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Quality of Life for Children With Functional Abdominal Pain: A Comparison Study of Patients' and Parents' Perceptions

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ABSTRACT

OBJECTIVE. Children with chronic abdominal pain of nonorganic origin, termed functional abdominal pain (FAP), experience school absences and social withdrawal and report impaired physical ability. The aim of this study was to assess patients' and parents' perceptions of health-related quality of life (QoL) for children with FAP.

METHODS. Between October 2002 and November 2003, 209 children (including 125 girls; age: 11.2 ± 3.5 years) and 209 parents were recruited from a pediatric referral center. At the time of their initial evaluations, participants completed a validated, health-related QoL instrument (Pediatric Quality of Life Inventory), which was scored on a scale of 0 (poor) through 100 (best). Children with FAP ($n = 65$) and their families were compared with control groups of healthy children ($n = 46$) and children with histologically proven inflammatory bowel disease (IBD) ($n = 42$) or gastroesophageal reflux disease (GERD) ($n = 56$).

RESULTS. Children with FAP had self-reported QoL scores (score: 78) that were similar to those for children with GERD (score: 80) or IBD (score: 84). Children with FAP had lower QoL scores than did healthy children (score: 88). Parents of children with FAP reported lower QoL scores, compared with their children's scores (scores: 70 vs 78).

CONCLUSIONS. Children with FAP reported lower QoL, compared with their healthy peers, and had the same QoL scores as did children with IBD or GERD. Parents' perceptions of QoL for children with FAP were lower than their children's self-reported scores. These findings highlight the clinical significance of FAP and may provide insight into one facet of the disease's biopsychosocial etiology.

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Key Words

abdominal pain, functional, children, parents, quality of life

Abbreviations

FAP—functional abdominal pain
IBD—inflammatory bowel disease
GERD—gastroesophageal reflux disease
PedsQL—Pediatric Quality of Life Inventory
PCDAI—Pediatric Crohn's Disease Activity Index
QoL—quality of life

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FUNCTIONAL ABDOMINAL PAIN (FAP) is a prevalent disorder affecting children and young adults. Children with FAP may experience significant school absenteeism, family disruption, and social withdrawal, and they may demonstrate features of anxiety and depression.¹ Patients who suffer from functional gastrointestinal disorders may also endure sleep difficulties, headaches, dizziness, and fatigue.² Because objective clinical end points are rarely available for gastrointestinal disorders, patient-based outcome assessments such as measures of health-related quality of life (QoL) and self-reported symptom ratings are increasingly important.³ Studies of adults suffering from functional disorders such as irritable bowel syndrome revealed self-reported QoL scores comparable to those reported by patients with inflammatory bowel disease (IBD).⁴ These data highlight the potentially debilitating severity of gastrointestinal diseases, regardless of the cause of abdominal pain.

There is evidence that FAP rarely is a self-limiting condition and, after 5 years, one third of children continue to experience symptoms.⁵ There is also evidence that FAP in childhood leads to increased health care utilization in young adulthood.⁶ Irritable bowel syndrome alone affects 70 million adults in the United States and is responsible for \$30 billion dollars each year in health care and lost wage costs.⁷

Children may experience FAP symptoms for >1 year before adequate relief is achieved.⁸ Extensive evaluation and delayed diagnosis of a disease with no well-accepted treatment create a frustrating dilemma for patients, parents, and practitioners. Data from Crushell et al⁹ suggest that parents' perceptions and acceptance of severe FAP as a biopsychosocial disease affect disease prognosis favorably. It is unknown whether frustration with diagnostic uncertainty and ineffective treatment modalities affects parents' perceptions of QoL or exacerbates the presentation of FAP symptoms.

Despite these associations and observations, there have been no formal studies of the perceptions of QoL among children with FAP and their families, compared with children with chronic organic gastrointestinal diseases associated with abdominal pain. The aim of this study was to assess health-related, self-reported, and parent-perceived QoL among children with FAP, compared with a control population of healthy children and a group of children whose abdominal pain was associated with organic causes such as IBD and gastroesophageal reflux disease (GERD).

