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Case Report
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Primary Leiomyosarcoma of Epididymis: One Case Report

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Abstract

Background: Leiomyosarcoma is a tumor that can develop in any organ that contains smooth muscles. Although leiomyosarcoma is common, epididymal localization of them is quite rare.

Case Report: A 79-year-old patient presented with mild pain in the right groin and scrotum for three years followed by a lump at the posterior aspect of the right testis. Ultrasonography and Magnetic Resonance Imaging (MRI) of the scrotum showed an irregular and heterogenous mass of 4 cm × 3 cm × 4 cm at the inferior aspect of right testis. High inguinal orchiectomy was performed under general anesthesia. The pathological diagnosis was a primary epididymal leiomyosarcoma.

Conclusion: Epididymal leiomyosarcoma is rare and difficult to diagnose pre-operatively. The final diagnosis of Leiomyosarcoma requires histologic examination. Resection must be extensive and complete. The effect of chemotherapy and radiation on the leiomyosarcoma of the epididymis remains unclear. Recurrence is common, so follow-up is necessary.

Keywords: Epididymal; Leiomyosarcoma; Diagnosis; Treatment

Background

Soft tissue sarcomas account for a relatively small proportion of systemic malignancies, common in the intestinal mucosa, retroperitoneum [1,2]. Leiomyosarcoma is a malignant mesenchymal tumor arising from the smooth muscle, the vascular smooth muscle, or the mucous muscle of the intestinal wall accounting for 5% to 10% of all soft tissue tumors [3,4]. The Leiomyosarcoma of peritesticular tissue were derived from the testicular tunica (48%), spermatic cord (48%), epididymis (2%), and dartos muscle and scrotal subcutaneoust issue (2%) [5]. Epididymal leiomyosarcoma is rare [6] and occurs in the smooth muscle surrounding the basement membrane of the epididymal duct [7]. We report a case of epididymal leiomyosarcoma.

Case Presentation

A 79-year-old patient presented with mild pain in the right groin and scrotum for three years followed by a lump at the posterior aspect of the right testis. No history of trauma, urinary tract infection, hematuria, dysuria, or surgery. The local examination revealed a hard mass about 5 cm × 3 cm × 4 cm fixed to the inferior aspect of right testis. Ultrasonography of the scrotum showed an irregular and heterogenous mass of 4 cm × 3 cm × 4 cm at the inferior aspect of right testis (Figure 1). Magnetic Resonance Imaging (MRI) showed an oval-like solid space occupying lesion about 3.5 cm × 3.5 cm × 4.0 cm (Figure 2). High inguinal orchiectomy was performed under general anesthesia. Gross pathological examination revealed a 4 cm × 3.5 cm × 3.5 cm solid tumor mass which the cut surface is grayish white with a crisp texture (Figure 3). On histopathology, the tumor was composed of pleomorphic spindle cells arranged in fascicles (Figure 4a) and the tumor cells are markedly heterogeneous, with pathological mitosis (Figure 4b), invading the albuginea testis and grade 1 (according to National Federation of French Cancer Centers and National Cancer Institute system). Immunohistochemistry showed tumor cells to be positive for Smooth Muscle Action (SMA), desmin (Des), h-Caldesmon, vimentin and EMA (Figure 5a, 5b) and negative for CD34, CD117, PLAP, a-inhibin, ki-67, DOG-1, Myogenin, MyoD, S100 and SOXIO. The pathological diagnosis was a primary epididymal leiomyosarcoma. After the operation, chest and abdominal Computed Tomography (CT) scans were performed, and tumor markers were detected. No abnormalities were found. He was not planned for any adjuvant therapy.

Discussion

We searched Pubmed with keyword: "Leiomyosarcoma" and "Epididymis" on November 11th, 2019

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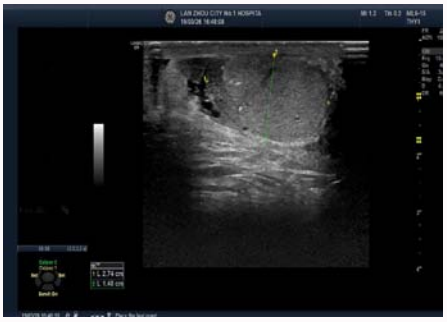

Figure 1: There was an irregular heterogeneous mass about 4 cm × 3 cm × 4 cm.

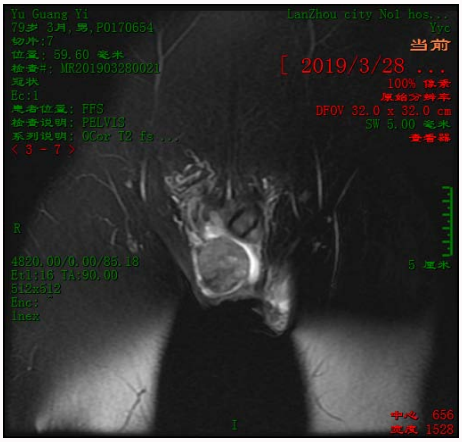

Figure 2: Oval-like solid space occupying lesion about 3.5 cm × 3.5 cm × 4.0 cm.

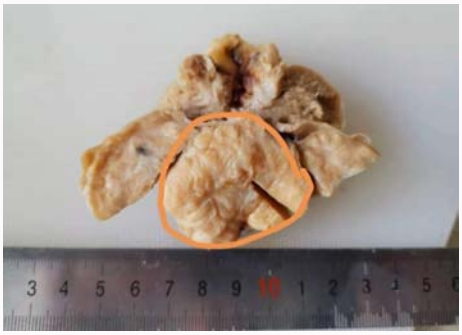

Figure 3: Tumor mass: 4 cm × 3.5 cm × 3.5 cm, the cut surface is grayish white with a crisp texture.

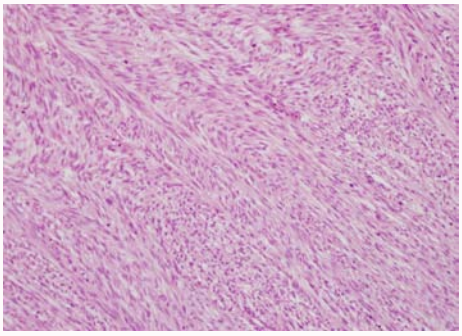

Figure 4a: The tumor was composed of pleomorphic spindle cells arranged in fascicles (HE x200).

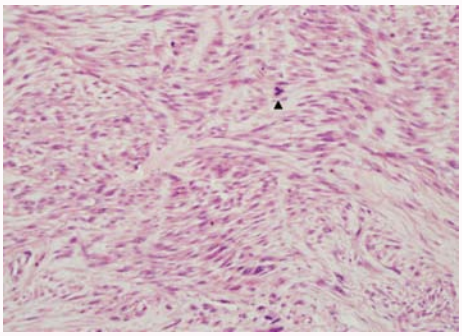

Figure 4b: The tumor cells are markedly heterogeneous, with pathological mitosis, as shown by the Black Arrow (HE x400).

Figure 5a: Desmin positive (immunohistochemistry x200).

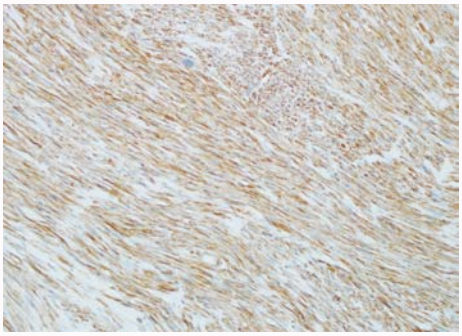

Figure 5b: H-caldemon positive (immunohistochemistry x400).

and could retrieve 24 cases of primary epididymal leiomyosarcoma that have been published. Kwaeetal [8] in 1949 claimed the first case report of primary leiomyosarcoma of epididymis. Epididymal leiomyosarcoma are more common in men aged 50 to 80 years. But it may also occur in children and in the young and the middle-aged [9]. Risk factors for testicular leiomyosarcoma include high doses of anabolic steroids, chronic inflammation, or past exposure to radiation [10], but there are no reported predisposing factors leading to epididymal leiomyosarcoma in the literature. The leiomyosarcoma of the epididymis is difficult to diagnose preoperatively and usually presents as a painless, hard mass that may cause discomfort. The mass is usually well-defined, lobulated, easily moving, and sometimes accompanied by epididymitis. The examination should begin with an ultrasound of the scrotum to determine the size and location, texture, and vascular distribution of the mass. The leiomyosarcoma of the epididymis appears as a solid mass with blood flow on the sonogram, but epididymitis has a similar sonographic appearance that needs to be identified. However, MRI may be better at locating the tumor and

understand their relationship to surrounding tissue in more detail [11].

The final diagnosis of Leiomyosarcoma requires a histological examination to differentiate between benign and malignant smooth muscle. The classic histologic features are rhomboid, fasciculate and braided arrangement of tumor cells, marked cell atypia, and obvious mitosis [12]. Immunohistochemistry: SMA (+), Desmin (+), S-100 (-), CD34 (-), CD117 (-) [13].

Treatment of choice is high inguinal orchiectomy. Lymph node dissection is not required. Resection must be extensive and complete [10]. The effect of chemotherapy and radiation on the leiomyosarcoma of the epididymis remains unclear [14].

The biological behavior of the Leiomyosarcoma is difficult to determine. It has been reported that tumor grade, stage, histological type, lymph node metastasis or distant metastasis are closely related to prognosis [15]. Recurrence is common, so follow-up is necessary [16].

The purpose of this article is to delineate the clinicopathologic features of epididymal leiomyosarcoma and spread awareness of the malignant nature of the disease, to improve the diagnosis and treatment of this disease.

Conclusion

Epididymal leiomyosarcoma is rare and difficult to diagnose pre-operatively. The final diagnosis of Leiomyosarcoma requires histologic examination. Resection must be extensive and complete. The effect of chemotherapy and radiation on the leiomyosarcoma of the epididymis remains unclear. Recurrence is common, so follow-up is necessary.

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