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CASE REPORT

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Tumor of the epididymis: an uncommon presentation of disseminated coccidioidomycosis

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ABSTRACT

Coccidioidomycosis is an endemic disease of arid regions in the Western hemisphere. Its clinical presentation varies from asymptomatic nodules on chest x-rays to disseminated disease. We present the case of a 48-year-old man with a hard and heterogeneous tumor in the posterior aspect of the right testis. Color flow doppler testicular ultrasonography was performed and two nodular masses in the tail of the right epididymis were identified. An epididymectomy was performed and histopathological examination revealed coccidioidomycosis. After diagnosis, the patient was successfully treated with fluconazol.

KEYWORDS: Epididymis. Mass. Disseminated coccidioidomycosis. Adults.

INTRODUCTION

Coccidioidomycosis is caused by the dimorphic fungus *Coccidioides*. Dimorphism is represented by two morphological forms: septate, branching hyphae with thick-walled arthroconidia in nature, and a parasitic form, spherules, in the susceptible host. Human infection occurs after inhalation of arthroconidia[1]. The disease occurs mainly in arid regions of Central and Southern California, Arizona, Western Texas, Southern New Mexico, Northern Mexico, and several desert regions of Central and South America[2]. In 75% of the cases, the primary infection passes inadvertently. Clinical presentations range from self-limited pneumonia (called "San Joaquin Valley fever") to disseminated disease. The main sites of dissemination are the skin, soft tissues and meninges; genitourinary dissemination is uncommon[3]. We describe a case of a male patient with a diagnosis of disseminated coccidioidomycosis with a painless scrotal mass.

CASE REPORT

A 48-year-old man from Northeast Nuevo Leon State presented with a history of a painless scrotal mass that progressively increased in size during the last 3 months. He denied dysuria, urethral discharge, weight loss, fever or diaphoresis. He has worked as a farmer for the last 20 years and his medical history was relevant for chronic alcohol consumption during the last 10 years (200 g/week) and a recent diagnosis of primary adrenal insufficiency treated with a daily dose of 10 mg of prednisone and fludrocortisone 0.1 mg per day in the past year. Vital signs were stable. Further examination revealed a hard and heterogeneous tumor in the posterior

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aspect of the right testis. Inguinal lymphadenopathy, local hyperthermia, skin lesions or color change were not present. Laboratory results showed a hemoglobin concentration of 13.1 g/dL, leukocytes 8.09 x10^9/L, platelets 199 x10^9/L, glucose 96 mg/dL, sodium 138 mmol/L, chlorine 100 mmol/L, potassium 4 mmol/L, calcium 8.4 mg/dL, urea nitrogen 16 mg/dL, creatinine 1.0 mg/dL, C-reactive protein 1 mg/L, alanine aminotransferase 36 UI/L, aspartate aminotransferase 30 UI/L, total bilirubin 0.8 mg/dL and albumin 3.8 g/dL. Color flow doppler testicular ultrasonography was performed and two nodular masses in the tail of the right epididymis with increased vascular flow were found (Figure 1). An epididymectomy was performed and histologic examination revealed a granulomatous epididymitis with scattered coccidioidal spherules present within the granulomas (Figure 2). The diagnosis of disseminated coccidioidomycosis was made and systemic antifungal therapy was initiated with fluconazole 400 mg once daily for 12 months. As the diagnosis of disseminated coccidioidomycosis usually relies on the histopathological identification, further serological tests or culture were not performed. Alterations in the lungs compatible with current pulmonary coccidioidomycosis were not found on the chest CT-scan. Involvement of other organs was systematically excluded in our patient as well, however, further evaluation for adrenal insufficiency will be carried out after discharge.

DISCUSSION

Coccidioidomycosis is an infection caused by the soil-dwelling dimorphic fungi *Coccidioides* spp. The endemic regions in Mexico are characterized by a dry climate, alkaline soil, summers with high temperatures, and low annual precipitation. This combination of environmental factors facilitates the spread of *Coccidioides* spores in the air. Coccidioidomycosis was a reportable disease in Mexico up to 1994 with an average of 1,500 cases reported each year. Since 1995, there are no data on the clinical burden of coccidioidomycosis[4]. The highest number of clinical cases are located in the Northern States on the US-Mexico border (Nuevo Leon, Tamaulipas, Chihuahua, Baja California, and Sonora)[5].