METHODS

Assessment Instrument

After informed consent and assent (when appropriate) were obtained, the Pediatric Quality of Life Inventory (PedsQL) was administered to children and their parents at a tertiary pediatric care center, at the time of initial

evaluation.^{10,11} The PedsQL is a health-related QoL tool; it uses generic core scales that comprise parallel child self-reports and parent proxy reports. The PedsQL has been validated among children as young as 5 years of age.

The 23-item PedsQL generic core scales encompass physical (8 items), emotional (5 items), social (5 items), and school (5 items) functioning. A 5-point response scale is used (0 indicates never a problem, and 4 indicates almost always a problem). Items are reverse-scored and transformed linearly to a scale of 0 to 100 (0 = 100, 1 = 75, 2 = 50, 3 = 25, and 4 = 0), with higher scores indicating better health-related QoL.

The PedsQL was read to children <7 years of age by the investigator. Parents and their children completed the PedsQL separately before physician evaluation. Separate reports were used because child self-reports are based on perceptions of internal states, whereas parent reports reflect the child's observable behaviors.

Study Population

Study patients designated as having FAP, GERD, or IBD were all children who presented to a pediatric tertiary referral center for initial evaluation of their gastrointestinal complaints. Eligibility criteria for children to enter the study were as follows: (1) age of 5 to 18 years, (2) male or female, (3) no history of attention-deficit disorder, (4) no history of cognitive delay, (5) comprehension of English, and (6) no concomitant chronic illness. Study patients designated as having FAP met consensus criteria for FAP known as ROME II criteria,¹² including at least 12 weeks of (1) continuous or nearly continuous abdominal pain for a school-aged child or adolescent; (2) no or only occasional relation of pain to physiologic events (eg, eating, menses, or defecation); (3) some loss of daily functioning; and (4) evidence that the pain is not feigned. Study patients designated as having GERD were subsequently diagnosed on the basis of endoscopic and histologic findings consistent with inflammation. Study patients designated as having IBD met both clinical and histologic criteria for either ulcerative colitis or Crohn's disease. A standardized scoring scale that is used routinely was available for Crohn's disease, ie, the Pediatric Crohn's Disease Activity Index (PCDAI).¹³ This was used to assess the severity of Crohn's disease at the time of the initial evaluation. Healthy control patients were recruited from a community-based, general pediatrics office at the time of a routine physical examination.

Outcomes

The primary outcome measure of the study was determination of QoL for children with FAP, compared with healthy control subjects and subjects with abdominal pain resulting from other chronic gastrointestinal disorders, such as IBD and GERD. Secondary outcome measures included parents' assessments of their children's

QoL. Additional outcome measures included differences in perceptions between children and their parents.

Data Analyses

Analysis of variance was used to compare continuous outcomes, and the χ^2 test was used to compare generic core scales between different study groups. Numerical data are expressed as mean \pm SEM. QoL scores were compared with independent-sample *t* tests. All *P* values are 2-sided, with *P* values of $<.05$ being considered statistically significant. The institutional review board of Atlantic Health System approved this study.

RESULTS

Study Group

Between October 2002 and November 2003, overall 209 children (including 125 girls; age: 11.2 ± 3.5 years) and 209 parents completed the PedsQL at the time of evaluation. Results for children with FAP ($n = 65$) and parents were compared with those for healthy children ($n = 46$) and control patients with organic causes of abdominal pain, ie, IBD ($n = 42$), Crohn's disease ($n = 33$; mean PCDAI score: 29 ± 11.2 ; range: 10–62.5), or GERD ($n = 56$). The groups were similar with respect to gender, race, parental marital status, and socioeconomic status (Table 1). Most parent respondents were mothers (82%). On direct questioning, mothers reported a positive self-history of either "nervous stomach" or migraine headache in 44% of FAP cases.

