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Case report

Solitary fibrous tumor/hemangiopericytoma of the penis

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ABSTRACT

Solitary fibrous tumor/hemangiopericytoma (SFT-HMP) is a rare spindle-cell mesenchymal tumor, thought to be of myofibroblastic origin. Penile SFT-HMP was mentioned in the literature in two separate case reports which were published in 2015 and 2017. We present the first case of SFT-HMP which is localized on corpus cavernosum of the penis. A 55-year-old man presented to our clinic with a small and gradually growing lesion on his penis for the past year, which recently caused difficulties during sexual intercourse. On physical examination; a well-shaped, nodular non-fluctuant, solid, painless mass, measuring 4x4 cm was palpated. Magnetic resonance imaging showed 5x5 cm mass located on the right corpus cavernosa. Under spinal anesthesia, surgical excision was performed. Pathologically, the tumor had an irregular architecture patterns and was characterized by hypercellular areas separated by thin-walled, branching vessels, lined with a single layer of flattened endothelial cells. SFT-HMP rarely occurs in genital tract and penile presentation is among the rarest. It should be classified and considered under penile masses, especially if the mass is well circumscribed, painless and slowly growing in nature. After differential diagnosis, surgical excision is mandatory.

Keywords: Hemangiopericytoma; penile mass; solitary fibrous tumor.

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Introduction

Solitary fibrous tumor/hemangiopericytoma (SFT-HMP) is a rare spindle-cell mesenchymal tumor, thought to be of myofibroblastic origin.[1] It has an intermediate behavioral pattern in other words it rarely metastasizes.[2] This tumor was described by Muray in 1942 as a pleural based lesion.[3] Thirty percent of SFT-HMP occurs in sites other than pleura (extrapleural). Deep muscles of proximal extremity, head, neck and retroperitoneum are common locations for extrapleural SFT.[4] Penile SFT-HMP was mentioned in the literature in two separate case reports which were published in 2015 and 2017.[5,6] We present the first case of SFT-HMP which was localized on corpus cavernosum of the penis.

Case presentation

A 55-year-old man presented to our clinic, complaining of a small gradually growing lesion on his penis for the past year, which recently caused difficulties during sexual intercourse. He did not have any problems related to voiding or erection. Patients' past medical history was unremarkable. On physical examination; a well-shaped, nodular non-fluctuant, solid, painless mass, measuring 4x4 cm was palpated. The mass was located on the root of the penis which was in association with the right corpus cavernosum. No palpable lymph nodes were detected. No abnormalities were detected on laboratory analysis. Ultrasound scan revealed a hypervascular, solid mass measuring 4x4 cm, which contained both hyperechogen and hypoechogen areas. Magnetic resonance imaging showed that the mass was 5x5 cm in size and located on the right corpus cavernosum (Figure 1a). The mass was heterogeneous and hyperintense on T2- weighted images and heterogeneous and isointense on T1- weighted images. The mass had marked homogeneous contrast enhancement (Figure 1b). No remark-

able lymph nodes or metastasis were noted. Decision of surgical excision was made. Patients' written consent for the operation was obtained.

Under spinal anesthesia, surgical excision was initiated via an incision over the mass. During surgical exploration, it was observed that the mass had clear boundaries with an evident capsule. The mass was originated from superficial wall of right corpus cavernosum. The mass was not in close contact with the spermatic cord or the urethra. Blind and sharp dissections were performed around the mass. Mass was sharply excised from corpus cavernosun, with a thin layer of corporal body wall. Cavernosal defect measuring 2x1 cm was sutured with an absorbable suture material. Artificial erection was performed and no chordee was observed. A vacuum drainage tube was placed to the surgical site and removed on the first postoperative day. Patient was discharged on the first postoperative day, uneventfully.

Macroscopically, the tumor was well circumscribed, encapsulated and measuring 5x5x3.5 cm. On sliced sections; the mass was whitish and brown in color and it was frequently multinodular and firm in nature. Focal hemorrhagic areas were frequently observed and the mass had non-infiltrative margins. Microscopically, the tumor had irregular architecture patterns and was characterized by hypercellular areas separated by thin-walled, branching vessels, lined with a single layer of flattened endothelial cells (Figure 2). Tumor cells were ovoid and spindle shaped. Tumor cells contained limited eosinophilic cytoplasms, had indistinct borders and dispersed chromatin within vesicular nuclei. Mitoses were generally scarce and rarely exceeded 3 mitoses per 10 HPF. Immunohistochemical staining revealed CD34 positivity. However, CD34 staining was relatively focal. CD99 and bcl2 immunostainings revealed diffuse and strong positivity.

Postoperative period was uneventful, in terms of surgical site complications, and patients' overall health. Chordee or erectile dysfunction was not observed during postoperative follow-up. An abdominopelvic magnetic resonance imaging was performed one year after the surgery and no local recurrence or distant metastasis was detected.

