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ID TEACHING CASES

Oculoglandular Tularemia From Crushing an Engorged Tick

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We report on an unusual case of oculoglandular tularemia acquired after crushing a tick removed from a dog. As a droplet sprayed into the patient's eye the eyelids became inflamed, and on the fourth day, a high fever started. Prompt antibiotic treatment prevented serious complication.

Keywords. oculoglandular; dog-related infection; tick-borne infection; tick-borne tularemia; tularemia.

A 25-year-old man removed an engorged tick from his dog. As he crushed the tick, a droplet accidentally sprayed into his left eye. Two days later, his left eyelids and the surrounding skin of the face became swollen and sensitive to touch. A lymph node became enlarged and moderately painful in front of the left ear. The conjunctivae were red with minimal purulent discharge, and his eyes started to intensively tear (Figure 1). He had a temperature of 37.2°C 3 days after the incident. His family doctor initiated treatment for erythema migrans with 500 mg of amoxicillin b.i.d. and an eye drop containing dexamethason and gentamycin. His temperature elevated to 39°C on the fourth day, and then he visited our clinic. Suspecting tularemia, we immediately changed the treatment to doxycycline 100 mg b.i.d. for 20 days and changed the eyedrops to tobramycine instead of the steroid and gentamycin combination. His routine laboratory tests showed a sedimentation rate of 5 mm/h; his white blood cell count was 3.3 G/L (normal range, 4–10.0 G/L), C-reactive protein was 16.3 mg/L (normal, <8.0 mg/L), procalcitonin was <0.05 ng/L (normal, <0.50), alanine aminotransferase (ALT), aspartate aminotransferase (AST) and gamma-glutamyltransferase (GGT) were normal, but total bilirubin was elevated: 44.2 µmol/L (normal, 5.1–17.1 µmol/L) and direct bilirubin was 11.1 µmol/L (normal: <6.8 µmol/L), tested on the fifth day after the incident. All the other blood tests were in the normal range. His temperature decreased rapidly and returned to normal within 3 days after the doxycycline treatment. He became symptom-free 2 weeks after the start of the treatment (Figure 2) and was healthy at the time of the 6-week follow-up visit.

The serum samples drawn on the fourth, 19th, and 42nd days after the accident were positive for *Francisella tularensis* antibodies examined by tube agglutination (in-house; 0, 1:160, 1:320) and immunofluorescence test (IgM: 0, 1:64, 1:64; IgG: 0, 1:256, 1:1024). Polymerase chain reaction was not performed because of the biohazard concern. All serum samples tested for *Borrelia burgdorferi* s.l. by Western blot and *Anaplasma phagocytophilum* indirect immunofluorescence tests remained negative for both IgM and IgG.

Tularemia is a zoonotic infection caused by *F. tularensis*. It is extremely contagious. The infectious dose in humans is 10 to 50 organisms [1]. Rodents, lagomorphs, and ticks are the main reservoirs. Ticks and biting flies are the most important vectors of transmission. Animal bites, contaminated water, processing or touching cadavers of wild game (especially hares), and tick bites are frequent routes of transmission. In Slovakia, a neighboring country to Hungary, it was found that 12.8% of tularemia cases were tick-borne [2]. There are 3 subspecies of *Francisella tularensis*; *holarctica* has less virulence in comparison with subsp. *tularensis*, which is missing from Europe but common in North America.

There are 6 major patterns of illness: ulceroglandular, glandular, oculoglandular, pharyngeal, typhoidal, and pneumonic [1]. Tick-borne spread is a leading form of transmission of ulceroglandular disease, the most common subtype of tularemia [3].

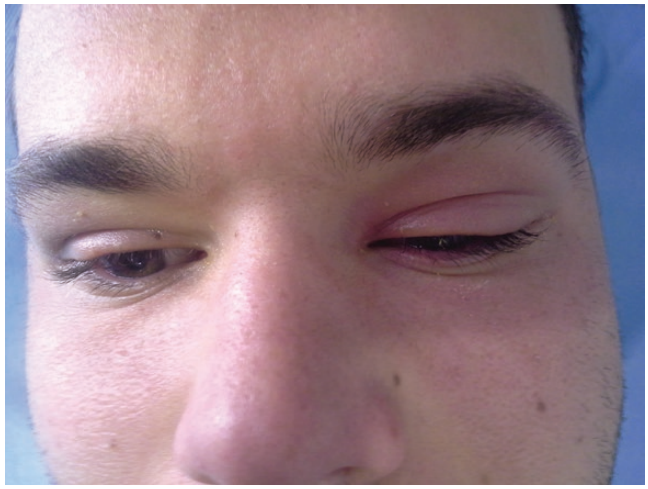

Figure 1. Oculoglandular tularemia; before initiating doxycycline treatment.

