UTILIZING DISCREPANCY THEORY TO QUANITATE QUALITY OF LIFE IN CHRONICALLY ILL CHILDREN

APPROVED BY SUPERVISORY COMMITTEE

Committee Chairperson's Name, Carlos Girod, M.D.

Committee Member's Name: Brian Feldman, M.D.

Committee Member's Name: Ramiah Subramanian, M.D., Ph.D.

UTILIZING DISCREPANCY THEORY TO QUANITATE QUALITY OF LIFE IN CHRONICALLY ILL CHILDREN

by

BRYN WEBB

DISSERTATION

Presented to the Faculty of the Medical School

The University of Texas Southwestern Medical Center at Dallas

In Partial Fulfillment of the Requirements

For the Degree of

DOCTOR OF MEDICINE WITH DISTINCTION IN RESEARCH

The University of Texas Southwestern Medical Center at Dallas

Dallas, Texas

May 25, 2007

Copyright

By

Bryn Webb

ABSTRACT

UTILIZING DISCREPANCY THEORY TO QUANTITATE QUALITY OF LIFE IN CHRONICALLY ILL CHILDREN

BRYN WEBB

The University of Texas Southwestern Medical Center at Dallas, 2007

Supervising Professor: Dr. Brian M. Feldman, Professor of Pediatrics, HPME and PHS, The Hospital for Sick Children, University of Toronto, Toronto, Canada

*As presented at the 2007 Pediatric Academic Societies' Annual Meeting in Toronto, Canada May 5-8, 2007

BACKGROUND: Quality of Life (QoL) is a ubiquitous yet poorly defined concept in clinical medicine. Many widely used instruments to measure QoL lack a theoretical basis and therefore may not provide accurate assessment. Multiple discrepancy theory advocates that net satisfaction is determined by evaluating the difference or 'gap' between current life circumstances and a standard of comparison. A QoL instrument based on discrepancy theory has been developed by our research group to evaluate children with chronic diseases.

OBJECTIVE: To evaluate the parent-child agreement for discrepancy theory items on the GAP QoL Questionnaire for children with chronic illness.

DESIGN/METHODS: Children and parents were recruited for this pilot study when attending regularly scheduled appointments in a variety of outpatient clinics at The Hospital for Sick Children (SickKids). Trained interviewers administered the GAP Questionnaire to parents- and to their children if older than age 10. Children and parents answered the questionnaire blind to each other's answers.

RESULTS: 77 participants (28 children, 49 parents: 24 parent/child pairs) were enrolled. The most important items for determining QoL generated by the GAP Questionnaire differed for children and parents (See Table). The overall weighted Kappa value for parent-child agreement on the GAP Questionnaire was 0.349 indicating moderate agreement. Items with the highest agreement were having pets (0.6962), getting along with brothers/sisters (0.5886), and the place religion has in your life (0.549).

Most Important Items			
Children		**Parents**	
Having good friendships	51.75	Being allowed to do all the things you like doing	34.38
Being happy most days	50	Getting told you have done a good job at something	33.92
Getting along with your parents	48.82	Being physically able to do everything you enjoy doing	32.75
Caring about your future	38.89	Enjoying time you spend with your family	31.9
Being able to take care of yourself	38.14	Having good friendships	31.27

Average weighted score listed.

CONCLUSIONS: Items used to determine important areas of discrepancy, as determined by the GAP Questionnaire, often differ between parents and children. Parent proxy reports are probably not desirable for highly personal and subjective constructs like QoL.

TABLE OF CONTENTS

PRIOR PUBLICATIONS AND PRESENTATIONS ……....7

LIST OF FIGURES ……...8

LIST OF TABLES …….....9

CHAPTER ONE – INTRODUCTION ……10

CHAPTER TWO-UTILIZATION OF THE GAP S QUESTIONNAIRE TO IDENTIFY DETERMINANTS OF QOL ……14

CHAPTER THREE – VALIDATION OF THE GAP S QUESTIONNAIRE …….31

CHAPTER FOUR – CONCLUSIONS AND RECOMMENDATIONS …….. 39

BIBLIOGRAPHY ……..40

VITAE …… …42

PRIOR PRESENTATIONS & PUBLICATIONS

Presentations:

Bryn Webb, **Pediatric Academic Societies' Annual Meeting,** Toronto, Canada, May 5-8, 2007.

Bryn Webb**, American Medical Association – Medical Student Section 4th Annual Student Research Poster Competition**, November 4, 2005, Dallas, TX.

Bryn Webb, Poster PresentationWinner, **42nd Medical Student Research Forum**, UT Southwestern Medical Center, 2004.

Publications:

Crawford GE, Holt IE, Whittle J, **Webb BD**, Tai D, Davis S, Marguiles EH, Chen Y, Bernat JA,Ginsburg D, Zhou D, Luo S, Vasicek TJ, Daly MJ, Wolfsberg TG, Collins FS. Genome-wide mapping of DNase hypersensitive sites using massively parallel signature sequencing (MPSS). Genome Res 2006; 16: 123-131.

