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Preference-Based Assessments

Can Independently Elicited Adult- and Child-Perspective Health-State Utilities Explain Priority Setting?

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ABSTRACT

Objectives: Time trade-off (TTO) utilities for EQ-5D-Y-3L health states valued by adults taking a child's perspective are generally higher than their valuations of the same state for themselves. Ceteris paribus, the use of these utilities in economic evaluation implies that children gain less from treatments returning them to full health for a specified amount of time than adults. In this study, we explore if this implication affects individuals' views of priority-setting choices between treatments for adults and children.

Methods: We elicited TTO utilities for 4 health states in online interviews, in which respondents valued states for a 10-year-old child and another adult their age. Views on priority setting were studied with person trade-off (PTO) tasks involving the same health states. We tested the ability of the subjects' TTO utilities to predict these societal choices in PTO.

Results: There are no significant differences between adult and child health state valuations in our study, but we do observe a substantial preference for treating children over adults in the PTO task.

Conclusions: Our findings suggest that perspective-dependent health-state utilities only explain a small part of views on priority setting between adults and children. External equity weights might be useful to better explain the higher priority given to children.

Keywords: EQ-5D-Y-3L, equity weighting, person trade-off, priority setting, quality-adjusted life-year model, time trade-off.

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Introduction

Economic evaluations of health interventions often use quality-adjusted life-years (QALYs) to express health benefits. QALYs are a utility measure with 1 QALY being the equivalent of 1 year in perfect health or, for example, 2 years in a health state with a utility of 0.5. It can, among others, be used to compare the value of different medical treatments. One way to elicit utilities of a health state based on the QALY model is to use the time trade-off (TTO) method. This method lets respondents consider living a fixed amount of time in some impaired health (typically 10 years) after which death follows and asks them how much of this time they are willing to give up to regain full health (FH). From these indifference between time in impaired health and FH, utility of impaired health can be approximated. For example, if someone expresses to be indifferent between 7 years in FH and 10 years in a wheelchair, the utility of living in a wheelchair is usually taken to be $7/10 = 0.7$. If instead, someone expresses to be indifferent between 10 years in FH and 10 years in a wheelchair, the utility of living in a wheelchair is 1, and both states are valued equally.

The TTO method is one of the main methods the EuroQol group uses in their EQ-5D valuation protocols.^{1,2} In that context,

researchers have shown increasing interest in valuing the health states of children.³ A separate instrument has been developed for this by the EuroQol group, known as EQ-5D-Y-3L,^{4,5} for which a valuation protocol has been published recently.⁶ The instrument is similar to the adult version, albeit with some slight differences in wording (eg, it uses child-appropriate examples for usual activities).

Several empirical results pose challenges to the use of EQ-5D-Y-3L. Importantly, the valuation protocol recommends the use of a child perspective, that is, valuation tasks are completed by adults considering the life of a 10-year-old child. Reasons for this recommendation are that it might be deemed unethical to have children compare health states with being dead and that the valuations tasks might be too cognitively demanding for children.⁷ Recent work has shown that the use of such a child perspective yields higher utilities for children than using the perspective typically used for adult EQ-5D instruments.⁸⁻¹⁰ This may be caused by respondents being less willing to give up time for children than for themselves.¹¹⁻¹³ Nevertheless, evidence for methods that do not rely on trading off lifetime is mixed. For the visual analog scale (VAS), Kind et al¹⁴ found that adults gave lower weights to children's health states than to their own or

other adults' health states, and Lipman et al¹¹ observed no systematic differences.

This study is motivated by potential effects that perspective-dependent utilities may have on priority setting between adults and children, all else being equal. For example, if utilities for the same reported level of problems, as classified according to the adult version of the EQ-5D-3L (eg, state 22222), are higher for children than adults, this means that there is less potential utility gained by moving a child from 22222 to FH than an adult (if utility is normalized as is usual). Consequently, ceteris paribus, this reduced potential utility gain for children because of higher utilities means that, for treatments that improve quality of life from a specific impaired health state to FH, cost-effectiveness ratios would get more favorable for adults relative to children (assuming the same duration of this improvement). In contrast, it implies that, for life-extending interventions with the same durations, cost-effectiveness ratios would get more favorable for children relative to adults (because a child's life for the same reported levels of problems has higher utility than an adult's life for the same reported levels of problems).

