Isolated Lacrimal Gland Tuberculosis: A Rare Clinical Entity in a Young Child

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Abstract: Discharging sinus in upper eyelid is one of rare manifestations of tuberculosis. The present report describes the youngest case of tubercular dacryoadenitis in an otherwise healthy child. A 4 year old male patient presented with discharging sinus in upper eyelid that developed after a spontaneously burst abscess in upper eyelid. Pus culture was sterile and the child did not improve with antibiotics. The child subsequently developed superotemporal orbital swelling and along with discharging sinus. Debridement and curettage of the necrotic tissue and sequestrum were done, histopathology showed the chronic granulomatous inflammation consistent with tuberculosis. The patient was managed with anti-tubercular therapy for 6 months. Atypical presentations of tuberculosis like discharging sinus in eyelid pose a difficult problem in diagnosis particularly in a young child. Spread of tubercular bacilli to the lacrimal gland is more commonly hematogenous or may occur directly from conjunctival tuberculosis and rarely from direct inoculation during trauma. This case report presents unusual development of tuberculosis with no other local or systemic focus of tubercular infection and any direct trauma. Tuberculosis should be kept as a possible differential diagnosis in cases of painless lid swelling, especially in a country like India where tuberculosis is endemic.

Key Words: Tuberculosis, lacrimal gland, granuloma

Introduction:
A Tuberculosis (TB) is a multisystemic infectious disease caused by Mycobacterium tuberculosis (MTB). TB is the leading infectious cause of morbidity and mortality worldwide. Approximately one third of the world’s population is infected by TB, of which 10% are symptomatic and other 90% have latent TB.[1] Multidrug resistant TB, human immunodeficiency virus (HIV) and global migration along with poor socioeconomic conditions, immunosuppression and general debility are predisposing factors leading to an increasing number of infections in both the developing and developed world.[2,3] Orbital tuberculosis is quiet uncommon, even in endemic countries where the incidence of pulmonary and extrapulmonary TB is remarkable.[4] Although, in majority of cases primary foci of TB is found, but there are cases in whom it is not found.[4,5] Tuberculous dacryoadenitis is an uncommon clinical entity with the diagnosis usually made on the basis of histopathological examination of the lacrimal gland.[6] Malignancy, developmental anomalies, and nontuberculous infections are the common orbital lesions noted in children.[7] Orbital TB may either be primary in which eye is the primary port of entry of the mycobacterium into the body, or it may be secondary as a result of hematogenous spread from a distant site. Primary TB of the eye is rare and inflammation of the uveal tract is the most common manifestation of the disease, due to its high blood supply.[8,9] Primary tuberculous dacryoadenitis is a rare entity and it is predominantly seen in females between 35 and 50 years of age, although it has been described in children.[5,10] This report describes the youngest case of isolated primary tuberculous dacryoadenitis developing in a healthy young male child with no other local or systemic focus of tubercular infection.

Case Report
A 4 years old boy presented to the Otorhinolaryngology OPD with complaints of left upper lid swelling since 5 months which was slowly progressive, and the swelling turned to an...
abscess and burst spontaneously giving rise to a discharging sinus since the last 3 months. He had received several courses of oral antibiotics from private practitioners prior to reporting to our hospital. There was no history of fever, cough or any history suggestive of rhinosinusitis. On examination the left eyelid was retracted and everted with protrusion of the lid mucosa due to edema (Figure 1). On probing of sinus tract it was extending deep to bone of orbital rim along with sequestrum. There was no limitation of ocular movements and the visual acuity was normal. Blood investigations complete hemogram, were within normal limits. On Contrast enhanced computer tomography (Figure 2) of the orbit and paranasal sinus showed bony erosion of the supero-lateral wall of the left orbital rim with mildly enhancing soft tissue component and thickening of left upper eyelid, findings were suggestive of osteomyelitis. Magnetic resonance imaging was suggestive of osteomyelitis involving the supero lateral bony orbital rim of the left orbit with intra-osseous and subcutaneous fluid collection suggestive of abscess. After pre-anesthetic work up and acquiring fitness the child was planned for debridement and curettage. Left upper lid was found retracted and everted and adherent to the underlying tissue. Sequestrum and necrotic tissue was curetted out (Figure 3). Debridement in the region of the lacrimal fossa was done, unhealthy periosteum was removed, lacrimal gland found fibrotic and send for biopsy. Histopathological examination of necrotic tissue sequestrum and gland tissue showed chronic inflammation consistent with tuberculosis. The patient was investigated for any systemic focus of tubercular infection, but the results were negative. There was no past history of tubercular infection or contact. The patient was started on anti-tubercular therapy. After follow-up of 6 months, the patient was asymptomatic, and there was no systemic manifestation of tuberculosis. The wound healed well, the lid retraction and edema resolved (Figure 4). At last follow-up he is doing well and all the signs and symptoms resolved completely.

