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Case Report

Fibroepithelial stromal polyp of bladder-a mimicker of sarcoma or angiomyxoma at uncommon location

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Abstract: The fibroepithelial stromal polyp is a benign polypoid proliferation of the stroma with overlying epithelium. Because the lesion contains atypical stromal cells, sometimes it can be overdiagnosed as sarcoma or with myxoid stroma, it can be misdiagnosed as angiomyxoma. The reported locations are mainly in the lower female genital tract, urethra, and rarely extragenital sites, such as the breast, and are exceptionally rare in the bladder. We encountered a 65-year-old man who presented with two small velvety, erythematous patches on the posterior bladder wall. The final diagnosis is a fibroepithelial stromal polyp of the bladder. Familiarity with this lesion will prevent overinterpretation of this benign lesion as a malignancy.

Keywords: Bladder, fibroepithelial stromal polyp, male

Introduction

The fibroepithelial stromal polyp is a benign polypoid proliferation of the stroma with overlying benign, usually squamous epithelium without acanthosis or papillary and unrelated to HPV infection. Diagnosing this lesion is mainly based on morphology. Clinically it is asymptomatic and can cause discomfort, bleeding, discharge et al. It is a benign lesion and can be cured by excision or regress after delivery. But it might recur if not completely exercised [1]. It was reported initially by Norris and Taylor in 1966 and is an uncommon but generally recognized lesion of the female genital tract [2]. Because the lesion contains atypical stromal cells, it can be mistaken for a sarcoma, mainly if it shows hypercellularity, pleomorphism, and atypical mitoses or angiomyxoma if with a myxoid background. The lesion can occur at any age but might be hormonally related and affects reproductive-age women. The most common sites are the lower female genital tract, urethra, and rarely extragenital sites, such as the nipple [3-5]. There have been rare cases in the male urinary tract and genital area [6-8], but it is not well-known in these areas. Other unusual locations, such as the anus and nasal cavity, have also been reported [9, 10]. Because it is not well known in the bladder, overinterpretation as a sarcoma or angiomyxoma is possible.

Case report

The patient was a 65-year-old male with a history of lower urinary tract symptoms due to prostatic hypertrophy on maximal medical therapy. He also had been experiencing hematuria. On office cystoscopy, he was found to have a 3 cm prostate mildly obstructing the urethra and two small velvety, erythematous patches on the posterior bladder wall. The lesion was resected without difficulty. During the operation, the lesion is described as a “small velvety patch on right posterior bladder wall with an erythematous area alongside it”. Two biopsies were taken of the right posterior bladder wall. The pathology laboratory received two (2) pink/tan soft tissue fragments measuring 0.1 and 0.2 cm in the most significant dimension. On the histology, the overlying urothelium was benign. The stroma was collagenous and hypocellular, containing scattered atypical and multinucleated cells, similar to the fibroepithelial stromal polyp well recognized in the female lower genital tract (**Figure 1**). The immunohistochemistry showed positive stains for Desmin, ER, PR (**Figure 2**)

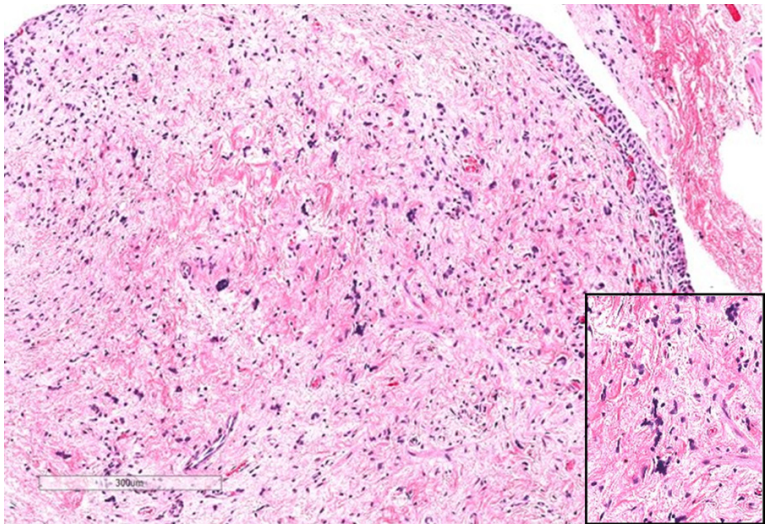

Figure 1. Higher magnification (10×) Bladder polyp containing a hypocellular stroma with atypical stromal cells (inset), similar to the fibroepithelial stromal polyp of the female genital tract.

