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Lacrimal Sac Rhinosporidiosis: An Unusual Case Report

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Abstract Rhinosporidiosis is a granulomatous disease that is a disease caused by Rhinosporidium seeberi. Though it occurs universally, the disease is widely prevalent in the tropics, especially in southern India and Sri Lanka. It usually affects the nasal mucosa and nasopharynx and rarely the conjunctiva, lacrimal sac, tonsils and skin. We present a case study of an isolated lacrimal sac rhinosporidiosis in a 15-year-old boy who was a resident of Terai. He presented with a diffuse left medial infraorbital swelling for a period of 14 months. He also complained of intermittent epiphora. External examination of the left eye revealed a diffuse, soft, and nontender swelling in the medial infraorbital region. Fine needle aspiration report was inconclusive. A computed tomography scan of the paranasal sinuses revealed an isodense lesion with mild enhancement within the preseptal compartment along the inferior aspect of the left orbit. A dacryocystectomy was performed and histopathology report confirmed rhinosporidiosis. There is no recurrence after 2 years of follow up.

Keywords: *Rhinosporidiosis, lacrimal sac, Rhinosporidium seeberi*

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1. Introduction

Rhinosporidiosis is a granulomatous disease that is caused by *Rhinosporidium seeberi*. [1] Initially described by Seeber in 1900 in an individual from Argentina, [2] rhinosporidiosis is endemic in India, Sri Lanka, South America, and Africa. [3,4,5,6] Cases from the United States and Southeast Asia, as well as scattered occurrences throughout the world, have also been reported. [3,4,5,6] The disease is widely prevalent in the tropics, especially in southern India and Sri Lanka. [6] In addition to humans, disease has been noted in cats, cattle, dogs, ducks, goats, horses, mules, parrots, and swan. [7] It usually affects the nasal mucosa and nasopharynx and rarely the conjunctiva, lacrimal sac, tonsils and skin. [8] The clinical presentation is usually the pinkish polypoidal bleeding mass in the nasal cavity. This is a case study of an isolated lacrimal sac rhinosporidiosis in a 15-year-old boy which is very unusual presentation of rhinosporidiosis.

2. Case Report

A 15 yrs old male presented to the ENT OPD with left medial infraorbital swelling for a period of 14 months. He also complained of intermittent epiphora. External local examination revealed a diffuse, soft, and nontender swelling in the medial infraorbital region. (Figure 1) Pressure over the mass produced a reddish mucopurulent discharge from the lower punctum. The overlying skin was normal. The vision and the extraocular movements were normal. There was partial patency of the left nasolacrimal passage which was ascertained by some regurgitation of fluid on performing sac syringing. A computed tomography scan of the paranasal sinuses revealed an isodense lesion with mild enhancement within the preseptal compartment along the inferior aspect of the left orbit (Figure 2). Fine needle aspiration report was nonconclusive.

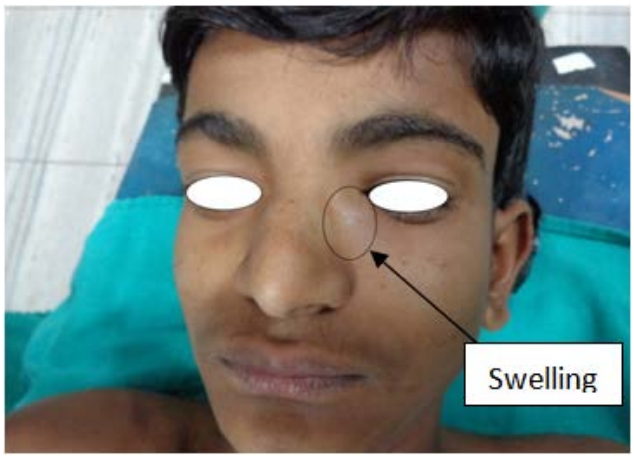

Figure 1. Clinical photograph showing the lesion

Patient was planned for excision of the mass. Since the diagnosis was still not clear it was confirmed intraoperative by opening the lacrimal sac. Multiple tiny pink, vascularized growth with papillary excrescences extension were seen (Figure 3 & Figure 4).

