Pediatric rheumatology

The Gap Study (GapS) interview - developing a process to determine the meaning and determinants of quality of life in children with arthritis and rheumatic disease

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Abstract

Objective

Quality of Life (QOL) is a ubiquitous yet rarely precisely defined term. QOL may be determined by the differences (gaps) between our current situation and our expectations. Contemporary methods of measuring QOL often do not take these gaps into consideration. We performed this study to develop items and methods for measuring and valuing these gaps in order to better determine individual QOL for children with rheumatic diseases.

Methods

We generated items from literature review, other QOL measures and interviews with pediatric rheumatology patients and their families. Gap-scales to measure the discrepancy between a child's current state and the expected or desired state were designed and tested iteratively in pilot interviews.

Results

Thirty-one children (mean age = 13.5 years, age range = 6-17 years) and 22 parents were recruited through pediatric rheumatology clinics. The process of item generation, reduction and preliminary formatting yielded a list of 72 items. We developed a 3-point categorical scale of importance and a vertical visual analog scale (VAS) to determine individual valuation of items. 5 gap-scales were developed to reflect different aspects of the discrepancy between the child's current and expected or desired states for different QOL items.

Conclusions

We have developed a QOL interview based on theory that we can now test to see if it will enrich our understanding of the determinants of QOL in pediatric rheumatology patients and other chronically ill children.

Key words

Quality of life, measurement, health outcomes, children, parents.

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Introduction

Advances in health care in recent decades have brought about a shift in therapeutic focus. Traditionally, there has been an emphasis on increasing longevity. For chronic illnesses like arthritis, there is a growing recognition of the importance of also improving the quality of life (QOL). The measurement of QOL may help to provide clinicians with a meaningful way to measure the impact of therapy for chronically ill patients.

There is no consensus about the definition of QOL, the construct of which has been a matter of much debate. For example, Gill and Feinstein in their critique of 75 publications of various QOL instruments found that only a minority actually defined QOL (1). Gill and Feinstein defined QOL as "inherently an attribute of the patient (or beholdee)" and therefore they recommended that QOL instruments should incorporate the subjective perceptions of the individual (1). This is consistent with the recent World Health Organization (WHO) definition of QOL as "the perceptions of individuals of their own position in life in the context of the culture and value systems in which they live, and in relation to their own goals, expectations, standards and desires" (2). QOL is probably best considered as a subjective, multi-dimensional, individually valued phenomenon.

It has been theorized that individuals determine their own QOL from the gap between their life circumstances and a standard for comparison (the 'multiple discrepancies theory' herein called the 'gap hypothesis') (3). Michalos has proposed six different 'gaps' that may be used to determine QOL (3). These gaps are defined as the difference between; 1) what one has and what one wants to have; 2) what one has and what is considered to be the ideal; 3) what one has and what a reference group has; 4) one's present circumstances and what one expects or expected them to be; 5) one's present QOL and the best QOL experienced in the past; 6) one's personal attributes and the attributes of one's environment (3). Of these, it is not yet clear which gap provides the most valid and meaningful measure of what individuals feel to be their QOL (4).

Much of the research on the concept and measurement of QOL has understandably been focused on adults, since children present special challenges to researchers. The perspectives of children are rarely sought because of the challenges posed by their differing levels of cognitive maturity. As such, the conceptual bases of many pediatric QOL instruments have been derived from adult research. However, if an individual's perspectives are a vital part of the determination of QOL, then the perspective of the child must be an important consideration in pediatric QOL research. It has also been suggested that caregiver reports may be important in providing significant additional information about a child's QOL (5).

The purpose of this study was to develop a measurement process of QOL that can be used to test the validity of the gap hypothesis for children with arthritis and other rheumatic conditions. In developing such a measurement process, we wish to determine for these children:

1. Which items best reflect QOL for use in a 'gap' interview?
2. What is the most understandable method for measuring personal QOL gaps?
3. What is the best method for ranking and weighting the importance of individual QOL items during the gap interview?

