

Since January 2020 Elsevier has created a COVID-19 resource centre with free information in English and Mandarin on the novel coronavirus COVID-19. The COVID-19 resource centre is hosted on Elsevier Connect, the company's public news and information website.

Elsevier hereby grants permission to make all its COVID-19-related research that is available on the COVID-19 resource centre - including this research content - immediately available in PubMed Central and other publicly funded repositories, such as the WHO COVID database with rights for unrestricted research re-use and analyses in any form or by any means with acknowledgement of the original source. These permissions are granted for free by Elsevier for as long as the COVID-19 resource centre remains active.

ELSEVIER

Contents lists available at ScienceDirect

Health Policy

journal homepage: www.elsevier.com/locate/healthpol



Does moral reasoning influence public values for health care priority setting?: A population-based randomized stated preference survey



Avram E. Denburg^{a,*}, Wendy J. Ungar^b, Shiyi Chen^c, Jeremiah Hurley^d, Julia Abelson^e

- ^a Department of Paediatrics, The Hospital for Sick Children, University of Toronto, Toronto, M5G 1X8, Canada
- ^b Institute of Health Policy, Management, and Evaluation, University of Toronto, Toronto, M5T 3M6, Canada
- ^c Biostatistician, Biostatistics, Design and Analysis, Peter Gilgan Centre for Research and Learning, The Hospital for Sick Children, Toronto, M5G 1X8, Canada
- ^d Department of Economics, McMaster University, Hamilton, L8S 4L8, Canada
- e Department of Health Research Methods, Evidence and Impact, McMaster University, Hamilton, L8S 4L8, Canada

ARTICLE INFO

Article history: Received 24 June 2019 Received in revised form 16 April 2020 Accepted 17 April 2020

Keywords: Health system priority-setting Resource allocation Public preferences Value-based healthcare

ABSTRACT

Objective: Preferences of members of the public are recognized as important inputs into health care priority-setting, though knowledge of such preferences is scant. We sought to generate evidence of public preferences related to healthcare resource allocation among adults and children.

Methods: We conducted an experimental stated preference survey in a national sample of Canadian adults. Preferences were elicited across a range of scenarios and scored on a visual analogue scale. Intervention group participants were randomized to a moral reasoning exercise prior to each choice task. The main outcomes were the differences in mean preference scores by group, scenario, and demographics.

Results: Our results demonstrate a consistent preference by participants to allocate scarce health system resources to children. Exposure to the moral reasoning exercise weakened but did not eliminate this preference. Younger respondent age and parenthood were associated with greater preference for children. The top principles guiding participants' allocative decisions were treat equally, relieve suffering, and rescue those at risk of dying.

Conclusions: Our study affirms the relevance of age in public preferences for the allocation of scarce health care resources, demonstrating a significant preference by participants to allocate healthcare resources to children. However, this preference diminishes when challenged by exposure to a range of moral principles, revealing a strong public endorsement of equality of access. Definitions of value in healthcare based on clinical benefit and cost-effectiveness may exclude moral considerations that the public values, such as equality and humanitarianism, highlighting opportunities to enrich healthcare priority-setting through public engagement.

© 2020 Elsevier B.V. All rights reserved.

What is already known on this topic

- Public preferences are recognized as important inputs into health care priority-setting, but there is little rigorous knowledge of them.
- The values that inform public preferences for health resource allocation are poorly understood.

E-mail address: avram.denburg@sickkids.ca (A.E. Denburg).

What this study adds

- Public values support prioritizing scarce health resources to children
- Structured moral deliberation reduces the intuitive priority given to children.
- There is strong public support for humanitarian principles, such as equality, to guide health care resource allocation across recipient ages and diseases.

1. Introduction

Faced with both scarce resources and pressures to keep pace with innovation, health systems in most developed nations empha-

^{*} Corresponding author at: Division of Haematology/Oncology, Department of Paediatrics, The Hospital for Sick Children, 555 University Ave, Toronto, Ontario, M5G 1X8, Canada.

size the comparative value of health interventions in payment policies and coverage decisions [1,2]. Such appraisals of value rely on assessments of clinical efficacy, economic efficiency and, increasingly, societal values. Growing recognition that democratic commitments warrant more room for public voice in health and social policy decisions – particularly when allocating public resources – has spurred attempts to incorporate citizens' values and preferences in health technology assessment (HTA) in an increasing number of developed countries [3]. While approaches to measuring clinical and economic value are well-specified, the means of identifying and assimilating relevant societal values in HTA are not [4,5].

To bridge this gap, a growing academic literature has sought to address the measurement and interpretation of societal value judgements for health system priority setting. Two contrasting approaches to the elicitation of societal values have prevailed: population-based surveys and exercises in deliberative engagement. Both admit of strengths and limitations. While survey methods are able to elicit preferences from a large swath of the public, they often preclude in-depth reflection and discussion about the complex ethical issues involved in setting priorities [5,6]. By contrast, deliberative engagement with patients or publics offers rich opportunities for nuanced and recursive consideration of the values that motivate allocative decisions, but from the bounded perspective of a small and select group of persons [7]. A few attempts have been made to marry deliberative and survey methods, either by bookending deliberative events with survey questions or by embedding opportunities for deliberation within survey designs [8-10]. We adapt an approach to the latter to investigate societal preferences for allocating health care resources to children as compared to adults.

1.1. Why children?

Age represents one of the most prominent issues explored in the literature on social values for health system resource allocation [11–16]. The focus on age stems in part from the utilitarian assumptions that anchor prevailing methods of health economic evaluation [17,18]. Quality-adjusted life years (QALYs) have played a dominant role in assessing the value of health interventions, as a universal metric for comparisons of benefit among different technologies that incorporate quantity and quality of life into a unified indicator. Closely allied to the use of QALYs as an outcome measure in applied health economic evaluation is the assumption of utilitarian QALY maximization as a normative goal – and thus decision-criterion – when selecting the best alternative among competing interventions [19].

The prominence of QALY maximization as a decision-criterion in applied health economic evaluation, including by HTA institutions internationally, has prompted theoretical and empirical inquiry into the strength of societal preference for QALY maximization [20–23]. Moral philosophers and social choice theorists have challenged the moral legitimacy of purely consequentialist approaches to health care rationing, including QALY maximization, citing the relevance of ethical concepts ranging from distributive justice, priority to the worst off, and rights-based entitlements in health resource allocation [24–29]. Empirical studies of public preferences for health care resource allocation often employ recipient age as a proxy for QALY gains. Studies of public preferences have yielded considerable evidence favouring priority to younger populations [13–16]. Such studies have often compared adults of various ages; select studies include a childhood age range among their comparators [15,30,31,32,33]. However, few studies have explored public preferences for health resource allocation decisions related to children as such. Moreover, the reasons underlying societal preference for the young remain incompletely understood. Crucial questions persist: Does QALY maximization drive precedence to the young or is it independent of life years gained? Are there other normative considerations at play? How do different moral ideas guide age-based prioritization by study participants? Are there specific values that inform allocative decision-making about children?

