

Original Article

Linguistic validation of a disease-specific quality of life measure for children and teenagers with cardiac disease

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Abstract Introduction: To anglicise an American – that is, English language – disease-specific health-related quality of life measure, using the Paediatric Cardiac Quality of Life Inventory, for children in the age group of 8–12 years and adolescents in the age group of 13–18 years with cardiac disease, and to assess conceptual equivalence of the American and British versions. **Methods:** A process of forward and backward translation of the measure was undertaken before focus groups and individual interviews with 40 participants – that is, 20 children/adolescents with cardiac disease and 20 parents of children/adolescents with cardiac disease – to determine their understanding of the meaning of the questions. **Results:** Interviews established that participants understood the meaning of the questions, although some found it difficult to explain the meaning of questions in which the language was explicit and wanted instead to answer the individual questions as they applied to them/their child. There was agreement that all versions of the questionnaire were relevant and comprehensive, and that the length of the questionnaires was acceptable and practical. **Conclusions:** The anglicised version of the Paediatric Cardiac Quality of Life Inventory appears to be a linguistically valid measure of health-related quality of life for children and adolescents with cardiac disease. The psychometric properties of the anglicised Paediatric Cardiac Quality of Life Inventory are now being tested in a multi-centre study in the United Kingdom.

Keywords: Health-related quality of life; linguistic validation

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SURGICAL AND MEDICAL ADVANCES IN THE LAST 30 years have resulted in dramatic improvements in survival rates of children with cardiac disease.¹ However, as a consequence, there is a growing population of children and young people with significant physical, neuro-developmental, and/or psychosocial morbidity because of their underlying condition and/or its management.^{2,3} There is increasing attention being given to the impact of this on quality of life, and the need for a disease-specific measure of health-related quality of life has become increasingly apparent.

In the United Kingdom, the importance of assessing quality from the patient perspective as a marker of the effectiveness of clinical care has recently been emphasised.^{4,5} Central to this is the use of patient-reported outcome measures, which are measures of a patient's health status or health-related quality of life.⁶ Since April, 2009, all providers of National Health Services funded care treating adult patients undergoing four specific surgical procedures – knee replacement, hip replacement, hernia repair, and varicose vein surgery – have been required to collect data using generic and disease-specific patient-reported outcome measures. Within the National Health Services, there is now a move to implement routine evaluation of health-related quality of life for all patient groups, both adult and paediatric, thus necessitating the development of valid and reliable measures.

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In direct response to the need for a measure of health-related quality of life for children and adolescents with cardiac disease, the Paediatric Cardiac Quality of Life Inventory has recently been developed in the United States.⁷ The measure exists in child, 8–12 years, and adolescent, 13–18 years, formats, with additional parent-proxy reporting for each age group, and extensive testing has indicated that all four forms are valid and reliable.⁸ However, external validity data are restricted to the United States. In view of the stability in the incidence of cardiac conditions in the paediatric population worldwide and the potential benefit of having an international tool to evaluate the impact of cardiac disease and its treatment on health-related quality of life, the aim of the current study was to develop a conceptually equivalent version of the Paediatric Cardiac Quality of Life Inventory for use in the United Kingdom. The specific aims were to anglicise the Paediatric Cardiac Quality of Life Inventory, using an approach of forward and backward translation; to undertake a qualitative evaluation of the resulting versions with a small sample of patients with cardiac disease and their parents, in order to identify any semantic and/or conceptual difficulties with the translations; and to make any necessary modifications to the questionnaires before further qualitative evaluation with another group of patients and parents. Successful completion of these stages would then enable the psychometric properties of the British versions of the Paediatric Cardiac Quality of Life Inventory to be tested nationally.

Methods

Translation

There is a consensus that the development and validation of quality of life measures in different countries must achieve four levels of cross-cultural equivalence, the first of which is conceptual equivalence.^{9,10} This is the extent to which items in the source – American English – and target – British English – languages are similar, both in terms of the semantic meaning and the underlying concept, which is the focus of this study.

The translation was an iterative process that followed the established guidelines of forward and backward translation.¹¹ In the first instance, members of the American and British research teams met to discuss each question on each version of the American questionnaire to clarify specific terms and concepts, and a consensus forward translation was agreed. Subsequently, a backward translation was undertaken independently by two American citizens living in the United Kingdom, and discussions were then held with members of both research teams to finalise a consensus version of all four forms of the Paediatric Cardiac Quality of Life Inventory in “British” English.

Assessment of conceptual equivalence by qualitative evaluation

We obtained ethical approval for the study from the Royal Brompton Harefield and National Heart and Lung Institute Ethics Committee, and individual consent was obtained from the parents of each child and from young adolescents aged 16 years and over. Children under the age of 16 years assented to participation.

