

Severe bleeding from misdiagnosed urethral prolapse in two young girls

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ABSTRACT

Introduction: Urethral prolapse (UP) is a rare and misleading pathology that occurs most commonly in young black females. It is the protrusion of the distal urethra through the external meatus with bleeding as the main symptom, followed by voiding disturbances. **Case Series:** The two cases presented here were referred to us with diagnosis of vulvar tumor and sexual abuse. The clinical diagnosis of strangulated UP with anemia from chronic hemorrhage was made for the first case and the diagnosis of UP with acute uncontrollable bleeding for the second. Surgical management with excision of the protruded urethra was successful and the postoperative follow-up was uneventful. The pathology report confirmed the diagnosis and no recurrence was noticed within a year. **Conclusion:** Urethral prolapse is often misdiagnosed in our environment. Severe cases need immediate surgical treatment that can be performed successfully with no risk of recurrence.

Keywords: Urethral prolapse, Misdiagnosis, Treatment, Resource-limited setting

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INTRODUCTION

Urethral prolapse (UP) is a rarely diagnosed pathology that occurs most commonly in young black females under the age of 10 (average age at presentation of four years) [1, 2]. It was first described in 1732 by Solingen [1]. Its misdiagnosis rates are high due to the rarity of this condition, among which rape and genital trauma are common, leading to false accusations and unjustified social and legal problems [2–5].

Clinically, it presents as a circular protrusion of the distal urethra through the external meatus. In some cases, it may be strangulated. The main presenting symptom is bleeding from the urethral stump considered by the parents as “vaginal” bleeding. Some voiding disturbances, such as dysuria, frequency, and burning sensations may be associated [1, 2]. On physical examination, a round doughnut-shaped mucosa is observed protruding from the urethral opening. The hymen is mostly in place as the children affected are usually not yet sexually active.

The cause of UP is still unknown, however, some theories have been postulated, the most popular one embracing a weakened attachment between the inner and the outer layers of the urethral walls, whose separation coincides with episodic increment in intra-abdominal pressure [1, 6].

The treatment modalities include sitz baths, topical antibiotics, topical estrogen cream, manual reduction

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under general anesthesia, and surgery [7, 8]. Surgery is indicated for cases of medical treatment failure. However, in severe cases like those presented here, immediate surgery management is mandatory to control severe bleeding or to avoid tissue necrosis. For many authors, surgery is the treatment of choice even in no emergency cases as it has a high cure rate [2, 8, 9].

The purpose of the presentation of these cases is to raise awareness among medical doctors of this rare and possibly life-threatening pathology of young girls of resource-limited setting, and to share our management procedure in a low resource setting.

CASE SERIES

Case 1

A 6-year-old female child was brought into consultation for vaginal bleeding with painful urination. The history of the disease revealed slight bleeding followed by dysuria and suprapubic tenderness evolving for about a week. No history of chronic cough nor constipation was found. The child complained to her grandmother with whom she lives in the village who considered her pain as being of traumatic origin and gave her paracetamol associated with a vulva wash with lukewarm soapy and salted water. Faced with persistent symptoms with worsening bleeding, she took her to the health center from where she was referred with the suspicion vulva tumor. On physical examination, we noted a temperature of 37.5 °C, a weight of 24 kg, a height of 118 cm, and pale conjunctivae. The external genitalia presented with a circular 15 mm in diameter and cyanotic mass surrounding the external meatus, with contact bleeding, and almost obstructing the vaginal orifice (Figure 1). Insertion of Foley catheter in the bladder through the center of the mass reinforced our diagnosis of UP which was complicated by strangulation and anemia. Laboratory tests revealed a hemoglobin level of 10g/dL, and normal blood coagulation tests. Surgical treatment conducted with the procedure described in case 2 and partially illustrated on Figure 2 was successful and postoperative follow-up was uneventful.

