

## Case Report: Conjunctival Rhinosporidiosis Associated to Scleral Ectasia

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### Abstract

Rhinosporidiosis is a rare fungal infection, with eye involvement in 10% of cases. The etiological agent is *Rhinosporidium seeberi*, from the mezomycetozoa class. It presents with diverse morphology including elevated lesions and polypoid lesions, with characteristic of non-granulomatous inflammation. Concomitant scleral ectasia may occur in rare episodes. This report presents a case of scleromalacia associated with bulbar conjunctival rhinosporidiosis in a young patient with a history of ocular trauma and therapeutic options.

**Keywords:** Rhinosporidiosis; Infection; Polyp; Conjunctiva; Therapy; Scleromalacia

### Abbreviations

LVA: Low Visual Acuity; RE: Right Eye; LE: Left Eye; IOP: Intraocular Pressure; RO: Rondônia

### Introduction

Rhinosporidiosis is an infection produced by *Rhinosporidium seeberi* and is characterized by elevated lesions, resembling polyps with white dots of varying size in the mucosa [1]. It has been classified using molecular biological tools in the Mesomycetozoa clade, with genetic heterogeneity demonstrated in both human and animals strains with several mechanisms of immune evasion [2].

Epidemiology is not well understood and appears to be an endemic infection of tropical climate mainly in India and Sri Lanka. Also, more than 90% of the affected patients report having attended ponds or lagoons [3]. It can affect throat, ear nose and genitalia [2,4].

It appears to be three times more frequent in men [3], affecting the nasal mucosa, and less frequently the eye. When in this site, it is called oculosporidiosis and represents about 10% of the cases of the disease [3,5]. The conjunctiva and lacrimal sac are the preferred sites in the eye, with less involvement of the sclera and lacrimal pathways [3,4]. There have been 13 cases of scleral ectasia associated with bulbar conjunctival involvement described in the literature [5].

The first case of rhinosporidiosis was described in 1900 in a 19-year-old rural worker with nasal cavity injury [6]. In Brazil few cases are described, being this one of the first in the state of Minas Gerais.

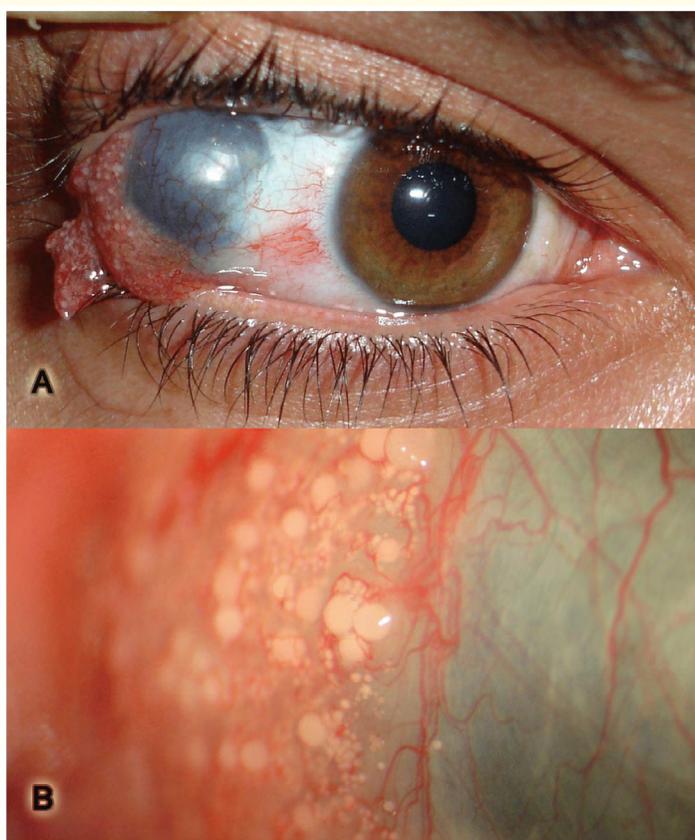
### Objective of the Study

The objective of this study is to describe a rare case of oculosporidiosis with scleral involvement and to spread the knowledge of this nosological entity.