English-speaking patients aged 8–18 years of age with cardiac disease, who were attending routine outpatient clinics at one of the two paediatric cardiac centres in the United Kingdom were identified from clinic lists. The intention was to hold a separate focus group at each centre for children in the age group of 8–12 years, adolescents in the age group of 13–18 years, parents of children, and parents of adolescents. We decided upon a minimum number of four participants for each focus group, necessitating the identification of at least four suitable participants for any one group – child or adolescent – from a particular clinic. Patients and their parents were sent a letter about the study two weeks before the scheduled clinic appointment, and this was followed-up with a telephone call. Focus groups were scheduled to take place after the clinic.

During the group discussion, each question on the appropriate version of the Paediatric Cardiac Quality of Life Inventory was read aloud and participants were asked to explain their understanding of the meaning of the question. Each question was prefaced with the phrase “Because of my (child’s) heart condition...”. Each participant was asked about each question and the first participant to answer each question was rotated around the group. Once all of the questions had been addressed, the group was asked for any further feedback about the wording or meaning of the questions, and whether they had any suggestions for further items. Each group session was tape-recorded.

Owing to the fact that in many clinics there were insufficient eligible patients for a focus group – that is, fewer than four individuals – individual interviews were held with some patients and/or parents, whereby each question was read aloud and they were asked to explain their understanding of the question. If any participant answered the question rather than explaining its meaning, they were asked probing questions to elicit their understanding of the question.

Results

During the translation phase of the study, 12 (50%) items on the child version and 9 (30%) items on the adolescent version required some changes, most of which were minor. However, some items required

greater amendment such as the adolescent item “I am self-destructive”, which was changed to “I engage in risk-taking behaviour”.

One focus group took place for each of the four groups – children, adolescents, parents of children, and parents of adolescents – and an additional 16 interviews were held, that is, four for each of the four groups. Table 1 shows the demographics.

In both the group and individual interviews, the main difficulty that participants had was in explaining the meaning of the question, rather than answering it; this was often because the meaning was explicit in the wording, and thus participants found it difficult to reword it. Table 2 shows the examples of responses given.

Participants found the questions clear, relevant, and comprehensive; they also found the length of the questionnaire to be acceptable. One question concerning the fear of dying generated more discussion; however, there was a high level of agreement that it was important to include this question. No modifications or further qualitative evaluations were required after the focus groups and interviews.

Discussion

The objective of this study – to perform a linguistic validation of an American, that is English language, disease-specific measure of health-related quality of life for children and adolescents with cardiac disease and their parents – was satisfactorily achieved. The forward and backward translation resulted in a measure that was comprehensible, relevant, and comprehensive for a British population, and which was additionally considered to be appropriate and acceptable in terms of content and length. The only difficulty with conceptual issues was for participants to reword the questions in a sufficiently different manner so as not to be repeating the words of a question in a different order, and it was evident that they found the questions easier to answer than to explain. It was important, however, to encourage explanation so as to ensure that the meaning of each item was clear. Although not within the remit of this study to investigate, one might hypothesise that questions that are underpinned by more abstract or complex concepts will be more liable to variable interpretation, irrespective of linguistic variations.

Selecting participants for the group interviews was more difficult than anticipated, and resulted in the need for some individual interviews to be undertaken. However, this allowed participants to be selected with greater diversity in terms of diagnosis, age, and ethnicity because there were no constraints on recruiting sufficient numbers from any one clinic.

Table 1. Demographics of participants.

Number participating in focus group	Age		Diagnosis (%)				Gender (%)		Ethnicity (%)		
	Number interviewed	Mean (standard deviation); years	Single ventricle	Biventricular repair	Transplant	Untreated congenital heart disease	Acquired cardiac disease with structurally normal heart	Male	Female	White	Black or Asian
Children	4	10.6 (1.5)	1 (11)	4 (44)	1 (11)	1 (11)	2 (22)	5 (56)	4 (44)	6 (67)	3 (33)
Adolescents	4	15.3 (1.2)	1 (9)	4 (36)	1 (9)	2 (18)	3 (27)	5 (45)	6 (55)	6 (55)	5 (45)
Parents of children*	4	NA	1 (10)	5 (50)	1 (10)	1 (10)	2 (20)	2 (20)	8 (80)	6 (60)	4 (40)
Parents of adolescents*	4	NA	1 (10)	5 (50)	1 (10)	1 (10)	2 (20)	3 (30)	7 (70)	7 (70)	3 (30)

*Diagnosis for parents indicates the diagnosis of their child

Table 2. Examples of responses from the group and individual interviews.

