Journal of Oncological Sciences 3 (2017) 135–136

Contents lists available at ScienceDirect

Journal of Oncological Sciences

journal homepage: https://www.elsevier.com/locate/jons

A paratesticular inflammatory myofibroblastic tumor and review of the literature

Ali Alkan [a, *], Serhat Toprak [b], Elif Berna Köksoy [c]

[a] Osmaniye Public Hospital, Medical Oncology, Osmaniye, Turkey
[b] Osmaniye Public Hospital, Pathology, Osmaniye, Turkey
[c] Ankara University School of Medicine, Medical Oncology, Turkey

ARTICLE INFO

Article history:
Received 13 August 2017
Received in revised form
7 September 2017
Accepted 14 September 2017
Available online 22 September 2017

Keywords:
Testis tumor
Inflammatory myofibroblastic tumor
Cancer

ABSTRACT

Inflammatory Myofibroblastic tumor (IMT) is a pseudosarcomatous inflammatory lesion originating from soft tissue or viscera. It has been reported in different solid organs, rarely in genitourinary system. Thirty-eight year old male patient presented with scrotal pain and lump. Ultrasonography revealed a 4.5 × 3.5cm ovoid heterogeneous mass related with epididymis. Patient was treated with right orchiectomy and the pathology confirmed the diagnosis of paratesticular IMT. Our case had similar clinical and pathological characteristics with other cases in the literature. Although IMT has a benign nature, due to inadequate experience and possibility of recurrence in other organs, patients should be followed after surgical resection.

1. Introduction

Inflammatory myofibroblastic tumor (IMT) is a pseudosarcomatous inflammatory lesion originating from soft tissue or viscera. Although it was initially reported in thoracic cavity, it has been documented in a variety of extrapulmonary sites, ranging from brain to bladder.[1] Genitourinary IMTs have been reported, accounting 9% of extrapulmonary IMTs. In addition, paratesticular IMTs have been rarely reported in the literature.[2–6] We present a young male patient who presented with paratesticular IMT and review of the literature.

2. Case report

A thirty eight year old male patient without any comorbidities, was presented with scrotal pain and a lump. There was no trauma, fever, symptoms of infection. On physical examination a 4 × 3cm non-tender, firm mass was palpated at the posterior of right testis. An inguinal hernia was detected. The skin of scrotum was normal and there were no pathological inguinal lymph nodes. Ultrasonography revealed a 4.5 × 3.5cm ovoid heterogeneous mass related with epididymis. Abdominal imaging was normal. AFP, BCG ve LDH levels were normal. Patient was treated with right orchiectomy, resection of mass and and repair of inguinal hernia. Pathological examination showed a 4.5 × 3.5cm globular mass with a capsulated external surface. Microscopic evaluation showed a myofibroblastic proliferation related with epididymis (Fig. 1). There were inflammatory cells, predominantly plasma cells and lymphocytes. There was a sclerotic fibrous stroma consisting of vascular structures and mononuclear cells. Immunohistochemistry was positive for actin, desmin, vimentin and negative for cytokeratins (AE1/AE3) and S100. Pulmonary and abdominal computed tomography was normal. Patient was followed without any further treatment. Patient has been followed with 3 monthly intervals and a workup with physical examination, scrotal/abdominal ultrasonography and chest x-ray. After 1 year of follow up, patient is free of recurrence.

3. Discussion

Inflammatory Myofibroblastic tumor (IMT) is a rare spindle cell proliferation which has been reported in the literature with different designations, such as inflammatory pseudotumor, plasma cell pseudotumor, xanthomatous pseudotumor, atypical myofibroblastic tumor, atypical fibromyxoid tumor, pseudosarcoma, plasma cell granuloma, and fibrous pseudotumor.[2] It is composed of myofibroblastic mesenchymal spindle cells with an inflammatory infiltrate of plasma cells, lymphocytes, and eosinophils.[7] The pathology has been reported in different organs, such as lung, skin, soft tissues, breast, gastrointestinal tract, pancreas, oral cavity, bone

* Corresponding author. Osmaniye Public Hospital, Osmaniye devlet Hastanesi, EK hizmet binası, tıbbi onkoloji bölümü, Merkez, Osmaniye, TR 80350, Turkey.
E-mail address: alkanali@yahoo.com (A. Alkan).
Peer review under responsibility of Turkish Society of Medical Oncology.

http://dx.doi.org/10.1016/j.jons.2017.09.002

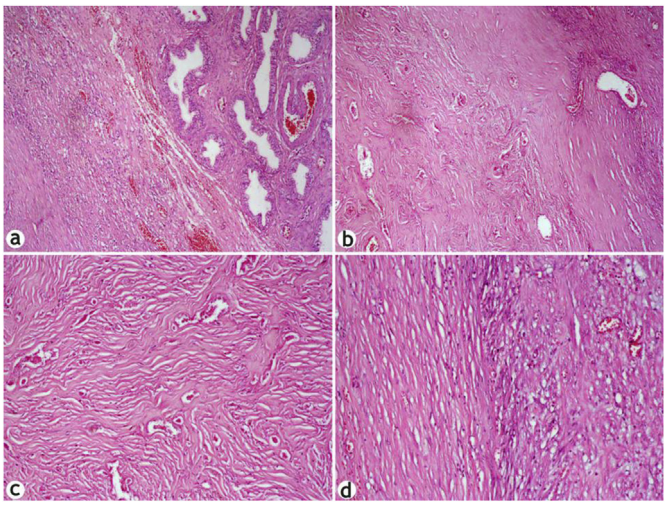

Fig. 1. a) Hex4; epididymis and myofibroblastic proliferation consisting of congested vascular structures, b) Hex10; sclerotic fibrous stroma with vascular structures, c) HEx20; Fibrotic background with mononuclear cells, d) Hex40; Myofibroblastic cells (right) and fibrosis (left).

and central nervous system.[3]

Genitourinary system is a rare location for this rare pathology. In the genitourinary tract, IMT has been reported in the kidney, urethra, prostate, ureter, and testis but is most frequently observed in the bladder. IMT has been reported in paratesticular structures in fewer than 10 cases in literature. The exact etiology is not known, but ischemia, infection and chronic irritation have been postulated as etiological factors.[8] Painless mass in scrotum is the usual presentation of paratesticular IMT. It has been most commonly reported in children or young adults. However, it has also been described in an elderly male patient.[9] Ultrasonography of the scrotum should be the initial imaging modality to differentiate testicular masses from paratesticular ones. During evaluation of paratesticular mass, more common pathologies should be considered. Varicocele, spermatocele, infections and primary malignant diseases should be excluded.[5]

According to cases, paratesticular IMT is considered as a benign pathology. Although IMT series in different organs have a 25% recurrence rate, aggressive and locally invasive paratesticular IMT has been rarely reported.[10] Thus, lesions occurring in the retroperitoneum, mesentery or mediastinum are considered potentially malignant and could recur after treatment. Optimal treatment strategy is wide resection of the mass with conserving testis. Due to benign nature of the disease and rare recurrences, adjuvant chemotherapy or radiotherapy are not recommended. In recurrent cases, resection of the mass is recommended. Radiotherapy or chemotherapy with methotrexate or steroids could be used.[10] Rearrangements involving the *ALK* locus on chromosome 2p23 have been documented in approximately 50% of IMTs. Distant metastases occur primarily in ALK-negative IMTs, but local recurrence occurs regardless of ALK expression.[11] In recurrent ALK positive cases, Crizotinib is an effective treatment modality.

In conclusion, our case had similar clinical and pathological characteristics with other cases in the literature. The patient was free of recurrence after 1 year and still under close follow up. Paratesticular IMT is a rare pathology and should be considered in the differential diagnosis of paratesticular masses. Although it has a benign nature, due to inadequate experience and recurrence potential in other organs, patients should be followed after surgical resection.

References

1. Coffin CM, Watterson J, Priest JR, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor). A clinicopathologic and immunohistochemical study of 84 cases. *Am J Surg Pathol*. 1995;19:859–872.
2. Harel MMJ. Paratesticular inflammatory myofibroblastic tumor in a pediatric patient. *Case Rep Urol*. 2014;2014:303678.
3. Dangle PP, Wang WP, Pohar KS. Inflammatory myofibroblastic tumor of epididymis: a case report and review of literature. *World J Surg Oncol*. 2008;6:119.
4. Tunuguntla H, Mishra A, Jorda M, Gosalbez R. Inflammatory myofibroblastic tumor of the epididymis: case report and review of the literature. *Urology*. 2011;78:183–185.
5. Rafeek N, Joseph LD, Rajendiran S, Narayanan CD. Inflammatory myofibroblastic tumor of spermatic cord. *Int J Surg Case Rep*. 2012;3:618–621.
6. Nistal M, Gonzalez-Peramato P, Serrano A, Reyes-Mugica M, Cajaiba MM. Primary intratesticular spindle cell tumors: interdigitating dendritic cell tumor and inflammatory myofibroblastic tumor. *Int J Surg Pathol*. 2011;19:104–109.
7. Coffin CM, Hornick JL, Fletcher CD. Inflammatory myofibroblastic tumor: comparison of clinicopathologic, histologic, and immunohistochemical features including ALK expression in atypical and aggressive cases. *Am J Surg Pathol*. 2007;31:509–520.
8. Brauers A, Striepecke E, Mersdorf A, Sohn M, Fuzesi L. Inflammatory pseudotumor of the epididymis. *Eur Urol*. 1997;32:253–255.
9. Chakrabarti N, Shetty R. Inflammatory myofibroblastic sarcoma of the spermatic cord. *Indian J Surg*. 2010;72:152–154.
10. Meis JM, Enzinger FM. Inflammatory fibrosarcoma of the mesentery and retroperitoneum. A tumor closely simulating inflammatory pseudotumor. *Am J Surg Pathol*. 1991;15:1146–1156.
11. Butrynski JE, D'Adamo DR, Hornick JL, Dal Cin P, Antonescu CR, Jhanwar SC, et al. Crizotinib in ALK-rearranged inflammatory myofibroblastic tumor. *N Engl J Med*. 2010;363:1727–1733.