LIST OF FIGURES

FIGURE ONE …….23

FIGURE TWO ……25

LIST OF TABLES

TABLE ONE ……24

TABLE TWO ……...26

TABLE THREE ……...28

TABLE FOUR ……..29

TABLE FIVE ……....36

TABLE SIX ……...37

TABLE SEVEN ……38

CHAPTER ONE
Introduction

UTILIZING DISCREPANCY THEORY TO QUANTITATE QUALITY OF LIFE IN CHRONICALLY ILL CHILDREN

The need for accurate measurement of quality of life (QoL) has become increasingly important as medicine advances. New treatments are now available for previously fatal conditions, and the measurement of QoL may serve as an endpoint in evaluating treatment efficacy. Additionally, detailed evaluation of QoL may aid both patients and physicians in setting priorities and decision-making regarding the use of medical interventions. QoL determination may provide additional information when rationing of resources is necessary or during end-of-life decision-making. (1)

Despite the well documented importance of QoL in the medical literature, confusion exists in the medical community as to the precise meaning of this term. Perhaps the main debate is whether QoL is a subjective or objective measure. Supporters of QoL as a subjective measure argue that only the individual can judge the quality of his or her life, and therefore, not only must this measurement be reported by him or her, but the values and judgments important in quantifying QoL must be up to the individual. Proponents of objective measurement believe the main determinants of QoL are common to all persons,

and therefore, QoL is not influenced by personal feelings, interpretations, or prejudice. (2)

Medical research to date has failed to properly define QoL, and hence, conclusions made about QoL in the medical literature may be inconsistent; for example the construct measured and called "QoL" in one setting may be inapplicable in a different setting that calls for a different construct. In Gill and Feinstein's critique of 75 articles on QoL measurements, only 15% of the reviewed articles actually define the term they purported to measure. (3) (Gill and Feinstein supported the subjective view of QoL.) More recently, Davis' review of pediatric QoL instruments identified eleven different types of definitions for QoL and health-related QoL. Additionally, only 3 of 38 instruments identified were constructed using an explicit theory of QoL. (4) These findings highlight the variability in available QoL instruments and the need for a unifying construct.

In response to the confusion in the medical community, the World Health Organization (WHO) formulated a definition of QoL. This definition describes QoL as an "individuals' perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards, and concerns." Furthermore, QoL is described as "a broad ranging concept, incorporating in a complex way individuals' physical health, psychological state, level of independence, social relationships, personal beliefs and their relationships to salient features of the environment." This WHO

definition highlights the multi-dimensional nature of QoL and supports the position that QoL is a subjective measure. (5)

Current widely used instruments to measure QoL focus on evaluating a series of items grouped within domains – as opposed to utilizing global measures. These domains are thought to represent the core components of QoL and often include: disease state and physical symptoms, functional status, psychological functioning, and social functioning. (6) Because the domains are pre-defined for all respondents (and the scoring of the items and domains has been pre-determined by the developers), most existing QoL measures might be considered as objective measures.

Jenney and Campbell stipulate that QoL domains must represent the priorities and perceptions of the individuals within the given population (7). It would seem, however – that for a truly subjective QoL measure – these domains must reflect the priorities of *each individual*. This makes the formulation of a subjective, generic instrument that measures fixed domains difficult for application to the individual patient.

The clearest way to measure subjective QoL, that addresses the preferences of the individual, is to use global scales. Currently used global measures include visual analog and time trade off scales amongst others. Measurement of time trade off stems from the field of health economics and assumes that a given person would prefer a shorter life of satisfactory health, or life, as opposed to a longer life with serious discomfort or handicap(8). Due to the simplistic nature of

these instruments (i.e. all elements of quality of life are considered together), critics of these measures argue that limited information may be gained.

The GapS interview questionnaire was developed by our group and is based on multiple discrepancy theory. Multiple discrepancy theory proposes that net life satisfaction is determined by evaluating the difference or 'gap' between current life circumstances and a standard of comparison. Michalos proposed six different gaps, which include the difference between: 1) what one has and what one wants, 2) what one has and what is considered to be 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, and 6) one's personal attributes and the attributes of one's environment (9). Calman applied this theory to QoL measurement when he suggested that QoL may be improved by either reducing expectations or increasing quality of the present life, i.e. narrowing the gap (10). The GapS questionnaire is a subjective measure, theory-based, that should provide much more detailed information about QoL than global rating methods.(11)

In this study we asked whether a "gap" approach – utilizing the GapS questionnaire – may provide an accurate assessment of subjective QoL in children with chronic disease when compared to QoL measured by other subjective global standards. We also attempted to determine the usefulness of a parent proxy when measuring QoL in children with chronic disease.

CHAPTER TWO
Utilization of the GapS Questionnaire to Identify Determinants of QoL

BACKGROUND

QoL is generally regarded as a multi-dimensional concept. QoL is not determined by a single factor, but is comprised of all areas of a person's life that he/she deems important. The decision about which aspects of life are important is an individual one. QoL is a subjective measure, and is best measured by one's own self. The use of parent proxies in pediatric patients further complicates the measure of QoL.

In this chapter, we attempt to utilize our new QoL tool, the GapS Questionnaire, to identify determinants of QoL in chronically ill pediatric patients. We also seek to examine parent-child agreement when using our instrument.

METHODS

PATIENT RECRUITMENT

Subjects were consecutively enrolled in this study when attending routine appointments in the Rheumatology, Diabetes, Epilepsy, Gastroenterology, and Cystic Fibrosis outpatient clinics, and the day-hospital outpatient unit at The Hospital for Sick Children (SickKids). Eligible participants included patients of any age and one parent. Patients ≥ 10 years of age completed all measures

independently and blind to their parent's responses. Subjects were excluded if they could not adequately read and speak English. This study was approved by the Research Ethics Board at SickKids; signed informed consent (and verbal assent where appropriate) was obtained for all participants.

