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Preference-Based Assessments

Does Changing the Age of a Child to be Considered in 3-Level Version of EQ-5D-Y Discrete Choice Experiment–Based Valuation Studies Affect Health Preferences?

Juan M. Ramos-Goñi, PhD, Anabel Estévez-Carrillo, MSc, Oliver Rivero-Arias, DPhil, Donna Rowen, PhD, David Mott, PhD, Koonal Shah, PhD, Mark Oppe, PhD

ABSTRACT

Objectives: There has been some debate about the choice of perspective and the age of the child considered when completing preference elicitation tasks in the 3-level version of EQ-5D-Y (EQ-5D-Y-3L) valuation protocol. This study aimed to clarify the impact on latent scale EQ-5D-Y-3L values of varying the age of the child experiencing the health state considered by respondents completing the discrete choice experiment (DCE) tasks of the protocol.

Methods: We conducted an online DCE with a representative sample of 1000 adults in the United Kingdom and 1000 adults in the United States. Respondents selected the health state they prefer from a series of DCE paired EQ-5D-Y-3L health state comparisons using their own perspective and that of a hypothetical child from the following age groups: "5-7 years old," "8-10 years old," "11-13 years old," and "14-15 years old." Data analysis was conducted using separate multinomial logit models for each perspective and country. We also estimated combined models including data from each possible pair of perspectives and used interactions between EQ-5D-Y-3L levels and perspective to determine whether any differences were statistically significant.

Results: No statistically significant differences in coefficients between perspectives were found in the United States. In the United Kingdom, there were differences between the own perspective and the 5 to 7 years old perspective (looking after myself level 3) and between the 5 to 7 years old perspective and the 8 to 10 years old perspective (usual activities level 3).

Conclusions: Our results suggest that there is minimal impact on latent scale values when using different ages of the hypothetical child in the current EQ-5D-Y-3L valuation protocol.

Keywords: discrete choice experiment, EQ-5D-Y, health-related quality of life, perspective, value set.

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Introduction

In the last 20 years, the incorporation of health-related quality of life (QOL) in the form of utilities has become an integral part of economic evaluations of medical treatments. The most widely used instrument to obtain health-related QOL utilities is the EQ-5D.¹ The EQ-5D is a generic instrument based on 5 dimensions: mobility, self-care, usual activities, pain/discomfort, and anxiety/depression. It has different versions including the 3-level version of EQ-5D (EQ-5D-3L), which has 3 response levels per each dimension (no problems, some, and unable/extreme), and the 5-level version of EQ-5D (EQ-5D-5L), which has 5 response levels per each dimension (no problems, slight, moderate, severe, and unable/extreme); finally, it also includes a visual analog scale. Sets of utility values for the health states described by the EQ-5D-3L and EQ-5D-5L instruments (referred to as "value sets"), which facilitate the estimation of

quality-adjusted life-years (QALYs), are available for a wide range of different countries.^{2,3}

In 2010, the EuroQol Group introduced the EQ-5D-Y, a version of the EQ-5D for younger populations aged 8 to 15 years.⁴ This instrument adapted the wording of dimensions and its levels from the original EQ-5D-3L, leading to the following dimensions—mobility, looking after myself, usual activities, pain/ discomfort, and worry/sad/unhappy—and the following levels: no, some/a bit, and a lot/very for the levels. The visual analog scale remained as part of the instrument. After the development of the instrument, a methodological research program was initiated to determine the most appropriate methods for generating a value set for this 3-level version of EQ-5D-Y (EQ-5D-Y-3L) instrument.⁵⁻⁷ This culminated in an international protocol to develop EQ-5D-Y-3L value sets, which has recently been published.⁸ Several research teams have completed or are currently undertaking valuation and methodological studies following this guidance.⁹⁻¹¹

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This valuation protocol, in a similar fashion to the protocol for EQ-5D-5L valuation studies,¹² recommends the use of a discrete choice experiment (DCE) and a composite time trade-off (C-TTO) to elicit preferences for EQ-5D-Y-3L health states. The idea is that the DCE informs the relative importance of attributes and levels on a latent scale and that the only role of the C-TTO is to anchor the latent scale DCE model on the QALY scale (ie, full health = 1; dead = 0).⁷ The EQ-5D-Y-3L valuation protocol suggested that adult respondents should be asked to state their preferences with respect to EQ-5D-Y-3L health states in the context of a hypothetical 10-year-old child.

