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Failed Conservative Management in a 6-year-old Girl with Urethral Prolapse: Is Always the Surgery the Solution?

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Abstract

Urethral prolapse (UP) is a rare condition in young female children, with an incidence of 1 in 3000. It commonly presents with vaginal bleeding or dysuria, and is characterized by a circular protrusion of the distal urethra mucosa through the external urethral meatus, forming a round, soft red-purple colored mass. A 6-year-old girl was admitted to our surgical department with symptoms of vaginal bleeding over the last 4 days. Physical examination revealed a doughnut-shaped mass prolapsing from the pudendal labia. Abdominal ultrasound, pelvic magnetic resonance imaging, and blood tests, including hormonal tests, were normal. We initially decided on conservative treatment with topical oestrogen cream and sitz baths. After 12 days, no improvement was observed, and surgical management was undertaken. The surgical approach involved the complete excision of the prolapsed tissue and mucosal to mucosal anastomosis with Vicryl 5/0 sutures. A cystoscopy was previously performed, and the results was normal. Postoperative follow-up over a 6-month period showed no recurrence or urethral stricture. UP in children should always be considered in cases of unexplained vaginal bleeding. Management remains controversial. Surgical excision is recommended for severe cases and in cases that conservative management is uneffective whithin 2-4 weeks.

Keywords: Urethral prolapse, children, urethral and vaginal problems, congenital disease

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Introduction

Urethral prolapse (UP) is a very rare condition in children, with an incidence of 1 in 3000. It most commonly occurs in prepubertal black females and is rare in Asia, with an incidence of 1 in 73,000 in Japan. UP was first described by Solinger in 1732 as a circular protrusion of the distal urethral mucosa, which everts through the meatal opening. Edema and congestion of the prolapsed tissues are seen, with a characteristic, round, soft, red-purple mass prolapsing from the pudendal labia. It commonly presents with vaginal bleeding or dysuria, or as an asymptomatic doughnut-shaped sign at the site of the urethral opening[1,2].

The differential diagnosis includes ectopic ureterocele, condyloma, vaginal polyp, ureteric cysts, and malignancies of the abdomen, vagina, or urethra, such as rhabdomyosarcoma. It is also important to mention that in some cases, it may cause concern regarding sexual abuse among health personnel[3,4].

In this article, we present a 6-year-old white girl from Greece who was admitted to our surgical department with bleeding, and a doughnut-shaped mass of red and purple tissue surrounding the urethra, obscuring the hymenal orifice.

Case Report

The patient was admitted to the hospital with vaginal bleeding that had been ongoing for the past three days. The bleeding was accompanied by symptoms such as itching, frequent urination, and dysuria. There was no history or clinical suspicion of trauma. On examination, the child appeared well, with no signs of distress. Vital signs were within normal limits. A thorough assessment was conducted by the surgical and endocrine departments to rule out other potential causes of vaginal bleeding.

An ultrasound was conducted as part of the clinical investigation, with no major clinical findings from the kidneys, bladder, or adrenal glands. The pediatric-type uterus had dimensions slightly above the normal age range, and the ovaries showed multiple follicles. No fluid was observed in the Douglas pouch.

Hormonal evaluation revealed results consistent with prepubertal status, including normal prolactin and dehydroepiandrosterone levels. Additionally, all malignancy markers were negative. Routine laboratory tests, including complete blood count, biochemical markers, and thyroid function tests, all returned normal results. Urinalysis showed the presence of 30-35 red blood cells per high-power field, 2+ hemoglobin, and traces of leukocyte esterase; without other signs of urinary tract infection.

Following consultation with the multidisciplinary team investigating suspected cases of sexual or other forms of abuse, tests for sexually transmitted diseases were requested and returned negative. No further investigation was deemed necessary.

The patient underwent an examination under general anesthesia, during which urethral mucosal prolapse was identified. The prolapse was reduced, followed by urethral catheterization using an 8 Fr Foley catheter. The hymen was found to be intact. The urethral mucosa prolapsed again, albeit to a lesser degree (**Figure 1**). Further hormonal evaluation and local therapy with oestrogel were scheduled; with a follow-up examination under anesthesia in one week.

The surgical procedure was performed under general anesthesia in a lithotomy position. After inserting an indwelling urinary catheter, four guiding sutures (Vicryl 4/0) were placed. The protruding urethral mucosa was resected with monopolar cautery, and the intervening tissue was sutured with Vicryl 5/0 stitches (**Figure 2**).

