

PARINAUD SYNDROME CAUSED BY *Bartonella henselae*: CASE REPORT

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SUMMARY

The authors report a case of Parinaud syndrome (conjunctivitis with pre-auricular satellite adenitis) caused by *Bartonella henselae*, the etiologic agent of Cat Scratch Disease. The etiologic assessment of this case was performed by serum indirect immunofluorescence reaction and allowed for a better therapeutics and follow up, avoiding ineffective antibiotics and surgical interventions.

KEYWORDS: Cat Scratch Disease; Parinaud Syndrome; *Bartonella henselae*.

INTRODUCTION

Oculoglandular Parinaud syndrome was originally described as an anatomic-clinical condition consisting of granulomatous conjunctivitis accompanied by pre-auricular satellite adenitis^{1,6}. The etiology of this syndrome was not clear at first, but Parinaud related its occurrence to previous contact with house pets⁶. Since then, several causal agents have been identified: *Francisella tularensis*, *Chlamydia trachomatis*, *Sporothrix schenckii*, *Mycobacterium tuberculosis*, *Treponema pallidum*, as well as some viruses¹⁷. Another pathogen, a gram-negative pleomorphic bacillus transmitted by cats, has been recently isolated and classified as *Bartonella henselae*⁴. Concurrently, laboratory tests to detect infections caused by this bacteria have been developed among which Serum Indirect Immunofluorescence is one of the most sensitive and specific^{8,20}.

However, given the technical difficulties to diagnose infections caused by *Bartonella henselae* and the infre-

quent occurrence of Parinaud syndrome, the cases confirmed through laboratory tests and documented are scarce in the pediatric literature⁴. The authors describe a case of Parinaud syndrome caused by *Bartonella henselae* in which the accurate diagnosis was instrumental to establish the course of the treatment and follow-up.

CASE REPORT

E.E.M., male, caucasian, nine years-old, previously healthy was taken to the Emergency Room because of a painful progressive tumor in the right pre-auricular region which had been developing for five days. During this period, the child had only one febrile episode (38°C), but showed no improvement after he had been prescribed analgesics and cefalexin at two other medical centers.

The physical examination revealed a good clinical condition except for conjunctival hyperemia without secretion in the right eye, periorbital edema and a hard-

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ened tumor (diameter 3.0 cm) accompanied by inflammatory signs in the affected area. Fibroelastic ganglia (diameter 1.0 cm) were observed in the submentonian region as well as right submandibular and upper cervical area.

Ultrasonography showed adenomegalies in the affected areas. Initial blood tests revealed slight leucocytosis (12,000 cells/dL) without band forms and amilase within range. Thorax and facial x-rays results were normal.

The patient was admitted to clarify the etiology and for closer follow-up of this case of Parinaud syndrome. Upon admission, when questioned about prior contact with cats, the patient reported he had been repeatedly scratched by a three-month-old cat that had been living in his house for a month.

Blood was collected to conduct cultures in tryptic soy broth and serological tests for citomegalovirus, toxoplasmosis, infectious mononucleosis, rubella and Aids. The Mantoux test was performed. In addition, Serum Indirect Immunofluorescence was proceeded to detect anti-*Bartonella henselae* antibodies. Serologic testing procedures were similar to those described previously⁸. Antigens and anti-sera were gently supplied by Dr. David Welch (Clinical Microbiology Laboratories, Oklahoma Medical Center, USA). The only positive result was the one for *Bartonella henselae* (IgG titer = 512).

Due to the intense inflammation with pain in the pre-auricular adenitis, analgesics and cotrimoxazole (40 mg/kg/day of sulphametoxazol) were prescribed. After ten days of treatment, the tumor had partially shrunk and both fever and other inflammatory signs had disappeared, so the patient was discharged. Three months later, he still showed slight enlargement of the pre-auricular ganglia, which only went away by the fifth month.

DISCUSSION

Cat Scratch Disease is a bacterial pathology frequent in countries with large number of household cats but less common in developing countries where this practice is not widespread^{6,15}. Although most patients report scratches by the animal, in some cases the sheer contact with saliva seem to suffice to transmit the disease.

In the habitual course of the disease papulous lesion develops on the inoculation site approximately four to six days after contact with a cat¹⁰. The enlargement of the

lymphatic drainage ganglia of the affected area lasts from one to seven weeks after inoculation and spontaneous resolution takes place within two to six months^{6,10}. The ganglial involvement may vary in extension and severity, single or multiple, large or small ganglia may occur. In general, there are minor inflammatory signs in the area although severe inflammatory reactions or cases of rapid development of the disease – as observed in our patient, are occasionally reported¹². These acute clinical manifestations led the patient's family to seek medical help three times in five days. The overall symptoms of the disease such as fever and malaise occur in 1/3 of the patients and are not very intense as in the case here reported^{6,17}.