Clinical presentation varies among patients from asymptomatic nodules on chest x-rays to life-threatening disease. Typically, coccidioidomycosis presents as a community-acquired pneumonia, but in 1% of patients, coccidioides can affect any other organ or tissue; this last condition is known as disseminated or extrapulmonary

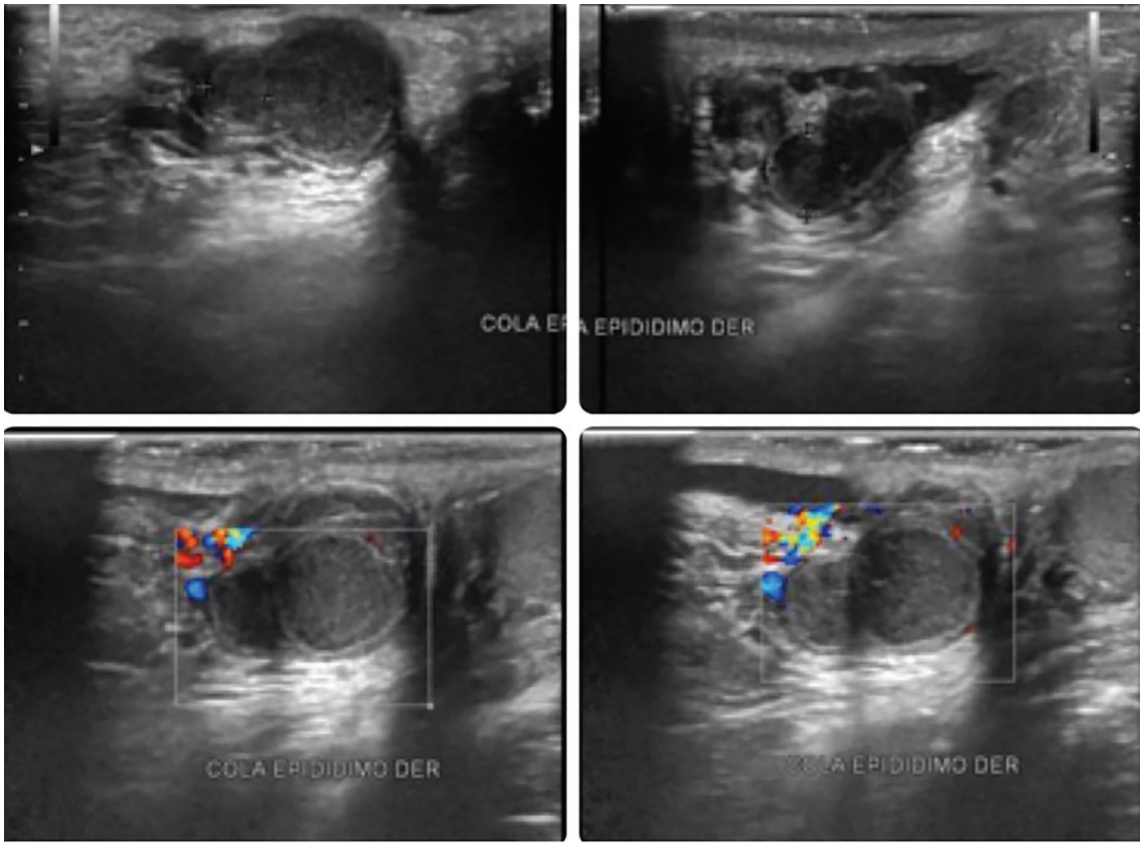

Figure 1 - Tumor of the epididymis due to disseminated coccidioidomycosis. Color flow doppler testicular ultrasonography revealed the presence of two nodular masses in the tail of the right epididymis with increased vascular Flow.