Discussion:
Dacryoadenitis is a rare manifestation of tuberculosis. It was first described more than on century back and since 1970, less than 10 cases have been mentioned in the English literature.[5,10] Helm and Holland reviewed 43 patients with histological evidence of orbital tuberculosis and found lacrimal gland involvement in only 8 (14%) patients.[8] Van Assen et al reported tubercular dacryoadenitis in a 41-year-old Somalian female.[11] Agrawal et al found only one case of lacrimal gland involvement out of 14 cases of orbital tuberculosis in India, during a period of 5 years.[12] 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.[13] Females in endemic areas aged between 35 and 50 years are predominantly affected by this manifestation, although it has been described in children. The chronic inflammatory lesions involving the orbit are usually of unknown aetiology. Clinically, these lesions may be mistaken for neoplasm and may simulate a pseudo tumour. Most of these inflammatory reactions are non-granulomatous. Truly granulomatous lesions rarely involve the orbit and tubercular involvement is particularly very rare. The presenting symptoms of tuberculous dacryoadenitis are usually a painless swelling of the eyelid, mimicking a benign, mixed tumour of the lacrimal gland. As in our patient, the presenting symptom is usually a painless swelling of the eyelid, mimicking a benign mixed tumour of the lacrimal gland. The spread of M. tuberculosis to the lacrimal gland is thought to be mainly haematogenous. Spread to the lacrimal gland also occurs directly from primary conjunctival tuberculosis.[6] Involvement of the lateral wall of the orbit suggests a haematogenous source of infection. The predominant imaging features of orbital tuberculosis are involvement of the orbital bony wall and lacrimal gland, with soft tissue inflammatory mass/abscess formation. The acid-fast bacilli may lie dormant in the lacrimal gland and become reactivated later when the body’s resistance decreases.[6] In our patient, the lacrimal gland involvement was probably of haematogenous origin. Isolation of M. tuberculosis is required for the definitive diagnosis, but positive culture 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.[11] Sclerotic type being more common as seen in our case. We believe that the differential diagnosis in patients with enlargement of the lacrimal apparatus should also include tuberculous dacryoadenitis, especially when originating from endemic areas. The systemic history of the patient, including past history of exposure to tuberculosis suggestive of pulmonary tuberculosis and nutritional status, should be completely evaluated in patients with lesions suggestive of tubercular aetiology. Involvement of the orbit as in our case suggests a hematogenous source of infection, however in our case history, examination and investigations did not show any systemic involvement. A combination of surgery and antituberculous drugs was reported successful in literature in the past, but now a day’s antituberculous drug treatment is adequate and the prognosis is excellent.[14] The treatment of these lesions is highly successful, without any sequelae, provided prompt antituberculous therapy is instituted. The need for knowledge about rare manifestations of tuberculosis is becoming greater since the incidence of tuberculosis is increasing due to HIV infection.

Conclusion
Tubercular dacryoadenitis should be kept as a possible differential diagnosis in cases of painless lid swelling, especially in a country like ours where tuberculosis is endemic. In the light of this, it is of great importance for physicians, especially in the developing country, to be conscious of rare manifestations of tuberculosis in other parts of body.

References: