

## First case report of isolated *aspergillus* dacryoadenitis

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We report a case of isolated *Aspergillus* dacryoadenitis. A 23-year-old male presented with dull ache, diffuse swelling in superolateral quadrant of the right orbit and proptosis for 4 months. Ocular examination showed conjunctival congestion, discharge in the fornix and palpable lacrimal gland (LG) mass. Routine hematological investigations followed by computed tomography scan of orbits were done. He did not respond to a course of systemic and topical antibiotics. Lateral orbitotomy with extended lid crease incision was performed with excision biopsy of LG. Abundant blackish material was found in the LG intraoperatively. The specimen was sent for histopathological examination (HPE). HPE report showed *Aspergillus*. Thorough ENT and systemic evaluation ruled out any other site with the fungus. To the best of our knowledge, this is the first case report of *Aspergillus* infection in LG.

**Key words:** *Aspergillus*, dacryoadenitis, lacrimal gland mass, orbit

Orbital aspergillosis is a relative uncommon orbital infection. Orbital aspergillosis usually arises from nasal and paranasal fungal sinusitis. Usually seen in immunocompromised individuals.

### Case Report

A 23-year-old male presented with dull ache, redness, diffuse swelling in superolateral quadrant of the right orbit and proptosis for 4 months [Fig. 1a]. He had no history of trauma, sinusitis, steroid intake, diabetes, and no other significant medical history. On examination, his best-corrected visual acuity of the right eye (RE) was 20/15, N6 and left eye (LE) was 20/20, N6. RE examination showed diffuse right upper lid edema. A firm, tender mass was palpated in the superotemporal quadrant of the orbit. Hertel's exophthalmometry showed 2 mm proptosis of the RE with 1 mm inferior displacement.

Extraocular movements were full. Slit lamp examination of RE revealed conjunctival congestion with discharge and rest of the anterior segment examination was within normal limit. Fundus examination was normal. LE examination was unremarkable. General and systemic examinations were normal. Routine hematological investigations were normal. Computed tomography (CT) scan showed a 17 mm × 19 mm × 20 mm well-defined, homogenous, enhancing lacrimal gland (LG) mass with no indentation of the globe [Fig. 1b-d]. No calcification or bony erosion was noted. Acute dacryoadenitis was suspected and was given a course of systemic and topical antibiotics (tablet amoxicillin and clavulanate potassium, 500 mg/125 mg and topical moxifloxacin eye drops). The patient did not respond. Lateral orbitotomy with extended lid crease incision was performed with excision biopsy of LG. Abundant blackish material was found in the LG intraoperatively [Fig. 2]. The specimen was sent for histopathological examination (HPE) in formalin and in saline for KOH preparation. HPE report showed lobules of benign LG acini surrounded by dense lymphoplasmacytic inflammation [Figs. 3 and 4]. Occasional necrotizing granulomas and pigmented island of narrow, septate, branching fungal hyphae along with few nonbudding spores suggestive of *Aspergillus* infection. The KOH preparation also showed filamentous, branching, and septate hyphae. Culture was done in sabouraud dextrose agar which isolated *Aspergillus*. Thorough ENT examination was done but did not reveal any evidence of sinus pathology. A diagnosis of isolated LG aspergillosis was therefore made. We started patient on systemic antifungals, i.e., tablet Itraconazole 200 mg TDS for first 3 days followed by maintenance dose twice day for 3 months. The patient was kept under follow-up of 1, 2, 6 weeks.

### Discussion

Our patient had clinical features of dacryoadenitis. The etiology of dacryoadenitis can be infectious and inflammatory. Infectious causes are viruses, bacteria, fungi, and parasites.<sup>[1]</sup> Though infectious causes include fungi, isolated fungal dacryoadenitis has never been reported.

*Aspergillus* is a ubiquitous fungus found particularly in soil and decaying vegetation. There are four main types of *Aspergillus*, i.e. *Aspergillus fumigatus*, *Aspergillus flavus*, *Aspergillus lentulus*, and *Aspergillus nidulans*. Of which *Aspergillus fumigates* and *A. flavus* are the most common fungal contaminants of the sinuses, and thus have the potential to infect the orbit. Cases of invasive sino-orbital aspergillosis in healthy individuals have often been reported from Sudan, India, and other tropical areas.<sup>[2]</sup> Since these areas share a hot and humid climate favorable for fungal

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Quick Response Code:	Website: www.ijo.in
	DOI: 10.4103/0301-4738.187678

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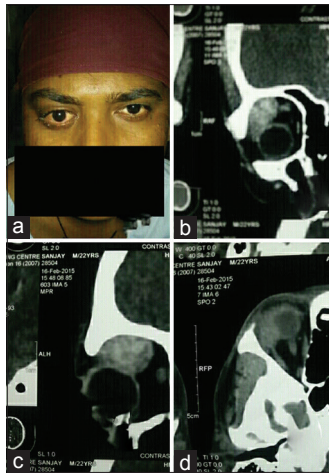
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Manuscript received: 02.09.15; Revision accepted: 20.05.16

