



Case report

“Vaginal” bleeding in prepubertal age: A rare scaring riddle, a case of the urethral prolapse and review of the literature

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ABSTRACT

The entities responsible for isolated vaginal bleeding are a broad spectrum of diseases. Vaginal bleeding in a prepubertal child is always treated as an alarming symptom both by parents and professionals. Most often, one of the first hypotheses is that a sexual abuse may be occurred and the clinical work up is oriented to explore it and eventually confirm or substantiate another diagnosis. Among the possible differential diagnosis urethral prolapse has to be considered.

We report the case of a 6 years-old Caucasian girl referred to our Service for a mild vaginal bleeding and with the suspicion of sexual abuse, excluded after the confirmation of the presence of urethral prolapse. The clinical picture and workup are described and compared with a review of the international literature on the issue.

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1. Background

The entities responsible for isolated vaginal bleeding are a broad spectrum of diseases, ranging from a simple urethral caruncle to rhabdomyosarcoma. Other differential diagnosis include hemorrhagic cystitis, hypothyroidism, precocious puberty, ovarian cyst presence, foreign body, exposure to exogenous sex steroids, and trauma which may or may not be associated with neoplasm, vulvovaginitis, urethral prolapse and sexual abuse (Table 1) [1].

Firstly described by Solinger in 1732, Urethral Prolapse (UP) is a rare disorder characterized by the circular protrusion of the distal urethral mucosa through the external meatus and forming an hemorrhagic sensitive vulvar mass that easily bleed, separated from the vagina [3]. According to the international literature, this disease usually seems to occur most commonly in prepubertal black females [4,5] and postmenopausal white women, even though the phenomenon has also been described in white female children [6], and very early in life [7,8]. Less frequently the urethral prolapse is strangulated [9,10].

In a case series of 58 white girls [6], this disorder was most common at the age of 3–5 years or 5–9 years, in very physically

active and rapidly growing young girls. Most prolapses occurred in the summer (53.4%). Only 2 of the girls were not of asthenic physique. Additionally other factors were found: perineal trauma in 12 and constipation, diarrhoea, or heavy cough in 10 girls. Most children were seen by a gynaecologist before the admission to surgical department.

In most of the cases the presenting symptoms is vaginal bleeding [11], but other symptoms have been described such as dysuria and variable difficulty in urination depending on the size of the protrusion and acute urine retention due to urethral strangulation.

The aetiology of this rare condition is still debated. Several theories have been proposed calling for a congenital or an acquired defect. The theory of the congenital origin includes weakness of the pelvic floor structures such as inadequate pelvic attachments and urethral hypermobility, but also urethral intrinsic abnormalities, e.g. abnormally patulous urethra, wide urethra or redundant mucosa [12].

From a structural point of view a weakened attachment between the inner longitudinal and outer circular-oblique smooth muscle layers of the urethra can easily determine a separation of the two muscle layers, for example during a rapid increase of the intra-abdominal pressure, and predispose to prolapse (Fig. 1).

Post-mortem examination in children have been reported by Lowe et al. [13]: most frequent anatomical defects detected were eversion of the urethral mucosa, vascular congestion of the corpus

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Table 1
Vaginal bleeding: differential diagnosis.

Category	Causes
Endometrial shedding caused by estrogen withdrawal	Endogenous
Vaginitis	Exogenous Non sexually transmitted disease (Shigella, Candida, Group A streptococcus) Sexually transmitted disease
Tumors	Foreign body Poor hygiene Clear cell adenoma Rhabdomyosarcoma (bladder, urethra, vagina) Endodermal carcinoma Mesonephric carcinoma Ovarian tumor Adrenal tumor Hemangioma Urethral papilloma
Urinary tract	Hematuria Hemorrhagic cystitis Urate crystals Ureteral Ectopia Ureteral Duplication Ureterocele Urethral prolapse Urinary tract infection Voiding Dysfunction Urinary incontinence
Trauma	Vaginitis Accidental (straddle injury) Abuse (inflicted trauma, sexual abuse, rape)
GI tract	Hematochezia Anal fissure Chron's disease
Metabolic	Hypothyroidism Liver cirrhosis Coagulopathy Precocious puberty McCune-Albright syndrome
Dermatosis Congenital/malformative	Ovarian cyst Lichen Sclerosus et Atrophicus Prune belly syndrome Malakoplakia[2]

spongiosum urethrae muliebris, and a cleavage plane between the inner longitudinal and outer circular-oblique smooth muscle layers of the urethra. The Authors sustained that the prolapse could result from poor attachments between the smooth muscle layers of the urethra in association with episodic increases in intra-abdominal pressure. The “abdominal pressure” hypothesis has been sustained by a clinical review of 12 cases, where 67% have had previous episodes of marked increases in intra-abdominal pressure.

The acquired conditions include trauma caused by sexual abuse, masturbation or malnutrition.

The physical examination usually allows attributing the urethral origin of the observed mass and the recognition that it is mucosal tissue.

The management of urethral prolapse ranges from medical therapy that consists of topical estrogen use to conservative surgical excision in whom medical therapies fail [14,15].

2. Literature review

At present, performing the search URETHRAL PROLAPSE, 779 items are retrieved, 696 on humans (1966–2008). Searching the



Fig. 1. Urethral prolapse: circumferential congested mucosa donut-shaped surrounding the urethral meatus.

quoted phrase URETHRAL PROLAPSE, 88 items were retrieved in the range period 1957–2009, 2 referred to veterinarian condition [16,17], 1 is an editorial of 1974 published in BMJ [18], 1 is a letter discussing the best treatment options by Douglas CP on his experience on 25 patients under the age of 8 [19], 1 on the pelvic support loss with defects similar to those observed in humans in 28 adult female squirrel monkeys [20], and 60 are not available (1957–2006) (Table 2).

Checking only for patients affected by UP, several manuscripts presented case(s) of UP in a more general record of cases.

We report the case of a 6 years-old Caucasian girl referred to our Service for a mild vaginal bleeding and with the suspicion of sexual abuse.

3. Case report

Paula (6 years old, Caucasian) was brought to the Emergency Room (ER) by her mother, late in the evening. The reason of entering the ER was described by Paula's mother with extreme anguish. “Paula came home after spending all the afternoon with her father, Mark. An hour later, while changing her for bed, I've noticed that her knickers were stained with blood”. She told she was afraid that her father could have sexually abused the child. She said to

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