Materials and methods

General approach

We wanted to include all the attributes necessary for the measurement of QoL for children with arthritis. These comprise: i) individual selection of those aspects of life ("items") that have an impact on life quality, ii) weighting the chosen items in order of their contribution to life quality (so that the most important items are given the most weight), and iii) valuing the "gap" between one's life situation and an individual standard for each of these items. Based on the "gap hypothesis" this should provide a comprehensive understanding of a child's life situation to allow the health care team to deter-

mine those areas of life and health that need intervention. In this study we interviewed children with rheumatic disease and their parents to help determine the best way to measure all of these attributes.

Subjects

Eligible subjects included English-speaking patients (and 1 of their parents) attending the rheumatology clinics at The Hospital for Sick Children (SickKids) – a tertiary care centre. Children below the age of 6 years were excluded because of the variability in understanding verbal and written language at this age and the difficulty in ensuring full understanding of the items during the interview. Subjects were identified and approached consecutively from the clinics. A trained interviewer enrolled the families and conducted the interviews as the families waited for their child's appointment.

The Research Ethics Board at SickKids approved the study. Written informed consent was obtained from all families. Verbal assent was obtained from children between the ages of 8 and 16 years.

The study was conducted in two phases. The first phase involved the development of the interview questionnaire or the Gap Study (GapS) Questionnaire. In this phase we first generated a comprehensive list of items pertinent to the quality of life of children with chronic illnesses. This was followed by item reduction and subsequent preliminary formatting of the items and the scales used to determine the perceived gaps. In the second phase of the study, the list of items and the scales were pilot tested for their wording, as was the scale used to generate rankings and weightings of the items.

Phase 1 – Development of the GapS Questionnaire

Item generation

Items were generated through literature review, semi-structured interviews with participants and discussion sessions with participants and with the investigators.

A Medline (1966-2005) search strategy was used to identify literature about QOL and specifically about QOL in children. Searches were restricted to English language publications. Original articles and existing measures were reviewed with the aim of identifying domains and items relevant to QOL measurement. Existing measures including the Juvenile Arthritis Quality of Life Questionnaire (6), Health Utilities Index (7), Childhood Health Assessment Questionnaire (8), Child Health Questionnaire (9), Pediatric Quality of Life Inventory (10), Medical Outcomes Study 36-Item Short-Form Health Survey (SF-36) (11) and the EQ-5D (12) were reviewed. This process yielded 3 separate lists of items generated by 3 members of the research team individually reviewing the literature. All items were then collated and duplicate items discarded. Additional items thought to be particularly relevant to a child's QOL were discussed at regular sessions with the investigators. For example, items like worrying, spirituality and sex were subsequently incorporated into the list of items. To ensure the comprehensiveness of the list, items were initially categorized into 8 domains, including family, school, friends, self-care/function, hobbies/interests, health, emotions and miscellaneous. These were then re-categorized under the domains body functions and structure, activity and participation, and environmental factors, according to the WHO's International Classification of Functioning, Disability and Health (ICF) (13).

Semi-structured interviews were conducted with individual children and their parents to ascertain their feedback about the appropriateness and importance of items. In most cases, the child was interviewed first followed by a discussion with the accompanying parent. This ensured that each child was allowed to express his or her opinion regarding the items in the questionnaire. The same interviewer conducted all interviews, which also helped to ensure consistency between interviews. Each item in the list was explored with an open-ended discussion using the prompts "whether this is important to you" and "what do you understand by this question?".

All participants were given the opportunity to suggest additional items important to their QOL. These additional items were explored in depth with participants to ensure that they were not already included in the original item list.

Item reduction

Before family interviews were conducted, the research team deleted similar items found during our literature review and combined some items to minimize the number of items to be reviewed during family interviews.

The retained items were subsequently presented to the families interviewed. Each item in the list was individually evaluated during these interviews that were used to identify further items considered unimportant or redundant. Items identified as being unimportant by several respondents were then reviewed by the research team, with consideration of deleting them from the item list. This was an iterative process in which the research team met between groups of family interviews to adjust the item list. Subsequent families were therefore presented with refined item lists until no further changes were suggested.

