A 41-year-old Somalian female inhabitant of the Netherlands presented with malaise and cervical lymph node swelling. Enlarged mediastinal, hilar and abdominal lymph nodes were found on CT scan. Subsequently the left lacrimal gland became swollen, accompanied by periostitis of the lateral orbit margin. *Mycobacterium tuberculosis* was cultured from lymph node tissue and the diagnosis of tuberculous dacryoadenitis with periostitis was made on CT images and histology. All lesions responded well to tuberculostatic treatment. Although tuberculous dacryoadenitis is a very rare manifestation of tuberculosis, it is still important to recognise this presentation, especially since the incidence of tuberculosis continues to increase in Western countries.

**CASE REPORT**

A 41-year-old Somalian woman, who had been living in the Netherlands for two years, presented at the internal medicine outpatient clinic with abdominal pain, nausea and anorexia. Her weight had decreased by about 10 kg in the past six months. She had no complaints of night sweats or fever. Six months earlier she had been treated with antibiotics and a proton pump inhibitor for a *Helicobacter pylori*-positive duodenal ulcer. This resulted in a clinical response of only short duration. Physical examination showed an adipose woman with two slightly enlarged cervical lymph nodes and epigastric pain on palpation. Further examination was unremarkable.

Blood tests demonstrated microcytic red blood cells (MCV 79 fL), without anaemia, and slightly elevated amylase of 290 U/L (normal <220 U/L), alkaline phosphatase of 133 U/L (normal 120 U/L) and γ-glutamyltransferase of 64 U/L (normal 45 U/L). ESR was 20 mm in the first hour and the white blood cell count was 4.4 x 10^9/L with a normal differentiation.

A gastroduodenoscopy was carried out, showing *Helicobacter*-negative bulbitis. Enlarged mediastinal and hilar lymph nodes were seen on chest X-ray and abdominal ultrasound revealed abdominal lymphadenopathy. Chest and abdominal CT scans could confirm these findings. A tuberculin skin test was positive (32 x 29 mm). Bronchial secretions did not show any acid-fast bacilli.

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A biopsy of one of the pathological cervical lymph nodes was ordered. In the mean time, our patient developed a non-tender swelling of the left orbital margin and she complained of diplopia, vertigo and headache. She had no excessive tear production, orbital oedema or redness. The neurologist and the ophthalmologist found no signs of cerebral or ocular involvement. A CT scan of the cerebrum and skull demonstrated swelling of the left lacrimal gland accompanied by soft tissue swelling outside the skull on the temporal side and bone destruction of the lateral orbit margin. Involvement of the cerebrum was suspected (figures 1a and b).

Differential diagnosis at this time consisted of tuberculous lacrimal gland involvement, lacrimal gland carcinoma and benign mixed tumour of the lacrimal gland. A biopsy showed a granuloma with Langhans'-type giant cells; cultures remained sterile. A cervical lymph node biopsy showed acid-fast bacilli and cultures revealed *Mycobacterium tuberculosis*. Sputum cultures also revealed *M. tuberculosis*, which was sensitive to all the common antituberculous drugs. In the mean time polymerase chain reaction (PCR) of the sputum for *M. tuberculosis* complex turned out to be positive. Thus, the final diagnosis was generalised lymph node and pulmonary tuberculosis with tuberculous dacryoadenitis, although a PCR of the lacrimal gland tissue was negative. The HIV test was also negative.

After the diagnosis was made on the acid-fast strains of the lymph node biopsies, the patient started treatment with a four-drug tuberculostatic regimen: pyrazinamide and ethambutol during the first two months, and isoniazid and rifampicin during a total of six months. Signs and symptoms resolved completely within a few weeks.

**DISCUSSION**

Dacryoadenitis is an inflammation of the main lacrimal gland. Pyogenic bacteria such as *S. aureus* and streptococci are the most common causes of the acute infection. Chronic infections of the lacrimal gland occur in tuberculosis, syphilis, leprosy, cysticercosis and schistosomiasis. Dacryoadenitis is a rare manifestation of tuberculosis. It was first described by Abadie in 1881. Since 1970, nine cases have been mentioned in the English literature. In a study investigating ocular involvement in 1005 patients with active systemic tuberculosis, no lacrimal gland involvement was observed. A series of 10,542 cases of tuberculosis demonstrated an incidence of ocular tuberculosis of 1.4%, but again no cases of dacryoadenitis were encountered. Females in endemic areas aged between 35 and 50 years are predominantly affected by this manifestation, although it has been described in children. It is most often found years after an old pulmonary or lymph node tuberculosis has resolved. It may, however, exist in newly diagnosed tuberculosis, even as the presenting symptom.

As in our patient, the presenting symptom is usually a painless swelling of the eyelid, mimicking a benign mixed tumour of the lacrimal gland. There may be periostitis of the orbit.
The spread of *M. tuberculosis* to the lacrimal gland is thought to be mainly haematogenous. Local spread with conjunctival tuberculosis as the source may be possible. Isolation of *M. tuberculosis* is required for the definite diagnosis, but positive cultures from lacrimal gland secretions or from fine-needle aspirations are extremely rare. Histopathological examination shows a typical granuloma, and this usually leads to the diagnosis, especially when other features of tuberculosis are present. Two histological types of dacryoadenitis can be distinguished, the sclerotic and the caseous type, the latter being extremely rare. The use of PCR for *M. tuberculosis* complex on tissue of the lacrimal gland in cases of tuberculous dacryoadenitis is not mentioned in the literature. In our patient it turned out to be negative. We speculate that tuberculous dacryoadenitis is a paucibacillary infection, comparable with cutaneous tuberculosis and tuberculids, in which the immunological reaction to the infection plays a crucial role. In individuals with highly suspected cutaneous tuberculosis, PCR for detection of *M. tuberculosis* is positive in 54 to 60%. A combination of surgery and antituberculous drugs was reported successful in literature in the past, but nowadays antituberculous drug treatment is adequate and the prognosis is excellent.

We believe that the differential diagnosis in patients with enlargement of the lacrimal apparatus should also contain tuberculous dacryoadenitis, especially when originating from endemic areas. The need for knowledge about rare manifestations of tuberculosis is becoming greater since the incidence of tuberculosis is increasing due to HIV infection and immigration of people from endemic areas to Western countries. Histopathology will usually lead to the diagnosis, while cultures rarely become positive for *Mycobacterium tuberculosis*.

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