



Views, obstacles, and uncertainties around the inclusion of children and young people's time in economic evaluations: Findings from an international survey of health economists

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ABSTRACT

People's time is a limited resource and, in economic evaluations that adopt a societal perspective, it is important that it is valued and accounted for. Yet, in economic evaluations of interventions for children and young people (CYP), attempts to take into account the opportunity cost of their time are rare. To understand why this is the case, we need to first understand what views health economists hold in relation to CYP time, and what challenges they face in incorporating this in their evaluations.

We planned and carried out an international survey of health economists. We used a combination of approaches to identify potential survey respondents (the survey's sampling frame), we developed a questionnaire that sought to capture respondents' views and practice through close- and open-ended questions, we piloted the questionnaire through a series of cognitive interviews, and we e-mailed unique links to the final version of the questionnaire to 1956 individuals in the sampling frame. We analysed data using quantitative (descriptive and inferential statistics) and qualitative (thematic analysis) methods.

We received 274 complete responses. Most respondents (87%) believe CYP time should be considered for inclusion in economic evaluations conducted from a societal perspective. However, they identify a number of obstacles to doing so, most prominently uncertainties around appropriate practice (e.g., when CYP's time should or should not be included in calculations), methodological gaps (e.g., what value to attach to CYP's time), and practical difficulties in measuring displaced time in CYP. Reporting on their own practice, most respondents found it challenging to consider CYP time in their studies, and stressed the need for clear guidance on when, and further research on how, to appropriately account for CYP's time in economic evaluations. We offer our views on how to move the topic forwards and make suggestions further research.

1. Introduction

Time, its allocation across different activities and its value have attracted considerable interest in social sciences (Gronau, 1977; Pentland et al., 1999). Being a fundamental and limited resource, time is a factor that impacts, directly or indirectly, on numerous decisions made by individuals and households (Becker, 1965; Grossman, 1972). Naturally, there has been a steady stream of research on various aspects of the topic, including on ways to record how people use their time (Gershuny and Sullivan, 2019), what preferences and determinants influence time allocation decisions (DeSerpa, 1971) and, increasingly, what (monetary)

value can be attached to people's time. Research on the latter has been actively pursued in different fields of inquiry—including education, transportation and health economics—often to support economic evaluations assessing the desirability of policies or programmes that are likely to release or take up more of an individual's time (Mackie et al., 2001; Shaw, 1992; Koopmanschap and van Ineveld, 1992).

In health care, economic evaluations—typically in the form of cost-effectiveness and cost-utility analyses—are increasingly carried out to inform funding decisions about care offered to adults and children (Drummond et al., 2015). There, all-encompassing evaluations conducted from a societal perspective should, in principle, account for a

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range of inputs, including patients' time (Drummond et al., 2005; Neumann et al., 2016). If, for example, the process of receiving care results in a patient giving up (additional) time which could have been spent in other activities (e.g., work or leisure), this time should be measured, assigned a value and included in calculations (Dranove, 1996; Gold et al., 1996; Posnett and Jan 1996). Thus, in cases where an evaluated programme of care takes up less of a person's time (e.g., due to a shorter procedure, quicker examination or less frequent appointments) and imposes a lower time-related cost to the individual than its comparator, ignoring time inputs would overlook these gains and provide an incomplete assessment of the programme's value (Russell, 2009; van den Berg et al., 2017).

In economic evaluations, time can be an input to an assessed intervention (e.g., time spent receiving care or engaging with a health-promoting activity), as well as an output of such activities (e.g., less time spent in ill health or extended survival due to the intervention) (Gold et al., 1996; Drummond et al., 2015; Neumann et al., 2016). Debates on how best to measure, value and account for time inputs have led to insights and recommendations (Brouwer et al., 1998; Krol and Brouwer, 2015; Sendi and Brouwer, 2004; Zhang et al., 2011) that have found their way into applied economic evaluations (Krol et al., 2013; Tranmer et al., 2005). However, these insights focus, almost exclusively, on working-age adults. Children and young people's (CYP, here defined as individuals up to 18 years of age) time also has an opportunity cost (Posnett and Jan 1996; Neumann et al., 2016), reflecting the loss of benefit or satisfaction from not being able to use this time for other activities (e.g., education, engaging with hobbies and leisure activities, socialising etc (Mullan, 2020)). However, attempts to account for this in applied work are rare and inconsistent (Andronis et al., 2019). To understand why this is the case, one needs to first understand health economists' views in relation to CYP's time and its inclusion in economic evaluation.

With this in mind, we carried out an international survey aiming to gather and explore researchers' views, practices, perceived obstacles and uncertainties in relation to CYP's time in economic evaluations. The remainder of this paper is structured as follows. In the next section, we give the broader context of this study and describe the methods we used to carry out the reported survey. This is followed by a summary of findings, with a particular focus on respondents views on uncertainties and gaps in existing methods. Drawing on these findings, we highlight key messages, discuss challenges and pinpoint areas for further research.