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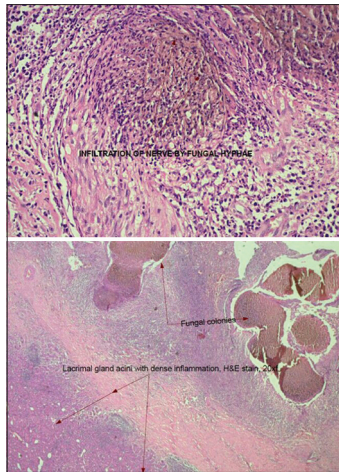
**Cite this article as:** Acharya I, Basa D, Kavitha M. First case report of isolated *aspergillus* dacryoadenitis. Indian J Ophthalmol 2016;64:462-4.



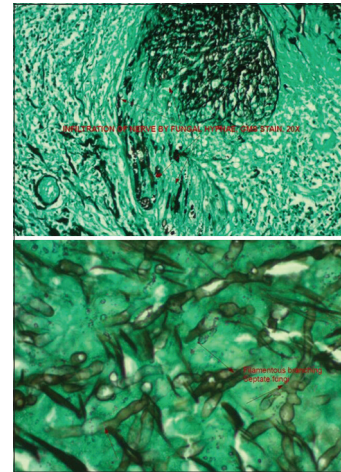
**Figure 1:** (a) Clinical photograph of the patient with right eye proptosis inward and downward. (b-d) Well-defined, homogenous, enhancing lacrimal gland mass with no indentation of the globe and without calcification



**Figure 2:** Intraoperative photographs showing exposure and excision of the lacrimal gland and black material within the lacrimal gland mass



**Figure 3:** Lacrimal gland acini with dense inflammation and fungal colonies (H and E)



**Figure 4:** Infiltration of nerve by fungal hyphae and filamentous branching septate fungi (Gram-stain)

growth and possibly a larger number of fungal spores in the environment, even healthy individuals are exposed to fungal spores resulting in increased risk of sino-orbital aspergillosis. Nevertheless, it is most commonly seen in immunocompromised hosts. Risk factors for aspergillosis are summarized in Table 1.

The spectrum of *Aspergillus* infection is complex and may present in four fundamental patterns:<sup>[5]</sup> Allergic, noninvasive, invasive, and fulminant. In nonimmunocompromised individuals, two modes (allergic, noninvasive) of presentation of *Aspergillus* are typically observed, both of which are usually less aggressive. Allergic *Aspergillus* sinusitis is a cause of chronic sinusitis seen in atopic but otherwise normal individuals<sup>[6]</sup> and is thought to be a combination of type I and type III immunological reactions to *Aspergillus* antigens via granulomatous inflammation. Noninvasive disease results in the formation of an aspergilloma (a fungus ball) and behaves like chronic sinusitis.<sup>[7]</sup> In contrast, immunocompromised patients typically demonstrate either invasive aspergillosis,

**Table 1: Risk factors for aspergillosis<sup>[3,4]</sup>**

Total neutrophils <1000/mm
T-cell defects, for example, AIDS
Defects of phagocytosis
Hematologic malignancy
Steroids or other immunosuppressive agents
Diabetes mellitus
Prosthetic devices or trauma
Excessive environmental exposure (e.g., nearby demolition or restoration of buildings, yard work, compost heaps)
Residence in endemic area (e.g., Sudan)
Advanced age

which is associated with granulomatous inflammation and fibrosis, or fulminant aspergillosis, which is characterized by diffuse vascular invasion, thrombosis, and ultimately tissue necrosis.<sup>[7]</sup> Showers of *Aspergillus* emboli from an infected valve or other source may infarct small vessels, while larger vessels may become thrombosed *in situ*.<sup>[5]</sup>

We need to put other causes of LG inflammatory diseases and neoplasms as differential diagnosis. Both CT and MRI have advantages in diagnosing orbital aspergillosis. The presence of dense intraluminal calcifications on CT scan is highly indicative of aspergillosis, but their absence does not rule it out since the calcifications may be present in only 50% of infected patients. When present, calcifications with a density >2000 HU are particularly suggestive of aspergillosis.<sup>[8]</sup> Fine-needle aspiration is one of the procedures to be indicated, especially in patients too ill to undergo more definitive biopsy or when an orbitotomy is not acceptable to the patient. Orbital aspergillosis in LG has never been reported in the literature till to date. Hence, one should suspect fungal infection in a case presenting as dacryoadenitis and not responding or improving with empiric antibiotic therapy.

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

#### Financial support and sponsorship

Nil.

#### Conflicts of interest

There are no conflicts of interest.

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