Preliminary formatting

Following the generation and reduction of the list of items, the preliminary version of the interview questionnaire consisted of all remaining items. Each item was constructed in the form of a statement using the "third person technique" to reduce values implicit in the wording of the item. An example of the third person technique is: "Some children have very many friends, while other children have very few friends. How about you?". The items were then pilot tested for clarity of language and format.

Concurrent with the process of item reduction, problematic questions and alternative phrasing of these questions were discussed with the investigators on an ongoing basis and changes were incorporated in subsequent family interviews until the "final" wording of the items was agreed upon. These were then pilot tested in the second phase of the study.

Phase 2 – Pilot Testing

In the second phase of the study the amended items, the 'gap' response scales and the importance ranking (by sorting and ranking on a vertical visual analog scale – VAS) were pilot tested in a different group of families recruited for this purpose.

As discussed above, some investigators feel that individual valuation or weighting of items must be considered when evaluating QOL (14). We pilot tested – for wording and feasibility – the following technique for eliciting importance ratings for each item:

Participants were asked to rate how important each item in the list was to their quality of life on a 3-point ordinal scale ("Don't care", "A little important", and "Really important"). After completion of the listed items, participants were invited to add up to 5 additional items, thought to be "really important" to their QOL that were not already present in our item list.

Each item was represented on a card. Cards of items selected as "really important" by each participant were then laid out on a table, and each participant was asked to sort these cards in order of importance to their QOL.

Each participant was then asked to place the cards corresponding to the top 10 most important items along a vertical VAS.

The vertical VAS consisted of a 50 centimetre Velcro strip on a white foam board, with divisions ranging from 0 (least important to one's QOL) to 100 (most important to one's QOL).

Each card was attached to the Velcro strip with a piece of red yarn at the point along the scale corresponding to the perceived importance weighting for that item.

In the context of the 'gap hypothesis', it is unclear which standard reference should be used in formulating the gaps (4). We asked participants (children and parents separately) to consider their/their child's current life situation for each of their 10 most important QOL items and compare it to where

1. "I think I should be…"
2. "I would really like to be…"

with the response options ranging from "much worse" to "much better".

We also asked the participants to compare their current life situation to *other children their age* and to state where

3. "I think I am…"
4. "I think I should be…"
5. "I would really like to be…"

using the same response options.

The response options were presented using different wording and in different ways (including ordinal scales, double-anchored VAS, VAS with equal markings along the scale, etc.) to ascertain respondents' preferences based on clarity, ease of use, and sensibility. The participants also provided feedback on the ranking and weighting of items. The process was an iterative one; the individual participant's feedback was incorporated into the QOL interview process for subsequent respondents.

Results

Thirty-one children and 22 parents were recruited. The most common diagnosis amongst the children was juvenile idiopathic arthritis in 17 children. Six children had juvenile dermatomyositis and 2 children had systemic lupus erythematosus. The remaining children had Behcet's disease, idiopathic thrombocytopenic purpura, wrist pain, relapsing polychondritis, mixed connective tissue disease and vasculitis. There was a preponderance of females amongst the patients (22/31 = 71%) as well as the parents (17/22 = 77%). The mean age of the patients was 13.5 years (SD 3.0; range 6 – 17 years), with the mean age of the parents being 41.9 years (SD 5.7; range 30 – 51 years).

Phase 1

In total, 17 families were interviewed for the first phase of the pilot study.

Item generation

In total, 105 items were generated from the literature review and discussion sessions. Family interviews yielded an additional 5 items that were deemed to be important by children and their parents. For example sex, independence and "fitting in with peers" were identified as important issues by the adolescents. Items like "remembering to take my medicines" and "living with my illness" were important to the younger children. All items were then categorized by the investigators in two ways: under 8 domains of QOL - family, school, friends, self-care/function, hobbies/interests, health, emotions and miscellaneous, and under the headings – impairments of body functions and structures, activity limitations, participation and environmental factors – of the ICF (13).