2. Methods

2.1. Survey approach and identification of potential participants

We planned and carried out a list-based web survey through a proprietary survey platform (Qualtrics XM, Provo, UTH, USA). List-based web surveys invite individuals in an existing or newly compiled list of potential respondents (sampling frame) to participate in an online survey by completing an electronic questionnaire (Fricker, 2017). Such surveys offer notable advantages over other common survey approaches (e.g., unrestricted self-selected surveys or intercept surveys): they allow the inclusion of potential respondents who cannot be reached through membership in mailing lists, they give greater control over who can access the survey, they allow sending targeted reminders, and they avoid reliance on external parties' willingness to distribute survey invites or host the survey link on their website (Fricker, 2017; Callegaro et al., 2015). However, list-based surveys can be notably more time consuming than other types of surveys, largely as they require assembling a sampling frame of potential respondents when one is not available.

The survey's target population was defined as individuals familiar with the general methodology used when undertaking economic evaluations in health care. As these are commonly researchers who identify as health economists, we use this term to denote our survey invitees, acknowledging that individuals in the target population may not

identify as health economists or individuals who identify as health economists may not be familiar with the general methodology of economic evaluation. A complete list of individuals in our target population is not available, thus the survey's sampling frame needed to be constructed by identifying as many individuals in the population as possible. To mitigate the chance of coverage error, we used a multi-modal approach to searching for individuals and their publicly available contact information (Alvarez and VanBeselaere, 2005). First, we searched websites of organisations with whom target respondents are likely to be affiliated. These included Departments/Centres/Units in Higher Education Institutions, research centres, professional associations, special interest groups and societies within and outside the UK. Additionally, to identify individuals who may not be affiliated with such organisations, we carried out general searches on the internet through a widely-used generic search engine (Google®), search engines of scholarly literature (Google Scholar® and Semantic Scholar®) and professional networking portals (ResearchGate®, Academia. com®) using the term 'health economist' on its own or in combination with other terms (e.g., research, profile, country name). We complemented internet searches with a bibliographic review of published literature aiming to identify authors of economic evaluations and health technology assessment studies. We searched PubMed and the Paediatric Economic Database Evaluation (PEDE) (Ungar and Santos, 2003), we selected 500 unique articles at random and we extracted information about each article's first and last author and correspondence email addresses. In addition, we carried out searches for authors of economic evaluations in the grey literature (conference papers and proceedings, dissertations and theses, government and official publications, reports, practice guidelines, working papers) through ProQuest®. Last, we listed individuals that the research team knew of (e.g., prominent researchers, researchers with an interest in economic evaluation) as well as individuals suggested by survey participants, who contacted us to suggest that the survey would be of interest to their colleagues. The final list included the name and publicly available contact information (email address) of potential invitees and, where available, information about employing institution or professional affiliation, professional title and country of residence. The project received ethics approval from the University of Warwick (BSREC 25/21–11).

2.2. Questionnaire development

The survey's questionnaire was developed iteratively by two researchers (LA, CM) with feedback and suggestions provided by the wider team of close collaborators (CD, EL, SP). The questionnaire comprised multiple-choice and rating scale questions, often accompanied by open-ended (free-text) questions, typically asking respondents to elaborate on their answers or suggest their own answers if these were not amongst the offered options.

There were six sections in the questionnaire: the first section comprised a set of filtering questions, designed to record information on respondents' self-perceived familiarity with economic evaluation and 'screen out' invitees whose answers suggested, according to pre-determined criteria, that they did not fit the target population. The three subsequent sections asked questions about respondents' views and practices centred around the inclusion of CYP's time in economic evaluation, where CYP was defined as individuals up to 18 years of age (United Nations General Assembly, 1989). The last section asked general questions about the respondent, including affiliation and country of residence (see Electronic Supplementary Material 1 for a printable version of the survey's electronic questionnaire).

As part of the survey development process, we piloted all participant-facing components (invitation, participant information sheet, consent form, questionnaire, prize draw sheet) with a convenience sample of 20 researchers who met the criteria of the target population. We sought feedback on different components of the survey, including the invitation to participate, the participant information leaflet, the consent process

and the survey's questionnaire. We collected feedback through written comments or cognitive interviews: we asked 15 respondents to provide written feedback in 'free-text' boxes while completing the survey and we carried out cognitive interviews with 5 respondents, in the form of 'think aloud' sessions with concurrent probing (Willis, 2004; Collins, 2014). All cognitive interviews were conducted via teleconferencing, recorded, and were subsequently transcribed. With the exception of cognitive interviews, feedback was provided anonymously, to minimise the chance of acquiescent bias (Vannette and Krosnick, 2017).

For the purposes of pre-testing the survey questionnaire, we prompted respondents to provide feedback, keeping in mind Tourangeau's model of the survey response process (Tourangeau et al., 2000). For instance, respondents were prompted to give feedback on whether questions were clear and unambiguous (comprehending the question); whether they were answerable, and information needed to answer them could be easily recalled (retrieving information); whether the questionnaire was free from leading questions and judgment was unobstructed (judgement); whether options in multiple choice questions were appropriate, and 'free text' boxes were provided where apt (response). Additional feedback was sought on the completeness and comprehension of the participant information sheet, which included important information about the survey (purpose, length, anonymity arrangements, data storage and security, ethics review and approval process). Piloting also gave an opportunity to identify potential issues around arrangements related to administering the survey (e.g., platform settings for survey distribution and anonymity, retrieval of responses etc.). All feedback was considered, and most observations were incorporated in the final version of the survey.

