

## CASE REPORT

### PARINAUD'S OCULOGLANDULAR SYNDROME ASSOCIATED WITH PARACOCCIDIOIDOMYCOSIS

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#### SUMMARY

The authors report one case of Parinaud's oculoglandular syndrome associated with *Paracoccidioides brasiliensis* infection. No other medical report of this condition was found in the medical literature available at Index Medicus and Medline.

The eye involvement has been rather uncommon in paracoccidioidomycosis and this report emphasizes the possibility of this kind of presentation making it also necessary to include paracoccidioidomycosis among the several known causes of Parinaud's oculoglandular syndrome.

**KEYWORDS:** Parinaud's oculoglandular syndrome; Conjunctivitis; Paracoccidioidomycosis.

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#### INTRODUCTION

Parinaud's oculoglandular syndrome is a curious medical condition characterized by granulomatous conjunctivitis associated with homolateral cervical and either pre or retro auricular lymph adenopathy<sup>16</sup>. It has been associated with several different infectious diseases<sup>1</sup>, especially to cat scratch disease (*Bartonella henselae*)<sup>22,24,27,28,29,32</sup>, but at times it has been reported in granulomatous chronic infectious like tuberculosis and atypical mycobacteria infections<sup>1</sup> and syphilis<sup>1</sup>. Fungal infections also have been associated to this syndrome especially sporotrichosis (*Sporothrix schenckii*)<sup>1</sup>, blastomycosis (*Blastomyces dermatitidis*)<sup>1,4</sup>, and at least on one occasion, to coccidioidomycosis (*Coccidioides immitis*)<sup>7</sup>.

Paracoccidioidomycosis is by far the most important endemic mycosis in South America<sup>9,13,27</sup>. The association between Parinaud's oculoglandular syndrome and paracoccidioidomycosis hasn't been reported so far.

We report here one typical case of Parinaud's oculoglandular syndrome, which had *Paracoccidioides brasiliensis* as its etiological agent. We further overviewed the literature about other possible infectious causes of this syndrome.

#### CASE REPORT

A 31-year-old black man was admitted to the hospital because of

cervical lymphadenopathy which had started five months earlier, associated with low grade fever, malaise and weight loss. Since the very beginning of his illness he had complained of pain and a purulent discharge in the right eye. His clinical picture evolved in a very indolent way and the patient only received topical therapy for a supposed bacterial conjunctivitis. As his clinical picture deteriorated a cervical lymph node biopsy was performed giving rise to the diagnosis of paracoccidioidomycosis and the patient was transferred to our service for treatment. At the physical examination one could see a thin man with cervical lymphadenopathy (Fig. 1-a), associated with increased right pre and retro auricular lymph nodes. Those lymph nodes were swollen, slightly painful and one located at the right cheek showed sinus drainage. The most prominent lymph node was aspirated (Fig. 1-b) and viscous yellow pus was obtained which was shown to be negative for fungi on direct examination. Another lymph node was further biopsied confirming the diagnosis of paracoccidioidomycosis. No mucosal lesions were seen but his ophthalmologic examination revealed prominent right eye conjunctivitis especially in the bulbar upper inner conjunctiva (Fig. 1-c). This lesion was biopsied and the histological pattern is shown in Fig. 2. Cultures at Sabouraud's medium were negative but serology by double radial immunodiffusion using GP 43 *Paracoccidioides brasiliensis* antigen rich fraction<sup>8</sup> was positive at a title of 1/32. A chest X-ray showed a diffuse alveolointerstitial infiltrate (Fig. 1-d) and the sputum analysis revealed the presence of *Paracoccidioides brasiliensis* forms. Other blood tests were unremarkable. The patient was given cotrimoxazole and showed continuous improvement, being discharged after 3 weeks.