Potentially, these implications could be entirely in line with societal preferences, suggesting that EQ-5D-Y-3L utilities elicited with adult and child perspectives can be used to inform policy makers about both the utility component of QALYs, as well as priority setting between adults and children. Yet, the conflicting evidence for TTO and VAS and the hypothesized reluctance to give up years for children in TTO suggest this discrepancy may have other causes. There is no clear evidence if these disparate adult and child TTO utilities are a correct representation of people's preferences for health states, nor if the implications of their differences are in line with how people would allocate resources between adults and children.¹⁵

One test that could be used is to investigate if the TTO utilities, obtained from using the EQ-5D-Y-3L instrument, are able to predict choices in priority setting of specific health improvements of different cohorts of society. That is, if the valuation of the same impaired health state is different for adults than for children, then the utility gain from a treatment that cures the health problem will also be different for adults and children. For example, if the health state "living in a wheelchair" is valued at 0.6 for children and at 0.5 for adults, then a treatment that brings the patient back to FH for 1 year would yield a utility gain of 0.4 for children and 0.5 for adults, assuming FH is valued at 1 for both adults and children. This means that a QALY maximiser would prefer to give such a treatment to the adult rather than the child. Nevertheless, we should keep in mind that although these TTO utilities might be informative in predicting societal choices, they need not be the only determinant. People may also have explicit age-based preferences, which can only be captured by direct comparisons among different age groups, such as adults and children, which are not included in TTO.¹⁵ These kinds of age-based preferences can be regarded as equity weights,¹⁶ which are a way of attributing more or less importance to health benefits achieved in some circumstances relative to others,¹⁷ in which the available evidence comparing children and adults suggests that children are prioritized over adults.^{18,19} Therefore, the health state values obtained with the EQ-5D valuation instrument are not expected to fully correspond to revealed prioritization when not controlling for this kind of equity weighting.

In this article, we test one of the implications of a difference in values for child health states and adult health states, namely, that medical treatments give more gains to adults than to children for the same levels of reported problems. We do this by investigating if respondents' prioritization preferences in the allocation of

scarce healthcare resources to specific medical treatments between children and adults in a person trade-off (PTO) task are consistent with the between-perspective differences in the corresponding health-state utilities as obtained by TTO.

Methods

Model

The PTO method²⁰⁻²² was originally developed to estimate social value of health states, but it can also be used to elicit to distributional weights attached to different groups in society, such as different generations.^{23,24} One possibility to do this is by using a 2-step procedure, in which the utility of a health state is first measured (eg, by a TTO task), followed by the estimation of equity weights by having respondents compare health improvements of 2 groups of people.²⁵ In this study we use the TTO to estimate health state utilities from both the adult and the child perspective and have the same respondents directly compare health improvements of groups of adults and children to estimate equity weights.

We apply a social welfare function, in which the social welfare W of the population is determined by aggregating the total number of QALYs of population groups g , who all get a specific weight α_g . Assuming all members have only one chronic health state in their life (this assumption is not necessary, but simplifies notation and is sufficient for our study, because in all tasks involving prioritization between groups, we only consider lifetimes with a single chronic health state), then W can be evaluated by the following equation:

$$W = \sum_{g=1}^n \alpha_g V(h_g, T_g) \quad (1)$$

in which $V(h_g, T_g)$ is the total utility of the considered period T_g spent in health state h_g , and n denotes the total number of groups in the society.

In the priority setting task, we let the respondent compare a group of 10-year-old children to a group of adults of the respondents' age, each receiving the same health improvement from h to FH lasting for 10 years. If we denote the utility of the 10-year-old children by $U_C(h)$, the utility of the adult by $U_A(h)$, $N = A$ the number of adults getting the treatment and $N = C$ the number of 10-year-old children, then we get the following equation:

$$10 \times \alpha_C \times [U(FH) - U_C(h)] \times C = 10 \times \alpha_A \times [U(FH) - U_A(h)] \times A \quad (2)$$

in which α_C is the weight given to children and α_A is the weight given to adults.

The utilities $U_C(h)$ and $U_A(h)$ can be elicited in the first step, for which we use a TTO task with either adult or child perspectives in our study. Usually, the linear QALY model is assumed in applications of TTO:

$$V(h, T) = T \times U(h), \quad (3)$$

in which $U(h)$ is the utility of health state h . The TTO method asks respondents for the number of years $T = X$ in FH such that they are indifferent to 10 years in h . This generates the following equation:

$$X \times U(FH) = 10 \times U(h). \quad (4)$$

Applying the usual scaling of $U(FH) = 1$, we obtain: $U(h) = X/10$. If a health state is considered worse than dead (WTD), the WTD

procedure is started, in which 10 years of lead time are added to the years of disease time. Indifference then yields:

$$U(h) = (X - 10)/10. \quad (5)$$

If we apply the estimates of $U_C(h)$ and $U_A(h)$ derived from the TTO task and again scale $U(FH)$ to 1 (this assumption is not necessary, but it simplifies notation and is sufficient for our study because in all tasks involving prioritization between groups, we only consider lifetimes with a single chronic health state), then we can solve for the ratio α_C / α_A , ie, the relative weight given to children vs adults:

$$\frac{\alpha_C}{\alpha_A} = \frac{[1 - U_A(h)]A}{[1 - U_C(h)]C} \quad (6)$$

It readily follows that this is equal to A/C if $U_C(h) = U_A(h)$. Alternatively, if $\alpha_C = \alpha_A$, then any difference between A and C would be due to age-specific differences in $U(h)$, as is evident when we solve Eq. (6) for A/C while setting $\alpha_C = \alpha_A$:

$$\frac{A}{C} = \frac{1 - U_C(h)}{1 - U_A(h)} \quad (7)$$

Therefore, we can use Eq. (7) to test if different equity weights are given to health improvements in children as opposed to health improvements in adults. As is common in applications of a health-related social welfare function,^{26,27} we assume that $\alpha_C = 1 - \alpha_A$, which gives the following:

$$\alpha_C = \frac{(1 - U_A(h))A}{(1 - U_C(h))C + (1 - U_A(h))A} \quad (8)$$

Appendix A in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002> shows how such a test would work using the answers of a fictive respondent to one of our PTO tasks as an example.