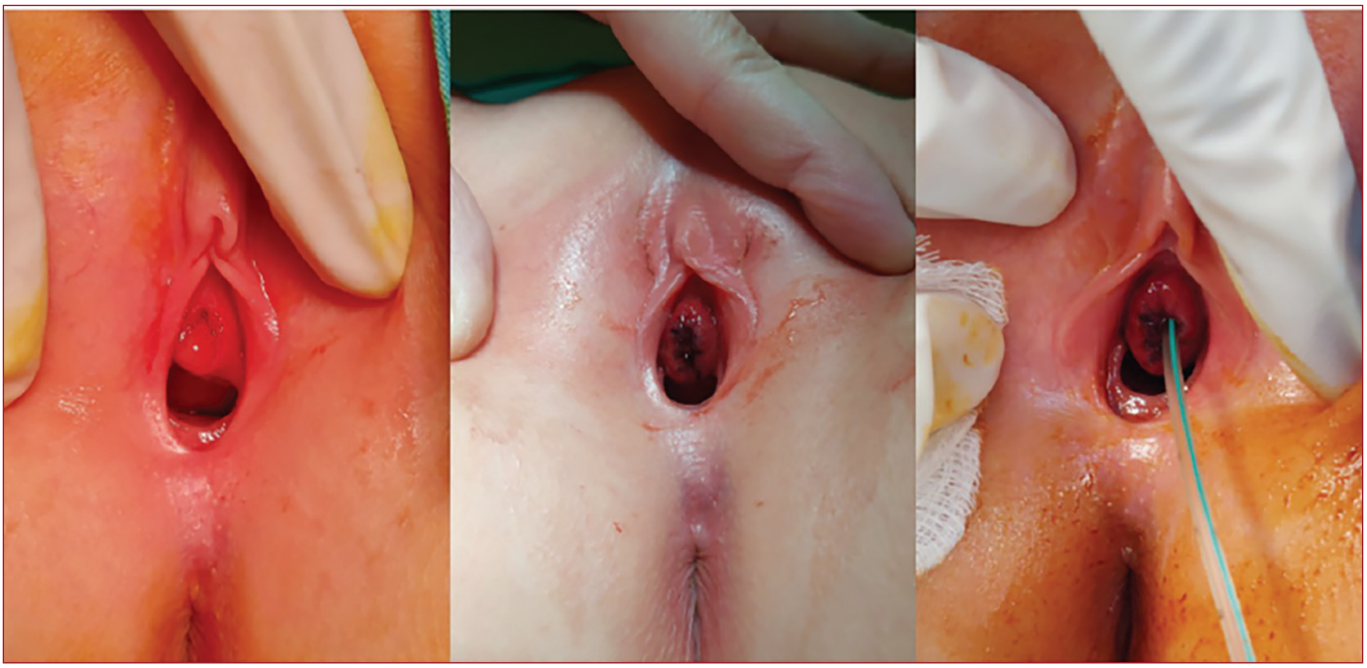

Figure 1. Examination under general anesthesia and catheterisation using an 8 Fr Folley catheter

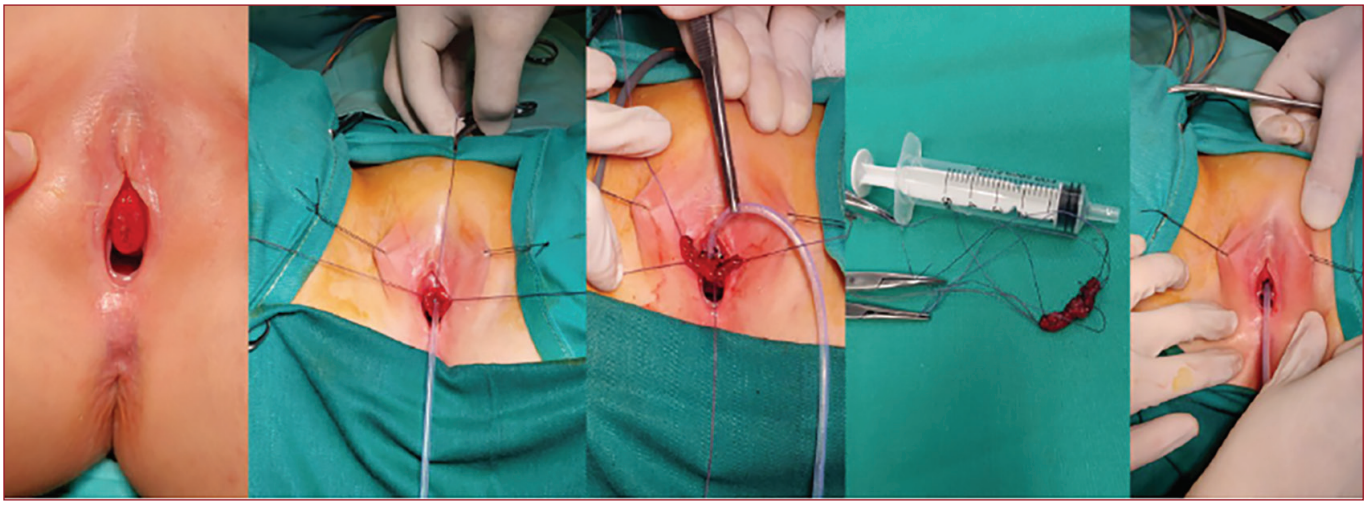

Figure 2. Surgical technique: Guiding suteres, resection of protruding urethral mucosa, suturing the remaining tissue after placement of Folley catherer