DATA COLLECTION.

A trained interviewer administered the Gap Study (GapS) questionnaire along with other measures and a standard interview script was utilized (See Figure 1). Parents completing the survey were instructed to respond to the best of their ability *from the perspective of their child*. Interviewers evaluated ease of data collection for each participant so that data gathered from potentially problematic interviews was identifiable.

THE GAP STUDY (GAPS) INTERVIEW QUESTIONNAIRE

Completion of the GapS tool by parent or child requires the fulfillment of 3 steps.

First, 72 items representing a wide variety of criteria thought to be important in QoL (e.g. "Number of friends you have") are ranked on a 3 point scale ("Don't care", "A little important", or "Really important"). At the end of the ranking, the participant is encouraged to add up to 5 "supplemental items" not listed in the survey that he or she feels are important to QoL.

Second, the participant is asked to pick as many as 10 "most important" items (which may include the supplemental items). These 10 items are ranked on a vertical visual analog scale (VAS). This VAS looks like a thermometer with a 0 to

100 scale bounded at 0 = "least important to one's QoL" and 100 = "most important to one's QoL". The subject is then asked to place cards – each with one of their "most important" items written on it – on the scale to give a weight for each item.

Third, for each of the picked items, the participant answers 5 questions on a horizontal VAS. Each question enquires about a different aspect of the gap between the subject's perceived current state and some desired or standard realization. Responses numerically range from -3 "Much worse" to +3 "Much better." The five questions are:

- Compared to where I am now, I would really like my situation to be…
- Compared to where I am now, I expect my situation to be…
- Compared to other kids my age, I think my situation is…
- Compared to other kids my age, I expect my situation to be…
- Compared to other kids my age, I would really like my situation to be…

Statistical Analysis

When applicable, the JMP 5.1.2 computer software program was used for statistical analysis (SAS Institute, Cary, NC).

Analysis of Gap Study (GapS) Items

The ranking and analysis of GapS study items by parents and children was done by four methods. First, using a 3-point (0-2) scale the rankings for each item were averaged. A rating of "Don't care" was assigned a 0, "A little important" was assigned a 1, and "Really important" was assigned a 2. Second,

the median score for each item was identified after using a 4-point (0-3) score. Like the first method, a rating of "Don't care" was given a 0, "A little important" was given a 1, and "Really important" was given a 2, but, for the 4-point scale, if the item was chosen as part of the top 10, it was scored 3. Third, the percentage of times an item was chosen for the top 10 was identified. Fourth, an average was calculated for each item using the thermometer weight of each of the top 10 chosen items. If the item was not chosen to the top 10, it was assumed to have a zero weight.

Analysis of Parent-Child agreement

Using data collected from the 24 parent-child pairs, Kappa values were calculated to measure non-random parent-child agreement for each of the 72 items. The 4 point, 0-3 scale described above was used. Unweighted Kappa, Kappa with linear weighting, and Kappa with quadratic weighting were calculated using a program by Dr. Richard Lowry (Vassar): http://faculty.vassar.edu/lowry/kappa.html. The overall agreement for each parent-child pair ranking all items was also analyzed by calculating Kappa values with this program.

RESULTS

Patient Characteristics.

This study enrolled 92 total participants, 60 parents and 32 children (Table 1). After reviewing remarks the trained interviewers made about the administration

of the questionnaire, the final data set included 77 participants (49 parents, 28 children, and 24 parent-child pairs). Most excluded surveys were grossly incomplete or removed because the subject had marked difficulty understanding the survey.

Analysis of Gap Study (GapS) Questionnaire Items

The ranking of GapS Questionnaire items for parents and children by averaging ratings for a 3 point 0-2 scale (Method 1) revealed the distribution shown in Figure 2. The items children ranked lowest were #69- Having sex as an important part of life (mean score= 0.36), #22- How much stuff (toys) you have (0.57), #15- Feeling pressured by other people (0.61), #17- Moving homes a lot (0.64), and #33- Your height (0.71). Parents also ranked item #69 lowest (0.21). Other low rankings by parents were #70- Missing work (0.42), #71- Being happy at work (0.48), #67- Whether or not you have a boyfriend or girlfriend (0.50), #72- Being able to do the things you need to do at work (0.50), and #68- Being happy with your boyfriend or girlfriend (0.58).

The ranking of median scores using a 4-point (0-3) scale (Method 2) revealed 3 highly important items for children, all with a median score of 3: #5- having good friendships, #6- getting along with your parents, and #44- being happy most days. For parents, the highest median score was a 2, and this score was shared by 40 items. The lowest ranked items for children each with a median score of 0 were again #69 and #17. Two additional items had a median score of 0.5: #22- How much stuff (toys) you have and #71- Being happy at

work. For parents, the following items had a median score of 0: #67- Whether or not you have a boyfriend or girlfriend, #68- Being happy with your boyfriend or girlfriend, #70- Missing work, #72- Being able to do the things you need to do at work, #71- Being happy at work, and #69- Having sex as an important part of life.