Discussion

Since hemangiopericytoma was first described in 1942 and solitary fibrous tumor in 1931; differentiation and classification of these two tumor types have been controversial.[7] Microscopically, presence of dilated and branching vessels and gene fusion between NAB2-STAT6, which results in overexpression of STAT6 protein, are common findings in both tumor types.[8] In 2013, World Health Organization (WHO) banned using the term hemangiopericytoma and hemangiopericytomas were classified as a part of extrapleural solitary fibrous tumors.[9] Recently, in 2016, WHO introduced the term SFT-HMP to describe these two soft tissue tumors.[3] Neuropathologists still prefer the term hemangiopericytoma, in reference to meningeal tumors, to emphasize their aggressive behavior compared with other SFT.[10] We also prefer to use the term SFT-HMP.

Clinical presentation of patients depends on location of the tumor.[8,11] Jia et al.[3] reported a case series including 20 patients with spinal osseous SFT-HMP and demonstrated that localized pain with an average duration of 4.8 months was the most common complaint. Among his patients, 65% of the patients had symptoms due to spinal cord compression and 5% had a palpable mass. Fernandez et al.[12] demonstrated that almost all of his patients presented with nonspecific abdominal or back pain. One of his patients was incidentally diagnosed while being evaluated due to her irregular menstrual cycles. Abdominal and pelvic SFT-HMP tumors were diagnosed in 6 of his patients. Ronchi et al.[2] reported a case which was presented with urinary retention and constipation due to a huge SFT-HMP on prostate gland. In a review published by Tanaka et al.[13], 36% of their patients had voiding difficulties, 32% had hematuria, 18% had incidental imaging findings, and 14% presented with lower abdominal discomfort. Castellani et al.[5] reported a penile SFT-HMP case which was presented with a slow growing painless mass. Our patients' primary complaint was difficulty during sexual intercourse. No coital problems have been reported in the literature regarding female patients with a genital tract SFT-HMP.[8]

In the literature, there is no specific laboratory test available for the diagnosis of SFT-HMP. Myoteri et al.[4] suggested using glucose levels due to the fact that 10% of the patients have hypoglycemia due to increased production of Insulin like Growth Factor 2. Differentiation between SFT-HMP and other solid tumors is also not possible using radiological studies. Rosenkrantz et al.[14] described the appearance of SFT-HMP in computed tomographic imaging as homogeneous mass with well-circumscribed margins, and without any sign of invasion. Shanbhogue et al.[15] stated that imaging heterogeneities are the limitations for the differential diagnosis of SFT-HMP which can be listed as; variety of enhancement, necrosis and hemorrhage. Fernandez et al.[12] demonstrated that malign tumors had a higher SUDmax value than benign tumors (11.4 in malignant tumors, 2.9 in benign tumors).

Tumor sampling via needle biopsy is a useful tool for the differential diagnosis of SFT-HMP. Tanaka et al.[13] obtained needle biopsies of vesical, and periprostatic mass in two cases and pathological report showed spindle-cell shaped proliferation, CD34 expression, positive Bcl-2, which are all in favor of SFT-HMP. Perioperative excisional biopsy confirming SFT-HMP followed by complete tumor resection is a viable option especially in oral tumors. Biopsy of paratesticular and sper-

