Lacrimal sac diverticulum associated with Rhinosporidiosis –Atypical presentation

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Abstract

Lacrimal sac diverticulum can be congenital, inflammatory or traumatic in origin, in our case inflammation secondary to Rhinosporidiosis was the cause for development of diverticulum. Rhinosporidiosis is a chronic granulomatous infection of mucous membrane caused by Rhinosporidiosis seeberi, which is a eukaryotic pathogen belonging in class Mesomycetozoea. Here patient presented as a case of persistent cystic swelling just below the medial canthus associated with intermittent epiphora and discharge. After all required investigations, surgical excision of the diverticulum was done preserving the lacrimal sac. As Rhinosporidiosis was diagnosed on histopathological examination, post surgery dapsone therapy was given for 6 months and followed for 1 year. Till date there is no recurrence.

Keywords: Diverticulectomy, Lacrimal sac diverticulum, Lower lid mass, Rhinosporidium Seeberi

Introduction

Lacrimal sac diverticulum is not a very commonly seen condition. It is cystic outpunching and commonly communicates with lacrimal sac or canaliculi and presented as permanent or intermittent swelling near the sac or just below the medial canthus. Lacrimal sac diverticulum may be congenital in origin or in acquired cases inflammation and trauma are the main causes.(1,2) Diverticulum most commonly developed from the infer-lateral wall of the lacrimal sac as this area gives least resistance for anything to expand.(1) It appears as a cystic swelling along the infero-medial rim of orbit or medial side of lower lid.(3) Diverticulum has either open or one-way valve like communication with lacrimal sac. Usually diverticulum remains asymptomatic, but once connection between sac and diverticulum became narrow or blocked then it leads to formation of lacrimal cyst. A large diverticulum may compress the lacrimal passage and cause symptoms like epiphora, swelling, recurrent dacryocystitis and orbital cellulites. Sometimes the diverticulum becomes infected and leads to formation of secreting fistula. Dacryolith may be formed inside the diverticulum.(1) The diverticulum may be a single cyst or sometimes it may consist of 2-3 lobules.(4)

Here we are presenting a case report of lacrimal sac diverticulum presented as a lower lid cystic mass with patent lacrimal passage. Diverticulum was associated with Rhinosporidiosis, which was diagnosed histopathologically.

Case Report

A 33 year male, social worker by occupation, visited to our OPD with complains of cystic swelling on medial side of left lower lid since 3 years. Accept that swelling patient was asymptomatic for 2 years, then gradually swelling stared increasing in size and also associated with intermittent epiphora and discharge from the same eye. Patient had no history of any trauma or surgery over sac area.

On examination, there was a cystic swelling just below the medial canthus, along the infero-medial orbital rim (Fig. 1). It was an oval, non tender cystic mass measuring 4x3 cm in size, surface of mass was smooth, skin over the mass was freely movable but it appears to be adherent to underlying tissue. The cyst was reducible on applying pressure over it. On doing lacrimal passage irrigation the passage was found to be patent but slight increase in the swelling was noticed. On CT scan, it appears as a small cystic mass along infero-medial orbital rim, abutting with nasolacrimal duct (Fig. 2a). Dacryocystography with omnipaque 350mg/ml was done that showed patent lacrimal passage with pooling of dye into diverticulum (Fig. 2b). All these indicate open communication between lacrimal sac and diverticulum. On otolarygological examination his nose and throat revealed no abnormality.

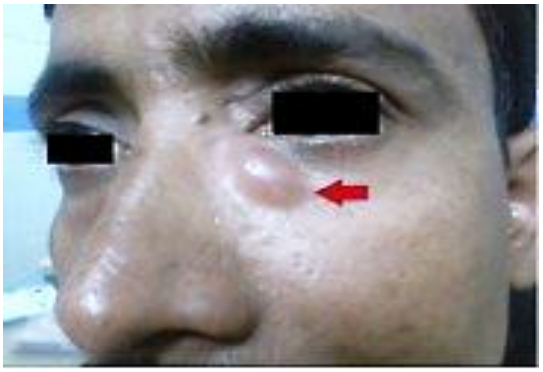

Fig. 1: Clinical photograph of patient showing lower lid mass just below the medial canthus

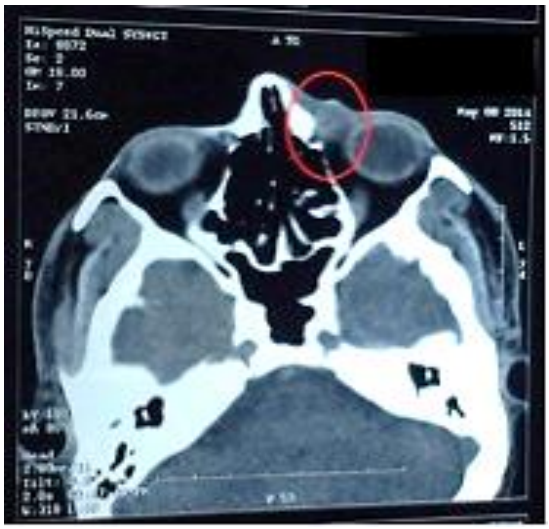

Fig. 2a: CT scan shows lacrimal sac diverticulum, abutting with nasolacrimal duct

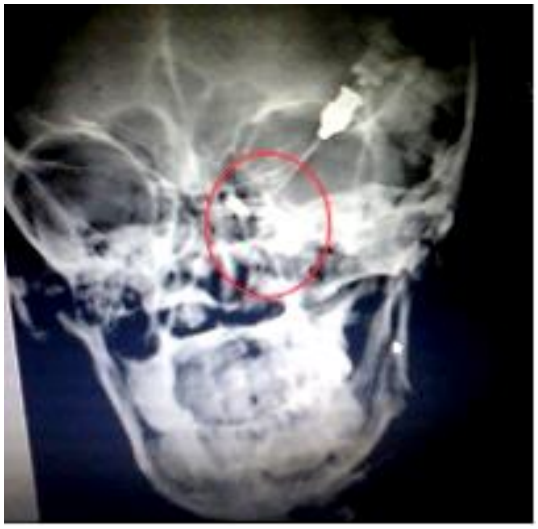

Fig. 2b: Dacryocystography shows patent lacrimal passage and communication between sac and diverticulum

Patient underwent the procedure of diverticulectomy under local anaesthesia. A curved incision medial to the swelling was given and the mass was explored. It was loosely adherent to the subcutaneous tissue and deeper structures. There was a small communication between the cystic swelling and lacrimal sac. Before excision syringing was done and then carefully the stump was tied and diverticulectomy was done, preserving the lacrimal sac. At the end of surgery lacrimal passage irrigation was done to confirm the patency of lacrimal passage and there was no leakage of fluid from the stump. The excised diverticulum was a single cyst, tan-gray in colour, measuring 1.2x 4x0.5 cm in size and filled with mucoid material (Fig. 3). On first post-operative day LPI was patent and there was no epiphora. Histopathologically lining of excised mass wall was similar with the nasolacrimal duct and lacrimal sac (Fig. 4a), also showing signs of chronic inflammation and Rhinosporidiosis infection (Fig. 4b). On the basis of histopathological finding post operatively patient was stared Dapsone 100 mg once daily for 6 months, and follow-up done monthly for 1 year, and found no recurrence.