Based on previous literature on age preferences, there is reason to expect a preference for young vs old. For instance, several studies reported more weight to be given to younger persons in case of life saving examples,²⁸⁻³⁰ whereas others found a similar result for quality-of-life improvements.³¹⁻³³ Explanations for this include higher productivity of younger people,^{34,35} the older people having already earned their “fair innings”^{36,37} and higher potential health gains because of a higher life expectancy³⁸ (if the study did not control for this). In this study, we investigate if such a preference, if replicated, can (partly) be attributed to perspective-dependent TTO utilities.

Experiment

Design and participants

Ethical approval for this study was given by the Ethics Review Committee of Erasmus School of Health Policy & Management on 25 February 2021 (number 21-004). Two trained interviewers performed video interviews with $n = 150$ Dutch citizens, each lasting between 25 and 60 minutes. These participants were recruited by Dynata, a survey company that has a large panel of respondents representative of the Dutch adult public in terms of age, gender, education, and geographic spread. Before starting the main experiment, we tested the software extensively ourselves. The interviewers then both performed 3 pilot interviews. Based on these 2 steps we made modifications to the operationalization. Furthermore, the interviewers clarified any uncertainty with respect to the task.

Respondents valued 4 health states with TTO and VAS using the perspective recommended in the valuation protocol for

EQ-5D-Y-3L⁶; that is, we asked these (adult) subjects to value health states considering a 10-year-old child. They also valued these states on behalf of someone else of the same age as the respondent. We used this other-adult perspective to ensure that the utilities can be used in PTO tasks (which are typically operationalized with a veil of ignorance). Furthermore, the use of another perspective prevents a change in perspective between the 2 tasks (ie, own vs other), which has been shown to affect valuations.¹¹ By asking adults to imagine another adult their own age, we are also able to control for potential variance related to age-related priority setting effects.³⁹

The EQ-5D-Y-3L consists of the dimensions “mobility,” “looking after oneself,” “doing usual activities,” “having pain or discomfort” and “feeling worried, sad, or unhappy.” Each of these dimensions is reported on with 3 levels, varying from no problems to a lot of problems.⁵ We denote a health state by a 5-digit-number, representing the level on each dimension corresponding to this health state (for instance, if someone has moderate problems walking about, no problems in selfcare, severe problems with usual activities, moderate pain, and being severely anxious, their health state would be denoted by 21323).

We valued the following 4 health states with both VAS and TTO: 11312, 22222, 32323, and a coma. The first 3 states were selected to cover a wide spectrum of severity. Furthermore, states 11312 and 22222 were also included in Kreimeier et al,⁸ facilitating comparison with their results. The state coma was included because it was part of one of the PTO robustness tasks (see section 2.2.5 and Appendix B in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002>) and was not described according to the EQ-5D-3L-Y classification system but as being unconscious. The order of the VAS and TTO blocks was randomized, just as the order of the perspectives within each block, and the order of the health states within each perspective. The order of the health states that a respondent received was always the same for the different tasks.

Introduction of experiment

The experiment started by having participants fill out some demographic characteristics (gender, age, and education), after which they completed the EQ-5D-Y-3L instrument to allow respondents to familiarize themselves with its descriptive system. Next, they filled in 2 practice VAS and TTO tasks with a 10-year duration, from the perspective of someone else of their age. This was done with a wheelchair example (adapted from Stolk et al¹), which was implemented to show respondents both the better than dead (BTD) and WTD procedure.

TTO

TTO was operationalized with the standard 10-year duration. The composite TTO was used, which uses the standard TTO procedure once it is determined that a health state is perceived to be BTD and a lead-time TTO for a health state that is regarded to be WTD. The lead time in FH was 10 years, which is commonly used in EQ-VT valuations.⁴⁰ We implemented a bisection elicitation procedure with 6 choices (the experiment was programmed in Shiny, and a demo version can be tried out here: <https://referencepoints.shinyapps.io/KeuzesOverGezondheid/>). A bisection procedure zooms in on an indifference value by taking the midpoint of an interval, which is decreasing in each consecutive choice as more information about the preference structure becomes available.⁴¹ For example, if 10 years in the impaired health states was preferred to death, then the duration in FH was increased to 5 years in the second choice (the midpoint between 0 and 10 years) and then to 2.5 years if 5 years in FH was preferred

or to 7.5 years if 10 years in the impaired health state was preferred. Repeating this procedure up to 6 times yields indifference at a precision of 0.5 years.

