



(CASE REPORT)



Parinaud's oculo-glandular syndrome secondary to sporotrichosis

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Abstract

Parinaud's oculoglandular syndrome (PCOS) is a rare eye disease caused by different etiological agents, including bacteria, viruses and fungi. PCOS is a special form of granulomatous conjunctivitis and is often associated with cat scratch disease, herpes simplex infection, sporotrichosis, blastomycosis, and coccidioidomycosis. Sporotrichosis is a subcutaneous mycosis caused by the fungus *Sporothrix schenckii* with global distribution and, in general, patients are treated on an outpatient basis. Since 1998, there has been an increase in cases in the State of Rio de Janeiro, Brazil, especially due to zoonotic transmission involving cats. PCOS is characterized by granulomatous conjunctivitis accompanied by adjacent pre-auricular lymphadenopathy and systemic symptoms such as sweating, feeling sick, and fever. Complementary tests including culture confirm the diagnosis of sporotrichosis. Its classic clinical presentation should always be remembered by specialists so that the correct diagnosis and treatment with antifungal agents is carried out early, so that the condition can be resolved without sequelae for the patients.

Keywords: Sporotrichosis; Parinaud's Oculo-Glandular Syndrome; Zoonoses; Granulomatous Conjunctivitis; Differential Diagnosis

1. Introduction

Parinaud's oculoglandular syndrome (PCOS) is considered a rare condition. Clinically, it is characterized by a unilateral granulomatous conjunctivitis that is usually accompanied by preauricular, submandibular, or cervical satellite lymphadenopathy. There may be association with unilateral conjunctival injection, foreign body sensation, and epiphora. Some patients may also develop ulceration of the conjunctival epithelium with mucus-purulent secretion [1-4]. The main etiology of PCOS is cat-scratch disease, which is caused by the bacterium *Bartonella henselae*. In addition to it, although less common, some viruses, parasites and fungi, such as sporotrichosis, can also cause the disease [1,3-5]. In 1898, Benjamin Schenck described sporotrichosis for the first time in the United States. The infection develops from the cutaneous inoculation of the dimorphic fungus *Sporothrix schenckii*. Lesions are usually restricted to the skin, subcutaneous tissue, and adjacent lymphatic vessels. Spread to other organs exists, but is rare [2,4,5,6]. The clinical forms of sporotrichosis are classified as fixed or localized cutaneous, cutaneous-lymphatic, disseminated cutaneous, mucosal and extracutaneous or systemic. The latter develops when there is inhalation of spores [3,4,6,7]. Cats play an important epidemiological role in the transmission and propagation of the disease, because in their skin lesions there is a large amount of infecting fungal cells. The fungus that causes sporotrichosis also inhabits soil, straw, wood and vegetables. Transmission occurs by direct inoculation of the fungus through scratching or biting infected animals or by minor trauma during activities related to gardening [4,5,7-9]. The diagnosis can be made from the association of clinical examination with complementary tests such as cytology, mycological culture examination, histopathology, serological

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tests, intradermal tests, inoculation in animals and polymerase chain reaction. Fungal culture, however, is the definitive method for diagnosing the disease. [8,10-12]. The drug of choice for treatment is itraconazole. It has fewer adverse effects when compared to other antifungal agents. The use of the drug must be extended for up to 30 days after clinical cure. Treatment can also be carried out with potassium iodide solution. [9,12-14].

2. Case report

L.A.P.B., male, 13 years old, student, born in Rio de Janeiro, Brazil, attended the ophthalmology consultation accompanied by his mother, who stated that her son had started febrile episodes associated with adynamia and ocular hyperemia with pruritus for 28 days, without spontaneous improvement. Referred by another ophthalmologist after treatment with topical antibiotic + ocular lubricant and Azithromycin orally without improvement. In his past pathological history he denied systemic comorbidities. He denied previous trauma, eye surgery and eye pathologies, as well as the regular use of eye drops and family members with a history of glaucoma. He claimed the family has pets at home, including 2 cats. 20/20 uncorrected visual acuity in both eyes. Biomicroscopy of the right eye (RE) showed multiple rounded and raised lesions in the lower tarsal conjunctiva, cul-de-sac and caruncle, with a granulomatous appearance and sessile base. Soft consistency, painless to palpation, with follicles around and small amount of secretion. (Figures 1 and 2) Left eye without changes.

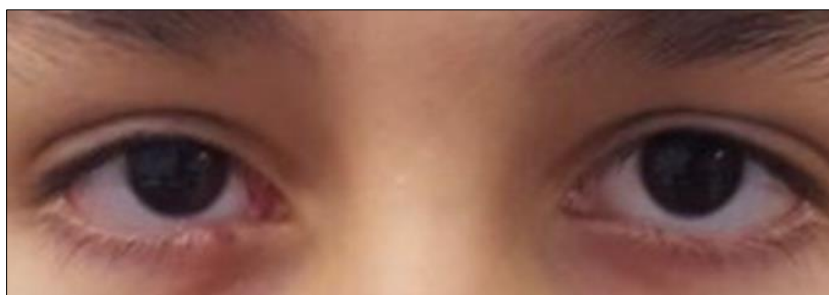


Figure 1 Left eye without changes



Figure 2 Right eye with alterations

Intraocular pressure: 10/11 mmHg (14: 30). Fundoscopy showed no changes. The patient had pre-auricular lymphadenopathy on the right, painless on palpation, slightly indurated, slightly reddened and warm, measuring approximately 2x2cm. (Figure 3)



Figure 3 Pre-auricular lymphadenopathy on the right, painless on palpation, slightly indurated, slightly reddened and warm, measuring approximately 2x2cm