Since the publication of the EO-5D-Y-3L valuation protocol. there has been some debate about the choice of perspective (10year-old child) and the implications of completing the elicitation task considering a child of different ages.¹³ During research reviewing the current literature about child health valuation,¹⁴⁻¹⁶ questions have been raised about the appropriateness of adopting this 10-year-old perspective on the EQ-5D-Y-3L valuation protocol. The perspective can be challenged on the basis of child perspective versus own perspective^{6,17} and also the age of the child within the child perspective. This is important in costeffectiveness models measuring benefits using QALYs in conditions affecting length of life and QOL from childhood into adulthood. The perspective of a 10-year-old child was selected for historical reasons; that is, it was used in the earlier studies exploring EQ-5D-Y-3L values,^{18,19} which was informed from previous literature.^{20,21} As far as we are aware, there is no solid evidence available about this to help decision makers make an informed decision about when a child perspective is selected or whether the specific age of 10 years is appropriate for the EQ-5D-Y-3L value set. To gain an understanding of the impact of changing the perspective within the current protocol, there is a need for an in-depth exploration of how the 2 components of the EQ-5D-Y-3L valuation protocol, namely, relative importance of dimensions (DCE) and scale anchor (C-TTO), behave when varying the age of the child experiencing the health state.

In this study, we evaluated the following research question: What is the impact on relative importance of dimensions (latent scale) for an EQ-5D-Y-3L value sets of varying the age of the child experiencing the health state considered by respondents completing the DCE tasks of the protocol?

Methods

Experimental Design

The DCE design described in the international EQ-5D-Y-3L valuation protocol included 150 pairs. Nevertheless, our study included a within-respondent design among perspectives and the original 15-pair/10-block design would have translated into 75 pairs (15×5) for each respondent, which was considered infeasible. Therefore, to reduce respondent burden while maintaining the within-respondent design, we reduced the original design. We used instead a 100-pair design per perspective, which was considered sufficient to model DCE data including the estimation of interaction terms. In other words, 100 pairs were enough for estimating a model including 20 parameters. Within each pair of the design, 2 attributes were kept at the same level (overlap) for each of the 2 choice options, to reduce the cognitive burden to respondents.²²

Participants responded to each DCE pair from their own perspective and from the perspective of a child in the following 4 age groups: "5-7 years old," "8-10 years old," "11-13 years old," and "14-15 years old." Therefore, each respondent completed each DCE pair from 5 different perspectives. The same 100-pair design

was used across each of the 5 perspectives to avoid any differences in models results because of differences in the underlying designs.

Each respondent completed 5 pairs \times 5 perspectives = 25 pairs in total. Within respondents, these 5 pairs were the same for the 5 perspectives (to avoid any differences in the models results for perspective because of differences in possible interactions between respondents and the design). This implied there were 100 pairs/5 pairs per respondent = 20 blocks of 5 pairs for each perspective. Each respondent was randomly assigned to 1 block that included the same 5 pairs for each of the 5 perspectives. The 25 pairs were randomized such that (1) the order of the perspectives seen by respondents was randomized and (2) the order of the pairs within each perspective was also randomized (see Fig. 1).

The 100 pairs that were included in the DCE were selected based on a simulation study, using data from a recent EQ-5D-Y-3L DCE study in Spain.¹¹ This Spanish study used the DCE design described in the international protocol article, which includes 150 pairs.⁸ We simulated 5000 DCE subdesigns, of 100 pairs each. For each of the 5000 subdesigns, we estimated the DCE model and its predictions using the Spanish responses and then calculated the error versus the predictions of the whole EQ-5D-Y-3L protocol DCE design (150 pairs, ie, the whole data set). The subdesign with the lowest prediction error was selected as the final subdesign to be used for this study. Therefore, in terms of prediction accuracy, the final subdesign was the closest possible design to the existing DCE design from the international EQ-5D-Y-3L protocol.

Quality Control

Three dominant pairs, where all dimension levels in one health state were logically better than the second health state, were added to the survey. Each respondent answered one dominant pair question using the first, third, and fifth perspective shown, in a random position between the other 5 pairs in each of these perspectives. These dominant pairs were added to allow for assessment of respondent engagement and were the same for each respondent. Therefore, all respondents completed 25 + 3 = 28 pairs in total.

The quality control for the data collection followed 2 rules to determine whether a respondent was sufficiently engaged while completing the survey. Based on previous experience, using EQ-5D-Y-3L and overlap design, we have estimated that respondents are able to complete each DCE task in approximately 8.5 seconds.¹¹ Based on this, we first excluded speeders—respondents who did not invest at least 4 minutes in completing the 28 DCE tasks. Second, we excluded respondents who incorrectly answered at least 2 of the 3 dominant pairs (ie, chose the dominated health state).

