Pediatric leiomyoma of the glans: a case report

V. BAGNARA¹, M. CASTAGNETTI², A.E. CALOGERO³, R.A. CONDORELLI³, R. CANNARELLA³, S. LA VIGNERA³

Abstract. – OBJECTIVE: The leiomyoma is a benign mesenchymal tumor originating from smooth muscle cells therefore its location is ubiquitous. The genitourinary system is not a common site and the glans localization in pediatric age has been described only three times in the literature to date.

CASE REPORT: We describe a case of an 11-year-old boy who presented with a painless, non-bleeding or itchy tumor of the glans. The surgical procedure consisted in the total removal of the mass. The histological study showed spindle cells with an eosinophilic cytoplasm while the immunohistochemical studies proved cells stained strongly positive for smooth muscle actin. The clinical follow-up for more than 5 years after surgery demonstrates the absence of recurrence and discomfort for the patient and a good aesthetic appearance of the glans.

RESULTS: Leiomyoma is a benign tumor that can originate anywhere there is smooth muscle. However, localization at the level of the glans can be treated with a total excision due to the presence of a cleavage plane with the surrounding tissues that allows a good reconstruction of the glans itself.

CONCLUSIONS: We propose that leiomyoma ought to be considered in the differential diagnosis of any glans mass in children.

Key Words:

Leiomyoma, Glans, Glans disease, Glans penis.

Introduction

Leiomyoma is a benign mesenchymal tumor originating from smooth muscle cells; therefore, its location is ubiquitous. Typically, it affects a single organ. However, also cases with localizations in multiple organs have been reported. The genitourinary system is not a common site. The kidney, especially the capsule, seems to be the most frequent location, and small tumors could

be randomly found, during surgery or autopsies! On the other hand, other organs of the genitourinary system can be affected, such as the ureter, the bladder, the urethra, the prostate, the seminal vesicles², the spermatic cord and the vas deferens, the epididymis³, the tunica albuginea, the testicle⁴, the penis⁵, the scrotum, the foreskin⁶ and the preputial frenulum¹.¬? Localization at the level of the glans, especially in children, is very rare³, with only three cases reported in literature to date. We herein describe the case of an 11-year- old boy who presented with a painless, non-bleeding or itchy tumor of the glans.

Case Presentation

The case herein reported concerns an 11-yearold boy who had noticed the appearance of a painless, non-bleeding or itchy growth of the glans in the absence of any trauma. At the clinical examination, the tumor appeared at the level of the right dorsal-lateral surface of the glans, reaching the balano-preputial sulcus, firm, painless, oval shaped with a maximum diameter of about 12 mm of a yellowish white color, hard consistency, firm, clear margins and slightly raised with respect to the mucous surface (Figure 1).

The surgical procedure consisted in submucosal isolation, by blunt dissection with micro-bipolar, of the neoformation, allowing its total "en bloc" removal, the saving of the mucous lining and the corpus spongiosum of the glans. The mass, in fact, was adherent but clearly dissociable both from the deep plane, consisting of the tunica albuginea of the corpus spongiosum of the glans, and from the superficial plane, consisting of the glandular mucosa (Figure 2). The subsequent reconstruction of the glans consisted of the juxtaposition of the glandular epithelium with separate stitches in 6/0 polydioxanone absorbable monofilament (PDSTM Ethicon Suture). The op-

¹Paediatric Surgery Unit, Policlinic G.B. Morgagni, Catania, Italy

²Section of Paediatric Urology, Department of Surgical, Oncological and Gastrointestinal Sciences, University Hospital of Padua, Padua, Italy

³Department of Clinical and Experimental Medicine, University of Catania, Catania, Italy



Figure 1. Clinical appearance of the lesion.

eration was performed under general anesthesia with penile block, without the use of tourniquets or adrenaline solutions. Only for better patient's postoperative comfort, a 12 Ch bladder catheter was kept in place for 24 hours.

The histopathological study described a well-circumscribed lesion due to the presence of a pseudocapsule of homogeneous appearance and consisting of fusocellular elements with eosinophilic cytoplasm, spindle nucleus with rounded edges. On immunohistochemical examination, the cellular structure showed high cytoplasmic positivity to specific smooth muscle alpha-actin and desmin while the S-100 protein resulted negative (Figure 3). The absence of cellular atypia, areas of necrosis and rare or absent mitosis finally allowed to exclude malignant nature of the lesion.

The clinical follow-up for more than five years after surgery demonstrates the absence of recurrence and discomfort for the patient and a good aesthetic appearance of the glans (Figure 4).

Discussion

Leiomyoma is a benign tumor that can originate anywhere there is smooth muscle. The origin of glans leiomyoma could be explained by the presence of smooth muscle cells in the blood vessel wall and in the trabeculae that separate the vascular lacunae of the corpus spongiosum⁹.

