminder flattens the uptake gradient between the health quintiles, resulting in a higher uptake in the lower health quintiles and a lower uptake in the higher health quintiles as compared with the universal reminder strategy.

Figure 3a shows the changes to the population health distribution associated with each of our three screening strategies relative to no screening, and Figure 3b looks more closely at the impact of the two redesign strategies as compared with the standard screening programme.

Compared with no screening, the screening programme in any of the three forms improves health across the distribution and widens health inequality in absolute terms, improving the health of the healthiest most and the least healthy least. Looking to Figure 3b, we see that compared with standard screening, the universal reminder is health improving across the distribution and further exacerbates absolute health inequality. By contrast, the targeted reminder as compared with standard screening reduces absolute health inequality by focussing additional benefits on the least healthy. It also reduces the health of some of the more healthy groups who benefit very little from the targeted reminder but still bear the health losses because of the opportunity cost of the strategy.

Table I. Standard cost-effectiveness results

	Bowel cancer-related cost (£)	Life years	QALYs	Incremental net health benefit (QALYs) ^a
No screening	278 793 874	50 577 384	41 762 818	—
Standard screening	350 872 069	50 634 273	41 806 794	40 372
Screening + targeted reminder	400 936 962	50 639 192	41 810 506	41 581
Screening + universal reminder	385 268 692	50 639 452	41 810 784	42 642

Results based on a lifetime model for a cohort comprising of 1 million 30 year olds.

^aIncremental to no screening.

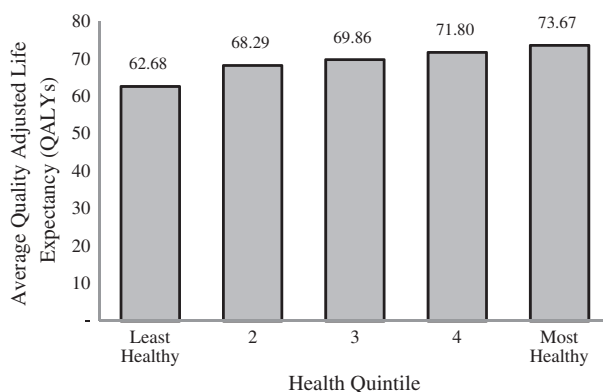


Figure 1. Baseline health distribution

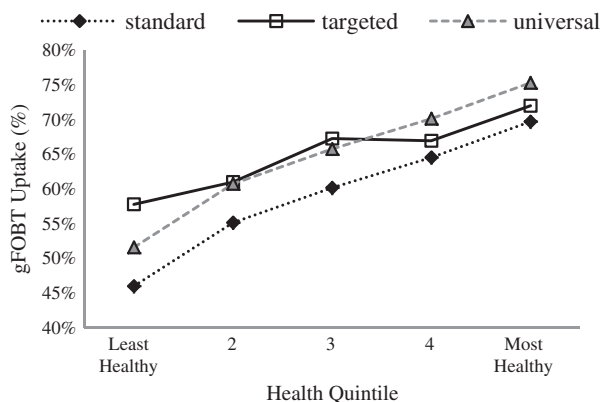


Figure 2. Guaiac fecal occult blood test (gFOBT) uptake distribution

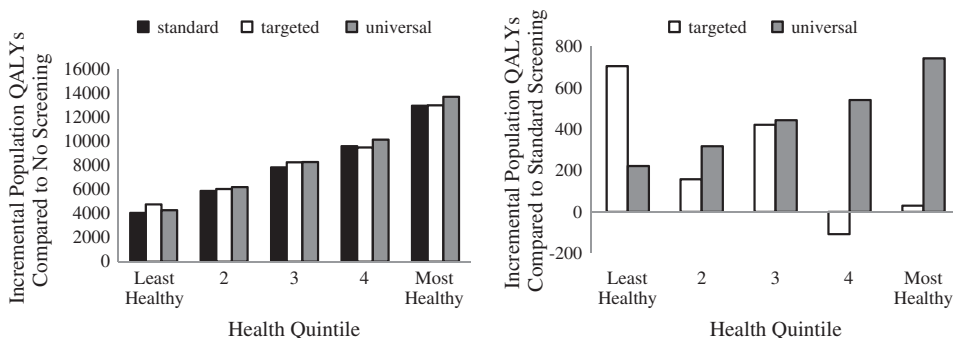


Figure 3. (a) Health compared with no screening (per million of population invited for screened). (b) Health compared with standard screening (per million of population invited for screening)

Combining the baseline health distribution and the estimated distribution of health changes associated with each of our screening strategies provides the overall health distribution associated with each strategy. Table II reports a range of absolute and relative inequality measures calculated for each strategy.

