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Benefits of primary surgical resection for symptomatic urethral prolapse in children

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Abstract *Objective:* Urethral prolapse (UP) is a complete eversion of the distal urethral mucosa through the external meatus. UP must be distinguished by examination from trauma, prolapsed ureterocele, tumors or sexual abuse. Its management remains controversial. The aim of the study was to promote the benefits of primary surgical management for UP.

Methods: A retrospective multicenter review of children who received surgery for UP between 1991 and 2011 was carried out. Non-complicated UP was primarily treated conservatively. A total of 19 patients were referred for complicated UP and underwent resection of the prolapsed urethral mucosa.

Results: The mean delay in diagnosis was 2.2 days (range 1–6) and the most common symptoms were vaginal spotting and bleeding. No predisposing factor was found, but most patients had a mean weight, height and BMI greater than the 50th percentile. All patients underwent surgery successfully. One patient experienced a complication, i.e., dysuria. There was no case of recurrence after a mean 28 months of follow-up.

Conclusion: Early detection is based on bedside examination. The first-line treatment strategy for uncomplicated UP should be conservative management. Surgical resection is safe and effective for patients with significant symptoms.

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Introduction

Urethral prolapse (UP) is a rare condition in children, with an incidence of 1 in 3000 [1]. It predominantly concerns black, premenarcheal girls [2,3], and the most common presenting symptom is vaginal bleeding [3]. Diagnosis relies on medical examination. Its cause is usually difficult to establish, and some cases are due to sexual abuse. UP was first described by Solingen in 1732 [4] but the optimal method of management remains controversial. Surgical resection has been recommended as an effective primary treatment strategy [5]; however, conservative management with prolapse reduction has recently been proposed [6] as an alternative strategy. The aim of the study was to promote the benefits of primary surgical management for UP.

Methods

Between January 1991 and December 2011, 28 girls aged 3–9 years presenting with urethral mucosal prolapse were referred to centers of pediatric surgery. All cases were reviewed retrospectively in an attempt to define delays in diagnosis, predisposing factors, definitive treatment, and complications from surgical management. This study was approved by the institutional review board. We reviewed the patients' position on the height and weight normogram, and compared them with individuals of the same age.

Most children were seen by a gynecologist before admission to our department. Diagnosis was confirmed in a medical examination by a senior pediatric surgeon. Typically, an inflamed and friable mass that closed the vaginal opening was observed (Fig. 1); the urethral orifice was found in its center. In cases of doubt, a catheter was used to confirm that the meatus lay at the center of the prolapsed mucosa. Bleeding from the normal introitus below the prolapsed urethral tissue was sometimes noted because of vaginal blood accumulation. Two particular forms of UP

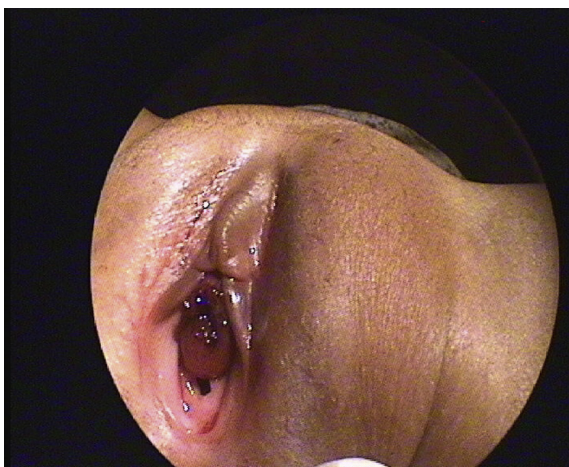


Figure 1 Preoperative view of urethral prolapse. A round edematous mass surrounds the urethral opening. Normal vaginal opening is observed below.

could be observed. Partial UP consists of incomplete presentation with non-circular prolapsed mucosa, while complicated UP is defined by pain and urinary symptoms such as urinary retention, bleeding or necrosis. In three cases, general anesthesia (GA) was necessary to perform adequate examination because the patients were agitated. The main differential diagnoses were ureterocele prolapse, vulvovaginitis, sexual abuse, and rhabdomyosarcoma.

Conservative treatment was intended in cases of incomplete presentations or absence of complications (e.g., necrosis, pain, and urinary symptoms). This consisted of reducing mucosal edema using sitz baths and anti-inflammatories. Conservative measures were sufficient for nine patients with asymptomatic UP, of whom five had partial type UP.

Surgical management was performed under GA in cases with symptoms. Surgery was conducted using a modified surgical technique described by Rudin et al. [5]. A Foley catheter is first introduced in the urethral meatus. Six holding sutures facilitate mobilization of the prolapsed mucosa from the urethral lumen. Four stay and temporary holding sutures (5–0 absorbable suture) are placed in each quadrant of the transection line, which is visible as a fibrous groove surrounding the prolapsed mucosa. In each quadrant, the mucosa is then gradually resected with cautery and the intervening tissue is sutured with two stitches. After 2 days, the Foley catheter was removed and the patient was discharged. Medical examination was performed 1 month after discharge and flowmetry control was performed 2–4 months later.

Results

Surgery was performed for 19 patients with an average age of 5.3 years (range 2–9 years), 17 of whom were black. Conservative measures had previously been unsuccessful for five of them after an average of 20.8 days (range 5–49 days). For the 14 other girls, surgery was chosen because of the diagnosis of complicated UP. Genital hemorrhage was the most common sign (17 of 19) of complicated UP. Bleeding was not profuse and no blood transfusion was necessary. Urinary symptoms were experienced in two cases and pain in six cases. The median delay in diagnosis was 2.2 days (range 1–6 days). Causative factors were highly suspected for four patients (two with chronic constipation and two with chronic cough), and one had severe cerebral palsy (patient 4). Of the 19 patients who received surgery, 18 had their height and weight recorded; all 18 patients except one (patient 4) had a mean weight above than the 50th percentile, and seven were above the 75th percentile. The mean height of 17 was above the 50th percentile and eight of these were above the 75th percentile. Eleven of 18 patients had a BMI above the 50th percentile and five of those were above the 75th percentile (Table 1).