The items chosen by children to be part of the top 10 with the highest percentages (Method 3) were again items #5 (57.14%), #6 (53.57%), and # 44 (53.57%). For parents, the items chosen to the top 10 with the highest percentage were #65- Being allowed to do all the things you like doing (43.75%), #12- Enjoying time you spend with your family (39.58%), and #14- Getting told you have done a good job at something (39.58%). 18 items for children were never chosen to be in the top 10. 5 items for parents also were never chosen (0%): #36- Having to go to the doctor or hospital at all, #45- Worrying about yourself, #50- How you like your personality, #70, and #72.

The most-important items for children and parents identified by the averaged thermometer weights (Method 4) are shown in Table 2.

Analysis of Parent-Child Agreement

The quadratic kappa values for each item ranged from 0.70 to -0.22 with an average of 0.18. Items with the highest and lowest quadratic kappa values are shown in Tables 3 & 4.

The quadratic kappa values calculated for each of the parent-child pairs ranged from 0.10 to 0.66 with an average of 0.35. There was no correlation

between age of the child and quadratic kappa for the parent-child pair (R square value 0.005, p-value 0.74).

DISCUSSION

Using data collected from our new Gap S Questionnaire, we have rated 72 possible determinants of QoL in chronically ill children. We have compared the data gathered from children to that which was gathered from parents to identify similarities and differences in the ranking of these determinants. We also measured parent-child agreement for each of the possible determinants and for each of the parent-child pairs.

In our study, the ranking of possible determinants of QoL by the first two methods highlighted components that were not contributory to QoL in our patient population. Notably, both parents and children ranked having sex as the least contributory item by Method 1; this may be a reflection of our population because most children were either pre-adolescent or early adolescent. Additionally, parents gave low ratings to other items that involved typical late-adolescent activities such as dating and work. Children ranked a variety of items as non-contributory including peer-pressure, moving homes, physical appearance, and owning possessions.

The ranking of items by Methods 3 and 4 identified components of QoL with the highest rankings given by children and parents. Children, not

surprisingly, felt having friendships and feeling happy were important components of having a good QoL. Both parents and children addressed the importance of a good parent-child relationship in impacting QoL. Interestingly, even though this survey was given to address the determinants of QoL in chronically ill children and a variety of issues related to illness were addressed, these health-related items for the most part were not ranked as highly contributory to QoL. It seems that determinants of QoL in chronically ill children may be remarkably similar to those expected in healthy children.

In our study, different items had vastly different levels of parent-child agreement. Items with the best agreement were items involving family structure- such as pets, siblings, and religion. The item with the worst agreement was having independence, but many items relating to friendships were also notable for poor agreement. Overall, parents were fair proxies for their children in completing the complete Gap S Questionnaire.

Although there are certain possible limitations to our study, we believe our results are justified. The participants in our study were not homogeneous; patients had a variety of diseases and were enrolled in multiple clinics at the Hospital for Sick Children. The variety in our patient population may be an asset, however, because we would like to utilize the Gap S Questionnaire to measure QoL in all pediatric patients living with chronic disease. An additional

possible limitation in our study is that all participants were recruited from a single hospital in Canada. On the other hand, The Hospital for Sick Children is a large, quaternary care facility located in the urban city of Toronto with patients from a variety of ethnicities and backgrounds. This diversity in patient population is an asset when assessing an instrument we wish to be applicable in many different patient settings in the future.

Here the Gap S Questionnaire has been utilized to identify determinants of QoL in chronically ill children. Data gathered from children as well as parent proxies was compared and the level of parent-child agreement was assessed.