Table II. Measures of inequality

Relative inequality indices	No screening	Standard	Targeted reminder	Universal reminder
Relative gap index (ratio)	0.17527 ^a	0.17592	0.17586	0.17596
Relative index of inequality	0.18607 ^a	0.18674	0.18668	0.18678
Gini index	0.03101 ^a	0.03112	0.03111	0.03113
Atkinson index ($\epsilon=1$)	0.00171 ^a	0.00172	0.00172	0.00172
Atkinson index ($\epsilon=7$)	0.01330 ^a	0.01337	0.01337	0.01338
Atkinson index ($\epsilon=30$)	0.06253 ^a	0.06281	0.06279	0.06283
Absolute inequality indices	No screening	Standard	Targeted reminder	Universal reminder
Absolute gap index (range)	10.98604 ^a	11.03064	11.02726	11.03325
Slope index of inequality	12.88747 ^a	12.94123	12.93691	12.94438
Kolm index ($\alpha=0.025$)	0.20281 ^a	0.20430	0.20416	0.20439
Kolm index ($\alpha=0.1$)	0.87801 ^a	0.88429	0.88371	0.88467
Kolm index ($\alpha=0.5$)	4.56391 ^a	4.58739	4.58587	4.58883

$\epsilon=1$ represents low relative inequality aversion, while $\epsilon=30$ represents high relative inequality aversion.

$\alpha=0.025$ represents low absolute inequality aversion, while $\alpha=0.5$ represents high absolute inequality aversion.

^aIndicates the most equal strategy.

All relative and absolute inequality measures calculated across a range of inequality aversion levels rank no screening as the least unequal and the universal reminder as the most unequal of the four strategies.

3.3. Social welfare results

We next combine our concerns for maximising health and minimising health inequality using social welfare analysis. We find that the estimated health distributions associated with both no screening and standard screening are generalised Lorenz dominated by those associated targeted and universal reminder strategies. This implies that both reminder strategies deliver more population health on average and a fairer distribution of health than the dominated strategies. Dominance does not apply between the targeted and universal reminder strategies, however, so we turn to our social welfare indices evaluated across a range of inequality aversion levels. The values of these indices are reported in Table III.

The social welfare indices show that where there is little or no concern for inequality, the universal reminder is the preferred strategy. However, as inequality aversion increases, the targeted reminder becomes the preferred strategy.

The results thus far have assumed an equal distribution of opportunity cost. Table IV reports the sensitivity of these results to alternative extreme assumptions. When all opportunity costs are borne by the least healthy subgroup, no screening and standard screening are no longer dominated.

Although the distribution of opportunity cost does not impact mean health, it does impact the distribution of health, and this is reflected in the social welfare measures. This is particularly evident at intermediate levels of

Table III. Measures of social welfare

Social welfare indices	Targeted reminder	Universal reminder
Mean health ($\epsilon=\alpha=0$)	69.30127	69.30233 ^a
Atkinson EDE ($\epsilon=1$)	69.18238	69.18331 ^a
Atkinson EDE ($\epsilon=7$)	68.37503	68.37510 ^a
Atkinson EDE ($\epsilon=30$)	64.94991 ^a	64.94796
Kolm EDE ($\alpha=0.025$)	69.09711	69.09794 ^a
Kolm EDE ($\alpha=0.1$)	68.41756	68.41767 ^a
Kolm EDE ($\alpha=0.5$)	64.71541 ^a	64.71350

EDE, equally distributed equivalent.

^aIndicates the strategy yielding the highest social welfare.

Table IV. Sensitivity to opportunity cost distribution

Social welfare indices	All opportunity cost borne by least healthy subgroup				All opportunity cost borne by healthiest subgroup	
	No screening	Standard	Targeted reminder	Universal reminder	Targeted reminder	Universal reminder
Mean health	69.25969	69.30006	69.30127	69.30233 ^a	69.30127	69.30233 ^a
Atkinson EDE ($\varepsilon = 1$)	69.14152	69.18056	69.18147	69.18252 ^a	69.18286	69.18373 ^a
Atkinson EDE ($\varepsilon = 7$)	68.33888	68.36800 ^a	68.36610	68.36734	68.37799 ^a	68.37769
Atkinson EDE ($\varepsilon = 30$)	64.92865 ^a	64.91468	64.89302	64.89892	64.95627 ^a	64.95350
Kolm EDE ($\alpha = 0.025$)	69.05688	69.09486	69.09556	69.09660 ^a	69.09793	69.09866 ^a
Kolm EDE ($\alpha = 0.1$)	68.38168	68.41112 ^a	68.40958	68.41074	68.42046 ^a	68.42020
Kolm EDE ($\alpha = 0.5$)	64.69578 ^a	64.68086	64.65951	64.66532	64.72148 ^a	64.71879

EDE, equally distributed equivalent.

