Published in final edited form as:

J Urol. 2008 October ; 180(4 Suppl): 1700–1704. doi:10.1016/j.juro.2008.03.117.

Parent Perspectives of Health Related Quality of Life in Adolescents With Chronic Kidney Disease Due to Underlying Urological Disorders: An Assessment Using the Child Health Questionnaire-Parent Form 50.

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Abstract

Purpose: Little is known about health related quality of life in adolescents with chronic kidney disease due to urological anomalies. We assessed generic health related quality of life in this population using a validated parent proxy instrument.

Materials and Methods: In this multicenter prospective cohort study the Child Health Questionnaire-Parent Form 50, a generic health related quality of life parent proxy instrument, was administered to 92 parents of adolescents 10 to 18 years old with chronic kidney disease. Mean summary measure and scale scores in adolescents with urological anomalies were compared to scores in those with other causes of kidney disease and in a representative American population sample.

Results: The cohort included 35 adolescents with an underlying diagnosis of congenital urological anomaly and 57 with another cause of kidney disease. There were no significant differences in mean scale or summary measure scores between the 2 diagnostic groups. When compared to a representative population sample, adolescents with congenital urological anomalies scored significantly worse on the questionnaire physical summary measure (44.5, 95% CI 39.9 – 49.1 vs 52.0, 95% CI 51.1–52.9, $p = 0.002$), and on the Child Health Questionnaire scales physical functioning, role physical, general health perceptions, family activities and parental emotional impact.

Conclusions: As assessed by Child Health Questionnaire-Parent Form 50, parents reported that adolescents with chronic kidney disease due to urological disorders scored lower on the physical summary measure than a population based reference sample and they reported a negative effect on family activities and parental emotional well-being. These findings suggest that the Child Health

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Questionnaire-Parent Form 50 could provide a family based assessment of generic health related quality of life in adolescents with urological disorders.

Keywords

kidney; quality of life; questionnaires; parent; adolescent

INTRODUCTION

Few groups have examined HRQOL in children with CKD due to underlying urological abnormalities. While great improvements have been made in survival, long-term functional outcomes in this group are compromised and there is a need to assess the factors affecting HRQOL in this population.¹ Approximately 57% of children with CKD have an underlying structural abnormality, such as posterior urethral valves, vesicoureteral reflux or dysplastic kidney.²

Compared to children with CKD resulting from other medical diseases, children with structural urological disease are clinically different in many ways. They are generally diagnosed earlier in life.³ While poor growth and development are frequently seen in CKD, it has its greatest impact on males with obstruction and dysplasia.⁴ In addition, urinary symptoms, including incontinence, the need for catheterization and delayed toilet training, may negatively affect quality of life in children with urological causes of kidney disease. Many of these children also experience associated anatomical abnormalities, multiple urological surgeries and hospitalizations.

Previous research has demonstrated that adolescents with CKD regardless of etiology have impaired functional health status. The reasons for this are being investigated and currently CKD associated anemia appears to be one of the strongest predictors of poor HRQOL in these children.¹ However, the effect of urological symptoms, such as urinary incontinence and bladder catheterization, has not been fully evaluated. There are few studies specifically examining those with underlying urological disease. In the urological literature studies suggest that enuresis may be negatively associated with self-esteem and emotional well-being, and recently 2 condition specific tools were developed to assess this impact.^{5,6} An incontinence symptom severity instrument was also recently developed for use in the adolescent population.⁷ Results of psychosocial outcomes in the bladder exstrophy population are mixed.^{8,9} With increasing prenatal diagnoses of congenital urological disease and a longer life span in children born with these abnormalities a family based approach is needed to assess the impact of these urological illnesses on quality of life and the outcomes of treatment for them.¹⁰

In a previous study we examined generic HRQOL in adolescents with CKD using CHIP-AE, a generic HRQOL instrument completed by the child.¹¹ In this study we examined generic HRQOL in the same cohort of adolescents with CKD, this time using the well validated generic HRQOL parent proxy instrument, CHQ-PF50.¹² Children with kidney disease due to underlying urological disorders generally have an earlier diagnosis of kidney disease, and may be more likely to experience social and physical sequelae. Thus, we hypothesized that parents of children with CKD due to underlying urological disorders would report that their

children had worse HRQOL in psychosocial and physical functioning compared to children with other causes of kidney disease and to a population based reference sample.

