CASE REPORT

Intravascular papillary endothelial hyperplasia of the bladder: Case report and review of the literature

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ABSTRACT

Intravascular papillary endothelial hyperplasia (IPEH), also known as Masson's tumor, is a benign unusual vascular lesion thought to arise from an organizing thrombus. Histologically, IPEH is characterized by papillary fronds lined by proliferating endothelium that may mimic angiosarcoma, and therefore the correct diagnosis may prevent unnecessary radical procedures. Involvement of the bladder is extremely rare, with only three cases reported in the literature. We report a case of IPEH arising in the bladder of a patient with history of prostate cancer treated with radiotherapy.

Key words: Bladder, intravascular papillary endothelial hyperplasia, Masson's tumor

INTRODUCTION

We present to the best of our knowledge the fourth reported case of intravascular papillary endothelial hyperplasia (IPEH) localized to the bladder. IPEH, also known as "Masson's vegetant hemangioendothelioma" or "Masson's hemangioma", is a rare benign incompletely understood process thought to be unusual reactive proliferation of endothelial cells arising from an organizing thrombus.[1] The skin and subcutaneous soft tissue are the most common sites; however, IPEH can arise virtually in any area of the body.[2] Generally, it has a slow growth rate.[1] Histopathologicaly, IPEH may mimic angiosarcoma, and the failure in distinguishing these entities may result in unnecessary radical surgery or irradiation.[3]

CASE REPORT

A 77-year-old African American male with locally advanced, poorly differentiated stage T2c, Gleason score 8 prostatic adenocarcinoma treated with hyperfractionated photon irradiation and neo-adjuvant hormonal therapy in 1993 presented for evaluation of intermittent painless gross hematuria noted 10 years after radiation therapy. He was started on hormonal therapy for biochemical prostate cancer relapse. His medical history was significant for atrial fibrillation for which he took Warfarin and Aspirin.

The patient underwent rigid cystoscopy which revealed a bladder mucosa with changes consistent with postradiation therapy, bleeding vessel near bladder neck, and two small bladder masses. These masses were approximately 1.0 cm in size and appeared to have some solid consistency. Clot evacuation and transuretheral resection of the tumors were performed. Microscopic examination of both lesions revealed submucosal dilated vessels with intraluminal anastomosing papillary fronds of fibrinous core lined by endothelial cells that showed occasionally mild to moderate pleomorphism with plump nuclei, occasional prominent nucleoli, and nuclear hyperchromasia without increased nuclear/cytoplasmic ratio. No mitotic activity or necrosis was identified. The muscularis propria was present and not involved [Figures 1-3]. Additionally, acute and chronic inflammation was noticed without reactive urothelial changes. These histological features were consistent with IPEH.

The patient has been followed for 1 year with no evidence of recurrence.

DISCUSSION AND LITERATURE REVIEW

The benign lesion that currently is known under the term IPEH was first described by Masson in 1923 in infected hemorrhoids of a 68-year-old man. Masson regarded the

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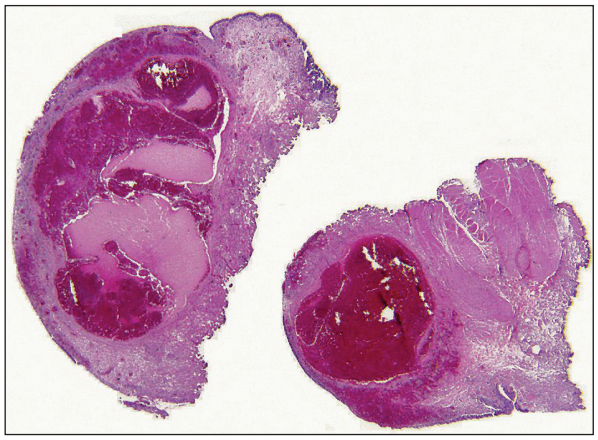

Figure 1: Low power view showing both lesions, H and E 20×

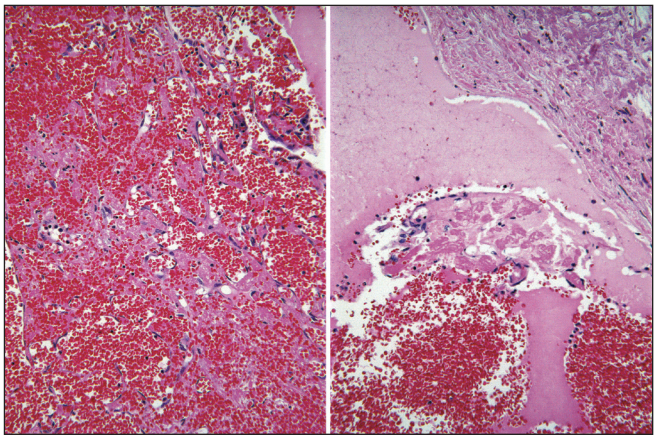

Figure 2: Composite high-power field, H and E 200×. Note the anastomosing papillary fronds.

