Case Report

Testicular sarcoidosis with bilateral scrotal swelling

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Abbreviations & Acronyms
ACE = angiotensin-converting enzyme
AFP = α-fetoprotein
CT = computed tomography
IL-2 = interleukin-2
LDH = lactate dehydrogenase
MRI = magnetic resonance imaging
β-HCG = β-human chorionic gonadotropin

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How to cite this article: Kimura S, Momozono K, Shimamatsu K, Noguchi M. Testicular sarcoidosis with bilateral scrotal swelling. *IJU Case Rep*. 2020; **3**: 12–14.

Received 15 July 2019; accepted 1 October 2019. Online publication 19 October 2019

Introduction: Sarcoidosis is a disease in which noncaseating granulomas form in several organs, particularly in the lungs and skin. Male genitourinary involvement in sarcoidosis is uncommon.
Case presentation: A 32-year-old male with painless bilateral scrotal swelling who was diagnosed with lung sarcoidosis presented to our hospital. Serum tumor marker levels were normal. Scattered hypoechoic mass lesions in both testes were noted on ultrasound examination. Biopsy of both testes revealed pathologically noncaseating epithelioid cell granuloma, and perihilar lymphadenopathy and a granulomatous lung nodule were found on chest computed tomography. Semen examination was performed after the biopsy, demonstrating oligospermia. A corticosteroid regimen was administered. After treatment, no abnormal accumulation in both testes was observed on gallium-67 scintigraphy, and semen examination demonstrated the mild improvement of the sperm count.
Conclusion: Treatments for testicular sarcoidosis vary, and malignancy and fertility must be considered.

Key words: corticosteroid, genitourinary sarcoidosis, oligospermia, scrotal mass, testicular sarcoidosis.

Keynote message

Although testicular sarcoidosis is uncommon, malignancy and fertility status must be taken into consideration.

Introduction

Sarcoidosis is a systemic granulomatous disease of unknown cause characterized by non-caseating epithelioid granulomas, and pulmonary, lymph node, skin, and eye complications are frequent. On the other hand, genitourinary sarcoidosis is rare. Approximately 0.2% of all sarcoidosis patients and approximately 5% of autopsy cases presented genitourinary sarcoidosis.[1,2] We report a case of testicular sarcoidosis in a 32-year-old Asian male with painless bilateral scrotal swelling.

Case presentation

A 32-year-old Asian male presented with a 1-month history of painless bilateral scrotal swelling. The patient was diagnosed with lung sarcoidosis (stage II) 2 years prior to presentation to our hospital and he took no regular medications. There was no family history of sarcoidosis. His height and weight were 171 cm and 87.4 kg, respectively. The physical examination revealed painless bilateral scrotal swelling and palpable 1- to 1.5-cm testicular masses. Initial blood tests, including complete blood count, electrolytes, renal function, and C-reactive protein levels, were within the normal limits, but slight liver dysfunction due to fatty liver was observed (AST 41 U/L, ALT 71 U/L, and γ-GTP 72 U/L). Serum levels of testosterone, follicle-stimulating hormone, luteinizing hormone, and prolactin were within the normal range. Serum tumor markers, including AFP, β-HCG, and LDH, were within the normal limits. The soluble IL-2 receptor level was mildly increased to 686 U/mL (normal range, 145–519 U/L)

and the serum ACE level increased to 26 U/mL (normal range, 8.3–21.4 U/L). The rapid plasma reagin test and Treponema pallidum hemagglutination assay were negative. Tuberculosis skin tests and serology findings regarding hepatitis C and B were also negative.

Ultrasonography revealed scattered hypoechoic mass lesions in both testes (Fig. 1). A nodular lesion with an internal cavity in the left upper lobe of the lung, hilar lymph nodes, and para-aortic lymph node swelling was noted on chest and abdominal CT (Fig. 2a,b). MRI demonstrated scattered T2 low-signal areas in both testes. There was an abnormal accumulation in both testes on gallium-67 scintigraphy. We suspected testicular sarcoidosis and testicular biopsy was performed. The lesions were identified by ultrasonography, and we incised the scrotum and the tunica albuginea of the testes. There were hard nodules with clear boundaries and the lesions were easily strippable. As the pathological diagnosis during the biopsy was suspected to be sarcoidosis without malignancy, we collected some nodules and finished the biopsy. Pathologically, noncaseating epithelial cells with multinucleated giant cells, and some normal spermatogonia and spermatocytes were observed in both testes, and we diagnosed the patient with testicular sarcoidosis without any tumor cells (Fig. 3). Semen examination was performed after the biopsy, demonstrating oligospermia (2×10^6/mL).