Primary Outcome

Children with FAP had self-reported overall QoL scores that were similar to those for children with GERD (78 vs 80; not significant) or IBD (78 vs 84; $P = .07$). There were no differences in QoL scores between Crohn's disease and ulcerative colitis. Children with FAP had lower QoL scores than did healthy children (78 vs 88; $P < .05$) (Fig 1). Children with FAP had physical scores lower than those for healthy children (73 vs 87; $P < .05$) and children with GERD (73 vs 85; $P < .05$) and similar to those for children with IBD (73 vs 81; not significant) (Table 2). Children with FAP had emotional scores lower than those for healthy children (77 vs 88; $P < .05$) (Table 2).

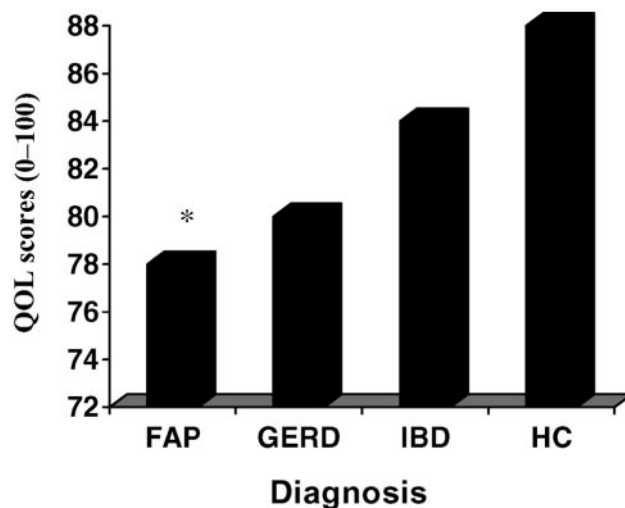


FIGURE 1
Children's self-reported QoL scores. HC indicates healthy children. * $P < .05$ versus HC.

Secondary Outcomes

Parents of children with FAP reported overall lower QoL scores, compared with their children's scores (70 vs 78; $P < .05$) (Fig 2). Parents of children with FAP reported the lowest QoL scores for their children, compared with other parents (children with IBD: 70 vs 78; $P = .07$; children with GERD: 70 vs 79; $P < .05$; healthy children: 70 vs 89; $P < .001$) (Fig 3). Parents of children with FAP reported lower social scores, compared with parents of healthy children (67 vs 93.2; $P < .001$), children with GERD (67 vs 88; $P < .05$), and children with IBD (67 vs 78; $P < .05$) (Table 2).

DISCUSSION

This study demonstrates the significant effect FAP has on QoL among children. Children with FAP have QoL lower than that of healthy children and similar to that of patients with demonstrable organic gastrointestinal diseases. The impairment in QoL is not limited to the patients, because parents perceive their children as having the lowest QoL, compared with all other groups.

The complex interaction of physiologic, psychological, and social factors known to play a role in functional bowel disorders may contribute to the lower QoL scores for children with FAP.¹⁴ There is evidence that children

TABLE 1 Demographic Features of Patient Population

	FAP	IBD	GERD	HC
Population, no.	65	42	56	42
Age, y ^a	10.9 \pm 2.8	12.9 \pm 1.3	11.8 \pm 3.1	11.9 \pm 4.6
Gender, % male	46	45	52	48
Symptom duration, mo ^b	18 \pm 6.4	16.1 \pm 2.3	13.3 \pm 4.5	NA

HC indicates healthy children; NA, not applicable.

^a No significant differences were noted between groups with respect to age.

^b No significant differences were noted between groups with respect to symptom duration.

TABLE 2 Individual PedsQL Scores for Different Diagnostic Categories

	PedsQL Scores							
	FAP		GERD		IBD		HC	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD
Child self-report								
Total	78.1 ^{a,b}	13.4	79.9	14.0	83.8	13.2	87.7	14.7
Physical	73.0 ^{b,c}	10.2	84.7	14.7	80.6	14.3	86.8	16.8
Emotional	77.3 ^b	10.3	83	15.1	78.1	16.5	88.6	19.7
Social	69.8	14.1	72.3	13.2	71.2	13.9	75.7	18.7
School	70.8	10.6	68.1	12.2	73.5	13.6	75.6	22.0
Parent report								
Total	70.1 ^{b,d}	7.9	78.6	10.3	77.8	12.1	88.7	15.1
Physical	76.9	11.8	83.9	13.3	81.5	11.5	78.1	14.0
Emotional	75.2	18.1	73.9	16.4	75	12.2	82.8	18.4
Social	67.1 ^e	12.3	87.5	14.2	77.5	14.7	93.2	16.7
School	72.2	15.8	73.9	15.1	74.5	12.1	68.9	15.8

HC indicates healthy children.

