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Case Report

Ocular Involvement as the Only Manifestation in Paracoccidioidomycosis

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Abstract

Introduction: Paracoccidioidomycosis is a systemic infection caused by Paracoccidioides Brasiliensis fungus that primarily affects the lungs. Ocular involvement is rare, in most cases having a secondary manifestation typically found in the eyelid and conjunctiva.

Patient and Clinical Findings: We reported a case of paracoccidioidomycosis in a female patient with a localized ocular manifestation.

Conclusions: This case shows the importance of including paracoccidioidomycosis as a differential diagnostic hypothesis in patients with only ocular manifestations that display tumors characterized by eyelid thickening.

Keywords: Conjunctival Infection; Conjunctival Melting; Paracoccidiodomycosis; Scleral Melting; Systemic Disease; *Paracoccidioides brasiliensis*

Abbreviations

CT: Computerized Tomography; PCM: Paracococcidioidomycosis

Introduction

Paracoccidioidomycosis is a granulomatous systemic infection that mainly affects the lungs, mononuclear phagocytic system, mucosa, skin, suprarenal glands, and can be disseminated through a hematogenous route (5.8). First described in 1908 by Adolfo Lutz, this disease also known as blastomycosis in South America, received the official term paracoccidioidomycosis in 1971 at the Symposium of Medellín [3]. It is caused by *Paracoccidioides Brasiliensis*, a thermos dependent dimorphic fungus that grows at ambient temperature in the soil as a permanent saprob in the form of mycelium, an infective form that can be inhaled, and at 35-37ºC it transforms into yeast, a pathogenic form responsible for causing lesions in the tissues of the host [3,8,9]. This infection affects more men than women (15:1) between 30 and 50 years old and can recur as a latent infection and is related to rural activity, in addition to being associated with smoking and alcoholism (3.9).

The disease is considered endemic in Latin America, occurring more frequently in countries of the South America, with the highest incidence in Brazil, a country in which the infection is the eighth leading cause of death among chronic or recurrent parasitic and infectious diseases [4,6,8]. The states of Sao Paulo, Rio de Janeiro and Minas Gerais have a higher incidence [3]. Ocular involvement is rare and, although some cases have had retinal and uveal involvement, cases normally involve the eyelid and conjunctiva [12,13]. For 80 years after the first report of paracoccidioidomycosis in 1908, only 44 cases with ocular involvement were described, most of them in Brazil [1]. Ocular involvement is most often a secondary

manifestation in the multifocal disease or, when described as a primary location, it is associated with systemic symptoms (fever and weight loss) [1,2,7].

Case Report

A 86 years old woman, housewife, born in Nipoa, coming from Poloni, resident of São José do Rio Preto in the interior of São Paulo, with no noteworthy medical history other than controlled hypertension. She was referred to a corneal specialist with a complaint of hyperemia for the past 2 months along with pain and secretion in the left eye for the past 15 days.

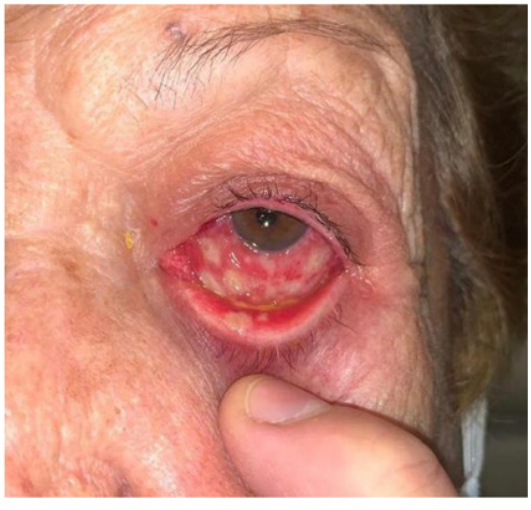

Figure 1

She had already been treated by another ophthalmologist who made the diagnosis of hordeolum and performed an unsuccessful drainage. Vigadexa and cylocort were used and there was no indication of improvement. Encountered during the physical examination were conjunctival hyperemia, bulbar edema, superior temporal conjunctival chemosis, ulcerated granulomas in the sclera, upper and lower eyelids and upper and lower tarsal conjunctiva lower eyelid, in addition to a diffuse infiltrate in the lower eyelid. There was no evidence of adenomegaly. The thickening found in the left lower eyelid was suggestive of a tumor, in view of this, a biopsy was performed to confirm the diagnostic hypothesis.