VAS

In principle, we could also have used VAS data for testing if perspective-dependent valuation reflects priority setting. The VAS is a horizontal or vertical rating scale where a respondent can evaluate health states where the endpoints are labeled “the best health you can imagine” and “the worst health you can imagine.”⁴² The value of h was taken to be the number it was assigned on the rating scale, divided by 100. Nevertheless, because dead was not valued on this scale, it was not anchored to the usual utility scale with $U(\text{dead}) = 0$ and $U(\text{FH}) = 1$. As such, all health states got a positive utility, without necessarily being regarded as BTD . Hence, comparability with the results of the TTO task is hampered by this limitation, and we caution against assigning too much weight to our results for this task. We therefore decided to report the results of the VAS task in Appendix C in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002> and focus on the TTO results in the main article. The same 4 health states as in the TTO task were valued on the VAS, again for both the adult-other and the child-other perspective. This enabled us to test if the VAS gave different predictions for the PTO task than the TTO, as formulated in hypothesis C1.

Priority setting preference elicitation with PTO tasks

Respondents completed 5 PTO tasks in random order (Table 1). Priority setting preferences were measured by 3 of these tasks, in which respondents had to choose between treating a group of 10-year-old children and a group of people of their own age (but not including themselves). Respondents considered a health improvement from an imperfect EQ-5D-Y-3L state, h_1 , to another state h_2 that dominated h_1 (ie, being better in at least 1 dimension and the same in all other dimensions). The patients' health would be improved for a period of 10 years, after which health restored to its original level. The second approach we used was a PTO task with lives saved,²⁴ in which we implemented a remaining life duration of 20 years for those whose lives were saved (PTO4). The advantage of this task is that it is not affected by potentially heterogeneous health-state valuations, while it also allowed for comparing views on priority setting between quality-of-life-improving treatments to life-enhancing treatments. The final PTO task was included as robustness check and involved an improvement from coma to FH for 10 years (PTOR). This task was added to test if priority setting would be similar if a state was used that we expected to differ less between the adult and child perspective and is described in Appendix B in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002>. Specifically, a coma was described as being unconscious but with the possibility to return to FH when awaking; hence, we expected this state to be valued about the same as death, that is, getting a utility of 0 for both adults and children.

Indifferences in the PTO tasks were elicited as follows. In the initial question, both groups consisted of 100 people, after which a bisection procedure of 5 more questions followed, where the number of patients of one of the groups was varied. After these questions, an indifference point was estimated to be the midpoint of cutoff points inferred from the binary choices (The respondents also had the opportunity to be indifferent in each question, in which case the iteration procedure stopped immediately, and the last value that appeared on the screen was stored as the indifference value). For instance, if the respondent chose to treat 100 persons of their own age in the first question, then the next

Table 1. Information of specific health state transition and duration used in each PTO task.

Question	Health improvement
PTO1	32323 to 11111 for 10 years
PTO2	11312 to 11111 for 10 years
PTO3	32323 to 11312 for 10 years
PTO4	Death to 11111 (lifesaving) for 20 years
PTOR	Coma to 11111 for 10 years

PTO indicates person trade-off

question would be 50 patients of their own age versus 100 patients of 10 years old. If they instead chose to treat 100 patients of age 10 years, then the next question would be 50 patients of age 10 versus 100 patients of their own age. After each question, the indifference region was narrowed down by taking the midpoint of 2 cutoff values, which was rounded to integers if needed. To illustrate this further, a possible choice sequence could be (chosen options in quotation marks) as follows: 1. 100A vs “100C”; 2. 100A vs “50C”; 3. “100A” vs 25C; 4. 100A vs “38C”; 5. “100A” vs 32C; 6. 100A vs “35C,” after which the estimated indifference value would be $(32+35)/2 = 33.5$. This meant that this respondent gave $100/33.5 = 2.99$ times as much weight to children as to adults (see Appendix D in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002> for all possible response options and their implied weighting of preference for prioritizing children vis-à-vis adults).

At the end of the interview, we asked some demographic questions (religiousness, whether they had children, and their annual gross household income, see Appendix E in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002>). Furthermore, we asked for their expected age of death, which was found to be related to TTO values for the adult perspectives in previous studies^{43,44} but not for the child perspective in another study.⁴⁵ We repeated this question for a 10-year-old child. Finally, we gave the respondents 3 statements related to their wider views about priority setting between adults and children, as measured on a Likert scale (1 = don't agree at all; ...; 7 = fully agree) (the complete instructions and some screenshots of the different tasks are provided in Appendix E in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002>).