As the patient wished to have children, we administered corticosteroids. He initially received 40 mg of oral prednisone/day, and the dose was reduced to a maintenance dose of 2.5 mg/day during the 7 months of treatment. On CT examination after treatment, the left lung nodule, hilar lymph node, and para-aortic lymph node had shrunk (Fig. 2c,d), and no abnormal accumulation in either testis was observed on gallium-67 scintigraphy. Mild improvement of the sperm count of 7×10^6/mL was noted by semen examination. We encouraged the patient to receive assisted reproductive treatments, such as intracytoplasmic sperm injection, as a treatment option for oligozoospermia, but he refused.

Discussion

Sarcoidosis is a disease of unknown etiology, characterized by the presence of noncaseating granulomas in multiple

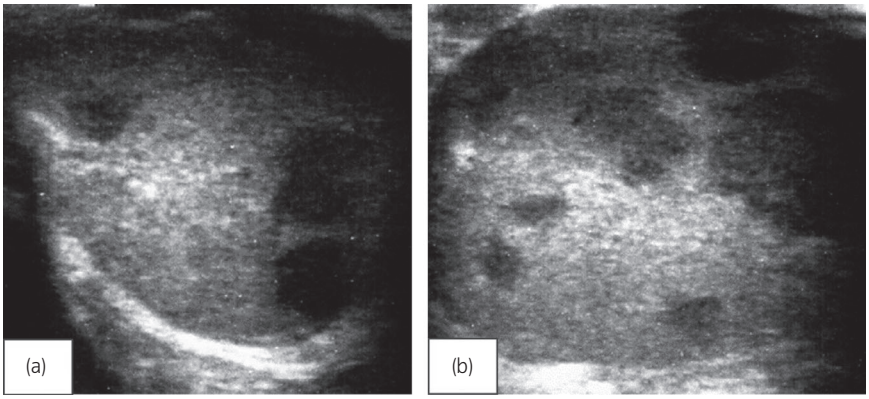

Fig. 1 Scrotal ultrasonography. Ultrasonography shows a scattered hypoechoic mass lesions in both testes (a, left testis; b, right testis).

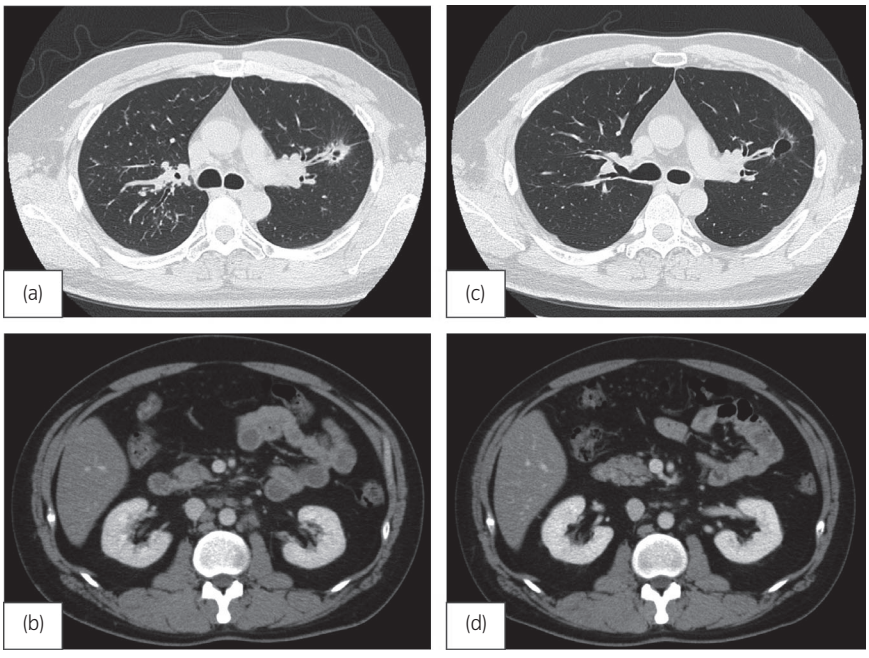

Fig. 2 CT scans of the chest and abdomen. A nodular lesion with an internal cavity in the left upper lobe of the lung (a), hilar lymph nodes, and para-aortic lymph node swelling (b) before corticosteroid therapy. A significant decrease in the size of the lung nodule (c), hilar lymph nodes, and para-aortic lymph nodes (d) after treatment.

