Primary Tuberculous Dacryocystitis: Two Case Reports and Review of Literature

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Abstract
Tuberculosis is one of the commonest infectious diseases affecting major population of developing countries. Lacrimal sac tuberculosis is one of the rare entities. We report two such cases of tuberculous dacryocystitis where intraoperatively unhealthy looking lacrimal sac clinched the diagnosis. Histopathological features such as caseous and epithelioid granulomas were suggestive of tuberculosis which was further confirmed with systemic evaluation. Anti-tubercular therapy was started, and patients were asymptomatic on follow up visits. A systematic Medline search was done, and we could find only 15 such cases reported in literature. A review was done, and summary of all cases are included in the article.

Keywords: Tuberculous; Dacryocystitis; Lacrimal sac; Epithelioid granulomas

Introduction
Tuberculosis is an infection caused by mycobacterium tuberculosis. It is the most common health problem in the developing countries with maximum cases reported from India [1]. It is one disease which can virtually affect any organ of the body. Lung is the most common affected organ due to its method of communication [1].

Ocular tuberculosis is not an uncommon form of tuberculosis. Ocular and periocular areas are involved either primarily through direct inoculation or secondarily by either hematogenous route or through adjacent paranasal sinuses. Prevalence of ocular tuberculosis is 0.4% to 9.4% [2]. It can present either as intraocular tuberculosis with posterior uveitis as the commonest presentation or can involve the adnexal structures of the eye which commonly presents as orbital periostitis [3,4].

Lacrimal sac tuberculosis is a rare presentation of ocular adnexal tuberculosis with very few cases reported in literature. Patients with tubercular dacryocystitis have very similar presentation to a case of chronic dacryocystitis. We report two such cases of Lacrimal Sac tuberculosis where intraoperative suspicion of unhealthy looking Lacrimal sac lead to the diagnosis. Simultaneously, a review of literature has also been done and all the cases of Lacrimal Sac tuberculosis as searched from medline are included.

Case Report
Case 1
A 35-year-old male presented with watering in left eye since a year. On syringing he was diagnosed to have Nasolacrimal Duct Obstruction (NLDO). Patient was advised a Dacryocystorhinostomy (DCR) surgery and he underwent the same (Figure 1a). Intraoperatively the sac was very thick and was looking unhealthy, therefore posterior flap of lacrimal sac was sent for histopathology which showed caseous granulomatous inflammation, but no acid-fast bacilli was demonstrated. There was a positive past history of pulmonary Koch’s for which patient never completed the treatment when asked retrospectively. There was no history of cough or chest pain in the past. On laboratory evaluation all parameters were within normal limit except for raised Erythrocyte Sedimentation Rate (ESR). On chest X-ray patient had marked Bronchoalveolar marking with infiltration seen in right upper zone (Figure 1b). He was diagnosed as a case of defaulter and was started on Anti Tubercular Therapy (ATT) for which he completed the course. He was fine after 9 months of follow up.

Case 2
A 73-year-old female patient who is a known case of chronic lymphocytic leukemia presented to us with complaints of watering since a year. On clinical examination patient had lacrimal sac mucocoele for which she underwent external dacryocystorhinostomy. Intraoperatively lacrimal sac wall was very thick and friable and was looking unhealthy so was sent for histopathological examination. On Histopathology, the sac wall showed multiple lymphocytes cuffed Epithelioid Granulomas with caseating necrosis and lymphocytic infiltration in the background suggestive of tubercular changes. On systemic evaluation total leucocyte count was raised and chest X ray showed opacities in the right middle lobe of the lung. On the basis of histopathological report patient was started on ATT. She was fine after 6 months of follow up.

Discussion
Ocular adnexal tuberculosis commonly presents as orbital periostitis or discharging sinuses. The adnexal structures can get affected

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<td>3 months of second DCR</td>
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<td>AFB positive on ZN stain</td>
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<tr>
<td>7</td>
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<td>3 months</td>
<td>Left eye</td>
<td>Granulomatous tissue at inferior canal</td>
<td>No</td>
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<td>Granulomas, epithelioid cells</td>
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<tr>
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<td>Khabbaz et al 2010 <code>13</code></td>
<td>39/M</td>
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<td>3 years</td>
<td>Left eye</td>
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<td>No</td>
<td>NO</td>
<td>Left sided sinusal lymph node</td>
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<td>Lecitinal sac, bone from anterior and rhinarial arch cells</td>
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<tr>
<td>9</td>
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<td>4 years</td>
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<td>Right sided DNM</td>
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<td>No</td>
<td>No</td>
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<td>Lecitinal sac</td>
<td>Granulomatous inflammation</td>
<td>M TB</td>
<td>CT showed dilated lacrimal sac</td>
<td>6 months of ATT</td>
<td>ATT</td>
</tr>
<tr>
<td>10</td>
<td>Mohindra et al 2010</td>
<td>60/M</td>
<td>Watery eye</td>
<td>3 months</td>
<td>Left eye</td>
<td>Edematous and congested nasal mucosa</td>
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<td>No</td>
<td>No</td>
<td>Watering persisted</td>
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<td>Granulomatous inflammation</td>
<td>M TB</td>
<td>CT showed dilated lacrimal sac</td>
<td>6 months of ATT</td>
<td>ATT</td>
</tr>
<tr>
<td>11</td>
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<td>25/F</td>
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<td>1 year</td>
<td>Right eye</td>
<td>WNL</td>
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<td>No</td>
<td>No</td>
<td>Watering persisted</td>
<td>Lecitinal sac</td>
<td>Granulomatous inflammation</td>
<td>M TB</td>
<td>CT showed dilated lacrimal sac</td>
<td>6 months of ATT</td>
<td>ATT</td>
</tr>
<tr>
<td>12</td>
<td>Suresh et al 2010</td>
<td>60/F</td>
<td>Watery eye</td>
<td>2 months</td>
<td>Right eye</td>
<td>WNL</td>
<td>No</td>
<td>No</td>
<td>No</td>
<td>Diabetes</td>
<td>No surgery done</td>
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<td>CT scan showed medial orbital blood clotting</td>
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<td>13</td>
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<td>No</td>
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<td>Lecitinal sac biopsy</td>
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<td>No</td>
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<tr>
<td>14</td>
<td>Gupta et al 2017 <code>16</code></td>
<td>13/M</td>
<td>Watery eye</td>
<td>1 year</td>
<td>Right eye</td>
<td>Granulomatous tissue at inferior turbinate</td>
<td>No</td>
<td>NO</td>
<td>Nasal mucosa</td>
<td>Consisting epithelioid granulomas</td>
<td>6 months of ATT</td>
<td>Tuberculin test was positive</td>
<td>CT showed dilated lacrimal sac with bacilli changes</td>
<td>6 months</td>
<td>Patient underwent DCR post ATT and doing fine after 3 months of follow up</td>
<td></td>
</tr>
<tr>
<td>15</td>
<td>Gupta et al 2017</td>
<td>50/F</td>
<td>Watery eye</td>
<td>6 months</td>
<td>Left eye</td>
<td>Tumor mass + swollen lymph nodes</td>
<td>NO</td>
<td>NO</td>
<td>No</td>
<td>Watering persisted</td>
<td>Nasal mucosa</td>
<td>Consisting epithelioid granulomas</td>
<td>M TB</td>
<td>CT showed dilated lacrimal sac</td>
<td>6 months of ATT</td>
<td>ATT</td>
</tr>
</tbody>
</table>

