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Sarcoidosis is a rare cause of infertility: A case report

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ABSTRACT

In this paper, we presented a patient, who applied to our clinic because of infertility. After an intensive investigation, we diagnosed systemic sarcoidosis with testicular involvement. Urogenital sarcoidosis is a rare and insidious condition, however, it can lead to infertility. Therefore, following the diagnosis, we applied systemic steroid therapy to the patient. Within one year, the patient had a child without assisted reproductive techniques. According to our experience, in this case, we concluded that infertility without an etiology should be investigated elaborately. Furthermore, urogenital sarcoidosis should keep in mind as a rare etiology.

Introduction

Sarcoidosis is idiopathic systemic granulomatosis of unknown etiology, characterized by the formation of immune granulomas (abnormal lumps of inflammatory cells) in the affected organs.[1] Although the lungs are the most affected organs, any organ can be involved.

Testicular dysfunction causing by urogenital sarcoidosis is very rare. Only about 60 cases have been reported in the literature. It is usually characterized by epididymis and testis involvement.[2]

In this paper, we decided to present a patient with urogenital sarcoidosis as a reminder of an etiology of infertility.

Case presentation

A 31-year-old man applied to our clinic with complaints of infertility. Both testicles were found in normal shape and adequate volume in physical examination. His sperm analysis showed azoospermia. Laboratory tests, including tumor markers and hormone levels (prolactin, testosterone, estradiol), were normal. However, scrotal ultrasonography (USG) showed heterogeneity in both testicles. Therefore, following USG, we performed scrotal MRI. It showed confluent hypoechoic areas in both testicles (Fig. 1).

Since he had intermittent cough for three weeks, a chest x-ray was performed. The x-ray revealed bilateral infiltrations in lungs. We decided to perform HRCT (High-Resolution Computed Tomography) because of a suspected diagnosis of sarcoidosis. HRCT showed scattered multiple nodular opacification sites and widespread fibrotic changes in both lungs (Fig. 2). The patient immediately was consulted to pulmonary diseases clinic. They decided to perform bronchoscopy and bronchoalveolar lavage. No malignancy was detected. Cultures were sterile, and the quantiferon test was negative.

We decided to perform an open testicular biopsy rather than open lung biopsy. No tumor cells presented in frozen section analysis. According to pathology; numerous granulomas without necrosis covering the entire parenchyma of the testis were observed (Fig. 3). PAS, GMS, and EZN stains showed no specific microorganism. Immunohistochemical studies showed no staining with PLAP and CD117. Morphologic findings may be compatible with sarcoidosis or idiopathic granulomatous orchitis. After consultation with pulmonary diseases clinic, initiation of treatment for sarcoidosis was decided. The patient responded to systemic steroid therapy within six months. Lesions in the lungs and the testes resolved within three months. One year later, without the assisted reproductive techniques, the patient had a child.

Discussion

In our case, we found intra-testicular lesions while we were investigating infertility. As a well-known fact; intra-testicular lesions should be approached as malignancy unless they are proven to be benign. Generally, radical orchiectomy was performed in one-third of the cases.

