



SÍNDROME OCULOGLANDULAR DE PARINAUD SECUNDÁRIA A ESPOROTRICOSE: RELATO DE CASO
PARINAUD'S OCULOGLANDULAR SYNDROME SECONDARY TO SPOROTRICOSES: CASE REPORT
SÍNDROME OCULOGLANDULAR DE PARINAUD SECUNDARIO A SPOROTRICOSE: REPORTE DE CASO


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RESUMO

Introdução: A Síndrome Oculoglandular de Parinaud é uma conjuntivite granulomatosa rara, acompanhada de linfadenopatia regional. É causada por etiologias variadas, sendo uma delas a esporotricose, que é uma causa rara e pouco relatada na literatura.

Objetivo: Relatar um caso da Síndrome Oculoglandular de Parinaud causada por esporotricose atípica, associada a dacriocistite, atendida por um serviço oftalmológico de urgência.

Métodos: O estudo foi realizado a partir da análise do prontuário, juntamente com a anamnese, exame oftalmológico e exames complementares da paciente. Foi realizada pesquisa bibliográfica com publicações de até 20 anos para fundamentação teórico-metodológica.

Resultados: Foi possível identificar algumas das alterações mais comuns na Síndrome Oculoglandular de Parinaud por esporotricose, consistindo em nódulos hiperêmicos na conjuntiva tarsal ou bulbar, relacionados à adenopatia endurecida ou fibroelástica, nas regiões submandibular e/ ou pré-auricular.

Conclusão: A esporotricose é uma doença de tratamento eficiente se identificada prontamente e deve ser sempre considerada como uma possível causa da Síndrome Oculoglandular de Parinaud. Este caso pode ser referência para profissionais da área, tanto como no diagnóstico quanto no manejo do paciente com tal diagnóstico sindrômico, já que é um caso raro e pouco visto.

Palavras-chave: esporotricose; síndrome oculoglandular de parinaud; conjuntivite granulomatosa

ABSTRACT

Introduction: Parinaud's Oculoglandular Syndrome is a rare granulomatous conjunctivitis, accompanied by regional lymphadenopathy. It is caused by varied etiologies, one of which is sporotrichosis, which is a rare cause and little reported in the literature.

Objective: To report a case of Parinaud's Oculoglandular Syndrome caused by atypical sporotrichosis, associated with dacryocystitis, attended by an emergency eye service.

Methods: The study was carried out from the analysis of the medical record, along with the anamnesis, ophthalmological examination and complementary examinations of the patient. Bibliographic research was carried out with publications of up to twenty years for theoretical and methodological foundations.

Results: It was possible to identify some of the most common changes in Parinaud's Oculoglandular Syndrome due to sporotrichosis. Such changes consist of hyperemic nodules in the tarsal or bulbar conjunctiva, related with adenopathy, hardened or fibroelastic, in submandibular and / or preauricular regions.

Conclusion: Sporotrichosis is a disease with an effective treatment if readily identified and should always be considered as a possible cause of Parinaud's Oculoglandular Syndrome. This case can be a reference for professionals in the field, both in diagnosis and in the management of patients with such syndromic diagnosis, since it is a rare and rarely seen case.

Keywords: sporotrichosis; parinaud's oculoglandular syndrome; granulomatous conjunctivitis

RESUMEN

Introducción: La síndrome oculoglandular de Parinaud es una conjuntivitis granulomatosa rara, acompañada de adenopatías regionales. Es causada por diversas etiologías, una de las cuales es la esporotricosis, que es una causa rara y poco reportada en la literatura.

Objetivo: Reportar un caso de Síndrome Oculoglandular de Parinaud por esporotricosis atípica, asociado a dacriocistitis, atendido por un servicio oftalmológico de emergencia.

Métodos: El estudio se realizó a partir de análisis de la historia clínica, junto con la anamnesis, examen oftalmológico y exámenes complementarios de la paciente. La investigación bibliográfica se realizó con publicaciones de hasta veinte años por razones teóricas y metodológicas.

Resultados: Se pudieron identificar algunos de los cambios más frecuentes en el Síndrome Oculoglandular de Parinaud por esporotricosis, consistente en nódulos hiperêmicos en la conjuntiva tarsal o bulbar, relacionados con adenopatías, endurecidas o fibroelásticas, en regiones submandibulares y / o preauriculares.