A number of studies have tried to tease apart the normative presumptions behind age-based rationing decisions. Tsuchiya et al. distinguish and test the evidence for different types of 'ageism' underlying societal preferences for health resource allocation: 1) 'health maximization ageism' (constant relative value of life-years, irrespective of age); 2) 'productivity ageism' (higher value of life-years in young adulthood, related to greater social and economic productivity); and 3) 'fair innings ageism' (emphasis on opportunity for equal aggregate lifetime health (or QALYs) through priority to those expected to experience less, such as the young or disadvantaged) [16]. Each of these approaches is outcome-oriented and concerned with health gains, though calibrated to prioritize different groups based on alternative ethical arguments.

'Health maximization ageism', which corresponds to pure QALY maximization, receives its strongest support in age-based stated preference studies that focus on life-saving interventions, or those that result in more aggregate QALYs for younger recipients. Even so, evidence for societal disavowal of pure QALY maximization exists, with some studies yielding majority preferences for equal allocation in the face of discrepant potential QALY gains across age groups [17,33]. Importantly, studies that test preferences for age-based allocation in terms of QALY maximization alone cannot discern whether distinct moral principles inducing priority to the young (be it children or younger adults) are at play.

When controlling for QALYs gained, the evidence in support of priority to younger groups is mixed. Stated preferences consistent with both 'fair innings ageism' and 'productivity ageism' are evident in select studies examining age-based trade-offs in the context of fixed benefits. When the duration of benefit across age groups is standardized – thereby in effect neutralizing 'health maximization ageism', or pure QALY maximization – consistent prioritization by age breaks down, with participants alternately preferring allocation to children, people in middle age, or equal allocation across age groups [15,16,30,34,35]. It is, however, often difficult to disentangle evidence in support of one of these forms of ageism from the other in the extant literature.

In addition to studies of age-based priority setting that focus on health outcomes, a number of studies explore the moral bases for allocative decision-making by examining the relevance of *causes*. Anand and Wailoo demonstrate weak societal preference for consequentialist rationing rules, including QALY maximization, through experimental rationing decisions that force trade-offs between hypothetical adult recipients of different ages [17]. Notably, they also empirically examine the relevance of deontological considerations in health care rationing, including personal responsibility for one's health state, socioeconomic status, and procedural considerations in priority setting. Their work demonstrates a disavowal by participants of pure QALY maximization, and highlights other salient normative considerations for potential incorporation into rationing exercises, including equality of treatment, individual rights and duties, and procedural fairness [17,36]. Relatedly, a limited body of evidence points to the impact of embedded moral reasoning on attenuated public preference for the young, suggesting that deliberation on a range of ethical principles can influence stated preferences for allocating resources based on age [10]. However, this evidence pertains to age variations amongst adults.

Despite this focus on age as a morally relevant variable, the extant literature contains little dedicated inquiry into allocative preferences regarding children *per se* [37]. This lack of evidence has contributed to a vacuum of both theoretical and context-specific knowledge about societal preferences related to the prioritiza-

tion of health system resources for children. The need for more and better knowledge of public values attached to health care priority-setting affecting children is underscored by inherent challenges associated with the assessment of child health technologies [38-40]. Health system funding decisions for children are often constrained by limited evidence for the clinical efficacy or economic efficiency of child health technologies [41]. Childhood diseases are typically rare, the conduct of research in pediatric populations is complex, and standard metrics of clinical and economic assessment fail to incorporate unique dimensions of childhood, such as family context and life-course impacts [1,42]. Greater uncertainty in clinical and economic domains may confer greater importance on public values in decisions about the prioritization of child health technologies. In addition, evidence of differences in public preferences for health resource allocation across the life course could strengthen standard approaches to the economic evaluation of health care technologies and services through enhanced incorporation of public values.

1.2. Objectives and aims

To better understand societal values for health resource allocation, we conducted a population-based stated preference survey with a nested randomized controlled moral reasoning intervention. Our objective was to generate evidence to inform economic evaluation and policymaking on health care priority-setting and payment reform in developed health systems. Uniquely, we sought to explicitly assess societal preferences for allocation to children, and to test the influence of structured deliberation – in the form of an individual-level moral reasoning exercise – on allocative preferences. Our principal aims were to: (1) understand the direction and strength of public preferences for health resource allocation between children and adults for varied treatment scenarios, (2) assess the impact of a moral reasoning intervention on the expression of such preferences, and (3) identify sociodemographic factors that impact the expression of public preferences on health resource allocation between children and adults. We also sought to test the divergence of participant preferences for children or adults from an assumption of age-neutrality, to understand the treatment scenarios within which significant preferences for either children or adults emerge. Finally, we aimed to characterize the principles that most influenced participants' allocative decisions, to gain a deeper understanding of the moral reasoning behind societal preferences for health resource allocation.

1.3. Hypotheses

In keeping with much of the prior literature on age-based resource allocation, we expected that control group participants would display an aggregate mean preference for allocation to children, particularly in scenarios where theoretical OALY gains were largest (i.e. cancer treatment and eating disorders therapy). In scenarios with fixed and equal life-year gains across children and adults (i.e. chronic disease drug, liver transplant), we hypothesized that participant preferences for children would be less pronounced, but persist on average, due to intuitive conceptions of 'fair innings'. We included a palliative care scenario to test the direction and strength of age-based preferences in a context focused on relief of suffering rather than hypothetical QALY gains, anticipating weak or no preference for allocation to children. In terms of participant sociodemographics, we presumed that younger adults and those with children of their own would preferentially favour allocation to children, due to temporal and emotional proximity to childhood states.

In contrast to prior evidence demonstrating diminished preference for younger adults induced through moral deliberation [10], we hypothesized that a moral reasoning exercise would increase the strength of public preference for allocation to children, as compared to adults. This hypothesis was predicated on insights from foregoing normative analyses of public policies for children, which have identified distinguishing characteristics of childhood - such as vulnerability, dependency, rarity, social distinction, and future potential – as drivers for policy development [43–48]. We postulated that the moral reasoning exercise, which incorporated a number of values-based considerations specific to children, would prompt participants to consider their allocative preferences in light of these unique concerns. Given the ethical tools to unpack intuitions about what children might deserve relative to adults, we anticipated that participants randomized to the moral reasoning arm would apprehend justifications for preferential allocation to children that may not be immediately apparent. We further hypothesized that participants in the intervention arm would display stronger aggregate preference for children across all the clinical scenarios tested, regardless of the duration of life years gained, as a result of their access to a varied palette of ethical principles by which to justify preferential allocation to children, including vulnerability, dependency, potential, and social distinction.

2. Methods

2.1. Study sample

We conducted a population-based stated preference survey of societal views on the prioritization of health resources among children and adults, administered to a non-probability sample of Canadian adults. Participants were recruited through letters of invitation emailed to a random sample from a panel of over one million Canadians maintained by a survey research firm. A limitation of non-probability sampling is the absence of general statistical theory to predict sample representativeness from survey design assumptions [49]. To minimize sampling bias and maximize the potential for national population representativeness, we employed interlocking quotas for stratified sampling (age, gender and region), balanced against Statistics Canada norms, and evaluated the composition of our final sample in comparison to the Canadian population on a range of sociodemographic characteristics. The survey was web-based and loyalty program rewards were offered to encourage participation.

2.2. Survey design and development

Development of the survey instrument drew on prior literature on the ethics of health resource allocation and social values relevant to child health policy. The principles included in our moral reasoning exercise were derived from a systematic review of the literature on social values relevant to child health and social policy, refined through an in-depth qualitative case study of health technology assessment and policymaking for children in Canada [47,48]. Iterative refinements to the survey were informed by a pilot phase with experts (n = 3) and laypersons (n = 2) and field testing with members of the public (n = 32). The final survey questionnaire directed respondents to assign numerical preference scores for the allocation of resources in different health care scenarios based on age-related criteria. It presented each participant with the same five hypothetical treatment scenarios (chronic blood disease, liver transplant, cancer therapy, palliative care, and eating disorder); these scenarios were intended to provide variation in disease characteristics such as acuity, morbidity, mortality, potential for cure, and nature of treatment (Fig. 1; see Supplementary File eFig. 2 for full reproduction of scenarios). Participants chose between funding for treatment among adult (average age 40) and child (average age

Scenario: Chronic Disease Drug

Imagine that two different medicines are available to manage an incurable chronic blood disease: one drug treats the child form of the disease, while a different drug treats the adult form of the disease. Without treatment, patients die of the disease within 6 months of diagnosis. With drug treatment, both child and adult patients can be expected to live about 20 years from diagnosis, after which they usually die from progressive complications of the disease.

Drug A	Drug B
A drug is available for patients with Child-onset Blood Disease .	A drug is available for patients with Adult-onset Blood Disease .
Patients are 10 years old, on average.	Patients are 40 years old, on average.
Patients live about 20 years with the treatment, at which point they usually die of complications from the disease.	Patients live about 20 years with the treatment, at which point they usually die of complications from the disease.
Without the drug, the disease causes death within 6 months.	Without the drug, the disease causes death within 6 months.

Each drug costs the same amount of money and would serve the same number of people. The health system can only afford to fund one of the two drugs at present, and cannot split the money between the two drugs.

Which drug should the health system director fund?

Please slide the bar to any point on the scale from -5 to +5 to show your strength of support for funding one of the drugs.

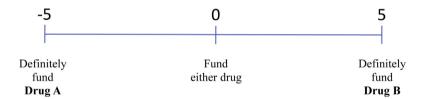


Fig. 1. Sample choice scenario.

Scenario: Chronic Disease Drug

Imagine that two different medicines are available to manage an incurable chronic blood disease: one drug treats the child form of the disease, while a different drug treats the adult form of the disease. Without treatment, patients die of the disease within 6 months of diagnosis. With drug treatment, both child and adult patients can be expected to live about 20 years from diagnosis, after which they usually die from progressive complications of the disease.

Each drug costs the same amount of money and would serve the same number of people. The health system can only afford to fund one of the two drugs at present, and cannot split the money between the two drugs.

Which drug should the health system director fund?

Please slide the bar to any point on the scale from -5 to +5 to show your strength of support for funding one of the drugs.

10) patients, from the perspective of a citizen advisor to a health system administrator. Preferences were captured as continuous variables on a visual analog scale (VAS) from -5 (full preference for children) to +5 (full preference for adults), with zero representing neutrality. We employed a VAS design to explore changes in the strength of participant preferences for children or adults in order to capture more subtle variation in preference than would have been possible with categorical choices, while still allowing for preference neutrality. Demographic data related to age, sex, income, education, employment, health status, and family structure were collected.

2.3. Intervention

Randomization of participants to either an intervention or control group was achieved via a least-fill approach, employing computational logic to assign respondents to the group with the lowest current quota count, which enabled random assignment with respect to stratification variables (age, gender, region). We subjected participants in the intervention group to a moral reasoning exercise prior to each choice scenario. The exercise presented subjects in the intervention group with a list of twelve ethical principles relevant to allocative decisions (Fig. 2). We fashioned principles to capture concepts identified as uniquely germane to health resource allocation involving adults and children, including 'fair innings', vulnerability, dependency, future potential, and distinction, as derived from a foregoing systematic review of social values relevant to health and social policy for children [47]. Balance was sought between principles that might inherently favour allocation to either children or adults. Participants in the intervention arm were asked to select the three principles that most influenced their choice in each scenario. Subjects in the control

Fund treatment based on evidence that it works

- "Fund treatments best proven to be safe and effective."
- "Since it is harder to study treatments in children, evidence is usually stronger for adult treatments."

Help everyone to live a full life

- "The older patients have had their turn."
- "We should take into account all the people affected by a patient's illness, including families and caregivers."

Relieve pain and suffering

- · "When it comes to relieving suffering, other factors shouldn't count."
- "We should always relieve pain when we can."

We should rescue those at risk of dying

- "Everyone deserves the same chance of rescue from life-threatening circumstances."
- "Saving someone's life is important, regardless of age."

Treat those society considers special

• "Children are a distinctly valued social group that deserves privileged treatment."

Give priority to rare diseases

- "Rare diseases are often neglected, so should receive special priority."
- "Childhood disease are often rarer than adult ones, so might be unfairly overlooked in health system planning."

Treat everyone equally

- · "All patients deserve equal access to medical care."
- "Both groups should have the same chance."

Treat those who are dependent on others

- "Children are dependent on their parents or caregivers, so their illness has direct impacts on the lives of others."
- "We should take into account all the people affected by a patient's illness, including families and caregivers."

Relieve pain and suffering

- "When it comes to relieving suffering, other factors shouldn't count."
- "We should always relieve pain when we can."

We should rescue those at risk of dying

- "Everyone deserves the same chance of rescue from life-threatening circumstances."
- "Saving someone's life is important, regardless of age."

Treat those society considers special

• "Children are a distinctly valued social group that deserves privileged treatment."

Give priority to rare diseases

- "Rare diseases are often neglected, so should receive special priority."
- "Childhood disease are often rarer than adult ones, so might be unfairly overlooked in health system planning."

Fig. 2. Moral reasoning exercise: Principles and rationales.

arm responded to the choice scenarios without exposure to a moral reasoning exercise. To minimize question order bias, we randomly rotated the order by which health care scenarios were presented to participants; in addition, we randomly rotated the order of principles within the moral reasoning exercise for the intervention cohort. Participants were prevented from revising their prior responses as they proceeded through the questionnaire.

2.4. Statistical analysis

Quantitative survey data were imported into SAS (Version 9.4) for analysis. Descriptive statistics were employed to characterize the respondent population and compare groups using the Student's *t*-test for continuous variables (age) and chi-squared for categorical variables. We compared select sociodemographic variables from the overall sample with general Canadian population demographics from the 2016 Statistics Canada Census of Population using one-sample proportion tests [50].

We employed a linear mixed-effects random intercept model, which allows for modelling of correlated continuous data, to analyze the strength of participant preferences for each of the scenarios presented and examine the impact of experimental group, scenario, and sociodemographic variables on mean preferences scores, while accounting for the correlation between repeated measures within the same subject. The model examined: 1) the difference in mean preference scores by group, scenario, and demographic characteristics and 2) the difference in mean preference scores between the intervention and control for each scenario. We analyzed the interaction of group and scenario on preference scores to understand whether group mean preference scores varied by scenario type, controlling for covariates (including age, geographic region, gender, language, education, employment, income, health, and family structure) (eMethods).

To further characterize the strength of participant preferences for children or adults, and explore potential choice uncertainty, we analyzed the proportions of respondents displaying any allocative preference beyond the bounds of a 'neutral' construct (zero midpoint), and modelled differences in neutral versus preferential responses as a binary outcome between groups and across scenarios through generalized estimation equation (GEE) modelling. Odds ratios with 95 % confidence limits were computed to indicate the likelihood of a neutral response for each scenario, using chronic disease as the reference scenario. GEE was also used to assess the likelihood of a neutral response by experimental group in each scenario, with moral reasoning as the exposure.

