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Long term follow up of ultrasound diagnosed bilateral testicular sarcoidosis: Case report and literature review

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ABSTRACT

Testicular sarcoidosis is a rare condition characterised by non-caseating granulomas located within the testicles. This case study reports a 44-year-old man who presented initially in 2011 with testicular pain and bilateral testicular sarcoidosis was confirmed on scrotal US. In 2019 he presented with erectile dysfunction. Scrotal ultrasound scan (US) showed bilateral testicular sarcoidosis that has not changed in size and nature when compared with US in 2011.

Case introduction

Sarcoidosis is a rare, multi-system, inflammatory condition characterised by non-caseating granulomas. The most affected regions include the lungs, skin, and eyes. Urological sarcoidosis is rare, having been reported in 0.2% of all sarcoidosis cases.[1] Due to the rarity of this condition, there are no strict guidelines for the diagnosis or management, apart from a consistent emphasis on it being a diagnosis of exclusion to ensure malignancy is ruled out. We present a 37-year-old Caucasian man with an 8-year history of bilateral testicular sarcoidosis.

Case description

A 44-year-old Caucasian man was seen in 2019 for the management of his 3 years history of erectile dysfunction. His clinical history revealed that in 2011, he was seen by another urologist for bilateral testicular pain. At that time, he had normal testes on clinical examination and normal testicular tumour markers. He had a history of lymphopenia with immune thrombocytopenia, psoriasis, and Gilbert's syndrome. A scrotal US scan showed multiple small hypoechoic avascular lesions in both testes measuring 3mm, suggestive of testicular sarcoidosis (Fig. 1). A scrotal T2-weighted MRI scan showed bilateral testicular multiple focal lesions which were relatively hypointense and measuring up to 3–4mm. Systemic sarcoidosis was ruled out by a regional sarcoidosis centre that recommended a testicular biopsy to confirm testicular sarcoidosis. The patient declined a biopsy and chose to have US follow up, however he unfortunately defaulted from it.

In 2019, his clinical examination showed normal testes bilaterally. Testicular tumour markers and testosterone levels were normal. A repeat scrotal US confirmed multiple small hypoechoic areas in both testes with no vascularity and no change in the size or nature of the lesions previously seen on the scrotal US in 2011 in keeping with previously diagnosed testicular sarcoidosis (Fig. 2). He did not want to have children, and his erectile dysfunction responded to Tadalafil 5 mg as required. Once again, systemic sarcoidosis was ruled out and he declined a testicular biopsy as he considered himself asymptomatic and no change in his bilateral testicular lesions on US over 8 years.

Discussion

Sarcoidosis is an idiopathic, multisystem, chronic condition characterised by noncaseating granulomas which occur most often in lymph nodes and the lungs. Other organs affected by sarcoidosis are the kidneys, skin, liver and spleen. It affects 1–6 of every 1000 people worldwide with black people affected 3–20 times higher than their white counterparts. Women are affected 10-fold compared to males.[1]

Genitourinary involvement is rare with only 60 cases described in literature. It most commonly affects the epididymis (73%), testis (47%),

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spermatic cord (8%) and prostate (3%) according to a review of the 60 cases described to date.[2]

A rare manifestation of sarcoidosis is hypothalamic/pituitary sarcoidosis that can present either as diabetes insipidus or with hypogonadotropic hypogonadism symptoms.[3] In this case, the patient was affected by erectile dysfunction after eight years of the initial diagnosis but did not have any other systemic features of sarcoidosis.

Reduced sperm quality has been previously reported in testicular sarcoidosis,[4] therefore for patients where future fertility is a concern, a sperm analysis can be carried out and repeated to continuously monitor. In this case fertility investigations were not carried out as our patient did not want to have children.

The diagnosis of sarcoidosis occurs through exclusion of other conditions such as tuberculosis, syphilis, actinomycosis, venereum granuloma, sperm granuloma and malignant lymphoma. Accurate diagnosis of testicular sarcoidosis is important as, often it is misdiagnosed for a neoplasm resulting in unnecessary orchiectomies. Ultrasonography is the imaging of choice for testicular sarcoidosis and was used for long term follow-up in this case. On US, testicular sarcoidosis typically shows bilateral testicular/epididymal hypoechoic lesions. This, combined with the absence of positive tumour markers and/or presence of systemic findings of sarcoidosis, can lead to a definite diagnosis.[2]

Testicular carcinoma has a strong relationship with sarcoidosis in comparison to other solid tumours with incidence being an estimated 100 times higher when compared to a white, young, male population.[5] Testicular biopsies should be carried out to confirm sarcoidosis and exclude malignancy.[4] In this case the US appearance of the bilateral testicular lesions was not suspicious of malignancy and testicular tumour markers were normal. Also, the patient declined a testicular biopsy twice especially after 8 years with no changes in the size and appearance of his bilateral testicular lesions.

Management of testicular sarcoidosis is dependent on individual patient needs in terms of the severity of disease and associated symptoms and the consideration of future fertility preservation. Spontaneous resolution of sarcoidosis over the interval of up to three years has been recorded previously in literature.[2] Interestingly in this case, the patient had no change to his disease over the course of 8 years.

