

# Childhood Urethral Mucosa Prolapse in Port Harcourt, Nigeria: An 11-year Experience

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## **ABSTRACT**

### **BACKGROUND**

*Childhood urethral mucosal prolapse occurs in girls between the ages of six months and eight years. It can be associated with significant morbidity with resultant urethral loss especially with poor management.*

### **OBJECTIVES**

*To review our experiences of managing urethral mucosal prolapse in children over an eleven year period and the outcome of treatment options.*

### **METHODOLOGY**

*This was a retrospective study of all pre-pubertal patients diagnosed and managed at the University of Port Harcourt Teaching Hospital from January 1<sup>st</sup>, 2003 to December 31<sup>st</sup>, 2013.*

### **RESULTS**

*A total of 13 patients were seen with childhood urethral mucosal prolapse. The mean age was 7.2 years with age range of 3-13 years. Twelve patients (92.3%) presented with genital bleeding, 11 (84.6%) patients had genital swelling while 10 patients (76.9%) of the patients presented with both symptoms. Urine culture was done for 11(84%) of the 13 patients and only 4 (36.3%) of the urine culture grew bacteria. Only 2 (15.4%) of the patients had mild symptoms and were managed conservatively with sitz bath and application of estrogen cream. Two out of the eleven patients who needed*

*surgical treatment were not done due to financial constraints. All surgical repairs were successful. There were no postoperative complications recorded in this study and no recurrence at follow up.*

### **CONCLUSION**

*This study shows that either medical or surgical treatment yields result. Medical practitioners who attend to children must be familiar with this condition to make prompt diagnosis.*

### **KEYWORDS**

*Childhood urethral mucosa prolapse; Port-Harcourt.*

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### **INTRODUCTION**

Urethral mucosal prolapse was first described in 1732, as a condition that 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.<sup>1,2</sup> This results in a congested and edematous reddish purple mass that bleeds. It is a rare condition that occurs most often in prepubertal girls and postmenopausal women and primarily in native African girls and white women respectively.<sup>1,3</sup> There have however been reports involving Hispanics and white girls.<sup>4,5</sup> This condition which is rarely diagnosed, can be associated with significant morbidity with resultant urethral loss especially with poor management.

Childhood urethral mucosa prolapse has an estimated global incidence of 1 in 3000.<sup>6</sup> Estimated isolated incidences in Nigeria include 19 cases in a 10 year period while a higher incidence of seven cases/year have been reported in Cote d'ivoire.<sup>7,8</sup>

Mucosal urethral prolapse is particularly commoner in the tropics than elsewhere<sup>2</sup>, with low socio-economic class being suggested as a predisposing factor. Though this assertion is unproven, the findings may be a reflection of the class of patients who generally attend such hospitals.<sup>9</sup>

Reports from studies indicate that UMP mucosal occurs more commonly in girls between the ages of six months and eight years, though isolated cases in adolescent girls have being reported.<sup>10</sup>

Since its description by Solingen, the aetiology of this condition is still not clearly understood but various predisposing factors have been identified.<sup>1</sup> These include; inadequate perineal muscular attachment, episodic increase in intra-abdominal pressure, urinary tract infections, female genital mutilations, urinary tract infection, estrogen deficiency and mucosal redundancy.<sup>10</sup> These factors lead to disruption of the muscular layers with resultant complete and circular eversion of the mucosa and eventual prolapse.<sup>10</sup> 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.<sup>10</sup>

Childhood urethral prolapse is predominantly asymptomatic and is often an incidental finding during routine examination. The most common presentation is vaginal bleeding, which is often misdiagnosed by primary care practitioners.<sup>11</sup> Other presenting features include discharge per vaginam, urinary retention, dysuria, frequency, introital pain or hematuria.<sup>2,10,12</sup> Laboratory and radiological evaluation are not necessary in most cases.<sup>13,14</sup> Differential diagnosis include urethral

caruncle, prolapsing urethrocele, condylomata, urethral papilloma, polyp, vaginal rhabdomyosarcoma, imperforate hymen and sexual abuse.<sup>13,14</sup>