Lastly, we quantified the proportion of respondents selecting each allocation principle overall and by scenario. We tested equality of proportions across scenarios to detect significant differences in the proportions of respondents selecting a given allocative principle. Chi-squared analyses were used to compare the proportions of participants selecting each moral reasoning principle in a given scenario, using one scenario (chronic disease) as a referent.

2.5. Ethical considerations

Ethics approval was granted by the Hamilton Integrated Research Ethics Board affiliated with McMaster University. Informed consent was obtained online as part of the survey panel opt-in process prior to individual survey initiation. The authors received no funding related to study design; collection, analysis, and interpretation of data; the writing of the report; or the decision to submit for publication.

3. Results

3.1. Study sample

Between April 18 and 24, 2017, a total of 12,803 individuals were screened for inclusion: 10,026 were deemed ineligible or declined to participate and 2777 individuals were randomized. Of these, we excluded 1048 for incomplete surveys. A total of 1729 (62.3%) respondents completed the survey; 173 were subsequently excluded for poor quality (e.g., racing). Our final sample included

1556 participants, with 773 in the intervention group and 783 in the control group (eFig. 1). Respondent characteristics were similar across groups (eTable 1). As compared with 2016 Canadian population census data, our sample evinced an over-representation of individuals with higher educational attainment.

3.2. Outcomes

We present our results in sequence corresponding to the hypotheses listed above. We begin with an exposition of control group results, to ascertain whether a baseline predilection for allocation to children was apparent in our sample, and how it varied across disease-specific scenarios. We then move to analysis of mean preference scores in the intervention group, and observed differences between the intervention and control group, to isolate the impact of the moral reasoning exercise on participant preferences. Our analysis of preference divergence from an ageneutral construct (-0.5 to +0.5) follows, to test the strength of allocative preference in either direction. Finally, we analyze participant choices about the moral principles guiding their allocative preferences, focusing on the frequency and patterning of principle selection across scenarios.

3.2.1. Allocative preferences

3.2.1.1. Control. Analysis of mean preference scores demonstrated a consistent aggregate preference by control group participants to allocate scarce health system resources to children across all trial scenarios (Fig. 3; Table 1). The strongest control group preference for children was observed in the cancer therapy (-1.77, 95 % CI -2.06 to -1.48, p < 0.0001) and eating disorders treatment (-2.01, 95 % CI -2.30 to -1.71, p < 0.0001) scenarios, in which the theoretical QALY gains were largest.

3.2.1.2. Intervention. Mean scores were statistically significantly higher (more positive) in the intervention group overall, suggesting a weaker preference for allocation to children in those subjected to the moral reasoning exercise (Fig. 3; Table 1). In the intervention group, a significant preference for allocation to children was retained in the cancer therapy (-0.83, 95 % CI -1.11 to -0.54, p < 0.0001) and eating disorder treatment (-1.11, 95 % CI -1.39 to -0.82, p < 0.0001) scenarios, but there was no age preference in the chronic disease drug, liver transplant and palliative care scenarios (Table 2).

3.2.1.3. Comparison. When analyzing the difference in mean preference scores between experimental groups for each scenario, the intervention had the largest absolute impact for the cancer therapy (0.94, 95 % CI 0.68–1.21, p < 0.0001) and eating disorder treatment (0.90, 95 % CI 0.63–1.16, p < 0.0001) scenarios, reflecting the strength of preference for children in these scenarios in the control group. The results demonstrated that younger respondents and parents had stronger preferences for allocation to children. (See eTables 2–3 for unadjusted and univariate model results.)

Where the benefits of an intervention were most obvious and different between adults and children – as they were in respect of life-years gained from cancer and eating disorders therapy – a choice to prioritize those benefitting most may have seemed easiest, even in the face of exposure to competing moral principles. This explanation is supported by theories of rational decision-making from the field of cognitive psychology that posit dual-process thinking, distinguishing intuition from reasoning [51–53]. The design of our study – including the survey format and pacing, question framing, and lay sample – may have induced intuitive responses from some participants, in the face of complex moral problems characterized by inherent uncertainty. In particular, participants may have allowed more obvious differentiators, such as discrepant benefits in length of life, to intuitively shape their preferences, rather

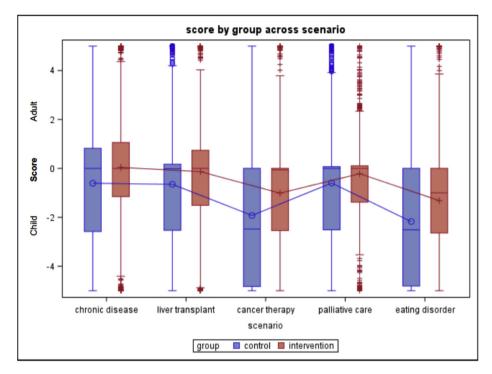


Fig. 3. Unadjusted group mean preference scores across scenarios.

Circle = mean; centre line = median; box = interquartile range (IQR: 1st and 3rd quartiles of the data); whisker (inner fences): lower = 1st quartile - 1.5SD, upper = 3rd quartile + 1.5SD; suspected outliers are noted with a circle (control group) or plus sign (intervention group) beyond the upper and lower inner fences.

Table 1Impact of demographic and experimental variables on mean preference scores: Multiple regression mixed model results.

Variable	Estimate	Standard Error	T value	p value
Intervention (vs control)	0.72	0.14	5.40	<0.0001
Scenario				<0.0001
Liver transplant (vs chronic disease drug)	-0.02	0.11	-0.16	0.87
Cancer therapy (vs chronic disease drug)	-1.30	0.11	-11.78	<0.0001
Palliative care (vs chronic disease drug)	0.05	0.11	0.41	0.68
Eating disorders treatment (vs chronic disease drug)	-1.53	0.11	-13.89	<0.0001
Group and scenario interaction				0.0021
• Intervention (vs control) and liver transplant (vs chronic disease drug)	-0.18	0.16	-1.16	0.25
• Intervention (vs control) and cancer therapy (vs chronic disease drug)	0.22	0.16	1.42	0.16
• Intervention (vs control) and palliative care (vs chronic disease drug)	-0.31	0.16	-2.00	0.05
• Intervention (vs control) and eating disorders (vs chronic disease drug)	0.17	0.16	1.11	0.27
Ontario (vs other regions)	0.02	0.10	0.23	0.82
Age categories				<0.0001
• 35 - 44 vs 18-34	0.35	0.14	2.39	0.02
• 45 – 54 vs 18–34	0.54	0.14	3.92	<0.0001
• 55+ vs 18–34	0.71	0.14	5.06	<0.0001
Female	0.12	0.09	1.30	0.19
English (vs French)	-0.28	0.12	-2.32	0.02
Education: some college or higher	0.04	0.13	0.29	0.77
Full-time employment	-0.04	0.11	-0.39	0.70
Medium-to-high income (vs low income)	-0.40	0.20	-1.99	0.05
Good-to-excellent health (vs fair or poor)	0.11	0.16	0.69	0.49
Married or living with partner (vs single or divorced)	0.06	0.10	0.55	0.58
One or more children (i.e. parenthood)	-0.40	0.11	-3.73	0.0002

Overall model significance: Log-likelihood ratio tests suggest the full model is significantly better than a null model (LR = 1049.38, p < 0.0001), but reveals no significant difference in fit between the full and parsimonious models (LR = 14.1, df = 7, p = 0.95).

