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Case Report

Solitary Fibrous Tumor of Testis: A rare Case with Review of Literature

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ABSTRACT

Solitary fibrous tumors (SFTs) more commonly arise in the pleura but recently, they have been reported in several extrapleural organs. Urogenital localization is rare, and only small numbers of cases of paratesticular SFT have been reported. An 81-year-old male with a history of colon carcinoma and complaint of testis swelling was referred for evaluation of a right paratesticular mass. Physical examination revealed a 2 cm oval-shaped paratesticular mass and herniation of intestinal loops in the right inguinal region after cough and Valsalva maneuver. An ultrasound examination was found in the upper pole of testis a well-defined hypoechoic mass in favor of testicular mass. It also revealed moderate to severe bilateral hydrocele and calcified wall in favor of benign lesion. In conclusion, SFT should be considered in the differential diagnosis of paratesticular masses and needs to be confirmed by IHC. CD34 and CD99 biomarkers are useful for confirmation of SFT.

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Introduction

The solitary fibrous tumor (SFT) is a less frequent spindle cell neoplasm stemming from mesenchymal cells. It was first described by Klemperer and Rabin.[1] In the past, most SFTs were known as hemangiopericytomas. Then after, pathologists used "solitary fibrous tumor" instead of "hemangiopericytoma".[2] SFTs more commonly arise in the pleura, but recently they have been identified in many extrapleural sites such as; meninges, oral cavity, nasal cavity, mediastinum, orbit, pelvis, retroperitoneum, kidney, vagina, vulva, urinary bladder, seminal vesicles, prostate, testis, and paratesticular region.[3-6]

To the best of our knowledge, the SFTs in the urogenital organs are rare and uncommon[7] and only a limited number of Paratesticular SFTs have been reported. So there are controversies on histopathogenesis of these tumors.[8-11] It is very important to make a differential diagnosis with the help of immunohistochemistry examinations. In the present report, we introduce a rare case of paratesticular SFT and its clinical, pathological and immunohistochemical characteristics.

Case Presentation

An 81-year-old male with a history of colon carcinoma and complaint of right testis painless swelling was referred for evaluation of a right paratesticular mass. In the past medical history, there was a transurethral resection of the bladder that the patient was not aware of the pathology report. The report was not available and no significant treatment was done.

Physical examination revealed a 2 cm oval-shaped paratesticular mass and herniation of intestinal loops in the right inguinal region after cough and Valsalva maneuver. Complete blood count and routine blood biochemistry reports were within the normal range. Testis tumor markers were evaluated and a mild increase in carcinoembryonic antigen (CEA) of serum was seen (7.6 ng/ml; reference range: 0-4 ng/ml). Alpha-

fetoprotein (AFP) was 101.5ng/ml, (reference range: up to 8.75 ng/ml), lactate dehydrogenase (LDH) was 290 IU/L (reference range: 225-500 IU/L), and beta-human chorionic gonadotropin (βhCG) was 0.95 mIU/ml (reference range: up to 2.24 mIU/ml). An ultrasound examination was found in the upper pole of testis a well-defined hypoechoic mass (9*7mm) in favor of testicular mass. It also revealed moderate to severe bilateral hydrocele and calcified wall in favor of benign lesion. Meanwhile left kidney simple cortical cyst (27mm) in ultrasound examination was detected. Size of each testis was 45*23 mm. The paratesticular tumor was excised and sent to the pathologist. Hematoxylin-Eosin stain showed a proliferation of spindle cells in storiform pattern with a few dilated vessels (Figure 1). Immunohistochemical examination showed the cluster of differentiation (CD) 99 positive and strong focally positivity CD34 in the neoplastic cells of the tumor but it was negative for B-cell lymphoma 2 (BCL-2), (Figure 2). Thus, the diagnosis of a paratesticular solitary fibrous tumor confirmed by histopathology and immunohistochemical findings. Follow-up after surgical treatment was not available and the patient's written consent to report the case obtained.

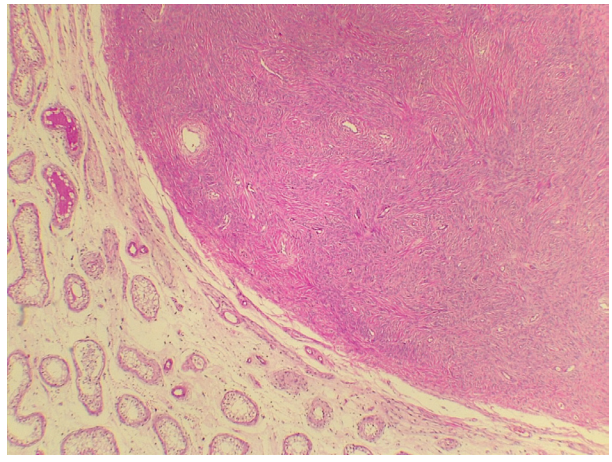

Figure 1: Hematoxylin-Eosin staining: Paratesticular SFT with spindle cells in storiform pattern (×40 magnification).