MATERIALS AND METHODS

Patient Population:

The Functional Outcomes in Adolescent CKD study is a prospective cohort study of physical and psychosocial functioning in adolescents with CKD. Participants were recruited from 7 pediatric nephrology centers at tertiary care hospitals in the United States, including The Johns Hopkins Hospital, Baltimore, Maryland; Children's Hospital of Philadelphia, Philadelphia, Pennsylvania; Montefiore Medical Center, Bronx, New York Medical College, Valhalla, North Shore University Hospital, Manhasset and Schneider Children's Hospital at North Shore-Long Island Jewish Health System, New Hyde Park, New York; and Robert Wood Johnson Medical School, New Brunswick, New Jersey.¹³ Institutional review board approval was obtained for the protocol at each participating center. Parental consent and child assent was obtained for each participant.

Eligibility criteria were 1) patient age 10 to 18 years, 2) attendance at a participating outpatient clinic during the study period of October 1998 to March 2003, 3) advanced stage 2 or stages 3–5 CKD according to the National Kidney Foundation Kidney Disease Outcomes Quality Initiative (as estimated by a glomerular filtration rate of less than 75 cc per minute per 1.73 m² by the Schwartz formula), dialysis treatment (hemodialysis or peritoneal dialysis) or patient was a stable kidney transplant recipient, and 4) child and parent or guardian was capable of reading English or Spanish at the fourth grade level.

CHQ-PF50:

CHQ-PF50, a generic HRQOL parent proxy instrument, was administered at baseline to parents or guardians of adolescents 10 to 18 years old with CKD. Additional information on demographics and medical history was obtained, including the primary diagnosis, child age and gender, hematocrit, albumin, transplant status, CKD stage, parent gender and race, and biological parent status.

CHQ is a set of generic quality of life instruments designed for children 5 to 18 years old.¹² CHQ measures 11 multidimensional scales and 2 summary measures. The parent form is available in 2 lengths (50 or 28 items) and a youth version is also available for adolescents 10 to 17 years old, which consists of 87 items. We used the parent form with 50 items, which requires approximately 20 minutes to complete. Scores can be analyzed separately as CHQ scale scores or combined for an overall physical and psychosocial summary measure score. Scales are scored from 0 to 100 with higher scores indicating better quality of life. The 2 summary measures are transformed, so that they may be compared to the mean $\pm$ SD of 50 $\pm$ 10 seen in the general American population.¹²

Normative values are available for the scale and summary measure scores, and benchmarks for the parent reported CHQ versions are also available for some conditions. Previous studies have tested construct validity, convergent validity and sensitivity to change. Internal

consistency was 0.62 to 0.94.¹⁴ CHQ focuses on functioning and in this way it is similar to SF-36®. The Appendix shows a description of CHQ-PF50 summary scores and subscales.

Statistical Analysis

Statistical analysis was performed using Stata® 9 statistical software. Outcome variables were defined as the scores obtained in each CHQ-PF50 summary measure and scale. Children receiving dialysis treatment were excluded because previous HRQOL research in children with CKD has shown a strong negative association between dialysis treatment and HRQOL.^{13,15} Potential covariates were child age and gender, hematocrit, albumin, transplant status, CKD stage, parent gender and race, and biological parent status. Associations between the hypothesized covariates and each diagnosis category using the t, Fisher exact and chi-square tests were explored. Bivariate comparison of mean summary measure scores and scale scores between the 2 diagnostic groups was performed using the t test. Based on the available fixed sample size there was approximately 64% power to detect a difference of 5 points, which was considered by the developers of CHQ-PF50 to be a clinically and socially significant difference between the 2 diagnostic groups. Bivariate comparison of mean summary measure scores and scale scores between the congenital urological disease group and an available published representative population sample with an age range of 5 to 18 years was also performed using the t test.

