

An Unusual Cause of Vulvar Hemorrhage: About 12 Cases

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Abstract

Original Research Article

Vulvar or genital hemorrhage is an alarming reason for consultation that worries parents a lot. Several etiologies of vulvar bleeding have been reported, including sexual abuse (common in our socioeconomic context) or urethral prolapse. Urethral prolapse is the complete eversion of the distal congestive urethral mucosa through the urinary sphincter. It is a benign condition that is uncommon in children and occurs in girls with a peak in frequency between the ages of four and six, mainly in the black population. The clinical presentation is dominated by vulvar bleeding, which therefore poses a medico-legal issue and a differential diagnosis with other causes of genital hemorrhage. The diagnosis is purely clinical, presenting as a more or less voluminous pink or purplish edematous circumferential pseudotumor bulge that bleeds spontaneously. No paraclinical examination is necessary. We retrospectively analyzed the records of 12 prepubertal patients aged between 5 and 12 years who consulted for genital hemorrhage either post-traumatic or spontaneous with urethral prolapse in the Pediatric Surgical Emergency urethral prolapse department, over a period of 18 years (1998 - 2020). In our series, the age of diagnosis ranged from 5 to 12 years with a median age of 08 years. Eleven patients were Caucasian and only one patient was from Senegal. Other causes of vaginal hemorrhage were ruled out, including sexual abuse or post-traumatic bleeding. The treatment of this affection is subject to great controversy, between surgical and conservative treatment, all our patients benefited from surgical treatment except one patient who benefited from medical treatment. The risk of complications, in particular urethral stenosis and recurrence, is not negligible in the face of poorly conducted medical treatment or surgery. In our series, the clinical outcome was favorable in all patients. The duration of hospitalization could be shortened by an early removal of the urinary catheter, thus a lower hospitalization cost.

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INTRODUCTION

Vulvar or genital hemorrhage is an alarming reason for consultation that worries parents a lot. Several etiologies of vulvar bleeding have been described, including sexual abuse (common in our socioeconomic context). In our study, the main etiology of bleeding, although rare, was urethral prolapse, which is diagnosed clinically. All other causes of genital bleeding in prepubertal girls -described in several studies such as vulvar trauma or foreign bodies- were ruled out [1-6].

Urethral prolapse is a rare condition in children with an incidence of (1/3000) [10]. It was first described in 1732 by Solingen. The therapeutic management is still subject to controversy. This pathology is predominant in prepubertal girls [12, 13] of sub-Saharan origin. The most frequent reason for consultation is vulvar bleeding [13]. It is caused by the complete eversion of the congestive urethral mucosa through the urinary sphincter.

Clinically, it presents as a more or less voluminous pink or purplish edematous circumferential bulge that bleeds readily. It is manifested by bleeding, dysuria, or vulvar swelling.

Its presentation is typical, and the diagnosis is clinical. The etiology is difficult to elucidate and may give rise to a medico-legal issue with the suspicion of sexual abuse. Two types of treatment exist, either medical or surgical. Surgical treatment remains the treatment of choice and consists of circumferential excision of the prolapsed part on a Foley catheter, followed by a mucosal suture with absorbable material using separate stitches, this is called the four-quadrant technique. Results are observed from 48 to 72 hours after removal of the bladder catheter [14].

MATERIAL AND METHODS

Our retrospective study concerns a series of 12 patients whose initial reason for consultation was vulvar hemorrhage with the diagnosis of urethral prolapse. All

patients were operated between February 1998 and October 2021, in the pediatric surgical emergency department of the children's hospital in Rabat. Several parameters were studied.

The age of the patients ranges from 5 to 12 years with an average age of 8 years.

The main reason for consultation was the sudden onset of vulvar bleeding with the fear of sexual abuse or trauma.

After eliminating other causes of vulvar bleeding (vulvar trauma, intravaginal foreign body, hormonal disorders, early puberty, etc.), we studied the medical and surgical history in search of chronic pathologies, such as constipation or bronchial asthma, which may be the cause of elevated abdominal pressure. The infectious status of these girls was also assessed, including the search for recurrent urinary tract infections and oxyuriasis (Figure 1). The gynecological history gives us information on the age of menarche. Clinical examination first looked for secondary sexual characteristics according to Tanner's classification, followed by a meticulous examination of the perineal region. The latter revealed the size and type of prolapse and its classification.