Statistical Analysis

The PTO data were first analyzed while assuming $U_A = U_C$. Eq. (8) then changes into the following:

$$p_c = \frac{A}{A+C} \quad (9)$$

In which p_c denotes the priority-setting ratio that does not allow for perspective-dependent health-state utilities. For respondents who favored treating 100 children over 100 adults in the first question, A would be 100 and C would be between 0 and 100, yielding $0.5 < p_c < 1$. Respondents favoring the treatment of 100 adults over 100 children would have $C = 100$ and $0 < A < 100$, giving $0 < p_c < 0.5$. Hence the variable p_c is symmetrical around 0.5, with respondents $0.5 < p_c < 1$ prioritizing treatments to 10-year-old children in the PTO task, whereas respondents with $0 < p_c < 0.5$ prioritize treatments to adults. A downside of this variable is that it is not linearly related to the intensity of preferences. For

Table 2. Summary statistics of background variables.

Variables	Percentage	Mean	SD	Quartile 1	Quartile 3
Age		51.8	15.20	40	64.25
Gender					
% Male	48.0				
% Female	51.3				
% Other	0.7				
Education*:					
Lower	17.4				
Middle	45.6				
Higher	36.9				
Health status: VAS		77.3	14.35	70	90
Expected age of own death		83.5	5.78	80	86
Expected age of death of child of 10 years		88.1	8.51	83	93
Has children	60.1 (61.3)	0.61	0.49		
Is religious	39.9 (40.7)	0.42	0.50		
Likert scale question about priority to children over adults		5.24	1.49	4	7
Likert scale question about importance of equal healthcare access		5.45	1.56	4.75	7
Likert scale question about importance of longevity relative to quality of life for a child		3.57	1.70	2	5

VAS indicates visual analog scale.

*Lower education: elementary school or prevocational secondary education; middle education: secondary vocational education or upper-level secondary school; high education: higher professional education or university.

instance, if someone is indifferent between treating 20 children and 100 adults, $p_c = 0.83$. If for someone else treating 40 children is equivalent to treating 100 adults, then their $p_c = 0.71$. Therefore, we first transform this measure using the function $\pi(\cdot)$, bounded between -100 and 100 , where -100 (100) reflects a preference for always treating the group of adults (children) irrespective of the size of each group. We obtain this measure by taking 100 minus the answer C given by those respondents giving more weight to children than to adults. For respondents giving more weight to adults than to children, we take their answer A minus 100 (see Appendix D in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002> for all possible answers). We estimate the mean of the answers on this scale and report the p_c corresponding to this mean.

We continue with estimating the equity weight α_c of Eq. (8), based on the answers given in the PTO task and the utilities of the health states, as estimated in the TTO. We test the effect of differential adult and child utilities on priority setting by comparing α_c and p_c . We do so for each PTO task using paired t tests, except for the lifesaving task, which only involved death and FH and hence was not influenced by any adult-child differences in health-state utilities (recall that we assumed death and FH to be valued the same for adults and children).

With these estimates we test the following null hypotheses:

Hypothesis 1: equity weights are the same for adults and children: $\pi(\alpha_c) = \pi(\alpha_A) = 0$ (t tests).

Hypothesis 2: priority setting is not affected by differential utilities for adults' and children's health states: $\pi(\alpha_c) = \pi(p_c)$ (paired t tests) (note that is equivalent to a test of $U_A[h] = U_C[h]$).

Hypothesis 3: equity weights are not affected by the health states used; ie, they do not differ between the first 3 PTO questions: $\pi(\alpha_c[\text{PTO1}]) = \pi(\alpha_c[\text{PTO2}]) = \pi(\alpha_c[\text{PTO3}])$ (paired t tests and Friedman test).

Hypothesis 4: equity weights are the same for allocations involving quality-of-life improvement and allocations involving life

extensions: $\pi(\alpha_c[\text{PTO1}]) = \pi(\alpha_c[\text{PTO4}])$, $\pi(\alpha_c[\text{PTO2}]) = \pi(\alpha_c[\text{PTO4}])$, $\pi(\alpha_c[\text{PTO3}]) = \pi(\alpha_c[\text{PTO4}])$ (paired t tests).

Regression analysis

We performed mixed effects regressions with subject random effects and fixed effects for the PTO task, with the transformation π of p_c as dependent variable:

$$\pi(i, q) = \delta_q + \text{PTO}_q \beta' + x_i \gamma' + \varepsilon_{i,q} \quad (10)$$

In which i (1-150) is the subject number, q (1,2,3,4) is the number of the PTO question, δ_q is a constant reflecting PTO4, PTO_q is a matrix containing the question dummy for PTO, x_i is a matrix containing the other variables (gender, age, own health rating, education, children, religion, subjective life expectancy of children and adults, TTO utilities for adults and children, order of PTO and TTO/VAS, and the Likert scale questions), and $\varepsilon_{i,q}$ is an error term.

Results

Summary statistics and data quality

Table 2 presents summary statistics of our sample. We performed a data quality check⁴⁶ for the TTO and VAS, as shown in Appendix F in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002>. Looking at the PTO data, we found that 56% gave more weight to the young in all 5 tasks, whereas 3.3% always gave more weight to the old. The highest (lowest) possible value of p_c was reached in 15.7% (2.4%) of the responses, and 8% (0%) of the respondents always chose to treat the young (old). Furthermore, the PTO weights were positively correlated with the answers given to the Likert scale question on priority setting for children ("When the budget for medical treatment is limited, children should receive priority over adults" [1 = don't agree at all;

Table 3. Mean utilities of TTO values from adults and children (SD in parentheses).

