Case report

Surgical Resection and Repair of Prolapse of the urethral mucosa in a Pubertal Girl: A Case Report.

Abstracts

~~Aim/~~**Background**: Urethral mucosa prolapse, though a rare condition seen in pubertal/adolescence girls, diagnosis should be considered in cases of unexplained vaginal bleeding in this age group. ~~Hence, need for clinicians to make appropriate diagnosis and offer a definitive management including surgical resection which is cost-effective. Urethral mucosa prolapse was first described in 1732 by Solinger.~~This condition is most commonly observed in prepubertal black girls and postmenopausal white women however, it can be seen in pubertal and adolescence girls. The majority of cases (80%) are in the paediatric age group (between the ages of 6 months and 8 years) and the incidence in this age group is 1:3000. This case report presents a 12-year-old pubertal girl from Makurdi, Benue State, Nigeria, who presented with genital swelling and vaginal bleeding associated with pain. Due to the severity of her symptoms, surgical resection and repair (meatoplasty) were performed.

Case Presentation: The patient presented with genital swelling, vaginal bleeding, and pain. A genital examination revealed moderate vaginal bleeding and a dark red, oedematous mass with a central dimple prolapsing through the external urethral meatus. The diagnosis was confirmed by passing a pediatric nasogastric tube through the dimple, draining clear urine. She underwent a urethral meatoplasty and had an uneventful recovery.

Conclusions: Although urethral mucosa prolapse is rare in pubertal/adolescent girls, it is important for clinicians to consider this diagnosis in cases of unexplained vaginal bleeding. Surgical resection provides a definitive and cost-effective treatment, with excellent outcomes as demonstrated in this case.

Comment [AH1]: Please, add 3-6 keywords.

Introduction~~/Aim~~

The Urethral mucosal Prolapse, was first reported in 1732 by Solinger[1-3]. It is described as a rare condition characterized by the circular protrusion of the distal urethral mucosa through the external urethral meatus. It commonly forms a hemorrhagic,doughnut-like shaped vulvar mass separated from the vagina, which easily bleeds. This condition is most commonly observed in prepubertal black girls and postmenopausal white women however, it can be seen in pubertal and adolescence girls[1-3]. The majority of cases (80%) are in the paediatric age group and the incidence in this age group is about 1:3000[3]. Although urethral mucosa prolapse is rare in pubertal/adolescence girls, diagnosis should be considered in cases of unexplained vaginal bleeding in this age group. Hence, need for clinicians to make appropriate diagnosis and offer a definitive management including surgical resection which is cost-effective.

Presentation of Case

Miss M.A was a 12-year-old girl. She presented at the Emergency Paediatric Unit of the Benue State University Teaching Hospital, Makurdi on 14/07/2021 with history of genital swelling around the urethral opening of five days duration. She first noticed it when bathing and reported to her mother a day before

presentation at the facility. The swelling was small but progressively increased in size. The mother asked her to applied petroleum jelly to help reduced the discomfort. A day prior to presentation, the bleeding increased in quantity and the episodes of bleeding became more frequent. She was feeling burning sensation in the genital region and pain. She also had 2 episodes of vaginal bleeding on the morning of presentation. The bleeding was slightly more than the previous episodes. She also had history of difficult and painful micturition. There was no urinary frequency. History was not suggestive of any sexual abuse, genital trauma or fever. She had no history of previous or recent female genital mutilation. The patient was referred to the Gynaecological Emergency Unit of the same facility with provisional diagnosis of suspected sexual assault to keep in view urinary tract infection.

Comment [AH2]: apply

Comment [AH3]: reduce

She was the second child out of 4 children of her mother in a polygamous family. The elder sister did not have a similar complaint. There was no family history of bleeding disorder. She was a Primary 5pupil. She was not on routine medication for any condition. There was no history suggestive of drug allergy.

She was of normal body built but anxious. Other general examination findings were normal. She weighed 34kg and her height was 1.30m giving her a body mass index (BMI) of 20.12kg/m^2. The systemic examination findings were essentially normal. Examination of the external genitalia revealed moderate bleeding from the mass on contact. Gentle separation of the labia (figure 1) revealed a 2cm by 2cm circumferential reddish mass protruding around the external urethral meatus (“doughnut shaped” mass) and it bleeds on contact. The urethral opening was visible at the center of the mass. An infant feeding tube was passed through the center of the mass and it drained clear urine. The hymen was intact. There was no abnormal vaginal discharge or any other lesion noted.