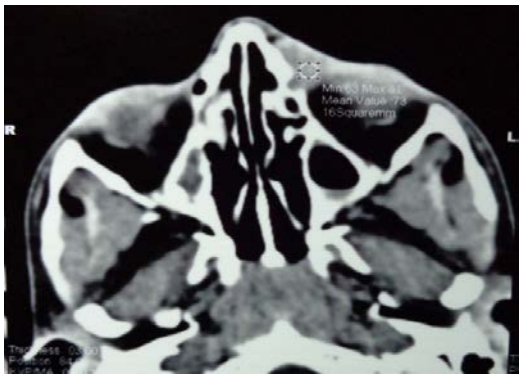

Figure 2. An axial CT scan of paranasal sinuses showing isodense lesion within left inferior aspect of the preseptal compartment

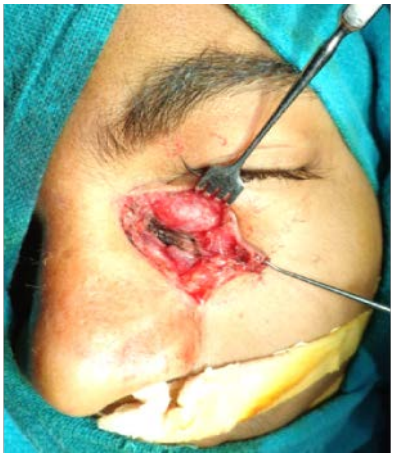

Figure 3. Intraoperative photograph showing lacrimal sac

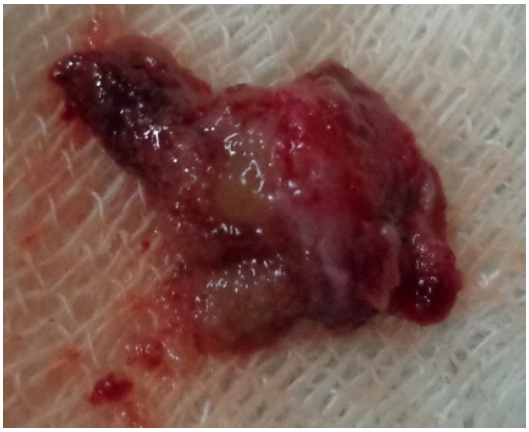

Figure 4. Gross photograph of specimen (incised lacrimal sac)

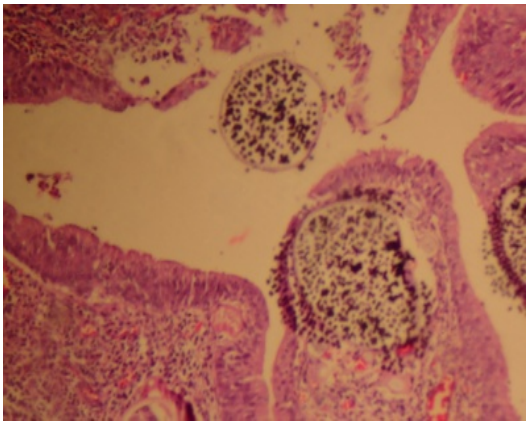

Figure 5. Histological section showing hyperplastic epithelium, multiple sporangia and trophocytes in the subepithelial region (H & E stain; X 100 magnification)

The sac was sutured and dacryocystectomy was performed. Care was taken to avoid spilling of spores during complete removal of the mass. Extension of growth in nasolacrimal duct were also removed en bloc along with the sac. The nasolacrimal duct wall was curetted after removal of growth. Specimen was sent for histopathology, bacterial, and fungal studies. The histopathological report was suggestive of rhinosporidiosis (Figure 5).

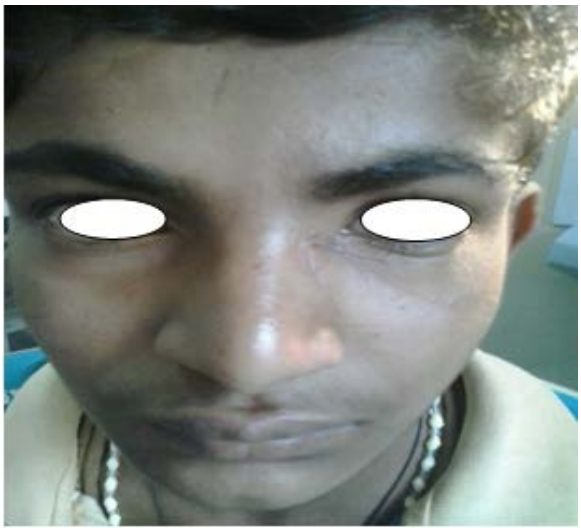

Figure 6. Postoperative photograph at 2 years follow up

Patient was kept on daily 100mg oral diaminodiphenylsulfone (DDS) (Dapsone) for 3 months. After 2 years of follow up there is no recurrences (Figure 6).