R²: The full model with predictors explained 9.7 % of within-subjects variance and 4.3 % of between-subjects variance, as compared with the null model (without predictors). Comparing the full model with a parsimonious model (non-significant predictors removed), the two models were almost identical. The parsimonious model explained 0.05 % more of the between-subjects variance; there was no difference in within-subjects variance explained.

than take the time and cognitive effort to work through conflicting moral choices in a rule-bound manner. This is likely particularly true of those unexposed to the moral reasoning intervention, and might explain why preference score differences between the intervention and control groups were largest in respect of cancer therapy and eating disorders treatment. It may also explain why variance in preference scores narrowed consistently across scenarios in the

intervention group as compared to the control group: it is possible that this reduction in variance represents a reduction in choice uncertainty, in the context of enhanced participant reliance on analytical reasoning induced by the study intervention.

3.2.1.4. Neutrality. Proportions of neutral response (score between -0.5 and +0.5) were consistently higher in the intervention group

Table 2Mean deviation of preference scores from zero difference between groups: Multiple regression mixed model results.

Scenario	Intervention		Control		Difference		
	Mean	95 % CI	Mean	95 % CI	Estimate	95 % CI	p value
Chronic disease	0.25	(-0.03, 0.53)	-0.47	(-0.76, -0.18)	0.72	(0.46, 0.99)	<0.0001
Liver transplant	0.05	(-0.23, 0.34)	-0.49	(-0.78, -0.20)	0.54	(0.28, 0.80)	< 0.0001
Cancer therapy	-0.83	(-1.11, -0.54)	-1.77	(-2.06, -1.48)	0.94	(0.68, 1.21)	< 0.0001
Palliative care	-0.02	(-0.30, 0.27)	-0.43	(-0.72, -0.14)	0.41	(0.15, 0.67)	0.0021
Eating disorder treatment	-1.11	(-1.39, -0.82)	-2.01	(-2.30, -1.71)	0.90	(0.63, 1.16)	< 0.0001

CI = confidence interval.

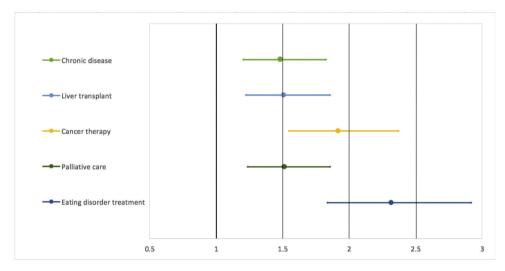


Fig. 4. Odds of preference neutrality: Intervention vs. control.

p < 0.0002 for all comparisons

across all scenarios (eTable 4). Univariate GEE analysis revealed the impact of the moral reasoning intervention on preference neutrality across children and adults, with neutral responses significantly more likely in the intervention group than the control group (OR = $1.69,\,95\,\%$ CI $1.45-1.96,\,p<0.0001$). When examining the impact of scenario, taking chronic disease as the referent, neutral responses were significantly more likely in the palliative care scenario (OR = $1.27,\,95\,\%$ CI $1.13-1.43,\,p<0.0001$) and significantly less likely in the eating disorder scenario (OR = $0.63,\,95\,\%$ CI 0.56 to $0.71,\,p<0.0001$). The multiple regression GEE model affirmed greater odds of preference neutrality in the intervention group as compared to the control group across all scenarios (Fig. 4; eTable 5).

Two explanations for greater apparent preference neutrality in the intervention group are possible. The greater predilection for neutrality in the experimental group may represent stronger endorsement of equality as a primary justification for resource allocation, induced by exposure to a range of humanitarian principles, including statements about equality, relief of suffering, and rule of rescue. Conversely, participants may have found it difficult to resolve competing moral intuitions through reasoned deliberation, particularly in the scripted and time-bound context of a survey. Greater preference neutrality in the intervention group could represent a tendency to reject a difficult moral choice between adults and children, one that pulled responses closer to zero and limited stronger preferences to scenarios where obvious benefits (such as differential QALY gains) made the moral leap less daunting.

3.3. Moral reasoning analysis

We made a conscious attempt to incorporate principles related to an array of individual and societal benefits and costs, to induce reflection on the range of moral reasons one might consider germane to health system resource allocation. We also sought balance between principles that might intrinsically engender preference for children or adults. Participants exposed to the moral reasoning exercise demonstrated remarkable consistency in their prioritization of principles for allocative decision-making. The top three principles guiding participants' allocative decisions were: 1) treat equally (54.3%–63.9%), 2) relieve suffering (39.6%–66.1%), and 3) rescue those at risk of dying (37%–40.8%). In all cases except palliative care, 'treat equally' ranked number one, with a proportion uniformly greater than 50%; subjects deemed 'relieve suffering' most important (66.1%) in the context of palliative care, followed closely by 'treat equally' (63.9%). The least endorsed principles (\leq 10%) for assigning allocative preference were: priority to rare diseases, priority to special populations, and priority based on societal productivity.

Despite consistency in ranking of principles, the proportion of participants selecting a given principle to guide allocative decisionmaking varied significantly across scenarios (Table 3). As compared to the chronic disease treatment scenario, the cancer therapy scenario prompted significantly more participants to cleave to principles that, in theory, justify preferential allocation to children: opportunity to live a full life (24.7 % vs 19.2 %, p < 0.0028), duration of benefit (34.5 % vs 19.5 %, p < 0.0001), and concern for special populations (11.9 % vs 7.4 %, p < 0.0026). They also tended to disavow principles favouring allocation to adults, including family responsibilities (14.2 % vs 24.7 %, p < 0.0001) and economic productivity (7.9 % vs 16 % p < 0.0001). A similar pattern was observed for the eating disorder treatment scenario, with the addition of vulnerability (36.1 % vs 21.9 %, p < 0.0001) to the principles endorsed. By contrast, in the palliative care scenario, participants displayed significantly more concern for equality of treatment (63.9 % vs 55.2 %, p < 0.0005) and relief of pain and suffering (66.1 % vs 39.6 %, p < 0.0005) 0.0001), and less concern for 'fair innings' (the entitlement to a full lifespan) or duration of benefit.

Table 3Participant selection of allocative principles by scenario.

Principle	Overall (%)	Chronic disease drug	Liver transplant	Cancer therapy	Palliative care	Eating disorder treatment	Aggregate p value ^b
Equal treatment	57.75	55.24	58.99	54.33	63.91 ^a	56.27	0.0008
Relief pain and suffering	45.33	39.59	40.75	40.10	66.11 ^a	40.10	<0.0001
At risk of dying	40.44	44.11	42.56	40.62	37.90	37.00	0.02
Capacity to benefit longer	24.89	19.53	24.71 ^a	34.54 ^a	15.91	29.75 ^a	<0.0001
Most vulnerable	24.71	21.86	18.76	22.51	24.32	36.09 ^a	<0.0001
Evidence that it works	24.14	25.87	26.65	23.42	20.44	24.32	0.04
Live a full life	20.65	19.15	17.98	24.71 ^a	17.21	24.19 ^a	<0.0001
Treat those dependent on others	17.46	16.56	17.34	18.50	16.30	18.63	0.65
Family responsibility	16.56	24.71	20.57	14.23 ^a	13.07 ^a	10.22 ^a	<0.0001
Other considerations	14.41	13.45	16.56	11.90	15.27	14.88	0.09
Productive people	10.45	16.04	14.10	7.89 ^a	7.50 ^a	6.73 ^a	<0.0001
Special people	10.25	7.37	9.96	11.90	11.00	11.00	0.04
Rare disease	7.37	9.96	7.63	7.24	6.34	5.69	0.02

a Statistically significant difference in proportion selecting the principle for indicated scenario compared to the chronic disease scenario at p < 0.01 level.

