STUDY PROTOCOL

The Children and Young People Quality of Life Study: A protocol for the qualitative development of attributes for capability wellbeing measures for use in health economic evaluation with children and young people [version 1; peer review: awaiting peer review]

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Abstract

Background: Health services face difficult choices regarding how to allocate scarce health and social care resources. Economic evaluation can aid this process of decision making by allowing competing healthcare interventions or services to be compared in terms of their costs and consequences. However, existing use of economic evaluation of health services largely focuses on comparing interventions in terms of their impact on health only, missing factors potentially important to broader wellbeing. The capability approach measures outcomes in terms of what people are able to do and be. It provides an alternative evaluative space for economic evaluation, which may be especially beneficial for populations such as children and young people, for whom health might not be the only outcome of importance to their wellbeing.

Study aims: This study aims to develop conceptual attributes and descriptive systems for capability wellbeing measures for children and young people to be used in economic evaluation, using qualitative methods with children and young people directly and their parents/guardians.
Methods: Qualitative interviews will be used with children and young people and parents/guardians to develop the conceptual attributes (items) and wording for the capability wellbeing measures. Recruitment will be undertaken through organisations such as schools and charities and online using Facebook. Interviews will be face-to-face or online and will focus on identifying factors important to children and young people's wellbeing to create an initial draft of the capability wellbeing measures. Subsequent interviews will focus on refining the wording used in the measures. Interviews will be analysed using methods of constant comparison, to explore similarities and differences in what participants considered important to children and young people's wellbeing and to develop appropriate wording for the measures based on participant responses.

Ethics: The study received approval from the University of Bristol Faculty of Health Sciences Research Ethics Committee (reference 77121).

Keywords
Protocol, capability wellbeing, children and young people, economic evaluation, qualitative research

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Introduction

Health services face increasingly difficult choices about what interventions to fund from available resources. These decisions are often informed by evidence derived through economic evaluation, where competing interventions are compared in terms of their costs and consequences, with a typical focus on maximising health gain (Drummond et al., 2015). However, there is growing recognition that this focus is too narrow (Brazier & Tsuchiya, 2015). An important alternative is to evaluate the capability wellbeing that is generated in the population (Helter et al., 2020). The capability approach is concerned with the freedom to achieve across all aspects of life that are valued (Sen, 1993) and outcomes are measured in terms of what a person can be and do in their life. A key advantage of the approach is that it can provide an alternative evaluative space for economic evaluation, capturing a broader set of benefits from health and care interventions instead of health functioning alone (Coast et al., 2008; Greco et al., 2016; Simon et al., 2013). Capability wellbeing measures for use in economic evaluation have previously been developed for the adult population (ICECAP-A) (Al-Janabi et al., 2012; Flynn et al., 2015), older people (ICECAP-O) (Coast et al., 2008; Grewal et al., 2006) and those at end-of-life (ICECAP-SCM) (Huynh et al., 2017; Sutton & Coast, 2014). All were generated using direct research with the relevant population to determine what is important to individuals within that population – specifically, using qualitative methods to inform the conceptual attributes (or items) contained within the measures. Capabilities and their development have been demonstrated to be particularly crucial in children (Anand & Roope, 2016), but as it stands, there are currently no capability wellbeing measures available for use with children and young people (CYP) in economic evaluation. Further, many of the health measures currently used in economic evaluations to generate health-related quality of life have simply been adapted from adult versions (Chen & Ratcliffe, 2015; Griebisch et al., 2005; Ungar & Gerber, 2010) with questionable appropriateness and sensitivity. Economic measures such as the CHU-9D that have been developed specifically for children have focused explicitly on the health context in the derivation of attributes (Stevens, 2009), which is arguably even less appropriate for CYP than for adults since it ignores broader factors potentially important to CYP’s wellbeing (Mitchell et al., 2021).

Aims and objectives

This study aims to develop a range of capability wellbeing measures for CYP covering different ages/developmental stages to be used in economic evaluation and to inform health and social care funding decisions. This protocol details the qualitative methods for the development of the conceptual attributes and wording for the capability wellbeing measures for CYP.

