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Case Report

A Rare Case of Hemangioendothelioma of Urinary Bladder

Abstract

Hemangioendothelioma is a vascular tumor of endothelial nature of intermediate grade. It most commonly arises from soft tissue of upper and lower extremities. We report a rare case of epithelioid hemangioendothelioma of the urinary bladder. Histologically, it was a vascular tumor formed by smaller capillaries lined by plump epithelioid cells having eosinophilic cytoplasm. Diagnosis was confirmed by immunohistochemistry, as the tumor cells were positive for CD34 and smooth muscle actin.

Keywords: *CD34, epithelioid cells, urinary bladder, vascular tumor*

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Introduction

Hemangioendothelioma is a vascular tumor of endothelial nature of intermediate grade. Tumors included in this group have the ability to recur locally and have some ability to metastasize but at a far reduced level compared to angiosarcoma.[1] Vascular neoplasms are uncommon in urinary bladder, especially there are only two reported cases of epithelioid hemangioendothelioma (EHE) of urinary bladder after extensive search.[2,3] We describe a rare case of EHE of the urinary bladder, in which was confirmed by histopathology followed by immunohistochemistry (IHC).

Case Report

A 48-year-old male presented with a history of episodic hematuria for the past 8 years. He was nondiabetic, normotensive, and euthyroid. No definite diagnosis and treatment was done for the past 8 years. On clinical examination, no abnormality was detected. Routine hematological and biochemical parameters were within the normal limit. This patient's renal parameters (blood urea and serum creatinine) were within normal limits at presentation. He had no history of smoking, medicinal exposure, occupational exposure, or any chronic infection of bladder, which are considered as significant risk factors for bladder tumor. His urine cytology on multiple occasions revealed markedly high red blood cell (RBC) count and occasional blood clot. There was no evidence of any malignant cells.

He was evaluated with ultrasound (USG) of kidney, ureter, and bladder (KUB) region and computed tomography (CT) scan of the abdomen. In USG, presence of a mass lesion was suggested at the left side of pelvis in the line of ureter. CT scan of KUB revealed a 2.2 cm × 2 cm enhancing calcified nodule attached to urinary bladder dome [Figure 1].

The patient was posted for cystoscopy, and during operation, solitary >2 cm solid raspberry-like highly vascular tumor was found at the dome of the bladder. A transurethral complete resection of the tumor was done by cystoscopy with biopsies from deeper tissue. Rest of the bladder was devoid of any abnormality.

Histopathological examination was carried out on formalin-fixed, paraffin-embedded tissue sections prepared from biopsy specimen. On histopathological examination, sections studied showed a vascular tumor formed by smaller capillaries lined by plump epithelioid cells having eosinophilic cytoplasm. Some of the individual cells showed a vascular lumen containing RBCs. Lining transitional epithelium was unremarkable. No evidence of malignancy was detected [Figure 2]. Immunostaining was performed by applying the streptavidin–biotin–peroxidase conjugate method in an automated stainer (Ventana BenchMark XT, Ventana

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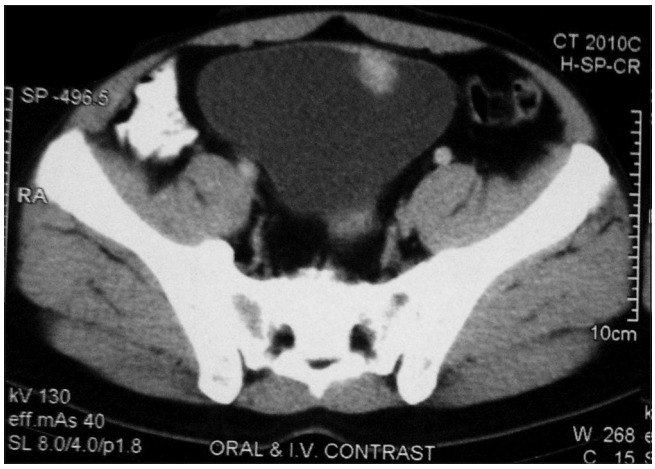

Figure 1: Contrast-enhanced computed tomography of the bladder showing an enhancing mass lesion involving the dome of the urinary bladder

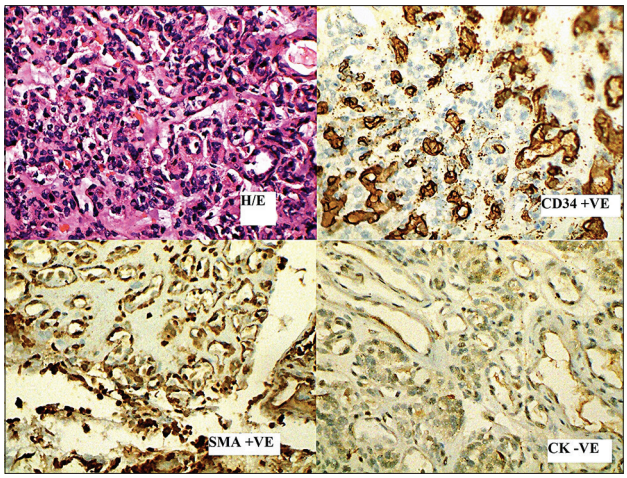

Figure 2: Photograph shows routine section of proliferation of plump endothelial cells (H and E, ×400) and positivity for immunostain CD34, smooth muscle actin, and negative immunostain CK (×400)

Medical Systems, Rosche) utilizing the manufacturer's protocol with prediluted ready-to-use antibodies (Dako, Glostrup, Denmark). On IHC, tumor cells were positive for CD34 and smooth muscle actin but negative for CK, EMA, CD-68, S-100, vimentin, and CD56 [Figure 2]. These features are consistent with EHE, ruling out possibility of other soft tissue and epithelial neoplasms. At 6-month follow-up, the patient was asymptomatic, and on USG, there was no recurrence in the bladder.

Discussion

EHE is a vascular tumor of adults which is characterized by an "epithelioid" or "histiocytoid" endothelial cell. They involve both superficial and deep soft tissue. They are characterized by rounded to slightly spindled endothelial cells, having eosinophilic cytoplasm with vacuolization with rounded nuclei. The behavior of EHE is intermediate between hemangiomas and conventional (high grade) angiosarcomas.[4-6] While EHE involves commonly the soft tissue of the extremities, visceral involvement may also occur.[7] The differential diagnoses that were considered based on the histopathological characteristic are granular cell tumor, inflammatory myofibroblastic tumor, EHE, and epithelioid leiomyosarcoma.

As there were no predisposing factors for transitional cell carcinoma in our case and the tumor has not progressed significantly since the past 8 years without any medical intervention, it suggests a possibility of benign tumor of urinary bladder.

Characteristic histopathologic features of EHE include plump epithelioid cells with eosinophilic hyaline cytoplasm, angiocentric pattern having cytoplasmic vacuoles representing primitive vascular lumina. The cells are arranged in cords and papillary tufts of plump cells within lymphovascular spaces.[6] Presence of RBCs in cytoplasmic vacuoles or primitive tumor-cell lined channels are highly suggestive of EHE and positive IHC for vascular markers and negativity to epithelial differentiation and other mesenchymal marker confirms the diagnosis.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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