Treatment options available are medical and surgical treatment. Medical treatment involves the use of sitz bath, local application of antibiotics and steroids.<sup>2,15</sup> This can be palliative with associated high failure rates and recurrence<sup>5</sup>. Surgical treatment is better for restoration of the functional anatomy.<sup>13</sup>

The aim of this study there is to review our experiences in the management of childhood mucosal urethral prolapse over an eleven year period at the University of Port Harcourt Teaching Hospital (UPTH). Despite being a rare condition, it is subject to various misdiagnosis, delayed treatment and management by medical practitioners who attend to children. Importantly this study aims to create the awareness of this condition and the importance of multi-disciplinary approach and referral to an eventual satisfactory cure of a rare diagnosis.

## MATERIALS AND METHODS

This was a cross sectional retrospective study of all the cases of pre-pubertal patients who were diagnosed and managed at the University of Port Harcourt Teaching Hospital from January 1st 2003 to December 31st 2013 at the University of Port Harcourt Teaching Hospital Port Harcourt, River State. Nigeria. The case files of all patients with childhood urethral mucosal prolapse between the stated periods were retrieved from the medical records department of the hospital for analysis. Ethical approval was obtained from the UPTH ethics committee. Information obtained from the patient's chart included age at presentation, clinical presentation, pre-operative evaluation, type of surgery and outcome. Data collected was entered into a spread sheet using SPSS 15.0 for Windows<sup>®</sup> statistical software which was also used for analysis. Results are presented as means with standard deviations and rates in tables and figures.

## RESULTS

During the eleven year study period, a total of 13 patients were seen with childhood urethral mucosal prolapsed (Table 1). All the patients were pre-pubertal African girls. The average age was 7.2 years with age range of 3-13 years. The clinical presentations were genital bleeding alone in 12(92%) patients, genital swelling alone in 11(84%) patients and both symptoms in 10(76%) of the patients. There was no history of urinary symptoms in any of the patients. Examination revealed congested and inflamed circumferential mass of a 0.5-2cm lump protruding around the urethral meatus. The urethral opening was clearly visible in all patients. None of the patients had previously had a female genital mutilation (FGM). Of the 13 patients, 11(84%) had urine culture as part of their preoperative evaluation; and 4 (36.3%) out of the eleven patients culture grew bacteria (Table 2). In all, two patients were managed conservatively with Sitz bath and application of oestrogen cream because their symptoms were mild even if the diagnosis was certain. Their proplase resolved successfully with no recurrence. Eleven patients needed surgical treatment however only nine had an eventual repair. Two were lost majorly due to financial constraints. All surgical repairs were successful.

The four quadrant excisional technique was applicable to eight of the 8 surgical patients and excision with a catheter in situ in 1 patient. There were no postoperative complications recorded in this study and no recorded case of recurrence at follow up. Consultants were involved in all the surgical cases.

## DISCUSSION

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.<sup>2</sup> The underlying cause of this condition remains uncertain, but lack of oestrogen is thought to have a role because of

**Table 1.** Characteristics of patients treated for urethral mucosal prolapse in eleven years (2003-2013)

AGE	FREQUENCY	PERCENTAGES
3-5	3	23.1
6-8	7	53.8
9-11	2	15.4
12-14	1	7.7
MODE OF PRESENTATION		
GENITAL BLEEDING	12	92.3
GENITAL SWELLING	11	84.6
URINARY SYMPTOMS	0	0

**Table 2:** Urine culture result of patients treated for urethral prolapse in eleven years (2003-2013)

	Frequency	Percent	Valid percent	Cumulative percent
Sterile	5	38.1	38.5	61.6
Not done	4	30.8	30.8	92.3
Klebsiellasp	3	23.1	23.1	92.3
Staph. aureus	1	7.7	7.7	100.0
Total	13	100.0	100.0	100.0

the preponderance of the condition in the pre-pubertal and postmenopausal age groups.<sup>12</sup> A common theory is that the problem arises as a consequence of poor attachments between the two layers of smooth muscle surrounding the urethra combined with episodic increases in intra-abdominal pressure.<sup>16</sup> Previous retrospective studies found that 50% of the patients included an antecedent history of cough, trauma or constipation or large for their age girls leading to straining and distention.<sup>5,16</sup> Despite this, in majority of cases and with this study, there were no obvious predisposing factors.