Survey Instrument and Implementation

Respondents completed the following sections:

- 1. Respondent information page explaining the aim of the project and consent form. If a respondent did not provide consent, the survey finished immediately and a message thanking the respondent for considering participating was shown.
- 2. Respondent demographic questions including geographical location, age, and gender to delimit the quotas defined to ensure the sample representativeness and 3 questions related to experience with illness
- 3. Self-completion of the EQ-5D-Y-3L instrument as a warm-up task, that is, to get familiarized with the instrument
- 4. An instructions page on how to complete the DCE and informing the participant that questions would be asked for

different perspectives (please see Appendix Material 1 in Supplemental Materials found at https://doi.org/10.1016/j.jval.2 022.03.001 for screenshots of the presentation)

- 5. 28 paired comparisons: 5 pairs for each of the 5 perspectives plus 3 dominant pairs. A pop-up alerts respondents when they were switching between perspective (please see Appendix Material 1 in Supplemental Materials found at https://doi. org/10.1016/j.jval.2022.03.001 for screenshots of the presentation).
- 5 debrief and additional background questions related to their perceptions on the adult/child perspectives and their experience with children with serious illness (see Appendix Material 2 in Supplemental Materials found at https://doi.org/10.1016/j. jval.2022.03.001 for details)
 - The left-right positions of the health states within a pair were also randomized to avoid potential left-right bias of respondents. To mitigate the occurrence of nonattendance, differences between choice options were emphasized using bold fonts.²²

Sample Size and Respondent Recruitment

A total of 50 observations per pair (per perspective) were considered enough^{23,24} to reliably estimate the discrete choice models, leading to a total sample size of 20 blocks \times 50 observations per block = 1000 respondents.

We aimed to collect data from 1000 respondents in the United Kingdom and 1000 respondents in the United States. An internet panel company in each country invited participants to complete the online survey. The panel company sent invitations to their panelist including a specific link, and the panelist chooses whether to access the survey and is free to exit the survey at any time. For the United Kingdom, quota representation was sought in terms of age (18 years and older), gender, and nation (England, Scotland, Wales, and Northern Ireland). For the United States, quotas included age, gender, and geographical location, according to the US census. Quotas were hard implemented in the survey, thereby ensuring representativeness.

Respondents included in the final sample received points from the survey company as a reward for their participation.

Statistical Analysis

Sample characteristics were described using proportions for categorical variables and mean and SD for age. We estimated conditional logit models using the clogit Stata command. We separately estimated models for each perspective in each country. Using the model coefficients, we calculated the relative weights of dimensions (relative attribute importance) in percent terms and plotted the results in a bar chart. Finally, to test whether the differences observed in the descriptive analysis were significant, we compared perspectives by estimating a model, which included only data from 2 perspectives (eg, own perspective vs "5-7 years old"), and we added interactions terms between the EQ-5D-Y-3L levels of each dimension and one of the 2 perspectives in comparison.

Formally, if n represents the respondent and j an option, the utility of the respondent n for the option j is modeled as

 $U_{nj} = V_{nj} + \varepsilon_{nj}$

where V_{nj} represented a linear decomposition of the utility of respondent n of the health state j and ε_{nj} represented the error in valuing that utility. Therefore, it is assumed that when a respondent chooses i over j, its utility for i is higher than the utility for j. Then the probability for option i can be estimated as follows: $P_{ni} = \Pr(\varepsilon_{nj} - \varepsilon_{ni} < V_{nj} - V_{ni}) \forall j \neq i.$ For the 2 by 2 perspectives comparison, we used the following decomposition of V:

$$V_i = b_1 MO2_i + b_2 MO3_i + b_3 SC2_i + b_4 SC3_i + b_5 UA2_i$$

 $+b_6UA3_j+b_7PD2_j+b_8PD3_j+b_9AD2_j+b_{10}AD3_j$

 $+b_{11}(MO2_j x Perspective_k) + b_{12}(MO3_j x Perspective_k)$

 $+b_{13}(SC2_i x Perspective_k) + b_{14}(SC3_i x Perspective_k)$

 $+b_{15}(UA2_i \times Perspective_k) + b_{16}(UA3_i \times Perspective_k)$

 $+b_{17}(PD2_i x Perspective_k) + b_{18}(PD3_i x Perspective_k)$

 $+b_{19}(AD2_i x Perspective_k) + b_{20}(AD3_i x Perspective_k)$

When comparing perspective k and m, $Perspective_k$ represented a dummy variable taking a value of 1 when the response was for the perspective k and 0 when it was for the perspective m. Note that only data from perspectives k and m were included.

We conducted these pairwise analyses between perspectives for each of the 5 perspectives, leading to $\begin{pmatrix} 5\\2 \end{pmatrix}$ = 10 models per country. To correct for multiple testing, we applied a Bonferroni correction. This allowed us to compare any statistically significant differences between relative importance of dimensions/levels among any 2 perspectives by looking to the 10 *P* values of the interaction terms between the perspective and the 10 main effects model coefficients. To facilitate the presentation of these results, we only report the *P* values.

For all models, the linear part consisted of using the responses to the DCE (1/0 expressing whether option A was selected instead of B) as the dependent variable and either regular or incremental dummies for levels 2 and 3 per each dimension as the independent variables. We used regular dummies (movements from level 1 for both levels 2 and 3 per each dimension) for the descriptive analysis as the rescaled 0 to 1 results express the relative importance of dimensions just looking at the movement between levels 1 and 3 of the coefficients. For the statistical tests, we used incremental dummies, because we wanted to test movements between consecutive levels, in other words, not only looking at the relative importance of dimensions but also at the relative importance of all dimensions and levels.