A Foley catheter (10 Fr) was placed for 72 hours and was removed postoperatively without complications. The patient voided well and was discharged. Clinical examinations at 1 week, 1 month, and 6 months postoperatively were all normal. A follow-up abdominal and pelvic magnetic resonance imaging performed after 4 months was also normal. A uroflowmetry was not performed, as our patient remained asymptomatic after the surgery. The parents, who are the patient's legal guardian, have been informed for this publication and have given their writen informed consent.

Discussion

The management of UP remains controversial. Some authors advocate for conservative treatment, which aims to reduce mucosal edema, improve local hygiene, and address estrogen deficiency by using a combination of sitz baths, topical estrogen cream, antibacterial wash/soap, and topical antibiotics. Others prefer surgical management. However, no clear treatment algorithm exists. Generally, surgical treatment is favored in children when symptoms are disturbing, such as in our patient, who was very anxious due to her bleeding for 15 days, or in cases when the prolapsed tissue appears ischemic, in which case surgery is beneficial[2].

In 2011, Holbrook and Misra[5] reviewed their 13-year experience with 23 black female children who had UP without symptoms (mild prolapse). Thirteen patients were successfully treated conservatively, while seven underwent prolapse reduction under general anesthesia. Only one patient had a recurrence after two years, which required further reduction in the operating theater. Additionally, two patients underwent partial reduction, but the prolapse resolved completely over an observation period of 3 months following the procedure[5].

A 2008 review by Hillyer et al.[6] reported on 34 female patients with UP, 30 of whom were successfully treated with surgery. Surgical excision of the prolapsed mucosa circumferentially over a Foley catheter was performed. All patients were discharged within 24 hours post-operation. No recurrence was noted during the follow-up period. A 2014 review by Ballouhey et al.[7] concluded that conservative management should be the first-line treatment for uncomplicated patients, while surgical resection is safe and effective for those with significant symptoms. In their study, 19 young girls with UP underwent surgery. The urethral mucosa was resected with cautery, and the intervening tissue was sutured with two sutures. A Foley catheter was inserted and removed two days after surgery. A medical examination performed 1 month after discharge and the flowmetry control 2 to 4 months later showed no recurrence or meatal stenosis[6,7].

Our patient was a Caucasian female with a doughnut-shaped mass prolapsing from the pudendal labia and bleeding for the past four days, initially thought to be vaginal bleeding. Blood tests, including hormonal evaluations, were normal, and an ultrasound did not reveal any abdominal mass. She had no history of constipation, asthma, or other conditions that could increase abdominal pressure. For these reasons, we initially chose conservative management with sitz baths and estrogen cream. After 12 days, there was no improvement, and the bleeding persisted, making the patient anxious. Given the lack of response to conservative treatment, we opted for surgical management. As supported by the literature, surgery is the preferred option in complicated cases and when symptoms persist. Although there is no universally accepted surgical technique, we followed the guidelines of Hillyer et al.[6] and Ballouhey et al.[7]: Resecting the urethral mucosa and leaving a Foley catheter in place for 3 days. The surgery was uneventful, with no postoperative complications.

Conclusion

UP is a rare condition in children with an unknown etiology and controversial management. Physical examination should always be the first step in establishing an accurate diagnosis. Surgical resection of the prolapsing urethral mucosa is a safe and cost-effective treatment, offering low recurrence rates, especially in children presenting with symptoms.

Ethics

Informed Consent: The parents, who are the patient's legal guardian, have been informed for this publication and have given their written informed consent.

Footnotes

Author Contributions: Yerolemidou E: Surgical and Medical Practices, Concept, Design, Analysis or Interpretation, Data Collection or Processing, Literature Search, Writing; Tzortzopoulou A: Surgical and Medical Practices, Concept, Design, Analysis or Interpretation, Data Collection or Processing, Literature Search, Writing; Ververidis M: Surgical and Medical Practices, Concept, Design, Analysis or Interpretation, Data Collection or Processing, Literature Search, Writing; Antonopoulou P: Surgical and Medical Practices, Concept, Design, Analysis or Interpretation, Data Collection or Processing, Literature Search, Writing; Achilleos O: Surgical and Medical Practices, Concept, Design, Analysis or Interpretation, Data Collection or Processing, Literature Search, Writing.

Conflict of Interest: The authors have no conflicts of interest to declare.

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