Item reduction

Although the length of the item list had to be reduced for practical purposes, we were careful to ensure that all the domains listed above were still represented by relevant items. Following an iterative process, with subsequent groups of families, we retained 58 items (Table I).

Preliminary formatting

During this process, patients identified:

1. that some of the questions were inappropriately combined. For example, they felt it necessary to consider friends and family separately as well as school and work separately.
2. problems with the wording of some items. For example, "Some kids have a lot of pain. Other kids do not have this problem. How happy are you with the amount of pain you have?" Many participants especially parents felt that the word "happy" was inappropriately used in this question. Suggestions included "How do you feel about…?" or "How about you?" as alternatives.
3. problems with the length of some questions. For example, "Some kids can have trouble doing things they need to do at school or work. Other kids don't have trouble doing the things that they need to do at school or work. How happy are you with being able to do the things you need to do at school or work?" This question, and a few others like it, was shortened.
4. questions which were inappropriate for younger children. For example, questions dealing with boyfriend/girlfriend relationships were considered inappropriate for very young children.

As a result of suggestions from fami-

Table I.

ICF classification	Items retained after item reduction process (58)	Items in final version of GapS questionnaire (72)
Impairments of body functions and body structures	1. How you look	Whether you like how you look
	2. Physical growth	Your height Your weight
	3. Being physically able to do things that you enjoy doing	Being physically able to do everything you enjoy doing
	4. Being mentally able to do things that you enjoy doing	*Deleted*
	5. Taking care of yourself	Being able to take care of yourself (Taking care of yourself means doing things like getting dressed, going to the bathroom and taking a bath without help)
	6. Taking medicines or getting treatments	Having to get treatment or take medicine
	7. Side effects of treatment	Feeling sick when you get medicine or treatment
	8. Going to the doctor or hospital	Having to go to the doctor or hospital at all Having to go to the doctor or hospital a lot
	9. Sleep	Sleeping well at night
	10. Your memory	Being able to remember things Remembering to take your medicines
	11. How smart you are	Whether you see yourself as being smart
	12. Appetite	*Deleted*
	13. Enjoyment of food	Enjoying your food
	14. Pain	Being in physical pain
	15. Fatigue	Your energy levels
	16. Other symptoms of illness (eg. nausea)	Feeling unwell because of your illness
	17. Your marks/grades in school	Getting good marks at school
	18. Missing school or work	Missing school Missing work
Activity limitations and participation restriction	19. Satisfaction with your school or job	Being happy at school Being happy at work
	20. Spending time with your friends and family	Being able to spend lots of time with your family Being able to spend lots of time with your friends Enjoying the time you spend with your family Enjoying the time you spend with your friends
	21. Interests and talents (e.g. music, dance, sports, art, clubs and groups etc.)	Being able to join in hobbies and after-school activities like dance, sports, art, music, clubs and groups
	22. Achieving your goals	Achieving your goals
	23. Receiving recognition for your talents/abilities	Getting told that you have done a good job at something
	24. Entertainment, leisure time (e.g. reading, watching TV, movies, playing video games)	Reading, watching TV or movies, or playing video games
	25. Going out (e.g. shows, restaurants, sporting events, shopping)	Going to shows, restaurants, sports events or shopping
	26. Going on vacation or to camp	Going away for vacation or camp
	27. Being able to do the things you need to do at school or work	Being able to do the things you need to do at school Being able to do the things you need to do at work
	28. Being allowed to do things that you enjoy doing	Being allowed to do all the things that you like doing
	29. Having to move homes	Having to move homes a lot

