**Atypical Presentation of Urethral Mucosal Prolapse in a Prepubertal Girl: A Case Report and Literature Review**

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ABSTRACT

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| Urethral mucosal prolapse is a rare benign condition that is often misdiagnosed by clinicians and associated with fear of sexual assault by parents. It occurs in both children and postmenopausal women. We report a case of urethral mucosal prolapse in a 6-year-old pupil who presented with vaginal bleeding for which the parents thought she was sexually assaulted in school; to buttress the need for sensitisation on urethral mucosa prolapse in schools and during community outreaches. Vaginal examination revealed a well-circumscribed doughnut shaped fleshy mass at the external urethral meatus, for which a diagnosis of urethral mucosal prolapse was made. Although surgery is indicated in some cases, she was managed medically with topical oestrogen cream and the symptoms resolved. Prepubertal urethral mucosal prolapse is a benign condition and often misdiagnosed; early diagnosis and management will allay parental fears and prevent sequelae. Female education and community sensitization will increase awareness level of the condition. |

***Keywords****:* Urethra, Mucosa, Prolapse, RSUTH

1. INTRODUCTION

Urethral mucosal prolapse occurs when the mucosa evaginates beyond the urethra meatus (Fiogbe et al., 2011). Although it is very rare in young women, occurs in children and postmenopausal women (Fornari et al., 2020). It is a circular complete eversion of the terminal urethral through the external urethral meatus (Igwebueze & Asimadu, 2015; John et al., 2015). It is reported mostly in the prepubertal black female population and the Caucasian postmenopausal women with a global incidence of 1:3000 in the paediatric group (Fornari et al., 2020; Jessop et al., 2016; John et al., 2015).Urethral mucosal prolapse has a bimodal age distribution, seen mostly in prepubertal girls aged 1-9 (the average age at presentation of 4 years) and postmenopausal women(Abhulimhen-Iyoha & Oguejiofor, 2017; Fornari et al., 2020).

The aetiology is unknown but several theories have been proposed such as genetics and intrinsic urethral anomalies (such as increased urethral motility, poor attachment between the inner longitudinal and outer circular smooth muscle layers of the urethra). Thus, the urethra when subjected to predisposing factors like increased intra-abdominal pressure from chronic cough, constipation or obesity can lead to urethral mucosal prolapse (Abolarinwa et al., 2013; Igwebueze & Asimadu, 2015; John et al., 2015).Poor hygiene and nutrition, poor and low oestrogen which might be the case in prepubertal age and postmenopausal period are other factors implicated (Abhulimhen-Iyoha & Oguejiofor, 2017). Often urethral mucosal prolapse and its complications are misdiagnosed as sexual assault creating anxiety in parents, care givers and attendants. We report this case to buttress the need for awareness creation on the topic.

2. CASE REPORT

We report a case of a 6-year-old basic three pupil who was brought to children emergency ward of our hospital on account of bleeding per vaginam 7 hours prior to presentation. Blood stain was noticed in her pants after urination in school. It was the first episode. The blood was dark red, not profuse and no history of passage of blood clots. There was no history of trauma to the external genitalia, or urinary tract, no history of sexual assault or fall astride. There was no history of bleeding from other body orifices. She had not developed secondary sexual characteristics. She had no history of chronic cough, constipation or child labour. There was no history of similar complaints in her older siblings. Her parents thought she was sexually assaulted in school and brought her to hospital for clarification. Her developmental milestones were appropriate for her age. She has not had any previous hospital admissions. She had no history of systemic illnesses and had not undergone any surgery. On examination at presentation, she was not pale, anicteric, acynnosed, afebrile (T=36.40 C), not dehydrated, no lymphadenopathy. Her respiratory rate was 19 cycles per minute. Her chest was clinically clear. Partial pressure of Oxygen was 98%. Cardiovascular and abdominal examination findings were normal. Vaginal examination revealed relatively normal female external genitalia, reddish circumferential periurethral mass at the external urethral meatus, soft and fleshy (Figure 1). The hymen was intact and no active vaginal bleeding. A size 6 urethral catheter passed through the meatus yielded clear urine. Her packed cell volume was 36%, blood group O rhesus D positive, white blood cell count was 9.1x109/l and platelet count were 284 x 109/L. A diagnosis of urethral mucosal prolapse was made for which she and her parents were reassured and counselled on the modality of management. She was placed on topical estrogen for two weeks, sitz bath twice per day, analgesics and haematinics. On four weeks follow-up visit, her clinical condition improved remarkably and the prolapse resolved.



Figure 1. Perineum showing fleshy circumferential mass at the urethral meatus (urethral mucosa prolapse)

3. discussion

Urethral mucosal prolapse is common among children and postmenopausal women (Gynecology, 2021). It is a rare benign condition that is often misdiagnosed by clinicians and associated with fear of sexual assault by parents. It is the protrusion of the urethra that is associated with various presentation such as blood stain in underwear, bleeding, partially thrombosed mucosa, urinary retention and pain (Fornari et al., 2020; Gynecology, 2021). Solingen described urethral mucosal prolapse first in 1732 (Abuhasanein et al., 2021; Ballouhey et al., 2014).

