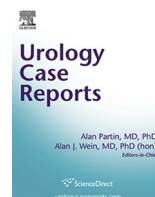




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Oncology

Serial Ultrasonography Assessments of a Testicular Infarction Mimicking Testicular Tumor in a Behcet Disease Patient

Zeid Al-Ani*, Syahminan Suut, Bella Huasen

Department of Radiology, North Manchester General Hospital, Delaunays Road, Manchester, M8 5RB, UK

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ABSTRACT

Behcet disease (BD), a vasculitic disease, may present with a broad range of systemic manifestations. Urologic complications are rarely described in the literature, but when they occur, they present as epididymo-orchitis. We describe a rare case of testicular infarction in a patient with BD followed up with serial ultrasound imaging. We highlight the diagnostic challenges when presented with testicular pain in a patient with BD and the potential consequences in the management.

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Introduction

Behcet disease (BD), a vasculitic disease, may present with a broad range of systemic manifestations. Urologic complications are rarely described in the literature, but when they occur, they present as epididymo-orchitis. We describe a rare case of testicular infarction in a patient with BD followed up with serial ultrasound imaging. We highlight the diagnostic challenges when presented with testicular pain in a patient with BD and the potential consequences in the management.

Case presentation

A 36-year-old male patient presented with a 1-day history of left-sided scrotal pain. There were no urinary symptoms or fever. There was no recent preceding injury or trauma. He had similar episodes of left testicular pain diagnosed as epididymitis several years ago but had remained well in the interim. His past medical history included a diagnosis of BD with scrotal and mouth ulcers and ocular involvement. This was stable and treated with steroids, cyclosporine, colchicine, and azathioprine.

Scrotal examination elicited tenderness of a swollen left testicle. No mass was palpable. Hematology revealed raised white blood cell count at $16.4 \times 10^9/L$. Urine and microbiologic analyses were unremarkable.

Germ cell tumor markers (lactate dehydrogenase, alpha-fetoprotein and human chorionic gonadotropin) were within normal range.

He was clinically diagnosed with epididymo-orchitis, and oral ciprofloxacin and doxycycline were commenced.

Ultrasound scan showed an isoechoic and well-defined abnormality in the upper pole of left testis, merging with a swollen and poorly defined epididymal head. This was a new finding compared with a previous ultrasound scan performed 4 years previously. Color Doppler assessment was unremarkable (Fig. 1).

There was a wide differential for the nature of this lesion, including the incidental finding of a testicular tumor. After multi-disciplinary input, a repeat testicular ultrasound scan was performed, which showed evolution of the testicular lesion becoming hypoechoic compared with the rest of the testis (Fig. 2).

The patient was reviewed in outpatient clinic after 3 weeks when he reported improvement in his symptoms and resolution of the testicular pain. Owing to the relative lack of symptoms and the concern for testicular malignancy, possibility of orchidectomy was suggested.

A third ultrasound scan was performed 4 weeks from initial presentation and showed further evolution of the left upper pole testicular lesion, which appeared smaller and more hypoechoic with well-defined borders (Fig. 3).

As the patient was well and reluctant to have orchidectomy, a conservative management approach was adopted.

Ultrasound scan performed 10 weeks from the first scan showed that the lesion had significantly decreased in size confirming the diagnosis of testicular infarction (Fig. 4).

Discussion

BD is a progressive vasculitic disease with a relapsing and remitting course. The prevalence in North America and Europe is

* Corresponding author. Tel.: +44-07842880529.

E-mail address: zeid82@yahoo.com (Z. Al-Ani).

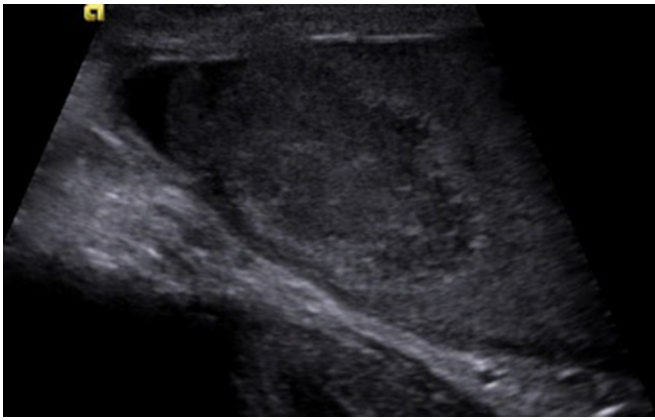


Figure 1. Isoechoic lesion in the upper pole of left testis with well-defined and thick wall is shown. The abnormality was hypovascular on color Doppler.

1 case per 15,000–500,000 population compared with 420 cases per 100,000 population in Turkey.^{1,2} The clinical manifestations presenting in most of the patients with BD are oral and genital ulcers, uveitis, and skin lesions. Other common clinical manifestations include arthritis, thrombophlebitis, and various neurologic syndromes. Less frequent complications include arterial thrombosis, systemic and pulmonary circulation aneurysms, colitis, epididymitis, and orchitis.³

The frequency of epididymo-orchitis in BD has geographic variation and differs between juvenile and adult patients. The highest frequency (44%) of epididymo-orchitis has been reported in Russia and the lowest (2%) in France. Epididymo-orchitis was noted in 11.3% of adult patients and 7.7% in children. The incidence of epididymo-orchitis was 31% in Iraqi but only 6% in Turkish patients.⁴ Zouboulis et al⁵ reported prostatitis and epididymo-orchitis with BD in 22% of cases.

