

# Isolated Lacrimal Gland Tuberculosis:

## A Case Report

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### Abstract

**Purpose:** To report a patient with orbital tuberculosis (TB) without any history of systemic involvement

**Case report:** A 41-year-old Afghani man who presented with progressive painless swelling in lateral side of the left upper eyelid and diplopia. He did not have history of fever or pulmonary disease. Histopathology revealed a granulomatous inflammation with caseation necrosis. Microbiological studies for finding Acid-fast bacilli and Polymerase chain reaction (PCR) results were negative. The patient completely responded to a course of anti-tuberculous treatment.

**Conclusion:** Although tuberculous dacryoadenitis is a very rare manifestation of TB, it is still important to recognize this presentation, specially when patient originating from endemic area.

**Keywords:** Orbit, Tuberculosis, Pathology Tests, Polymerase Chain Reaction

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### Introduction

Orbital tuberculosis (TB) is quiet uncommon, even in endemic countries where the incidence of pulmonary and extrapulmonary TB are remarkable.<sup>1,2</sup> Although, in majority of cases primary foci of TB is found, but there are cases in whom it is not found.<sup>1-3</sup> A definite diagnosis depends on positive culture of the organism; but it is a cumbersome and time consuming process.<sup>1-3</sup> For these reasons, several attempts have been made to improve the diagnosis of Mycobacterium TB by molecular or biological techniques. In this case report we present a case of unilateral lacrimal gland TB, which has not the history and evidence of pulmonary or other site involvement.

### Methods

We encountered a 41-year-old Afghani man who has painless and progressive swelling of the lateral side of the left upper eyelid for 6 months. The patient did not have history of fever, night sweats or weight loss. He had a history of sinus surgery due to chronic sinusitis eight months ago. A firm, non-tender mass could be palpated in the lacrimal gland area and due to mass effect; downward displacement of the left globe was evident (Figure 1). The visual acuity (VA) was  $20/20$  in both eyes but the patient had severe vertical diplopia. No abnormalities were found in the anterior segment and ocular fundus examination.

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In orbital CT scan, the left lacrimal gland was enlarged with a relative well demarcated border. There was not any evidence of orbital wall erosion or periosteal reaction. There was not any calcification inside the mass (Figure 2). Orbital MRI showed a well-defined homogenous mass at the expected location of the left lacrimal gland extending inferiorly. On T1-weighted images, the gland was isointense or slightly hyperintense relative to muscle tissue and hypointense relative to fat. On the T2-weighted images, the mass showed hypointensity (Figure 3). There was mild to moderate enhancement after contrast injection.

According to clinical and imaging findings we considered a list of differential diagnosis for our patient including: benign mixed tumor

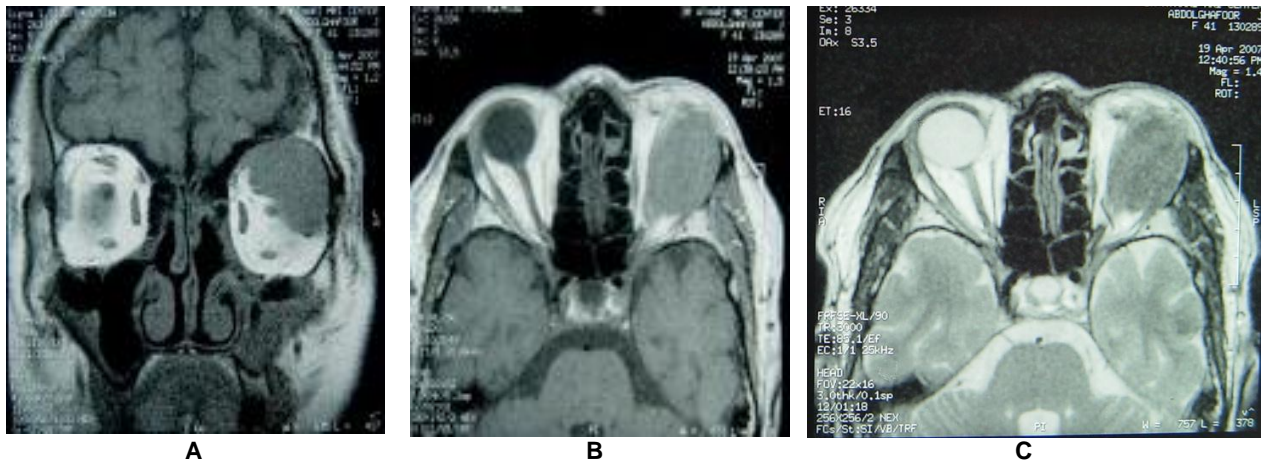
of the lacrimal gland, lacrimal gland carcinoma, lymphoproliferative involvement of lacrimal gland and inflammatory disorders. An incisional biopsy was performed through an eyebrow incision. Histologic examination showed chronic necrotizing granulomatous inflammation. Foci of caseation necrosis were surrounded by epithelioids, histocytes and some multinucleated langhans and foreign body type giant cells (Figure 4). This histologic picture was suggestive for TB; but, Zeihl-Neelsen stain for Acid-fast bacilli was negative. Also, we applied a nested polymerase chain reaction (PCR) technique with primers coding for MPB64 gene, which it is specific for Mycobacterium Tuberculosis. The PCR result was negative.