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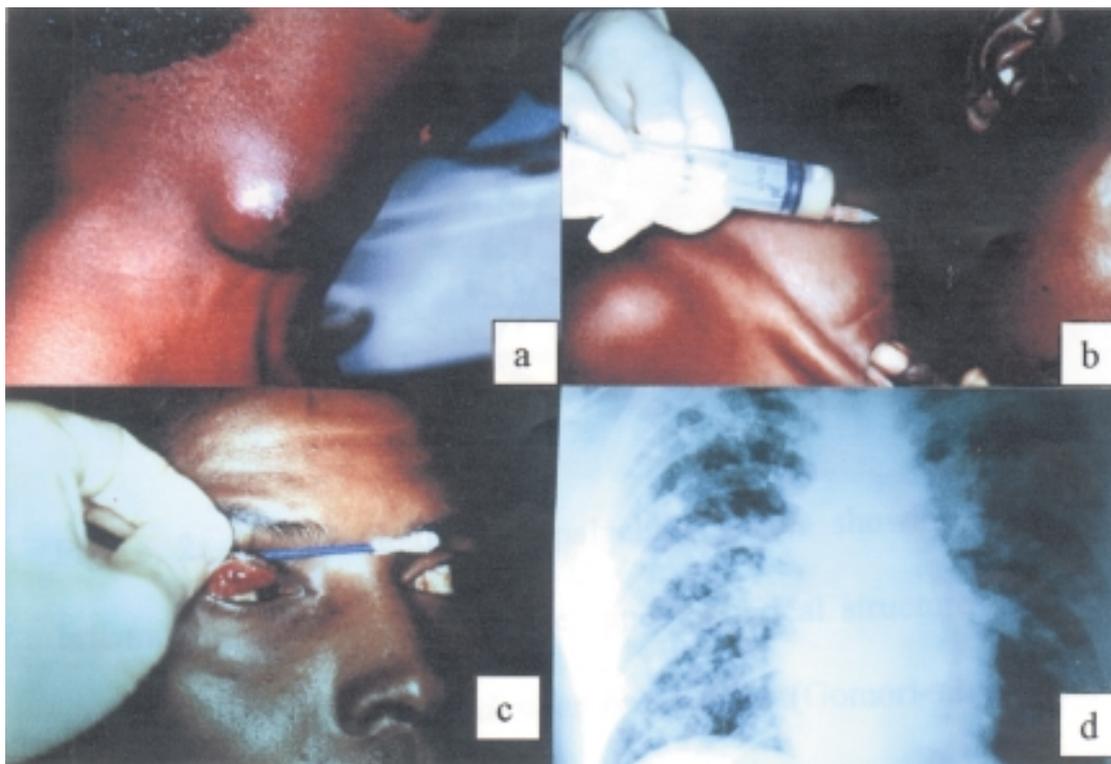
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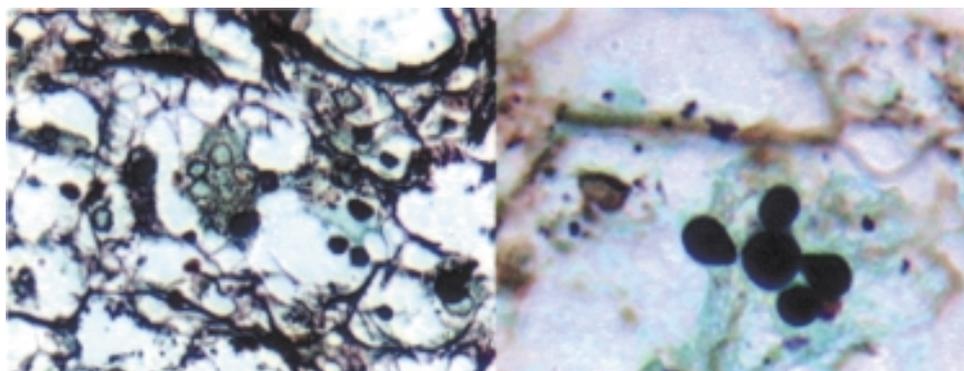
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**Fig. 1** - a) Cervical lymphadenopathy showing swollen lymph nodes, b) aspiration showing a viscous yellow pus, c) prominent right eye conjunctivitis specially in bulbar upper inner conjunctiva and d) chest X-ray showing a diffuse alveolointerstitial infiltrate.



**Fig. 2** - Histopathology of palpebral lesion showing granulomatous inflammation of connective tissue and spherical structures with budding compatible with *Paracoccidioides brasiliensis* (Gomori-silver stain X 200 and X 400).

## DISCUSSION

The purpose of this paper is to emphasize the peculiar ophthalmologic aspects of this case of paracoccidioidomycosis, which is compatible with Parinaud's oculoglandular syndrome<sup>1,31</sup>.

