

Rhinosporidiosis of the lacrimal sac in a tertiary care hospital of India - A retrospective case study

Sharmistha Behera, Ravindra Kumar Chowdhury, Jayashree Dora

Purpose: Though rhinosporidiosis of the lacrimal sac is a rare disease across the globe, the frequency with which these patients come to the outpatient department in western Odisha is quite alarming. This study was undertaken to upgrade the knowledge about the clinical profile and management of rhinosporidiosis of the lacrimal sac. **Methods:** This is a retrospective study comprising 32 clinically diagnosed and histopathologically proved cases of lacrimal sac rhinosporidiosis who were managed with dacryocystectomy with meticulous excision. Intraoperative copious irrigation with 5% povidone-iodine for 5 min and postoperative dapsone therapy for 3–6 months had been administered to all the patients. The mean follow-up period was 16.7 months. The study was conducted over 5 years from August 2015 to July 2020. **Results:** *Rhinosporidium seeberi*, an aquatic protistan parasite, was found to be the causative agent. Males and females were affected equally. Children less than 10 years of age comprised 56.2% (18 cases). History of pond bathing was found in 100% of cases. The most common presentation was boggy swelling over the lacrimal sac. The involvement was unilateral in all the cases. None of the patients were found to have nasal involvement. In 65.6%, the lesion was limited within the sac. Recurrence was noted in 25% of cases. **Conclusion:** Rhinosporidiosis of the lacrimal sac should be excluded in all patients presenting with boggy swelling of the lacrimal sac with a history of pond bath. The recurrence can be minimized by meticulous excision, intraoperative betadine, and postoperative dapsone therapy.

Key words: Dapsone, lacrimal sac, rhinosporidiosis

Rhinosporidiosis is presumably a waterborne disease-causing granulomatous infection of the mucous membrane.^[1] The causative agent, *Rhinosporidium seeberi*, is an aquatic protistan parasite, located phylogenetically between the fungal animal divergence.^[2] It belongs to a novel group of fish parasites that infect fish as well as amphibians. The nose and nasopharynx (78%) are the most common sites of affection in humans, followed by conjunctiva and lacrimal apparatus (15%).^[3] Twenty-six percent of ocular rhinosporidiosis occurs in the lacrimal sac either with conjunctival or nasal involvement.^[4] Conjunctival polyp (77.6%) is the most common ocular manifestation. The southern part of India and Sri Lanka are the endemic zones of rhinosporidiosis due to their hot tropical climate.^[5] In India, the ocular manifestation of rhinosporidiosis is rarely encountered outside the coastal areas of Kerala, Tamil Nadu, and Puducherry. However, West Bengal, Madhya Pradesh, Rajasthan, Odisha, Bihar, and Maharashtra are the places where sporadic occurrence has been reported.^[6–8] The climate in Odisha is moderate; it lies on the eastern coast of India and comes wholly under the tropical zone. However, extreme climatic conditions are experienced in its western districts such as Bolangir, Sambalpur, and Sundergarh. The summer temperature in this part varies from 20°C to 45°C, while in winter, it is between 13°C and 32°C.^[8]

Department of Ophthalmology, V.S.S. Institute of Medical Sciences and Research, Burla, Sambalpur, Odisha, India

Correspondence to: Dr. Ravindra Kumar Chowdhury, Assistant Professor, Department of Ophthalmology, V.S.S. Institute of Medical Sciences Research, Burla, Sambalpur - 768 017, Odisha, India. E-mail: ravindrachowdhury@gmail.com

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We present the clinical profile, histopathology, and management of 32 cases of lacrimal sac rhinosporidiosis in a tertiary care hospital that mainly caters to the western districts of Odisha, India. This is the first reported retrospective study of lacrimal sac rhinosporidiosis in this part of India as per the literature review. Unilateral presentation, non-involvement of adjacent structures such as conjunctiva or nasal mucosa, and history of pond bath in all the cases are interesting to note.

Methods

Thirty-two clinically diagnosed and histopathologically proved cases of lacrimal sac rhinosporidiosis who were managed with dacryocystectomy with meticulous excision were included in the study. This is a retrospective study carried out between August 2015 and July 2020. A team of three ophthalmologists and two pathologists were the researchers. To minimize the interobserver variation, a standard clinical definition with histopathological confirmation was followed. Soft, fluctuant boggy swelling of the lacrimal sac was suspected to be due to rhinosporidiosis [Fig. 1]. The clinical diagnosis was confirmed histopathologically when there was the presence of innumerable sporangia of all

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Figure 1: Boggy swelling of the lacrimal sac area in different patients

sizes in different stages of maturation in the subepithelial layer with inflammatory cells such as lymphocytes, plasma cells, histiocytes, and polymorphs covered by proliferating stratified columnar epithelium [Fig. 2]. Sporangia were found to have a double-layered refractile chitinous wall with sporangiospores. The detailed clinical profile, demography with special attention to the history of pond bath, and mode of management of each patient were taken into consideration. Patency of the lacrimal passage was checked. All the patients were referred to an otorhinolaryngologist to look for any nasal involvement. Dacryocystectomy (DCT) was done in all the clinically diagnosed patients. During excision of the lacrimal sac, special care was taken to avoid the spilling of spores. After complete removal of the sac, 5% povidone-iodine was applied for 5 min in all cases. Post-operatively, Dapsone in a dose of 100 mg once or twice daily for 3–6 months had been given after excluding Glucose-6 phosphate enzyme deficiency. The mean follow-up period was 16.7 months with a range of 12–36 months.