RESULTS

The cohort included 35 adolescents with an underlying diagnosis of congenital urological anomaly and 57 with another cause of kidney disease. Detailed information on the underlying urological diagnosis in 20 of 35 participants was retrospectively collected at 1 center. The most common diagnosis was posterior urethral valves (9 patients), followed by vesicoureteral reflux (5), dysplastic or hypoplastic kidney (3), obstructive uropathy (2) and cloacal outlet obstruction (1). In the category of other causes of kidney disease the underlying diagnoses were chronic glomerulonephritis in 15 patients, focal segmental glomerulosclerosis in 10, polycystic kidney disease in 6, autoimmune disease in 2, other in 19 and unknown in 5. Table 1 lists the characteristics of the study participants. Mean age and gender of the child, gender of the parent, biological parent status, hematocrit, transplant status, albumin and CKD stage were similar between the 2 diagnostic groups. However, the subgroup with congenital urological disease had a statistically significantly larger proportion of white parents (88.6 vs 57.1, $p = 0.002$).

There were no significant differences in mean scores for any CHQ-PF50 summary measure or scale between the 2 diagnostic groups. However, when compared to a representative population sample, parents reported that adolescents with congenital urological anomalies scored lower on the CHQ-PF50 physical summary measure (44.5, 95% CI 39.9–49.1) vs 52.0, 95% CI 51.1–52.9, $p = 0.002$), and on the physical functioning, role physical, general health perceptions, family activities and parental emotional impact scales (table 2).

DISCUSSION

In this analysis of generic HRQOL in adolescents with CKD due to underlying urological anomalies parents reported that their children had poorer HRQOL than that reported by parents of children in the general population. This was observed in many areas of HRQOL, including physical functioning. There were no statistically significant differences seen in HRQOL between adolescents with CKD due to urological anomalies and HRQOL in those with other causes of kidney disease. In addition to urological symptoms such as incontinence and bladder catheterization, poor HRQOL outcomes in this population may also be due to renal disease. This was a group of adolescents with CKD and it has been demonstrated in previous studies that one of the strongest predictors of HRQOL in children with CKD is anemia.¹ Poor growth and development are common in children with CKD, which may also have a role in the impaired QOL outcomes.

Previously we had analyzed HRQOL data on the same cohort of children using the child reported generic HRQOL instrument CHIP-AE.¹¹ That analysis also demonstrated that children with congenital urological disease did not have lower HRQOL than their peers with other causes of kidney disease. Findings in that study were consistent with the current analysis when comparing diagnostic groups to each other but the results differed in other important ways. CHIP-AE analysis showed no significant HRQOL impairment in children with congenital urological abnormalities compared to population based norms. In contrast, our new analysis demonstrates that CHQ-PF50 detected statistically significant impairments in multiple HRQOL domains, especially those related to physical functioning of the child, and the impact on the parent and family life. This suggests that CHQ-PF50 may be more sensitive than CHIP-AE for detecting impaired HRQOL in children with urological disease. These differences may also be in part explained by the different health status instruments that were used and the different perspectives of the child and parent respondents.

There are many well validated, adolescent appropriate tools to assess generic HRQOL in adolescents with urological disease but to our knowledge the most valid tool to use has not been identified. Some of the most widely used instruments are CHQ, PedsQL™, Functional Status II-R and CHIP-AE.^{12,16–18} Some instruments are completed by the child and others are completed by a parent as a proxy for the child. Interestingly although parents often make health care decisions for their children, they are frequently discordant when reporting QOL. In general parents and children tend to have more agreement when rating physical HRQOL compared to emotional or social HRQOL.¹⁹ Parents may also report that illness in a child has more negative impact on QOL than the child reports.¹⁹ This may be due to different perspectives, life experiences and expectations between parents and children. While child and proxy reports may be discordant, they also complement each other.