Health state	TTO Adults	TTO Children
11312	0.68 (0.32)	0.70 (0.30)
22222	0.75 (0.29)	0.73 (0.24)
32323	0.24 (0.60)	0.20 (0.61)
Coma	-0.43 (0.41)	-0.42 (0.38)

TTO indicates time trade-off.

...; 7 = fully agree) for all 5 tasks (Kendall's τ , P 's < .01), suggesting that the PTO data reflect respondents' attitude toward trade-offs between treating adults and children.

Health-state valuations

Table 3 presents statistics on the health-state valuations. The TTO utilities are similar for adults and children, contrary to the usual finding of higher utilities for children. The differences are insignificant for all health states (P 's > .25).

PTO Tasks

Table 4 reports the ratios p_C and α_C corresponding to the means of $\pi(\cdot)$. Below, we describe the hypothesis test results.

Hypothesis 1 (equity weights are the same for adults and children: $\pi(\alpha_C) = \pi(\alpha_A) = 0$). For all PTO questions, $\pi(\alpha_C)$ was higher than 0 ($P < .01$ for all), rejecting Hypothesis 1.

Hypothesis 2 (priority setting is not affected by differential utilities for adults and children: $\pi[\alpha_C] = \pi[p_C]$). Formal tests indicate that the differences between α_C and p_C were not significant for PTO2 ($P = .17$), but they did differ for PTO1 and PTO3 ($P < .01$), partly rejecting Hypothesis 2.

Hypothesis 3 (equity weights are not affected by the health states used: $\pi[\alpha_C(\text{PTO1})] = \pi[\alpha_C(\text{PTO2})] = \pi[\alpha_C(\text{PTO3})]$). A Friedman test revealed that the transformed weights $\pi(\alpha_C)$ were different between the 3 quality-of-life-improving tasks when using TTO values ($P < .01$), rejecting Hypothesis 3 when correcting for differential health state values for adults and children as measured with the TTO task. Testing task-by-task, PTO1-TTO and PTO2-TTO did not differ ($P = .85$), but PTO3-TTO gave significantly lower transformed weights than PTO1-TTO and PTO2-TTO (P 's < .01).

Hypothesis 4 (equity weights are the same for allocations involving quality-of-life improvement and allocations involving life extensions: $\pi[\alpha_C(\text{PTO1})] = \pi[\alpha_C(\text{PTO4})]$, $\pi[\alpha_C(\text{PTO2})] = \pi[\alpha_C(\text{PTO4})]$, $\pi[\alpha_C(\text{PTO3})] = \pi[\alpha_C(\text{PTO4})]$). Comparing the transformed equity weights from the lifesaving PTO tasks (PTO4) to the other 3 PTO tasks, we find that weights were higher in the lifesaving task (P 's < .02 for all). This implies Hypothesis 4 can be rejected, with stronger equity weighting for lifesaving treatments than quality-of-life-improving treatments.

The results of the mixed effects regressions are reported in Table 5. Model I only includes the PTO dummies and the order variable, Model II adds the demographics to model I, finally, Model III also includes the TTO utilities. Two of the 3 PTO question dummies involving health improvements (PTO1 and PTO3) are significant at $\alpha = 5\%$. This highlights that the specification of the health states is a relevant criterion in priority setting. Age and the dummies for middle and high education have a significant positive coefficient, indicating that older and more educated people give more weight to the treatment of children than younger and less educated people, respectively. Of the TTO utilities, only the utilities of health state 32323 are significant, with the sign for adult utilities being negative, whereas it is positive for child utilities. This means that respondents who value state 32323 better for adults give less weight to children in the PTO tasks and vice versa for children. This result is contrary to the theoretical prediction of our model because the utility gain when the health of the group of adults is improved from 32323 to a better health state is lower if the utility of 32323 is higher, making the treatment of the group of children more attractive. A similar reasoning holds for child utilities, in which a higher utility of state 32323 would be expected to make treating children less attractive.

Discussion

Earlier work has shown that TTO utilities for EQ-5D-Y-3L health states valued by adults taking a child's perspective are generally higher than their valuations of the same state for themselves.^{8,9} Our objective was to replicate these differences and test if they can explain views on priority setting between treatment of adults and treatment of children. In this section, we will discuss the results of this explorative study, as well as alternative interpretations of these results (often related to the limitations of our experiment). We observed small differences between valuations in child and adult perspective in the TTO task. This result deviates from the observations of Kreimeier et al⁸ and Dewilde et al,⁹ who found considerably larger differences, with child valuations higher than adult valuations. One alternative explanation could be that we used the other perspective for both the valuation of the adults' and the valuation of the children's health states, whereas Kreimeier et al⁸ used the self-perspective for adults and the other perspective for children, and Dewilde et al⁹ used the self-perspective for both adults and children (Appendix Table G1 in Supplemental Materials found at <https://doi.org/10.1016/j.jval.2023.08.002> in the supplemental material provides a comparison of the design and average utilities for the Dutch respondents of these 2 studies [which were kindly shared by the authors of these studies after personal communication] with ours). Hence, our choice of perspective may be considered a limitation of the study, and a potential explanation for the (lack of) differences observed here and in other work suggests that utilities elicited with tasks in which adults decide for themselves. It implies that our results cannot be straightforwardly compared with