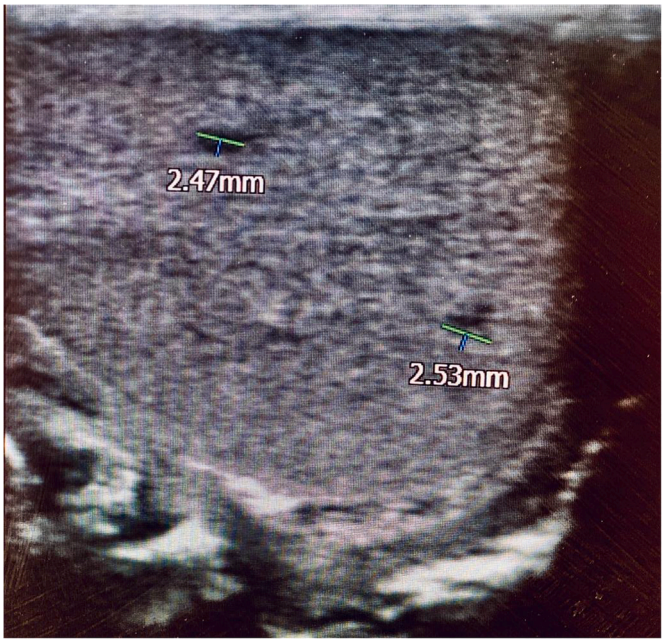

Fig. 2. Ultrasound scan in 2019 showing no significant change in size, appearance, and distribution of the avascular hypoechoic lesions in both testes.

Corticosteroids are used for those with unresolving disease, for patients where organ function is affected or in the presence of hypercalcaemia. Corticosteroids are used in testicular sarcoidosis as they have been shown to reduce testicular lesion size and alleviate metabolic repercussions and symptoms of sarcoidosis. Surgical intervention can be considered in cases where corticosteroids do not control symptoms or

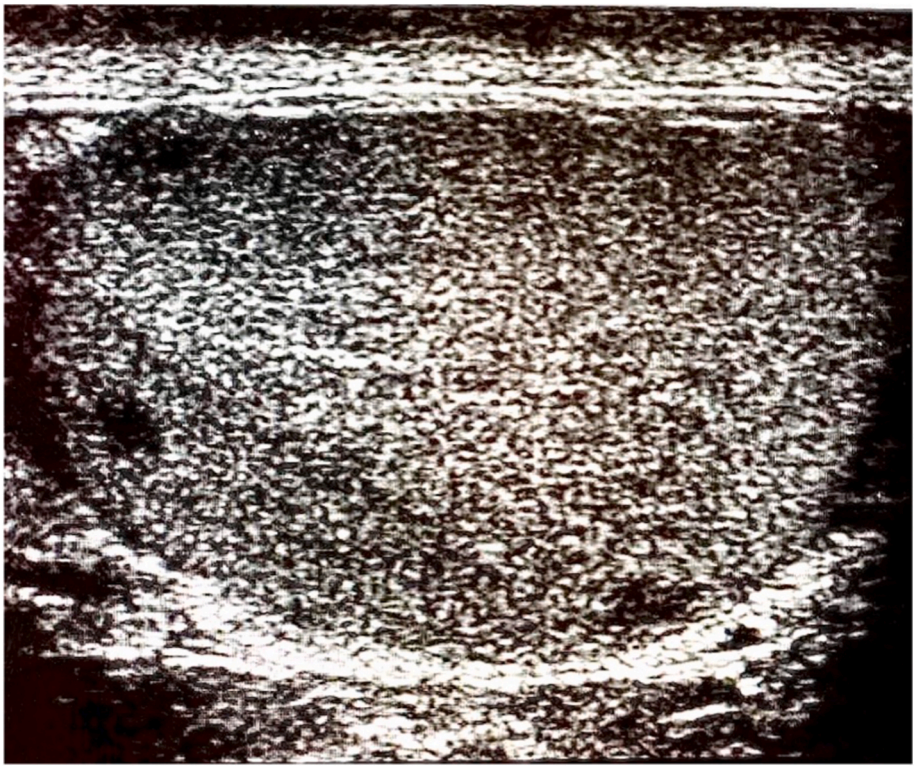

Fig. 1. Ultrasound scan in 2011 showing multiple small hypoechoic focal lesions with no internal vascularity.

disease progression.[2] In this case, corticosteroid management was not needed as the patient was asymptomatic until 2019, after which his erectile dysfunction was sufficiently treated with Tadalafil. It was agreed that serial ultrasound observation is sufficient after systemic sarcoidosis was ruled out both in 2011 and 2019. On both occasions, the patient declined testicular biopsy due to lack of symptoms and lack of clinical testicular change.

In conclusion, testicular sarcoidosis is an important differential diagnosis to consider despite its rarity as it is often mistaken for testicular malignancy resulting in unnecessary orchiectomies. Diagnosis can be made using sonography, absence of positive tumour markers and histology of testicular biopsies. Investigations to check for systemic sarcoidosis are also important to further confirm diagnosis and to assess the management.

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