4. Discussion

A principal finding of our study is the consistent preference for allocation to children across health care scenarios in the overall cohort. This finding is in keeping with much of the extant evidence on societal preferences for allocation to the young, but adds depth and specificity in relation to health care resource allocation to children *per se*, as most prior literature primarily examines the normative relevance of age variations among adults [13,14,17,34,35]. Our focus on allocative trade-offs between children and adults allowed for sustained examination of societal preferences related to children in the face of changing health conditions and outcomes, distinguishing it from prior literature.

The strength of the preference for children in our study varied by scenario, and was influenced by a number of factors. It was greatest in relation to cancer therapy and eating disorders treatment. These were the only scenarios where the gain in life-years from treatment was not bounded by the natural history of the condition and equalized across hypothetical programs, but tagged to normal life expectancy. Consequently, there was a clear expected difference in the benefit to be gained (as measured in length of life) between the idealized adult and child populations of interest. This may represent a preference for life years gained rather than children per se. Tsuchiya et al. have demonstrated altered patterns of age-based allocative preference in response to changes in the duration of benefit from a given intervention [16]. In the face of life-long benefits (e.g. life-saving interventions), their study participants gave consistent precedence to younger groups when allocating scarce resources. By contrast, when juxtaposed with life-long benefits, fixed benefits induced a preference pattern that privileged young to middle adulthood. Our results may indicate similar moral intuitions behind participant choices, though with the notable difference of an equal allocation option. Where pure QALY maximization was possible – namely, in the cancer therapy and eating disorders treatment scenarios – participants seemed to adjudge the life-long benefits that would accrue to children too large to overlook. Conversely, in the scenarios with fixed benefits, preference for allocation to children was weaker in the control group, and a trend toward preference for equal allocation emerged in the intervention cohort. An alternative explanation could be that participants perceived scenario-specific differences in duration of benefit more readily than other benefits that, though not explicitly mentioned, also retain moral relevance in the context of these and similar scenarios. Such benefits could include reduction in inequality of outcomes – for instance, founded on inherent characteristics of the disease (rarity) or population (vulnerability, dependency) – or equitable chances to live a full life ('fair innings') [54–59].

Interestingly, our results at once confirm and challenge prior evidence that suggests decision-making from an impersonal vantage point (e.g. a budgetary decision-maker) makes it easier to discriminate in favour of one group as against another. Nord et al. elicited allocative preferences from study participants within two different assumed perspectives: a 'veil of ignorance', in which the participants themselves might need the health intervention in question one day; and a health system administrator role, in which they were tasked with decisions about resource allocation to others from a budgetary standpoint [15]. They found that the tendency to privilege younger people in allocative decisions was more evident amongst those who assumed the latter perspective, and argued that the degree of emotional remove induced by the shift in perspective might account for this difference. Evidence from our study for the moderating effect of a moral reasoning exercise on participant preferences for allocation to children, from an administrative perspective, suggests the opportunity for ethical reflection mitigated participant willingness to discriminate between groups on the basis of age. This may reflect choice uncertainty and a resultant instinct for preference neutrality, rather than evidence of real, rich moral deliberation. Alternatively, this may represent a 'depersonalizing' effect of moral deliberation on allocative decision-making, one sufficient to impact societal preferences for health resource allocation - but in the opposite direction to that observed by Nord et al.

Counter to our hypothesis, exposure to a moral reasoning intervention diminished participant preferences for allocation to children. Evidence of a moderating effect of moral reasoning on allocative preference for children suggests the opportunity for ethical reflection mitigated participant willingness to discriminate between groups on the basis of age. This relationship may have been driven by a more varied set of ethical principles with which to draw conclusions. Interestingly, this is the same consideration that motivated our hypothesis of *increased* preference for children through moral reasoning. We presumed that the ethical nuances attached to funding health interventions for children - including distinguishing features such as vulnerability, dependency, neglect. and future potential, that stood out in prior normative analyses of child health and social policy - would come through more clearly to participants when exposed to a range of principles touching on them [43–47,54–58].

Notably, the strength of preference for equal allocation increased among those randomized to the moral reasoning exercise. One interpretation of this is that the intervention prompted participants to evaluate the relationship between their moral intuitions and their stated preferences, and induced a reasoned judgment about the importance of equality as a guiding principle for resource allocation. An alternative explanation is that, despite

b Test of equality of proportions across scenarios.

the study intervention, it remained cognitively difficult for participants to reason through the uncertainty inherent in weighing competing moral principles. The greater predilection for neutrality in the experimental group may have represented retreat to equality in the face of difficult choices.

4.1. Strengths and limitations

Our study admits of potential limitations. In relation to the survey instrument, the brief statements used to evoke the meaning of each principle may have failed to capture its ethical nuances. Different framing of the principles may have led to different patterns of preference among participants; however, randomly varying the sequence of principles mitigated any ordering effect bias [60,61]. Use of a continuous scale overlapping zero to measure the strength of allocative preferences in the context of a binary choice task (fund either intervention A or B) may have led some participants to misinterpret zero as a choice to allocate equal sums of money to each population, rather than agnosticism about which intervention to fund. We sought to mitigate this potential limitation through simple, explicit scenario instructions; however, it is possible that misapprehension of the 'neutrality of preference' state by some participants impacted our results. The study was not designed to measure the time it took to read the principles in the moral reasoning intervention separately from completing the choice tasks. This could have given a proxy sense of whether participants read and engaged with the ethical ideas and choices involved. The assembly and composition of our study sample may have also limited the external validity of our results. Values endorsed by the Canadian public may not reflect those of other polities. The use of covariateadaptive randomization to form balanced treatment groups with respect to relevant covariates has both benefits and inherent shortcomings; use of stratified randomization may have permitted some selection bias and only balanced a limited number of potentially relevant covariates [62]. More specifically, the survey firm generated cohorts from an opt-in panel of eligible Canadians recruited and incentivized through brand loyalty points programs. Some of the inherent selection bias associated with online opt-in panels was evident, privileging those with higher education who are web literate. However, the lack of statistically significant impacts of education and income on preference scores in the multivariate model attenuates theoretical concerns about sociodemographic bias.

Our study also has important strengths. It is, to our knowledge, the first to experimentally examine the values behind health and social policy decisions about children, their valence in relation to adult needs and priorities, and their stability in the face of moral deliberation. Its randomized design and large, heterogeneous population-based sample allowed for robust conclusions about the effect of the intervention on participant preferences. Our findings demonstrate a convincing relationship between exposure to a range of relevant moral principles and the priorities set. This affirms the complexity of such decisions and the impact of ethical deliberation on them. These findings challenge reflexive trust in survey-based preference elicitation, and imply the need to complement such modalities with deliberative modes of public engagement on questions of social importance, such as the allocation of scarce public resources.

4.2. Implications for research and policy

In policy terms, our work both supports and challenges conceptions of value in health care based primarily on QALY maximization. Dominant modes of health economic evaluation privilege interventions that maximize aggregate individual and societal utility, typically in the form of the duration and magnitude of benefits reaped and the size of the population reached. Our results

demonstrate a stronger preference for children in the hypothetical scenarios (cancer and eating disorders) in which the duration of benefit accrued mainly to children; this may represent an underlying motivation by participants to maximize benefits. However, we also observed a strong inclination for equality and humanitarianism amongst study participants, regardless of experimental group. The three principles deemed most salient to priority-setting were equal treatment, relief of suffering, and rule of rescue. None of these prioritizes age as a relevant variable; each starts from a belief in the intrinsic value of all human life, and resists grading that value by social context or circumstance. Our results, therefore, suggest the need to incorporate other values into health care priority-setting, as some value frameworks for funding decisions have begun to do [63]. The salience of these findings to priority-setting efforts in the context of public health emergencies - including access to scarce ventilators or drugs in the context of the COVID-19 pandemic – is readily apparent. They furnish empirical evidence for the adjudication and refinement of ethical frameworks for triage and resource allocation during the pandemic based on public values in Canada [64,65]. Efforts to incorporate a broader range of public values and preferences in health and social policy decisions also involve inherent risks. Among them is the need to confront and grapple with public attitudes that conflict with the fundamental normative or legal principles that structure most democratic societies, including basic commitments to the protection of individual rights and nondiscrimination [66]. Future research to explore this tension and ways to resolve or address it is warranted.

Allied to this, our study affirms the importance of process in health care priority-setting exercises. The opportunity to explicitly consider varied normative ideas appears crucial to informed allocative decisions based on public values. The observed changes in public preferences in the face of competing moral principles imply the impact, and potential relevance, of structured opportunities for moral reasoning when making such consequential decisions. This includes the potential value of deliberative public engagement to health policymaking, particularly in domains where distinct ideas about the public good may compete. Specific to children, few efforts to incorporate public deliberation in research and policy on child health and social policy priorities have prevailed to date. In the face of resource scarcity, evidence-informed child health and social policies will depend on the careful elicitation and integration of public values. This and allied work bear potential relevance in social policy domains beyond health, including public attitudes to 'deservingness' attached to welfare state distributional policies in domains as diverse as labour markets and education [67–,68,69,70,71]. Crucial future areas of inquiry and application include the inclusion of child voices in research and policymaking, and the exploration of social values for priority setting within public policies and programs for children.

5. Conclusions

Our study underscores the relevance of age in public preferences for the allocation of scarce health care resources, extending evidence of this calculus to trade-offs involving children. Nevertheless, it demonstrates the mutability of such preferences in the face of structured moral deliberation. This finding has three main repercussions. It furnishes a critical lens for the interpretation of stated preference surveys; introduces a note of caution into dominant modes of health care funding allocation decisions premised on utility maximization; and implies the value of deliberative methods as a complement to both. We observed a strong inclination for equality and humanitarianism amongst study participants, regardless of experimental group. These moral impulses prevailed over consequentialist logic, including priority to the young founded on

aggregate benefit. The stability of these principles in the face of changing allocative preferences signals their importance as public values – but it also hints at the complexity of values-based decision-making. The public seems, at face value, to believe in equality of access and defend a set of shared human entitlements to care. It also seems to assign intuitive priority to children. The challenge of reconciling these convictions demands processes nimble enough to negotiate this paradox. Spaces for moral deliberation – whether in large-scale surveys or focused qualitative engagement – are essential to arrive at health care priorities that reflect what we collectively hold dear.

Contributions

AD conceived and designed the study. SC and AD performed the statistical analysis. AD prepared the first draft of the manuscript. All authors contributed to study design, critically revised the manuscript, and approved the final version. As corresponding author, AD accepts responsibility for the work, had full access to the data, and controlled the decision to publish. AD attests that all listed authors meet authorship criteria and that no others meeting the criteria have been omitted.

Declaration of Competing Interest

None.

Acknowledgements

During the period of this research, AD was supported by grants from the Canadian Institutes of Health Research, Pierre Elliott Trudeau Foundation, Canadian Child Health Clinician Scientist Program, and Pediatric Oncology Group of Ontario.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:https://doi.org/10.1016/j.healthpol.2020.04.007.

References

- [1] [Anonymous] Details omitted for double-blind reviewing.
- [2] Wong CA, Perrin JM, McLellan M. Making the case for value-based payment reform in children's health care. JAMA Pediatrics 2018;172(6):513–4.
- [3] [Anonymous] Details omitted for double-blind reviewing.
- [4] Stafinski T, Menon D, Philippon D, McCabe C. Health technology funding decision-making processes around the world: the same, yet different. Pharmacoeconomics 2011;29(6):475–95.
- [5] [Anonymous] Details omitted for double-blind reviewing.
- [6] Dolan P, Cookson R, Ferguson B. Effect of discussion and deliberation on the public's views of priority setting in health care: focus group study. BMJ 1999;318:916–9.
- [7] [Anonymous] Details omitted for double-blind reviewing.
- [8] Fishkin JS. Democracy and deliberation. New Haven, Conn: Yale University Press; 1991.
- [9] Luskin R, Fishkin JS, Jowell R. Considered opinions: deliberative polling in Britain. British Journal of Political Science 2002;32:455–87.
- [10] Johri M, Damschroder LJ, Zikmund-Fisher BJ, et al. Can a moral reasoning exercise improve response quality to surveys of healthcare priorities? Journal of Medical Ethics 2009;35:57–64.
- [11] Lewis PA, Charny M. Which of two individuals do you treat when only their ages are different and you can't treat both? Journal of Medical Ethics 1989;15:28–34.
- [12] Busschbach JJ, Hessing DJ, de Charro FT. The utility of health at different stages in life: a quantitative approach. Social Science & Medicine 1993;37:153–8.
- [13] Johannesson M, Johansson PO. Is the valuation of a QALY gained independent of age? Some empirical evidence. Journal of Health Economics 1997;16:589–99.
- [14] Tamura M, Kawata C, Hashimoto M. An empirical study of the fairness of allocation of health care resources. Japanese Journal of Health Economics and Policy 1995;2:55–70.
- [15] Nord E, Street A, Richardson J, et al. The significance of age and duration of effect in social evaluation of health care. Health Care Analysis 1996;4:103–11.