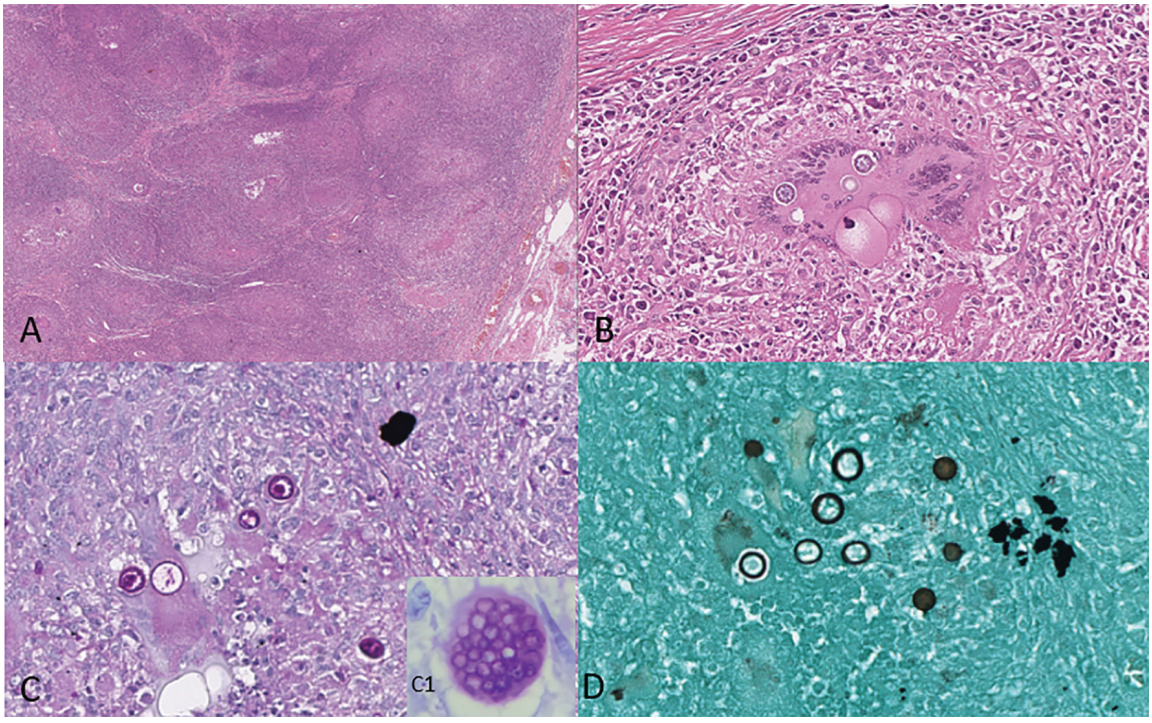

Figure 2 - Coccidioidal epididymitis in a 48-year-old man. Diffuse epididimitis with multiple granulomas observed in light microscopy and at low magnification (5 x) (A), at high magnification (B), several fungal structures with a round shape, double wall, and smaller oval structures inside (endospores) can be observed along with giant multinucleated cells. PAS (C) and Gromori-Grocott (D) (40 x) staining highlighting the spherules and endospores. In oil inmersion (C1, 100 x) the endospores can be individually distinguished.

coccidioidomycosis. Remarkably, 99% of patients with a disseminated form of the disease are immunocompromised[6]. The main risk factors are corticosteroid therapy, organ transplantation, hematogenous malignancy, chronic renal disease, chronic liver disease and pregnancy[3]. Our patient had a previous diagnosis of primary adrenal insufficiency and chronic systemic steroid consumption was documented. Although extremely rare, *Coccidioides* has involved the adrenals in necropsy reviews of disseminated coccidioidomycosis[7]. However, to the best of our knowledge, only three cases of adrenal insufficiency due to this fungal infection have been reported in the literature[8-10]. Adrenal function in patients with disseminated coccidioidomycosis has not been evaluated and it is not clear if an entity such as subclinical adrenal insufficiency exists in this population. Further studies are needed, in order to establish the best diagnostic and treatment approach of this group. The etiology of adrenal insufficiency in our patient was not well documented before consultation and this will be reevaluated after completion of treatment.

Alcohol is one of the most used drugs worldwide and it is well-known that it affects the immune system, with even moderate amounts influencing immune responses. Although alcohol can alter the function of all cell populations involved in the innate and adaptive immune responses, the effect in many cases is a subclinical immunosuppression that becomes clinically relevant only after a secondary insult[11].

Excessive alcohol consumption was identified in our patient, interestingly this has been recently associated with an increased risk of tuberculosis in a metanalysis[12], however, tuberculosis has not been evaluated in this patient.

In a previous study, Adam *et al.*[3] retrospectively reviewed 150 cases of extrapulmonary coccidioidomycosis and found that visceral organ involvement, as occurred in our patient, was the rarest presentation. Our patient only reported a painless epididymal tumor, which represents an atypical presentation of an unusual form of disseminated coccidioidomycosis.

Since the introduction of first-generation triazoles, current guidelines suggest that treatment of patients with extrapulmonary soft tissue coccidioidomycosis (not associated with bone infection) should consist of daily doses of fluconazole or itraconazole[1]. Patients with coccidioidomycosis of the epididymis can receive a combined strategy of medical and surgical procedures for the involved organ, but good responses with surgical resection or medical treatment alone have also been documented in specific populations[13]. To the best of our knowledge, no randomized controlled studies studies have evaluated and compared outcomes between these management strategies, in this respect, a strong evidence-based treatment recommendation cannot be made.

Coccidioidomycosis of the epididymis is not common. The diagnosis should be suspected mainly in immunocompromised patients who have signs or symptoms of epididymitis and a history of traveling to or living in an endemic area. The best treatment strategy of isolated genital tract disease has not been well defined and most clinical decisions are based on reported experiences. Future evidence-based treatment strategies for this unusual presentation should be documented.

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CONFLICT OF INTERESTS

The authors declare that they have no conflict of interests.

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