Table I continues

Table I. *(continuation)*

ICF classification	Items retained after item reduction process (58)	Items in final version of GapS questionnaire (72)
Environmental factors	30. Satisfaction with the home you live in	Your life at home The house you live in
	31. Satisfaction with where you live (eg. Community, city, country, etc)	The area you live in
	32. The things you own (eg. Toys, books, clothes, games, etc)	How much stuff (toys) you have How much you like your stuff (toys)
	33. People pressuring you (e.g. parents or peers)	Feeling pressured by other people
	34. How you behave at home and at school	Your behaviour at school Your behaviour at home
	35. Worry about yourself	Worrying about yourself
	36. Worry about other people	Worrying about other people you know
	37. Personality (being funny, being shy, being outgoing, etc)	How you like your personality [Your personality is about what you're like as a person (shy, outgoing, funny, serious)]
	38. Your mood	Being happy
	39. Self esteem	How you feel about yourself
	40. How you feel about your sexual orientation	*Deleted*
	41. How you feel about your sexual identity	How you feel about being a boy or a girl
	42. Coping with problems	Being able to cope with your problems
	43. How you feel about your future	Caring about your future
	44. Culture/ethnicity	*Deleted*
	45. Spirituality/religion	The place religion has in your life
	46. Social conscience (e.g. caring about politics, social issues, environment)	Caring about the environment
	47. How much money you and your family have	How much money your family has
	48. The number of friends you have	Number of friends you have
	49. Your ability to make new friends	Being able to make new friends
	50. Relationships with peers (how you treat each other, supportiveness, closeness, etc.)	Getting along with other kids your age
	51. Having a boyfriend/girlfriend or being single	Not having a boyfriend or girlfriend
	52. Relationship with boyfriend/girlfriend	Being happy with your boyfriend or girlfriend
	53. Having pets	Having pets
	54. Relationship with parents/guardians	Getting along with your parents
	55. Relationships with your brothers/sisters	Getting along with your brothers/sisters
	56. Relationships with extended family members	Getting along with your grandparents, aunts, uncles or cousins
	57. Relationships with your teachers or babysitters	Getting along with your teachers
	58. Relationships with your friends	Having good friends
		*Being able to fit in *How much food you have to eat *Living with your illness *Being able to learn new things *Having independence (e.g. being able to drive)

*Denotes additional items identified by families to be important.

lies, amendments to the items were made during phase 1 – the final item list comprised a total of 72 items (Table 1).

Phase 2

In total, 14 families were interviewed for phase 2 of the study.

Pilot testing

No further amendments to the structure and wording of the items were required during this phase. Regarding the response sets, issues that arose included:

1. Similarities between response stems for the 'gap' scales. For example, children reading through the response stems quickly were not able to discern the differences between comparisons made to their current situation as opposed to comparisons made to other children their age.
2. VAS modifications for the 'gap' responses. When families were presented with different response options, feedback was generally positive about using the VAS for answering questions. Parents, in general, liked including a range of negative to positive numbers marked evenly on the VAS. Many children, however, found multiple divisions on a VAS confusing. The best compromise was to use a linear scale with 7 divisions.
3. Standardization of anchors at either end of the VAS. Each VAS was double-anchored. To maintain consistency, attempts were made to standardize the anchors used across items. Initially, word anchors only were used – "Much better" and "Much worse" at either end. To make this easier for the younger respondents smiling and frowning faces were added to reinforce the anchors.

Importance ranking

The 3-point ordinal scale for importance circumvented the need to develop different item lists for different ages. By allocating an importance rating to individual items, children self-selected age-appropriate items. After picking cards corresponding to their nominated "Really important" items, most children were able to sort up to 10 cards in order of importance but had difficulty when there were more than 10 cards. Subsequently, all children had no difficulty with placing the 10 cards along the vertical VAS once the task was clearly explained.

The performance characteristics of the GapS questionnaire in relation to other valid QOL measures was not part of this pilot study; it is currently being evaluated.

Discussion

We were able to develop an interview questionnaire as well as a technique of ranking and weighting items, to be used to elicit QOL in a way consistent with the gap hypothesis. Our process involved children and their parents in order to better reflect the very subjective nature of QOL.