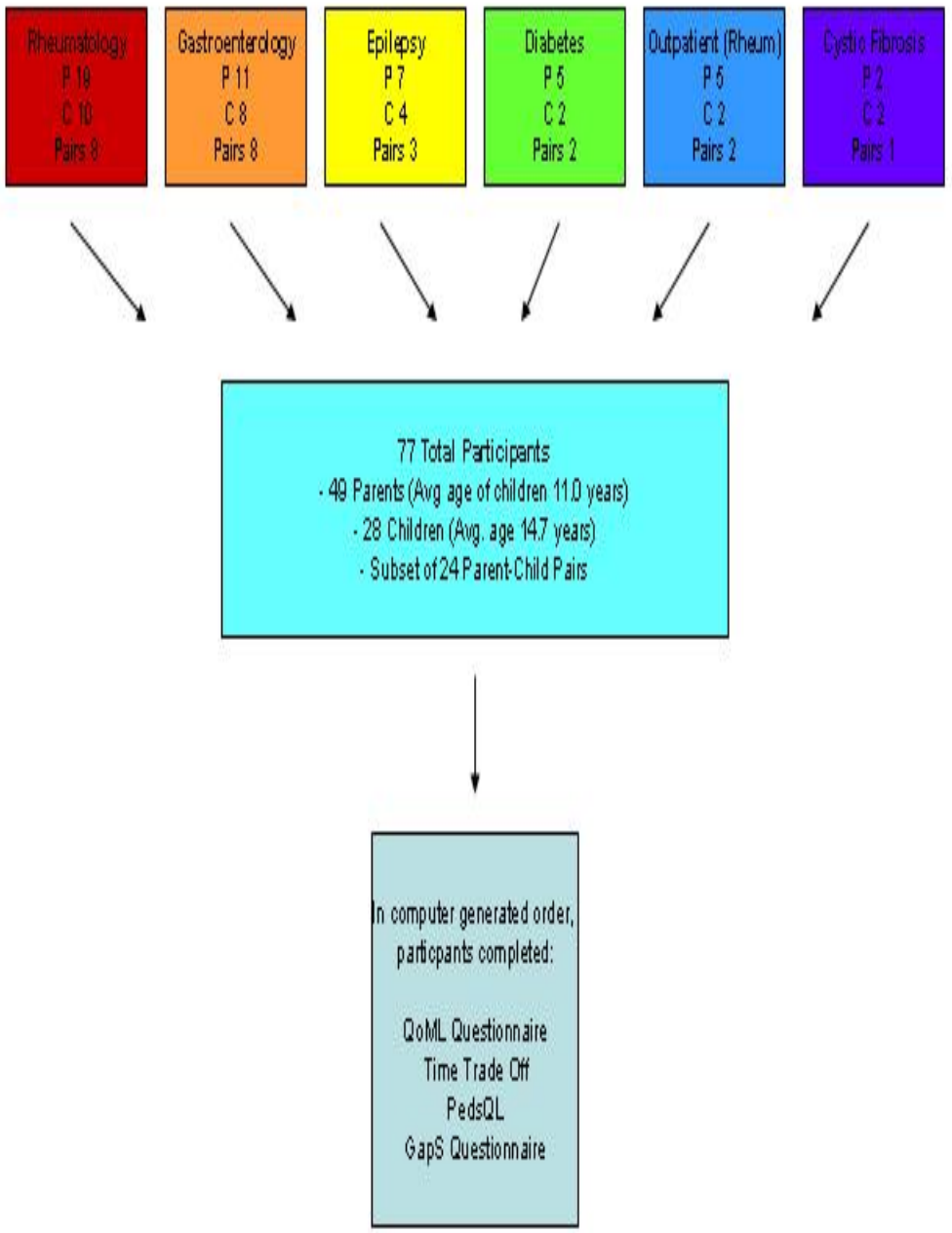

Rheumatology
P 19
C 10
Pairs 8
Gastroenterology
P 11
C 8
Pairs 8
Epilepsy
P 7
C 4
Pairs 3
Diabetes
P 5
C 2
Pairs 2
Outpatient (Rheum)
P 5
C 2
Pairs 2
Cystic Fibrosis
P 2
C 2
Pairs 1
77 Total Participants
- 49 Parents (Avg age of children 11.0 years)
- 28 Children (Avg. age 14.7 years)
- Subset of 24 Parent-Child Pairs
In computer generated order, participants completed:
QoML Questionnaire
Time Trade Off
PedsQL
GapS Questionnaire

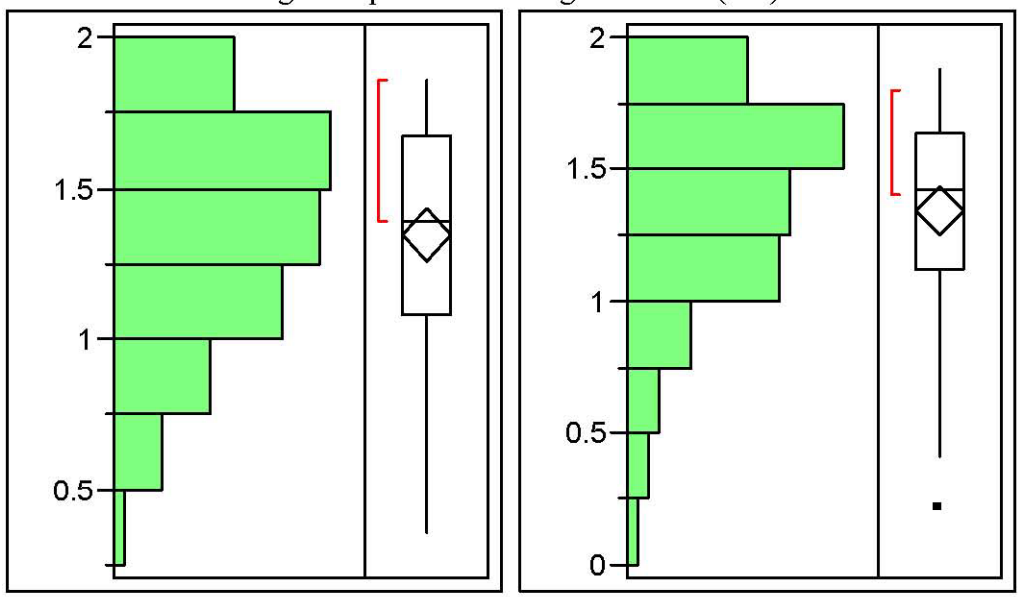
TABLE 1

Participant Characteristics

	Parents	Children	Parent-child pairs
Avg age of children	11.0 years	14.7 years	14.4 years
Age range of children.	3.7 months to 18 yrs 5.4 months	10 years 5 months to 18 years 5.4 months	10 years 5 months to 18 years 5.4 months
# of males/ # of females	25/24	16/12	13/11
Recruited from			
Rheumatology	19	10	8
Gastroenterology	11	8	8
Epilepsy	7	4	3
Diabetes	5	2	2
Outpatient	5	2	2
Cystic Fibrosis	2	2	1

FIGURE 2

Distribution of Averaged Gap S Items Using a 3-Point (0-2) Scale



The y-axis is the averaged item score (0-2 range). The x-axis illustrates the frequency of items with that average.