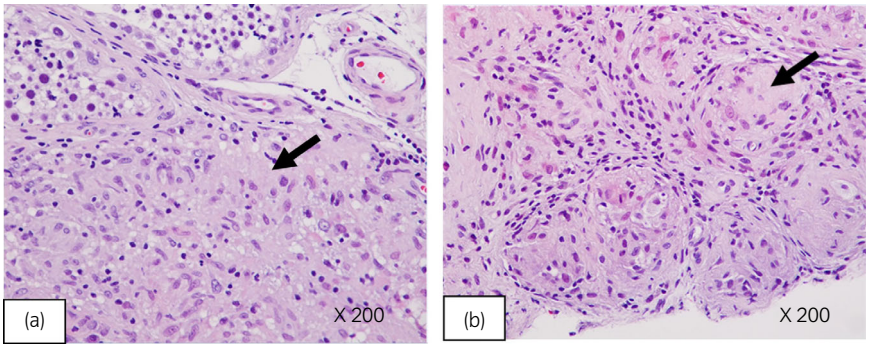

Fig. 3 Histopathology of the testicular biopsy. Noncaseating epithelial cells with multinucleated giant cells (black arrow), and some normal spermatogonia and spermatocytes (a, right testis; b, left testis; magnification ×200).

organs. Genitourinary sarcoidosis, including the epididymis and testis, accounts for less than 0.2% of all clinically diagnosed cases.[1,2] The most common age and race of patients are 20–40 years old and African-American, respectively, with Asian or Caucasian patients and those older than 40 years being less frequent.[3] Patients with testicular sarcoidosis usually present a nodular, diffuse, painless mass in the unilateral testis.[1,4] Cases of azoospermia due to testicular sarcoidosis have also been reported.[5,6]

The most important differential diagnosis for a scrotal mass is testicular malignancy. Both testicular sarcoidosis and testicular malignancy develop in patients of the same age group. In addition, a possible association between sarcoidosis and testicular malignancy has been reported.[7] There was an approximately 100-fold increase in incidence compared with that in a general population of young white men. For patients suspected of having testicular sarcoidosis, malignancy must be considered. In the diagnosis of testicular sarcoidosis, ultrasonography, MRI, and gallium-67 scintigraphy can identify solid lesions and help localize them without clarifying their nature. Serum laboratory tests for tumor markers associated with testicular malignancies (AFP, β-HCG, and LDH) and serum ACE can be helpful in different diagnosing testicular cancer and sarcoidosis. The serum ACE level is increased in 75% of untreated patients with sarcoidosis, but not in the testicular cancer patients.[8] It should be noted that patients with sarcoidosis have been reported to have mildly increased serum AFP levels due to sarcoid activity in the liver.[9]

Treatment of testicular sarcoidosis varies, and malignancy and fertility must be considered. Conservative approaches involving biopsy for diagnosis are recommended when patients present with bilateral scrotal masses or there is a strong clinical suspicion of sarcoidosis and the serum testicular tumor markers are negative. Orchiectomy is usually performed for the patients presenting with a unilateral testicular mass or those with positive tumor markers.[10] The effects of testicular sarcoidosis on fertility have not been examined, but fibrosis and occlusion of the ductus epididymis may cause azoospermia. In cases of oligospermia or azoospermia, corticosteroid therapy may improve the sperm counts through the regression of obstructive epididymal granulomas.[11] However, the amount and period of corticosteroid therapy are controversial, and there are few studies on this issue.

Conclusion

Although testicular sarcoidosis is uncommon, many factors, including the risk of malignancy, tolerance of surgery, and fertility status, should be considered in its treatment.

Conflict of interest

The authors declare no conflict of interest.

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