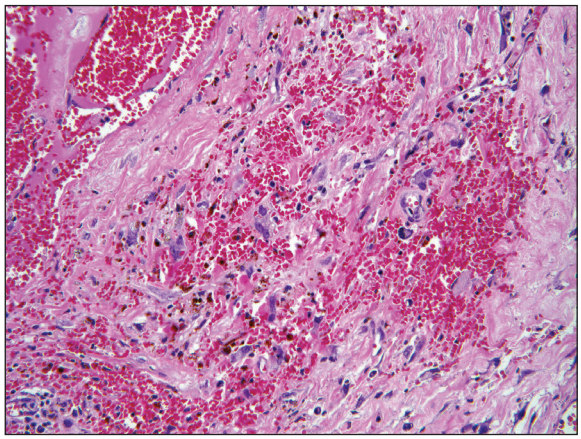

Figure 3: High-power field, H and E 400× showing mild to moderate nuclear atypia.

lesion as true benign neoplasm that exhibits degenerative changes such as necrosis and thrombosis as it outgrows its blood supply and termed it as "Hémagioendothéliome végétant intra-vasculaire."[2,4-6] Henschen thought that it was a primary endothelial proliferation arising in response to inflammation and stasis in a vascular bed.[2] A plethora of terms had been used for this entity until 1976 when it was first coined as "Intravascular Papillary Endothelial Hyperplasia" by Clearkin and Enzinger who suggested that the thrombosis takes place before the papillary proliferation and that the thrombotic material provides a matrix for its development.[1] In their opinion, slowing of blood flow, stasis, and thrombosis appears to be prerequisites for the development of the lesion.[1] In 1990, this suggestion was supported by Albert and Kahn who also thought that IPEH is a peculiar form of an organizing thrombus by finding similar progression of the immunophenotype of the endothelial cells in IPEH and organizing thrombus using immunohistochemical studies.[7] Both are primarily positive for ferritin, and histiocytic markers, eventually gain vimentin positivity, and finally with maturation, demonstrate positivity only for factor VIII-related antigen.[7] IPEH has been most commonly detected in the veins of deep dermis or subcutis of the head, neck, fingers, and trunk, but can occur in any area of the body.[2] Multifocal IPEH in the retroperitoneum and spine has also been described.[8]

A previous series by Tavora *et al.*[9] reports the first three known cases of IPEH involving the urinary bladder. The patients in these cases were older than our patient, aged 81–83, with a markedly shorter times elapsing between therapy and presentation (range 2–17 months). They presented with a similar history of radiation therapy for tumors of endometrium, prostate, and bladder. It seems that radiation effect might predispose to the development of IPEH, further supporting the concept of the reactive nature of this lesion. On cystoscopy, the tumors presented as raised lesions and were all small in size (<1.0 cm) as it was in our patient. Histologically, similar to our case, these lesions were seen in the submucosa and had mild atypia without mitosis, necrosis, or solid growth pattern. Fibrin deposition was noted in our case as well as in one of the previous cases.

With regard to the differential diagnosis it is very important to discriminate IPEH from angiosarcoma because both could be preceded by history of radiation.[10] IPEH can mimic angiosarcoma especially when there is passive extension into the surrounding tissue after vessel rupture. However, the intravascular location of most of the lesion along with the reactive changes in the vessel wall is suggestive of rupture.[2] In addition, angiosarcoma exhibits significant mitotic activity, marked pleomorphism, solid sheet formation, and necrosis.[3] In the bladder, the size and the depth of invasion also could be used to differentiate between the two entities.[9]

The rare extravascular papillary endothelial hyperplasia that has been described in other locations arising from

organizing hematomas tends to be well circumscribed and lacks the irregular anastomosing channels and the malignant features of angiosarcoma.[2,10]

CONCLUSION

We are presenting to the best of our knowledge the fourth case of bladder IPEH in the literature. IPEH in our case differs from the previous three cases by presenting as two separate bladder masses and occurring after longer period of time following radiation therapy. It is important to consider IPEH in the differential diagnosis of vascular lesions of the bladder in order to prevent unnecessary surgical or radiation intervention.

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