Conclusión: La esporotricosis es una enfermedad de tratamiento eficaz si se identifica fácilmente y siempre se debe considerar como una posible causa del síndrome oculoglandular de Parinaud. Este caso puede ser una referencia para los profesionales del campo, tanto en el diagnóstico como en el manejo de pacientes con dicho diagnóstico sindrómico, ya que es un caso raro y poco visto.

Palabras clave: esporotricosis; síndrome oculoglandular de parinaud; conjuntivitis granulomatosa

INTRODUCTION

Sporotrichosis is an infection caused by fungi of the genus *Sporothrix* and occurs more commonly in tropical and subtropical regions (Orofino-Costa et al., 2017). It is an anthrozoosis that can be transmitted by human contact with infected plants or animals, especially cats, or by inhaling spores through the upper respiratory tract (Niklitschek et al., 2015; Orofino-Costa et al., 2017; Queiroz-Telles et al., 2019). The estimated prevalence of sporotrichosis in Brazil ranges from 0.1% to 0.5% (Chakrabarti et al., 2014).

The clinical manifestations of sporotrichosis were divided into four groups, according to Sampaio and Lacaz: lymphocutaneous, fixed cutaneous, multifocal or disseminated, and extracutaneous (de Abreu Ribeiro et al., 2010; Morris-Jones, 2002). Thus, the most common involvement is lymphocutaneous (Ramos-e-Silva et al., 2007). Among the extracutaneous manifestations, there is ocular involvement, which is the mucosa most commonly affected by sporotrichosis (Morris-Jones, 2002; Orofino-Costa et al., 2017). When infection of the ocular mucosa and regional lymph nodes occurs at the same time, there is Parinaud's Oculoglandular Syndrome (Morris-Jones, 2002).

Parinaud's Oculoglandular Syndrome is described as unilateral conjunctivitis and pre-auricular, submandibular and cervical lymphadenopathy on the side ipsilateral to conjunctivitis (Ramos-e-Silva et al., 2007). The condition is usually associated with the bacterium *Bartonella henselae*, but it can also be caused by other chronic granulomatous infections, such as tuberculosis and syphilis, viral infections, such as the herpes simplex virus, and other fungal infections, such as blastomycosis and coccidioidomycosis (Ferreira et al., 2014). Despite being less prevalent compared to infection by *Bartonella henselae*, infection by *Sporothrix* can also cause Parinaud's Syndrome, as in the clinical case.

An ophthalmological and neurological examination is indicated for these patients, including visual acuity, field of view, examination of the pupils and fundoscopy. In addition to ocular manifestations, the individual with Parinaud's Oculoglandular Syndrome also has conjunctivitis associated with ipsilateral lymphadenopathy in the submandibular and preauricular regions. Lymphadenopathy can extend to lymph nodes in the cervical and parotid regions (Paul Yan, 2015).

The diagnosis of sporotrichosis and consequently as a cause of Parinaud's Syndrome is made through the correlation between clinical, epidemiological and laboratory aspects. The diagnosis can be made in the following ways: microscopy; histopathological study; culture; intradermal test with sporotrichin; molecular detection; detection of antibodies (Paul Yan, 2015). The most sensitive and gold standard method for the diagnosis of sporotrichosis is culture.

The objective of this case report is to report a case of Parinaud's Oculoglandular Syndrome caused by atypical sporotrichosis, associated with dacryocystitis, attended by an emergency eye service, that can be used as a reference for professionals in the field since more comprehensive studies are still scarce. The importance of reporting the case of this disease is highlighted as it is a rare pathology.

1. METHODS

The study was initiated from the care of a patient seen at an ophthalmologic emergency service. Ophthalmic clinical examination, anatomopathological examination performed after clinical suspicion and complementary data reported by the patient were used as data. After confirming diagnosis and treatment, a bibliographic review was made on the subject, analyzing the most relevant articles published in the last 10 years in indexed journals. The keywords used for the search were: "Sporotrichosis", "Parinaud's oculoglandular syndrome" and "Granulomatous conjunctivitis". All ethical issues were respected in the development of the study.

