

BILATERAL EPIDIDYMAL LEIOMYOMAS: ATYPICAL CLINICAL PRESENTATION, UTILITY OF SCROTAL ULTRASOUND AND SURGICAL CONSIDERATION

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ABSTRACT

Epididymal tumours are rare occurrences and even more so are bilateral diseases. Patients with epididymal tumours are commonly brought to clinical attention with painless scrotal mass. We present a rare case of bilateral epididymal leiomyomas with an atypical clinical presentation of painful scrotal swelling masquerading as recurrent epididymo-orchitis.

In this article, the clinical, sonographic and histological findings are described. Common differential diagnosis of epididymal tumours and their imaging features are also discussed.

Introduction

Epididymal leiomyomas are one of the commonest epididymal tumours.¹ Patients may present with painful scrotal swelling which may be regarded as epididymo-orchitis due to tender scrotal swelling on physical examination. Our patient was found to have bilateral epididymal tumours on ultrasound which was arranged due to the atypical clinical course of epididymo-orchitis. Awareness of possible bilateral epididymal tumours is important for clinicians in formulating their surgical plan in order to preserve testicular function especially in younger adult males.

Case Presentation

A 74-year-old man presented with repeated episodes of left sided scrotal pain and swelling for about 1.5 year. Physical examination found tender left epididymal thickening and mildly tender left testis. Clinical exami-

nation of the contralateral hemiscrotum showed a 1cm non-tender nodule at right epididymis. Right testis was unremarkable. The patient was otherwise well without any alarming clinical feature. He was treated as left epididymo-orchitis with repeated courses of Levofloxacin but to no avail.

The patient underwent ultrasound examination due to the atypical clinical course, which revealed a 3.0 x 2.7 x 3.4cm well-defined heterogeneous hypoechoic roundish solid lesion in left epididymal tail. Mild intralesional Doppler signal was noted within this lesion. (Fig. 1a & 1b) Mild mass effect of this lesion results in upward displacement of left testis. The smaller lesion detected clinically at contralateral epididymis showed similar imaging characteristics as the left epididymal lesion, measuring 1.0 x 0.8 x 0.9 cm. (Fig. 1c & 1d) Otherwise, the bilateral testicles and spermatic cords were unremarkable with no abnormally increased vascularity or altered echogenicity to suggest epididymo-orchitis.

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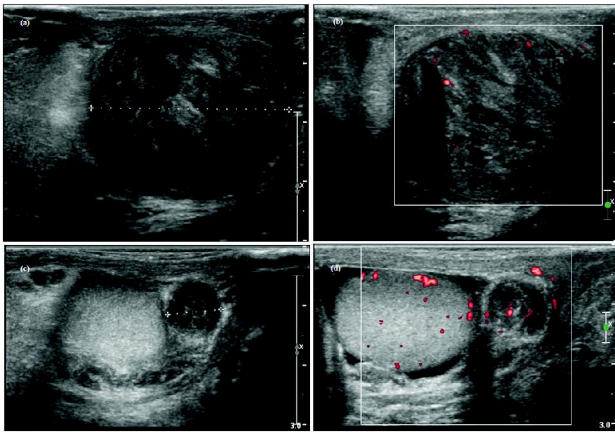


Figure 1: (a) Grayscale ultrasound image shows a 3.0 x 2.7 x 3.4 cm well-defined heterogeneous hypoechoic roundish solid lesion in left epididymal tail. (b) Doppler ultrasound study shows mild intralesional vascularity. (c) Right epididymal lesion with similar sonographic features as left sided lesion. (d) Mild vascularity is also noted in right epididymal lesion.

