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Case Report

Acute lacrimal gland swelling with intracranial extension and without any neurological features – A rare case report

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Abstract

We report a case of young adult female with swelling in the superolateral aspect of left orbit for two weeks which was diagnosed as tubercular dacryoadenitis with intracranial extension without any neurological features. Tubercular dacryoadenitis is very rare but still makes an important differential diagnosis of lacrimal gland swellings especially in endemic areas like India. Few cases of tubercular dacryoadenitis have been reported in the past; but not a single case with intracranial extension in young adults with short history and without neurological symptoms. Although radiological investigations are routinely done in orbital lesions, it is advisable do so even in acute cases to look for intracranial extension before the appearance of neurological features. Tubercular dacryoadenitis though rare, should be kept as a differential diagnosis of acute or chronic lacrimal gland swellings in endemic areas.

Keywords: Tubercular dacryoadenitis, Intracranial extension, Acute presentation of tuberculosis

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Introduction

Tubercular dacryoadenitis is very rare but still makes an important differential diagnosis of lacrimal gland swellings especially in endemic areas like India. Tuberculosis may present as pulmonary or extra pulmonary form. Dacryoadenitis represents an extra pulmonary form of tuberculosis which may occur either because of haematogenous or direct spread from the paranasal sinuses, conjunctiva or rarely from direct trauma. Tubercular infection usually has chronic presentation but this case presented with short history of painless lacrimal gland swelling with intracranial extension without any neurological features, an unusual presentation of tuberculosis with no local or systemic focus of tuberculosis or direct trauma.

Case history

A 24 year old female presented to our hospital with heaviness and diffuse swelling in the superolateral aspect of left orbit for last two weeks. Heaviness was present all the time not associated with blurring of vision, redness, watering. On examination, the best-corrected visual acuity was 20/20 in both eyes. The patient was orthophoric, anterior and posterior segments were normal of both the eyes and ocular movements were full with no proptosis. But on palpation of the left eye, a firm swelling was found in the superolateral aspect of orbit. The swelling seemed to be arising from the lacrimal gland, it was not compressible or reducible. A provisional diagnosis of acute bacterial dacryoadenitis was made. The patient was given systemic broad spectrum antibiotics

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and anti-inflammatory drugs for a week. The patient reported after 1 week with no improvement in symptoms in fact the size of swelling had increased and had become cystic [Fig. 1]. Since no response was seen with the present treatment further investigations were done.

Complete haematological profile, liver functions, renal functions, USG abdomen, X-ray chest were done and found to be normal. ESR was raised (50 mm), Montoux was positive (20 mm) and FNAC showed chronic granulomatous reaction with multinucleated giant cells. A sample of the aspirated fluid was sent for microbiological investigations which did not report any bacteria on staining or growth on culture after 72 h. CECT orbit and Brain and PNS reported 2.6 × 1.6 cm mass lesion involving the superolateral quadrant of left orbit arising from the orbital part of left lacrimal gland with erosion of the bones of the roof and extending intracranially abutting the left frontal lobe [Fig. 2a and b]. On the basis of above findings with no response to systemic antibiotics, a high suspicion of lacrimal gland tuberculosis was made and also because of intracranial extension, anti-tubercular therapy (ATT) was started. AKT-4 kit daily was started for 2 months. It contains ethambutol hydrochloride 800 mg + isoniazid 300 mg, pyrazinamide 750 mg, and rifampicin 450 mg.

The swelling decreased significantly within three weeks of starting ATT [Fig. 3]. A repeat CECT orbit and brain was done after 6 weeks of starting ATT. Significant reduction in the size of the intracranial extension as well as the size of the lacrimal gland was reported [Fig. 4]. Also at this time growth was seen in LJ culture media which showed Acid Fast Bacilli on staining confirming our diagnosis of Tubercular dacryoadenitis [Fig. 5]. We have two molecular based tests for detection of Tuberculosis at our hospital, namely Xpert MTB/Rif Assay which is an automated in vitro diagnostic test using nested real time PCR for qualitative detection of M-TB complex and INH and RIF resistance and Genotype MTBDR plus (HAIN assay) which is a molecular genetic assay for detection of M-TB complex and RIF resistance. However WHO clearly states the recommendation that HAIN be used in Smear positive pulmonary samples; this excludes its use in our sample. WHO also specifies that the conditional recommendation with very low quality evidence for use of Xpert MTB/Rif assay in extrapulmonary samples. This leaves the option only of conventional PCR for our sample which is currently unavailable in our set up and the patient was unable to get it done outside due to financial reasons.