Surgical resection was performed after an average of 7 days (range 1–49 days). The average duration was 2.1 days and 20.8 days, respectively, for patients with primary surgical management and initial conservative measures. A spontaneous favorable course was observed after 2 days of hospital stay (range 2–5 days). The total hospital stay was

Table 1 Characteristics of patients with surgical management.

Patient	Age (years)	PCM	Adverse event	Weight (%)	BMI (%)	Delay in diagnosis (days)	Delay before surgery (days)	Total hospital length of stay (days)
1	4	N	Chronic cough	90	25	1	1	3
2	6	N	None	90	75	1	2	4
3	8	Y	None	50	5	2	8	10
4	9	N	Cerebral palsy with chronic constipation	25	75	1	1	4
5	9	N	Chronic constipation	None	None	1	2	3
6	4	N	None	75	75	5	3	2
7	3	N	Chronic cough	50	25	2	4	3
8	6	N	None	50	50	2	4	3
9	5	Y	Urinary tract infections	85	50	4	7	8
10	3	N	Upper respiratory illness	75	90	1	1	2
11	5	Y	Chronic urinary tract infections	90	90	4	49	10
12	7	Y	None	75	85	4	35	11
13	3	Y	None	100	100	2	5	5
14	5	N	None	50	50	1	2	3
15	7	N	Recent urinary tract infections	90	75	6	2	5
16	5	N	Upper respiratory illness	90	75	1	1	2
17	5	N	Upper respiratory illness	75	50	2	2	3
18	4	N	None	75	85	1	2	2
19	3	N	None	50	75	2	3	4

Abbreviations: PCM = previous conservative measures; Y = Yes; N = No.

4.6 days (range 3–11 days); it was shorter for patients with primary surgical management (3.0 days) than for those treated with initial conservative measures (8.8 days). One complication was observed: urinary pain with dysuria in one case treated with initial conservative management on the second postoperative day. The mean follow-up was 28 months (1–52 months); no recurrence or urethral stenosis was observed.

Discussion

This series illustrates the management of UP in children. Surgical management appears to be secure with few complications, with the main problem being diagnosis confirmation. GA was sometimes required to perform adequate medical examination and surgical treatment was then performed during the same time. Reducing the delay in diagnosis and in surgical treatment seemed to improve postoperative course.

The pathophysiology of UP is not clear. One theory is poor attachment between the smooth muscle layers of the urethra [7] in association with recurrent episodes of increased intra-abdominal pressure [2,7]. Predisposing factors like cough and constipation [8] have been suggested, as having above-average height and weight for the age [9]. High BMI affects intravesical pressure in adults but this has not been proven in the pediatric population [10]. In our series, four patients had a previous history of severe

constipation or cough. Most patients in the present study were heavy and large for their age with BMIs above the 50th percentile, confirming that overweight may contribute to this condition.

The management of UP is still controversial and opinion is divided on the benefits of conservative versus surgical treatment. Conservative therapy includes sitz baths, topical estrogen cream or anti-inflammatory preparation, and appears to be effective for most patients with mild symptoms. However, its effectiveness is limited (33% in Trotman and Brewster's study [11] and 38% in Holbrook and Misra's [6]), and the recurrence rate is high (26% in Rudin's study [5]).

Surgical resection has been presented as a safe treatment modality [5]. Complications include meatal stenosis and dysuria [5,11] and the recurrence rate seems lower than with conservative management (5% in Hillyer et al.'s study [2]). The present study confirms the low complications and recurrence rate with this technique. We recommend performing flowmetry after 2–4 months to detect meatal stenosis.

Holbrook and Misra [6] have proposed an alternative to surgical resection, which consists in reduction of UP under GA. With this technique, complete reduction was achieved in only three of seven patients, and one required further reduction 2 years later. The remaining four patients with partial reduction presented with improvement in symptoms and surgical treatment was not warranted. No data regarding overweight as a predisposing factor for UP

recurrence were available. Because this technique requires GA, it appears to be less cost-effective than surgical management [2].

Given these considerations, non-complicated or incomplete presentations of UP could be treated conservatively with ambulatory management. Unnecessary procedures can be avoided for patients with UP that is incidentally found and in whom reduction under GA would not be beneficial in terms of global effectiveness and risks associated with GA.

In the present study, we found a relationship between delay before surgery and total hospital stay length. Pre-operative hospital stay costs could be avoided. Patients with complicated UP often present with a main complaint of vaginal pain or dysuria; for children with these more significant symptoms, we recommend that resection should be performed as the primary treatment. This would allow prompt management with a single admission and single GA, enabling quick relief of pain and problematic symptoms. Therefore, it would be more cost-effective and could contribute to avoid repeated admissions and parental or child anxiety.

Conclusions

UP is a rare condition in children. In our study, the affected patients were mainly black, young premenarcheal girls who appeared to be large and heavy for their age. The underlying cause is unknown but increase in intra-abdominal pressure is suspected, and the most common presentation is genital bleeding. Diagnosis relies on physical examination and GA is sometimes required. Awareness of UP is important to avoid delayed therapy and repeated examinations. Conservative management is suitable for non-symptomatic patients, while surgical resection offers safe and cost-effective improvement. Therefore, surgery remains the primary treatment strategy for patients with significant symptoms and the reference treatment.

Conflict of interest

None.

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