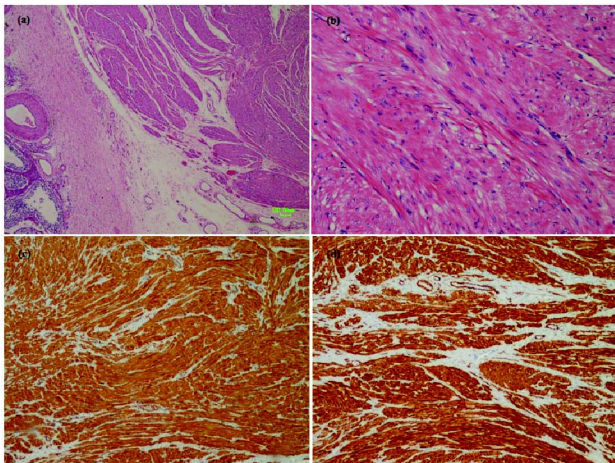


Figure 2: (a) A well-circumscribed tumour with adjacent testicular parenchyma. The tumour is composed of interlacing fascicles of spindle cells with eosinophilic cytoplasm and elongated 'cigar-shaped'. (H&E; original magnification, 4X) (b) The spindle cells show no significant cytologic atypia. Mitotic activity is inconspicuous and tumour necrosis is absent. (H&E; original magnification, 20X) (c) Tumour is alpha-smooth muscle actin positive (Immunohistochemical stain; original magnification, 10X) (d) Tumour is h-caldesmon positive (Immunohistochemical stain; original magnification, 10X)

Left orchidectomy was subsequently performed to resect left epididymal tumour in view of significant symptom to the patient. Intraoperative finding revealed that the tumour was well-demarcated and separated from normal left testis and left spermatic cord. Histopathology of the specimen showed a circumscribed left epididymal nodule composed of interlacing fascicles of spindle cells having eosinophilic cytoplasm and

elongated "cigar-shaped" nuclei; no significant cytologic atypia was observed in these spindle cells. Mitotic activity was inconspicuous and tumour necrosis was absent. Immunostudy showed the neoplastic cells were diffusely positive for smooth muscle markers. The features were consistent with epididymal leiomyoma. (Fig. 2) The left testis and left spermatic cord included in the specimen were unremarkable. Since the contralateral epididymal lesion was static for about 3 years besides being clinically asymptomatic and similar in the benign sonographic features as the larger resected left epididymal leiomyoma, it was decided that clinical observation for this lesion would be appropriate, with a presumed diagnosis of a contralateral synchronous epididymal leiomyoma.

Discussion

Primary epididymal tumours are rare intra-scrotal neoplasms of mesenchymal origin and most of them are benign in nature.¹ The three commonest types of benign epididymal tumours are adenomatoid tumour, leiomyoma and papillary cystadenoma, accounting for 73%, 11% and 9% of these tumours respectively.¹ Epididymal leiomyoma is the second most common primary epididymal tumours, representing 6% of all epididymal tumours.¹ It is a benign neoplasm derived from smooth muscle cells and commonly seen in the uterus of female subjects although other organs in the genitourinary tract (e.g. renal capsule, bladder, etc.) can also be affected.² Bilateral synchronous epididymal leiomyomas are extremely rare and only a small number of cases have been reported in the medical literature so far.³ To the best of our knowledge, there is no published report of bilateral synchronous epididymal leiomyomas in the indexed medical journals over the past ten years. Most of the patients with bilateral epididymal leiomyomas presented with bilateral painless scrotal masses with a protracted course of gradual enlargement.⁴ Our patient, however, presented with a painful left-sided scrotal swelling masquerading as recurrent epididymo-orchitis apart from the asymptomatic contralateral lesion. Clinicians should therefore be aware of this entity and ultrasound examination in atypical cases of scrotal swelling or pain could help clarify the underlying pathology.

Sonographic features of the aforementioned three commonest epididymal tumours could be variable. Adenomatoid tumours can appear as oval well-circumscribed homogeneous isoechoic to hyperechoic lesion although predominantly cystic appearance is also possible.⁵ Leiomyoma can appear as well-circumscribed heterogeneous hypoechoic lesion with multiple recurrent shadows.⁵ It can also demonstrate intralesional calcifications similar as uterine leiomyoma; alternatively, it may also show predominantly cystic appearance. Papillary cystadenoma commonly appears as predominantly solid mass with small cystic spaces but may also be largely cystic with small papillary projections.⁵ The important associated background history of von Hippel-Lindau disease would provide a clue to the diagnosis.

Surgical treatment for epididymal leiomyoma includes epididymectomy or orchidectomy. Given the benign nature and possible bilateral occurrence of epididymal leiomyoma, a more conservative surgical approach would be the treatment of choice in order to preserve testicular function. However, in cases where the tumour adheres to testicle rendering exclusion of malignancy unreliable by gross examination alone, more radical resection with orchidectomy would be an optimal treatment.

Conclusion

In summary, we present a rare case of bilateral epididymal leiomyomas masquerading as epididymo-orchitis at initial presentation. The atypical clinical presentation, utility of ultrasound in clarification of clinical findings and surgical options have been discussed above. Clinicians should be aware of this rare entity which can at times mimic epididymo-orchitis and possible bilateral occurrence which is an important consideration in gauging the radicality of surgical resection.

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