The granulomatous material was collected for swab and culture for fungi, and the presence of *Sporothrix schenckii* was diagnosed. Treatment was started with Itraconazole 200mg orally, 1x/day. After 4 months of treatment, he showed progressive improvement in his condition with his regular follow-up maintained by the ophthalmology department. (Figure 4)



Figure 4 Improvement in his condition

3. Discussion

PCOS is a syndrome marked by the presence of nodular lesions in the bulbar and tarsal conjunctiva with a granulomatous appearance. Conjunctivitis can present as mild to severe, with ulcers or necrosis [3,6,8,9,11]. When present, adenopathy may or may not be painful, and its size and consistency are also variable. Other nonspecific signs and symptoms may accompany PCOS, such as fever, hyperemia, conjunctival follicular reaction, foreign body sensation and mucoid or purulent secretion [7,10,15]. The patient in the present report had systemic symptoms such as fever and prostration. started approximately 1 month ago, an intraocular foreign body sensation in the RE and with ipsilateral preauricular lymphadenopathy. The presence of granulomatous lesions in the tarsal conjunctiva of the right eye associated with the presence of follicles led the diagnosis to some possibilities. PCOS can be caused by several types of pathogens. Among them, the most common is the gram-negative bacillus *Bartonella henselae*, responsible for cat-scratch disease [6,13-15]. Other bacteria such as *Chlamydia trachomatis*, *Listeria monocytogenes*, *Treponema pallidum*, and *Mycobacterium tuberculosis* can also cause the disease. Fungi such as *Paracoccidioides brasiliensis*, *Cryptococcus neoformans* and *Sporothrix schenckii* and viruses such as Herpes simplex type 1 and Epstein-Barr are also etiologies of PCOS, but are less common [3,5,9,15]. The case described shows a rarer etiological condition, but which should always

be taken into account when faced with a patient with unilateral granulomatous conjunctivitis associated with the presence of follicles, preauricular lymphadenopathy, and systemic symptoms such as fever.

In countries with hot and humid climates, the incidence of sporotrichosis is higher. In these countries, transmission by traumatic inoculation with plants or soils with fungi of the *Sporothrix* genus is also prevalent. Currently, however, zoonotic transmission by biting, scratching or direct contact with infected cats is notable [8,12-14,15]. Therefore, it is essential that a detailed anamnesis be carried out in order to find out if there is contact with domestic animals by affected patients as performed in our case. The clinical manifestations of sporotrichosis are divided into cutaneous and extracutaneous. The localized cutaneous clinical form is the most common presentation. It presents a benign course of a subacute or chronic nature, being characterized by polymorphic lesions of the cutaneous and subcutaneous tissues, with involvement of the adjacent lymphatic channels that can suppurate, drain and ulcerate. [8,10,11,15-17]. Mucous involvement is uncommon and preferentially affects the tarsal conjunctiva. PCOS is the representation of this infection, which is most often caused by local trauma. Involvement of intraocular structures, on the other hand, occurs mainly by hematogenous dissemination [5,7,8,14,15]. This more severe condition was not present in the patient, since only his tarsal conjunctiva was affected, with no involvement of segment structures. Posterior and other intraocular parts. When not properly treated, patients can evolve with loss of visual function, and it is essential to reinforce the importance of the correct and early diagnosis of this condition. A complete anamnesis is essential in the elucidation of cases, since the history of professional or domestic contact with animals or contaminated materials is routinely found and leads to diagnostic suspicion. [3,4,7,10]. In the case described, contact with cats was crucial for clinical suspicion and in directing diagnostic possibilities.

For diagnostic confirmation, detection of the etiologic agent can be done directly or indirectly. In direct examination, the Gram stain method may be helpful. Histopathological examination can also help, but is less effective compared to fungus-specific culture. Serological tests, although little used, have shown good sensitivity and specificity. [3,9,11,13]. Polymerase chain reaction (PCR) can also be used, despite being less accessible. In the case of sporotrichosis, the best method for diagnosis is the detection of the etiologic agent in an appropriate culture medium. [5,9,11,12,14]. Fungal culture from the biopsy fragment was the fundamental test for elucidating the etiological agent related to the patient's condition, since both sporotrichosis and bartonellosis have similar epidemiological and clinical characteristics, which can make the differential diagnosis difficult. In the treatment of sporotrichosis, potassium iodide was the first drug described for a long time. Currently, the drug of choice is Itraconazole. The recommended dose is 100 to 200 mg per day orally, with a duration of 3 to 6 months of treatment [8-10,13]. Intravenous amphotericin B is the drug of choice for systemic sporotrichosis and the disseminated form of the disease, and is also indicated in cases that have not responded adequately to potassium iodide and Itraconazole [9,12,13,15]. In our case, the patient was treated with Itraconazole 200 mg once a day for 4 months, with improvement in his condition and an outpatient follow-up was performed without complications.

4. Conclusion

PCOS should always be considered in the differential diagnosis of conjunctivitis with an evident granulomatous reaction and ipsilateral lymph node involvement. Sporotrichosis generally has a good prognosis, especially when diagnosis and treatment are done early, which allows for a significant improvement in the signs and symptoms of the condition, in addition to reducing the chances of complications. In the past pathological history, knowledge of previous comorbidities, family history and epidemiological data are essential for directing diagnostic hypotheses and for choosing the best complementary methods to elucidate the condition. After that, the therapeutic conduct and follow-up of the patients can be carried out properly without leaving sequelae for them.

Compliance with ethical standards

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Disclosure of conflict of interest

The author, the co-authors, the guiding professor and the reviewer suggested for evaluation of the work entitled Parinaud's Oculo-Glandular Syndrome Secondary to Sporotrichosis. Do not present any form of conflict of interest regarding the above.

Statement of informed consent

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

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