A strength of this study is that we used a generic HRQOL instrument, which allows comparison not only to normal population based data, but also across disease states. For example, in a recent study using CHQ-PF50 to assess HRQOL in adolescents after transplantation the mean physical functioning score after liver transplantation was 88.8 and the general health perceptions score was 49.8.²⁰ In adolescents with kidney transplants the physical functioning score was 80.7 and the general health perceptions score was 50.4.

Benchmarks for other disease states may also be compared to our data. The mean physical summary measure score in adolescents with CKD due to urological anomalies in our study is 44.5 (95% CI 39.9–49.1). In comparison, the mean physical summary measure score in children is 42.1 for juvenile rheumatoid arthritis, 47.7 for asthma, 47.7 for epilepsy and 57.6 for attention deficit hyperactivity disorder.¹²

Although generic tools are useful for these comparisons, they may not be sensitive enough to identify more subtle impairments related to specific symptoms. A disease specific measure may be more sensitive to detect clinical changes due to treatment or differences among subgroups with a particular disease. A limitation of this analysis is the paucity of data on surgical history and urological symptoms, which would have been helpful to further characterize the urological disease subgroup and potentially identify additional causes of impaired HRQOL. In an ongoing study we are prospectively administering the CHIP-AE and the CHQ parent and child forms in a group of children with well defined urological disease and symptoms. In addition, a symptom specific tool or module of disease specific questions could be developed.

In this era of increasing emphasis on evidence-based medicine patient based assessment of outcomes will become increasingly important. Generic HRQOL instruments might be useful in research and potentially in clinical settings. Instruments such as CHQ-PF50 may allow pediatric urologists to quantify what many observe through clinical experience. They may also lead us to new ways to help improve care in these patients. A valid approach to measuring HRQOL in this population will be important in the future to assess the success and usefulness of different surgical, medical and behavioral interventions in children with congenital urological disorders. For example, potential applications of this knowledge could be to plan the timing of interventions to occur before an age when adverse effects on HRQOL become prominent and help provide information to families and physicians to use in the medical decision making process.

CONCLUSIONS

As assessed by CHQ-PF50, parents reported that adolescents with CKD due to urological disorders scored lower on the physical summary measure than a population based reference sample and they also reported a negative effect on family activities and parental emotional impact. These findings suggest that CHQ-PF50 could provide a family based assessment of improvements in generic HRQOL after urological interventions. Further research could explore the reasons for impaired HRQOL in physical functioning to identify modifiable factors predicting HRQOL.

ACKNOWLEDGMENTS

Marie Diener-West provided statistical advice and Korina FlorCruz assisted with the manuscript.

Study received institutional review board approval at each participating center.

Supported by the Kidney and Urology Foundation of America, NIH Roadmap Johns Hopkins Multidisciplinary Clinical Research Career Development Award Grant K12 HD049104 (JLD), a National Kidney Foundation of Maryland Professional Development Award (JLD) and NIH/NIDDK UO1 DK066174 Chronic Kidney Disease in Children CKiD (ACG, SLF).

Appendix:: Description of The CHQ-PF50 (Adapted from the CHQ Manual12):

Scales and Summary Measures	Description
Physical Functioning	Limitations in physical activities such as sports, mobility, and self-care due to health problems.
Role/Social Limitations due to Emotional or Behavioral Difficulties	Limitations in school work or activities with friends due to emotional or behavioral problems.
Role/Social Limitations due to Physical Health	Limitations in school work or social activities due to physical health problems.
Bodily Pain and Discomfort	Severity and frequency of bodily pain or discomfort.
General Behavior	Severity and frequency of behavioral difficulties.
Mental Health	Frequency of depressed, anxious or positive affect.
Self-Esteem	Satisfaction with school and athletic ability, relationships with friends and family, appearance and overall life.
General Health Perceptions	Parent's assessment of the child's general health compared to other children, and expectations for the child's health in the future
Emotional Impact on Parent	Severity of suffering and worry experienced by the parent due to the child's health, emotional, behavior or learning problems.
Time Impact on Parent	Limitations in time the parent had for his or her own personal needs due to the child's health problems.
Family Activities	Impact of child's health or behavior problems on family activities, conflict or arguments in the home.
Physical Health Summary Measure	Scale scores are standardized, aggregated and normed to generate a score reflecting overall physical health status.
Psychosocial Health Summary Measure	Scale scores are standardized, aggregated and normed to generate a score reflecting overall psychosocial health status.

