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BMJ Open Developing a cerebral palsy-specific preference-based measure for a six-dimensional classification system (CP-6D): protocol for a valuation study

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ABSTRACT

Introduction Cerebral palsy (CP) is a lifelong condition. The CP quality of life (CPQOL) instrument is a frequently used disease-specific instrument to assess health-related quality of life (HRQoL) in people with CP, but it cannot be used to generate quality-adjusted life years (QALY) which are the basis of cost utility analysis (CUA). Generic utility instruments (such as the EQ-5D or SF-6D) that are used to value HRQoL may be insensitive to small but important health changes in children with CP. This study aims to generate a preference-based scoring algorithm for the CP six dimensions (CP-6D), a classification system developed from the CPQOL.

Methods and analysis A discrete choice experiment with duration (DCEto) will be administered to value health states described by the CP-6D classification system. These health states will be presented to members of Australian general population and parents of children with CP via an online survey. Conditional logit regression will be used to produce the utility algorithm for CP-6D.

Ethics and dissemination The Griffith University Human Research Ethics Committee approved for the study (reference HREC/number 2018/913). The developed algorithm can be applied to previous and future economic evaluation of interventions and treatments targeting people with CP which have used either the CPQOL or CP-6D.

INTRODUCTION

Health economic evaluations are widely used to value interventions, treatments, procedures and policies. The most prevalent methods in economic evaluation are cost-effectiveness analysis (CEA) and cost utility analysis (CUA),¹ where CUA can be considered a special case of CEA.² CEA compares the costs and outcomes resulting from an intervention relative to one or more comparators and estimates the incremental cost of each additional unit of outcome. CUA builds on this by using a summary health outcome measure that considers both mortality and quality of life, usually through the quality-adjusted life years (QALY).³ CUA is intended to enable comparison across disease areas and

Strengths and limitations of this study

- Developed a rigorous protocol to undertake the valuation of the CP-6D.
- Utility values generated from this study, using the DCE method, will be estimated for all interventions and treatment for people with CP in which CPQOL or CP6D have been applied.
- The DCEto method will help anchoring the preference value sets on a 0–1 scale for both general population and people with CP.
- The difference in preferences between the general population and people familiar with CP will be compared.

hence to be of more use to policymakers. For this reason, CUA is widely recommended as the primary method of economic evaluation by healthcare reimbursement agencies.⁴

The QALY allows comparison of different health interventions or treatments over a time interval. To estimate QALYs resulting from an intervention, health utility weights (or health state values) are required. These utility values are attributed to health states, typically described by a classification system, and are anchored on a scale from 0 to 1, where 0 represents dead and 1 represents full health.⁵ Health state values are usually generated from preference-based instruments that use preference elicitation techniques such as time trade-off (TTO), standard gamble (SG) and discrete choice experiment (DCE)⁶ to assign a value to each health state described by the underlying classification system.

Preference-based instruments can be either generic-specific or condition-specific.⁷ Conventionally, generic instruments have been more widely used, as they allow estimation of QALYs based on consistent health state classifications and utility data across all conditions and treatments. However, as these

instruments are designed to be generic, they may lack the ability to capture changes in important aspects of some health conditions, particularly for those smaller generic instruments which may consist of only a handful of domains to describe health. Therefore, there is a growing trend towards condition-specific instruments, which are likely to be more reflective of all important domains of health and more sensitive to changes in health-related quality of life (HRQoL) in some diseases or disabilities.⁸

There are two key elements of a preference-based instrument: (1) a classification system that provides a system for the full spectrum of health defined by a discrete number of health states and (2) a utility value set that provides a value for each health state as described by the classification system.⁷ Recently, a new health state classification system, the cerebral palsy six dimensions (CP-6D), derived from the CP quality of life (CPQOL) instrument has been developed (Bahrampour *et al*, under review, 2019). The CPQOL is designed to measure HRQoL in children and adolescents with CP.⁹ For developing the CP-6D, the parent/proxy version was used as there was a wide range of age groups from 1 to 17 year olds in the trials that administered CPQOL; however, the survey was adjusted based on the common questions in all versions. To develop the CP-6D, statistical methods, including factor and Rasch analyses, were used. These methods were applied to identify the dimensions and the item to represent each dimension for the CP-6D. The dimensions of the CP-6D are social well-being and acceptance; physical health; communication; pain and discomfort; manual ability and sleep.