Parinaud's oculoglandular syndrome has been characterized by the combination of unilateral granulomatous conjunctivitis and enlarged

regional lymph nodes<sup>1</sup>. These lymph nodes usually located at the preauricular, submaxillary, or submandibular areas are often tender<sup>1,31</sup>. By far the most prevalent cause has been cat scratch disease caused by *Bartonella henselae* infection<sup>22,24,29,32</sup>, but other infectious agents have also been associated.

Blastomycosis<sup>1,4</sup>, tularemia<sup>19</sup>, tuberculosis and atypical mycobacteria infection<sup>1</sup>, actinomycosis<sup>1</sup>, syphilis<sup>1</sup>, listeriosis<sup>1</sup>, chancroid<sup>1</sup> and

lymphogranuloma venereum<sup>1</sup> have been reported as causes of this syndrome. Other associated infections include *Pasteurella multocida*<sup>1,3</sup>, *Yersinia enterocolitica*<sup>1,17</sup>, *Yersinia pseudotuberculosis*<sup>23</sup>, *Pseudomonas mallei*<sup>1</sup>, *Herpes simplex type I*<sup>15</sup>, *Epstein-Barr virus*<sup>25,26</sup> and *Rickettsia conori* infections<sup>1,21</sup>.

There is no other previous report of Parinaud's oculoglandular syndrome associated with paracoccidioidomycosis according to a Medline search as well as the medical literature available in Index Medicus.

BELFORT Jr. *et al.*<sup>6</sup> described a possible case of Parinaud's oculoglandular syndrome caused by *Paracoccidioides brasiliensis* but this report didn't mention it as that, just emphasizing the simultaneous attack of conjunctiva and the draining lymph nodes which by the way have been considered the usually typical characteristics of that syndrome.

BURNIER & SANT'ANNA<sup>14</sup>, described two cases of eyelid paracoccidioidomycosis, which were the first sign of the disease. Importantly, in both cases the first clinical diagnosis made was not a fungal infection, but of a neoplastic. According to this report no lymphadenopathy was observed so the diagnosis of Parinaud's oculoglandular syndrome could be ruled out.

SILVA *et al.* in 1988<sup>30</sup> reported six cases of ocular paracoccidioidomycosis causing a heterogeneous group of lesions, including two cases of conjunctivitis. These authors also updated what had been published up to that time regarding ocular paracoccidioidomycosis but again no description could fit with Parinaud's oculoglandular syndrome.

Conjunctivitis has been considered the most frequent clinical form of ocular lesion in human paracoccidioidomycosis considering the extreme rarity of eye involvement in this disease<sup>5,7,27,30</sup>.

Another infrequent but severe ocular form of this disease is coriorretinitis with only four cases described as of 1986<sup>2</sup>.

Several hypotheses have been raised to explain ocular paracoccidioidomycosis. According to experimental models, the severity and frequency of ocular lesions in animals, intracardially inoculated with *P. brasiliensis* cultures, suggest haematogenic dissemination of the fungus during septicemic stages<sup>12,18</sup> but no one could rule out the possible role of lymphatic dissemination<sup>14</sup>, self-inoculation from other fungi-rich lesions<sup>1</sup>, contiguous cutaneous spread<sup>14</sup> or even direct inoculation from natural sources<sup>30</sup>.

The practical importance of a syndromic diagnosis like Parinaud's oculoglandular syndrome lays in the diagnostic track it gives, considering the limited array of diseases associated with that syndrome.

On the other hand, ophthalmologists and pathologists must be aware of its occurrence especially in endemic regions in order to avoid diagnostic pitfalls.

Besides other ophthalmologic syndromes like chorioretinitis<sup>2</sup>, anterior uveitis<sup>30</sup>, choroidal lesions<sup>10, 30</sup>, blepharitis<sup>30</sup>, oculomotor nerve lesions<sup>20</sup> and conjunctivitis<sup>6,11,14,20,30</sup>, *P. brasiliensis* can cause typical Parinaud's oculoglandular syndrome.

## RESUMO

### Síndrome oculoglandular de Parinaud associada à paracoccidioidomicose

Os autores relatam um caso de síndrome oculoglandular de Parinaud associada à infecção por *Paracoccidioides brasiliensis*.

Nenhum outro relato desta condição foi encontrado na literatura médica disponível pelo Index Medicus e Medline.

O envolvimento ocular é incomum na paracoccidioidomicose e este relato enfatiza a possibilidade deste tipo de apresentação, tornando-se necessário incluir a paracoccidioidomicose entre as várias causas conhecidas de síndrome oculoglandular de Parinaud.

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