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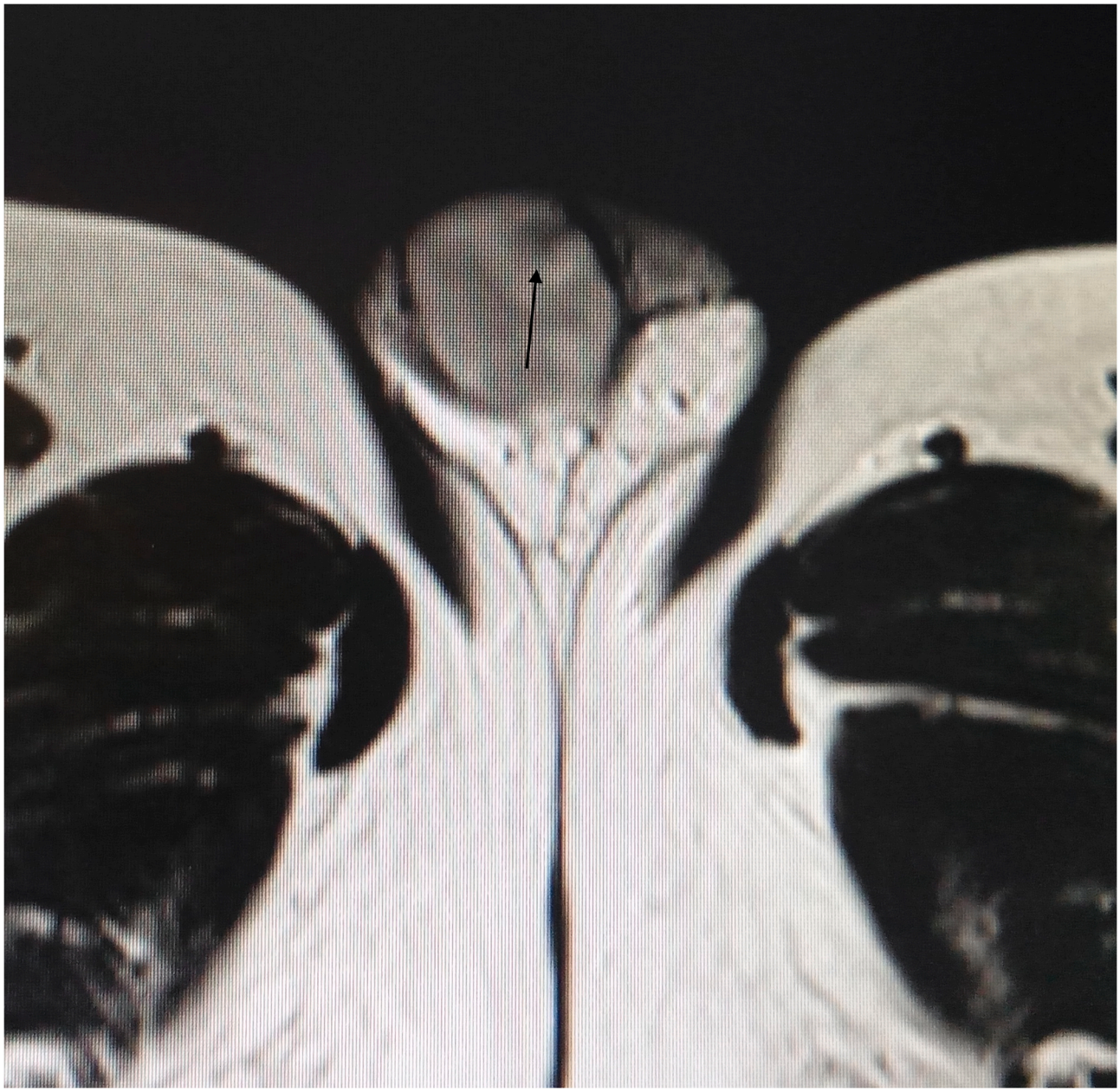

Fig. 1. MRI showed confluent hypoechoic areas in both testicles.