The mean age of presentation in this study was 7.2years; this was similar to results from previous studies<sup>7,8</sup>. The role of urinary tract infection in the pathogenesis has been emphasized in some studies as a predisposing factor.<sup>10</sup> In this study, majority of the culture were sterile and this calls for further look into the role of U.T.I as a cause or due to the condition itself as suggested. The barbaric act

of FGM suggesting damage and or disruption to the perineal muscular attachment of the urethra was not seen in this study. This may be a reflection of the area of study. This finding also suggests that the role of FGM in the aetiology of childhood urethral mucosal prolapse is negligible. The most common presentation of childhood urethral mucosal prolapse in this study is genital bleeding. This presented as spotting on the underwear or diapers. This is in keeping with the findings from previous studies.<sup>10,18</sup> It is however at variance with the study done in London, in which a periurethral mass was the most common presentation.<sup>12</sup> These findings are erroneously ascribed to sexual abuse of the child and some practitioners make this diagnosis or have a high index of suspicion in this age group until a urethral prolapse is definite.<sup>4,11</sup> The diagnosis was easily made in 11 of the patients based on the presentation of periurethral mass (doughnut shaped mass) and genital bleeding.

Both medical and surgical options of treatment were carried out in this study. Two patients were successfully managed with estrogen cream and sitz bath as an alternative to surgical approach with no reoccurrence. This finding collaborates other studies where either sitz bath or the use of estrogen cream as a medical approach of cure has a place in the management of childhood urethral mucosal prolapse.<sup>5,12</sup> Both patients presented with only genital bleeding. It is therefore advised that medical approach should be individualized and offered to patients with mild symptoms.

Surgical treatment is believed to be better for restoration of the functional anatomy especially in cases of severe prolapse and necrosed mucosa.<sup>13</sup> The most popular surgery done in this center was the 4 quadrant excision. The other, being ligation of the mucosa over a catheter was performed in one patient. Other surgical methods available include; excision of mucosa with a catheter in situ, cauterization of the mucosa, cryosurgery and reduction of prolapse through a vagina lesion followed by suturing the circular muscle

fibers around the distal urethra.<sup>2,13,17</sup> The 4 quadrant excisional technique was introduced and more popular over the excision over the catheter due to the latter's disadvantage in the reduction of the proximal mucosa which made mucosal to mucosal anastomosis difficult, resulting in high risk of post-operative meatal stenosis.<sup>14</sup> In this study there were no post-operative complications or recurrence. The surgical outcome in this study is similar to that of a previous study done in New Jersey, USA.<sup>14</sup> This study shows that either medical or surgical approach is favourable unlike studies that challenge conclusions of proponents of both approaches. The key is to individualize the patient for the best management option. Though there was no reoccurrence in this study, it should be noted that it can occur in both forms of treatment. Proponents of surgical treatments as the primary treatment have suggested, cost effectiveness with a single admission, reducing anxiety and repeated medical attention compared to the medical approach. In contrast, the exposure of the young girls to anaesthesia, post-operative stenosis, dysuria and reoccurrence are possible side effects of surgical treatment.

## CONCLUSION

Urethral mucosal prolapse can definitely be diagnosed without laboratory or radiologic evaluation by demonstrating that the oedematous tissue surrounds the meatus circumferentially. Girls with mild symptoms can be managed conservatively. For girls with more significant symptoms, we recommend the simple four quadrant surgical excision technique under general anaesthesia. The key in the management is to individualize the patient and offer the best treatment option.

Proper evaluation is needed and mothers counseled to report to hospitals at the notice of genital bleeding and swelling. Also, awareness should be created so that all medical practitioners who attend to children will be familiar with this condition.

The need to assist families with financial constraints is advocated as the loss of follow up

could mean future disaster to the physical and mental health of the girls lost.

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