Table 2

Most Important Items for Parents and Children Determined by Averaged Thermometer Weights (Method 4)

Parents			Children		
Item #	Item Description	Weighted Avg	Item #	Item Description	Weighted Avg
65	Being allowed to do all the things you like doing	34.38	5	Having good friendships	51.75
14	Getting told you have done a good job at something	33.92	44	Being happy most days	50.00
25	Being physicially able to do everything you enjoy doing	32.75	6	Getting along with your parents	48.82
12	Enjoying time you spend with your family	31.90	57	Caring about your future	38.89
5	Having good friendships	31.27	26	Being able to take care of yourself	38.14
44	Being happy most	30.85	53	Achieving your	28.50

	days			goals	
11	Being able to spend lots of time with your friends	30.46	25	Being physically able to do everything you enjoy doing	27.82
13	Enjoying the time you spend with your friends	26.70	41	Remembering to take your medications	27.21
6	Getting along with your parents	22.29	43	Getting good marks at school	26.96
16	Having pets	21.85	12	Enjoying time you spend with your family	26.04

Table 3

Items With Highest Kappa Values for Parent-Child Agreement

Item #	Item Description	Quadratic Kappa
16	Having pets	0.70
7	Getting along with your brothers/sisters	0.59
49	The place religion has in your life	0.55
42	Being able to learn new things	0.52
28	Having energy	0.51
68	Being happy with your boyfriend or girlfriend	0.50
40	Being able to remember things	0.50
19	Your life at home	0.48
17	Moving homes a lot	0.45
70	Missing work	0.44
61	Being able to join in hobbies and after-school activities	0.43

TABLE 4

Items with Lowest Kappa Values for Parent-Child Agreement

Item #	Item Description	Quadratic Kappa
66	Having independence	-0.22
46	Worrying about other people you know	-0.21
2	Being able to make new friends	-0.12
13	Enjoying the time you spend with your friends	-0.09
1	Number of friends you have	-0.08
12	Enjoying time you spend with your family	-0.07
30	Living with your illness	-0.05
10	Being able to spend lots of time with your family	-0.05
58	Caring about the environment and what is happening in the world around you	-0.04
48	Whether you like how you look	-0.04
62	Reading, watching TV or movies, or playing video games	-0.03
52	Your behavior at home	-0.02
38	Enjoying your food	0
22	How much stuff (toys) you have	0

14	Getting told you have done a good job at something	0

CHAPTER THREE
Validation of the GapS Questionnaire

BACKGROUND

As a new QoL instrument, the next step in the development of the Gap S Questionnaire is to determine its construct validity. Construct validity seeks to determine whether a theoretical concept, which is not "operationally defined", may be measured by a specific device or tool. One method of validation utilizes correlation measures and factor analysis. If two instruments measure the same (or similar) construct(s), a positive correlation between them is predicted.(12)

Here we attempt to validate the GapS Questionnaire by comparing it to two other global measures of QoL, the Quality of My Life Questionnaire and Time Trade Off.

METHODS

PATIENT RECRUITMENT AND DATA COLLECTION

Patients and parents were recruited to participate in this study as described in Chapter 2. For this portion of the study, data collected from the GapS Questionniare was compared to data collected from other QoL measures.

Additional Measures

The Quality of My Life (QoML) Questionnaire

The QoML Questionnaire is a global subjective assessment of QoL and health-related QoL (HR-QoL). The questionnaire consists of two questions which are answered on horizontal visual analog scales. Scores may range from "The Worst" to "The Best." The first question asks "Overall, my life is…" and measures overall QoL. The second questions asks "Considering my health…" and measures HR-QoL.

Time Trade Off (TTO)

Originally developed by Torrance et al (1972), TTO is a utility measurement that quantifies how many years of remaining life a person would trade for a perfect health or life state. Patients in our study were assumed to have a remaining life expectancy of 70 years regardless of diagnosis. Patients were asked in an alternating fashion if they wished to trade off large numbers or small numbers of years until a point of indecision was identified. The TTO score is calculated as

The Number of Years at Point of Indecision / Remaining Years of Life at Current Health State.

Scores may range from 0 (life state equivalent to death) to 1 (perfect life state).

Statistical Analysis

The Jmp 5.1.2 computer software program was used for statistical analysis. With the data collected from children and parent proxies, R squared values were calculated to determine correlations (proportions of variance accounted for)

between the 5 gap scales of the GapS Questionnaire and the other measures. Correlations were made using weighted and unweighted gap scales. Unweighted gap scales were determined by averaging the gap scale ranking for all of the top 10 items. Weighted gap scales were determined by calculating the mean of the gap scale ranking multiplied by the thermometer weight for all of the top 10 items.

RESULTS

QUALITY OF LIFE MEASURES

The average scores for both parents and children for the Quality of My Life Questionnaire and Time Trade Off measures are listed in Table 5.

CORRELATIONS BETWEEN GAP SCALES OF GAP S QUESTIONNAIRE AND OTHER MEASURES

Correlations between the unweighted Gap scales and the Quality of My Life Questionnaire and Time Trade Off measures for children and parent proxies are shown in Table 6. Correlations for weighted Gap scales are shown in Table 7.

DISCUSSION

In this section, data gathered by children and parent proxies for the Gap S Questionnaire was compared to other global measures of QoL. Both weighted and unweighted gap scales were examined.