^aIndicates the strategy yielding the highest social welfare.

inequality aversion ($\varepsilon = 7$ or $\alpha = 0.1$) where we see that the preferred screening strategy changes from standard screening when all the opportunity cost is borne by the least healthy group, to universal reminder when opportunity cost is equally distributed, to targeted reminder where all the opportunity cost is borne by the healthiest group.

3.4. Adjustment for alternative social value judgements results

Our results so far have assumed all inequality is unfair; Table V reports the sensitivity of our results to all eight possible sets of social value judgements regarding which inequalities are deemed unfair that can be made in this example.

The sensitivity analysis suggests that in this example, value judgements around the fairness of variation associated with area level deprivation are pivotal in determining the preferred strategy.

Finally, we explore the sensitivity of our social welfare indices calculated for the nondominated strategies to the choice of inequality aversion level as shown in Figure 4a and b. These figures show the difference between the EDE of the alternative strategies. The threshold level of inequality aversion at which the targeted reminder becomes the preferred strategy is eight for the Atkinson EDE and 0.12 for the Kolm EDE. At these levels of inequality aversion, a decision maker would be willing to sacrifice 1000 potential QALYs among the population of 1 million 30 year olds in order to achieve the more equal distribution of health offered by the targeted screening strategy.

Table V. Sensitivity of preferred screening strategy decision to the choice of social value judgements

Social value judgment			Preferred strategy based on social welfare index					
IMD	Ethnic diversity	Gender	Atkinson EDE ($\varepsilon = 1$)	Atkinson EDE ($\varepsilon = 7$)	Atkinson EDE ($\varepsilon = 30$)	Kolm EDE ($\alpha = 0.025$)	Kolm EDE ($\alpha = 0.1$)	Kolm EDE ($\alpha = 0.5$)
Fair	Fair	Fair	U	U	U	U	U	U
Fair	Unfair	Fair	U	U	U	U	U	U
Fair	Fair	Unfair	U	U	U	U	U	U
Fair	Unfair	Unfair	U	U	U	U	U	U
Unfair	Fair	Fair	U	U	T	U	U	T
Unfair	Unfair	Fair	U	U	T	U	U	T
Unfair	Fair	Unfair	U	U	T	U	U	T
Unfair	Unfair	Unfair	U	U	T	U	U	T

IMD, index of multiple deprivation; EDE, equally distributed equivalent; U, universal reminder; T, targeted reminder.

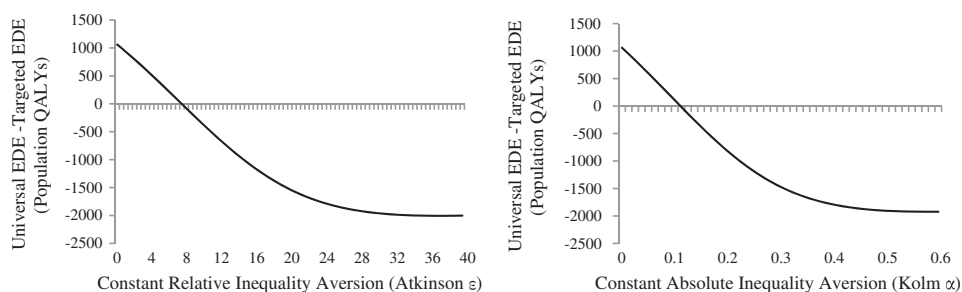


Figure 4. (a) Sensitivity to level of relative inequality aversion. (b) Sensitivity to level of absolute inequality aversion

4. DISCUSSION

4.1. Distributional cost-effectiveness analysis

The results from the model show that although the national BCSP has a small per person benefit, this benefit is substantial at a population level. This is to be expected for a population health intervention such as this, where the majority of people screened will not have bowel cancer, and some of the people who develop bowel cancer may not participate in screening. So despite large individual benefits accruing to people who participate in screening and have their bowel cancer detected early, these benefits accrue to only a relatively small number of people and are averaged across the whole population, giving a small expected per person benefit among the general population.