Case 2

A 4-year-old female child was referred in emergency by a general practitioner from a nearby health facility for “vaginal bleeding.” The history of the disease revealed a three days vulva pain that the child reported to her elder sister who did not take it seriously. The bleeding started abruptly in school during break and the child complained to her mistress who took her to the health center where the diagnosis of sexual abuse was suspected, hence her referral. On physical examination, her clothes were soaked with blood. The temperature was 36.7 °C, a height of 95 cm, a weight of 16 kg, and a pulse rate 124/min. There was a hemorrhagic mass in



Figure 1: Cyanotic thrombosed urethral prolapse.

the anterior region of the vulva. The examination after a vaginal tampon soaked in adrenaline solution shows the urethral origin of the mass, certainly a prolapse, as the center admitted easily the insertion of Foley catheter. The exploration of the posterior fornix of the vulva shows an intact hymen. Laboratory tests showed a hemoglobin level of 12.8 g/L and normal clotting profile. Given the hemorrhagic nature of the prolapse, we decided for a surgical treatment under general anesthesia. A complete and circumferential excision of the prolapsed urethra was done, followed by muco-mucus interrupted suture using Vicryl 4/0 suture (Figure 2). The picture of the vulva at the end of the procedure shows the catheter within a repaired urethra (Figure 3). The catheter was removed on postoperative day 3 and the child discharged. She was reviewed at 1 month and at 12 months following surgery and no complication was noticed. Histologic exam of the stump confirmed the diagnosis.



Figure 2: Resection of the prolapsed urethra under an indwelling Foley catheter and traction to easily identify the mucosa.



Figure 3: Postoperative aspect.

DISCUSSION

Our two cases were young black girls from poor families, with ages 4 and 6 years. Many authors had made the same observations [1, 7, 8]. No explanations have been made to why this pathology is common among children of the black race. However, concerning age, prepubertal girls lack estrogen which plays a role in strengthening the pelvis. This lack of estrogen may therefore constitute an exposure factor to UP. This may also explain some degree of success observed when estrogen cream is used to manage UP.

The main symptom was bleeding, present in the two cases. Blood loss from UP can be life-threatening, needing resuscitation by transfusion [2]. For case 1, the bleeding was chronic resulting in anemia. The prolapse was large with contact bleeding and cyanosis, and signs of strangulation. In Holbrook series, the majority presented with a vulvar mass (8/21), followed by bleeding (6/21) [7]. As the bleeding is more likely to be an alarming sign than a mass, it is possible that in our resource-limited and rural setting, some cases of UP are still undiagnosed. Active search of UP through systematic exam of young girls under 10 may contribute to have an UP prevalence closer to the real situation. Mothers should also be encouraged to examine their young girl child and seek medical advice for any vulvar pathology.

Trauma and sexual abuse are the most frequent causes of bleeding in young girls in our environment and this may explain the wrong referral diagnosis of case 2. The misdiagnosis of UP was found to generate social and legal problems [10, 11]. If the health care provider who sees the child first is not capable of making the diagnosis of UP, the consequences can be drastic not only for the child but also for the society.

According to many authors, diagnosis of UP is clinical and requires capacity of detection from the health care professional [2, 6, 10]. These capacities can only be acquired through picture visualization or successful diagnosis and management of a previous case. Also, given the low suspicion index of the disease because of its rare nature among clinicians, most of the cases are likely to be missed or misdiagnosed in clinical setting. In Anveden series, only 21% of referred cases had a correct diagnosis [3]. However, with a little knowledge or experience, on UP, diagnosis can be made clinically as we did for our two cases before pathological confirmation.

Our two cases of UP were treated surgically. From literature review, initial treatment of mild forms includes sitz bath, topical estrogen cream, and antibiotics. Many authors have done this with success [1, 7, 10]. However, in severe cases with uncontrollable bleeding and/or tissue thrombosis, there is no room for medical treatment and surgery is mandatory [1, 6, 12]. Our case 1 had tissue thrombosis and anemia from chronic bleeding and the second had profuse uncontrollable bleeding. First line surgical treatment was therefore indicated for both during which a removal of the prolapsed urethra (with

an indwelling Foley catheter), after a circumferential incision followed by muco-mucosal suture was done. The postoperative follow-up was uneventful and no recurrence was found at 12 months. For many authors, surgery is the treatment of choice for UP [2, 8, 9].

