



Lower urinary tract symptoms in children with anorectal malformations with rectoperineal fistulas



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ABSTRACT

Aim: The aim was to describe the frequency of lower urinary tract symptoms (LUTS) in children with anorectal malformations with rectoperineal fistulas (ARM-P), as compared with healthy controls based on gender.

Method: LUTS were defined using the 2014 definitions of the International Children's Continence Society. Data were collected at 2 tertiary pediatric surgery centers in 2 countries from all children aged 4–12 years who had undergone an operation for ARM-P.

Results: A total of 24 girls and 33 boys, with a median age of 8 (4–12) years, were eligible and compared with 165 controls. Of the patient group, 4 (17%) girls had 8 urinary tract anomalies (UTA), and 8 (24%) boys had 13 UTA. There were no gender differences in LUTS among the patients. The frequency of urinary tract infections was higher among the patients (5/24 girls and 7/55 boys) than the controls (1/55 and 4/110) ($p = 0.009$). More patients (5/24 girls and 5/33 boys) than controls (1/55 and 2/110) used daily urinary medications ($p = 0.009$ and $p = 0.007$, respectively). Patients with UTA reported urinary infections more frequently (3/4 girls and 4/8 boys) than those without UTA (2/20 girls and 0/25 boys) ($p = 0.018$ and $p = 0.002$, respectively).

Conclusion: Children with ARM-P had more LUTS than controls, and patients with concomitant UTA had more LUTS than patients without UTA. Therefore, children with ARM-P are suggested to have routine follow-up for both UTA and LUTS.

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Lower urinary tract symptoms (LUTS) are reported to significantly reduce the quality of life of children and adults with anorectal malformations (ARM) [1,2] and of otherwise healthy children [3–5]. The International Children's Continence Society (ICCS) defines LUTS as continuous or intermittent urinary incontinence, storing symptoms such as urgency or emptying problems, and lower urinary tract infections [6–8]. In recent studies, these particular terminology and definition of LUTS have been used, thus facilitating comparisons among different cohorts.

Screening for urinary tract anomalies (UTA) is generally recommended for children born with ARM [9–10], and concomitant malformations in the urinary tract are reported to be present in 30%–50% [11–13]. The prevalence of LUTS among the various groups of ARM or specific ARM is, however, not clear. One study on children with ARM, excluding those with perineal fistula (PF), reported a higher prevalence of lower urinary tract dysfunction (LUTD), excluding

infections, in younger children than older children [14]. A recent study on males with PF, including both children and young adults, reported a similar frequency of urinary incontinence in patients as controls [15]. To our knowledge, there are no reports particularly examining LUTS in children of both genders with PF regarding LUTS. Thus, the primary aim of this study was to report the frequency of LUTS in girls and boys with PF and to compare these findings with healthy children. The secondary aim was to assess whether children with PF and concomitant urinary tract anomalies had more LUTS than PF-patients with normal urinary tracts.

1. Materials and method

1.1. Patients

All newborn children with PF who were referred to 2 tertiary pediatric surgery centers in 2 Scandinavian countries from January 1998 and January 2000, respectively, and obtained follow-up until December 2008 were included in the study. The eligible populations in the 2 countries have similar socioeconomic conditions with free health care for all residents. The ARM were subtyped according to the Krickbeck classification, as suggested by the International Conference for the Development of Standards for the treatment of Anorectal Malformations in

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2005 [9]. Excluded from the study were those with anal stenosis. The results were collected when the children were between 4 and 12 years of age. The studied group then consisted of 41 children from one center and 25 children from the other center. These 66 children, 30 girls and 36 boys, fulfilled the inclusion criteria.

1.2. Controls

The controls were collected from different preschools and schools in the region of one of the centers. Questionnaires regarding LUTS were answered by the parents of children who were between 4 and 12 years of age. Children with colorectal or renal diseases and children who had prior urinary tract operations were not asked to respond. The answers were collected in sealed envelopes and placed in closed collecting boxes in each school or sent to the department to preserve anonymity. Only questionnaires that were completely answered were included.

1.3. Questionnaires

A questionnaire with 10 questions (Table 1) based on the ICCS definitions of incontinence, emptying habits, and urinary infections [6] (Fig. 1) was used for registering LUTS among both patients and controls. For the patients, at one of the centers, the questionnaire was used during counseling, while at the other center, the questionnaire served as a template for collecting the information retrospectively from the medical charts.

