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Persistent Congenital Paraurethral Cyst, a Rare Cause of Dysuria in a Girl

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Abstract

Congenital paraurethral cyst rarely occurs. Its natural history is spontaneous regression, leading to conservative management adoption. We report an exceptional case of a 13-year-old female with a persistent congenital paraurethral cyst, leading to dysuria. We surgically excised the cyst, and the outcomes were unremarkable 20 months postoperatively.

Keywords

Congenital Paraurethral Cyst, Persistent, Dysuria, Girl, Surgical Excision

1. Introduction

Congenital paraurethral cyst (CPUC) is a rare condition, and its incidence is reported to range from one in 1038 to 7246 live female births [1]. It is due to ductal obstruction of paraurethral glands, Skene's glands [2]. The diagnosis is usually made in neonate, during the physical examination, with investigations needed only for atypical presentation to exclude differentials [3]. The natural history of CPUC is regression, as influence of maternal hormones decreases. Its persistence is exceptional [4] [5]. Management of CPUC during the neonatal period is conservative, when surgery (excision or drainage) is indicated for persistent or lately diagnosed CPUC [3]. We report a girl thirteen years old who presented dysuria due to a persistent CPUC.

2. Case Presentation

A thirteen-year-old girl was received in our department for dysuria and interlabial mass. The mother reported that soon after birth, a vulvar mass was identified in the patient. At the maternity, she was reassured that the mass would soon

regress, and no investigations were requested. However, the mass persisted, and since infancy, it progressively increased its size, but no medical nor traditional management was sook. During the past months, the patient experienced progressive dysuria, which worsened two weeks before the consultation in our department. Then parents consulted at a district medical center, where she was referred. The patient had no medical history; she had her menarche a year ago.

On physical examination, vitals were within normal ranges. An interlabial globular pink mass was identified. It was soft, not painful, and was located on the right side of the urethral meatus, distorted, and deviated on the left side. Its largest diameter was 20 mm (**Figure 1**). Assumed diagnoses were CPUC or prolapsed megaureter. An abdominopelvic ultrasound (US) was ordered and did not identify any urinary tract anomaly. A bacteriological examination of urines did not show any bacteria.

The patient was hospitalized at our department. Initial management included parenteral paracetamol (15 mg/kg/6hours) as analgesic, and placement of a Foley catheter to allow better emptying of the bladder. The same day, she underwent surgical excision of the cyst under general anesthesia, using cautery. The implantation of the cyst was sutured with 4/0 Vicryl using interrupted sutures (**Figure 2**). Postoperatively, intravenous paracetamol was continued, along with ibuprofen (10 mg/kg/8hours). The immediate postoperative course was uneventful, and she was discharged 24 hours later, with per oral paracetamol and ibuprofen for 7 days. Histology of the excised tissue revealed presence of a urothelial tissue with Malpighian metaplasia and unspecific sclero-inflammatory reaction. After 20 months of follow-up, no recurrence nor complication was noted, the physical examination was normal.

3. Discussion

Congenital paraurethral cyst is a rare congenital malformation of the urinary

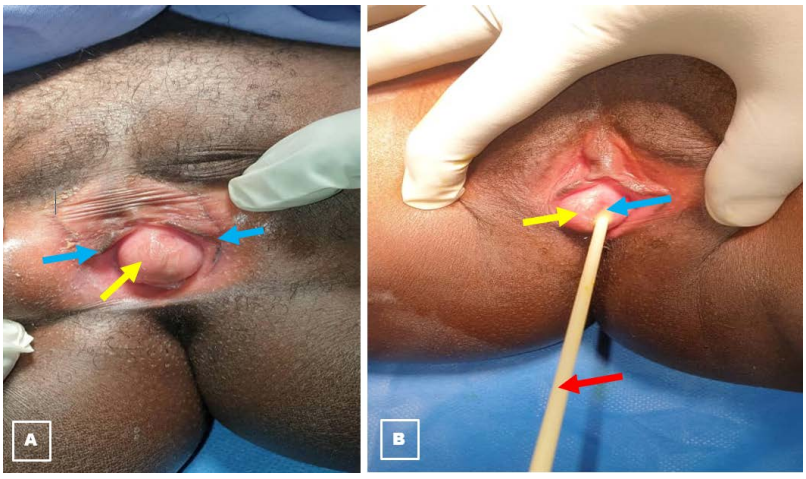

Figure 1. Physical examination. Before insertion of a Foley catheter (A), note the interlabial (yellow arrow) with retracted labia minora (blue arrows). After insertion of the catheter (B), the mass (yellow arrow) originates right to the urethral meatus (blue arrow).

