# Parinaud's oculoglandular syndrome and possibly causing cortical cataract

Síndrome oculoglandular de Parinaud como possível causa de catarata cortical

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# Abstract

According to the World Health Organization, cataract is the leading cause of blindness and visual impairment throughout the world. However, the etiology of cataracts often remains unknown. This report describes the development of cortical cataract in a patient after Parinaud's oculoglandular syndrome caused by the fungus Sporothrix schenckii.

Keywords: Conjunctivitis; Cataract/etiology; Sporothrix; Ocular motility disorders/complications; Case reports

# Resumo

De acordo com a Organização Mundial de Saúde, a catarata é a principal causa de cegueira e deficiência visual em todo o mundo. No entanto, a etiologia das cataratas frequentemente permanece desconhecida. Este relato descreve o desenvolvimento de catarata cortical em um paciente após Síndrome oculoglandular de Parinaud causada pelo fungo Sporothrix schenckii. **Descritores:** Conjuntivite; Catarata/etiologia; Sporothrix; Transtornos da motilidade ocular/complicações; Relatos de casos

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### **INTRODUCTION**

ccording to the World Health Organization, cataract is the leading cause of blindness and visual impairment throughout the world<sup>(1)</sup>. It is often associated with aging, but may be caused by endocrinological disease, trauma, drug therapy or infections, or be of idiopathic etiology<sup>(1)</sup>. Infectionrelated lens opacification may be due to the microorganism itself or, most commonly, secondary to ocular immune inflammatory response or corticotheraphy side effects<sup>(1)</sup>.

Cataract following virus aggression is well established in the literature<sup>(1)</sup>. However, this is to our knowledge the first report of cataract probably caused by conjuntival Sporothrix schenckii infection.

In this study, we report the clinical aspects of the first documented case of cortical cataract following Parinaud's oculoglandular syndrome secondary to infection with S. schenckii. The epidemiological aspects, diagnostic methods and drug therapy of this rare disease are also discussed.

#### **Case report**

MSG, 59, diabetic, born and raised in Rio de Janeiro, RJ, was seen at Hospital das Clínicas in Vitória (a state capital of Southeastern Brazil) in december 2009 due to conjunctival hyperemia in the left eye (OS) with 20 days of evolution. The patient reported previous onset of painless nodule-postular lesions in both upper limbs: five lesions along the lymphatic chain on the left side and one on the right side. On occasion of the first examination at our service, one of the lesions on the left side was still perceptible but the others had scarred. Painful preauricular, cervical and supraclavicular lymphadenopathy was also observed ipsilaterally to the ocular conjunctivitis.

When questioned, the patient reported having suffered scratches and bites from a stray cat adopted a few weeks earlier. The animal, according to the patient, appeared sick and had multiple skin tumors.

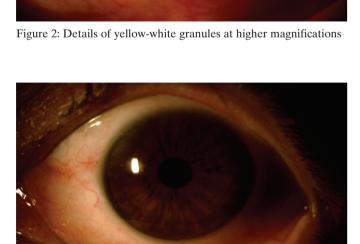
The patient also mentioned being treated by four different ophthalmologists over the course of the disease. Each one prescribed seven-day courses of one of these drugs: amoxicillin, erythromycin, vigamox<sup>®</sup> (0.5% moxifloxacin) or fenidex<sup>®</sup> (0.05mg dexamethasone + 5mg chloramphenicol + 0.25mg tetrahydrozoline). However, no clinical improvement was observed.

The best-corrected visual acuity (VA) was 20/20 in the right eye (OD) and 20/50 in the OS. The examination was hampered by constant epiphora. The OD was normal on biomicroscopy. The OS presented mucopurulent discharge and intense chemosis associated with the presence of diffuse yellow-white granules in the bulbar conjunctiva (figures 1 and 2). There were no corneal or lens findings and no anterior chamber reaction. The intraocular pressure was 10mmHg in both eyes.

The diagnosis of S. schenckii was established based on a conjunctival swab collected from the affected eye and submitted to histopathological analysis and growth in specific culture. It was then introduced specific therapy with administration of 6mL 25% potassium iodide solution every 12 hours at the Department of Infectious Diseases, but turned out to be allergic to iodine. As a result, the patient was prescribed itraconazole at 100mg/day for 60 days. This was extended for another 30 days in order to achieve a satisfactory therapeutic response, with complete resolution of the eye infection (figure 3) and of the lesions in the upper limbs.

granules in the bulbar conjunctiva; note the presence of secretion in the conjunctival fornix and the absence of cataract





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After 3 months of follow-up and coinciding with the end of therapy, the patient complained of reduced VA in the OS. Upon examination, a small anterior cortical cataract was observed, with loss of contrast and a best-corrected VA of 20/50. In 2011, the patient reported an even greater loss of VA in the OS, with more intense and diffuse cortical cataracts. However, for family reasons, the patient decided to postpone surgery. A year later, the patient returned and requested surgical treatment of the affected eye. The best-corrected VA was 20/25 in the OD and 20/70 in the OS. Incipient nuclear sclerosis was seen in the OD on biomicroscopy, while the OS presented a diffuse cortical cataract (nuclear 2+/4+ and posterior subcapsular 1-2+/4+). Up to this point, no anterior chamber reaction or any other inflammatory sign had been observed in the OS.

The patient then underwent phacoemulsification with intraocular lens implantation without complications. The anterior capsule and lens fragments extracted were submitted to histopathological analysis. Despite thorough analysis, no traces of fungus were found in the sample.