An assessment of urethral mucosal prolapse in a pubertal girl was made. Her parents were counseled on the need for surgery and informed written consent was obtained. Results of investigations were normal. The surgery was planned for the next morning. This was done under general anaesthesia using the modified Kelly-Burnham procedure. The bladder was emptied of clear urine with a size 10 Foley’s urethral catheter and retained.

The assistant surgeon retracted the labia majora with the fingers to expose the external urethral meatus. With this, the prolapsed mucosa was mobilized from the urethral lumen. Four stay and temporary holding sutures using vicryl number 3/0 suture were placed in each quadrant of the transaction line at 12, 3, 6 and 9 o’clock positions of the prolapsed urethral mucosa. In each quadrant, the mucosa was then carefully resected with scalpel blade. The excision of the prolapsed mucosa was started with the quadrant between stay stitches at 12 o’clock and 3 o’clock positions. It was held with a Mosquito’s artery forceps; a gentle outward traction was applied and the urethral catheter was retracted to the opposite side by the assistant. A vertical incision was made just before the stay stitch at 12 o’clock position through the full thickness of the prolapsed mucosa to the level of the normal urethral mucosa. The similar incision was made just before the stay stitch at 3 o’clock position. The flap thus formed between the stay stitches at 12

o'clock and 3 o'clock positions was transected at the level between the normal urethral mucosa and prolapsed mucosa transversely. Care was taken to avoid cutting through the urethral catheter that was insitu. The normal urethral mucosa in this quadrant was sutured to the periurethral mucosa with two interrupted stitches using vicryl No. 3/0. The mirror procedure was performed on the quadrant between 3 o'clock and 6 o'clock positions. The prolapsed mucosa together with the stay stitch at 3 o'clock position was excised. Similarly, the normal urethral mucosa in this quadrant was sutured to the periurethral mucosa with two interrupted stitches. The mirror procedure was repeated on the remaining quadrants. The surgical area was observed for any evidence of bleeding and the urethral catheter was left in situ for continuous urinary drainage for 72 hours.

She had a smooth recovery from anaesthesia. She was allowed oral intake once she was fully conscious. The operative findings and prognosis were explained to her parents. She was reviewed daily on the Ward and her condition remained stable. On the third postoperative day, the urethral catheter was removed. She was observed for a day without catheter and was able to void freely. The mother was counselled regarding prognosis and the need for regular follow up. She was subsequently discharged home on the fifth day postoperatively. She had regular follow up. The histology report showed features consistent with urethra mucosa. There was no recurrence of the urethral mucosa prolapse or complication during the 3 years of follow up.

Figure 1: Showed a circumferential dark reddish mass protruding around the external urethral meatus ("doughnut shaped" mass)

Figure 2: During surgical resection and repair

Comment [AH4]: This figure is not appearing within the text. Please, refer to it.

Discussion

Urethral mucosal prolapse is a benign condition characterized by an eversion of the urethral mucosa, in form of a pink or purple, hemorrhagic, cyanotic or necrotic mass of variable volume which bleeds continuously or on contact.[1-3] This bleeding is often the main mode of presentation.[1-3] Prolapse of the urethral mucosa occurs when the rest of the urethra remains in its normal position but the mucosa becomes loosened from its submucous attachment and is gradually extruded through the external urethral meatus.[2] The benign nature of this disease contrasts with the anxiety of parents who very often attribute it to sexual abuse thus causing serious medico-legal issues.[2,4,5] Urethral mucosa prolapse is a rare often misdiagnosed condition in young girls or teenagers predominantly of the black race. It was first

described nearly 250 years ago by Solingen.[4,5] The exact incidence is not well known, with a suggested incidence of 1 in 3000.[1,3,4]

The exact pathophysiology of urethral mucosa prolapse is not clear. One theory is of poor attachment between the inner longitudinal and outer circular smooth muscle layers of the urethra, in association with recurrent episodes of increased intra-abdominal pressure.[1,5,6] Predisposing factors like chronic cough and constipation have been suggested, as having above average height and weight for the age.[4-6] Over weight and obesity affect intravesical pressure in adults but this has not been proven in the paediatric population.[4] Other predisposing factors include oestrogen deficiency, poor nutrition, trauma to the urethra, urinary and vaginal infections;excess urethral mucosa and physical exertion.[4-6] Race and socioeconomic status may play a role due to its preponderance among Africans and girls from lower socioeconomic status.[1,7,8] Certain cultural practices like female genital mutilation may be a predisposing factor in the development of urethral prolapse.[6]

