

ORIGINAL RESEARCH

Open Access



Conservative management of urethral prolapse in African girls: a report of 15 cases

Ndeye Fatou Seck*, Ndeye Aby Ndoye, Florent Tshibwid A Zeng, Mame Bounama Diop, Lissoune Cissé, Ibrahima Bocar Wellé, Aloïse Sagna and Gabriel Ngom

Abstract

Background: Urethral prolapse is a rare disease, mainly occurring in African prepubertal girls. Its etiology remains unclear; however, some risk factors have been reported. The diagnosis is made clinically. The treatment can be conservative or surgical.

Methods: We report our experience in the service of pediatric surgery at Albert Royer National Children's Hospital Centre. We conducted a retrospective descriptive cross-sectional study, on which considered patients were managed for urethral prolapse in our service from 2014 to 2019.

Results: Our study has included 15 girls whose mean age was 4.08 years (1.17–10). Two risk factors (chronic cough and constipation) were found in 20%. Genital hemorrhage was the main symptom (73.3%), and suspicion of sexual abuse was documented in 13.3%. The clinical finding was classical (donut-shaped vulvar mass) in all patients. All patients underwent conservative management, which was successful in 73.3%. Patients with failed conservative management were treated surgically by resection of the prolapsed mucosa on a Foley catheter. No complication was reported after surgical treatment, and after a 13-month mean follow-up, no recurrence was reported in all patients.

Conclusions: Conservative management is an efficient option for urethral prolapse. It has the advantage of avoiding general anesthesia with its potential complications and restricted availability in sub-Saharan Africa. Surgical management should be reserved for failed conservative management.

Keywords: Urethral prolapse, African girls, Conservative management, Albert Royer, Senegal

1 Background

Urethral prolapse (UP) is a circumferential eversion of the urethral mucosa through the urethral meatus [1]. It is a rare condition, occurring in 1/3000 children, which is predominantly found in African girls aged from 4 to 6 years [2]. Its cause is still unclear. No investigation is necessary as its diagnosis is essentially clinical [3]. Management of UP is still controversial as some authors suggest a conservative management while others favor a surgical approach [2, 4]. Its outcome is generally

favorable, with some exceptions, irrespective of prior conservative or surgical management [5].

Even though UP is documented to be predominant in Africans, African reports on the subject are rare. This has led us to report our experience to describe epidemiological, diagnostic, therapeutic, and evolutionary aspects of UP in African girls who have been managed at Albert Royer National Children's Hospital Centre of Dakar, in Senegal.

2 Methods

We conducted a retrospective descriptive cross-sectional study over 6 years, between 2014 and 2019. Our research was conducted in the pediatric surgical service of Albert Royer National Children's Hospital Centre of Dakar, in

*Correspondence: ndeyefatou2789@gmail.com

Service of Pediatric Surgery, Albert Royer National Children's Hospital Centre, Université Cheikh Anta Diop, 5035 Dakar, Senegal

Senegal. It is one of the three public pediatric surgical services in Dakar and neighboring environs, providing service to a pediatric population of more than a million [6].

We included female children aged less than 15 years. Exclusion criteria were: (1) missing data or records and (2) de novo surgical therapy.

Parameters studied in our review included age at diagnosis, symptoms and their duration, comorbidities, clinical findings, results of investigations, management, and outcomes. Quantitative variables were expressed in mean (age), qualitative ones in frequencies (comorbidities, chief complaint, symptoms, clinical findings, successful conservative management, and complications). Data were registered and analyzed with Epi Info™. Results are presented as text, table, and figure.

Conservative treatment consisted of local application of estrogen cream twice to thrice daily, for 2 weeks, along with a nonsteroidal anti-inflammatory drug (niflumic acid, 15 mg/kg twice daily for 3 days) and sitz baths with lukewarm water for 15–20 min twice a day. Patients were reviewed in the clinic each week. The persistence of the prolapse after 14 days of conservative treatment was considered as a failure of the latter, which was the indication of surgical treatment. This was done by placing a urethral Foley catheter, followed by surgical excision of the prolapsed mucosa using electrocautery. The margin of the urethral mucosa was not sutured. Postoperative pain was managed with paracetamol 15 mg/kg qid for 3 days. Sitz baths were conducted at home, with a lukewarm bath as in conservative management for 7 days.

Ethics approval for processing this retrospective study was obtained from the Committee of Albert Royer National Children's Hospital Centre.

3 Results

3.1 Participants

Seventeen patients with UP were identified over the 6 years, two of whom were excluded as they received surgical therapy ab initio. Our review included 15 female patients, giving a frequency of 2.5 cases per year. The mean age was 4.08 years, ranging from 1.17 to 10 years. The mode was 6 years.