Using the unweighted gap scales, we are able to see that for children the best correlations are between Gap 1 and Gap 2 and the second question of the Quality of My Life Questionnaire, which measures HR-QoL. For parents, the best correlations are seen with the Gap 3 scale. However, for both parents and children, even the best correlations between the gap scales and other QoL measures do not account for the majority of the variance. This most likely means that the Gap S Questionnaire and other global measures of QoL are measuring somewhat different constructs.

With the weighted gap scales, the gaps with the best correlations for children and parent proxies are similar. Notably, for parents, many correlations improve when using the weightings, but for children this is not the case. This may reflect some difficulty for children in giving thermometer weightings for their top picked items.

The differences seen between parents and children using the different gap scales may be intuitive. Gap scales 1 and 2 utilize an internal reference ("compared to where I am now"), whereas the remainder of the scales use an external reference ("compared to other kids my age"). Because parents are external themselves, and thus acting as parent proxies, one might expect higher correlations with other measures when using gap scales with external references. Similarly, for children the simplest scale may be to compare their desires or wishes to their current status state using the internal reference scales of gaps 1 and 2. Additionally, some children may have not yet developed the capability to accurately compare themselves to other kids, thus making gap scales 3 to 5 more difficult to utilize.

The data shown here represents the first steps in determining the construct validity of the Gap S Questionnaire. Here all top 10 items chosen were included in the weighted

and unweighted gap scales analyzed. Much more analysis needs to be done to determine the accuracy of this instrument in measuring QoL, including analyzing gap scales with fewer top 10 items chosen. This construct validation will help to determine whether an instrument based on discrepancy theory may provide an accurate determination of QoL in pediatric patients with chronic disease.

TABLE 5

Average Scores for Comparison QoL Measures for Children and Parent Proxies

	Parents	Children
QoML Questionnaire:		
Avg QoL score	7.30	8.04
Avg HRQoL score	6.40	6.73
AVG TTO SCORE	0.83	0.85

Children Top 10	R-squared	P-value							
	Gap 1 U		Gap 2 U		Gap 3 U		Gap 4 U		Gap 5 U	
QoFML Corrected Overall Score	0.003911		0.001529		0.053012		0.011218		0.028914	
HrQOL Corrected Overall Score	0.135365	0.059	0.159113	0.0393	0.000278		0.072031		0.086918	
TTO Years at Point of Indecision	0.059695		0.002884		0.00052		0.006168		0.000018	

Parents Top 10										
	Gap 1 U		Gap 2 U		Gap 3 U		Gap 4 U		Gap 5 U	
QoFML Corrected Overall Score	0.022398		0.000744		0.194328	0.0022	0.000301		0.030728	
HrQOL Corrected Overall Score	0.131751	0.0122	0.089549		0.17047	0.0043	0.033842		0.084776	
TTO Years at Point of Indecision	0.024658		0.002665		0.146558	0.0086	0.014332		0.046285	

Key

0.10 < R squared < 0.15

0.15 < R squared < 0.20

The 5 Gaps

1) Compared to where I am now, I would really like my situation to be...

2) Compared to where I am now, I expect my situation to be...

3) Compared to other kids my age, I think my situation is...

4) Compared to other kids my age, I expect my situation to be...

5) Compared to other kids my age, I would really like my situation to be...

Children Top 10	R-squared	P-value							
	Gap 1 W		Gap 2 W		Gap 3 W		Gap 4 W		Gap 5 W	
QoFML Corrected Overall Score	0.010263		0.005862		0.034503		0.002963		0.022991	
HrQOL Corrected Overall Score	0.110372	0.0904	0.149235	0.0465	0.000171		0.069483		0.073335	
TTO Years at Point of Indecision	0.035279		0.007434		0.000754		0.001958		0.000008	

Parents Top 10										
	Gap 1 W		Gap 2 W		Gap 3 W		Gap 4 W		Gap 5 W	
QoFML Corrected Overall Score	0.012346		0.002121		0.231042	0.0007	0.000301		0.018194	
HrQOL Corrected Overall Score	0.159454	0.0054	0.124645	0.0149	0.190413	0.0024	0.049581		0.111691	0.0232
TTO Years at Point of Indecision	0.041822		0.011428		0.139912	0.0104	0.00606		0.070576	

Key

0.10 < R squared < 0.15

0.15 < R squared < 0.20

0.20 < R squared < 0.25

The 5 Gaps

1) Compared to where I am now, I would really like my situation to be...

2) Compared to where I am now, I expect my situation to be...

3) Compared to other kids my age, I think my situation is...

4) Compared to other kids my age, I expect my situation to be...

5) Compared to other kids my age, I would really like my situation to be...

CHAPTER FOUR
Conclusions and Recommendations

Conclusions:

- Parents and children rank the importance of many GapS items differently.
- The overall parent-child agreement for the complete GapS survey is variable. (Quadratic kappa range indicates slight to substantial agreement).
- Certain GapS survey items have moderate to substantial agreement. Others have extremely poor agreement.
- Different Gap scales better approximate other QoL measures for parents and children. For parents, the Gap 3 scale is best, whereas Gaps 1 and 2 may be a better measure for children.
- More analysis is needed to determine the validity of the GapS instrument.