CONCLUSION

Urethral prolapse is a rare pathology of young black girls, often misdiagnosed in our environment. Bleeding remains the most common and alarming symptom but complications of strangulation and thrombosis are likely to occur. Misdiagnosis in our setting is generally associated with unnecessary social and legal tension. Increasing awareness of physicians in low resource countries on proper identification and management of UP is capital. In severe cases, surgical treatment with complete and circumferential excision of the prolapsed urethra followed by muco-mucosal suturing is safe, simple, and without any recurrence risk.

REFERENCES

- Gill BC, Kim ED. Urethral Prolapse. 2018. [Available at: <http://emedicine.medscape.com/article/443165-overview>]
- Ndour O, Aalle K, Faye Fall AL, et al. Urethral mucosa prolapse in young girls: A series of 12 patients and review of literature. [Article in French]. *Afr J Urol* 2017;23:359–63.
- Anveden-Hertzberg L, Gauderer MW, Elder JS. Urethral prolapse: An often misdiagnosed cause of urogenital bleeding in girls. *Pediatr Emerg Care* 1995;11(4):212–4.
- Shavit I, Solt I. Urethral prolapse misdiagnosed as vaginal bleeding in a premenarchal girl. *Eur J Pediatr* 2008;167(5):597–8.
- Shurtleff BT, Barone JG. Urethral prolapse: Four quadrant excisional technique. *J Pediatr Adolesc Gynecol* 2002;15(4):209–11.
- Holbrook C, Misra D. Surgical management of urethral prolapse in girls: 13 years' experience. *BJU Int* 2012;110(1):132–4.
- Falandry L. Prolapse of the urethra in black girls. Personal experience in 11 cases. [Article in French]. *Med Trop (Mars)* 1994;54(2):152–6.
- Lui C, Lin Y, Chen X, Li S, Zhu H. Urethral prolapse in prepubertal females: Report of seven cases. *J Obstet Gynaecol Res* 2018;44(1):175–8.
- Yddoussalah O, Oudrhiri M, Sumba H, et al. Urethral prolapse in young girls: A report of eight cases. *J Urol Res* 2018;5(2):1101.
- Diouf C, Diallo I, Mbaye F, et al. The urethral prolapse, a rare cause of genital bleeding in girls: Report of three cases. *Med Sur Urol* 2016;5(2):162.
- Abhulimhen-Iyoha BI, Oguejiofor CA. Urethral prolapse: An important differential diagnosis of bleeding from the vulva in pre-pubertal African girls. *Port Harcourt Med J* 2017;11(2):96–8.
- Sanda GO, Soumana A, Oumarou H. The urethral mucosal prolapse in young girls: A series of 22 cases and a literature review. *Afr J Urol* 2012;18(2):93–6.

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Author Contributions

Kenfack Bruno – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Atem Bethel Ajong – Design of the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Noubom Michel – Design of the work, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Data Availability

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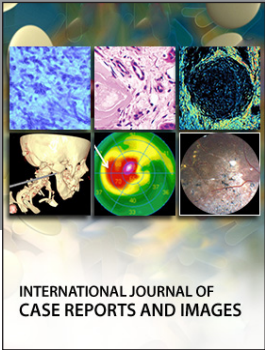
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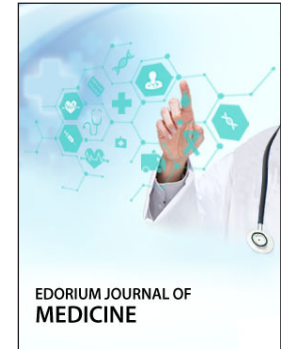
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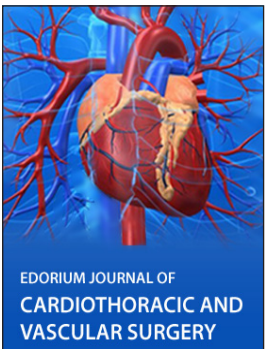
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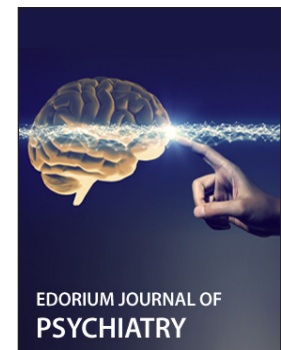
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