



Contents lists available at ScienceDirect

Tzu Chi Medical Journal

journal homepage: www.tzuchimedjnl.com

Pathology page

Leiomyoma of the female urethra: A case report

Cheng-Sheng Chen^a, Chien-Chin Chen^{b, c, *}^a Department of Urology, Chia-Yi Christian Hospital, Chiayi, Taiwan^b Department of Pathology, Chia-Yi Christian Hospital, Chiayi, Taiwan^c Department of Cosmetic Science, Chia Nan University of Pharmacy and Science, Tainan, Taiwan

ARTICLE INFO

Article history:

Received 4 November 2014

Received in revised form

13 November 2014

Accepted 17 November 2014

Available online 28 February 2015

A 27-year-old young aboriginal woman from the Atayal tribe was found to have bilateral polycystic ovaries and irregular menstruation without a history of pregnancy. She complained of dysuria and a slowly growing perineal tumor was identified 1 week later and as a result she came to our urology department for help in May 2014. During the physical examination, a fleshy purple red tumor at the urethral meatus, about 2 cm at its largest diameter, was found. Urine analysis showed neither pyuria nor hematuria. Cystourethroscopy revealed that the urethral tumor was located at the distal urethra without intravesical abnormalities (Fig. 1A); we then performed total excision of the urethral tumor. A histological examination revealed one well-defined nodular submucosal tumor composed of elongated spindle-shaped smooth muscle fibers that were distributed as sweeping interlacing bundles (Fig. 1B and C). No nuclear atypia, tumor necrosis, or active mitosis was found. The overlying urothelium was flat and nondysplastic. Immunohistochemically, the spindle cells were diffusely positive for smooth muscle actin (Fig. 1D) and desmin, but negative for S-100 and CD117. Urethral leiomyoma was diagnosed. Her urethral mucosa was then reconstructed continuously. After the operation, the patient recovered uneventfully.

Leiomyomas may proliferate in any tissue where smooth muscle cells are present and have been found to manifest in various unusual locations including the ovaries, vulva, bladder, and urethra. Although leiomyomas are the most common benign soft tissue tumor in the lower urinary tract, reported cases are rare and there have been approximately 40 such cases associated with the urethra, with more women than men being affected [1–5]. The true incidence rate is unknown. Patients with urethral leiomyomas often present with a urinary tract infection, the presence of a mass, dyspareunia, obstructive urinary retention, or irritative lower urinary tract symptoms. The clinical differential diagnoses of a urethral tumor include urethral polyp, urethral diverticulum, urethral prolapse, Gartner's duct cyst, caruncle, inverted papilloma, leiomyoma, carcinoma, lymphoma, and metastatic tumor. Pathologically, the urethral leiomyoma must be differentiated from other spindle cell tumors, especially leiomyosarcoma, which shows marked pleomorphism, increased cellularity, and frequent mitoses. Although extragastrointestinal stromal tumors and neurogenic tumors are extremely rare in lower urinary tract, we did include them within the differential diagnoses; immunohistochemically, these tumors should express CD117 or S-100, respectively.

The definite etiology of urethral leiomyoma is unknown, but the fact that they are known to undergo enlargement during pregnancy and shrinkage after delivery implies the presence of a possible hormone effect [6,7]. Surgical excision is the curative treatment. On reviewing the literature, most cases seem to have a benign post-operative course and in total only two recurrences of the tumor have been reported [7]. The urethral leiomyoma affecting our patient was located in the distal urethra, which is an uncommon site for this tumor. Although the tumor has a benign biological behavior, it is difficult to diagnose clinically and a pathological examination is an essential part of the diagnosis. Complete excision is adequate treatment.

Conflicts of interest: none.

* Corresponding author. Department of Pathology, Chia-Yi Christian Hospital, 539, Zhongxiao Road, Chiayi, Taiwan. Tel: +886-5-2765041x7521; fax: +886-5-278-1961. E-mail address: hmarkc@gmail.com (C.-C. Chen).

<http://dx.doi.org/10.1016/j.tcmj.2014.11.004>

1016-3190/Copyright © 2015, Buddhist Compassion Relief Tzu Chi Foundation. Published by Elsevier Taiwan LLC. All rights reserved.

