

## Case Report

# Lacrimal sac rhinosporidiosis mimicking soft tissue sarcoma: a case report

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### ABSTRACT

Rhinosporidiosis is a granulomatous disease caused by *Rhinosporidium seeberi*, usually affecting the nasal mucosa. Involvement of other sites of body in rhinosporidiosis especially, the lacrimal sac involvement is very rare. Hence, we report a case of lacrimal sac rhinosporidiosis in an adult male patient who presented with infraorbital swelling.

**Keywords:** Rhinosporidiosis, Lacrimal sac, Sporangium

### INTRODUCTION

Rhinosporidiosis is a granulomatous disease caused by *Rhinosporidium seeberi*, usually presenting as polypoidal growth involving the nasal mucosa and nasopharynx.<sup>1</sup> It rarely affects other sites like lacrimal sac, tonsils and the skin. The disease is endemic in India (especially in Tamil Nadu) and Sri Lanka.<sup>1</sup>

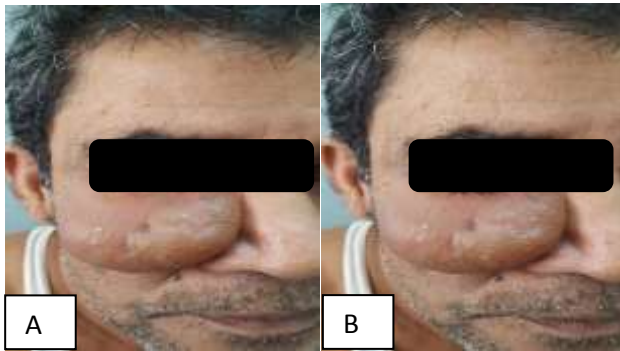
We present a case of rhinosporidiosis involving lacrimal sac and the overlying skin.

### CASE REPORT

A 58 year old gentleman presented to the department of otorhinolaryngology, VIMS and RC, with complaints of right infraorbital painless, insidious onset and progressive swelling of one year duration. For the same swelling he had undergone endoscopic dacryocystorhinostomy (DCR) under general anesthesia four months prior to the presentation. However seven days after the surgery the swelling increased in size to the extent of 5×8 cm (vertical and horizontal respectively). He also had noticed right nasal discharge on pressure over the swelling. He

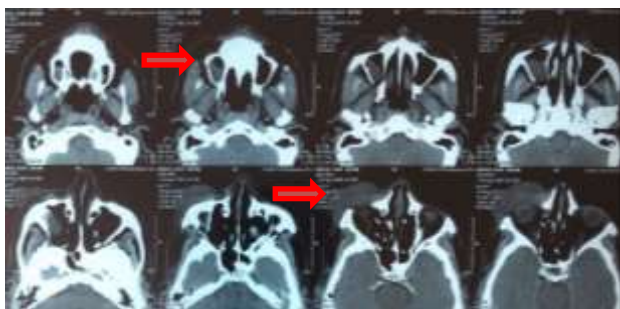
did not suffer from nose block, nose bleed, decreased sense of smell, excess watering of right eye, ear or throat related symptoms. He had never sustained trauma to facial structures. He had no comorbid conditions except for systemic hypertension for three years for which he was on regular medications.

On examination he had normal vitals and general physical examination did not reveal any clinical abnormalities. Thorough ENT examination was carried out. Anterior rhinoscopy showed deviated nasal septum to right with spur with left inferior turbinate hypertrophy. Diagnostic nasal endoscopy showed mulberry nodule at the level of axilla of right middle turbinate. Synechiae observed between nasal septum and right middle turbinate. Ears and throat were normal. A soft solitary non-tender, non-pulsatile, non-reducible but compressible swelling of 5×8 cms was noted sagging down in the right infra-orbital region extending vertically from lower eyelid down to the level of right ala of the nose. The swelling extended horizontally from the lacrimal sac area to the lateral canthus. There was Pau-de-orange appearance of lower portion of skin over the swelling. Ophthalmic examination was normal except for the above mentioned swelling in infraorbital region.



**Figure 1: (A) and (B) Pedunculated infraorbital swelling.**

Computerized tomography of nose and paranasal sinuses showed right maxillary opacity right concha bullosa with a lobulated fairly well defined heteroattenuating predominantly hypoattenuating lesion in the right preseptal region in the lower eyelid with extension into the right nasolacrimal duct region. Frontal, ethmoidal, sphenoid sinuses were normal. The osteomeatal complexes were normal on both sides. Cribriform plates, pterygoid plates, recti muscles and optic nerves were intact.