^a No significant difference, compared with the GERD and IBD groups.

^b Significantly lower, compared with the healthy control group ($P < .05$).

^c No significant difference, compared with the IBD group.

^d Significantly lower, compared with the healthy control and GERD groups ($P < .05$).

^e Significantly lower, compared with all parents ($P < .05$).

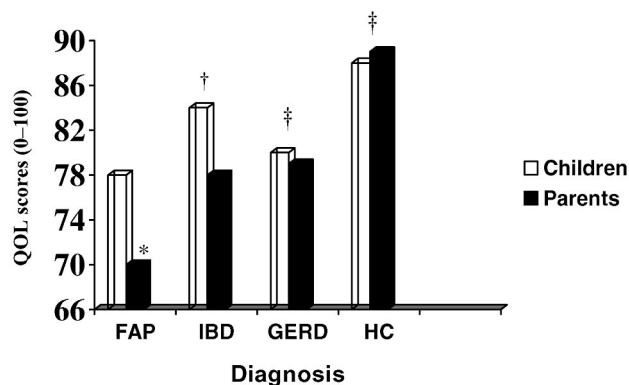


FIGURE 2

Differences in perceptions of QoL by children versus parents. HC indicates healthy children. * $P < .05$; † $P = .06$; ‡ = not significant.

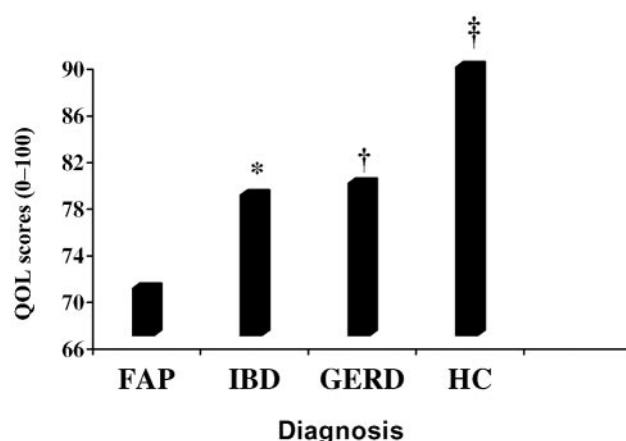


FIGURE 3

QoL for children as scored by parents. HC indicates healthy children. * $P < .05$; † $P = .06$; ‡ = not significant.

with FAP have visceral hyperalgesia, a condition that could be demonstrated with a gastric barostat.¹⁵ The same investigators reported that children with hyperalgesia had an increased tendency toward anxiety, compared with healthy children, and that the degree of anxiety increased as the duration of symptoms increased. This anxiety may contribute to the impaired QoL we report.

Patient-based outcome assessments, such as measures of health-related QoL and self-reported symptom ratings, are increasingly deemed to be important end points in caring for patients.³ Using the National Longitudinal Study in Adolescent Health, in which data on self-reported symptoms were available for 20 000 adolescents, investigators sought a relationship between frequent abdominal pain and depression.¹⁶ In that study, 16% of all adolescents were at risk for depression, but the risk of depression increased to 45% among adolescents with

abdominal pain. Compared with healthy children with “rare” abdominal pain (less than once per week), those with pain >4 times per week reported increased incidence of missing school >10 times per year, decreased sense of energy, increased crying, and greater feelings of sadness and loneliness. Children with frequent abdominal pain were less likely to participate in active sports and school activities and more likely to consider their lives a failure, compared with those with no abdominal pain. These negative associations with frequent abdominal pain might have effects on the lower emotional scores children with FAP reported in the study.