Table 4. Equity weights measured in the PTO task (ratios of means of $T(p_C)$ and $T(\alpha_C)$, SD in parentheses). PTO task

Weight	32323-11111 (PTO1)	11312-11111 (PTO2)	32323-11312 (PTO3)	Death - 11111 for 20 y (PTO4)	Coma-11111 (PTOR)
p_C	0.65 (0.68)	0.67 (0.67)	0.65 (0.68)	0.70 (0.66)	0.61 (0.73)
α_C	0.63 (0.71)	0.64 (0.75)	0.55 (0.82)	0.70 (0.66)	0.60 (0.74)

PTO indicates person trade-off.

Table 5. Results of mixed effects regression on $\pi(p_c)$ elicited from PTO tasks.

Dependent variable: $\pi(p_c)$ derived from PTO task Model	Coefficient (SD in parentheses)		
	I	II	III
Constant	0.741 (0.046)*	0.057 (0.252)	-0.148 (0.272)
Order [†]	0.001 (0.030)	0.023 (0.031)	0.019 (0.032)
Dummy PTO1 32323-11111 (reference: PTO4 Death-11111)	-0.037 (0.019) [‡]	-0.033 (0.020)	-0.033 (0.020)
Dummy PTO2 11312-11111	-0.028 (0.019)	-0.024 (0.020)	-0.024 (0.020)
Dummy PTO3 32323-11312	-0.049 (0.019) [§]	-0.045 (0.020) [‡]	-0.045 (0.020) [‡]
Interviewer dummy		0.026 (0.030)	0.046 (0.030)
EQVAS Own health today		0.002 (0.001)	0.002 (0.001)
Female (reference: male)		0.055 (0.034)	0.049 (0.035)
Age		0.004 (0.001)*	0.004 (0.001)*
Middle education [¶] (reference: low education)		0.089 (0.041) [‡]	0.085 (0.044)
High education [‡] (reference: low education)		0.102 (0.042) [‡]	0.105 (0.044) [‡]
Religious (reference: not religious)		0.000 (0.029)	0.005 (0.030)
Has as at least one child (reference: no children)		-0.002 (0.033)	0.006 (0.034)
Income (in categories)		0.011 (0.010)	0.010 (0.010)
PRIORITY_CHILDREN**		0.042 (0.011)*	0.043 (0.011)*
ACCESS ^{††}		0.015 (0.009)	0.019 (0.009) [‡]
ONGLIFE ^{††}		0.001 (0.010)	0.004 (0.010)
Expected age of death adults		-0.003 (0.004)	-0.000 (0.004)
Expected age of death child		0.000 (0.003)	-0.001 (0.003)
TTO adult utility 11312			0.070 (0.073)
TTO adult utility 22222			0.041 (0.071)
TTO adult utility 32323			-0.108 (0.038) [§]
TTO adult utility Coma			-0.093 (0.049)
TTO child utility 11312			-0.023 (0.072)
TTO child utility 22222			-0.070 (0.074)
TTO child utility 32323			0.088 (0.037) [‡]
TTO child utility Coma			0.046 (0.055)

Log restricted likelihood: model I: 107.508; model II: 65.114; model III: 54.483. Wald Chi squared: model I: 7.55, $P = .109$; model II: 64.84, $P < .001$; model III: 79.95, $P < .001$
PTO indicates person trade-off; TTO, time trade-off; VAS, visual analog scale.

*Significant at the 0.1%-level.

[†]Order: order of PTO and TTO/VAS.

[‡]Significant at the 5%-level

[§]Significant at the 1%-level.

^{||}EQVAS: own health measured with a VAS.

[¶]Middle education: education level higher than elementary school or prevocational secondary education, but less than higher vocational education.

[‡]High education: higher vocational education or university.

**PRIORITY_CHILDREN: "when the budget is limited, children should receive priority over adults."

^{††}ACCESS: "I think it is important that everyone has equal access to healthcare, if that means fewer life will be saved in total."

^{††}ONGLIFE: "I think it is important that a sick child can live as long as possible, even if it lowers their quality of life."

tasks in which adults decide for someone else (ie, another adult or child).¹¹ The potential effects of using self- and other-perspectives may have implications on comparisons and transitions between existing EQ-5D value sets that should be explored in future work. Nevertheless, our results on the child health states considered in isolation are still different from the results of Kreimeier et al,⁸ although they also used the other perspective for this part.