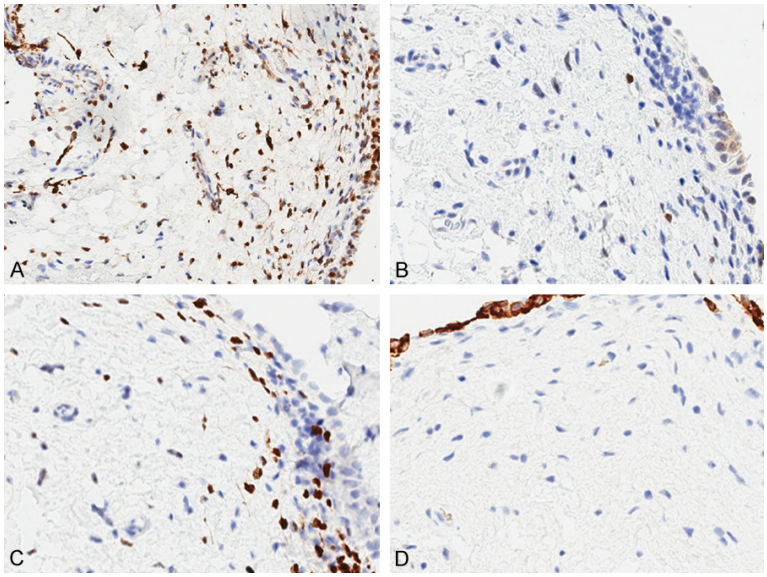

Figure 2. Immunohistochemistry stain of high magnification (20×). A. Desmin stains positive of epithelium and stromal cell. B. ER stains stromal cells and focal epithelium. C. PR stains stromal cells. D. CK5/6 stains only epithelium cells.

and vimentin, negative for cytokeratin (**Figure 2**), SMA, HMGA2, S100, myogenin (not shown). The patient was treated with excision and has been a follow-up for more than two years showing no sign of recurrence.

Discussion

Fibroepithelial stromal polyp, formerly known as pseudo sarcoma botryoides or sarcoma botryoides-like lesion [2]. It is a rare benign stromal polyp with polypoid growth of atypical fibroblasts in the myxoid stroma. The surface is covered by epithelium. Atypical fibroblasts with bizarrelooking can be overinterpreted as sarcoma [11]. The myxoid stroma of this entity can be misinterpreted as angiomyofibroblastoma, myofibroblastoma angiomyxoma. Angiomyofibroblastoma is a well-circumscribed neoplasm composed of epithelioid fibroblastic cells and a richly vascularized myxocollagenous stroma. Sometimes it coexists with mast cells and adipocytes in the virginal area and male glans penis [12, 13]. The entity usually expresses ER, PR, desmin, and smooth muscle actin by immunohistochemistry [14]. A more aggressive lesion with more local recurrence is deep angiomyxoma. Angiomyxoma is a rich vascularized neoplasm with low cell density, positive for ER and PR, desmin, and might be positive for CD34. But they grow in deep vulvovaginal, pelvic, and perineal tissues and are large (often 10 cm or more) and bear a rearrangements in HMGA2 [15]. The lesion is more spindly and aggressive with infiltrative growth, which is not seen in any other stromal tumors in the above differential diagnosis. Myofibroblastoma is another benign stromal tumor with short spindle cell in bundle or fascicles [16] with a similar immunohistochemistry character as angiomyxoma but characterized by loss of Rb expression. The morphologic and immunohistochemical overlap between cellular angiofibroma, mammary type myofibroblastoma, and angiomyofibroblastoma, fibroepithelial stroma polyp is not critical. The only essential lesion is sarcoma and angiomyxoma, which must be ruled out.

Since fibroepithelial stroma polyp is a rare benign stromal polyp, it is easily underrecognized or overinterpreted. This is particularly true when found in less expected locations such as the urinary tract in our case [7, 8, 17]. The pathogenesis needs to be better understood. Some researchers think it is a reactive hyperplastic process because the margin of this lesion is not well defined. The lesion in the female genital tract has been suggested to arise from the subepithelial stromal cell [18]. Other evidence supporting a reactive process is a series of seven cases of the glans penis. The lesions were strongly associated with condom catheter use [19]. Others have reported the lesion in a very young patient and suggest a congenital process [20]. Another theory in the female genital tract is that the lesions are hormonally related. The hypothesis is based on the stromal cells expressing estrogen and progesterone receptors and responding to hormones which is consistent with our result in **Figure 2** [21, 22]. Additional support for the lesion being hormonally related is the frequency in pregnancy with the disappearance after delivery [23]. The lesion in the bladder suggests the pathogenesis is related to inflammation [6]. The clinical appearance of the lesion in our patient was consistent with chronic inflammation or carcinoma in situ. Chronic inflammation in the bladder is often related to recurrent urinary infections and can be associated with cystitis cystica. Carcinoma in situ has a typical velvety, flat appearance and is managed by biopsy, fulguration and post cystoscopy intravesical BCG installation. Fibroepithelial polyps are usually managed by endoscopic resection and surveillance cystoscopy.

Conclusion

The benign fibroepithelial stromal polyp is an uncommon lesion usually seen in the lower genital tract of reproductive-aged women. The presence in the bladder of a male is scarce. Recognition of the lesion will prevent overdiagnosis as a malignancy. Particularly in hypercellularity, cytological atypia increased mitotic figures, and atypical mitoses, the lesion may be misdiagnosed as a sarcoma. Aggressive angiomyxoma may also be considered if the stroma is edematous and myxoid. Diagnosis relies predominantly on histopathology. Immunostaining shows variable staining for desmin, estrogen receptors, progesterone receptors et al. Treatment is excision and will cure the disease.

Disclosure of conflict of interest

None.

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