Results

In all 32 patients, the involvement was unilateral. Both right and left eyes were affected in equal numbers. The disease was more common in patients below 10 years of age [Table 1]. Males and females were affected in equal numbers.

Non-tender boggy swelling of the lacrimal sac area was the chief presentation in all 32 cases (100%). Epiphora in eight cases (25%), epistaxis in six cases (18.7%), and polypoidal growth on the swelling in one case (3.1%) were the other manifestations [Table 2]. The swelling was found to be extending both above and below the medial canthus in 28 cases (87.5%) and was limited to the area below the medial canthus in four cases (12.5%). Blockage of lacrimal passage was

Table 1: Age and sex distribution of lacrimal sac rhinosporidiosis

Age in years	Number of males	Number of Females	Total (n=32)
<10	10	8	18 (56.2%)
11-20	2	4	6 (18.7%)
21-30	2	2	4 (12.5%)
>30	2	2	4 (12.5%)
Total	16	16	32

Table 2: Clinical presentations of lacrimal sac rhinosporidiosis

Presentation	Number of cases	Percentage (n=32)
Boggy swelling over lacrimal sac area	32	100
Epiphora	8	25.0
Epistaxis	6	18.7
Polypoidal growth over the sac area	1	3.1

observed in eight cases. Nasal or conjunctival involvement was not found in any of the patients. All the patients had a history pond bath.

The lesion was confined to the lacrimal sac in 21 cases (65.6%), and extension outside the sac into the subcutaneous space was marked in 11 cases (34.4%). On dissection of the lacrimal sac, pink vascularized growth with finger-like extensions was observed in all the cases [Fig. 3]. Bleeding was remarkable during the excision of the sac. Recurrence was noted during follow-up in eight cases (25%).

Discussion

Lacrimal sac rhinosporidiosis is usually suspected in patients presenting with boggy swelling over the lacrimal sac region with a feeling of "bag of worms."^[9-11] Nuruddin *et al.*^[12] found 100% of patients having a feeling of boggy swelling over the sac area and epiphora in 22.2%. In the present study, such swelling over the lacrimal sac area was found in 100% of cases and epiphora in 25%. The spread of rhinosporidial infection is pericanalicular and perisacular.^[13] This might be the reason why only one-fourth of the patients presented with epiphora despite lacrimal sac involvement. Epistaxis in 18.7% of cases without nasal involvement might be due to trickling of blood through the nasolacrimal duct.

In the present study, lacrimal sac rhinosporidiosis was most frequent in children less than 10 years of age. This is in contrast to the findings of Nuruddin *et al.*,^[12] who had reported a higher prevalence in the age group of 25–34 years. This disparity may be attributed to the higher number of children having the habit of bathing in pond water in this part of the country.

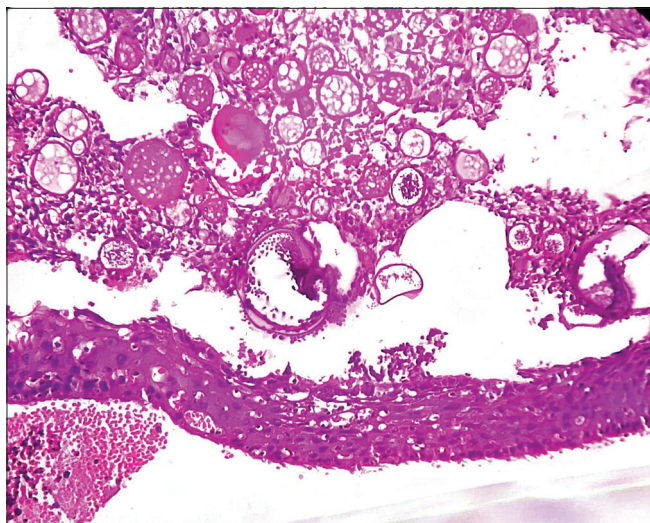


Figure 2: Histopathological picture of the lesion showing proliferating stratified columnar epithelium of lacrimal sac with sporangia of all sizes in different stages of maturation in the sub-epithelial layer with inflammatory cells (H and E stain, 200x)

Further, the narrow lacrimal drainage system in children can be a contributing factor. Males and females were found to be affected equally in our study.