3. Discussion

Rhinosporidiosis is presumably a waterborne disease, caused by *Rhinosporidium seeberi.* Though it occurs universally, 88% of cases are reported from India and Sri Lanka. [6] It is an aquatic protistan parasite in taxonomic classification of recent studies. It is currently included in a new class the Mesomycetozoea. [9] It usually involves the nasal mucosa and rarely the conjunctiva, lacrimal sac, tonsils, and skin. Primary ocular rhinosporidiosis occurs in 10% of cases. [10] The conjunctiva was the most common site of infection in 76 (92.68%) of the cases. The lacrimal sac was affected only in six (7.32%) cases. [11] The pathway of transmission of rhinosporidiosis remains unclear. As the most affected sites are nose and eye, it has been suggested that infection occur while bathing in common pond i.e. water borne. It is true for our case also as there is history of taking bath in small pond. In this case, lacrimal sac alone was involved, it being a hollow protected viscous, the infection either would have reached the sac from nose or eye through lacrimal canaliculi without affecting the nose or conjunctiva. [12] Epiphora is unusual in nasolacrimal rhinosporidiosis because the spread of infection is pericanalicular and perisaccular. [8] However, our patient had complaints of intermittent epiphora with partially blocked nasolacrimal apparatus. Dacryocystectomy has been described in the literature for lacrimal rhinosporidiosis [11]; this case was also managed by dacryocystectomy. It is essential to administer Dapsone postoperatively to tackle the local subepithelial and

subcutaneous spread and to prevent recurrence [13]. The role of Dapsone in reducing the rate of postoperative recurrence is attributed to an arrest of maturation of the spores and an accentuated granulomatous response with fibrosis after Dapsone therapy [14].

4. Conclusion

Though lacrimal sac rhinosporidiosis is rare, in all cases of medial infraorbital pathology, one needs to keep a clinical diagnosis of lacrimal sac rhinosporidiosis in mind whenever patient belonging to tropical endemic region.

References

[1] Ashworth, J.H., On Rhinosporidium seeberi with special reference to its sporulation and affinities, *Transactions of the Royal Society of Edinburgh*, 53, 301-42, 1923.

[2] Seeber, G.R., Un neuvo esporozoario parasito del hombre: dos casos encontrades en polipos nasales. *Thesis, Universidad Nacional de Buenos Aires*, 1900.

[3] Gaines, J.J. Jr, Clay, J.R., Chandler, F.W., Powell, M.E., Sheffield, P.A., Keller, A.P. 3rd, Rhinosporidiosis: three domestic cases. *South Med J*, 89(1), 65-7, 1996.

[4] Hospenthal, D.R., Uncommon Fungi and Prototheca. In: Mandell, G.L., Bennett, J.E., Dolin, R., eds. *Mandell, Douglas, and Bennett's Principles and Practice of Infectious Diseases*. 7th ed. Philadelphia, Pa: Churchill Livingstone Elsevier; 3365-3376, 2010.

[5] Kwon-Chung, K.J., Bennett, J.E., Rhinosporidiosis, In: *Medical Mycology*, Philadelphia, Pa: Lea & Febiger, 695-706, 1992.

[6] Karunaratne, W.A.E.: *Rhinosporidiosis in Man*, Athlone, University of London, 1964.

[7] Kennedy, F.A., Buggage, R.R., Ajello, L.. Rhinosporidiosis: a description of an unprecedented outbreak in captive swans (Cygnus spp.) and a proposal for revision of the ontogenic nomenclature of Rhinosporidium seeberi. *J Med Vet Mycol*. May-Jun 1995; 33(3):157-65.

[8] Nerurkar, N.K., Bradoo, R.A., Joshi, A.A., Shah, J.,Tandon, S., Lacrimal Sac rhinosporidiosis: A case report. *Am J Otolaryngol*, 25, 423-6, 2004.

[9] Fredricks, D.N., Jolley, J.A., Lepp, P.W., Kosek, J.,Relman D.A., Rhinosporidium Seeberi: A human pathogen from novel group of aquatic protistan parasites, *Emerg Infect Dis*, 6, 273-82, 2000.

[10] Mukherhee, P.K., Rhinosporidiosis (oculosporidiosis), Current Ocular therapy. Philadelphia, PA, Saunders, 77-79, 1995.

[11] Shrestha, S.P., Hennig, A., Parija, S.C., Prevalence of rhinosporidiosis of the eye and its adnexa in Nepal, *Am J Trop Med Hygiene*, 59, 231-34, 1998.

[12] Jain, S.N., Aetiology and incidence of rhinosporidiosis, *Indian J Otolaryngol*, 19, 1-21, 1967.

[13] Nair, K., Clinical trial of diaminodiphenylsulphone (DDS) in nasal and nasopharyngeal rhinosporidiosis, *Laryngoscope*, 89, 291-95, 1979.

[14] Job, A., Venkateswaran, S., Mathan, M., Krishnaswami, H., Raman, R., Medical therapy of rhinosporidiosis with Dapsone, *J Laryngol Otol* 107, 809-12, 1993.