Urethral mucosa prolapse is mostly asymptomatic but it can present with vaginal bleeding causing spotting on the underwear or diapers, periurethral mass, dysuria, haematuria, frequency, and urgency especially in prepubertal and post-menopausal cases.[6-8] Vaginal examination commonly will reveal a pinkish/purplish or reddish circular urethral mucosa at the external meatus with a dimple on it.[2,9-11] Catheterization of the bladder often confirms the diagnosis when urine is drained.[11-13] In adolescent and postmenopausal patients, with urethral mucosa prolapse, vaginal bleeding associated with voiding symptoms are fairly common[1,6,7]and this was the experience in this patient. If the prolapsed urethral mucosa is large, the mucosal mass may become strangulated, which results in venous obstruction, thrombosis and necrosis of the prolapsed tissue.[1,6,7,14] This is because; the swelling and congestion create a purse string effect around the distal urethra impeding venous return and vascular congestion. If left untreated this may lead to strangulation and eventual necrosis of the protruding tissues. Most of the symptoms of urethral mucosa prolapse can readily be ascribed to sexual abuse and this may cause anxiety to the parents as was observed in this case.[1,7,15] Hence, the treating physician should be aware of the fact that until sexual abuse is ruled out and diagnosis is confirmed, parents will likely expressed worries.

Diagnosis of urethral mucosa prolapse is essentially clinical and is made by visualization ofa circular protrusion of the distal urethra through the external meatus and catheterization of the central opening within the prolapsed urethral mucosa.[6-8] Extensive investigations is not necessary to make the diagnosis; however, ultrasound scan can be done to exclude other abnormalities and even malignancy of the kidneys and urinary bladder.[6-8]

The differential diagnosis of this condition is urethral caruncle which is described as a fleshy outgrowth of the distal mucosa usually originating from the posterior lip; however, urethral caruncles are focal, while urethral mucosa prolapse are circumferential.[1,13-14]Other differential diagnoses of a paediatric inter-labial

mass include ectopic urethrocele, Gartner's cyst, rhabdomyosarcoma, urethral papilloma and urethral diverticulum.[5-7]

Treatment modalities range from conservative management to various operative interventions. Surgical treatment remains the gold standard as it allows for the restoration of functional anatomy, especially in cases of severe prolapse, significant bleeding with necrotic mucosa or failed medical management.[5,7,8] The modified Kelly-Burnham operation in which the prolapsed mucosa is excised in quadrants; and the healthy mucosa is reapproximated with interrupted absorbable sutures is the preferred surgical technique which was done for this patient. The advantage with this technique is that, the urethra hardly retracts inward with associated difficulty to trace it for mucosal to mucosal anastomosis. The technique is also associated with low risk of post-operative bleeding, recurrence and meatal stenosis.[6-8]Ligation around a Foley catheter is no longer used because of such complications as recurrence, infections and postoperative pain.[9,10] Other surgical techniques include Keefe vagina/urethra placation, surgical reduction maintained with mattress sutures, manual reduction and cautery excision.[6-8] Complete and circumferential surgical excision of prolapsed mucosa ensures good results within 24 to 72 hours[9,10].

Medical treatment includes local hygiene, sitz bath, topical antibiotics, steroids or oestrogen cream.[6-8] Most of the success with the medical treatment is with the topical application of oestrogen cream. Oestrogen cream can be applied locally for 2 to 3 times a day in the outpatient if necessary.[2,3,6] The essence of the sitz bath is to decrease swelling and oedema and antibiotics use is to treat/prevent local infection.[15] Medical management can be associated with high failure rates and a recurrence rate of 67%.[7-9]

Postoperative complications following urethral prolapse repair may include urethral stenosis and urinary incontinence.[1,4,10] To help prevent urethral scarring, some authors prefers to perform this procedure on healthy oestrogenized tissue and ensure complete approximation of the urethral mucosa to the vaginal epithelium to prevent retraction of the urethral mucosa.[12,13,15] Postoperative urinary incontinence may occur owing to the diminished urethral length following excision; however, this is rare.[14,]

Conclusion

Although rare, urethral mucosa prolapse is noticeable in the black pediatric population and has excellent outcomes when treated surgically. It is though rare in pubertal/adolescence girls, diagnosis of urethral mucosa prolapse should be considered in cases of unexplained vaginal bleeding even in pubertal/adolescence girls. Diagnosis is primarily clinical, and surgical resection provides a definitive and cost-effective treatment. The key in the management is to individualize the patient and offer the best treatment.

