Case Report

Female urethral cavernous hemangioma – An unusual cause of hematuria: A rare case report

Shivraj N. Kanthikar, Dhiraj B. Nikumbh, Nandkumar V. Dravid, Chakor M. Rokade

Department of Pathology, ACPM Medical College and Hospital, Dhule, Maharashtra, India

ABSTRACT

Hemangiomas are benign vascular lesion, most commonly seen in liver and skin whereas rarely found in genitourinary system. Urethral hemangiomas are mostly found in males. To the best of our knowledge, in females only handful of case reports has been described in the literature. We report a case of urethral hemangioma in a 28-year-old female presented with history of intermittent hematuria. Cystourethroscopy examination revealed vascular mass of 2 cm × 2 cm at anterior urethral meatus. Surgical excision of mass with fulguration of base with diathermy was performed under general anesthesia. Final diagnosis on histopathology was given as cavernous type of urethral hemangioma. In spite of its benign nature, these lesions have a tendency to recur. In more extensive lesions or recurrence, open exploration with resection of involved tissue is always needed. Treatment of hemangiomas depends on size and site of the lesion and follow-up is needed to avoid recurrence. Histomorphological diagnosis of the lesion is always warranted in view of different treatment modalities.

Key words: Hemangioma, hematuria, urethra

INTRODUCTION

Bleeding per urethra is a common urological symptom which needs urgent evaluation to exclude malignant etiology. Hemangiomas are most commonly found in the liver and skin, and rarely reported in the prostate, bladder, ureter and perineum. Urethral hemangiomas are exceedingly rare in females. They presented as an erythematous, inflamed mass surrounding the urethral meatus.[1] Urethral hemangiomas are seen predominantly in males with handful of case reports was described in females.[2] It has been suggested that they originate from unipotent angioblastic cells which fails to develop into normal blood vessel. The most common histopathological type is cavernous hemangioma.[3] The cardinal sign is presence of bleeding, means either gross or microscopic hematuria or urethrorrhagia.[4]

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Urethral hemangiomas are rare, and only 20 cases have been reported in the literature.[1] We present a case report of cavernous hemangioma of urethra in young adult female in view of its rarity.

CASE REPORT

A 28-year-old female came to Surgical Department of our hospital with the chief complaint of intermittent bleeding during urination since 1 month. Cystourethroscopy revealed a small brownish colored nodular growth of 2 cm × 2 cm at anterior meatus of urethra. Hematological investigation revealed mild anemia with hemoglobin level of 9.8 g%. Iron studies and coagulation profile were within normal limits. Ultrasound sonography abdomen-pelvis showed no significant pathology. After clinical and thorough investigations, excision of the urethral nodular mass was undertaken under general anesthesia. Surgical excision along with fulguration of base with diathermy was done and specimen sent for histopathological examination.

Grossly

Received brownish colored nodular mass of 2 cm × 2 cm, well circumscribed, soft and spongy. On cutting open, blood oozes out.

Address for correspondence: Dr. Shivraj Nagnath Kanthikar, Department of Pathology, ACPM Medical College and Hospital, Sakri Road, Dhule - 424 001, Maharashtra, India. E-mail: skanthikar@gmail.com

Histopathology

Sections showed tissue bits lined by transitional epithelium underneath showed a tumor. Tumor composed of variably-sized blood-filled vascular spaces lined by flattened endothelial cells separated by thin fibrous septae [Figure 1]. Focal areas show papillary endothelial hyperplasia. Active thrombosis was not seen in vessels. The final histopathological diagnosis given was cavernous hemangioma of the urethra. After surgical intervention, the patient is on regular follow-up with no complaints.

DISCUSSION

Urethral hemangiomas are extremely rare condition and occur at any age group. Most of the cases are encountered in males and only handful of case reports has been reported in females.[5] Presentation of urethral hemangiomas depends on the site and size of the lesion. Hemangiomas of the anterior urethra may present as urethral bleeding and lesions located in the proximal urethra usually present as hematuria, urinary retention with blood clots.[5]

Hemangiomas are believed to be congenital in origin, arising from the embryonic rest of the unipotent angioblastic cells that fail to develop into normal blood vessels. Others have implicated a theory of local varicosity and chronic irritation.[2] It may coexist with external hemangiomas and congenital disorders like Sturge-Weber or Klippel-Trenaunay syndrome.[6] Large lesions may present with obstructive urinary symptoms or protrude through the urethral meatus.[7]

Urethrocystoscopy is an excellent diagnostic method that supports in identification of the characteristics, friability, size, location and number of hemangiomas.It also assists in planning of possible therapeutic and surgical options in different cases.[3,8]

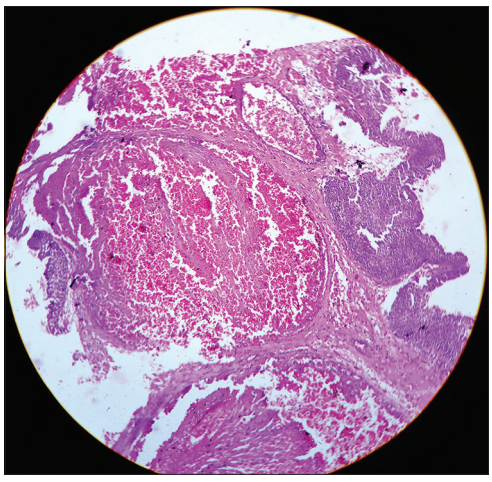

Figure 1: Light microscopy showing transitional epithelium of urethra underneath blood filled cavernous vascular spaces with flattened endothelial cells (H and E, ×100)

Regarding differential diagnosis, fibro-epithelial polyps, foreign body granulomas, leiomyomas, periurthral abscess, urethral warts, and malignant tumours such as adenocarcinoma, transitional cell tumors, sarcomas, melanomas and squamous cell carcinomas should be considered.[4,8] Histopathologically we can easily differentiates these lesions.

Immunohistochemistry (IHC) has limited role in hemangiomas due to its histomorphodiagnosis. IHC has role in the diagnosis of borderline vascular tumors as epithelioid hemangiomas and malignant conditions like angiosarcoma due to spindle cell predominance. The IHC for desmin, CD-34, CD-68, S-100, epithelial membrane antigen, chromogranin, and cytokeratin were negative but positive for endothelial cell markers CD-31 and Factor VIII in such tumors.[9] Different treatment modalities have been described in urethral hemangiomas, such as transurethral resection and fulguration of base of tumor, laser ablation, cryotherapy, selective embolization, steroids, local radiotherapy and open surgical excision.[4,5,7,10] Symptomatic and large lesions require complete excision, along with fulguration of the base either by diathermy or laser.[11]

In our case, lesion was present at anterior meatus of urethra; surgical wide excision along with fulguration of the base with diathermy was done. Earlier studies revealed that if complete resection of the mass was done with available good surgical techniques, chances of recurrences are less and with excellent prognosis.

CONCLUSION

Urethral hemangiomas are benign in nature, with tendency to recurrence. Regular follow-up of patient is always required. Urethral hemangiomas are very rare in females. It is essential to choose good diagnostic, accurate histopathological and easily available treatment modalities especially for younger patients to prevent their recurrences. Histopathological study is always warranted in view of treatment differs. IHC has limited role in hemangiomas, whereas it's helpful in borderline and malignant lesions of blood vessels.

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