3.2 Main findings

Risk factors were found in three patients (20%): Two had a chronic cough, and one had chronic constipation.

Genital hemorrhage was the main chief complaint in 73.3% of cases. Presenting complaints are reported in Table 1. Clinical examination found, in all patients, a circular pinkish mass above the vaginal introitus. The mass was surrounding the urethral meatus through which a Foley catheter could be introduced. In one patient, bleeding was present during the clinical examination (Fig. 1).

Table 1 Symptoms of urethral prolapse found in patients managed at our service from 2014 to 2019 (N=15)

| Symptom | Number | Percentage |
|---------------------------|--------|------------|
| Genital hemorrhage | 11 | 73.3 |
| Urogenital trauma | 3 | 20 |
| Genital mass | 2 | 13.3 |
| Suspicion of sexual abuse | 2 | 13.3 |
| Hitching | 2 | 13.3 |
| Hematuria | 2 | 13.3 |
| Urinary incontinence | 1 | 6.7 |

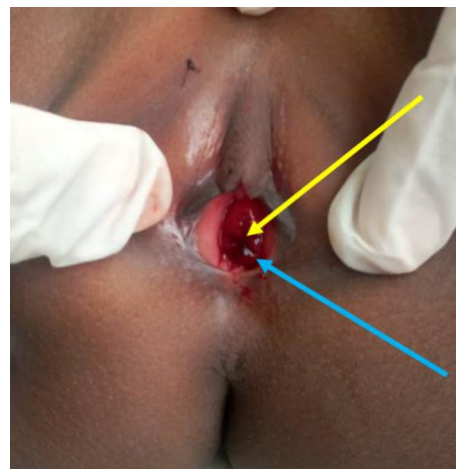


Fig. 1 A patient with urethral prolapse. Notice the urethral meatus (yellow arrow) at the center of the prolapsed mucosa, which the lower part is ischemic and bleeding (blue arrow)

Associated comorbidities were found in two patients (13.3%) including umbilical hernia and rectal prolapse. Full blood count (FBC) has been ordered in all patients, and none had anemia. Microbiologic examination of urine was asked in four patients, and no infection has been detected for all four cases.

Conservative treatment was initiated in all patients and was successful in 11 of them (73.3%). The remaining four patients (26.7%) showed persistence of the prolapse and therefore underwent surgical excision of the prolapsed mucosa. The Foley catheter was removed 3–6 days postoperatively with a mean of 4 days. Patients stayed in clinics for a mean time of 8.75 h (2–24 h). Outcomes were marked by dysuria in two patients after Foley catheter removal, which regressed spontaneously within hours. This corresponds to grade I of the Clavien–Dindo classification of surgical complications.

The mean follow-up was 13 months (6–17 months), and no recurrence has been noted in patients

conservatively managed, neither in those who underwent surgical excision.

4 Discussion

With 2.5 cases per year, our in-hospital frequency of UP is similar to another African study [7]. However, an African multicentric study found a much higher frequency with 7 cases per year [8]. In western countries, UP is quite rare with in-hospital frequency ranging from 2.1 to 3.9 cases per year [9], and it is scarce and more frequent in African and African-descent populations [1]. Our study population had a mean age of 4 years, which is not different from reports of several authors who found a mean age ranging from 4 to 6 years [5, 7, 10, 11].

The etiology of UP is unknown; however, some risk factors have been described, such as chronic constipation or cough, bearing heavy things, perineal trauma, sexual abuse, urogenital infections, malnutrition, and urethral mucosa excess [9, 12]. In our study, three patients (20%) presented risk factors. Some authors reported risk factors in 4.8–69% [5, 10, 11]. Other authors did not find reported risk factors [13]. We think these differences are due to the lack of systematic data collection as all studies were retrospective. Cohort studies will be helpful to highlight risk factors. Another evoked risk factor is the poor level of estrogen [14, 15]. Estrogen receptors (ERs) were identified in the smooth muscle and connective tissue of the female urethra, which suggests that these tissues recognize estrogen and have a response to its action [16]. Some authors suggest that estrogen seals urethral mucosa to the underlying submucosa so that the lack of estrogen leads to poor connection between mucosal and submucosal layers of the urethra, which in turn results in UP [14]. A study reported lower serum estrogen levels in premenopausal women experiencing pelvic organ prolapse (POP) compared to those without POP [16]. This is the justification of hormonal therapy in UP.

Genital hemorrhage was the main complaint, found in 11 patients (73.3%). Our findings are confirmed by other authors, in either western or African countries, with genital bleeding ranging from 81.2 to 100% [5, 10, 13, 17]. In the African settings, parents often link genital hemorrhage to sexual abuse or urogenital trauma, which are sometimes suggested as the etiology, as found in 13.3% of our patients. Other African studies reported similar proportions [3, 7]. Care should be taken to make the right diagnosis despite these wrong orientations from parents.