Protocol

Study design

The research will be split into two iterative phases. Phase 1 will focus on developing the conceptual attributes for the CYP capability wellbeing measures. Phase two will focus on ensuring that the conceptual attributes are fully developed, and meaningful wording is generated for each measure so that it is appropriate and relevant for each group of CYP.

Sampling and recruitment

Phase 1:

To develop capability measures for CYP, a broad range of CYP and their parents/guardians will be recruited. The age of CYP sampled will range from 0–18 years. Those with parental responsibility will be recruited either in addition to, or as a proxy for, the CYP’s views. The sampling criteria for this phase of the research are as follows:

Inclusion criteria:

- CYP aged 0–16 from a range of backgrounds and/or those with parental responsibility.
- CYP aged 16–18 from a range of backgrounds.

Exclusion criteria:

- CYP aged 5 and under as interviews are not recommended with children of this age (Arbuckle & Abetz-Webb, 2013).
- CYP and/or those with parental responsibility who are unable to understand study information and/or communicate in English.

A multi-method, purposeful approach (Patton, 2002) to sampling will be taken, aiming for maximum variation (Merkens, 2004) with those sampled including CYP from different age groups, socio-economic backgrounds, gender, ethnicity, household composition and those with and without health conditions. A gradual selection sampling approach will be taken, which involves assessing who to approach next based on the nature of data previously collected (Flick, 2009). All CYP aged 6 to 15 will be recruited through their parents/guardians and young people aged 16 to 18 who are able to provide informed consent will be recruited directly. Parents/guardians will act as a proxy for CYP aged 5 and under.

Several sampling strategies are planned to recruit CYP and/or their parents. Strategy one involves sampling through organisations including children’s centres, schools, and charities. Access to organisations will be gained through contacting the appropriate gatekeepers. Where organisations agree to help with study recruitment, they will be asked to assist in identifying potential participants and delivering recruitment material to parents/guardians. In some circumstances recruitment will be undertaken in person by the researcher and in these cases recruitment material will be handed to potential participants directly. Recruitment materials include a study information sheet, with versions available for parents/guardians but also for CYP of different ages at different key stages of education, a study invitation letter and study screening questionnaire. Potential participants will be asked to complete
and return a copy of the screening questionnaire if they are interested in participating after reading the study information. The questionnaire is designed to collect information on the characteristics of the CYP interested in participating in the study to ensure that CYP from a broad range of backgrounds are involved, to enhance the generalisability of the developed measures. The questionnaire also asks for potential participants’ contact details to allow the researcher to get in touch to discuss study participation.

A second strategy was added in response to COVID-19 and the difficulties with recruiting via gatekeepers in organisations such as schools due to organisational closures. Social media platforms, specifically Facebook, will be used to advertise the study to potential participants via a Facebook ‘targeted ads’ post. The targeted ads feature allows information on the study to be targeted at participants on Facebook who meet the study inclusion criteria, for example, parents/guardians of CYP in a particular age bracket. An account in the study name will be set up on Facebook from which posts on the study can be shared. The posts will provide information on the study and contact information for the research team. Potential participants will be asked to contact the researcher via Facebook or email to express an interest in taking part. The researcher will respond to interested participants with a copy of the study information sheet and study screening questionnaire and ask the potential participant to complete and return these if they would like to participate.

Snowball sampling (Kuper et al., 2008) will be used across both strategies to access siblings of children already recruited, with the permission of their parents/guardians and if those CYP are also happy to take part.

Phase 2:
A subset of recruited CYP and parents/guardians will be re-interviewed to check the coverage and wording of the conceptual attributes for each of the measures. Participants taking part in phase 1 will be asked to consent to potentially being recontacted to take part in phase 2. We may also recruit new participants for phase 2 if we are unable to recontact enough participants from phase 1 or if we need further participants to complete the aims of the research, using either of the two recruitment strategies outlined above.

Data collection
Phase 1:
In-depth interviews with CYP and/or those with parental responsibility will be undertaken. These interviews are designed to explore what is important to CYP from the CYP’s own perspective and/or that of their parent/guardian in terms of their current wellbeing and future well-becoming (future opportunities for development), to inform the content of the conceptual attributes for the measures. Data collection will take place either face-to-face or online, with the latter approach being the only approach used whilst COVID-19 restrictions are in place.