Rhinosporidiosis is a waterborne disease, and the organism is suspected to be present in stagnant water, for example, in ponds. However, it may also be transmitted via air and dust.^[13,14] In our study, all of the patients had a history of pond bath, which is consistent with most of the studies. The probable route of entry of this organism to the lacrimal sac is canaliculi and nasolacrimal duct. However, none of our patients presented with conjunctival involvement or any nasal involvement. This is an interesting variation.

Radiographic investigation such as CT dacryocystography was recommended before surgery to delineate the involvement of lacrimal sac and nasolacrimal duct by Pushker *et al.*^[11] However, not all our patients were exposed to such investigations to avoid the harmful effect of radiation and dye. In some patients, we performed dacryocystography where pooling of dye was seen. In most of the cases, we assessed the involvement clinically and did a thorough curettage and cauterization of the nasolacrimal duct to prevent recurrence.

The definitive treatment for rhinosporidiosis of the lacrimal sac is dacryocystectomy (DCT).^[4,9,10,11] Modified dacryocystorhinostomy (DCR) has been tried by different authors with success to prevent postoperative epiphora.^[12,15] However, as the DCT is an apparently safe and curative procedure in lacrimal sac rhinosporidiosis, we did not attempt DCR.

It is recognized that surgical removal of the lacrimal sac is difficult due to severe bleeding during the excision of lacrimal sac rhinosporidiosis. Thus, recurrence of the lesion is frequent. The main cause of recurrence of the lesion in lacrimal sac rhinosporidiosis is the inability to remove the spores of rhinosporidium completely. Intraoperative maneuvers such as 5% betadine soakage for 5 min after excision of the sac, electric cauterization of the surrounding healthy area, and application of 1–5 mL of amphotericin B (0.15%) have been tried by different authors with success.^[16-18] Arsecularatne *et al.*^[16] reported metabolic inactivation of endospores on exposures to povidone-iodine. Postoperative dapsone therapy with 100 mg once/twice daily for 3–6 months has been found to prevent recurrence as it is known to arrest the maturation of spores and

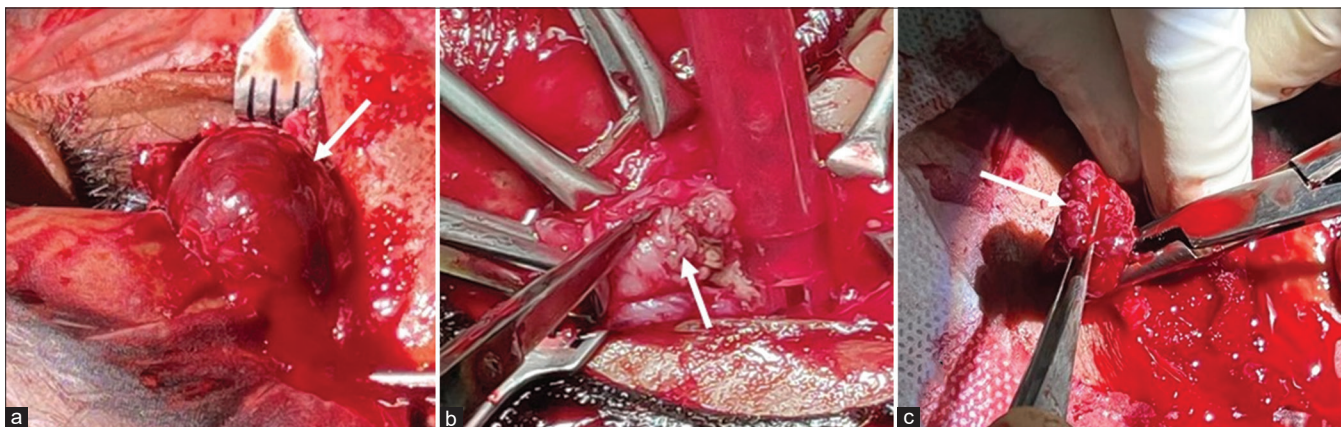


Figure 3: Intraoperative photographs of Rhinosporidiosis of the Lacrimal sac. White arrow showing (a) Pink vascularized growth; (b) Irregular finger-like extensions; (c) Vascular growth studded with gray-white dots

promote fibrosis in the stroma.^[17] This drug should, however, be used after ruling out drug allergy and G6PD deficiency.^[19]

In the present study, recurrence was observed only in eight cases (25%). The recurrence was more commonly seen when the extension was outside the sac. The result of this study does not agree with the observation of earlier studies which say recurrence is inevitable.^[9,20] We believe that intraoperative application with 5% povidone-iodine and postoperative dapsone therapy is effective in decreasing recurrence in our study. A limited period of follow-up might also be attributed to the low recurrence in our study. A larger sample size with a longer period of follow-up may be necessary to prove this fact.

Conclusion

Rhinosporidiosis of the lacrimal sac should be excluded in all the patients presenting with boggy swelling of the lacrimal sac with a history of pond bath. The recurrence can be minimized by meticulous excision, intraoperative betadine, and postoperative dapsone therapy.

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Conflicts of interest

There are no conflicts of interest.

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