2. CASE REPORT

Patient M.A.S., female, 30 years old, was first seen in December 2018, complaining of pain and edema in the topography region of the lacrimal sac on the right, in addition to hyperemia and itchy eyes. She reports a previous diagnosis of conjunctivitis and dacryocystitis, who has been on treatment with oral and topical antibiotics for a month, with therapeutic failure until then. The patient denies systemic and ocular comorbidities or other complaints. She reports close contact with cats, one of whom has a skin lesion. On examination, she had 20/20 visual acuity in both eyes. Biomicroscopy of the right eye shows diffuse hyperemia, transparent cornea, anterior chamber formed without chamber reactions, negative fluorescein test, important follicular reaction, in addition to the presence of small hyperemic nodules in the lower tarsal conjunctiva. She also had indurated lesions in the upper eyelid of the right eye and in topography of the lacrimal sac, with drainage of purulent secretion, compatible with dacryocystitis. Left eye without changes. Fundoscopy without changes in both eyes. Preserved eye movement in both eyes. Ectoscopy with lymph nodes in the right submandibular region, which were painful on palpation. A biopsy of the eyelid lesion and swab of the tarsal secretion were performed, and the fungus *Sporothrix sp* was found. From this the diagnosis of the Parinaud's oculoglandular syndrome was confirmed, with sporotrichosis as the cause. Then Itraconazole 200mg once daily for 3 months was started.

After 3 months of treatment, the patient returned for follow-up with complete resolution of the case. The patient attended the medical appointment without complaints, absence of conjunctival hyperemia and regression of dacryocystitis. He had only an indurated lesion in the upper eyelid on right eye.



Figure 1 – Dacryocystitis with drainage of purulent secretion in the right eye.



Figure 2 – Upper eyelid indurated lesion in the right eye.

3. RESULTS AND DISCUSSION

The Parinaud's oculoglandular syndrome caused by sporotrichosis contamination occurs through *Sporothrix sp* and occurs through injuries or scratches with spikes or splinters from contaminated vegetables, biting or licking infected animals and handling previously contaminated soil. It can also happen without any previous trauma, as in cases of inhalation of fungal spores, but they are even more rare (de Abreu Ribeiro et al., 2010). In the clinical case presented, the transmission of sporotrichosis occurred through contact with an infected cat. It is worth mentioning that the main form of contamination is the traumatic inoculation of soil, plants and organic matter contaminated with fungus (Madureira et al., 2018). The incidence of cat scratch disease is 2.4-2.7 per 100,000 people, since it is the main cause of Parinaud's oculoglandular syndrome (Paul Yan, 2015).

Sporotrichosis, on the other hand, has a worldwide distribution, being even more prevalent in tropical and subtropical areas, being even more common in urban areas than in rural areas. Its main characteristic is the occurrence of isolated epidemics, as an example that occurred in the city of Rio de Janeiro, Brazil, in 2009 (da Silva et al., 2012; Niklitschek et al., 2015).

Sporotrichosis is a neglected disease with no obligation to notify, what makes difficult to know its epidemiological data (Chakrabarti et al., 2015; Neto et al., 2021). The disease has been reported in countries such as the United States, Mexico, Peru, Colombia, China, India and Australia (Chakrabarti et al., 2015; Bonifaz & Tirado-Sánchez, 2017). In Europe, the countries that reported the most cases until 2017 were France, Italy and Spain. Among the countries, Brazil has the highest number of cases, mainly in the states of Rio de Janeiro, São Paulo, Minas Gerais, Paraná and Rio Grande do Sul, however, it has a low incidence and is still considered a rare disease (Bonifaz & Tirado-Sánchez, 2017).

Brazil is an endemic area for sporotrichosis (Barros et al., 2004). Although there are few case reports in the literature, it is known that the incidence of sporotrichosis has a current tendency to increase in endemic areas, which reinforces the importance of the present study. The similarity with other ophthalmologic diseases makes that ophthalmologists often mistakenly start the treatment with antibiotics, as happened with the patient M.A.S., which delays the definitive treatment and increases the chance of complications (Yagamata et al., 2017).

In a study of a series of cases of Sporotrichosis generating Parinaud's oculoglandular syndrome, most patients were female, as was the case in the present report. Despite denying eye trauma, all study patients had a pet cat at home diagnosed with

sporotrichosis, which was likely the source of contamination. This scenario is repeated in this case and is similar to the epidemiological data about the disease, which has as its predominant profile of contamination adult women involved in domestic activities. This epidemiological profile can be justified by the greater exposure of these people to close contact with pets (Ribeiro et al., 2020; Barros et al., 2004).

In the case presented, it was possible to identify some of the most common changes in Parinaud's Oculoglandular Syndrome due to sporotrichosis. Such changes consist of hyperemic nodules in the tarsal or bulbar conjunctiva, usually unilateral, associated with adenopathy, hardened or fibroelastic, in submandibular and / or preauricular regions (Shields et al., 2017).