## Case Report

A 17 years old white male, resident of Buritis-Rondônia (RO), came to the ophthalmology urgency unit of the Santa Casa de Misericórdia in Belo Horizonte, with a complaint of “growing bruinsing” for the past 6 months. He reported trauma with an air gun 6 months before and since then noticed a growing eye injury, accompanied by insidious discomfort and low visual acuity (LVA). He denied previous eye injuries, visual deficits or comorbidities and had the habit of bathing in ponds. Visual acuity of 20/25 in right eye (RE) and 20/20 in left eye (LE) with pinhole. He already had exams performed in his city: Fluorescein Angiography and Optical Coherence Tomography without pathological findings, sorology for infectious and rheumatological diseases as well as blood tests without abnormalities.

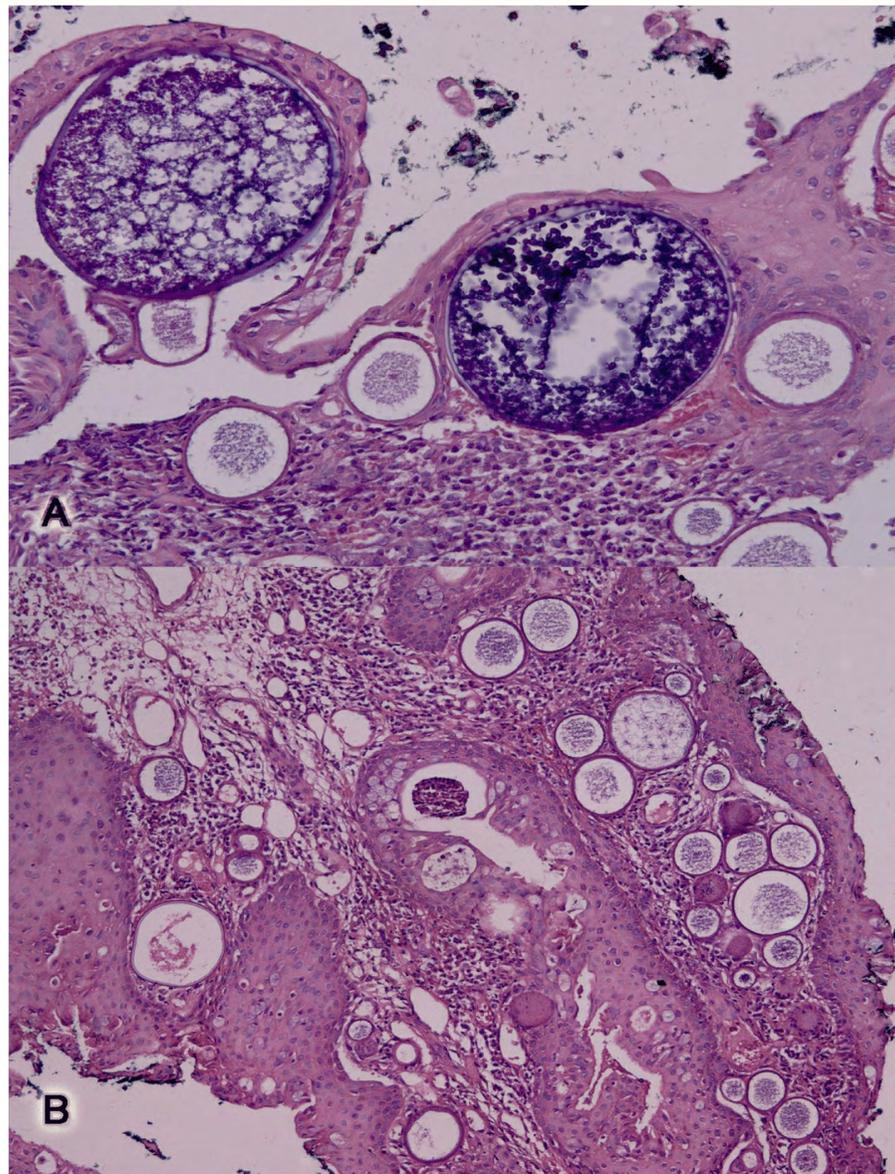
Biomicroscopy showed a nodular lesion with important scleral thinning, with a blackish coloration, about 1.2 x 1.0 cm, and a vascularized polyploidy lesion revealing content with multiple yellow and white micropoints (Figure 1). He had transparent corneas and lens, pupils were isochoric and photoreagent. Intraocular pressure (IOP): 12 and 13 mmHg, macules with physiological brightness, nerves with precise limits and physiological digging.