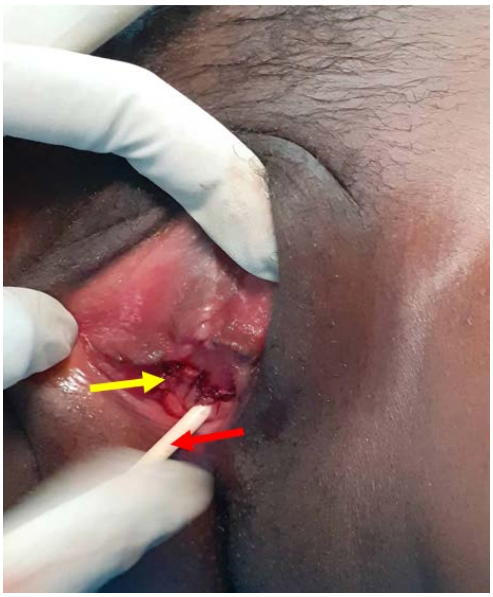

Figure 2. Intraoperative view. After surgical excision of the mass, no residual mass was identified (yellow arrow), as the Foley catheter (red arrow) remained inserted.

tract and an exceptional etiology of dysuria in girls [4]. It is secondary to obstruction of paraurethral glands, homologous to the male prostate, which drains into the female urethra through their ducts. Skene's glands are the largest two glands of 6 to 30 that secrete a mucoid material on sexual stimulation into the distal two-thirds of the female urethra [6]. Etiology of the ductal obstruction is still controversial. Advocated hypothesis include: influence of maternal estrogen, stenotic or inflammatory obstruction, and urothelial dislocation [5].

During the newborn examination, genitalia examination is mandatory. When properly performed, it allows the diagnosis of this congenital lesion which presents as an interlabial golden mass (due to its milky content), lateral to the urethral meatus, and non-painful [1]. The natural history of CPUC is regression as the child grows due to the reduction of maternal hormones impregnation, as suggested by authors supporting the hormonal hypothesis [7]. No additional investigations are necessary unless the patient presents atypically, such as in our case, where persistence up to 13 years old is exceptional. In such cases, differentials should formally be excluded before attempting any surgical option [1]. One of the possible differentials is prolapsed ectopic uretereterocele, which must be ruled out by urinary tract (UT) US [8]. In our case, no anomaly of the UT was detected by the US. Another possible differential is urethral prolapse, frequently encountered in African and African descent girls, as well as in our context [9]. In our patient, it was ruled out after clinical examination, as the mas was lateral to the urethral meatus. Other causes of interlabial mass must be ruled out: botryoidal rhabdomyosarcoma, imperforated hymen with subsequent hydrocolpos, hymenal cyst, Gartner duct cyst, and urethral polyp or diverticulum [1] [10].

Management of CPUC is controversial [7]. Considering its natural history and absence of symptoms, many authors favor conservative management with close

monitoring, especially in newborns. In a series of five patients, Japanese authors reported spontaneous regression from 76 to 304 days [1]. However, other authors praise operative management, arguing that interlabial mass in the neonate can be a stressful situation for parents, and its surgical removal guarantees the absence of recurrence [7]. Several operative options include needle aspiration, incisional drainage, marsupialization or surgical excision of the lesion [3] [11]. Optimal management of CPUP should consider both natural history and the possibility of persistence or complications, such as meatal obstruction, which occurred in our patient. Therefore, neonatal and asymptomatic CPUC should be managed conservatively, and persistent or symptomatic lesions should be operatively managed [1]. In our case, persistence until 13 years old and dysuria due to mass effect on the distal urethra and urethral meatus, we chose surgical excision. Outcome of CPUC is excellent, so that no recurrence was reported in the literature, neither after conservative nor surgical management [12].

4. Conclusion

Congenital paraurethral cyst is a rare condition. Usually asymptomatic and spontaneously resolving, it can be persistent and lead to dysuria, as shown in our patient. In such cases, surgical excision is the cornerstone of management, with excellent outcomes.

Consent for Publication

A written consent for publication was obtained from the patient's parents.

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Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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