WNL: Within normal limits
ATT: Anti Tubercular Therapy
M TB: M Tuberculosis
NAAT: Nucleic Acid Amplification Test

Table 1: Values
by tuberculosis either through hematogenous route or contiguous spread from the adjacent paranasal sinuses [5]. It is mainly categorized into 5 subtypes namely; classical periostitis, orbital tuberculosis or soft tissue swelling without bony destruction, orbital tuberculosis with evidence of bony destruction, orbital spread from paranasal sinuses and dacryoadenitis [3]. It is very difficult to categorize tuberculous dacryocystitis in any of these categories as there is a high possibility of missed diagnosis as all lacrimal sac specimen are not sent for histopathology. Secondly, all patients diagnosed with chronic dacryocystitis should undergo nasal endoscopic evaluation which also helps in picking up the unhealthy mucosa and sending biopsy at the same time, but it is not possible due to limited resources. Histopathology definitely plays an important role in such situations and helps the clinician to reach a proper diagnosis.

We did a complete medline search in English literature and found only 15 cases which presented primarily as tuberculosis of lacrimal sac. All cases have been published in literature presented with a similar complaint of watering and discharge with no prior systemic history of tuberculosis or any other systemic involvement suggestive of immunocompromised status of the patient (Table 1). After reviewing all the cases we found a female predominance (71.4% of cases) as seen in other cases of chronic dacryocystitis too. The mean age of presentation as seen from Table is 30.9 years (4.5 years-60 years) with no specific predilection for any side (right eye was involved in 50% of cases).

The diagnosis of tubercular dacryocystitis is difficult to make because of its common presentation as chronic dacryocystitis and there-fore, there is a huge variation seen in making the diagnosis of lacrimal sac. The first case was reported in 1947 by Anderson where he got a very similar picture like Boeck’s disease which later on demonstrated as tuberculous infection [7]. Similarly, in 1961 Sigelmann et al. reported a case of a 4.5 years old child where child presented with lacrimal sac abscess within 4 days of the surgery and they demonstrated tubercular bacilli in the drained-out material [8]. Later on, in about 40% of the cases as reported, intraoperatively sac wall was sent for further histopathological evaluation which showed caseative necrotic changes along with granulation tissue. In 2 cases the patient presented with simultaneous lymphadenopathy and diagnosis was made on fine needle aspiration cytology whereas in other 5 cases patients had inflamed nasal mucosa with granulation where biopsy showed presence of mycobacterium tubercle.

In this article we are describing two different cases which had almost similar presentation of watering and discharge but with different systemic history. The first patient was a diagnosed case of pulmonary Kochs but was a defaulter as he did not complete the course of Anti Tubercular Treatment (ATT) and hence, there is a likely possibility that the lacrimal sac was involved as a secondary site of tuberculosis which has not been reported in literature so far. The second case was already an immunocompromised patient who was getting treatment for leukemia and tuberculosis has a proven affinity for such patients. These two scenarios have not been reported in literature as first case is supposed to be a case of milliary tuberculosis which affects multiple organs but lacrimal sac as the site is not reported till date and in second case due to decreased immunity, patient got affected. It has been an established fact that chances of getting tuberculosis are higher in such patients but there has been no case reported till date.

Conclusion

Lacrimal sac tuberculosis is rare but probably an under reported entity due to its similar presentation like a case of chronic dacryocystitis. Treatment in most of the cases remains Dacryocystorhinostomy and high index of suspicion should be kept in mind specially in endemic regions and unhealthy looking lacrimal sac. In developing countries there is a high possibility of missed diagnosis as all lacrimal sac specimen are not sent for histopathology. Secondly, all patients diagnosed with chronic dacryocystitis should undergo nasal endoscopic evaluation which also helps in picking up the unhealthy mucosa and sending biopsy at the same time, but it is not possible due to limited resources. Histopathology definitely plays an important role in such situations and helps the clinician to reach a proper diagnosis.

References