As the literature remains inconclusive about which gap is most appropriate for estimating an individual's QOL, we elected to consider the gaps implicit in the WHO definition of QOL. Our chosen response stems allow an individual to consider their current situation in relation "to their own goals, expectations…and desires" (2). Additionally, the process we developed incorporates different standards against which children may compare themselves (i.e. their current situation or their peers' situations). We anticipate that the incorporation of 5 different gaps will allow us to determine – in future research – which gap, if any, is most predictive of what an individual feels globally to be their QOL.

As Gill and Feinstein have suggested, the measurement of QOL should incorporate the individual's subjective perceptions (1). One way of doing this is by allowing individuals to self-select items in quality of life measures as is done in the Patient Generated Index (PGI) (15), the McMaster-Toronto arthritis (MACTAR) patient preference questionnaire (16) and the Juvenile Arthritis Quality of Life questionnaire (JAQQ) (6). This results in the individualization of such measures, which may enhance responsiveness. The PGI measures the impact of a specific illness on the areas of patients' lives that they themselves consider important (15). The MACTAR allows patients to preferentially identify and rank their most important problems (16). The JAQQ – a measure of heath-related quality of life in juvenile arthritis patients, also allows patients to volunteer their own items so that the JAQQ can take into account items not included in the instrument itself or that become problems later (6).We allowed respondents to identify and rank items important to their QOL. In addition, respondents were also given the opportunity to nominate additional items not included in our item list, in an effort to more closely adhere to the recommendation made by Gill and Feinstein (1).

We feel that selecting, ranking and weighting the importance of items ought to be considered in determining the QOL of an individual. Value rankings have been introduced into a number of QOL measures because of the recognition that not all determinants of QOL are equally important or equally valued by people. This allows an individual to subjectively rank different determinants for him/herself. The 3-point ordinal scale worked quite well for allowing children to identify what was "really important" to their QOL. This also allowed us to retain a single item list rather than formatting versions for different ages. With the incorporation of importance ranking, children of different ages were able to pick out what was important to them accordingly, hence exercising their right for individual selection and prioritization. We were also able to quantify the child's perspective of the relative magnitude of importance of the "really important" items using the vertical VAS. Our preliminary results suggest that value rankings can be feasibly incorporated into QOL measures for children.

The gap approach was used as a conceptual basis for developing our QOL elicitation process. As Calman has suggested, QOL "measures the difference at a particular moment in time, between the hopes and expectations of the individual and that individual's present experiences" (17). It is possible for individuals to have a good quality of life despite major health problems (18). They may achieve this through reducing their expectations or by de-

creasing the value placed on those aspects of life that fall short of expectations (17). Hence, if the gap hypothesis is correct, an individual's quality of life may be dependent on their interpretation of the size of the gap and the value of the gap for different aspects of life. We hope that in future research we will determine whether our QOL interview process will provide health care providers with a feasible method of obtaining such QOL information directly from patients. If so, we will be able to validate the gap hypothesis – and we will be able to provide QOL information that is more in keeping with the construct put forward by the WHO than current so-called "health-related" QOL measures.

Additionally, to improve an individual's QOL, it may be important that the care or treatment will "narrow the gap between a patient's hopes and expectations and what actually happens" (17). If successful, our approach may be useful in determining those life aspects that could most beneficially be improved in order to maximize QOL.

The current study was conducted with pediatric rheumatology patients. The interview questionnaire may be limited in its generalizability to other patient populations. However, the composition of the research team included health care professionals from other medical and surgical disciplines to ensure the wider applicability of items in the questionnaire. Future studies will also address the validity of the GapS Questionnaire in a diverse clinical sample. Additionally, respondents were allowed to nominate supplemental items, further individualizing the GapS Questionnaire. The gaps measured in this questionnaire may not be a complete representation of an individual's QOL. Further research will be necessary to determine whether the GapS Questionnaire based on the gap hypothesis, does indeed provide a valid measure of QOL.

Conclusions

In summary, we have developed an innovative interview method for determining QOL in children with chronic rheumatologic conditions, based on the gap hypothesis. We will use this method in future research aimed to validate the gap hypothesis, and to develop a new, theory-based, method for determining QOL for this as well as other populations of chronically ill children.

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