When we compare 2 perspectives using DCE model as explained earlier, it is important to check each perspective model first. This is necessary to evaluate whether the scale plays a role in any differences found. In other words, the DCE results include both relative importance of levels/dimensions and scale. The scale is related to level of agreement between respondents (the closer observed choice probabilities to 100%, the larger will be the scale).²⁴ Therefore, when the scale is different between perspectives, it not possible to determine whether any observed significant differences are due to differences in scale or relative importance. In contrast, when the scale among perspectives is similar, significant differences are only because of differences in relative importance.

Results

We collected a sample of 1245 respondents in the United Kingdom and 1358 respondents in the United States. Of these

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Table 1. Sample characteristics.

Variables	United Kingdom		United States	United States			
	QC excluded* (n = 233)	Estimation (n = 1012)	QC excluded* (n = 323)	Estimation (n = 1035)			
Age, mean (SD)	36.27 (12.3)	49.66 (17.41)	39.45 (12.13)	49.7(16.73)			
Age groups (%) 18-24 25-29 30-39 40-49 50-59 60-69 70+	17.17 14.16 36.91 15.45 9.01 6.01 1.29	7.81 7.71 14.53 17.79 19.76 19.17 13.24	5.57 8.67 46.13 22.29 8.98 4.95 3.41	4.25 5.41 27.34 16.14 12.46 15.56 18.84			
Gender (%) Male Female Other Prefer not to say	66.95 33.05	49.7 50.2 0.1	59.13 40.56 0.31	48.7 51.01 0.19 0.1			
Have a child (%) 0-4 years old 5-7 years old 8-10 years old 11-13 years old 14-15 years old 16 years or older	64.38 19.7 15.9 20.6 19.7 6.9 12.9	58.79 6.4 6.9 7.7 7.6 4.9 40.4	64.71 15.2 22.3 23.5 19.5 10.8 8.1	55.75 11.6 76.0 15.4 12.4 6.2 26.1			
Experience with illness Personal (%yes) Relatives (%yes) Others (%yes)	37.34 53.22 38.2	30.43 61.76 27.67	45.82 41.49 28.17	30.43 43.47 24.93			
Self-reported EQ-5D-Y (%) Mobility (walking about) No problems Some problems A lot of problems Looking after myself No problems Some problems A lot of problems Doing usual activities No problems Some problems A lot of problems Having pain or discomfort No pain or discomfort Some pain or discomfort Feeling worried, sad, or unhappy Not worried, sad, or unhappy Very worried, sad, or unhappy Very worried, sad, or unhappy VAS, mean (SD)	66.95 23.18 9.87 72.10 21.03 6.87 67.38 23.61 9.01 56.65 33.48 9.87 51.50 31.76 16.74 68.54 (19.46)	80.43 16.40 3.16 91.21 7.41 1.38 76.38 19.27 4.35 54.74 40.02 5.24 49.70 40.12 10.18 72.04 (19.16)	66.95 23.18 9.87 72.10 21.03 6.87 67.38 23.61 9.01 56.65 33.48 9.87 51.50 31.76 16.74 68.54 (19.46)	80.48 15.46 4.06 90.43 7.54 2.03 80.58 16.33 3.09 53.62 39.13 7.25 59.03 34.40 6.57 72.04 (19.16)			
Exclusion criteria Nonspeeders and \geq 2 inconsistencies Speeders and $<$ 2 inconsistencies Speeders and \geq 2 inconsistencies QC indicates quality control; VAS, visual analog scale.	36.5 36.0 27.5		46.1 28.2 25.7				

respondents, 233 and 323 respondents in the United Kingdom and the United States, respectively, were removed from the analysis based on the quality control rules (fast completers and those failing ≥ 2 of the 3 dominance tests). Therefore, our final representative samples sizes (quotas were achieved as described earlier) consisted of 1012 in the United Kingdom and 1035 in the United States (Table 1). From the included respondents, 58.79% in

the United Kingdom and 55.75% in the United States were parents/ guardians and had at least one child.

Models per perspective and country are presented in Table 2. Results have been rescaled to a scale ranging between 0 and 1 to facilitate comparability between the model parameters and to express the relative importance of dimensions in percent terms. The scales for each model are reported at the bottom of the table. The

Table 2. Model coefficients rescaled to 0-1 and their original scale by perspective and country.