The development of the second element of this preference-based instrument, the health state value set, is the focus of the present study. However, there is considerable debate regarding who should determine the value of each health state,¹⁰ as it is believed that people with the condition and the general population may value the dimensions of health differently.¹¹ Conversely, and perhaps more traditionally, it has been argued that it is the general population preferences that should be considered and used to determine the value of the utility health states. This line of argument is supported by two considerations. First, it is the general population who are the taxpayers (or insurance fund members) and it is their money that will be used to fund new interventions and treatments, so it is ultimately this broader population base whose preferences should be considered. Second, the general population would be representative of all diseases and conditions and therefore the valuation might be considered more objective.¹⁰

The goal of this study is to generate a preference-based scoring algorithm for the CP-6D. To measure utility values of the health states defined by the CP-6D classification system, an algorithm will be developed using statistical models. This paper describes the methodology that will be used to collect data on preferences and develop the utility values for the CP-6D. When complete, the algorithm will enable data collected from any study using the CPQOL

to be used in the economic evaluation of treatments and interventions for people with CP by converting responses from the CPQOL to a HRQoL utility value necessary for deriving QALYs.

AIMS

1. To value health states generated from the classification system (CP-6D) with both a sample of the general population and a sample from people with CP registered with Queensland CP, using a DCE.
2. To determine the difference between general population and people with CP health state utility values.

METHODS AND ANALYSIS

Preference and elicitation methods

Both ordinal and cardinal preference-based elicitation methods can be used for preference elicitation. SG and TTO are the most widely used cardinal methods to value health states. Both are choice-based approaches. The SG task, asks participants to choose between a certain health state (the health state being valued) and a scenario with a probability (p) of gaining the best possible health outcome (full health) and a probability ($1 - p$) of the worst possible health outcome (usually immediate death).¹² For health states better than dead, TTO asks individuals about how many years of living in full health followed by an immediate death they would be willing to trade compared with living in a health state for definite number of years. It determines that the individual is willing to accept living in a full health state for fewer years relative to being sick for more years. However, it has been argued that risk aversion and time preference can bias individual's response using the SG and TTO, respectively.^{13 14} Additionally, it has been shown that these tasks are highly complex¹⁵ and sensitive to mode of administration,¹⁶ hence there has recently been a shift towards ordinal methods such as the DCE approach.¹⁷ DCE has gained popularity in health economic research since it uses ordinal response to estimate interval measures¹⁸ and can potentially allow for more flexible characteristics of the utility function.¹⁹

In this study, DCE with duration (DCEtto) will be used to elicit preference for health states generated and described by a new CP specific instrument (CP-6D). The DCE model asks participants to choose their preferred health state over hypothetical alternatives.²⁰ However, the values derived from a standard DCE (which compares health states without duration) are not anchored on a utility scale from 0 to 1 and therefore cannot be used to measure QALY directly. The values generated from the DCEtto, however, can directly anchor their relative preference onto the utility scale with the inclusion of a duration attribute.^{21 22}

Each of the CP-6D domains (physical health, social well-being and acceptance, communication, pain, sleep and manual ability) consists of five levels. In this study, the choice sets for the DCE will be generated from levels of