Invitations to participate in the survey were sent out via e-mail. The message explained the purposes of the study, asking for their participation and providing a link to a webpage where they could find more information about study (including anonymity arrangements and ethics approval) and, should they wish, participate. Two targeted reminders were sent to invitees who had not initiated or completed the survey, followed by a last reminder notifying invitees of the survey closing date.

2.3. Data analysis

Answers to multiple choice and Likert-type questions giving rise to categorical and ordinal outcome variables were first summarised and examined through frequency tables. Relationships between variables were then interrogated through multi-way contingency tables and regression analysis (Long and Freese, 2006; Hoffmann, 2016). We used logistic regression for binary outcome variables, multinomial logistic regression for categorical variables, and ordered (cumulative) logistic models for ordinal variables (Agresti, 2013). We tested for significance of predictor variables using Wald and likelihood ratio tests.

Answers to open-ended questions, given as comments in free-text boxes, were analysed through thematic analysis, with codes and themes developed inductively (Braun and Clarke, 2006; Coast, 2017). Two researchers (LA, CM) read through all comments and each of them constructed codes and applied them to text independently. Codes were then discussed, modified, and formed the basis for agreeing on a first set of themes. Each researcher subsequently applied the resulting codes and themes to respondents' answers independently and the coded text was examined for discrepancies between coders. The last step involved re-examining the codes and themes, discussing and resolving disagreements, and agreeing on the coding and theme categorisation for each piece of available text.

3. Findings

3.1. Survey responses

Email invitations to complete the survey were sent out to a total of 2027 individuals. Of these invitations, about 4% (71 emails) were not

delivered, leaving an 'effective sample' of 1956 survey invitees, of whom 369 (19%) initiated the survey. Amongst those, 282 invitees (76%) completed and submitted their responses. Eight of the 282 respondents were subsequently 'screened out' based on their answers to screening questions, leaving a set of 274 complete responses (14% of the effective sample).

3.2. Respondent characteristics

In the first part of the survey's questionnaire, respondents were asked questions about demographic characteristics and familiarity with methods of economic evaluation. A detailed list of answers to these questions is given in Table 1.

In brief, if we were to construct a 'typical' respondent, this person would work in academia ($n = 227$, 83% of all respondents), identify as a health economist ($n = 224$, 82%), have a lot of experience conducting economic evaluations in health care ($n = 140$, 51%), be very familiar with the general methodology for conducting economic evaluations ($n = 195$, 71%) and, over the last five years, would have always or almost always worked on or led economic evaluations of health care technologies ($n = 118$, 42%).

3.3. Views around time and its inclusion in economic evaluation

In the main body of the questionnaire, respondents were asked to indicate their agreement with various statements. Respondents were asked to consider an individual's time as an input to (rather than an outcome of) a health-improving intervention (e.g., time spent seeking or receiving care, time spent in health-promoting activities). Answers are summarised in Table 2.

Support for the view that "time is a valuable resource that, in principle, should be considered for inclusion in economic evaluations carried out from a societal perspective" was near-universal, with approximately 98% of respondents ($n = 268$) indicating that they strongly agree or somewhat agree with this statement. Similar levels of agreement (96%) were observed in relation to the statement "adults' time spent receiving care should be, in principle, considered for inclusion in economic evaluations carried out from a societal perspective". Support for the third statement, "CYP's time spent receiving care should be, in principle, considered for inclusion in economic evaluations carried out from a societal perspective", was slightly lower. In total, 238 respondents (87%) somewhat agreed or strongly agreed with this statement, though only 61% of the 199 respondents who strongly agreed that time in general is a resource worth considering for inclusion in an economic evaluation also strongly agreed that this is the case when it comes to CYP's time.

3.4. Reasons CYP's time is rarely included in economic evaluations in health care

As part of the questionnaire, respondents were also asked to indicate why, in their opinion, CYP's time is not often included in economic evaluations carried out from a societal perspective. Various options were provided, as well as an opportunity to suggest additional reasons. Respondents could select as many of the options as they wished. Table 2 shows selected options and their frequency. The commonest answers were "attaching a value to CYP's time is challenging" (selected by 223 respondents) and "measuring CYP's time is difficult or impractical" (selected by 126 respondents). Taken together, these answers pointed to practical and methodological impediments, a sentiment that was also expressed in other parts of the survey. Fifty-seven respondents (21% of those asked) suggested additional reasons and provided further explanation as 'free-text' comments. These answers were studied, coded, and categorised under one or more overarching themes (see Table 2). Thirty-one (54%) of the free-text responses elaborated on answers selected from the available options, typically providing further thoughts

Table 1
Respondent characteristics.