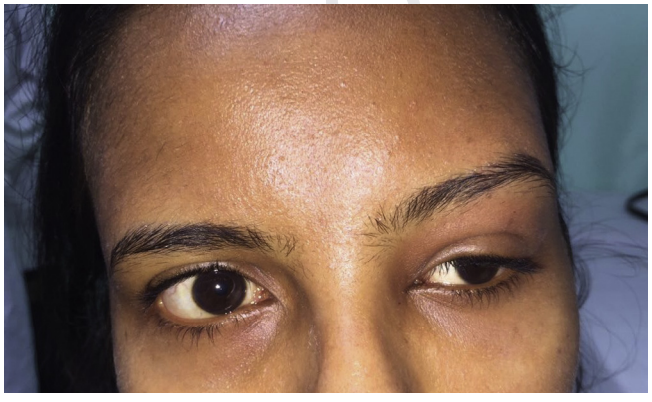

Fig. 1.

After completing 2 months of AKT 4, we started with AKT 2 (rifampicin 450 mg, isoniazid 300 mg) daily for next 6 months. Patient is responding to the treatment which is still continuing.

Discussion

Our patient had tubercular dacryoadenitis presenting as acute lacrimal gland abscess with intracranial extension. Orbital tuberculosis usually presents in five forms: classical periostitis, orbital soft tissue involvement, and cold abscess with or without bony destruction, orbital tuberculosis spread from the paranasal sinuses and tuberculous dacryoadenitis. Orbital tuberculosis has been reported as enlarging orbital lesions,[1] lacrimal abscess and discharging sinus.[2]

Dacryoadenitis can have varied aetiology. It can either be infectious caused by viruses, bacteria, fungi and parasites or inflammatory. Infectious dacryoadenitis can be acute or chronic.[3,4] Inflammatory type, usually presents as chronic dacryoadenitis as seen in sarcoidosis, Wegener's granulomatosis[5] thyroid ophthalmopathy, Sjögren's syndrome and orbital pseudotumor. Chronic dacryoadenitis may present as painless eyelid swelling which may simulate a benign mixed tumour of the lacrimal gland.

Very few cases of tubercular dacryoadenitis have been reported in literature. Out of 10,542 cases of tuberculosis reported from the United States, 1.4% had ocular tuberculosis with no lacrimal gland involvement.[6] During a five year study in India, only one case of lacrimal gland involvement was seen in 14 cases of orbital tuberculosis.[7] In another study on 1005 patients with systemic tuberculosis, lacrimal gland involvement was not observed in any of the cases.[8]

A 41-year-old Afghani man with painless and progressive lacrimal gland swelling of six months duration was also reported but no case till now of tubercular dacryoadenitis of 3 weeks duration and intracranial extension has been reported.[9] The clinical presentation can be indistinguishable from bacterial dacryoadenitis, hence, a high index of suspicion should arise in cases where antibiotics fail to produce a response. Although intracranial extension of orbital tuberculosis with extradural abscess has been reported if allowed to progress,[10] to the best of our knowledge, no case has been reported till now of acute tubercular dacryoadenitis with intracranial extension in such a short duration.

Tubercular dacryoadenitis should be kept in the differential diagnosis of acute lacrimal gland swellings without any symptoms of systemic tuberculosis as seen in our case to avoid misdiagnosis. For definitive diagnosis, PCR and isolation of Mycobacterium tuberculosis is required, but it is rare to get positive culture from lacrimal gland secretions or from fine needle aspirations. In our case too, PCR is currently unavailable in our set up and the patient was unable to get it done outside due to financial reasons and no AFB was reported on FNAC but culture on LJ media reported M. Tuberculosis after 4 weeks. Hence, whenever in doubt it is better to wait for the culture report for at least 6–8 weeks.

Erosion of roof of orbit with intracranial extension had occurred within 3 weeks of initial presentation without any neurological signs in our case, thus CECT of brain and orbit should be done in all cases of orbital swellings.

Tubercular dacryoadenitis may present as isolated acute painless lacrimal gland swellings. It is also important to note

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