**Figure 1:** A: Patient with polyploidy lesion with important scleral thinning; B: Reddish-pink, pedunculated, mobile, richly vascularized lesions.

Complete excision of the lesion was performed followed by scleral recoating from donor scleral graft in order to strengthen the exposed ocular wall. The scleral thinning altered the insertion of the rectus lateralis muscle. During surgery he received one intravenous dose of profiatic antibiotic (cefazolin) and then used seven days of topic antibiotic (moxifloxacin). After ninety postoperative days, it evolved with optimal adaptation of the scleral flap and without signs of recurrent conjunctival lesions.

The pathology of the lesion revealed a non-granulomatous lympho-histiocytic inflammatory infiltrate with edema and numerous fungal structures such as sporangia, endospores and spores already free (Figure 2).



**Figure 2:** A - Histopathological exams in higher magnification showing endospores and spores already free. B- Histopathological exams at lower magnification showing non-granulomatous lympho-histiocytic inflammatory infiltrate with edema and numerous fungal structures.

## Discussion

This study reports one of the earliest cases of oculosporidiosis with scleral involvement in Brazil. This disease should be suspected whenever a young patient comes from an endemic area or has conjunctival polyps and/or chronic dacryocystitis. There are reports of the disease in patients from Espírito Santo and Goiás making this entity important to ophthalmologists, because when diagnosed and treated promptly, it avoids potentially serious sequelae [4,6].

This chronic infection seems to have a male predilection of 3:1 and individuals between the second and third decades of life. Oculosporidiosis is closely related to water; as more than 90% of those infected report bathing in ponds or rivers, which is why it is suspected of aquatic transmission by direct contact of the parasite with the mucous membranes [3]. It is also believed that the transmission can occur due to exposure to dust from the fields and consequent inhalation of the pathogen [4].

The conjunctiva is the ocular focus of major involvement, followed by the tear sac, eyelids and sclera. In the literature, more than 90% of the reported cases refer to conjunctival involvement, another 9% of the lacrimal pathways and rarely the sclera. It is likely that scleral involvement occur due to enzymatic substances from the body corroding it [5]. The exclusion of other causes of scleral melting of non-infectious and infectious origin should always be performed [6]. The differential diagnosis of conjunctival polypoid lesions includes pyogenic granuloma, epitheliomas, papillomas, angiomas, angiofibromas, paracoccidioidomycosis, cryptococcosis, aspergillosis and, more rarely, amyloidosis [4]. Serologic tests should be performed in order to exclude non-infectious causes of scleromalacia such as scleromalacia perforans. For the ophthalmologist the examination in the slit lamp facilitates the diagnosis due to the typical morphology of the lesions being described as pathognomonic of the rhinosporidium, even with different presentations and sites such as flattened, polypoid, arising from the fornices or the bulbar conjunctiva itself [7].

The standard treatment is surgical excision. The literature presents results with very low recurrence rates in conjunctival involvement and higher indexes when the lacrimal sac is under study. It is believed that the highest recurrence rate is not caused by rhinosporidium tropism through the lacrimal sac, but because of the technical difficulty in removing the tissue affected by the pathogen in its entirety. In addition to surgical excision, cauterization with 2% silver nitrate is controversial, as well as the use of amphotericin B and 5% Povidone Iodine [1,8-11]. Corneal scleral transplant is described for the treatment of staphylomas, however, there are case reports of treatment with excision and cauterization of the base followed by cryotherapy, which evolved with complete resolution of the staphyloma without the costs of the transplants [5]. The use of dapsone 100 mg once or twice daily for three to six months seems to reduce the relapse rate by delaying spore maturation and promoting fibrosis of the lesions [7].

## Conclusion

Rhinosporidiosis is a rare disease and ocular involvement accounts for 10% of cases. The epidemiology and the form of contamination have not yet been clarified. Hence, the morphology of the lesion is fairly typical and easily diagnosed on biomicroscopic examination, provided that the attending physician is familiar with the diagnosis. Treatment is usually surgical, presenting very low recurrence rates when the lesion is completely excised.

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