## DISCUSSION

Sporotrichosis is caused by the dimorphic fungus *S. schenckii*. Though distributed worldwide, is considered endemic in many tropical and subtropical regions<sup>(2,3)</sup> and is the most common subcutaneous mycosis in Latin America<sup>(4,5)</sup>. Once considered a rural disease, itis increasingly common in cities, largely due to feline epizootics<sup>(4,6)</sup>.

The infection is usually acquired through inoculation of the fungus following skin trauma by contaminated plants<sup>(7)</sup>. However, a zoonotic form has been spreading in recent decades, transmitted mostly by cats. Thus, since the late 1990s, over 2000 cases in humans and 3000 cases in animals have been diagnosed in the state of Rio de Janeiro alone<sup>(6)</sup>.

Although patients with sporotrichosis often present significant skin changes, the disease usually follows a relatively mild course without affecting organs other than the skin, the mucosa and subcutaneous tissue<sup>(6)</sup>. The lymphocutaneous form is the most common presentation<sup>(3,8,9)</sup>. It begins with a nodular or ulcerated lesion at the site of inoculation of the fungus and then may follow a trajectory characterized by regional lymphatic infarcts subsequent to the lymph nodes, marked clinically by nodular lesions which ulcerate and drain. This spectrum is known as ascending lymphangitis<sup>(3,8)</sup>. The disease is not a difficult to treat, but is associated with indirect social costs due to work absenteeism, distress during the active stage and the unsightly appearance of the scars.

In rare cases, the fungus may have the conjunctiva as the main site of attack<sup>(3)</sup>, causing a unilateral granulomatous conjunctivitis with ipsilateral cervical lymphadenopathy, known as Parinaud's oculoglandular syndrome (POS)<sup>(10)</sup>. Most eye infections caused by sporotrichosis involve both the conjunctiva and the periorbital tissues<sup>(3,11)</sup>, such as orbital and palpebral abscess. However, suppurative chronic dacryocystitis, scleritis, iridocyclitis, retinochoroiditis and even endophthalmitishave been described<sup>(9)</sup>. Disseminated infection and local trauma are believed to contribute to eye infection<sup>(3,12,13)</sup>.

Conjunctival disease is characterized by nodular lesions which are occasionally ulcerated and accompanied by ipsilateral preauricular, submandibular or cervical satellite lymphadenopathy. In addition, fever, purulent discharge and follicular conjunctival reaction may be present<sup>(10,14)</sup>. The disease progresses with ocular manifestations ranging from mild to very severe<sup>(10)</sup>. Conjunctival involvement is rarely bilateral<sup>(10)</sup>.

POS may have several etiologies, but the most common is bartonellosis (cat scratch disease) caused by the bacterium *Bartonellahanselae*<sup>(14)</sup>. As the name suggests, the disease occurs after a cat bite or scratch and is characterized by fever and lymphadenopathy<sup>(14)</sup>. Considering the epidemiological profile of our patient, a differential diagnosis was imperative. The diagnosis was confirmed by the detection of *S. schenckii* in material from the conjunctiva submitted to histopathological analysis and grown in specific culture following standard procedures<sup>(6)</sup>.

The appearance of unilateral cortical cataract shortly after the occurrence of POS in an eye with previouslytransparent crystalline raises the hypothesis of a causal relationship with the fungus *S. schenckii*. Changes in the crystalline occur in cases of chronic uveitis or associated with steroid therapy. However, these factors were not observed in our case since the period of use of corticosteroid usewas short andno anterior chamber reaction was detected throughout the entire period of observation. If uveitis occurred at some point between consultations, it must have been short-lived and mild.

It should be mentioned that uveitis secondary to ocular sporotrichosis, although rare, has been associated with exuberant iridocyclitis and unfavorable outcomes, including endophthalmitis and eye loss<sup>(9,11)</sup>. It commonly occurs after hematogenous spread from systemic or ocular trauma injuries<sup>(11)</sup>.

Since the opacification of the crystalline occurred only in the previously infected eye and it could not be explained by any inflammatory process, fungal aggression is a plausible etiology. This is supported by the finding by Vieira-Dias et al.<sup>(3)</sup> of *S. schenckii* in the aqueous humor of a 12-year old girl with signs of ocular and cutaneous sporotrichosis. Likewise, Benz et al.<sup>(15)</sup> isolated the microsporidian *Encephalitozoon cuniculi* from samples of aqueous humor and crystalline after their extraction, implicating it as the agent responsible for the development of cortical or mature cataracts in cats. The absence of previous uveitis in 5 of the 19 eyes in their study strengthens the hypothesis of direct fungal aggression and supports our findings.

According to a paper published in 2004 by Vasavada et al.<sup>(16)</sup> the etiology of cataract is predominantly idiopathic. Atopy is the most common comorbidity or risk factor. The lack of knowledge of etiologic factors has important implications for the prevalence of both senile and congenital/infantile cataracts and, consequently, for public health care.

# CONCLUSION

Research efforts should concentrate on the search for the causal factors of cataracts and on the development of preventive strategies and targeted therapeutic approaches. In our study is not possible to ascertain the causal relationship between the development of cataracts and the Parinaud's Oculoglandular Syndrome, but it is tempting to speculate this association considering the clinical evolution of our case. Cataracts classified as idiopathic may be caused by a misdiagnosed infection.

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