Variable (regu	ılar dummies)	United S	tates				United Kingdom					
		Own	5-7 years old	8-10 years old	11-13 years old	14-15 years old	Own	5-7 years old	8-10 years old	11-13 years old	14-15 years old	
Mobility (walking about)	No problems to some problems	0.042	0.054	0.062	0.050	0.053	0.087	0.027	0.094	0.058	0.056	
	No problems to a lot of problems	0.165	0.141	0.156	0.142	0.162	0.185	0.120	0.168	0.152	0.148	
Looking after myself	No problems to some problems	0.043	0.028	0.017	0.022	0.032	0.039	0.019	0.036	0.027	0.044	
No problems to lot of problems	No problems to a lot of problems	0.139	0.106	0.095	0.133	0.132	0.165	0.104	0.130	0.123	0.147	
Doing usual No problems to activities some problems	0.065	0.064	0.064	0.074	0.066	0.080	0.088	0.086	0.092	0.082		
	No problems to a lot of problems	0.193	0.171	0.169	0.196	0.193	0.173	0.177	0.194	0.177	0.190	
Having pain or discomfort	No to some No to a lot of	0.099 0.272	0.134 0.299	0.119 0.285	0.095 0.246	0.125 0.268	0.097 0.257	0.146 0.331	0.110 0.272	0.128 0.303	0.122 0.279	
Feeling worried, sad, or unhappy	Not to a bit Not to very	0.051 0.230	0.073 0.283	0.089 0.295	0.077 0.283	0.081 0.246	0.068 0.220	0.083 0.269	0.096 0.235	0.082 0.246	0.072 0.236	
Range of the latent scale*		[1;-7.55]	[1;-5.65]	[1;-7.73]	[1;-6.62]	[1;-6.70]	[1;-4.59]	[1;-4.26]	[1;-4.38]	[1;-4.60]	[1;-4.88]	

Note. P values of the comparison are reported in Table 3.

*Larger range indicates higher level of agreement between respondents' responses.

analyses showed some differences between coefficients after rescaling in both countries. In addition, the scale in the United Kingdom was similar between perspectives meaning that the level of agreement between respondents did not differ among the 5 perspectives. Nevertheless, in the United States, the level of agreement in preferences regarding the 5-7 years old child perspective was the lowest among all perspectives, whereas for the own and the 8- to 10-year-old child it was the highest (Table 2).

The relative importance of dimensions per perspective also showed some differences (Fig. 2). The mobility dimension seemed to be more important for the own perspective than for any of the child perspectives in the United Kingdom, whereas this is not the case in the United States. Having pain/discomfort seemed to be more important for the youngest age group (5-7 years old) than for any other age group in both countries. In the United States, the dimension "looking after myself" is less important in the younger age groups from 5 to 10 years, whereas in the United Kingdom this is only the case for the youngest age group (ie, 5-7 years old).

In the confirmatory analysis, when running the models including interactions comparing 2 by 2 perspective and after a Bonferroni correction, all P values are nonsignificant in the United States suggesting there is no evidence in this setting that preferences differ when varying the age of the child (Table 3). Four P values were significant in the United Kingdom. Two significant P values were shown when comparing the own perspective with the youngest age range (5-7 years old); those were related to the movement from no problems to some problems in the mobility dimension, making it 7% lower on the own perspective, and related to the movement from "some problems" to "a lot of problems" in the looking after myself dimension, making it 6% lower in the own perspective. The other 2 significant P values were shown when comparing the 5- to 7-year-old group with the 8- to 10-year-old group, and they were related to the movements from "no problems" to "some problems" in the mobility dimension, making it 10% lower in the 5- to 7-year-old group and related to the movement from "some problems" to "a lot of problems" in

the doing usual activities dimension, making it 6% lower in the 5to 7-year-old group.

That there were only 4 of a possible 100 significant differences in modeling results in the United Kingdom and none in the United States was in line with the responses to the debrief question: "Please indicate whether you agree or disagree with the following statement: My answers to the questions changes when the age of the child described as experiencing the health states changed." Notably, 37% in the United Kingdom and 31% in the United States stated that they responded differently if the age of the child changed (Table 4). When asked about improving the health of children or adults, 32% of respondents in the United Kingdom and 40% of respondents in the United States stated that improving the health of a child should be given higher priority than improving the health of an adult, and most respondents stated that both children and adults should be given the same priority (Table 4).

Discussion

"What perspective is appropriate in an EQ-5D-Y-3L valuation protocol?" is a broad question that should be discussed from normative, ethical, methodological, and empirical angles. Other authors have covered normative discussions, and in their conclusions, they have suggested further research including the aim of this study.¹³ In particular, we have concentrated on assessing the relative importance differences because of perspective changes, rather than differences in the anchor on the (1) full health and (0) dead scale. Therefore, this study has not explored the normative arguments around selecting the age of the child nor assessed the impact on time trade-off values. Further research on these issues is warranted. This study has investigated whether relative preferences for health state descriptions affecting children are different when the adult undertaking the online survey is asked to imagine different child ages in a sample of respondents from the United States and the United Kingdom.

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Figure 1. Experimental design.