Another explanation for the different findings is that, although we used a bisection procedure to elicit indifferences, Kreimeier et al⁸ used the EQ-5D titration procedure. The difference between these procedures is that the titration procedure does not have a fixed number of choices; instead, the number of years in FH is increased or decreased in fixed steps until the respondent switches from the one

option to the other. This may result in different valuations, eg, because of strategic answering to reduce effort.⁴⁷ Moreover, a recent study¹¹ used a bisection procedure in a student sample and also found much smaller differences in TTO values between perspectives. More research is needed to further investigate this potential limitation of our study, for example, by performing a within-subject comparison of the bisection and titration procedure. Replication studies in other countries are also encouraged to test if our findings may have been country specific because our study relied solely on Dutch respondents, which limits the external validity of our results. Note that such studies may also consider using a different duration for TTO tasks. In our study we stuck to the durations recommended in the EQ-5D-Y-3L valuation protocol, but the 10-year duration (and lead time)

causes part of the TTO gauge duration to involve adult years when a child perspective is used.¹⁵

Given this lack of disparity between the 2 perspectives, it was hardly surprising that these valuations could not explain the priorities expressed in the PTO. Nevertheless, the priority given to children in the PTO was so strong that this could probably also not be explained by individual valuations if we would have found a difference between adult and child valuation. In fact, the typical finding of higher valuations for child health states than for adult health states predicts the opposite from our results, that is, more priority should then be given to treatment of adults than treatment of children. The results from this first experimental study on EQ-5D-Y-3L utilities and priority setting provide little evidence in favor of simply aggregating QALYs, and external equity weights might be considered to incorporate intergenerational fairness concerns (we assumed throughout that the utility of FH was the same for adults and children. A preference to treat children rather than adults in a PTO task may however also be the consequence of the utility of FH being valued higher for children than for adults. Therefore, future research is recommended that tests this assumption by asking the public to directly compare a fixed period in FH for an adult and for a child).

This finding is in line with earlier findings of a higher weight being given to health gains in children than in adults^{23,24,39,48} and to the more general findings of age-related equity weighting.^{28,31,38,49,50} Such equity weights might be implemented by first specifying a formal criterion for incorporating equity concerns into economic evaluations⁵¹ (eg, the proportional shortfall or end-of-life premiums have been used in some countries^{19,52,53}) and then eliciting the weights for which sophisticated methodology, often embedded in social welfare theory, has already been proposed.^{32,39,54-57} Nevertheless, our finding of different equity weights for different health states and for quality-of-life improvements vs lifesaving treatments (different valuations of QALY gains that involve life extensions than equally sized QALY gains involving quality-of-life improvements have been found in several previous studies.⁶⁰⁻⁶³), could be interpreted as suggesting that these weights may be context-specific; hence, the derivation of such external equity weights is not straightforward. Note that the variance between quality-of-life improvements vs lifesaving treatments may also be related to the relatively small sample size and the small number of tasks included in this first experimental study. Furthermore, for 2 of the 3 quality-of-improving PTO tasks, we found that the implied equity weights were affected by the perspective-dependent TTO utilities. This suggests that, in equity measurement exercises, such as the ones performed here, it may be unadvisable to assume that utilities are the same for children and adults, keeping in mind the proof-of-concept nature of our study.

In line with the recommended perspective for valuation of EQ-5D-Y-3L, we asked respondents to complete the valuation task considering a 10-year-old child. One may argue that the age of the child imagined may affect valuation because childhood, including what aspects of health are considered important, may differ a lot by age. Indeed, Reckers-Droog et al¹³ concluded that health-state preferences for a 10-year-old child may not be representative of preferences for the full EQ-5D-Y-3L age range, which would imply that the reliance on only 10-year-old children is a limitation of our study. Yet, Ramos-Goñi et al⁵⁸ suggest that little effect of age difference in children was found in influencing differences between perspectives in the US sample, whereas some differences were found between the own perspective and the 5 to 7 years old perspective in the UK sample, and the 5 to 7 years old perspective and 8 to 10 years old perspective during EQ-5D-Y-3L valuation. Given these mixed findings, we recommend future studies to replicate our study using different childhood ages.

Finally, this study collected data using video interviews, and at this stage the equivalence of video and in-person interviews is not yet clear.⁵⁹

Conclusions

In this study we tested if differences in adult- and child-perspective health-state valuations obtained by TTO were related to priority-setting decisions when budget must be allocated between children and adults. Unexpectedly, and potentially related to the limitations of this first explorative study, we found no significantly different health utilities between these perspectives. Still, perspective-dependent variation in health-state valuation could partly explain priority setting between adults and children in PTO questions. Separately elicited equity weights that reflect the relative weight given to children vis-à-vis adults may however be considered to supplement health-state utilities to make judgments that better reflect societal preferences for healthcare allocations. In other words, simply aggregating QALYs elicited with different perspectives does not seem to reflect society's preferences in trade-offs between children and adults in the case of scarce healthcare interventions.

Author Disclosures

The authors reported no conflicts of interest. The views expressed in this paper are those of the authors and do not necessarily reflect the views of the EuroQol group.

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