No additional examinations were necessary: 02 of our patients had pelvic ultrasound, only one had a plain abdominal x ray, and all of them had a pre-op workup (blood count, serum electrolytes and blood crasis). No patient underwent colposcopy.

Eleven out of 12 patients received surgical treatment in the first line, only one patient received medical treatment. The surgical technique of choice used for all patients was that described and modified by Rudin *et al.*, The technique consisted of the introduction of a Foley catheter through the urethral meatus, with traction of the urethra by 6 wires that highlighted the prolapsed mucosa (Figure 2). Then 4 marker wires are placed to define 4 quadrants. Circumferential excision of the prolapsed mucosa is then performed quadrant by quadrant with progressive muco-mucosal suture (Figure 3 & 4). The foley catheter was kept for 5 days. All patients were discharged under local care and antibiotic prophylaxis with good evolution.



Figure 1: Urethral prolapse and oxyuriasis



Figure 2: Exposition of the urethra



Figure 3: Muco-mucosal suture



Figure 4: Circumferential excision of the prolapsed

RESULTS

It appears from our research that urethral prolapse as a cause of vaginal bleeding remains a very rare condition. The largest series reported is that of Soderstrom HF *et al.*, (86 cases collected between 2001 and 2011). The diagnosis of urethral prolapse as a cause of vaginal bleeding was not retained in any patient of their series. Other series concerning urethral prolapse in girls have been reported by Da Silva-Anoma *et al.*, (65 cases in nine 9 years [5, 6], Rudin, J. E *et al.*, (58 cases in 15 years) [17]; Jerkins GR *et al.*, (40 cases in 19 years) [18]; Doletskii Sla *et al.*, (39 cases in 19 years) [12], and Hillyer S *et al.*, (34 cases in 23 years) [19].

Our series of 12 cases was collected over a period of 19 years with an epidemiological profile including one patient of black race of Senegalese origin and a variable socio-economic level. 6/12 of our patients were in the pre-puberty period.

The age of diagnosis varies between 05 and 12 years with a median age of 08 years.

Concerning our patients predisposing factors: none of our patients had a history of early puberty or sexual abuse, only one patient had a history of vulvar trauma. In the context of urethral prolapse, none of the patients had a history of asthma or pneumopathies that could cause chronic cough, one patient had chronic constipation, only one had recurrent urinary tract infections, and one had oxyurosis confirmed intraoperatively.

Symptoms and Clinic

The reason for consultation is vulvar bleeding for all patients.

Clinical examination allowed the diagnosis of urethral prolapse in all patients, which appeared in the majority of cases as a circumferential, pseudo-tumoral bulge, more or less voluminous, pinkish or purplish, edematous, bleeding on contact.

Paraclinical examinations: 3 patients benefited from an abdominal ultrasound; one patient benefited from a plain abdominal x ray.

Regarding the therapeutic management, one patient benefited from a clinical examination under general anesthesia with manual reduction of the prolapse followed by medical treatment with estrogen creams and sitz bath. No patient underwent colposcopy.

11 patients underwent surgical treatment in the first line, according to the surgical technique described by Rudin. First, a catheterization of the urethral meatus was performed by inserting a Foley catheter, followed by a resection of the prolapsed mucosa around the perimeter, followed by a muco-mucosal suture (between healthy urethral and vulvar mucosa), using the four-quadrant technique. The urinary catheter was removed after 5 days and the patients were discharged under local care, after an average hospital stay of 5 days.

The postoperative course was favorable for all of them.

The patients were seen after one month and then at 6 months: no case of urethral stenosis, urinary retention, or recurrence was reported. The hymen was intact in all patients.

DISCUSSION

UP as a cause of vaginal bleeding is a very rare condition in prepubertal girls. In our series all patients consulted for vulvar bleeding. The main reason for concern that alarmed the parents was sexual abuse, which in our socioeconomic context is very common (concern about hymen rupture). In all patients, sexual abuse was ruled out by clinical examination as well as vulvar trauma.