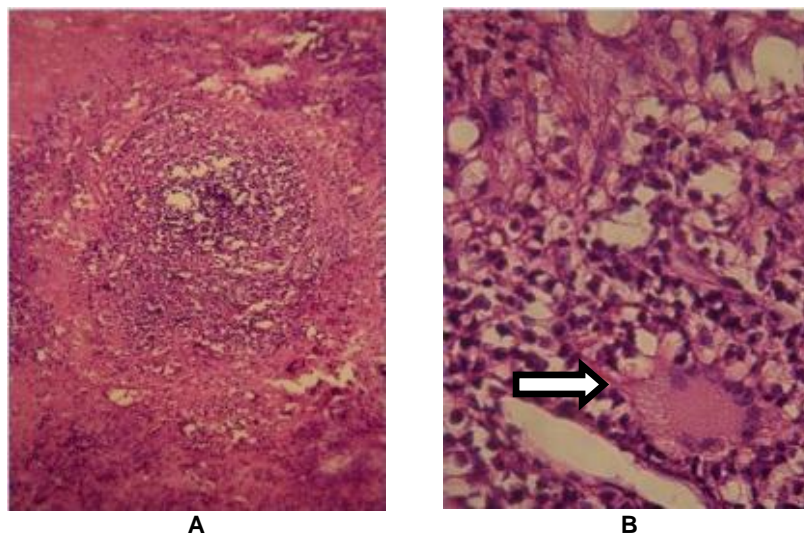
**Figure 1.** Showing swelling involving of supratemporal site and inferior displacement of the left eye



**Figure 2.** Coronal CT scan of the patient showing a soft tissue lesion in the region of the left lacrimal gland



**Figure 3.** Magnetic resonance imaging scans of the brain in coronal (A) and axial (B, C) projections, showing a homogenous orbital mass with extension to posterior and inferior



**Figure 4.** Histopathology picture showed chronic necrotizing granulomatous inflammation and Foci of caseation necrosis (A). Multinucleated Langhans and foreign body type giant cells (B)

All physical examinations were normal and there was no pathologic finding in the chest X-ray. Smear and culture from sputum was performed, but all the laboratory results were negative for Mycobacterium Tuberculosis. However, the Mantoux test was positive with an induration size of 18x20 mm.

The patient was from Afghanistan, which is one the most endemic sites of TB in the world. According to this background and significant positive result of Tuberculin test and histopathology report, anti-tuberculous treatment was started with four drugs anti-tuberculostatic regimen: pyrazinamide

(500 mg/day) and ethambutol (200 mg/day) during the first two months, and isoniazid (200 mg/day) and Rifampicin (150 mg/day) during a total of six months.

Signs and symptoms resolved completely within a few weeks after treatment.

### Discussion

In recent years, TB has known as a serious public health problem, with the potential for an increase in the prevalence of unusual pictures of disease.