Data collection with CYP will use participatory rural appraisal (PRA) techniques to encourage CYP to participate and engage with the research (Fargas-Malet et al., 2010; Veale, 2005). CYP will be provided with an A3 sheet of paper featuring five concentric circles and asked to draw or take (using a polaroid camera funded through the grant) a picture of themselves and place this in the centre circle. The CYP will then be asked to draw or write things of importance to them on sticky labels and place these onto the A3 paper around the picture of themselves in the centre circle. Adapted from hierarchical mapping methods used in existing measure development (Canaway et al., 2019), the CYP will be asked (if able) to place the things of most importance closest to their image (and to the inner circles) and factors of less importance further away (in the outer circles). The method is designed to elicit discussion around the factors of most importance to the CYP, allowing these to be explored further with the CYP during the interviews. Where interviews are undertaken online, the activity will be posted out to CYP in advance of their interview alongside instructions which the CYP can either read through themselves or with the help of a parent/guardian.

Separate interview topic guides will be developed for the parent/guardian and CYP interviews, and the topic guides will evolve as the research progresses to include new topics of interest. CYP and parents/guardians will be interviewed separately where possible (with parents/guardians in the vicinity for CYP interviews), although all parents and CYP will be given a choice as to whether parents/guardians will be present whilst the CYP is interviewed.

Phase 2:
Phase 2 of the study will use semi-structured interviews to check the coverage of the attributes developed in phase 1 and to generate meaningful and user-friendly wording and images to feature in the measure. CYP and parents/guardians will be asked whether they think that all the things that are important to CYP’s wellbeing have been identified in the attributes emerging in phase 1. In the CYP interviews, questions on the measure wording will focus on whether their understandings of the wording match the intended meaning of a particular attribute. Parents/guardians will be asked what they understand about the wording and asked how they think that their child will interpret it. All CYP and parents/guardians will also be asked about alternative options for the formatting of the questions and questionnaire, including the use of images to aid understanding of the questions. The interview topic guides will be adapted as data collection and analysis progress and are likely to change quite quickly as decisions around wording for each attribute are made based on previous findings. As this phase of data collection nears completion and the wording and presentation of the questions for the final measures become clearer, CYP and parents/guardians might be asked to pilot a partial or complete version of the questionnaire. This will be carried out using thinkaloud techniques, whereby the CYP or parent/guardian is asked to complete the latest version of the measure whilst ‘thinking aloud’ or verbalising their thoughts as they complete the questionnaire (Ericsson & Simon, 1993). This final, iterative aspect of this phase of research will help us to decide when the measure can be finalised and taken forward to validation.
Data analysis and sample sizes for phases 1 and 2:
Interviews for both phases of the research will be audio-recorded with permission from participants, and transcribed. Transcripts will be coded in NVivo11 qualitative data analysis software to identify key concepts and themes emerging from the interviews in terms of what is important to CYP’s wellbeing and well-becoming (future wellbeing). A coding structure will be developed based on the codes and themes emerging and will be applied to all interview transcripts, with codes being updated as analysis progresses. In-depth interviews (phase 1) will be undertaken and analysed iteratively and in batches to allow emerging findings to influence the topics explored in future interviews. In phase 2, the semi-structured interviews will be conducted and analysed iteratively to allow the proposed wording to be constantly refined in light of earlier participant feedback. CYP and parent/guardian responses will be analysed separately. Analysis will use constant comparison methods, whereby new data are continually compared to existing data to increase understanding (Strauss & Corbin, 1990). Analytic accounts will be created for batches of interviews to compare the responses of participants under key themes, and to identify differences in participants’ views, with these accounts eventually combined to compare the perspectives of all participants. This is particularly important for exploring where there may be differences in what is considered important by/to CYP (for example, whether factors of importance differ according to age groups), to determine how many separate childhood measures (with different conceptual attributes) will be needed. Recruitment, data collection and analysis will continue until data saturation has been reached (for both phases of the research) – that is, the point at which no new themes are emerging from the data (Bowen, 2008) and the underlying theory around what is important to the particular CYP group is fully developed as the basis for measure generation.