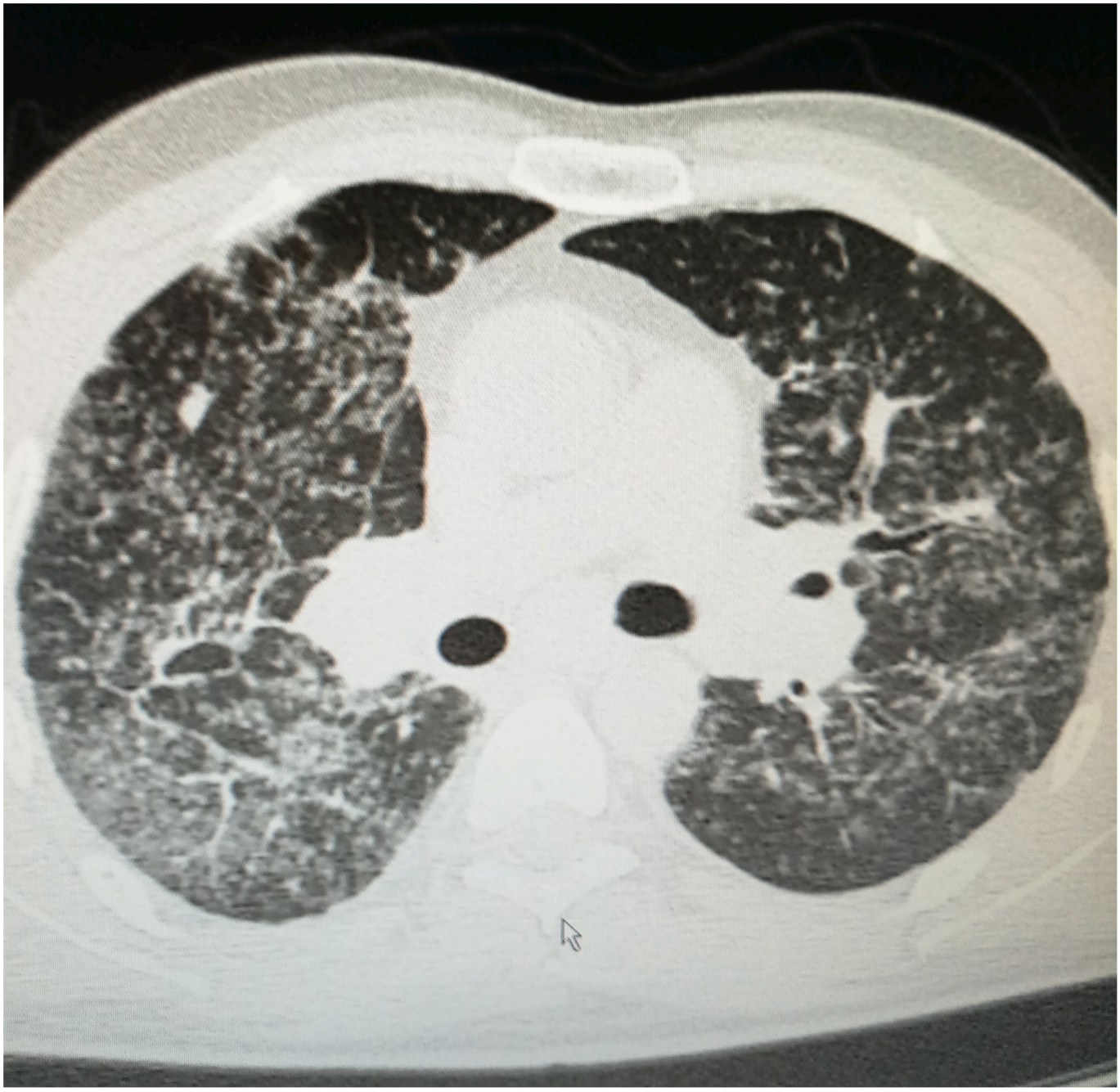

Fig. 2. HRCT showed scattered multiple nodular opacification sites and widespread fibrotic changes in both lungs.

However, bilateral heterogeneity and negative tumor markers suggest lymphoma instead of a malignancy. Moreover, bilateral testicular infarct, connective tissue diseases, and sarcoidosis should be kept in mind as non-neoplastic lesions.[3]

In general, the diagnosis of sarcoidosis based on clinical, radiological, and histopathological findings in biopsy specimens, and exclusion of all other pathologies.[4] In our case, the diagnosis made with an open testicular biopsy along with clinical and radiological findings.

Treatment of sarcoidosis depends on its severity. Most patients have spontaneous resolutions in 2 years. In severe sarcoidosis, medical treatment is needed. Immune suppressive agents are commonly used for first-line treatment. They improve radiological, symptomatic, and metabolic manifestations of the disease.[5] Because of the morphological findings, we decided to initiate oral steroid treatment.

Conclusion

Although testicular sarcoidosis is a rare condition, it may result in infertility. Therefore, clinicians should carefully investigate infertility patients with unknown etiology and systemic symptoms. After excluding the malignancies, sarcoidosis should be kept in mind in bilateral testicular infiltrations.

Compliance with ethical standards

All authors declare that he/she has no conflict of interest.

Ethical approval

All procedures performed in studies involving human participants

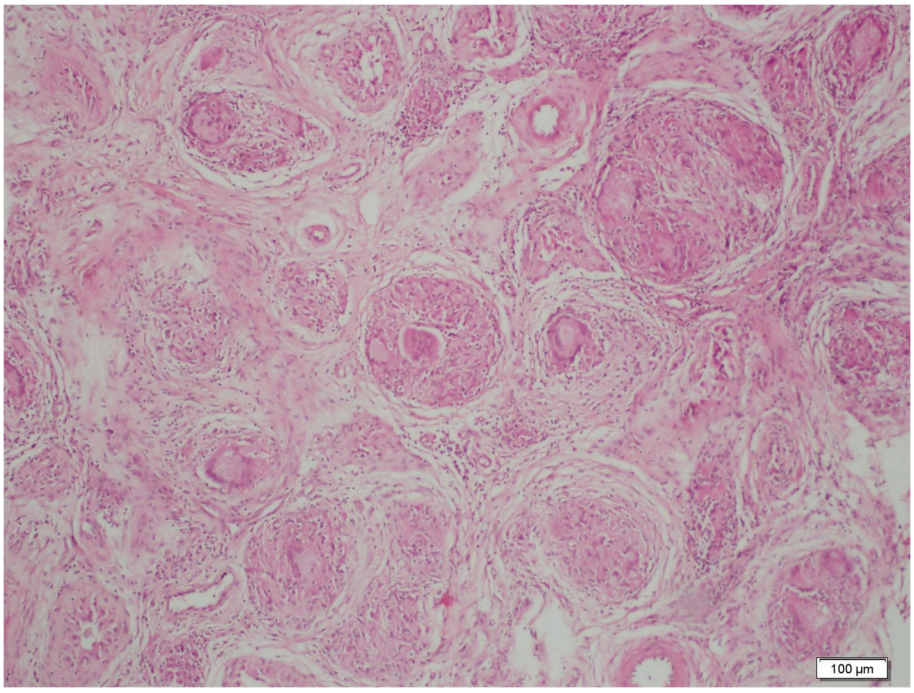

Fig. 3. Microscopic examination of small granulomas composed of epithelioid cells with scattered Langhans giant cells and lymphocytes (H&Ex100).

were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent

Informed consent was obtained from all individual participants included in the study.

Grant information

No grants submitted.

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