The etiology of epididymo-orchitis in patients with BD is not fully understood. Vasculitis causing inflammation has been proposed, but there is lack of histologic data. Infection has also been implicated; however, urinary cultures have consistently been negative in case series, and inflammation subsides with administration of anti-inflammatory drugs.^{4,6} Clinical presentation in different case series and reports was mainly as testicular pain, with testicular mass being less common.^{7–10}

Testicular infarction is a rare entity, with <50 reported cases.⁸ Although vasculitis was reported as a cause for testicular infarction in few cases before, none of these patients had BD. Case reports

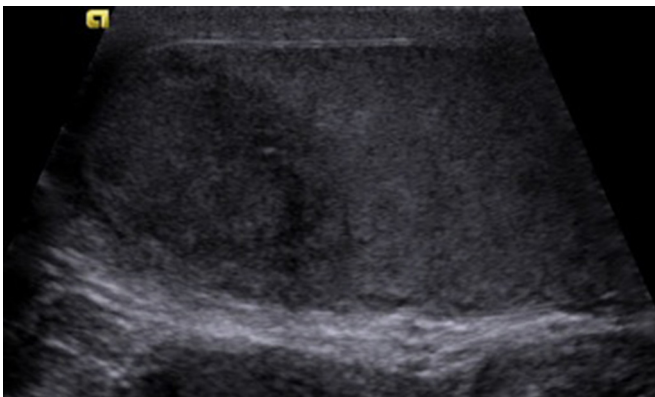


Figure 2. Second ultrasound scan 2 weeks from presentation is shown. The lesion in the upper pole of left testis showed marked evolution compared with previous scan.

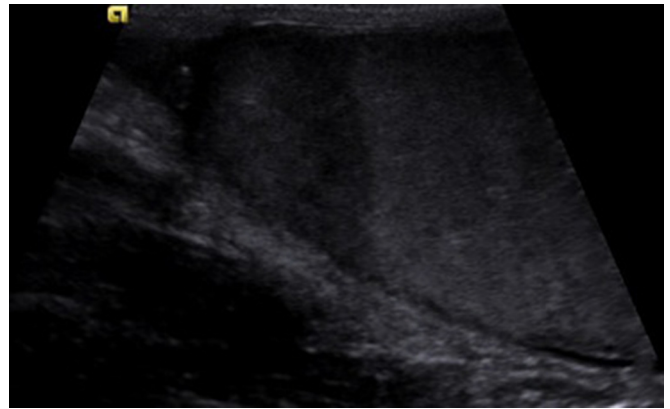


Figure 3. Third ultrasound scan 4 weeks from presentation is shown. This showed further evolution of the left upper pole testicular lesion becoming smaller and more hypoechoic.

of polyarteritis nodosa as a cause of testicular infarction are described.^{9,10} In one case, a patient had bilateral testicular infarction and orchidectomy with subsequent androgen hormone replacement. In another case report, a 19-year-old man presented with unilateral testicular swelling and pain. The initial diagnosis of epididymo-orchitis was altered to testicular neoplasm after ultrasonography. Histologic examination after orchidectomy showed testicular vasculitis.¹¹

Furthermore, there are 2 case series describing testicular infarction secondary to vasculitis. In one series of 19 cases of testicular infarction with associated vasculitis, 14 showed polyarteritis nodosa features with transmural necrotizing inflammation of small-medium arteries. In 4 cases, vasculitis was granulomatous, and 1 case was lymphocytic; in most patients, the preoperative impression was testicular cancer.⁷ In another case series of 10 testicular infarctions retrieved from the pathology records of one institution, giant cell vasculitis was identified as an etiologic factor in one patient.⁸

The diagnosis of BD is difficult, and diagnostic criteria includes recurrent oral ulcerations at least 3 times in 1 year with 2 of the following: recurrent genital ulcerations, eye lesions (uveitis or retinal vasculitis) observed by an ophthalmologist, skin lesions (erythema nodosum, pseudofolliculitis, papulopustular lesions, and acneiform nodules) in adult patients not on corticosteroids, and a positive “pathergy test” read by a physician within 24–48 hours of testing.¹²

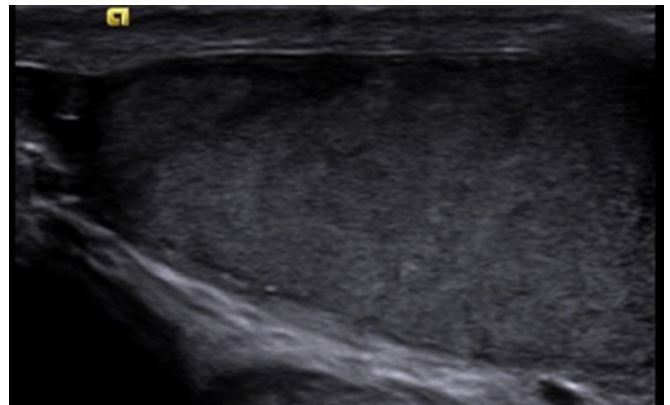


Figure 4. Ultrasound scan 10 weeks from first scan is shown. The lesion had significantly decreased in size.

Conclusion

Ultrasonography remains a good modality for investigating testicular pain and swelling. Awareness of BD and other vasculitis patients' urologic complications (epididymo-orchitis and testicular infarction) is important, as the latter may be mistaken for testicular tumors. Orchiectomy should be avoided because of the need for androgen replacement therapy and various psychological factors. In asymptomatic and clinically well patients, a conservative monitoring approach should be considered before a diagnosis becomes definitive.

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