Ethics
Ethical approval
Ethical approval for the study was gained from the University of Bristol Faculty of Health Sciences Research Ethics Committee (reference 77121).

Consent process
Recruitment will be operated on an ‘opt-in’ basis, where potential participants will contact the research team if they are interested in being involved in the research, allowing individuals to consider study participation in their own time. Participants who are approached in person about the study will be asked to take the study information away with them and asked whether they would be willing to later be contacted by a researcher to see if they are interested in participating using the contact details provided in the screening questionnaire. Written informed consent will be obtained from all parents/guardians and CYP directly involved in the research, prior to the research commencing. All parents/guardians with CYP aged under 16 will be asked to provide informed consent on their behalf. CYP will be asked to assent to taking part; they will be asked to complete and ‘sign’ an informed consent form to ensure that they are happy to participate in the research and to avoid potential coercion (Alderson & Morrow, 2011). For online interviews, informed ‘e-consent’ and assent will be taken using an emailed version of the relevant consent form and participants will be asked to provide a typewritten, scanned handwritten signature or electronic representation of a handwritten signature to complete the form.

The researcher will take steps to ensure that the CYP and parent/guardian understands the research and what they are being asked to do, by reading through the information sheet with them and checking their interpretations. Decisions will be made in conjunction with parents/guardians and the CYP as to whether CYP are able and willing to take part in interviews. Participants will have the opportunity to ask any questions and have them answered prior to the interview commencing. Participants will be reminded at the beginning and throughout the research that they are free to withdraw at any time without having to give a reason. Obtaining consent from participants will be an ongoing process, and formal written consent will be taken again for those participants who are also taking part in phase 2. Participants will be informed in the study information sheets and consent forms that if they decide to withdraw either themselves and/or their child from the research, they will need to notify a member of the research team within of a week of the interview to ensure that their interview data has not already been analysed and used to inform future interviews.

Potential risks of the research
We do not anticipate that the research involves any great risks to participants; however, steps will be taken to minimise potential issues. To prevent CYP feeling coerced into participating in the research, the researcher will explain the aims and processes of the study to the CYP and ensure that they understand the study before they are asked whether they would like to take part. The researcher will take time prior to the interview to introduce themselves to the CYP, go through the study information sheet and to ensure any questions the CYP have are answered. The researcher will consider the language that they use when asking the CYP whether they would like to be involved, avoiding wording which implies that they are expected to participate (Harcourt et al., 2011). This relates to the importance of creating balanced power relations between CYP and adult researchers during research (Alderson & Morrow, 2011).

It is possible that the interviews might uncover potentially sensitive topics for the CYP or parent/guardian. Should this happen, the participant will be given the opportunity to take a break from the interview or withdraw altogether. We will also ensure that CYP are aware that they do not have to answer all questions and can pass on some if they want to. Should any safeguarding issues with a child/young person arise, the researcher will stop the interview and make contact with the relevant safeguarding persons in the organisation through which the CYP was recruited or, if recruited online, with someone at an appropriate agency who can help them – for example, a local authority first response team (within local authorities, children’s social care should act as the principal point of contact for safeguarding concerns relating to children (HMGovernment, 2018)) or the police if they are at immediate risk. The researcher will ensure prior to an interview that they have full personal and contact information available for each participant so that these can be passed on to the appropriate persons/organisations should there be any safeguarding issues.
Plans for the dissemination of research findings
Research findings will be disseminated through presentations at national and international conferences and to relevant policy making bodies, through publications in relevant journals, and, for the measures themselves, through the ICECAP website.

Discussion
There are currently no existing measures of capability wellbeing that can be used in economic evaluation with CYP to inform health and social care funding decisions. This study aims to develop capability wellbeing measures for this population, using qualitative methods with CYP and parents/guardians to generate the conceptual attributes for the measures and to refine the wording and presentation to make the measures meaningful and appropriate for use.

Data availability
No data are associated with this article.

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