However, localization at the level of the glans is very rare and only a few cases have been described in the literature to date. A systemic review of the medical literature using the keywords "leiomyoma", "glans", "glans disease" and "glans penis" showed only 3 cases of pediatric glans leiomyoma reported up to June 2021. The first case of glans leiomyoma was reported by Belis in 1979 and concerned a 3-year-old child. Since then, only two other cases have been described by Stehr et al¹⁰ in 2000, in a 12-year-old teenager, and by Redman in 2000 in an 8-year-old boy11 (Table I). However, in the case reported by Stehr et al¹⁰ the excision of leiomyoma was only partial but the follow-up at 18 months showed the absence of symptoms. Other sporadic cases of glans leiomyoma have been described in adults.



Figure 2. Appearance of the tumor after glans mucosa detachment (A); Appearance of the glans after the "en bloc" excision of the tumor (B); Tumor after excision (C).

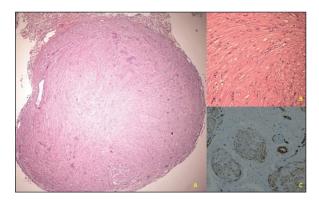


Figure 3. (H&E stain) Spindle cells with an eosinophilic cytoplasm at low (A) and high magnification (B); Immunohistochemical staining for smooth muscle actin (C).

In all cases of glans' localization, the treatment consisted in a total or partial excision of the tumor.

In no case a recurrence or malignant transformation is reported. In our experience, the total "en bloc" exeresis of the neoformation was possible by the presence of a cleavage plane that allowed an accurate and complete dissection of the lesion, saving the mucosa and the erectile tissue, corpus spongiosum of the glans, both adherent but not affected by the lesion and, therefore,

dissociable. The saving healthy tissue is certainly necessary for a more accurate reconstruction of the glans and limit serious and significant impairments both aesthetically and functionally.

In fact, after one year, the glans depression, residual on removal, and the surgical scar were barely visible. Furthermore, the boy did not suffer from discomfort, such as pain and/or itching, nor loss of sensitivity.

Conclusions

Although leiomyoma localized in the glans is a very rare condition, it must be kept in mind in the differential diagnosis of tumors that can occur here, such as plexiform neurofibroma, smooth muscle cell hamartoma, schwannoma, nerve sheath myoma and fibrohistiocytoma^{11,12}. The early and complete exeresis of this tumor is necessary to clarify the diagnosis and prevent further growth. In fact, even if confined to the subcutaneous layer, the growth of this tumor could lead to further difficulties in the excision and subsequent reconstruction of the glans with possible major repercussions both on an aesthetic and on a functional point of view.



Figure 4. Clinical appearance of the glans penis at one year follow-up.

Table I. Reported cases of leiomyomas of the penis in child.

Author	Year of publication	Patients' age (yr)	Location	Dimensions (cm)	Local recurrence
Belis et al ¹	1979	3	Glans penis	1.5	No
Stehr et al10	2000	12	Glans penis	< 1	No
Redman et al ⁶	2000	8	Glans penis	1	No
Our case	2021	11	Glans penis	1.2	No

Conflict of Interest

The Authors declare that they have no conflict of interests.

Informed Consent Statement

Informed consent was obtained from all subjects involved in the study.

References

- Belis JA, Post GJ, Rochman SC, Milam DF. Genitourinary leiomyomas. Urology 1979; 13: 424-429.
- Mendrek M, Bach C, Gaisa NT, Vögeli TA. Leiomyoma arising from the right seminal duct/seminal vesicle—Report of a rare case and review of the literature. Andrologia 2019; 51: e13174.
- Ozden O, Orhan D, Karnak I. Epididymal leiomyoma: an unusual intrascrotal tumor in a child. J Pediatr Surg 2009; 44: e5-7.
- Kullolli VS, Kullolli S, Pawar S, Gautam D. Leiomyoma of testis -a case report. Indian J Surg 2011; 73: 233-235.

- Liu SP, Shun CT, Chang SJ, Chen J, Hsieh JT. Leiomyoma of the corpus cavernosum of the penis. Int J Urol 2007; 14: 257-258.
- 6) Leoni S, Prandi S, Mora A. Leiomyoma of the prepuce. Eur Urol 1980; 6: 188-189.
- Moch H, Cubilla AL, Humphrey PA, Reuter VE, Ulbright TM. The 2016 WHO Classification of Tumours of the Urinary System and Male Genital Organs-Part A: Renal, Penile, and Testicular Tumours. Eur Urol 2016; 70: 93-105.
- Dehner LP, Smith BH. Soft tissue tumors of the penis. A clinicopathologic study of 46 cases. Cancer 1970; 25: 1431-1447.
- Christ GJ. The penis as a vascular organ. The importance of corporal smooth muscle tone in the control of erection. Urol Clin North Am 1995; 22: 727-745.
- Stehr M, Rohrbach H, Schuster T, Dietz HG. Leiomyom der Glans Penis [Leiomyoma of the glans penis]. Urologe A 2000; 39: 171-173.
- Redman JF, Liang X, Ferguson MA, Savell VH. Leiomyoma of the glans penis in a child. J Urol 2000; 164: 791.
- Bartoletti R, Gacci M, Nesi G, Franchi A, Rizzo M. Leiomyoma of the corona glans penis. Urology 2002; 59: 445.