| | Count | Percentage ^a |
|--|-------|-------------------------|
| How would you best describe yourself? | | |
| As someone who primarily carries out research in health economics. | 215 | 76.24% |
| As someone who sometimes carries out research in health economics. | 64 | 22.70% |
| As someone who never or nearly never carries out research in health economics. | 3 | 1.06% |
| In the last five years, how often has your research role involved leading or carrying out economic evaluations of health care interventions. | | |
| Never or almost never | 19 | 6.81% |
| Rarely | 13 | 4.66% |
| Sometimes | 52 | 18.64% |
| Often | 77 | 27.60% |
| Always or almost always | 118 | 42.29% |
| In the last five years, how often has your research role involved leading or carrying out research on methodological aspects related to economic evaluation of health care interventions. | | |
| Never or almost never | 28 | 10.04% |
| Rarely | 36 | 12.90% |
| Sometimes | 84 | 30.11% |
| Often | 80 | 28.67% |
| Always or almost always | 51 | 18.28% |
| How familiar do you feel you are with the general methodology for conducting economic evaluations in health care? | | |
| Unfamiliar | 0 | 0.00% |
| Somewhat familiar | 15 | 5.47% |
| Familiar | 64 | 23.36% |
| Very familiar | 195 | 71.17% |
| How familiar would you say you are with the categories of costs and outcomes that should typically be included in analyses carried out from different perspectives. | | |
| Unfamiliar | 1 | 0.36% |
| Somewhat familiar | 10 | 3.65% |
| Familiar | 61 | 22.26% |
| Very familiar | 202 | 73.72% |
| How familiar would you say you are with measurement, valuation and inclusion of individuals' forgone time (e.g., paid or unpaid work, leisure etc.) in economic evaluation. | | |
| Unfamiliar | 5 | 1.82% |
| Somewhat familiar | 44 | 16.06% |
| Familiar | 110 | 40.15% |
| Very familiar | 115 | 41.97% |
| In what capacity do you primarily carry out research in health economics?^b | | |
| As an employee of an academic institution. | 227 | 82.85% |
| As an employee of a non-governmental, not-for-profit organisation. | 19 | 6.93% |
| As an employee of a governmental organisation. | 8 | 2.92% |
| As an employee of a private, for-profit, company or organisation. | 5 | 1.82% |
| As a self-employed researcher. | 3 | 1.09% |
| As a student. | 6 | 2.19% |
| Other | 6 | 2.19% |
| I don't know or I prefer not to answer. ^c | 0 | 0.00% |
| How would you best describe yourself?^b | | |
| As a health economist. | 224 | 81.75% |
| As a health care professional. | 24 | 8.76% |
| As a researcher in a different field or discipline. | 52 | 18.98% |
| Other, please specify: | 6 | 2.19% |
| I don't know or I prefer not to answer. ^c | 0 | 0.00% |
| How would you describe yourself in relation to your experience in undertaking economic evaluations in health care? | | |
| I have a lot of experience in conducting economic evaluations in health care. | 140 | 51.09% |
| I have some experience in conducting economic evaluations in health care. | 117 | 42.70% |
| I have little or no experience in conducting economic evaluations in health care. | 15 | 5.47% |
| I don't know or I prefer not to answer. | 2 | 0.73% |
| In which country are you based? | | |
| United Kingdom | 97 | 35.40% |
| United States | 57 | 20.80% |
| Australia | 27 | 9.85% |
| Canada | 18 | 6.57% |
| Netherlands | 7 | 2.55% |

Table 1 (continued)

| | Count | Percentage ^a |
|--------------|-------|-------------------------|
| Germany | 4 | 1.46% |
| Brazil | 3 | 1.09% |
| Italy | 3 | 1.09% |
| South Africa | 3 | 1.09% |
| Other | 17 | 6.23% |
| Missing | 46 | 13.87% |

^a Calculated as the number of times an option was selected over the total number of respondents.

^b Respondents can select multiple options. Percentage given does not add up to 100%.

^c Indicates option was exclusive and not selectable in combination with other options.

regarding challenges in measuring and attaching monetary values to CYP's time. A further 25 answers (44%) mentioned requirements and specifications of economic evaluation as factors that influence researchers, including established guidelines and 'uninterested' decision-makers. As an example, respondents from the UK mentioned the influence of National Institute of Health and Care Excellence guidance (NICE, 2013) and the rarity of a true societal perspective as issues preventing the inclusion of CYP's time in economic evaluations. For example, a respondent expressed the view that "*Rarely (in the UK) are we able to comprehensively include all the elements a societal perspective demands. [...] This kind of leads into a narrower than intended version of a societal perspective being used in practice.*"

Several respondents mentioned gaps and uncertainties in the methods for the valuation and inclusion of the opportunity cost of CYP's time in economic evaluations, as well as practical obstacles to doing so. Commonly mentioned methodological gaps related to appropriate techniques for valuing CYP's time, while frequently mentioned uncertainties were around whether the opportunity cost of displaced time is already reflected in other parts of the evaluation (e.g., in parents/guardians' costs or in outcomes), as well as whether the time of CYP, who are not typically in paid employment, is worth considering as part of a societal perspective. Comments about practical challenges typically mentioned additional expense, time and ethical approvals needed for collecting additional data on displaced time from CYP.

Respondents who reported being familiar with methods associated with the measurement, valuation, and inclusion of time in economic evaluation were significantly more likely to select the option "measuring CYP's time is difficult or impractical" ($p = 0.01$) and provide additional comments about methodological uncertainties ($p = 0.05$), likely as their familiarity with such methods provides more awareness of the underlying methodological challenges and gaps. People with a lot of experience conducting economic evaluations were significantly more likely to select the options "many researchers do not explicitly think about CYP's time" ($p = 0.01$) and "many researchers think only adults' time should be included in economic evaluations" ($p = 0.05$). Results of statistical analyses are given in Electronic [Supplementary Material 2](#).

3.5. Views on when it is justifiable to leave CYP's time out of an economic evaluation

Respondents were also asked to indicate situations in which, in their opinion, it may be justifiable to not include displaced CYP time in an economic evaluation carried out from a societal perspective. Answers are summarised in [Table 2](#).