Table 1 The cerebral palsy six dimensions classification system

| Dimension* | Description |
|----------------------------------|---|
| Social well-being and acceptance | They feel <i>very unhappy</i> about how they are accepted by people in general They feel <i>unhappy</i> about how they are accepted by people in general They feel <i>neither happy nor unhappy</i> about how they are accepted by people in general They feel <i>happy</i> about how they are accepted by people in general They feel <i>very happy</i> about how they are accepted by people in general |
| Physical health | They feel <i>very unhappy</i> about the way they get around They feel <i>unhappy</i> about the way they get around They feel <i>neither happy nor unhappy</i> about the way they get around They feel <i>happy</i> about the way they get around They feel <i>very happy</i> about the way they get around |
| Communication | They feel <i>very unhappy</i> about the way people communicate with them They feel <i>unhappy</i> about the way people communicate with them They feel <i>neither happy nor unhappy</i> about the way people communicate with them They feel <i>happy</i> about the way people communicate with them They feel <i>very happy</i> about the way people communicate with them |
| Pain and discomfort | They are <i>very upset</i> about the amount of pain they have They are <i>moderately upset</i> about the amount of pain they have They are <i>somewhat upset</i> about the amount of pain they have They are <i>slightly</i> about the amount of pain they have They are <i>not at all upset</i> about the amount of pain they have |
| Sleep | They feel <i>very unhappy</i> about how they sleep They feel <i>unhappy</i> about how they sleep They feel <i>neither happy nor unhappy</i> about how they sleep They feel <i>happy</i> about how they sleep They feel <i>very happy</i> about how they sleep |
| Manual ability | They are <i>very unhappy</i> about the way they use their hands They are <i>unhappy</i> about the way they use their hands They are <i>neither happy nor unhappy</i> about the way they use their hands They are <i>happy</i> about the way they use their hands They are <i>very happy</i> about the way they use their hands |

*Bahrapour *et al*, Under-review, 2019.

these six domains with the addition of one attribute representing duration. The ordinal levels for all dimensions (except pain and duration) are very unhappy, unhappy, neither unhappy nor happy, happy and very happy. The ordinal levels for the pain domain are very upset, moderately upset, somewhat upset, slightly upset and not at all upset (table 1). These levels are derived from the wording of the original CPQOL instrument.

The duration attribute contains five levels (1, 3, 5, 7, 10 years). The upper limit of 10 years was used as it is commonly used as the fixed period of life years lived in a less than full-health health state in TTO valuations.²³

DCE tasks

A full factorial design is a combination of all attributes and their levels. Health states included in the DCE tasks will be generated using a combination of levels across dimensions, which will involve a mixture of high levels for some dimensions and low levels for other dimensions. The combination of attributes and levels for a full fractional in this study (six dimensions of the CP-6D each with five levels) would however result in $5^6=15\ 625$ health

states and when adding duration would result in $78\ 125\ (5^7)$ health state profiles and over a billion ($78\ 125 \times 78\ 125$) possible pairwise combinations of any two health state and duration combinations. Each pair choice presents two scenarios in which the respondents are asked to choose their preferred health state to live until they die. An example of the DCE pairwise task is presented in table 2.

Given the number of health profiles it would not be appropriate to present all combinations to participants. As such, a practical subset of health states will be selected (reduced number of health states) and used in the experiment while optimising the efficiency of the design. Specifically, a D-efficient design will be used to increase the efficiency of data collection.

In experimental design, choices should be selected that can examine both main effects (the effect of each independent variable on dependent variable) and possible interactions (preference for an attribute based on the level of another). However, in a DCEtto the disutility of levels of the instrument through interactions with