In one study, vaginal bleeding after sexual abuse was reported in only 11% of girls. There appears to be a reluctance to seek help when child abuse occurs [1, 7]. In other studies, the cause of vaginal hemorrhage remains unexplained [8-10]. One author concluded that the lack of identification of the cause of bleeding leads to the conclusion that the etiology of most cases is vulvovaginitis.

The exact etiology of UP in our series remains unknown, several theories have been evoked. Abnormalities of the structures that make up the pelvic floor, such as weak muscle fiber attachments, or a problem with urethral mobility such as hypermobile urethra syndrome as well as intrinsic abnormalities of the urethra, such as a dilated urethra, or an excess of urethral mucosa. Other theories include intrinsic abnormalities of the urethra, neuromuscular disorders and urethral malposition, or connective tissue abnormalities. All of these abnormalities may be the cause of urethral prolapse. It is therefore difficult to explain exactly the origin of urethral prolapse but a thorough research of risk factors has allowed us to get closer to it.

The most popular theory is that proposed by Lowe *et al.*, [20], who described the cause as cleavage between the oblique circular muscle layer and the smooth longitudinal layer due to increased intra-abdominal pressure. This situation has been raised in several studies and in particular in a study of 34 girls identified in the department of pediatric urology in Brooklyn, New York with a figure of 69% [12]. Elevated abdominal pressure is seen in bronchial asthma, chronic constipation, abdominal trauma, or recent surgery.

Regarding overweight: previous studies by Valerie *et al.*, [22] and Sugerma *et al.* have reported that overweight children are prone to develop urethral prolapse by the mechanism of increased abdominal pressure. In our series none of these factors were found in our patients, except for one patient who presented with post-traumatic vulvar bleeding and only one patient out of twelve presented with recurrent urinary tract infections, which raises another question: cause or consequence?

Indeed, according to some authors, infection/poor hygiene has also been proposed as a factor, although this is still disputed.

According to most authors, the age of the patients varies from 2.5 to 10 years with an average of 5.08 years [24-26].

The average age of our patients is 8 years. Prolapse is most frequent at the age of 5 years.

There is a classification of urethral prolapse that includes four types: Grade 1: minimal or partial prolapse, without inflammatory signs. Grade 2: circumferential prolapse, with edema. Grade 3: edematous mass prolapsed through the labia minora. Grade 4: severe bleeding, or necrosis or ulceration of the prolapse

In our series: 4 girls had a grade 1, 5 had a grade 2 and 3 patients had a grade 3.

Richardson *et al.*, reported in a series of 162 adult and pediatric patients that vulvar bleeding remains the most frequent mode of disclosure with a rate of 88% in girls. This is the case in our study, where all patients consulted for vulvar bleeding.

No further investigations to search for the etiology were performed. In their series of 58 cases, Rudin *et al.*, performed cystoscopy in 38 patients which revealed no abnormalities except for bladder inflammation. On the other hand, urodynamic examinations were performed in 18 girls and found neurogenic abnormalities in 7 of them. In our series, the anatomopathological study of the resected part is not specific as shown in the study of Rudin *et al.*,

The optimal management of PU remains controversial and depends on the severity of the symptoms (extent of vaginal bleeding and urinary signs). Conservative treatment appears to be effective for most patients with minimal symptoms: small size of the prolapse, little bleeding, but it has no place in cases of abundant bleeding or when the prolapse is large.

Surgical resection has been recommended as first-line treatment [23].

In our series the treatment was medical followed by manual reduction in one patient, and surgical in the remaining 11 girls. The postoperative follow-up was 100% favorable, no patient presented urethral stricture, nor urinary retention, nor recurrence. The surgical management in first intention seems to be the best compromise (short duration of hospitalization with no case of recurrence).

CONCLUSION

Genital bleeding is a frequent reason for emergency room visits in prepubertal girls. It appears that the management of genital bleeding in prepubertal girls is still poorly understood by health care providers.

The diagnosis of urethral prolapse is a rare condition, more common in the black population. The etiology is not yet elucidated but several theories have been reported such as increased intraabdominal pressure. The average age of the patients in our study was 8 years. The treatment of choice remains surgical with resection of the prolapsed mucosa which has given very good results with 0 cases of recurrence in our study. Medical treatment is reserved for cases where the symptomatology is minimal. The diagnosis is mainly clinical.

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