Over 78% of respondents ($n = 215$) believed it would be justified to exclude CYP's time in situations when the amount of forgone time is small enough to be considered negligible, and almost 74% ($n = 202$) that it would be reasonable when there is little or no difference in forgone or saved time between compared interventions. These were the most common answers chosen, suggesting that researchers may be willing to exclude these costs when they feel they are likely to be inconsequential.

Table 2
Respondents' views.

| | Count | Percentage ^a |
|---|-------|-------------------------|
| Time is a valuable resource that, in principle, should be considered for inclusion in economic evaluations carried out from a societal perspective. | | |
| Strongly disagree | 2 | 0.73% |
| Somewhat disagree | 1 | 0.36% |
| Neither agree nor disagree | 3 | 1.09% |
| Somewhat agree | 69 | 25.18% |
| Strongly agree | 199 | 72.63% |
| Adults' time spent receiving care should be, in principle, considered for inclusion in economic evaluations carried out from a societal perspective. | | |
| Strongly disagree | 0 | 0.00% |
| Somewhat disagree | 4 | 1.46% |
| Neither agree nor disagree | 7 | 2.55% |
| Somewhat agree | 56 | 20.44% |
| Strongly agree | 207 | 75.55% |
| Children and young people's time spent receiving care should be, in principle, considered for inclusion in economic evaluations carried out from a societal perspective. | | |
| Strongly disagree | 1 | 0.36% |
| Somewhat disagree | 13 | 4.74% |
| Neither agree nor disagree | 22 | 8.03% |
| Somewhat agree | 111 | 40.51% |
| Strongly agree | 127 | 46.35% |
| Children and young people's time is rarely valued and included in economic evaluations carried out from a societal perspective. Why do you think this may be? ^b | | |
| Many researchers think that only adults' time should be included in economic evaluations. | 72 | 26.28% |
| Many researchers think that CYP's time is not valuable or its value is very small. | 93 | 33.94% |
| Measuring CYP's time is difficult or impractical. | 126 | 45.99% |
| Attaching a value to CYP's time is challenging. | 223 | 81.39% |
| Many researchers think accounting for CYPs time is not essential, as it has rarely been done in existing economic evaluations. | 79 | 28.83% |
| Many researchers do not explicitly think about CYP's time when they consider which inputs/costs to include in their analyses. | 117 | 42.70% |
| Other reason(s), please explain | 57 | 20.80% |
| Themes ^c | | |
| Due to methodological uncertainties | 24 | 42.11% |
| Due to practical challenges | 6 | 10.53% |
| Due to requirements and specifications of economic evaluation | 25 | 43.86% |
| Elaborates on available option(s) | 31 | 54.39% |
| Other | 6 | 10.53% |
| I don't know or I prefer not to answer. ^d | 2 | 0.73% |
| In which situations do you think it may be justifiable to leave CYP time out of an economic evaluation carried out from a societal perspective? ^b | | |
| When the amount of forgone time is small enough to be considered negligible. | 215 | 78.47% |
| When there is little or no difference in forgone or saved time between compared interventions. | 202 | 73.72% |
| When forgone time relates to very young children. | 93 | 33.94% |
| Other reason(s), please explain: | 21 | 7.66% |
| Themes ^c | | |
| Potentially incompatible with established methods in economic evaluation | 4 | 19.05% |
| When CYP's time is not a forgone resource | 4 | 19.05% |
| Inclusion is not in line with requirements or specific situation | 8 | 38.10% |
| Methods are unavailable | 2 | 9.52% |
| Elaborates on available reason(s) | 1 | 4.76% |
| Other | 8 | 38.10% |
| It is never justifiable; CYP's time should always be included in economic evaluations carried out from a societal perspective. ^d | 11 | 4.01% |
| I don't know or I prefer not to answer. ^d | 6 | 2.19% |

^a Calculated as the number of times an option was selected over the total number of respondents.

^b Respondents can select multiple options. Percentage given does not add up to 100%.

^c Themes developed by research team and applied to free-text answers provided by respondents using thematic analysis. Answers may fall under multiple themes. Percentage given does not add up to 100%.

^d Indicates option was exclusive and not selectable in combination with other options.

Eleven respondents (4%) indicated that it is never justifiable to exclude CYP's time from an economic evaluation carried out from the societal perspective, while six (2%) selected "I don't know or I prefer not to answer".

Free-text comments were provided by 21 respondents. In these were references to situations where accounting for CYP's time may be unsuitable for a particular study (e.g., incompatible with aims or population of interest), where this might be already reflected in other parts of the economic evaluation (and thus posing a risk of 'double-counting'), and where CYP's time receiving care may not be perceived to have an opportunity cost. Further comments elaborated on selected options and mentioned the lack of clear guidance in the literature.

Finally, 34% of respondents (n = 93) said that disregarding CYP's time in a societal economic evaluation might be justifiable when displaced time relates to very young children. These respondents were asked to determine the age they considered to be a reasonable cut-off value below which children would be too young to consider, alongside their reasoning for their answer. Of the 93 respondents, 35% (n = 27) selected age 5, with 55% of answers giving values between 4 and 6 years of age (inclusive). Almost universally, these respondents gave the start of primary education as the reason for selecting this age, with several elaborating that this provides a methodological basis for valuing forgone school time. A few respondents pointed to laws relating to part-time and full-time work, while others suggested ages based on developmental milestones, the relative level of independence of the child, and thus their freedom to choose how they spend their own time.