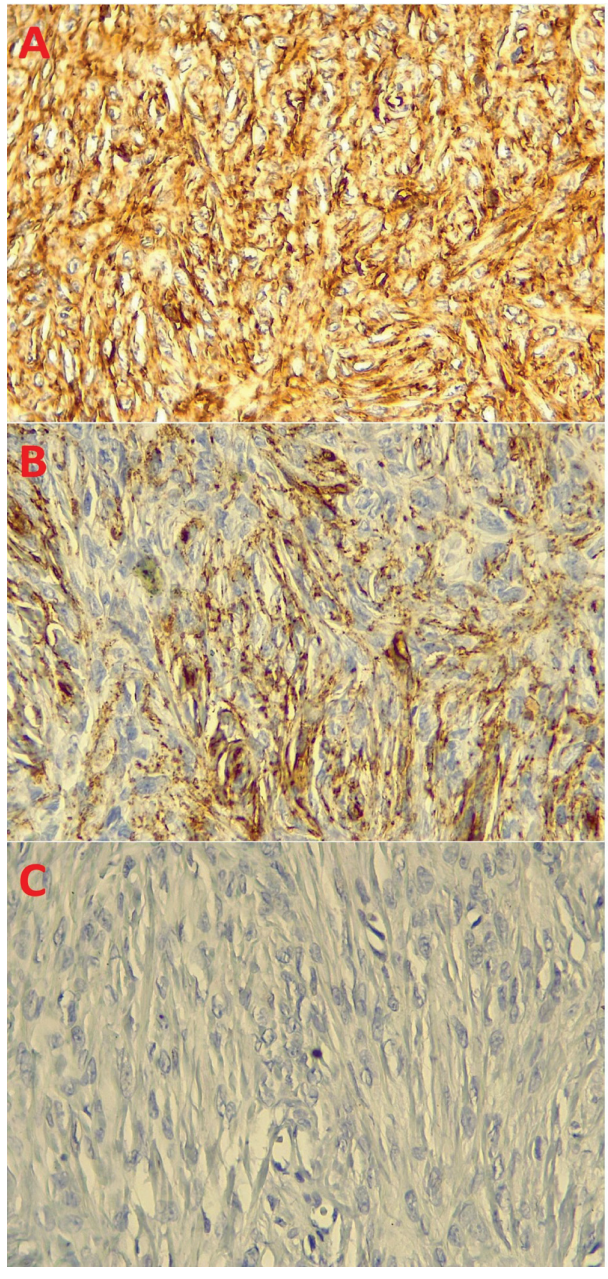

Figure 2: Immunohistochemistry staining: A) CD99 positivity, B) CD34 focal positivity, and C) BCL-2 negativity (×400 magnification).

Discussion

SFT is a mesenchymal neoplasm and it was first reported in 1931.[1] SFT is pathologically characterized by spindle cell proliferation. Although it is suggested that SFTs are limited to the pleura, they have been increasingly identified in many extrapleural sites.[3-6] Currently, only a small number of SFT cases have been recognized in the urogenital organs, testis and paratesticular region.[7-11] Immunohistochemistry has a prominent role in accurate diagnosis of tumor and ruling out of differential diagnosis.[12]

Our IHC results showed focal positivity for CD34 which is compatible with previous studies.[7, 13-16] CD34 immunoreactivity has been reported strongly in most SFTs. Although CD34 is considered as the most prominent feature in the diagnosis of SFTs,[17] about 5-10% of SFTs do not show any positivity for CD34.[18] Moreover, malignant SFTs mostly showed less CD34 reactivity. The histological criteria in favor of malignancy are larger tumors (more than 5 cm) with hypercellularity, nuclear pleomorphism, necrosis, more than 4 mitoses/10 HPF, and infiltrative borders.[19]

Other positive immunoreactivities in the present case include CD99. On the other hand, IHC examination showed negative expression of BCL-2 in our case while in contrast to our result, previous studies generally showed positivity for BCL-2.[13-15] Moreover, C Gengler & L Guillou showed positive reactivity for BCL-2 (30%), CD34 (80-90%), CD99 (70%), EMA (30%) and actin (20%) and negative reactivity for Desmin, cytokeratin and S-100 protein.[19] Despite the fact that the above mentioned IHC markers are strongly suggested in SFT diagnosis, Chmielecki and colleagues[19] reported a novel fusion gene, NAB2–STAT6, to characterize the SFTs. Several studies also revealed that STAT6 can be used for differential diagnosis of SFTs.[20-22]

As most of the SFTs are benign and well encapsulated,

complete removal of the tumor with negative margins is the common treatment for SFTs [23,24] and those with malignant characteristics need more therapeutic measures.[9] However, several studies have demonstrated that radiotherapy and chemotherapy, with ifosfamide and adriamycin, was effective.[9,25] The role of radiotherapy and chemotherapy is uncertain and further study is needed.[7] Therefore, differentiating the malignant SFTs from benign ones using IHC and histopathology can help to choose the most effective way of management.

Conclusion

SFT should be considered in the differential diagnosis of paratesticular masses and needs to be confirmed by IHC. CD34 and CD99 biomarkers are useful for confirmation of SFT.

Conflict of Interest: None declared.

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