1.4. Operation methods and follow-up

Both centers operate according to the PSARP procedure as described by Dr. Pena [16]. The anal sphincter complex was defined by electromyostimulation, and the mobilized rectum was placed in the center of the sphincter complex. A diverted colostomy was used if subtyping of PF could not be initially assured. The information about the level of the PF was reported only occasionally in the charts, and therefore all PF were grouped together in the analysis.

There were always two surgeons performing the operations, and at least one was an experienced pediatric colorectal surgeon. Antibiotic prophylaxis was routinely administered during surgery. According to the different routines at the two hospitals, intravenous antibiotic prophylaxis was administered for another 24 or 72 h postoperatively. If there were any signs of infection, a prolonged or new antibiotic treatment was prescribed.

All children were examined 2–4 weeks after the operation, and a dilatation program was started, if necessary.

1.5. Examination and follow-up of the urinary tracts

UTA were defined as any abnormal finding on ultrasound, voiding cysto-urethrography (VCUG), or urodynamic investigation. In both centers, urinary tract ultrasonography was routinely performed. VCUG was routinely performed in one of the centers, and in the other center, patients with hydronephrosis and/or urinary tract infections had VCUG. Voiding uretero reflux (VUR) was diagnosed either during routine examination with VCUG or after urinary tract infections. The principle was to treat VUR (>grade 2) with antibiotic prophylaxis and perform a surgical procedure only in case of repeated urinary tract infections.

1.6. Examination of sacrum and spinal cord

At one of the centers, an ultrasound of the sacrum and spinal cord was performed within 3 months of birth; if there were any signs of pathology or if neurogenic bladder was suspected, a magnetic resonance imaging (MRI) and urodynamic investigation were performed. In the other center, during the study period, an MRI, a urodynamic investigation, or both were performed if there were any clinical signs of skeletal or neurological pathology.

1.7. Study design

This study is a clinical follow-up, including both descriptive and comparative data. Information regarding PF, concomitant malformations, and symptoms was collected during regular patient follow-ups. LUTS registration was performed according to the definitions of ICCS [6]. The terms used in this study are summarized in Fig. 1.

2. Statistical considerations

Fisher's exact test was used for dichotomous outcomes between all groups. If a statistical difference was found, the results were analyzed with post hoc tests for symptom ranking. A p-value of <0.05 was considered significant. All statistical computations were performed by a statistician using the computer program, R version 2.15.2. Multiple comparisons were adjusted using the false discovery rate procedure.

3. Ethical considerations

The regional research ethics committee approved the study (registration number 2010/49) for one center, and the institutional board at the hospital approved it at the other center. Data are presented in such a way that it is impossible to identify any single patient.

Table 1

Questionnaire used for registering lower urinary tract symptoms (LUTS) in patients and controls.

Lower urinary tract symptoms (LUTS)
1. Are you/is your child able to control voiding? a) Yes, always or at least most of the time b) No, there is continuous urinary incontinence
2. Do you/does your child experience any urinary incontinence during the daytime? a) Yes b) No
3. If there is any daytime urinary incontinence, which type? a) Stress incontinence (when jumping, coughing, etc.) b) Postvoiding dribbling c) Urge incontinence (cannot stop urinary flow before reaching a toilet) d) Incontinence during different situations (mixed)
4. Do you/your child have incontinence during the night? a) Yes, every night or several times a week b) Sometimes (1–10 times/month) c) Seldom or never
5. Do you/your child experience difficulties with bladder emptying? a) Yes b) No
6. If there is a problem with bladder emptying, the problem is a) straining b) hesitancy c) feeling of incomplete emptying d) other _____
7. How often do you/does your child void? a) 1–3 times/24 h b) 4–8 times/24 h c) >8 times/24 h
8. Have you/has your child ever had any urinary tract infection that has been treated with antibiotics? a) Yes b) No
9. If you had/your child had any urinary tract infection treated with antibiotics, how many times did it occur? a) 1–2 times b) >2 times during lifetime c) Several times last year
10. Do you/does your child take any daily medication for the urinary tract at this time? a) Yes b) No If yes, which type of medication do you/does your child take? _____

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