Statistical analyses showed that those who often or always conduct economic evaluations were significantly more likely to answer that it is justifiable to leave CYP's time out when the amount of forgone time is small enough to be considered negligible (p = 0.02) and when there is little or no difference between interventions (p = 0.05). On the other hand, respondents who often or always conduct methodological research related to economic evaluation were more likely to select the option "when forgone time relates to very young children" (p = 0.03). (see Electronic [Supplementary Material 2](#)).

3.6. Practice in relation to CYP's time and economic evaluations

The next set of questions asked respondents about their own practice and experience. Answers are summarised in [Table 3](#). Asked whether they had ever conducted one or more economic evaluations of health care interventions related to CYP, 156 respondents (57%) answered positively, with most indicating that, at least some of them were carried out from a societal perspective. We asked respondents who have carried out at least one economic evaluation related to CYP that did not adopt a societal perspective to indicate reasons for this choice. Perhaps unsurprisingly, the most frequent answer was "a different perspective was recommended by decision-makers", pointing to the importance of meeting decision-makers' requirements, where these are available.

Respondents who reported having carried out one or more of economic evaluations from a societal perspective were asked to answer a further set of questions about the most recent such study that they undertook or were involved in. Seventy-two respondents did not include CYP's time as part of their analysis and were asked their reasons for not doing so. More than a third (36%, n = 41) of them said this was because "it is not clear how to value CYP's time", whilst just over 20% (n = 23) said "it is not clear how to measure CYP's time". Sixteen respondents (14%) indicated that "it is not clear how to include CYP's time", and another 16 (14%) answered that the additional time displaced by receiving care was small enough to be considered negligible. Other answers provided referred to lacking sufficient data, the type of intervention or population making the inclusion of such forgone time more challenging or unnecessary, or that they simply did not consider it. These answers largely fell in line with earlier questions regarding reasons for the infrequent consideration of this CYP's time.

Those who often, always, or almost always conduct applied research

Table 3
Respondents' practice.

| | Count | Percentage ^a |
|---|-------|-------------------------|
| Have you ever designed, carried out, or contributed to an economic evaluation assessing one or more interventions related to CYP? | | |
| No, never. | 118 | 43.07% |
| Yes, I have carried out one to four economic evaluations related to CYP. | 123 | 44.89% |
| Yes, I have carried out five or more economic evaluations related to CYP. | 33 | 12.04% |
| Were any of those economic evaluations carried out from a societal perspective, either as part of the primary or secondary analyses? | | |
| No, none of them. | 55 | 35.26% |
| Yes, some of them. | 63 | 40.38% |
| Yes, all of them. | 38 | 24.36% |
| Please think of the last economic evaluation of interventions related to CYP that was not carried out from a societal perspective. What was (were) the main reason(s) for not adopting a societal perspective in that analysis? ^b | | |
| A different perspective was appropriate to capture key costs and benefits. | 49 | 26.34% |
| A different perspective was recommended by decision-makers. | 55 | 29.57% |
| A different perspective was selected to match comparable evaluations. | 20 | 10.75% |
| Data available made adopting a societal perspective impossible. | 46 | 24.73% |
| Other | 13 | 6.99% |
| I don't know or I prefer not to answer. | 3 | 1.61% |
| Please think of the most recent economic evaluation related to CYP carried out from a societal perspective that you undertook or were involved in. How would you best describe this study? ^b | | |
| Cost-utility analysis | 49 | 48.51% |
| Cost-effectiveness analysis | 44 | 43.56% |
| Cost-benefit analysis | 17 | 16.83% |
| Cost of illness analysis | 13 | 12.87% |
| Cost analysis | 6 | 5.94% |
| Other | 8 | 7.92% |
| How was CYP's time accounted for in this analysis? | | |
| By considering forgone or saved time as a cost or cost-saving and including this in cost calculations. | 18 | 62.07% |
| By considering forgone or saved time as a benefit or loss of benefit and including this in outcomes calculations. | 6 | 20.69% |
| Other, please explain: | 5 | 17.24% |
| How was CYP's time measured in this analysis? | | |
| CYP's time was measured through bespoke data collection forms (e.g., questionnaires, diaries) developed or adapted for the purposes of the particular study. | 9 | 50.00% |
| CYP time was retrieved from other sources (e.g., databases of routinely collected data). | 5 | 27.78% |
| Other | 4 | 22.22% |
| What value was attached to a unit of CYP's time in this analysis? | | |
| CYP's time was not valued. | 1 | 5.56% |
| Other, please explain: | 16 | 88.89% |
| Themes ^c Monetary value related to wages or productivity | 8 | 38.10% |
| Related to education costs | 4 | 19.05% |
| Other - work in progress | 3 | 14.29% |
| Unclear | 2 | 9.52% |
| Other | 4 | 19.05% |
| I don't know or I prefer not to answer. | 1 | 5.56% |
| What was (were) the main reason(s) for including CYP time in outcomes calculations? | | |
| Please explain: | 5 | 83.33% |
| Themes ^c Appropriate for perspective | 2 | 40.00% |
| Pragmatic reasons | 1 | 20.00% |
| Other | 2 | 40.00% |
| I don't know or I prefer not to answer. | 1 | 16.67% |

^a Calculated as the number of times an option was selected over the total number of respondents.