Clinical findings are classically made of a doughnut-like pinkish mass, surrounding the urethral orifice, which can be easily catheterized, and this was found in all of our patients as many other authors did [7, 8, 13, 18, 19]. With time, prolapsed mucosa gets ischemic, ulcerated,

or necrotic [11, 12]. This evolution can explain mucosal bleeding found in two girls of our series.

Management of UP can be conservative or surgical. Several authors recommend conservative management as the first level of treatment, indicating surgery only in recurrent UP or failed conservative management of symptomatic UP [10, 12, 19]. Conservative management uses estrogen-based cream along with anti-inflammatory drugs and Sitz bath [11]. In our study, all patients initially underwent conservative management, which failed in 26%. The frequency of failed conservative management has been reported from zero to 80.6% [5, 11]. These differences can be due to the use of different topical creams. While estrogen cream is used, along with other measures, failure of conservative treatment is lower (0–38%) [11] compared to a study where many patients benefited from steroid cream alone (80.6%) [5]. Further studies should be done to better assess the impact of the nature of the topical cream on failed conservative management of UP in girls.

Surgical management of UP is mainly done by resection of the prolapsed mucosa with or without sutures [10]. Reduction under general anesthesia [19] or ligation on Foley catheter [8, 20, 21] has been suggested by some authors. In our patients, we used surgical resection without sutures, making hemostasis with electrocautery. Several authors used the same method with satisfying results [8, 13].

In our study, a Foley catheter was removed within 3–6 days. Among reported studies, some authors removed the catheter immediately after the surgery [8] or from 2 to 3 days [7, 13]. We preferred to keep the Foley catheter for at least 3 days to prevent urethral stenosis, as documented by other authors [7].

After surgical management, some complications can occur. In our study, temporary dysuria was reported in two patients. Some authors reported postoperative complications assessed by Clavien–Dindo classification as grade I: persisting dysuria and urethritis and grade IIIb: meatal stenosis requiring dilatation [11]. Failed surgical management has been reported in 17.8% [5]. This shows that surgical treatment has its potential complications and is not always successful. The place of conservative management as first line in the management of pediatric UP cannot further be emphasized.

4.1 Limitations

Our study has some limitations. The first is the lack of duration of conservative management, and the number of medical visits before UP gets cured conservatively. These limitations are since this information was not on medical files.

5 Conclusions

Urethral prolapse is a rare condition affecting mainly prepubertal African girls. Its main symptom is genital hemorrhage which can be wrongly attributed to sexual abuse, which can be ruled out by good interrogatory and physical examination. Its conservative management is possible, with long-term good results. It avoids general anesthesia with its potential complications and restricted availability in sub-Saharan Africa. Surgical management should be reserved for patients with failure of conservative management.

Abbreviations

ERs: Estrogen receptors; FBC: Full blood count; POP: Pelvic organ prolapse; UP: Urethral prolapse.

Acknowledgements

The authors thank the surgical team of Albert Royer National Children's Hospital Centre for managing all patients. The authors thank Dr. Alagie Bladeh (Medical Doctor attached to the Division of Pediatric Surgery at Edward Francis Small Teaching Hospital, Banjul, Gambia) for proofreading the manuscript. The author FTAZ is grateful to Else-Kröner-Fresenius-Stiftung, Holger-Poehlmann-Stiftung and the NGO Förderverein Uni Kinshasa e.V., fUNIKIN through the excellence scholarship program "Bourse d'Excellence Bringmann aux Universités Congolaises, BEBUC", which funds his specialization in pediatric surgery.

Author contributions

NFS contributed to conception, manuscript redaction, and data collection. NAN contributed to Conception, study design, and critical revision of the manuscript. FTAZ contributed to interpretation of results and manuscript redaction. MBD contributed to data acquisition, analysis, and interpretation. LC contributed to conception and data acquisition. IBW contributed to data analysis and interpretation. AS contributed to conception and critical revision of the manuscript. GN contributed to study design and critical revision of the manuscript. All authors have approved the submitted version of the manuscript and gave their agreement to personally be accountable for the author's own contributions and ensured that questions related to the accuracy or integrity of the work were appropriately investigated, resolved, and the resolution documented in the literature. All authors read and approved the final manuscript.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Availability of data and materials

Data are available from the corresponding author on a reasonable request.

Declarations

Ethics approval and consent to participate

Ethics approval for processing this retrospective study has been obtained from the Committee of Albert Royer National Children's Hospital Centre. Consent to participate was waived by the institutional ethics committee seeing the retrospective aspect of the study.