Recommendations/ Future Studies:

The next step in development of the Gap S Questionnaire will be questionnaire simplification and item reduction. At present, the Gap S Questionnaire is a lengthy instrument. Because of the ability to add supplemental items, items with low ratings and poor agreement may be eliminated. Additionally, after further analysis, certain gap scales that may be redundant or non-contributory should be thrown out.

The updated version of the Gap S Questionnaire will then need to be tested in a variety of clinics to make sure it is accurate in measuring QoL in pediatric patients living with a variety of chronic diseases.

Bibliography

1. **Eiser C, Morse R** 2001 The measurement of quality of life in children: past and future perspectives. J Dev Behav Pediatr 22:248-256
2. **Dijkers M** 1999 Measuring quality of life: methodological issues. American journal of physical medicine & rehabilitation / Association of Academic Physiatrists 78:286-300
3. **Gill TM, Feinstein AR** 1994 A critical appraisal of the quality of quality-of-life measurements. Jama 272:619-626
4. **Davis E, Waters E, Mackinnon A, Reddihough D, Graham HK, Mehmet-Radji O, Boyd R** 2006 Paediatric quality of life instruments: a review of the impact of the conceptual framework on outcomes. Developmental medicine and child neurology 48:311-318
5. 1995 The World Health Organization Quality of Life assessment (WHOQOL): position paper from the World Health Organization. Social science & medicine (1982) 41:1403-1409
6. **Spieth LE, Harris CV** 1996 Assessment of health-related quality of life in children and adolescents: an integrative review. Journal of pediatric psychology 21:175-193
7. **Jenney ME, Campbell S** 1997 Measuring quality of life. Archives of disease in childhood 77:347-350
8. **Hunt SM** 1997 The problem of quality of life. Qual Life Res 6:205-212
9. **Michalos A** 1985 Multiple discrepancies theory (MDT). Social Indicators Research 16:347-413

10. **Calman KC** 1984 Quality of life in cancer patients--an hypothesis. Journal of medical ethics 10:124-127

11. **Gong GW, Young NL, Dempster H, Porepa M, Feldman BM** 2007 The Quality of My Life questionnaire: the minimal clinically important difference for pediatric rheumatology patients. The Journal of rheumatology 34:581-587

12. **Cronbach LJ, Meehl PE** 1955 Construct Validity in Psychological Tests. Psychological Bulletin 52:281-302.

Curriculum Vitae

EDUCATION

M.D. University of Texas Southwestern Medical School, 2007
B.S. University of Texas at Austin, Major: Molecular Biology, 2002

HONORS

Medical School:
American Pediatric Society/Society for Pediatric Research Student Fellowship, 2006
Minnie L. Maffett Scholarship Award sponsored by the Texas Federation of Business and Professional Women's Club, 2006
NIH DNA Day Ambassador, 2005
NIH IRTA Research Fellowship for Medical Students, 2004-05
UT Southwestern Medical Student Research Forum 1st Place Poster Winner, 2004
NIH Summer Research Fellowship for Medical Students, 2003
UT Southwestern Medical Student Summer Research Fellowship, 2002

Undergraduate:
Phi Beta Kappa, 2002
National Merit Scholarship at the University of Texas at Austin
Summer Honors Undergraduate Research Program at Harvard Medical School, 2001
University of Texas at Austin Undergraduate Research Fellowship, 2000
Summer Undergraduate MSTP Research Program at the University of Iowa, 2000

BASIC SCIENCE RESEARCH PROJECTS

- High-throughput identification and analysis of DNase hypersensitive sites. Advisor: Dr. Francis Collins at the NIH-National Human Genome Research Institute (NHGRI). 2004-05.
- Characterization of a death receptor-induced, but caspase-independent pathway of programmed cell death. Advisor: Dr. Michael Lenardo at the NIH- National Institute of Allergies and Infectious Diseases (NIAID). 2003.
- Analysis of the role of ch-IAP1 (inhibitor of apoptosis protein) in v-Rel transformation. Advisor: Dr. Henry Bose at the University of Texas at Austin. 2000-02.
- Generation of a Sideroflexin 3 knockout construct as a model of sideroblastic anemia. Advisor: Dr. Mark Fleming at Harvard Medical School. 2001.
- Analysis of the ability of the type III secretion system of Salmonella typhimurium to delay assembly of host NADPH oxidase in human macrophage. Advisor: Dr. William Nauseef at the University of Iowa College of Medicine. 2000.

CLINICAL RESEARCH PROJECTS

- Quantification of quality of life in chronically ill children. Advisor: Dr. Brian Feldman at The Hospital for Sick Children at the University of Toronto. Fall 2006.
- An analysis of the effects of bedside case presentations on patients' perception of their medical care at a large county hospital. Advisor: Dr. Carlos Girod at the University of Texas Southwestern Medical School. 2006-07.

- Clinical analysis of pharmacologic interactions between phenytoin and temozolomide in brain tumor patients. Advisor: Dr. Karen Fink at University of Texas Southwestern Medical School, Department of Neurology. 2002.

PUBLICATIONS

Crawford GE, Holt IE, Whittle J, **Webb BD**, Tai D, Davis S, Marguiles EH, Chen Y, Bernat JA,Ginsburg D, Zhou D, Luo S, Vasicek TJ, Daly MJ, Wolfsberg TG, Collins FS. Genome-wide mapping of DNase hypersensitive sites using massively parallel signature sequencing (MPSS). Genome Res 2006; 16: 123-131.