The aetiology is unknown but several theories have been proposed such as genetics and intrinsic urethral anomalies (such as increased urethral motility, poor attachment between the inner longitudinal and outer circular smooth muscle layers of the urethral). Thus, the urethra when subjected to predisposing factors like increased intra-abdominal pressure from chronic cough, constipation or obesity can lead to urethral mucosal prolapse (Abolarinwa et al., 2013; John et al., 2015). Poor hygiene and nutrition, poor and low oestrogen which might be the case in prepubertal age and postmenopausal period are other factors implicated (Igwebueze & Asimadu, 2015; John et al., 2015)

The condition might be asymptomatic but the most common presentation is vaginal bleeding with associated periurethral mass (Abolarinwa et al., 2013; Fornari et al., 2020).These were the two presenting complaints of our patient.

Complicated cases can present with urinary symptoms; frequency and dysuria following congestion, strangulation and necrosis (Abolarinwa et al., 2013; John et al., 2015). These symptoms were not noticed in index patient.

The characteristic finding is a doughnut shape periurethral mass, that may be tender and bleeds on contact. If congested the mucosa might appear bright red or dark and cyanosed. It can be differentiated from trauma, polyps, prolapsed urethrocele, tumours, caruncles or sexual abuse by examination (Ballouhey et al., 2014)

Diagnosis of urethral mucosal prolapse is clinical, confirmed by the presence and positive catheterisation of a central opening in the protruding mass yielding sprouts of urine as was done for our patient. Extensive investigation is not necessary to make the diagnosis (Fornari et al., 2020; John et al., 2015)

The options of treatment can be either medical or surgical management. Medical management is the first line of treatment for asymptomatic and mild symptomatic cases. It includes the use of sitz bath, local antibiotic, steroid or topical Oestrogen cream. It was the first-line management for index patient and the symptoms resolved. Medical management is not always as effective as surgical methods and has a recurrence rate of 26-67% (Abhulimhen-Iyoha & Oguejiofor, 2017; Abolarinwa et al., 2013). In a few cases, topical oestrogen complications occur such as breast buds, vulva hyperpigmentation, vulva erythema, fire downy labial hair and vaginal bleeding. However, these features are benign and usually subsides with discontinuation of their use. Her parents were educated on this when she developed some of the complications. Although not used in index patient, the surgical option is used when the medical method fails, and often for severe cases with significant bleeding, thrombosis or gangrenous changes. However, in some regions, surgical management is routinely undertaken because the long-term follow-up of children is erratic and unreliable (Jessop et al., 2016). More so, the surgical method has a low recurrence rate of 5% (Abolarinwa et al., 2013; Pulavarthi & Vijayan). The surgical procedure serves to remove the non-viable tissue and restore the two muscle layers of the urethral to their normal anatomic state of apposition.(Ballouhey et al., 2014; Jessop et al., 2016). Surgical treatment involves complete surgical excision with various modifications, in which the prolapsed mucosa is excised after insertion of stay-sutures and the mucocutaneous junction is reapproximated with absorbable sutures. The surgical procedures available are: modified Kelly-Burnan operation, Keefe vaginal/urethral plication, manual reduction, ligation over a urethral catheter, cryosurgery, cautery excision (Ballouhey et al., 2014). Stewart reported a simple ingenious method of suturing the neo-meatus after prolapse excision by transfixing the distal urethral transversely and vertically which avoids retraction of the fresh endo-urethral mucosal circumference.

Like other operative interventions, surgical management for Urethral Mucosal Prolapse can be complicated by urethral stenosis, urinary incontinence, recurrence, meatal stricture and anaemia. (Hill et al., 2016; Rudin et al., 1997)

Our patient presented with vaginal blooding and periurethral mass which the parents thought she was sexually abused. The case of urethral mucosal prolapse can be diagnosed from the clinical presentation and the classical examination findings. There is need to create awareness of this condition in schools perhaps during school health programme for teachers, students to be aware of the condition. Such awareness should also be created in the community by health care professionals and during Parent -Teachers meeting for parents to also be acquainted with this gynaecological condition in children. The parents of our patient were not aware of the condition and affirmed they had never heard of it. This buttresses the need for awareness creation on occurrence of urethral mucosa prolapse in children in schools and during community outreaches.

4. Conclusion

Prepubertal urethral mucosal prolapse is a benign condition and often misdiagnosed; early diagnosis and management will allay parental fears and prevent sequelae. Female education and community sensitization will increase awareness level of the condition and enhance early presentation and management. The clinicians should also be familiar with this condition to avoid misdiagnosis.

**DISCLAIMER (ARTIFICIAL INTELLIGENCE)**

Authors 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 this manuscript.

Consent

All authors declare that ‘written informed consent was obtained from the patient’s parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial office/Chief Editor/Editorial Board members of this journal.

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