Usually, primary infection develops into an asymptomatic self limited pulmonary

granuloma that resolves and remains dormant. With reactivation, the bacilli may disseminate and involve any part of the body.<sup>5,6</sup> Secondary infections may occur with or without associated pulmonary TB.<sup>1-3</sup>

The spread of *Mycobacterium tuberculosis* to the orbital cavity is thought to be hematogenous, but, local spread from sinus or conjunctival TB, may be possible.<sup>3,4</sup> Intraorbital TB presents in two forms: tubercular osteoperiostitis and orbital space occupying lesions.<sup>3,4</sup> The disease may involve soft tissue, lacrimal gland, periosteum, or bone of the orbital wall.<sup>1,5</sup> According to histopathologic finding, dacryoadenitis can be distinguished in two types: the sclerotic and the caseous type, the latter being extremely rare.<sup>4</sup>

According to the literature, Ocular TB occurs in 1.4-18% of patients with established TB.<sup>6,7</sup> Although, the majority of these are cases of asymptomatic choroiditis,<sup>7</sup> but involvement of almost every ocular and adnexal tissue has been described. Orbital TB is more commonly seen in children<sup>1,2</sup>; female are affected more than males<sup>1,2,4</sup> and the lesion is usually unilateral.<sup>8</sup> For unknown reasons the left orbital involvement is more common than the right one.<sup>1,2</sup>

In a series of 10,542 cases of TB reported from the United states, 1.4% had ocular TB, but none of the patients had lacrimal gland involvement.<sup>4</sup> In a review of 43 patients with histological evidence of orbital TB, Helms and Holland found lacrimal gland involvement in only 8 (14%) patients.<sup>8</sup> In another study on 1,005 patients with active systemic TB no lacrimal gland involvement was observed.<sup>2</sup> However, The exact incidence of intraorbital extraocular TB has not been documented.<sup>1-4</sup>

Isolation of *Mycobacterium tuberculosis* is required for the definite diagnosis of Tuberculous dacryoadenitis, but positive cultures from lacrimal gland sections or from fine-needle aspiration is extremely rare.<sup>1-5</sup> In one study even with typical histopathological features and systemic confirmation of TB, acid fast bacilli could only be demonstrated in the lacrimal gland secretions in one of the eight reported patients and the diagnosis of lacrimal gland TB remained presumptive in the remaining patients.<sup>3</sup> According to Madge et al

study, among the 79 cases of orbital TB, only 19 (24%) showed a positive culture for *Mycobacterium tuberculosis*.<sup>7</sup>

Tuberculin skin testing (Mantoux test) are commonly used to detect exposure to asymptomatic infection (latent infection) and utilized to support diagnosis of TB. Tuberculin skin test develops 2-10 weeks after the initial infection and it may not be valuable in the endemic area where a positive test is seen in 67-90% of normal healthy individuals.<sup>5</sup>

PCR is a rapid technique for TB diagnosis, with a high specificity for pulmonary (98% if Acid-Fast bacilli positive, 40-77% if Acid-Fast bacilli negative) and extrapulmonary (93.7-100%) disease.<sup>7</sup> The use of PCR for *Mycobacterium tuberculosis* complex on tissue of the lacrimal gland in suspected cases of tuberculous dacryoadenitis is reported in ophthalmic literature.<sup>2,4</sup> According to the reports PCR has been successfully used to identify 8 cases of orbital Tuberculosis<sup>7</sup>; but, it turned to be negative in our patient. Some authors speculate that tuberculous dacryoadenitis is a paucibacillary infection, comparable with cutaneous TB and tuberculids, in which the immunological reaction to the infection plays a crucial rule.<sup>9,10</sup> For example in cutaneous TB, PCR for detection of *Mycobacterium tuberculosis* is positive in only 54 to 60% of patients.<sup>9,10</sup>

In the past, combination of surgery and anti-tuberculous drugs was recommended in literature,<sup>3,5</sup> but in our patient, only anti-tuberculous drug treatment was sufficient and the prognosis was excellent.

## Conclusion

Tuberculosis is not a disease of the past. Occurrence of the extrapulmonary TB is increasing in patients with human immunodeficiency virus infection. In our opinion, the differential diagnosis in patients with enlargement of the lacrimal gland should also contain tuberculous dacryoadenitis, specially when the patient originating from endemic areas. Orbital TB should be considered in these cases, even when microbiological or PCR evidence is not forthcoming, in the initial stages.

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