^b Respondents can select multiple options. Percentage given does not add up to 100%.

^c Themes developed by research team and applied to free-text answers provided by respondents using thematic analysis. Answers may fall under multiple themes. Percentage given does not add up to 100%.

related to economic evaluation were significantly more likely to have carried on or contributed to an economic evaluation of an intervention related to CYP ($p = 0.00$), as were those who indicated they are very familiar with the literature around inclusion of time in economic evaluation ($p = 0.08$), and those who identify as health economists ($p = 0.01$). A possible explanation is that researchers with greater experience and familiarity with methods in conducting economic evaluations are more likely to have conducted and economic evaluation related to CYP (or vice versa). Additionally, those who often, always, or almost always conduct methodological research into aspects related to economic evaluation were significantly more likely to have conducted an economic evaluation related to CYP using a societal perspective ($p = 0.05$) (Electronic Supplementary Material 2).

The 29 respondents who indicated they had taken CYP's time into account as part of their analysis were asked further questions about related methodological aspects. Of those asked, 18 (62%) included time by considering it as a cost or cost-saving. One respondent did not apply an estimated 'unit cost' or value to this time, whilst others used a variety of *ad hoc* estimates, including costs associated with schooling, adult average wages, or a proportion of adult average wages. A further six respondents (21%) indicated that CYP's time was considered as a benefit or loss of benefit, including it within outcomes calculations. The remaining five respondents answered that displaced CYP's time was factored in differently, including in a descriptive way, as both a cost and an outcome, or as a decrement applied to CYP's future earnings.

A proportions test showed that the inclusion of CYP's time appeared to be more common in countries outside the UK, the US, Canada, or Australia ($p < 0.01$). CYP time was considered by 80% of respondents who had conducted societal paediatric economic evaluations in "Other" countries (compared to 26% of UK-based respondents, 17% in Canada, 16% in the US, and 14% in Australia).

3.7. Views about areas for future research

Respondents were asked to consider the extent to which they agreed with a series of statements pertaining to the availability of methods relating to the measurement, valuation, and inclusion of CYP's time in economic evaluations. Responses to this question are available in Table 4.

Excluding the 45 individuals who selected "I don't know or I prefer not to answer", 60% of respondents indicated that they (somewhat or strongly) disagreed that methods for the measurement of CYP's forgone time are available and well-developed, 84% (somewhat or strongly) disagreed with a similar statement about valuation of CYP's time, and 73% (somewhat or strongly) disagreed about methods and guidance being available on when and how CYP's time should be reflected in an economic evaluation. Correspondingly, most respondents assigned high priority to research on how to value and when to include CYP's time in an economic evaluation carried out from a societal perspective (49% and 55%, respectively), and thought of research on ways to measure CYP's time to be of medium priority (51%).

4. Discussion

In this article, we report the findings of an international online survey aiming to understand health economists' views and practice in relation to the inclusion of CYP's time in economic evaluations. To our knowledge, this is the first survey on this topic.

The survey was administered online and adopted a list-based approach. List-based surveys require assembling the study's sampling frame, they offer greater control over who is invited to complete the survey (especially compared to unrestricted self-select or intercept surveys), which in turn can improve reach and diversity (Fricker, 2017). This aligns with the purpose of our study, which was to gather a range of health economists' views by capturing as many respondents as possible, rather than to test a specific hypothesis against a pre-specified level of

Table 4

Views about areas of future research.

| | Count | Percentage ^a |
|---|-------|-------------------------|
| Methods for the measurement of forgone time in CYP are available and well-developed. | | |
| Strongly disagree | 41 | 14.96% |
| Disagree | 96 | 35.04% |
| Neither disagree nor agree | 51 | 18.61% |
| Agree | 36 | 13.14% |
| Strongly agree | 5 | 1.82% |
| I don't know or I prefer not to answer. | 45 | 16.42% |
| Methods for the valuation of forgone time in CYP are available and well-developed. | | |
| Strongly disagree | 87 | 31.75% |
| Disagree | 105 | 38.32% |
| Neither disagree nor agree | 29 | 10.58% |
| Agree | 8 | 2.92% |
| Strongly agree | 0 | 0.00% |
| I don't know or I prefer not to answer. | 45 | 16.42% |
| Methods for the inclusion of forgone time in CYP are available and well-developed. | | |
| Strongly disagree | 54 | 19.71% |
| Disagree | 115 | 41.97% |
| Neither disagree nor agree | 36 | 13.14% |
| Agree | 22 | 8.03% |
| Strongly agree | 3 | 1.09% |
| I don't know or I prefer not to answer | 44 | 16.06% |
| If you consider methods to be unavailable or inadequate, please indicate what priority you would attach to research into different methodological aspects? | | |
| Research into data collection methods to facilitate measuring CYP's time.^b | | |
| Low level of priority | 29 | 15.03% |
| Medium level of priority | 98 | 50.78% |
| High level of priority | 63 | 32.64% |
| I don't know or I prefer not to answer | 3 | 1.55% |
| Research into methods for the monetary valuation of CYP's time.^b | | |
| Low level of priority | 20 | 8.85% |
| Medium level of priority | 91 | 40.27% |
| High level of priority | 111 | 49.12% |
| I don't know or I prefer not to answer | 4 | 1.77% |
| Research on when and how to incorporate CYP's forgone time in economic evaluations.^b | | |
| Low level of priority | 22 | 10.43% |
| Medium level of priority | 69 | 32.70% |
| High level of priority | 116 | 54.98% |
| I don't know or I prefer not to answer | 4 | 1.90% |

^a Calculated as the number of times an option was selected over the total number of respondents.