Table 2 An example of a discrete choice experiment choice pair

| Domain | | |
|-----------------------------------|---|--|
| Social well-being and acceptance | You feel <i>happy</i> about how you are accepted by people in general | You feel <i>very happy</i> about how you are accepted by people in general |
| Physical health | You feel <i>happy</i> about the way you get around | You feel <i>unhappy</i> about the way you get around |
| Communication | You feel <i>unhappy</i> about the way people communicate with you | You feel <i>very happy</i> about the way people communicate with you |
| Pain and discomfort | You are <i>slightly upset</i> about the amount of pain you have | You are <i>very upset</i> about the amount of pain you have |
| Sleep | You feel <i>very happy</i> about how you sleep | You feel <i>very unhappy</i> about how you sleep |
| Manual ability | You are <i>very unhappy</i> about the way you use your hands | You are <i>happy</i> about the way you use your hands |
| Duration | Living in this health state for 3 years and then die | Living in this health state for 1 year and then die |
| Which health state do you prefer? | Health state A <input type="checkbox"/> | Health state B <input type="checkbox"/> |

duration would also be measured. For this study, a design will be developed using the design generator software Ngene,²⁴ the design that will be based on D-efficiency criteria to select pairwise choice sets. The design will be generated to capture the two-factor interactions involving duration with duration anchoring the DCEtto on a scale of full health (1.0) and dead (0.0). The DCE will be designed without any priors for the pilot study, after that the priors generated from the pilot study will be applied to the final design.

It has become standard to ask each respondent to complete between 8 and 12 choice sets when using DCE to value health states generated from a multiattribute instrument. This range has been considered to maximise data collection per respondent without incurring significant responder bias such as to undermine the quality of the data. In this study, 12 choice sets per responder was chosen. The full range of choice sets will use a block design with 12 choice sets in 20 blocks so as to obtain responses with respect to 240 health states from both the general population and for people with CP. To prevent order bias by respondents, the sequence of choice pairs will be randomised within each block. The blocking will help the balance in the levels of attributes²⁵ and will also ensure that the number of respondents per block is equal.

Survey

Participants

In this study, a sample from the general population in addition to the parents/proxies of a child with CP sample will be recruited. Therefore, a value set for each group can be presented and the preference differences between the two groups can also be obtained.

General population and CP registry group

An online survey will be administrated to an Australian general population from May 2019. The survey contains several sections, in which the beginning section is an introduction to the research and the respondents will be asked

to provide consent in order to continue with the rest of the survey. After the person accepts to be a part of the study, the next section will require participants to provide demographic data (age, gender, education, income) and health status using the AQOL-4D (the assessment quality of life-4 demension),²⁶ which will allow a determination of whether the sample are representative of the Australian population. Next, the participants will be asked to describe their own health state using the CP-6D, with the next section containing the DCEtto tasks (ie, 12 plus an addition choice sets).

In the beginning of each section, there will be an introduction and a guide on how to complete the questions. Consistency will be checked using one of the choice tasks asked twice in the DCEtto, and at the end of the DCEtto tasks the individuals will be asked to rate the difficulty of the questions on a scale from 1 to 4.

Specifically, for the CP population demographic questions will also include the level of Gross Motor Function Classification System (GMFCS)²⁷ instrument and the Manual Ability Classification System (MACS)²⁸ instrument. These two scales describe the severity of the disease and CP functioning. The GMFCS, which is based on an individual's movement, is a multilevel categorisation technique that has five levels specifying the rate of how much help a person with CP needs and identifies whether the person needs a wheelchair or can walk independently.²⁷ The MACS describes how much assistance the person needs to use their hands.^{28 29} Both tools are widely used for the purpose to capture the child's functional level in CP studies³⁰

Sample size and recruitment

The respondents will be recruited from an existing Australian online panel administrated by Survey Engine,³¹ which is a survey company with expertise in online DCEs. Each respondent will be paid a small amount to complete the survey (approximately AUD\$10 each). The respondents

will be anonymous, and only de-identified data will be provided to the researchers. The respondents will be presented a web link to access the survey and this will enable them to complete the tasks at their convenience. An online panel is a cost-effective way to recruit respondents and has been widely used in general population valuation studies.³²

Statistical efficiency is a key focus of experimental design and is a major factor in determining necessary sample size. In similar work, a sample size of 1000–2000 has been demonstrated to produce small confidence intervals, even if the experimental design is not maximally efficient.²⁵ Based on this, a sample of 2000 people from the general population will be recruited. A study by Lancsar and Louviere stated that more than 20 respondents per choice set is required to estimate reliable models.³³ In our study using a sample of 2000 individuals means that there will be more than 20 respondents per choice set which is consistent with the Lancsar and Louviere study.