Sample question	Child responses	Parent of child responses
Because of my (child's) heart condition, adults around me (them) are overprotective	Adults won't let you do things that you want to because they think you can't because of your heart People don't let me do things because they don't want me to hurt my heart Sometimes adults worry about me too much and worry about letting me do stuff	Because of his heart people don't let him do things Adults fuss over her because of her heart She is not allowed to do things – adults worry about her and are scared to let her do things
Because of my (child's) heart condition, other people treat me (him/her) differently	They might treat you differently to how they treat other people Other people treat me not the same as they treat others – because I am different and I have something wrong with me	Because of his heart problem people feel sorry for him and act differently towards him People treat her differently to others because they worry about her heart or think she can't do things
	Adolescent responses	Parent of adolescent responses
Because of my (child's) heart condition, I (he/she) feel(s) different from everybody in a bad way	I feel generally different because I have something wrong with my heart and this is not good I feel different to how others feel because of my heart and this makes me feel bad	His heart condition makes him feel that he is not the same/as good as others He is different from others and he feels that he is not as good as they are because of his heart
Because of my (child's) heart condition, other people are uncomfortable around me (him/her)	People find it hard to be around me and be sociable because I have something wrong with my heart People don't like to talk about my heart problems and don't know what to say	People don't know how to react to her because she has something wrong with her heart Other people feel awkward and don't know what to say because of her heart

There was some regional diversity within the sample, and patients living in different parts of England were selected. There were no obvious differences in the interpretation of questions between participants from different regions, although it is acknowledged that regional differences may exist within the larger area of the United Kingdom – a generic problem with questionnaire use in any country.

Many questionnaires developed for use with an American population are used with a British population without any translation, which raises questions about their validity for use in the United Kingdom. For those that do undergo some linguistic modification, the details of how this process is conducted are not always specified.¹² The Paediatric Cardiac Quality of Life Inventory has been validated in the United States; however, before embarking on an expensive and time-consuming process of testing in the United Kingdom, it was important that the measure be known to be conceptually equivalent. Other linguistic validation studies have used techniques of cognitive probing,¹⁰ whereby participants answer the items and are asked questions about their answers; however, we were specifically interested in determining the conceptual equivalence of the two versions rather than addressing the response patterns or reasons for them. For this study, it was clear from initial discussions between the two research teams that some of the American wording did not hold the same meaning in the United Kingdom. Furthermore, some phraseology

was notably different from that used in the United Kingdom, and although this may not have obscured the meaning it may have resulted in participants being less willing to answer the questions. Finally, it is important that children and parents, rather than just health professionals, be involved in the linguistic validation to ensure comprehension of the items from different user perspectives.

Having achieved satisfactory linguistic validation of all forms of the Paediatric Cardiac Quality of Life Inventory for use with a British population, as well as endorsement of the instrument's relevance and comprehensiveness, testing of the measure to assess its psychometric properties in the United Kingdom is now underway.

References

1. British Cardiac Society. Grown-up congenital heart (GUCH) disease: current needs and provision of service for adolescents and adults with congenital heart disease in the UK. *Heart* 2002; 88 (Suppl 1): i1–i14.
2. Bellinger DC, Wypij D, duPlessis AJ, et al. Neurodevelopmental status at eight years in children with dextro-transposition of the great arteries: the Boston Circulatory Arrest Trial. *J Thorac Cardiovasc Surg* 2003; 126: 1385–1396.
3. Mahle WT, Clancy RR, Moss EM, Gerdes M, Jobs DR, Wernovsky G. Neurodevelopmental outcome and lifestyle assessment in school-aged and adolescent children with hypoplastic left heart syndrome. *Pediatrics* 2000; 105: 1082–1089.
4. Department of Health. High quality care for all: NHS next stage review final report. London, 2008.

5. Department of Health. Equity and excellence – liberating the NHS. London, 2010.
6. Department of Health. Guidance on the routine collection of Patient Reported Outcome Measures (PROMs). London, 2008.
7. Marino BS, Shera D, Wernovsky G, et al. The development of the pediatric cardiac quality of life inventory: a quality of life measure for children and adolescents with heart disease. *Qual Life Res* 2008; 17: 613–626.
8. Marino BS, Tomlinson RS, Wernovsky G, et al. Validation of the pediatric cardiac quality of life inventory. *Pediatrics* 2010; 126: 498–508.
9. Anderson RT, Aaronson NK, Leplege AP, Wilkin D. International use and application of generic health-related quality of life instruments. In: Spilker B (ed). *Quality of Life and Pharmacoeconomics in Clinical Trials*, 2nd edn. Lippincott-Raven, Philadelphia, 1996; 613–633.
10. Quittner AL, Sweeny S, Watrous M, et al. Translation and linguistic validation of a disease-specific quality of life measure for cystic fibrosis. *J Pediatr Psychol* 2000; 25: 403–414.
11. Behling O, Law K. *Translating questionnaires and other research instruments: problems and solutions*. Sage, Thousand Oaks, CA, 2000.
12. Hutchings HA, Upton P, Cheung WY, et al. Adaptation of the Manchester–Minneapolis Quality of Life instrument for use in the UK population. *Arch Dis Child* 2007; 92: 855–860.