The sample taken from the bulbar conjunctiva was negative for signs of malignancy and the morphological diagnosis indicated

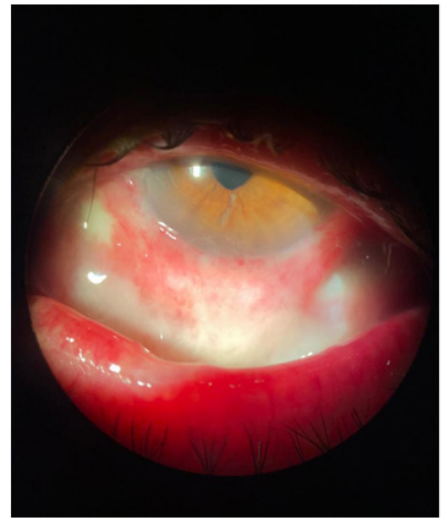

Figure 2

fungal conjunctivitis, where the fungal morphology was Paracoccidioidomycosis.

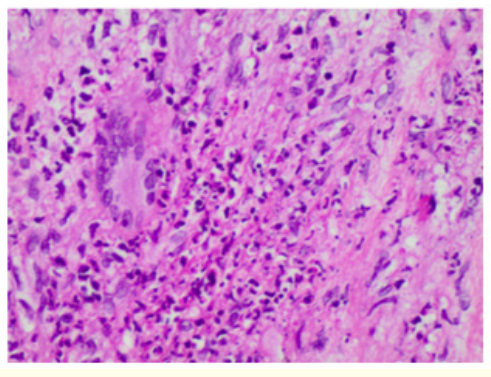

Figure 3

Therefore, treatment with oral itraconazole twice a day was started indefinitely. Due to non-adherence to medication, the patient worsened, returning with necrosis of the sclera and conjunctiva, associated with worsening of the lower eyelid thickening and increase in scleral lesions in the left eye. With correct adherence to treatment, there was an improvement with reduction of granulomas and healing. Subsequently, an orbital tomography was requested, which showed a slight swelling of the soft parts of the left

eyelid region, with increased contrast enhancement of the anterior aspect of the eyeball in the corneal topography on the left, which may be associated with an inflammatory or infectious process. The patient followed up with an infectious disease specialist who requested other tests such as serology for paracoccidioidomycosis, complete blood count, urea and creatinine dosage, serology for varicella zoster, abdominal ultraound, chest CT scan, paranasal sinuses CT scan and serology for toxoplasmosis. The exams, with the exception of the chest CT, showed no change, indicating preserved renal function despite long-term treatment and an absence of paracoccidioidomycosis antibodies. Computed tomography of the chest showed a solitary calcified micronodule with residual granuloma appearance in the lower lobe of the left lung, which could not be associated with infection and was too small to be punctured. Visual acuity (20/30) remained unchanged since the beginning of the patients history. Finally, upon referral to retinal specialists, a full retinal exam was performed and there were no alterations.

Discussion

PCM can be acute (juvenile) or chronic (adult). The former is less frequent (20% cases) affecting mainly children, adolescents and young adults and is characterized by fever, loss of weight, hepatomegaly, splenomegaly, adenomegaly and rarely involving the lungs. The latter represents 80% of cases which have a slow progression and can take years to develop, occurring between the ages of 30 and 50 in rural workers, affecting lungs, mucous membranes, lymph nodes, adrenals, central nervous system and eyes, among others organs (5.11). Despite affecting individuals between 30 and 50 years of age, the infection is acquired in the first two decades of life, affecting mainly men who are currently or have already been involved in some rural activity related to soil management [3,6,9]. Infection may also be associated with trauma caused by infected material (soil, dust, wood) [15,16]. It is believed that the lower prevalence in females is linked the suppression of the transformation of the mycelium into yeast by estrogen, so women would be protected from the manifesting of the disease but not from infections [6]. Ocular involvement can be clinically and histopathologically similar to carcinoma, so the anatomopathological examination should be performed to confirm the diagnosis.

Conclusion

The incidence of infection in women has rarely been described in the literature to date, which, in addition to making it difficult to diagnosis, raises doubts about the origin of the infection. Additionally, the absence of associated highlights the rarity of the case, since all cases of ocular involvement presented in the literature are secondary to other clinical manifestations. Ocular involvement in paracoccidioidomycosis is mostly characterized by involvement of the eyelid or the conjunctiva and in a few cases involvement of the retina has been described. The peculiarity of the case allows us to conclude that the infection by paracoccidioidomycosis must be considered in women, even if they do not have other systemic manifestations and are unrelated to rural activities.

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