The published valuation protocol for the EQ-5D-Y-3L suggested using a 10-year-old child as the perspective for valuing the health states described by the instrument.² Our results suggest that choosing a different child age for respondents to consider when completing the elicitation task has minimal differences between DCE modeled latent scale values across the perspectives used: "5-7 years old," "8-10 years old," "11-13 years old," and "14-15 years old." In addition, no substantial differences were found when the respondents considered the health states to apply to themselves (ie, "own" perspective). Therefore, we would recommend that there is no immediate need to change the perspective in the DCE side of the current protocol in the United Kingdom and United States. Nevertheless, given the observed differences between the published EQ-5D-Y-3L value sets in Japan Slovenia and Spain,⁹⁻¹¹ it is possible that the impact of different child ages and perspectives may vary across countries. This brings up the need for testing this approach in different cultures and languages.



Figure 2. Relative importance of dimension by perspective and country.

	The Second Process										
(incremental dummies)											
	Own vs 5-7 years old	Own vs 8-10 years old	Own vs 11-13 years old	Own vs 14-15 years old	5-7 years old vs 8-10 years old	5-7 years old vs 11-13 years old	5-7 years old vs 14-15 years old	8-10 years old vs 11-13 years old	8-10 years old vs 14-15 years old	11-13 years old vs 14-15 years old	
Mobility (walking about) No problems to some problems Some problems to a lot of problems	.754 .063	.520 .144	.781 .188	.594 .768	.743 .693	.973 .596	.828 .121	.717 .891	.912 .246	.801 .310	
Looking after myself No problems to some problems Some problems to a lot of problems	.456 .321	.227 .372	.347 .571	.711 .721	.645 .922	.845 .122	.709 .18	.791 .147	.405 .214	.571 .835	
Doing usual activities No problems to some problems Some problems to a lot of problems	.783 .177	.843 .184	.632 .765	.766 .766	.939 .985	.454 .297	.570 .102	.502 .306	.623 .106	.855 .554	
Having pain or discomfort No to some Some to a lot of	.186 .324	.445 .481	.818 .278	.076 .301	.579 .781	.122 .923	.655 .964	.323 .708	.315 .746	.046 .959	
Feeling worried, sad, or unhappy Not to a bit A bit to very	.373 .445	.074 .394	.171 .185	.067 .832	.373 .929	.637 .575	.352 .332	.675 .638	.972 .290	.648 .126	
Interaction term $ imes$ perspective	United Kingdom										

interaction term ~ perspective	onited Kingdom										
(incremental dummies)	Own vs 5-7 years old	Own vs 8-10 years old	Own vs 11-13 years old	Own vs 14-15 years old	5-7 years old vs 8-10 years old	5-7 years old vs 11-13 years old	5-7 years old vs 14-15 years old	8-10 years old vs 11-13 years old	8-10 years old vs 14-15 years old	11-13 years old vs 14-15 years old	
Mobility (walking about) No problems to some problems Some problems to a lot of problems	.000* .066	.612 .150	.047 .304	.039 .295	.000* .695	.092 .422	.104 .433	.015 .682	.012 .696	.947 .985	
Looking after myself No problems to some problems Some problems to a lot of problems	.085 .000*	.897 .081	.283 .014	.997 .041	.119 .057	.524 .218	.087 .096	.354 .496	.895 .789	.285 .673	
Doing usual activities No problems to some problems Some problems to a lot of problems	.263 .070	.578 .199	.969 .204	.553 .825	.101 .002*	.254 .594	.598 .044	.610 .012	.258 .290	.534 .140	
Having pain or discomfort No to some Some to a lot of	.316 .100	.211 .721	.214 .570	.361 .110	.806 .050	.814 .291	.923 .946	.991 .366	.731 .056	.739 .317	
Feeling worried, sad, or unhappy Not to a bit A bit to very	.648 .352	.024 .502	.798 .575	.723 .668	.008 .800	.483 .715	.916 .617	.048 .912	.009 .807	.546 .894	

Note. This table reports the P values of the comparisons of the coefficients from Table 2.

*Significant results are only the ones < 005 because of the Bonferroni correction (please see Methods section).

The absence of significant results across all perspectives comparison in the United States is in agreement with the lower percent of respondents stating that they did change their responses when the age of the child changed. In addition, having different levels of agreement, implying different preference strengths (and therefore scales) between perspective makes it more difficult for the models to capture significant results. Although in the United Kingdom there were similar levels of agreement between the perspectives, the scales of the models per perspectives were similar. Therefore, the analyses were more capable of capturing differences in the relative importance of dimensions. Additionally, the significant differences in the United Kingdom involved 5-7 and 8-10 years old perspectives and may indicate that there could exist a lower limit for the age of the child where below this limit health preferences differ. Nevertheless, this limit seems to be younger than 5 years old, which is beyond the age range recommended for the EQ-5D-Y-3L instrument.