Received 29 May 2020; editorial decision 5 August 2020; accepted 12 August 2020.

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DOI: 10.1093/ofid/ofaa363

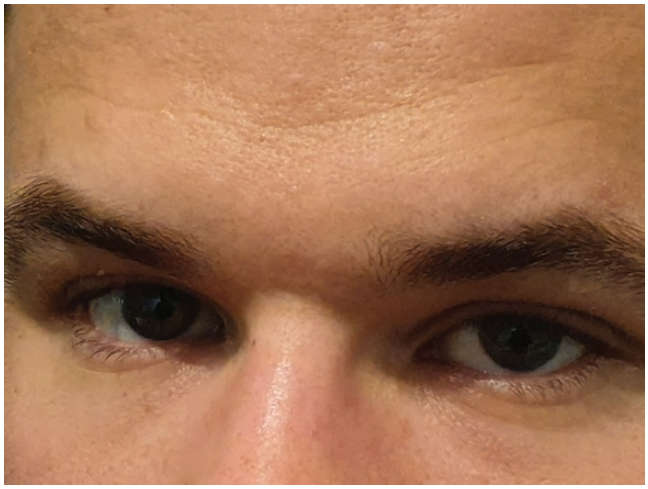

Figure 2. Oculoglandular tularemia; 2 weeks after initiating doxycycline treatment.

In 2018, 18 countries in the European Union/European Economic Area reported 441 cases of tularemia, 358 (81%) of which were confirmed [4]. The overall notification rate was 0.07 cases per 100 000 population. The incidence in Hungary varied between 17 and 140 cases per year during the period of 2014–2018, which means 0.2–1.4 cases per 100 000.

We found only 1 retrospective study [5] dating back 72 years, analyzing 1374 tularemic cases in Japan, where 5 similar cases were mentioned. This paper does not describe the route of infection, but crushed ticks removed from domestic dogs were the source.

In a recent study in which the role of domestic dogs in tularemia was examined, only 4 of the 1814 tularemia cases were related to contact with infected ticks, but none of them were oculoglandular [6]. Oculoglandular tularemia accounts for <5% of all cases and is the rarest of the subtypes [3]. Untreated tularemia can be fatal in North America [7], but in Europe mortality is exceptional; we could not find any such published case. The oculoglandular form can result in loss of vision [8]. Our case suggests that if there is the slightest suspicion of tularemia, effective antibiotic treatment should be started without any delay, long before the diagnostic serological test result is available. The first-line treatment of tularemia is streptomycin and gentamycin, especially in serious cases (if the patient has no meningitis). These antibiotics are preferred in the United States as cases are more serious there. In milder cases, quinolones and doxycycline are the drugs of choice. As our patient has no serious general symptoms and we treated him in the very early phase of his infection, we used doxycycline because of its favorable side effect profile [1]. For local treatment, gentamycin, ciprofloxacin, and tobramycin eye drops may help the healing process [8, 9].

Our patient has an elevated serum (mainly unconjugated) bilirubin level. Although tularemia may complicate granulomatous hepatitis [10], in our patient this was not the case, as he had no elevated liver enzymes. Later, in a repeated test, results showed similar bilirubin levels with otherwise normal hepatic function. Based on these characteristics, we diagnosed him with Gilbert's syndrome, which had no connection to the tularemia. Gilbert's syndrome is a common hereditary benign condition that affects the bilirubin conjugation pathway with no liver damage.

Acknowledgments

Patient consent statement. The patient whose photograph is included in this paper gave written consent to Dr. Lakos to use the photo in which his eyes appear and describe his illness in a way in which his identity is not revealed. The written and signed consent form is on file with the corresponding author (Dr. Lakos) of this manuscript.

Ethical approval. The manuscript conforms to the standards currently applied in Hungary. The study does not include factors necessitating ethical committee approval.

Potential conflicts of interest. The authors have no conflict of interest, and the work has not been supported by any firm. All authors have submitted the ICMJE Form for Disclosure of Potential Conflicts of Interest. Conflicts that the editors consider relevant to the content of the manuscript have been disclosed.

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