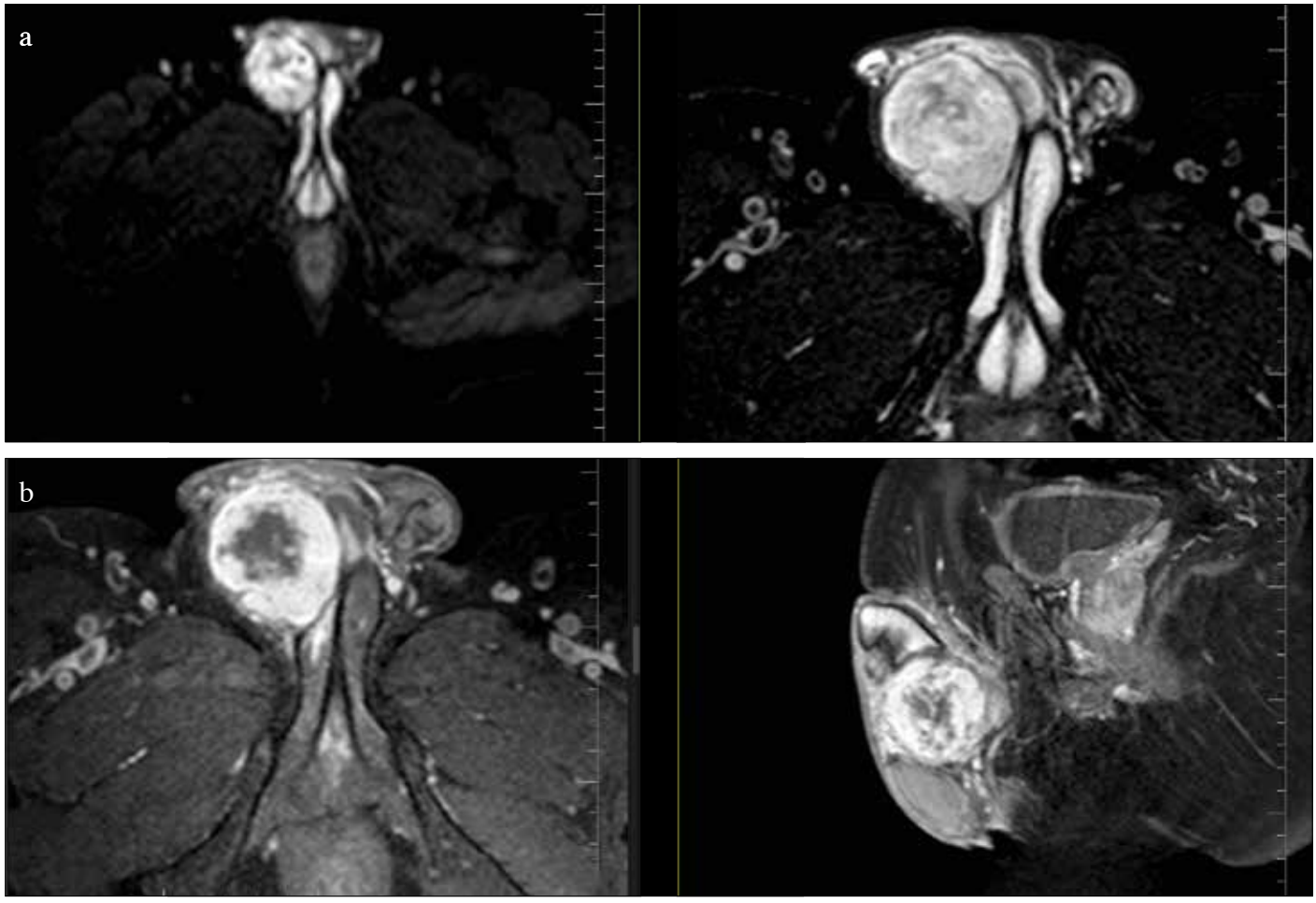

Figure 1. a, b. MRI image of the penile mass

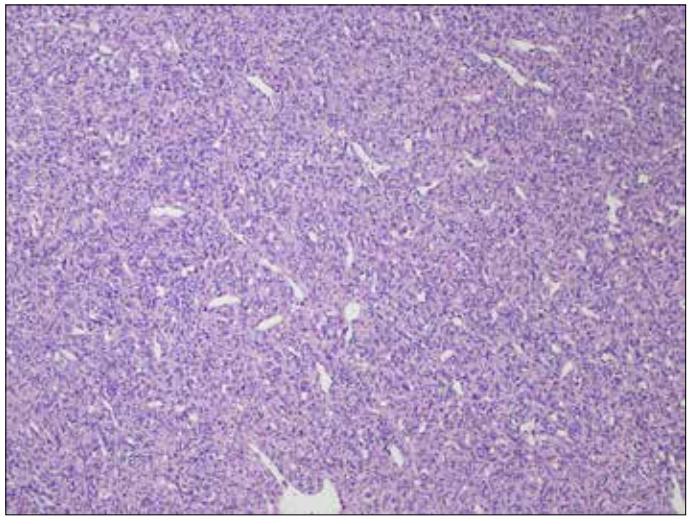

Figure 2. Microscopic appearance of the mass during pathologic evaluation

matic cord masses is not recommended due to the possibility of spreading the cancer cells. Non-surgical intervention was not a choice for our case, due to the complaints of the patient about his sexual life. That's why, we did not perform any kind of biopsy prior to surgery.

Authors suggest close follow up for at least five years for patients who had received a treatment for SFT-HMP in oral cavity, deep extremity, female genital tract, but a standard follow- up proposal is not available.[3,7-9] Individual follow-up strategies usually include radiological studies and physical examination. At postoperative first year, we performed an abdominopelvic MRI and no local recurrence was detected.

In conclusion, SFT-HMP rarely occurs in genital tract and penile presentation is among the rarest. It should be classified and considered under penile masses, especially if the mass is well circumscribed, painless and slowly growing in nature. After differential diagnosis, surgical excision is mandatory. Close follow up is recommended due to possibility of local or distant recurrences. Further studies with long-term results are needed for follow up strategies and adjuvant treatment regimens.

Informed Consent: Written informed consent was obtained from patients who participated in this case.

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