This study is not exempt of limitations. The differences in the relative importance of dimensions shown in the descriptive analysis in the United States could be confounded by the differences in the levels of agreement within the sample for the different perspectives. In addition, there is a limitation because of the internal consistency of the DCE. In other words, when a

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Table 4. Background questions.

	United Kingdom (%)	United States (%)
Should priority be given to improving the health of children, adults or neither? Children should be given higher priority Adults should be given higher priority Neither, both children and adults should be given the same priority Please indicate whether you agree or disagree with the following statement: My answer to the questions changes when the age of the child described as experiencing the health states changed.	31.92 2.37 65.71	39.9 6.18 53.92
Agree Neither agree nor disagree Disagree	26.88 36.17 36.97	37.00 32.75 30.25
Do you have a degree or equivalent qualification? No Yes	47.23 52.77	33.56 66.44
Which of the following descriptions best describes your main activity? Employed or self-employed Retired Student Looking after home or family Long-term sick or disabled None of the above	54.54 25.79 4.35 7.02 4.45 3.85	56.53 28.50 2.13 2.22 10.62
Has any child known to you ever experienced serious illness? Yes—but I do not know that child very well Yes—and I have a close relationship with that child No Would rather not say	10.57 13.14 74.51 1.78	14.49 16.23 67.54 1.74

respondent completes the same DCE, some variation in the responses would be are expected.^{25,26} This means that even when we repeat the DCE without modifying the perspective we would expect to see some differences between elicited values. Although this may limit the interpretation of the study results, its impact on the conclusions is only important when differences are shown. Therefore, in our case, given that we have found that changing the age of the child only produced very limited significant results, we cannot ensure that the differences observed are due to the change on the perspective and not due to the consistency of the DCE itself.

Another limitation was the potential link between complexity of the study design and respondent fatigue. Having the same 5 questions from 5 different perspective could lead to fatigue and random choices. To minimize the impact of this limitation, we randomized the order in which the perspectives were presented. Therefore, the potential noise caused by limitation is shared across all perspectives (see Appendix Material 3 in Supplemental Materials found at https://dx.doi.org/10.1016/j.jval.2022.03.001). In addition, we emphasized both the perspective being completed using bold and bigger font size and all changes between perspectives using pop-up boxes to highlight that the perspective had changed.

There is also the possibility that perspective did not lead to differences in which health state from the pair was preferred out of the DCE pairs shown, but that this does not imply that there are not differences in how respondents value states when the perspective changes. Therefore, different results may be found when using different elicitation methods such as time trade-off or potentially using different pairs within a DCE. The use of existing panels to recruit respondents raises concerns of representativeness of nonobservable characteristics because this method only includes respondents who are part of internet panels. Nevertheless, this does not necessarily imply that this has affected our results. Finally, qualitative research has examined the impact of perspectives²⁷ but the impact of different age of the child within the child perspective was not explored. Nevertheless, our study is a quantitative study that has not assessed qualitatively whether respondents regard health states differently for different age perspectives, and research assessing this would be able to examine further whether the results observed here reflect peoples underlying preferences.

Conclusions

Our results suggest that changing the age of the child imagined in DCE tasks designed to generate an EQ-5D-Y-3L value is unlikely to affect elicited values. Therefore, there is no clear need to change the currently adopted perspective in the EQ-5D-Y-3L valuation protocol on the basis of this evidence. Similarly, a change in the age of the child due to, for example, normative reasons, could be safely implemented, if necessary, without concerns in the resulting latent scale elicited values.

Supplemental Materials

Supplementary data associated with this article can be found in the online version at https://dx.doi.org/10.1016/j.jval.2022.03.001.

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REFERENCES

- Kennedy-Martin M, Slaap B, Herdmann M, et al. Which multi-attribute utility instruments are recommended for use in cost-utility analysis? A review of national health technology assessment (HTA) guidelines. *Eur J Health Econ.* 2020;21(8):1245–1257.
- EQ-5D-5L, validation: standard value sets. EQ-5D. https://euroqol.org/eq-5dinstruments/eq-5d-5l-about/valuation-standard-value-sets/. Accessed XXX.
- EQ-5D-3L, validation. EQ-5D. https://euroqol.org/eq-5d-instruments/eq-5 d-3l-about/valuation/. Accessed XXX.
- Wille N, Badia X, Bonsel G, et al. Development of the EQ-5D-Y: a childfriendly version of the EQ-5D. Qual Life Res. 2010;19(6):875–886.
- Kreimeier S, Oppe M, Ramos-Goñi JM, et al. Valuation of EuroQol fivedimensional questionnaire, youth version (EQ-5D-Y) and EuroQol five-

dimensional questionnaire, three-level version (EQ-5D-3L) health states: the impact of wording and perspective. *Value Health*. 2018;21(11):1291–1298