Disclaimer (Artificial intelligence)

Author(s) hereby declare that NO generative AI technologies such as Large Language Models (ChatGPT, COPILOT, etc) and text-to-image generators have been used during writing or editing of manuscripts.

REFERENCES

1. Armand Mayala Ma Mayala et al. Diagnosis and management of a urethral prolapse in a 6-year-old girl: a case report. Pan African Medical Journal. 2021;39(284). 10.11604/pamj.2021.39.284.30954. Available online at: https://www.panafrican-med-journal.com//content/article/39/284/full

2. Saida Hidouri, Sabrine Ben Ammar, Sana Mosbahi, Mohamed Ali Chaouch , Besma Gafsi d, Mongi Mekki . A case report of urethral prolapse in young girls: Clinical manifestations and surgical management.International Journal of Surgery Case Reports 120 (2024) 109879.

3. Akadiri O, Ajenifuja KO, Bakare B. Urethral Mucosa Prolapse in an 18-year-old Adolescent. Case Report in Obstetrics and Gynaecology 2013; Article ID 231708, 3 pages

4. Jalloh M, Heibig J, Gaye O, Ghaul W, Yankeleich G, Ndoye M, et al. Urethral Prolapse: Surgical and Social Consideration in Senegal. Case Report in Urology2022; Article ID 551416, 3 pages

5. Fornari A, Gressler M, Murari JCL. Urethral Prolapse: A Case Series and Literature Review. Journal of Obstetrics and Gynaecology of India 2020;70:158-162

6. Yddoussalah O, Qudrhiri M, Sumba H, Karmoun T, Elkhader K, Kuntani A et al. Urethral Prolapse in Young Girls. J Urol Res 2018:5(2),1101

7. John CO, Alagbeleye JO, Enyindah CE. Childhood Urethral mucosas prolapse in Port Harcourt, Nigeria. An 11 year Experience. The Nigerian Health Journal 2015;15(2):79-83

8. Dieth AG, Tembely S, Mieret JC, Toure A, Yaokreh JB, Kouame YGS et al. Urethral Mucosal Prolapse in girls in Abidjan (Cote d'Ivoire). Int J Women's Health Care 2016;1(1):1-3

9. Fiogbe MA, Hounnou GM, Koura A, Agossou-Voyeme KA. Urethral mucosas prolapse in young girls. A report of nine cases in Cotonou. Afr J Paediatr Surg 2011;8:12-14

10. Igwebueze OI, Asimadu EE. Premenarcheal Urethral Mucosa Prolapse in Enugu, Southeast, Nigeria. Five case series. J Women's Health Care 2015;4(4):243

11. Yddoussalah O, Odudrhiri M, Sumba H, Karmouni T, Elkhader K, Koutani A et al. Urethral Prolapse in young Girls. A Report of Eight Cases. J Uro Res 2018;5(2):1101

12. Pulavarthi S, Vijayan S. Prolapse of the Urethral mucosa in a pubertal Indian girl. Int J Reprod Contracept Obstet Gynaecol 2018;7(8):3397-3399

13. Gbobo, Isesoma, and Goddy Bassey. 2021. “Urethral Prolapse in Prepubertal Girls in Port Harcourt, Nigeria: A Multicentre Study”. Journal of Advances in Medicine and Medical Research 33 (19):120-25. https://doi.org/10.9734/jammr/2021/v33i1931086.

14. Ballouhey Q, Galinier P, Gryn A, Grimando A, Pienkowski C, Fourcade Let al. Benefits of primary surgical resection for symptomatic Urethral Prolapse in children. Journal of Paediatric Urology 2014;10:94-97

15. Aprile, A., Ranzato, C., Rizzotto, M. R., Arseni, A., Da Dalt, L., & Facchin, P. (2011). “Vaginal” bleeding in prepubertal age: a rare scaring riddle, a case of the urethral prolapse and review of the literature. Forensic Science International, 210(1-3), e16-e20.