^b Respondents only shown this option if they selected *strongly disagree*, *disagree*, or *neither agree nor disagree* with the corresponding statement in the prior question.

confidence. We received over 270 responses (14% of the total number of invitees); however, as with every survey where the exact size and synthesis of the population of health economists is not known, it is not possible to know how well the respondents matched this population, or the extent to which the reported views are representative and exhaustive. In addition, the survey was only available in English, which might have precluded the participation of some invitees.

We found that respondents do think of time as a resource that is worth considering for inclusion in economic evaluation, with most strongly agreeing with relevant statements. However, respondents were not as unequivocal when asked specifically about CYP's time. Compared to adults' time, fewer respondents agreed that it is an important resource that ought to be considered for inclusion in economic evaluations carried out from a societal perspective. Answers also indicated various views on barriers to the inclusion of CYP's time in such evaluations. More than two-thirds of the respondents attributed this to challenges in attaching a monetary value to CYP's time, with other frequently indicated reasons including difficulties with measuring CYP's time as well as the fact that, often, researchers simply do not think about including CYP's time in their studies. Reasons put forward directly by respondents pointed to uncertainties about when it is appropriate to include CYP's

time-related costs in an analysis, practical challenges largely in relation to data collection, and lack of clear guidance and appropriate methods.

Largely, answers about situations where disregarding CYP's time-related costs would be justifiable pointed to pragmatic considerations. Most respondents thought that such costs may be inconsequential—and therefore can be excluded—when time spent on treatment is small (both in absolute terms or as difference between compared interventions) and when CYP whose time is considered are very young (typically, less than 4 years old). With regards to practice, about 1 in 10 of all respondents had carried out an economic evaluation related to CYP where they considered forgone CYP time, with such time typically valued using *ad hoc* unit cost estimates (typically linked to adult wage figures or estimates of the daily cost of schooling).

Looking across the survey, there were several recurring points. First, responses revealed some ambivalence around when it is appropriate to account for CYP's time in an economic evaluation. Uncertainties centred around topics such as when CYP's time ought to be part of an economic evaluation carried out from a given perspective and when its inclusion may be inconsequential, superfluous, or even detrimental. For example, respondents mentioned ambiguity around when CYP's time has a discernible opportunity cost that is worth considering in an economic evaluation, or uncertainty on the possibility that inclusion of CYP's time as a cost may lead to 'double-counting' (i.e., if loss of time is also reflected (implicitly or explicitly) in the 'outcomes' side of the evaluations). To tackle such uncertainties, there is a need for an approach that encourages empirical research (e.g., whether treatment time considerations may be reflected in outcome valuation), lays out the findings for interpretation and debate, and produces recommendations that are clear, accessible, and, where possible, adaptable to different requirements and jurisdictions.

Equally importantly, respondents highlighted gaps in the economic evaluation 'playbook', which have a bearing on practice. Prominent amongst them was the lack of insights into appropriate estimates of the opportunity cost of CYP's time. The latter was hardly surprising: this is a gap that has been often highlighted in the literature, usually accompanied by calls for research on the topic (Andronis et al., 2019; Tsimicalis et al., 2006; Ungar and Gerber, 2010). In the absence of better solutions, it is often suggested that the value of CYP's time can be approximated by average wage figures (or some proportion of it) (Posnett and Jan 1996; Neumann et al., 2016) and we found that such estimates have been used in most economic evaluations mentioned in this survey. To fill challenging methodological gaps, one may need to think creatively. For example, rather than using arbitrary values, it may be possible to infer the monetary values individuals place on their time through elicitation. Encouragingly, findings of outcome valuation studies suggest that CYP can successfully engage with stated preference elicitation exercises (Ratcliffe et al., 2011; Guerriero et al., 2018; Dalziel et al., 2020), however, further research will be needed to understand whether (and if so, under what conditions) this is the case in the specific context of valuation of time. Such insights should be also complemented by additional meaningful work on how time and its value is perceived amongst different age groups and for different displaced activities (e.g., 'committed', 'contracted' or 'free' time). Other possible approaches should also be considered (e.g., inferring the value of lost education time with reference to impact on attainment and, subsequently, loss of future earnings) that may, in some cases, complement direct elicitation (Torgerson et al., 1994). Inevitably, normative questions are expected to arise (for example, whether decision-makers and society would be prepared to take into account CYP's views and, if not, whose values ought to be count), which will need to be discussed and debated.

Importantly, to move the topic forwards, gaps and uncertainties will need to be pinpointed and laid out. We feel that understanding researchers' views and bringing these to the fore is a useful first step in this direction.

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Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Data availability

Anonymised data underpinning the reported study can be accessed following a formal request, in accordance with terms and conditions set by the NIHR (Funder) and the University of Warwick (Contractor).

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.socscimed.2023.116179>.

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