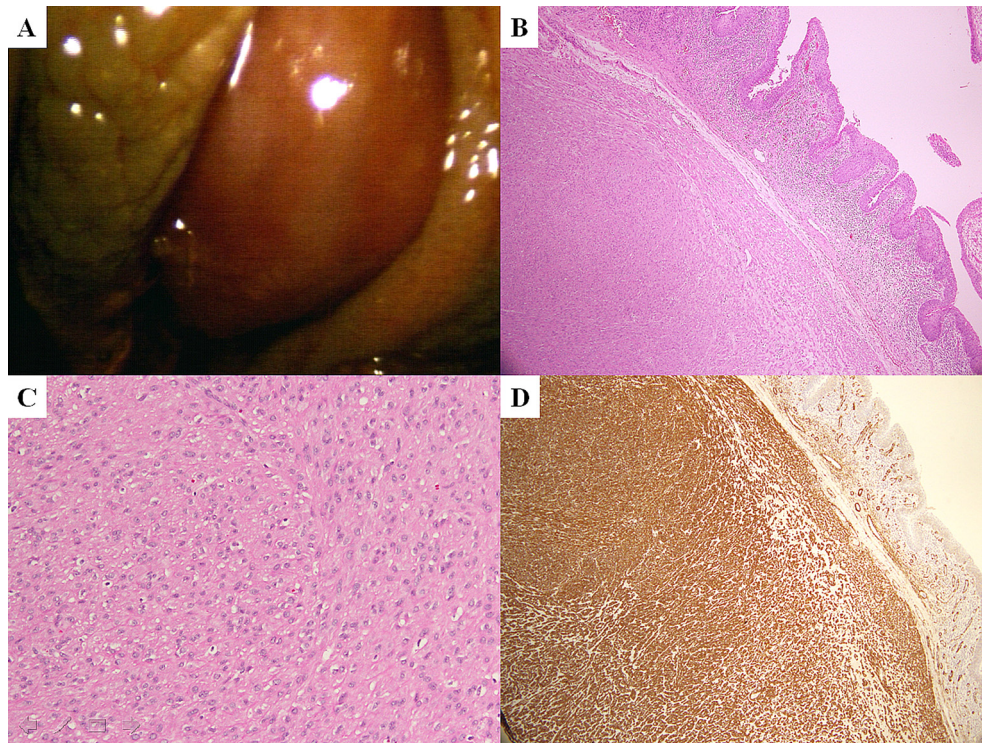


Fig. 1. (A) A fleshy purple red tumor is found to occupy the urethral meatus. (B) Microscopically, there is a well-circumscribed nodular mass in the subepithelial region with overlying nondysplastic urothelium and dense lymphocyte infiltration (hematoxylin and eosin $\times 40$). (C) The tumor is composed of aggregates of elongated spindle-shaped cells that are arranged as sweeping interlacing bundles (hematoxylin and eosin $\times 200$). (D) No nuclear atypia, tumor necrosis, or active mitosis is found. Immunohistochemically, the spindle cells were diffusely positive for smooth muscle actin ($\times 40$) and desmin, but negative for S-100 and CD117.

Further reading

- [1] Shield DE, Weiss RM. Leiomyoma of the female urethra. *J Urol* 1973;109:430–1.
- [2] Ozel B, Ballard C. Urethral and paraurethral leiomyomas in the female patient. *Int Urogynecol J Pelvic Floor Dysfunct* 2006;17:93–5.
- [3] Diószeghy G, Kiss A, Kondás J. Leiomyoma of the female urethra. *Int Urol Nephrol* 1998;30:603–7.
- [4] Lee MC, Lee SD, Kuo HT, Huang TW. Obstructive leiomyoma of the female urethra: report of a case. *J Urol* 1995;153:420–1.
- [5] Chong KM, Chuang J, Tsai YL, Hwang JL. A rapidly growing paraurethral myoma with profuse bleeding from a mucosal vessel: report of a case. *Gynecol Obstet Invest* 2006;61:87–9.
- [6] Fry M, Wheeler Jr JS, Mata JA, Culkun DJ, St Martin E, Venable DD. Leiomyoma of the female urethra. *J Urol* 1988;140:613–4.
- [7] Shen YH, Yang K. Recurrent huge leiomyoma of the urethra in a female patient: a case report. *Oncol Lett* 2014;7:1933–5.