With respect to the sampling of the Australian population with experience of CP, parent/proxy of children with CP will be recruited from an existing Australian CP registry. Based on previous research conducted in this population, an estimated number of people with CP who can be recruited from this registry is $n=300$. For this population, a proxy (eg, parents or guardian)³⁴ will be asked to complete the survey, as the child may lack the cognitive ability to complete all tasks within the survey. This is consistent with other studies in which a proxy has been employed to complete the tasks.³⁵

Pilot study

The survey will be soft-launched using a sample of 100 from the general population and 30 people from the CP population. The piloting will start May 2019 starting with the CP group. If no changes are made, the full data collection will occur including data collected prior and subsequent to the soft launch. The first dataset will be collected to

1. Pilot the classification system using parents/proxies opinions for validation of the six domains.
2. Check question difficulty, clarity and understanding by individuals. To this end, two questions will be added asking the participants how difficult they found the questions on a scale of 1–4 (difficulty in answering and difficulty in understanding the questions).
3. Indicate the feasibility of the duration levels.
4. Assess the time spent by each individual on the whole survey and for each DCE task. This will be used to determine participant burden and the extent to which response behaviour changes during the survey.
5. Determine the functioning of the whole survey. The pilot study will show practical issues when completing the tasks and indicates if any revisions are required including if the block design and randomisation of task ordering are operating as designed.

Patient and public involvement

No patient involved.

Analytical plan

To estimate health state values and determine the coefficients for main effects and interactions between the main effects, regression models will be fitted. This will include the conditional logit, as outlined by McFadden,³⁶ and mixed logit to potentially account for correlation of error terms in individual respondents^{37,38} (ie, the likely correlation among the multiple responses (ie, choices) provided from each individual). The final model selection will be determined based on model fit where model fit will be assessed with respect to log likelihood ratio χ^2 or McFadden's pseudo R^2 .³⁷

DCEtto is based on random utility theory, which states that the utility value of an attribute in a scenario can be explained by both fixed and random components.³⁹ As such, using the coefficients from the best-fitted model, values for the health states of the utility-based instruments can be estimated. The scoring algorithm will be developed using the model introduced by Bansback *et al*,²¹ where an extra attribute for duration is included in our design. Algorithms will be developed to convert responses for the quality of life instrument to utility-based instruments based on the coefficients of the selected models. The data will be analysed using Stata and R.

Potential incomplete data will be explored and further decision will be made based on the type of missing data. Probably a multiple imputation will be done, however the data need to be gathered first.

The health state values resulted from this study can be used to calculate QALYs in economic evaluation of treatments and interventions for people with CP, in which CPQOL has been used.

ETHICS AND DISSEMINATION

There are no known health or safety risk associated with participants in any aspect of the study. At the DCE opening however, there is a consent form that allows participant to enter the survey voluntarily and ensure that they will be fully informed about the aim of the study.

This study will estimate utility values for the new CP-specific instrument (CP-6D). This would be the first CP-specific preference-based instrument to value utility for people with CP from the Australian population. The algorithm developed from this study can be used to generate health state values for any study that has previously used CPQOL or plans to do so in the future, as the CP-6D was derived from CPQOL. The utility values generated from the algorithms developed in the present study can then be used to estimate QALYs for cost utility analysis of new treatment or interventions that are aimed for people with CP. The distribution of the results of this study will be through publication in academic journals and presenting in conferences.

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Patient consent for publication Not required.

Ethics approval For the ethics of this study, an ethical approval was given by Griffith University Human Research Ethics Committee (GUHREC) (Reference number: 2018/930).

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