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Case Report

Tularemia and the oculoglandular syndrome of Parinaud

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A B S T R A C T

An 18-year-old female came to our clinic with complaints of a tender lump just under her jaw on the left side and another lump in front of her left ear, ecchymosis around the eye and some redness in the eye at the same side. After administering antibiotic therapy for two days we suspected of tularemia and referred the patient to the Infectious Diseases Department. A serum sample was taken and a fine needle aspiration biopsy was performed. The patient was diagnosed with tularemia, the oculoglandular syndrome of Parinaud. Tularemia should also be kept in mind for differential diagnosis in patients with both ocular and glandular symptoms in endemic regions like Turkey and the appropriate therapy should be initiated immediately.

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Tularemia is a zoonotic disease caused by *Francisella tularensis*. Tularemia is seen in six clinical forms: ulceroglandular, glandular, oculoglandular, pharyngeal, typhoidal, or pneumonic. The oculoglandular form, one of the rarest forms (up to 4% only) may be acquired by direct contact with infected materials or by aerosolized contaminated particles.¹⁻³

An 18-year-old female came to our clinic with complaints of a tender lump just under her jaw on the left side and another lump in front of her left ear, ecchymosis around the eye and some redness in the conjunctiva at the same side. She had gone to a hospital before our clinic and had taken some antibiotics for infected lymphadenopathies and some eye drops for conjunctivitis. However, her complaints were still persisting with a decreased redness in her eye. In our physical examination we palpated a 1 x 1 cm sized mobile lymph node in left preauricular region and a 3 x 1.5 cm sized fixed lymph node in left submandibular area, both tender to touch. We also detected periorbital ecchymosis with a decreased redness in conjunctiva. The patient was hospitalized and began ampicilline-sulbactam (1 g, q8h, IV) treatment with the diagnosis of suppurative lymphadenitis

with accompanying conjunctivitis. After 2 days the symptoms had not relieved and we referred the patient to the Infectious Diseases Department for further investigation with the suspicion of oculoglandular tularemia (as it is endemic in our region). The patient was then put on doxycycline (100 mg, q12h, PO) therapy. The microagglutination test result was positive for tularemia with a titer of 1:160. The patient took oral doxycycline (100 mg, q12h, PO) for about 10 days and was discharged with oral therapy after her symptoms declined. She received the therapy for up to 21 days, and was considered successfully treated in the follow-up.

Tularemia has become endemic in our region Sivas, Turkey, for the last 2 years and only one oculoglandular form of tularemia was seen among 30 cases (3.33%) diagnosed with the microagglutination method. As seen in our patient, the first symptom may be conjunctivitis and can be treated as a common isolated disease. Despite topical treatment, the disease continues to develop and the patient is not diagnosed until he/she has swollen lymph nodes; or is even misdiagnosed as a conjunctivitis accompanying a parotitis or a parotid neoplasm.^{4,5}

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Parinaud's oculoglandular syndrome should be considered in the differential diagnosis in a patient presenting with unilateral granulomatous conjunctivitis, painful preauricular, and submandibular lymphadenopathy combined with systemic symptoms of general malaise and fever. Parinaud syndrome is virtually synonymous with cat-scratch disease although several other agents have been implicated, including tularemia, sporotrichosis, tuberculosis, and acute *Chlamydia trachomatis* infections.⁶ According to the literature, our patient is the fourth case with oculoglandular syndrome reported. Firstly, Mezricka⁷ described the oculoglandular syndrome in a patient with tularemia in their region in 1963. The second case was a 9-year-old boy reported by Halperin et al.⁸ in 1985. The last case was an 18-year-old male reported by Thompson et al.⁹ in 2001. Our patient was in the pediatric age group as the other two cases reported.

In the daily practice of an otolaryngologist, it is not usual to diagnose a patient with the oculoglandular syndrome of Parinaud. Tularemia should be considered in the differential diagnosis of the patients with both ocular and glandular symptoms in endemic regions like Turkey.

Conflict of interest

All authors declare to have no conflict of interest.

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