- Mott DJ, Shah KK, Ramos-Goñi JM, Devlin NJ, Rivero-Arias O. Valuing EQ-5D-Y-3L health states using a discrete choice experiment: do adult and adolescent preferences differ? *Med Decis Making*, 2021;41(5):584–596.
- Shah KK, Ramos-Goñi JM, Kreimeier S, Devlin NJ. An exploration of methods for obtaining 0 = dead anchors for latent scale EQ-5D-Y values. Eur J Health Econ. 2020;21(7):1091–1103.
- Ramos-Goni JM, Oppe M, Stolk E, et al. International valuation protocol for the EQ-5D-Y-3L. *Pharmacoeconomics*. 2020;38(7):653–663.
- Prevolnik Rupel V, Ogorevc M, IMPACT HTA HRQoL Group. EQ-5D-Y value set for Slovenia. *Pharmacoeconomics*. 2021;39(4):463–471.
- Shiroiwa T, Ikeda S, Noto S, Fukuda T, Stolk E. Valuation survey of EQ-5D-Y based on the international common protocol: development of a value set in Japan. *Med Decis Making*. 2021;41(5):597–606.
- Ramos-Goñi JM, Oppe M, Estévez-Carrillo A, et al. Accounting for unobservable preference heterogeneity and evaluating alternative anchoring approaches to estimate country-specific EQ-5D-Y value sets: a case study using Spanish preference data. ViH. In press.
- Oppe M, Devlin NJ, van Hout B, Krabbe PF, de Charro F. A program of methodological research to arrive at the new international EQ-5D-5L valuation protocol. *Value Health*. 2014;17(4):445–453.
- Lipman SA, Reckers-Droog VT, Kreimeier S. Think of the children: a discussion of the rationale for and implications of the perspective used for EQ-5D-Y health state valuation. *Value Health*. 2021;24(7):976–982.
- Mulhern B, Norman R, Street DJ, Viney R. One method, many methodological choices: a structured review of discrete-choice experiments for health state valuation. *Pharmacoeconomics*. 2019;37(1):29–43.
- Rowen D, Rivero-Arias O, Devlin N, Ratcliffe J. Review of valuation methods of preference-based measures of health for economic evaluation in child and adolescent populations: where are we now and where are we going? *Pharmacoeconomics*. 2020;38(4):325–340.
- Lenny A, Shah K. PNS208 a review of the methods used in valuation studies of child/adolescent health-related quality of life using EQ-5D-Y and CHU-9D. *Value Health.* 2020;23(suppl 2):S676.
- Prevolnik Rupel V, Ramos-Goñi JM, Ogorevc M, Kreimeier S, Ludwig K, Greiner W. Comparison of adult and adolescent preferences toward EQ-5D-Y-3L health states. *Value Health*. 2021;24(9):1350–1359.
- Kind P, Klose K, Gusi N, Olivares PR, Greiner W. Can adult weights be used to value child health states? Testing the influence of perspective in valuing EQ-5D-Y. *Qual Life Res.* 2015;24(10):2519–2539.
- Craig B, Greiner W, Brown DS, Reeve BB. Valuation of child health-related quality of life in the United States. *Health Econ.* 2016;25(6):768–777.
- Torrance GW, Feeny DH, Furling WJ, Barr RD, Zhang Y, Wang Q. Multiattribute utility function for a comprehensive health status classification system: Health Utilities Index Mark 2. *Med Care*. 1996;34(7):702–722.
- **21.** McCabe C, Stevens K, Roberts J, Brazier J. Health state values for the HUI 2 descriptive system: results from a UK survey [published correction appears in Health Econ. 2006 Jul;15(7):761]. *Health Econ.* 2005;14(3):231–244.
- Jonker MF, Donkers B, de Bekker-Grob E, Stolk EA. Attribute level overlap (and color coding) can reduce task complexity, improve choice consistency, and decrease the dropout rate in discrete choice experiments. *Health Econ*. 2019;28(3):350–363.
- Lancsar E, Louviere J. Conducting discrete choice experiments to inform healthcare decision making. *Pharmacoeconomics*. 2008;26(8):661–677.
- Hensher DA, Rose JM, Greene WH. Applied Choice Analysis: A Primer. Cambridge, United Kingdom: Cambridge University Press; 2005.
- Purba FD, Hunfeld JAM, Timman R, et al. Test-retest reliability of EQ-5D-5L valuation techniques: the composite time trade-off and discrete choice experiments. *Value Health*. 2018;21(10):1243–1249.
- Gamper EM, Holzner B, King MT, et al. Test-retest reliability of discrete choice experiment for valuations of QLU-C10D health states. *Value Health*. 2018;21(8):958–966.
- 27. Powell PA, Rowen D, Rivero-Arias O, Tsuchiya A, Brazier JE. Valuing child and adolescent health: a qualitative study on different perspectives and priorities taken by the adult general public. *Health Qual Life Outcomes*. 2021;19(1):222.