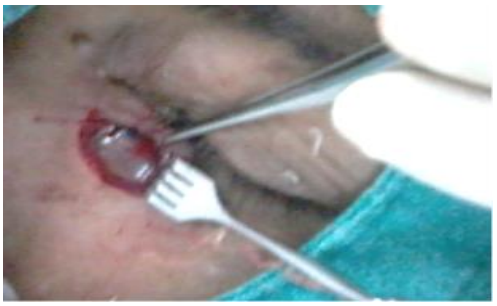

Fig. 3: Intraoperative photograph showing cystic mass

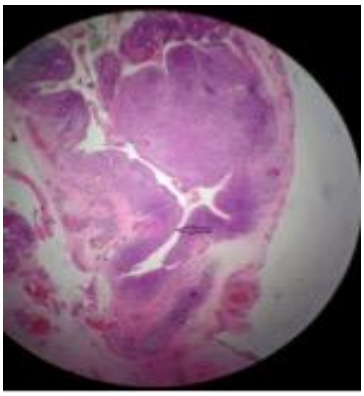

Fig. 4a: Histopathological examination of the mass (H &E, 10X). Arrow points to the pseudo stratified epithelium

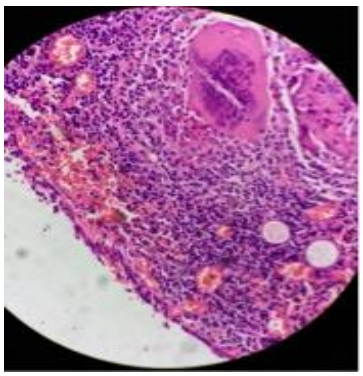

Fig. 4b: Histopathological examination of the mass (H&E, 40X) shows spoangia containing spores

Discussion

Lacrimal diverticulum involves the lacrimal sac, resulting from either a developmental malformation or an abnormality produced by local trauma and inflammation. In our case histopathological examination shows mass wall composed of pseudo stratified cuboidal and columnar epithelium, this confirms that mass is

related with lacrimal sac and nasolacrimal duct.[5] The presence of focal ulceration and stromal infiltration with plasma cells, lymphocytes, neutrophils and few eosinophils indicate chronic inflammation. Presence of numerous foreign body giant cells enclosing many sporangia with thick chitinous wall, some of which containing spores, confirmed the diagnosis of Rhinosporidiosis.

Rhinosporidiosis is a chronic granulomatous infection of mucous membrane mostly involving the nose, pharynx, oral cavity, conjunctiva, rectum and external genitalia caused by the organism Rhinosporidium Seeberi. Rhinosporidium Seeberi was initially thought to be a fungus but now classified in a new class, the Mesomycetozoea, along with the organisms that cause similar infections in amphibian and fish.[7] It usually manifests as vascular friable polyps that arises from the mucous membrane and on histopathology, large chitinous lesions filled with spore like bodies are seen.[6]

Ocular rhinosporidiosis also known as Oculosporidiosis commonly affects conjunctiva, lacrimal sac, canaliculi, lid and sclera.[7] Involvement of Lacrimal sac is mostly because of ascending infection from the nose and infection spreads diffusely underneath the skin.[8] Rhinosporidiosis of lacrimal sac manifest as a swelling over the sac area, lower lid mass, sac diverticulum, recurrent bleeding from nose.[9]

CT scan and dacryocystography shows connection between diverticulum and lacrimal passage, imaging techniques are also necessary for the proper diagnosis of lacrimal sac diverticulum and its management.[3,10]

Treatment of choice for lacrimal sac rhinosporidiosis is dacryocystectomy. In our case there was no history of epistaxis from nose and, or serosanguineous discharge from eye, the mass had cystic feeling and on otolaryngological examination revealed no pathological finding in nose and throat. As there was no clinical suspicion for Rhinosporidiosis, instead of dacryocystectomy, Diverticulectomy was done to preserve the lacrimal sac. It was diagnosed only by histological examination.

Conclusion

In case of Acquired lacrimal sac diverticulum, along with other causes we have to look for rhinosporidiosis even though patient does not present typical features of the Rhinosporidiosis. By doing proper evaluation and appropriate investigations, we can save the sac if the passage is patent.

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