Targeted and universal reminder strategies to increase uptake of bowel cancer screening both appear to be worthwhile in terms of improving population health. In the base case analysis, both would be viewed as welfare increasing compared with no screening or standard screening for a broad range of social welfare functions reflecting different views on health inequality. The universal reminder resulted in a greater population health improvement than the targeted reminder but was less attractive in terms of its impact on increasing health inequalities. In our base case analysis, the universal reminder would be the preferred intervention at the lower end of the range of inequality aversion values considered, but the targeted reminder could become preferred at high levels of health inequality aversion.

Although all three configurations of the screening programme are health inequality increasing compared with no screening, augmenting the current screening programme with a targeted reminder reduces health inequality. By contrast, augmenting the current screening programme with a universal reminder slightly increases health inequality as compared with the standard screening programme alone. Some aspects of the intervention-generated inequality due to the screening programme arise because of inequalities in uptake of gFOBT and follow-up colonoscopy. However, some of the health inequality impact arises through differing rates of morbidity and other-cause mortality (not related to bowel cancer directly). Because we are interested in lifetime health, as measured here using QALE, detecting cancer earlier and thereby preventing a cancer-related fatality will inevitably deliver a larger health gain in social groups with relatively high QALE (Hauck *et al.*, 2002).

4.2. Sensitivity analyses

No screening and standard screening could be ruled out on the basis of generalised Lorenz dominance, but this was sensitive to an assumption about the distribution of the opportunity cost. The ranking produced by social welfare indices was sensitive to the type and level of inequality aversion. Furthermore, alternative social value judgements about the fairness of variation associated with the different population characteristics impact our choice of preferred strategy.

4.3. Other approaches

A number of methods have previously been proposed in the literature for including health inequality concerns in economic evaluation. These typically involve either weighting health gains differently for different groups in

the population (Nord *et al.*, 1999) or weighting overall health gains directly against overall changes in health inequality in the context of ‘multicriteria decision analysis’ (Baltussen and Niessen, 2006). Both these types of method can be replicated using the DCEA framework by imposing the relevant restrictions on the fairness adjustment process and on the form and parameters of the social welfare function. We therefore see DCEA as encompassing these previous equity weighting methods within a more general framework that provides decision makers with more detailed information about health inequality impacts, rather than as a rival alternative approach.

An important emerging source of empirical literature on incorporating health inequality impacts into economic evaluation in low and middle income countries is the ‘extended CEA’ framework (Verguet *et al.*, 2014). This approach is similar in spirit to DCEA, although simplifies the analysis by (i) focusing on a single distributional variable (wealth quintile group) rather than analysing multiple distributional variables, (ii) setting aside the issue of opportunity costs falling on the health budget by assuming the intervention is funded by the tax system, and (iii) presenting results as a disaggregated ‘dashboard’ of costs and consequences by social group rather than using inequality indices and social welfare functions to explicitly analyse trade-offs between improving health and reducing unfair health inequality.

4.4. Conclusion

The DCEA framework outlined in this paper demonstrates how concerns for unfair health inequality can be taken into account when evaluating health care interventions funded within a fixed health budget. Transparency about value judgements and sensitivity analysis to reflect alternative value judgements is a key feature of the proposed framework. This form of analysis is particularly relevant when considering redesign options for preventive health care programmes to ameliorate intervention-generated inequalities, as in the case of the NHS BCSP. Data requirements for such analyses are nontrivial. However, credible DCEAs are currently feasible in at least some real-world settings, and further analyses will become possible in future as more evidence on distributional outcomes starts to emerge in the era of ‘big data’. Evidence on distributional outcomes can be obtained through evidence synthesis research exploiting networks of patient-level trial datasets, as well as the application of heterogeneous treatment effect estimators to large observational datasets. Research is needed, for example, to synthesise evidence on the social distribution of the effects of different approaches for increasing the uptake of screening and other forms of preventive care, with a view to gauging how far estimates can be generalised from one setting to another. More empirical work is also required to determine a realistic distribution of opportunity costs (plausibly reflecting the impact of likely disinvestment decisions in the health service) and to elicit reasonable ranges of values for societal levels of absolute and relative inequality aversion as well as social value judgements on what should be deemed as fair and unfair variations in health.

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