- [16] Tsuchiya A, Dolan P, Shaw R. Measuring people's preferences regarding ageism in health: some methodological issues and some fresh evidence. Social Science & Medicine 2003:57:687–96.
- [17] Anand P, Wailoo A. Utilities versus rights to publicly provided goods: arguments and evidence from health care rationing. Economica 2000;67:543–77.
- [18] Brock DW, Daniels N, Neumann PJ, Siegel JE, et al. Ethical and distributive considerations. In: Neumann PJ, editor. Cost-effectiveness in health and medicine, 2nd edition Oxford, UK: Oxford University Press; 2017. p. 319–42.
- [19] Torrance GW. Measurement of health state utilities for economic appraisal. Journal of Health Economics 1986;5:1–30.
- [20] Donaldson C, Baker R, Mason H, et al. The social value of a QALY: Raising the bar or barring the raise? BMC Health Services Research 2011;11(8):1–8.
- [21] Williams A. QALYS and ethics: a health economist's perspective. Social Science & Medicine 1996;43:179–804.
- [22] Weinstein M. A QALY is a QALY is a QALY—or is it? Journal of Health Economics 1988;7:489–91.
- [23] Dolan P, Shaw R, et al. QALY maximisation and people's preferences: a methodological review of the literature. Health Economics 2005;14:197–208.
- [24] Scheffler S, editor. Consequentialism and its critics. Oxford: Clarendon Press; 1988.
- [25] Rawls J. A theory of justice. Boston: Harvard University Press; 1971.
- [26] Scanlon TM. Rights, goals, and fairness. In: Scheffler S, editor. Consequentialism and its critics. Oxford: Clarendon Press; 1988. p. 74–92.
- [27] Sen AK. Development as freedom. Oxford: Oxford University Press; 1999. p. 87.
- [28] Sen AK. Personal utilities and public judgements: or, what's wrong with welfare economics. Economic Journal 1979;89:537–58.
- [29] Kolm SC. Rational normative economics vs 'social welfare' and 'social choice'. European Economic Review 1994;38:721–30.
- [30] Rodriguez E, Pinto JL. The social value of health programmes: Is age a relevant factor? Health Economics 2000;9:611–21.
- [31] Carlsson F, Daruvala D, Jaldell H. Preferences for lives, injuries, and age: a stated preference survey. Accident Analysis and Prevention 2010;42:1814–21.
- [32] Johansson-Stenman O, Martinsson P. Are some lives more valuable? An ethical preferences approach. Journal of Health Economics 2008;27:739–52.
- [33] Nord E, Richardson J, Street A, et al. Maximizing health benefits versus egalitarianism: an Australian survey of health issues. Social Science and Medicine 1995;41(10):1429–37.
- [34] Cropper ML, Aydede SK, Portney PR. Preferences for life saving programs: how the public discounts time and age. Journal of Risk and Uncertainty 1994;8:243–65.
- [35] Dolan P, Tsuchiya A. Health priorities and public preferences: the relative importance of past health experience and future health prospects. Journal of Health Economics 2005;24:703–14.
- [36] Wailoo A, Anand P. The nature of procedural preferences for health-care rationing decisions. Social Science and Medicine 2005;60:223–6.
- [37] Petrou S. Should health gains by children be given the same value as health gains by adults in an economic evaluation framework? In: Ungar WJ, editor. Economic evaluation in child health. Oxford, UK: Oxford University Press; 2010. p. 71–87.
- [38] [Anonymous] Details omitted for double-blind reviewing.
- [39] [Anonymous] Details omitted for double-blind reviewing.
- [40] Keren R, Pati S, Feudtner C. The generation gap: differences between children and adults pertinent to economic evaluations of health interventions. Pharmacoeconomics 2004;22(2):71–81.
- [41] Janvier A, Bauer KL, Lantos JD. Are newborns morally different from older children? Theoretical Medicine & Bioethics 2007;28(5):413–25.
- [42] Jenson J, Fraser M. Social policy for children and families: a risk and resilience perspective. London: Sage Publications Inc.; 2006.
- [43] Halfon N, Hochstein M. Life course health development: an integrated framework for developing health, policy, and research. Milbank Quarterly 2002;80(3):433–79.
- [44] Mayall B. Towards a sociology of child health. Sociology of Health & Illness 1998;20(3):269–88.
- [45] [Anonymous] Details omitted for double-blind reviewing.
- [46] [Anonymous] Details omitted for double-blind reviewing.
- [47] Cornesse C, Blom AG, Dutwin D, et al. A review of conceptual approaches and empirical evidence on probability and nonprobability sample survey research. Journal of Survey Statistics and Methodology 2020;8(1):4–36.
- [48] Statistics Canada. Census profile; 2016. Census. Available: http://www12.statcan.gc.ca/census-recensement/2016/dp-pd/prof/index.cfm?Lang=E (Accessed 2018 Apr.23).
- [49] Kahneman D. A perspective on judgment and choice mapping bounded rationality. American Psychologist 2003;58:697–720.
- [50] Stanovich KE, West RF. Individual differences in reasoning: Implications for the rationality debate? Behavioral and Brain Sciences 2000;23:645–65.
- [51] Stanovich KE. Who is rational? Studies of individual differences in reasoning. Mahwah, NJ: Erlbaum; 1999.
- [52] Williams A. Intergenerational equity: an exploration of the 'fair innings' argument. Health Economics 1997;6:117–32.
- [53] Culyer A, Bombard Y. An equity framework for health technology assessments. Medical Decision Making 2012;32:428–41.
- [54] Gormley W. Voices for children: rhetoric and public policy. Brookings Institution Press; 2012.
- [55] Mayall B. Values and assumptions underpinning policy for children and young people in England. Children's Geographies 2006;4(1):9–17.

- [56] Winquist E, Bell CM, Clarke JTR, et al. An evaluation framework for funding drugs for rare diseases. Value in Health 2012;15:982–6.
- [57] Tsuchiya A. Age-related preferences and age weighting health benefits. Social Science & Medicine 1999;48:267–76.
- [58] Desser AS, Olsen JA, Grepperud S. Eliciting preferences for prioritizing treatment of rare diseases: the role of opportunity costs and framing effects. Pharmacoeconomics 2013;31(11):1051–61.
- [59] Rabin M. Psychology and economics. Journal of Economic Literature 1998;36:11–46.
- [60] Zhao W. A better alterative to stratified permuted block design for subject randomization in clinical trials. Statistics in Medicine 2014;33(30): 5239–48.
- [61] [Anonymous] Details omitted for double-blind reviewing.
- [62] White DB, Lo B. A framework for rationing ventilators and critical care beds during the COVID-19 pandemic. JAMA 2020; (March).
- [63] Emanuel EJ, Persad G, Upshur R, et al. Fair allocation of scarce medical resources in the time of Covid-19. New England Journal of Medicine 2020; (March), http:// dx.doi.org/10.1056/NEJMsb2005114.
- [64] [Anonymous] Details omitted for double-blind reviewing.

- [65] Oorschot WV. Who should get what, and why? On deservingness criteria and the conditionality of solidarity among the public. Policy & Politics 2000;28(1):33–48.
- [66] Jensen C, Petersen MB. The deservingness heuristic and the politics of health care. American Journal of Political Science 2017;61(1):68–83.
- [67] Larsen CA. The political logic of labour market reforms and popular images of target groups. Journal of European Social Policy 2008;18(1):50–63.
- [68] Naumann E, De Tavernier W, Naegele L, Hess M. Public support for sanctioning older unemployed-a survey experiment in 21 European countries. European Societies 2019:1–24.
- [69] Knijn T, van Oorschot W. The need for and the societal legitimacy of social investments in children and their families: critical reflections on the Dutch case. Journal of Family Issues 2008;29:1520–42.
- [70] Lundberg O, Yngwe MÅ, Stjärne MK, et al. The role of welfare state principles and generosity in social policy programmes for public health: an international comparative study. The Lancet 2008;372:1633–40.
- [71] Cannella GS, Swadener BB. Contemporary public policy influencing children and families. International Journal of Educational Policy, Research, and Practice: Reconceptualizing Childhood Studies 2006;7:81–93.