Consent for publication

A consent form has been obtained from the parent of the patient whom the image is used as Fig. 1 in the article.

Competing interests

The authors declare no to have no known conflict of interests that could influence that work.

Received: 11 October 2021 Accepted: 13 May 2022

Published online: 15 June 2022

References

- Ballouhey Q, Galinier P, Gryn A, Grimaudo A, Pienkowski C, Fourcade L (2014) Benefits of primary surgical resection for symptomatic urethral prolapse in children. *J Pediatr Urol* 10(1):94–97
- Liu C, Lin Y, Chen X, Li S, Zhu H (2018) Urethral prolapse in prepubertal females: report of seven cases. *J Obstet Gynaecol Res* 44(1):175–178
- Agarwal S, Lall A, Bianchi A, Dickson A (2008) Uro-genital bleeding in premenarcheal girls: dilemmas of child abuse. *Pediatr Surg Int* 24(6):745–746
- Abouzeid H, Shergill IS, Al-Samarrai M (2007) Successful medical treatment of advanced urethral prolapse. *J Obstet Gynaecol* 27(6):634–635
- Ninomiya T, Koga H (2017) Clinical characteristics of urethral prolapse in Japanese children. *Pediatr Int* 59(5):578–582
- Bureau de L'État civil et des Projections démographiques (2021) Population du Sénégal: Année 2020. Direction des Statistiques démographiques et sociales Division du Recensement et des Statistiques démographiques, p 7. Available at <https://www.ansd.sn/index>. Accessed 08 Nov 2020
- Ndour O, Malle K, Fall ALF, Ndoeye NA, Nibagora J, Ngom G et al (2017) Le prolapsus de la muqueuse urétrale chez la fille: À propos de 12 cas et revue de la littérature. *Afr J Urol* 23(4):359–363
- Da Silva-Anoma S, Dibi Bertin K, Ossenu O, Atafi GD, Yao D, Roux C (2001) Le prolapsus muqueux de l'urètre de la fillette en Côte D'Ivoire. *Ann Urol* 35:60–63
- Rudin JE, Geldt VG, Aleceev EB (1997) Prolapse of urethral mucosa in white female children: experience with 58 cases. *J Pediatr Surg* 32(3):423–425
- Hillyer S, Mooppan U, Kim H, Gulmi F (2009) Diagnosis and treatment of urethral prolapse in children: experience with 34 cases. *Urology* 73(5):1008–1011
- Holbrook C, Misra D (2012) Surgical management of urethral prolapse in girls: 13 years' experience. *BJU Int* 110(1):132–134
- Fiogbe MA, Hounnou GM, Koura A, Agossou-Voyeme KA (2011) Urethral mucosal prolapse in young girls: a report of nine cases in Cotonou. *Afr J Paediatr Surg* 8:12–14
- Sandaa GO, Soumanaa A, Oumaroub H (2012) Le prolapsus muqueux de l'urètre chez la fillette: a propos de 22 cas colligés en dix ans et une revue de la littérature. *Afr J Urol* 18:93–96
- Jessop ML, Zaslau S, Al-Omar O (2016) A case of strangulated urethral prolapse in a premenopausal adult female. *Case Rep Urol* 2016:1802623
- Baker KC, Oakman C (2006) Thrombosed urethral prolapse in a premenopausal woman. *J Pelvic Med Surg* 12:171–173
- Lang JH, Zhu L, Sun ZJ, Chen J (2003) Estrogen levels and estrogen receptors in patients with stress urinary incontinence and pelvic organ prolapse. *Int J Gynaecol Obstet* 80:35–39
- Diouf C, Diallo I, Mbaye F, Ndoeye NA, Faye AL, Ndour O, Ngom G (2016) The urethral prolapse, a rare cause of genital bleeding in girls: report on three cases. *Med Sur Urol* 5(2):162
- Hilali M, Nour M, Talha H, Oubejja H, Erraji M, Zerhouni H et al (2014) Prolapsus urétral chez la fille à propos de cinq cas. *Arch Pédiatr* 21(5):569
- Ballouhey Q, Abbo O, Sanson S, Cochet T, Galinier P, Pienkowski C (2013) Hémorragie génitale de la petite fille: penser au prolapsus urétral. *Gynécologie Obstét Fertil* 41(6):404–406
- Anveden-Hertzberg L, Gauderer MW, Elder JS (1995) Urethral prolapse: an often misdiagnosed cause of urogenital bleeding in girls. *Pediatr Emerg Care* 11(4):212–214
- Okorie CO (2013) Urethral prolapse: contemporary report on a modified ligation over a urethral catheter treatment approach. *Nephrourol Mon* 5(4):866–869

Publisher's note

Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.