Most of the time, its clinical presentation leads to edema and hyperemia, as seen in the case report, which can cause diagnostic confusion. Some cases may have associated infections, such as dacryocystitis or choroiditis. In addition, other symptoms may be present, such as malaise, fever, itching, photophobia and foreign body sensation (BARNES et al, 2005).

In addition, such pathology does not usually lead to changes in visual acuity or changes to funduscopy. Changes in Parinaud's oculoglandular syndrome can be seen in more detail from biomicroscopy. Despite the characteristic lesions observed by this examination, it is only possible to define the diagnosis and the etiologic agent of the disease through anatomopathological examination (Paul Yan, 2015).

It is known that culture is the gold standard for establishing the diagnosis of sporotrichosis (Mahajan, 2004). The agent *Sporothrix schenckii* can be cultured from skin biopsies or other clinical specimens that can be collected by swab, as done in this report. After being collected or biopsied, this pathogen is placed in a culture medium, which in this case was Sabourad Agar with dextrose, at room temperature, taking from 4 days to 2 weeks to visualize visible growth. It is not necessary to use Gram stains, as the identification of this pathogen is made by direct visualization of the characteristic morphology (Ribeiro et al., 2020).

Regarding the treatment of Parinaud's oculoglandular syndrome due to sporotrichosis, drug therapy is essential, as its spontaneous resolution is extremely rare (Ribeiro et al., 2020). Previously, several drugs were used to treat this syndrome, such as potassium iodide, which became the treatment of choice. However, potassium iodide has not been shown to be effective for the treatment of extracutaneous forms of the disease, which is why it is no longer used, giving way to antifungals of the azole class (Barros et al., 2004; Mahajan, 2014; Kauffkman et al., 2007).

The ideal treatment for Parinaud's oculoglandular syndrome caused by sporotrichosis is with oral antifungal agents. Itraconazole is generally used, with doses ranging between 100 and 200 milligrams per day. The duration of treatment is variable, with 6-week treatments up to 6-month treatments found in the literature (Biancardi et al., 2019; de Abreu Ribeiro et al., 2010; Ferreira et al., 2014; Madureira et al., 2018). In the clinical case presented, the dose of 200mg per day for 3 months was used, and the treatment was successfully concluded. Therapeutic success has been achieved in both young and elderly patients who have undergone this therapy for the condition in question.

In the series of cases described by Ribeiro, Itraconazole, at a dose of 200mg per day, was used for treatment for 3 to 6 months, with a positive therapeutic response after the third month of treatment. Resolution of the condition was also reached at the end of the third month of treatment for the patient in this case report (Ribeiro et al., 2020).

CONCLUSION

Case reports on Parinaud's oculoglandular syndrome secondary to sporotrichosis are essential, as they treat neglected, little-known and rare diseases. Furthermore, despite the low incidence, an epidemiological growth of this pathology has been identified in recent years, which makes it even more important to report it, highlighting its characteristics and clinical management.

Despite being considered as rare, Parinaud's Oculoglandular Syndrome requires attention and prior knowledge from the ophthalmologist, especially considering the cases that present with granulomatous conjunctivitis, which can lead to greater complications for patients, since such symptoms are also present in other entities, generating diagnostic doubt, which can lead to a delay in the start of treatment. Furthermore, without the correct treatment, in addition to not achieving therapeutic success, the duration of the clinical picture is prolonged, which reduces the patient's quality of life.

Still, it is essential to collect a good anamnesis, considering previous diseases, professional career and epidemiological situation, which will assist in the diagnosis. A complete physical examination, in addition to the eye examination, is also essential for identifying characteristic signs, such as regional lymphadenitis.

As Parinaud's oculoglandular syndrome is an entity that has more than one etiology, an anatomopathological examination is necessary to identify the agent and correct institution of the specific treatment. In the case of the patient in question, the clinical picture was prolonged, due to an incorrect diagnosis made previously in another service, which delayed the implementation of effective therapy. However, right after entering the emergency service, the accurate diagnosis and adequate management were substantial for the complete resolution of the condition, without leaving sequelae. Prompt recognition of the disease and the implementation of treatment accelerate the patient's clinical improvement and prevent complications.

Sporotrichosis is a disease with an effective treatment if readily identified and should always be considered as a possible cause of Parinaud's Oculoglandular Syndrome. This case can be a reference for professionals in the field, both in diagnosis and in the management of patients with such syndromic diagnosis, since it is a rare and rarely seen case.

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