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Long-term disease-specific quality of life in adult anorectal malformation patients[☆]

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Abstract

Background: Fecal and urinary incontinence may differently influence various aspects of quality of life (QOL). The main aim of the present study is to determine whether fecal and urinary incontinence measured at time 1 of the study will predict QOL at time 2 (after 4 years), above and beyond the prediction already explained by fecal and urinary incontinence at time 2.

Methods: Thirty-six adult patients from the Italian Parents' and Patients' Association for Anorectal Malformations answered items about urinary and fecal incontinence at time 1 of the study and completed the Hirschsprung Disease/Anorectal Malformation Quality of Life questionnaire after 4 years from the first questionnaire. Two sets of hierarchical regression analyses were conducted with fecal and urinary incontinence serving as predictors of QOL and the different areas of QOL from the Hirschsprung Disease/Anorectal Malformation Quality of Life serving as outcome variables.

Results: The principal findings indicated that fecal continence is a strong predictor of QOL in the areas of social functioning, emotional functioning, and body image and that urinary incontinence predicted sexual functioning.

Conclusions: It seems that one's past experience with fecal incontinence is extremely relevant to current QOL, especially for body image. Urinary incontinence contributed less in explaining QOL in our patients, but because it is very relevant for sexual functioning, it should not be disregarded.

Quality of life (QOL) has become an important issue in the medical community as well as in support groups for the chronically ill. Pediatric surgeons working with anorectal malformation (ARM) patients have great responsibilities not only in regard to the surgical procedure itself, but also in following-up with patients. Many patients who undergo repair of an ARM complain of some degree of functional defecating disorder and urinary incontinence. These 2 problems may have a significant influence on the QOL of ARM patients.

A number of studies have focused on the influence of fecal incontinence and its long-term consequences on QOL in children and adolescents [1-8], whereas fewer studies have considered the same issue in adult patients [6,9-12].

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Even fewer have focused on the influence of urinary incontinence on QOL in patients with ARMs [11], even though it is reported that more than 40% of all patients with ARMs have urinary tract anomalies [6]. Over time, fecal and urinary incontinence may influence various aspects of QOL differently.

Measures of QOL rely primarily on subjective data reported by patients. Similarly, other measures potentially correlated with QOL such as perceived severity of the malformation or functional control are also based on self-reports. This raises the issue of how to verify the reliability of these self-reports in terms of their stability over time and in terms of the validity of the correlations with other variables.

The main aim of the present study was to determine, in a sample of Italian adults who underwent an operation for ARM in the neonatal period, whether fecal and urinary incontinence as measured at time 1 would predict ARM-specific QOL at time 2 (4 years later).

In particular, we aimed to:

1. determine the frequencies of specific types of ARMs (ie, fistula types), using the Krickenbeck classification system, the degree of fecal and urinary incontinence reported by patients, as well as patients' perceptions of the severity of their malformations;
2. examine the correlations between fecal and urinary incontinence as measured at time 1 of the study and various aspects of QOL measured at time 2 using a QOL measure that also includes questions about fecal and urinary incontinence to determine whether fecal and urinary incontinence are stable over time and with which areas of disease-specific QOL they are most highly correlated; means and standard deviations (SDs) for the QOL areas for each fistula type will also be reported;
3. examine correlations among patients' perception of severity of the malformation and QOL of life areas; and, finally,
4. determine whether fecal and urinary incontinence measured at time 1 of the study will predict the different aspects of QOL at time 2 (after 4 years), above and beyond the prediction already explained by fecal and urinary incontinence at time 2. In analyzing these relationships, we controlled for the perception of the severity of the malformation.

1. Materials and methods

All patients who participated in this study belong to the Italian Parents' and Patients' Association for Anorectal Malformations (AIMAR). Members of this association are people residing in Italy who were born with ARMs and who underwent surgical procedures for these malformations in surgical centers across the country.

In 2003, 62 adult patients from the AIMAR association completed a brief questionnaire and an informed consent form sent by mail. Patients were asked to provide demographic information, as well as information about fecal and urinary incontinence and their specific type of malformation (ie, fistula type) (for the AIMAR questionnaire, see Aminoff et al [13]).

With respect to fecal incontinence, patients were asked to report how frequently they were soiling during a week using a 4-point response scale with 1 = never soiling, 2 = occasionally soiling (<3 times a week), 3 = frequently soiling (>3 times a week), and 4 = totally incontinent. For urinary incontinence, patients were asked to report how continent they were using a 3-point response scale with 1 = totally continent, 2 = occasionally incontinent, and 3 = totally incontinent. To assist patients in identifying their malformation type, they were asked to choose one alternative from a list of anomalies based on the Krickenbeck classification system on fistulas [14]. They were also asked to report whether they had associated or sacral problems using a dichotomous yes/no response scale.

In addition, patients were asked to report whether they have ever followed a bowel management program. Possible answers for this question were "yes," "no," "it's not necessary," or "I do not understand this question." If patients had followed a bowel management program, they were asked at what age they started the program and whether the program substantially changed their QOL. Patients were also asked to choose, from a list of possible therapeutic aids, which treatment they were using to keep themselves clean. They were given the possibility to choose more than one alternative from a list that included enemas, laxatives, natural products, or no need for treatment.

Four years later, in 2007, all the adult patients who answered the first questionnaire were asked to complete a second questionnaire and informed consent form, once again sent by mail. The time 2 assessment included the adult version of the Hirschsprung Disease/Anorectal Malformation Quality of Life (HAQL) questionnaire and a measure about patients' perception of the severity of their malformation. The HAQL is a 42-item, disease-specific QOL questionnaire developed by Hanneman and colleagues [15] in the Netherlands. It assesses 11 areas of QOL including dietary modification to reach a laxative diet (eg, eating special food on purpose to get thin stools) or a constipating diet (eg, eating special food on purpose to get thick stools), the presence of diarrhea (eg, thin stools >4 times a day), presence of constipation (eg, thick stools), fecal incontinence (eg, soiling during the day or night), urinary incontinence (eg, loss of urine before reaching the bathroom), social functioning (eg, impaired social activities), emotional functioning (eg, being ashamed of leaving the classroom to go to the bathroom/feeling embarrassed), physical symptoms (eg, having abdominal pain), body image (eg, feeling less attractive), and sexual functioning (eg, less interest in sex). For each item, respondents were asked to indicate the frequency of occurrence in the past week using a 5-point scale ranging from 1 (never) to 5 (always). Reliability for the