Additional data from the present study revealed that children with FAP had QoL scores similar to those for children with active IBD, whose PCDAI scores indicated moderate to severe disease activity at the time of initial

evaluation. This finding is consistent with a previously published adult report describing no difference in QoL between patients with a functional bowel disorder syndrome and those with IBD.⁴ In limited pediatric data assessing the QoL of children with IBD, self-reported concerns pertained to treatment options and future health care implications.¹⁷ It is conceivable that similar concerns exist among children with FAP who have undergone extensive evaluation and endured ineffective medical treatments for prolonged periods.⁸

In our study, parental perceptions were especially revealing with respect to the significant discrepancy between QoL scores reported by children with FAP and those reported by their parents. Although Walker and Greene¹⁸ indicated that mothers of children with recurrent abdominal pain displayed greater anxiety, depression, and somatization, Garber et al¹⁹ noted that mothers of children with recurrent abdominal pain reported more child somatic symptoms than did the children themselves. A study of parental responses to child illness behavior revealed that parents encouraged children to adopt a sick role for gastrointestinal symptoms more than for upper respiratory disease.²⁰ In their report, Crushell et al⁹ described how parental perception has a significant role in disease progression and noted that parents' acceptance and perception of FAP as a biopsychosocial illness decreased recurrence of symptoms in severe cases. Parental reinforcement of illness behaviors because of the perceived decreased QoL may represent one component of the biopsychosocial etiology of FAP.

Also noted in the present study is the finding that not only parents of children with FAP but also families of children with GERD and IBD reported lower perceptions of QoL for their own children, compared with parents of healthy control children. These findings are consistent with other studies involving parents of children with chronic diseases, who may be in need of more psychoeducational support interventions.²¹

Parental social concerns might benefit most from targeted education, because they contributed most to the decreased PedsQL scores that parents of children with FAP reported. Specific questions on the PedsQL included "getting along with others," "other kids not wanting to be his or her friend," "getting teased by other children," "not able to do things other children his or her age can do," and "keeping up when playing with other children."

Social learning may also play a powerful role in the etiology of functional bowel disorders among children.²² In our study, >44% of mothers reported "nervous stomach"; these parental experiences and perceptions of their own functional bowel disorders may establish a behavior model from which children with similar symptoms draw cues.²²

In this study, we found that children with FAP have a lower QoL, compared with healthy children. In addition,

children with FAP have the same QoL as children with other chronic gastrointestinal diseases. These findings suggest that, regardless of the cause of abdominal pain, QoL is impaired at the time of the initial evaluation. Parents' perceptions of a significantly decreased QoL may reflect the severity of the disease itself, frustration with the evaluation process and treatment modalities, or the parents' personal experience with functional symptoms. The disparity in QoL assessments between parents and their children may be a factor in the psychosocial genesis of the disease process. Prospective epidemiologic studies focusing on the true prevalence of FAP in childhood and its effect on QoL, including psychological profiles, need to be performed.

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CONGENITAL GLUTAMINE DEFICIENCY

“An article in this issue of the *Journal* illustrates, once again, the potential for wider implications of what is probably a rare metabolic disorder. Haberle and colleagues report on two unrelated infants who had dramatically reduced levels of glutamine due to a deficiency of glutamine synthetase, the enzyme that catalyzes the conversion of glutamate to glutamine. Both infants had profound cerebral disease that included malformations of the brain, and both died during the neonatal period. . . . [A]n interesting and important feature of the report by Haberle and colleagues is that the condition was identified in the infants because of reduced glutamine levels, rather than increased glutamate levels. Presumably, their normal levels of glutamate reflect the many other pathways available for glutamate degradation. It is very unusual to diagnose an amino acid disorder solely on the basis of a decrease in the level of a specific amino acid. Consequently, the interpretation of amino acid analysis is usually directed toward increased, rather than decreased, levels. Could amino acid disorders be missed because of this selectivity? The present report dramatically shows the value of closely observing low as well as high levels. A more comprehensive interpretation of amino acid findings will probably result in identifying other as yet unknown genetic causes of disease—and will continue our march toward a more basic understanding of human disease.”

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