Cat Scratch Disease is mostly confined to the inoculated limbs and their satellite ganglia. However, in 10% of the cases, considered "atypical" there might be systemic infection with internal organ involvement such as commitment of the central nervous system (encephalitis, myelitis and radiculitis) or the peripheral nervous system (neuritis)^{13,18}. There have been even fewer documented cases of Cat Scratch Disease causing tonsillitis, retinitis, hepatitis, pneumonia, paravertebral abscess, vasculitis, nodous erythema and maculopapular exanthem^{2,5,7,9,11,19,21,22}. The present case of Parinaud syndrome is also considered an atypical presentation of Cat Scratch Disease because of its unusual site of inoculation of *Bartonella henselae* – the eye, although the infection was confined to a limited area^{14,17}.

The etiological agent in 90% of the cases of Cat Scratch Disease is *Bartonella henselae*, a gram-negative bacillus of the Bartonellaceae Family, Bartonella Genus, Alpha 2 of the Alpha-Proteobacterial subgroup, and *henselae* species¹⁴. Until 1993, this agent was classified as *Rochalimae henselae*. Previously, another bacteria – *Afipia felis* – had been mistakenly identified as the etiological agent of the disease, albeit it may have been involved in a very limited number of cases¹⁴.

Clearly, the diagnosis of Cat Scratch Disease had been quite challenging until recently. The criteria in use then were clinico-laboratorial, requiring presence of three of the following items: 1) history of contact with a cat (12 months or younger) and occurrence of a lesion on the inoculation site; 2) positive skin test (Hangar Rose); 3) negative laboratory tests for other etiologies of lymphadenopathy, and 4) histopathological characterization in biopsed material (granulomatous tissue)⁴. In the acute phase Warthin-Starry dye may detect the bacillus⁴. The problems with these criteria are: cases of patients whose skin lesion has already disappeared, poor standardization of skin test, and the need for an invasive procedure to obtain material for histologic exam⁴.

The laboratory methods for the diagnosis of Cat Scratch Disease developed after 1993 are not only more reliable but also capable of addressing the problems listed above³. The Serum Indirect Immunofluorescence Reaction has a sensitivity range of 82-88% and 86-90% specificity (for an IgG cut $\geq 1/64$)^{3,4,8,20}. The IgM detection is not very sensitive though very specific since the presence of these antibodies is very brief. In the present case the IgG titer was 512 while the IgM antibodies were not detected. The interpretation of these lab results must take into account the clinical data observed^{6,14}. The Serum Indirect Immunofluorescence Reaction for *Bartonella henselae* is the test recommended by Centers for Disease Control and Prevention for a diagnosis of Cat Scratch Disease^{8,20}. Other exams like the enzyme-linked immunoabsorbent assay (ELISA) and polymerase chain reaction (PCR) have been studied and appear promising as to their diagnostic potential^{3,4,12}.

The evolution of Cat Scratch Disease is generally favorable, even in atypical cases such as the present report of Parinaud syndrome^{6,16}. Therefore, the habitual prescription includes only symptomatic medication. Nonetheless, in cases of internal organ involvement and/or unusual severity as our patient's painful and rapidly progressive ganglionic infarctation, antimicrobial treatment with riphampicine, gentamicine, or cotrimoxazole is indicated and promote the healing of the lesions^{14,16}. The prescription of cotrimoxazole seemed to relieve the patient's symptoms, but other authors have questioned its efficacy^{2,16}.

In conclusion, the authors utilized the Indirect Immunofluorescence Reaction to clarify a case of Parinaud syndrome, which was caused by *Bartonella henselae*. The patient's therapeutics could be best oriented under this established diagnosis.

RESUMO

Síndrome de Parinaud causada por *Bartonella henselae*: relato de caso

Os autores relatam um caso de Síndrome de Parinaud (conjuntivite com adenite satélite pré-auricular) causada por *Bartonella henselae*, o agente etiológico da Doença da Arranhadura do Gato. O esclarecimento etiológico deste caso foi feito através da reação de imunofluorescência indireta o que permitiu um adequado enfoque terapêutico e acompanhamento do paciente, evitando o uso de antibióticos inadequados e intervenções cirúrgicas.

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