Abbreviations and Acronyms:

CHIP-AE	Child Health and Illness Profile-Adolescent Edition
CHQ	Child Health Questionnaire
CHQ-PF50	CHQ-Parent Form 50
CKD	chronic kidney disease
HRQOL	health related quality of life

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Table 1:

Characteristics of the study groups:

Characteristic	Congenital urological anomaly (n=35)	Other diagnoses (n=57)	p-value	Statistical test used
Age (years) Mean (SD)	14.1 (1.86)	14.0 (2.08)	0.85	t-test
Child Sex (% male)	27 (77.1%)	33 (57.9%)	0.06	Chi squared test
Parent Sex (% female)	29 (82.9%)	47 (82.46%)	0.96	Chi-squared test
Biological parent (%)	32 (91.4%)	54 (94.7%)	0.67	Fisher's exact test
Parent race (% white)	31 (88.6%)	32 (57.1%)	0.002	Fisher's exact test
Hematocrit Mean (SD)	34.0 (4.29)	34.6 (5.42)	0.62	t-test
Transplant (%)	22 (62.9%)	31 (54.4%)	0.43	Chi-squared test
CRF (%)	13 (37.1%)	26 (45.6%)	0.43	Chi-squared test
Albumin Mean (SD)	4.1 (0.38)	4.1 (0.50)	0.70	t-test
CKD Stage (%)				
Stage I	3 (10%)	12 (21.4%)	0.06	Fisher's exact test
Stage II	10 (33.3%)	19 (33.9%)		
Stage III	14 (46.7%)	14 (25%)		
Stage IV	1 (3.33%)	10 (17.9%)		
Stage V	2 (6.7%)	1 (1.8%)		

Table 2:

CHQ-PF50 Summary Measure and Scale Means Comparing the Congenital Urological Anomaly Group to a Representative U.S. Population Sample (adapted from CHQ Manual, Pages 264–266)¹²

CHQ-PF50 Summary Measures and Scales	Congenital Urological Anomaly (n=35) Mean (95% CI)	Representative U.S. Population Sample (n=380) Mean (95% CI)	p-value (t-test)
Physical Summary Measure	44.5 (39.9–49.1)	52.0 (51.1–52.9)	0.0023
Psychosocial Summary Measure	50.3 (47.4–53.3)	51.0 (50.0–51.9)	0.72
Physical Functioning	86.1 (78.4–93.8)	96.2 (94.7–97.7)	0.01
Role Physical	79.0 (67.8–90.3)	93.6 (91.6–95.5)	0.01
General health perceptions	44.8 (39.4–50.3)	73.1 (71.3–74.9)	0.0000
Bodily pain	78.9 (72.2–85.5)	81.6 (79.5–83.6)	0.45
Family activities	77.3 (70.0–84.5)	89.7 (87.9–91.6)	0.0002
Role Emotional/behavior	86.7 (79.2–94.2)	92.9 (91.1–94.7)	0.0536
Parental impact-time	85.3 (78.4–92.2)	87.9 (85.9–89.9)	0.47
Parental impact-emotional	59.9 (48.7–71.1)	80.4 (78.3–82.4)	0.0008
Self-esteem	75.1 (69.3–80.8)	79.7 (78.0–81.4)	0.12
Mental health	75.4 (71.4–79.5)	78.5 (77.1–79.8)	0.19
Behavior	77.8 (72.7–82.8)	74.6 (72.9–76.2)	0.26