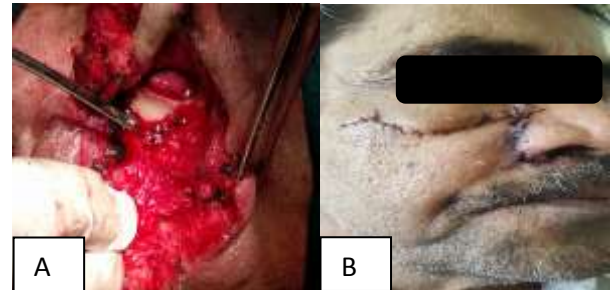
**Figure 2: CT brain and paranasal sinuses—coronal view; arrow heads indicating right maxillary sinus opacity and right infraorbital swellings respectively.**



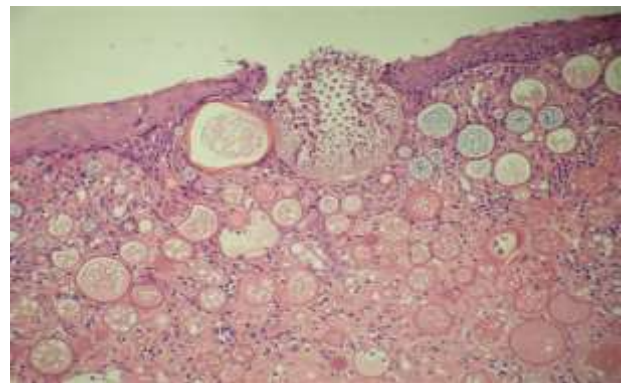
**Figure 3: CT brain-sagittal view; arrow head indicating infraorbital swelling.**

A right lateral rhinotomy and inferior meatal antrostomy followed by right maxillary polypectomy was performed using Weber Ferguson incision. Right lower eye lid swelling was explored and non-capsulated areas of granulation tissue were noted. On extension of dissection medially there was evidence of whitish spores with

mulberry appearance over the lacrimal sac. All the abnormal tissue in the lower eyelid with spores was removed along with excision of the lacrimal gland and a part of nasolacrimal duct. The specimen was sent for histopathology which revealed rhinosporidiosis with inflammatory maxillary sinus polyp with skin involvement.



**Figure 4: (A) Intraoperative image; (B) postoperative image.**



**Figure 5: Histopathological examination with H and E stain reveals sporangium with sporangiospores.**

Patient was started on oral dapsone 100 mg twice daily and has been asked to follow up after 1 month

## DISCUSSION

Rhinosporidiosis, a chronic granulomatous disease caused by *Rhinosporidium seeberi* was first identified in 1892 by Malbran of Buenos Aires.<sup>1</sup> The first detailed account of rhinosporidiosis was given by Seeber in 1900 followed by description of the life cycle of the fungus, *Rhinosporidium seeberi* in 1923 by Ashworth.<sup>1</sup>

The infection is acquired by bathing in ponds contaminated by animal feces.<sup>2</sup> Although the disease is endemic in India and Sri Lanka, it has also been identified in more than 70 countries including South America, South Africa, United States, England and Egypt.<sup>1,2</sup>

The disease is characterized histologically by granulomatous reaction, pseudocystic abscesses, and fibrosis around the causative organism.<sup>3,4</sup> The peak

incidence is seen in second and third decade with a male preponderance.<sup>5</sup>

The mucous membranes of the nose and nasopharynx is commonly affected; however any mucus membrane can be affected.<sup>1</sup> Anteriorly, the disease commonly involves the anterior nares, the nasal cavity, the inferior turbinates, septum and floor. Posteriorly, it can present as polyps in the nasopharynx, larynx, and soft palate.<sup>2</sup>

Ocular manifestations are seen in 15% of cases of rhinosporidiosis, mainly involving the bulbar and palpebral conjunctiva.<sup>2</sup> However, lacrimal sac involvement is a rare presentation of the rhinosporidiosis. In India, ocular rhinosporidiosis involves conjunctiva (69%), lacrimal sac (24%), canaliculi (4%), and lids and sclera (4%).<sup>6</sup> Lacrimal sac rhinosporidiosis although rare, can present as swelling over the sac area or lower eyelid, epiphora, epistaxis, or widened nasal bridge.<sup>7</sup>

Our patient, presented with a painless infraorbital swelling with minimal nasal manifestations.

The definitive diagnosis is by histopathological examination where in the organism is always found in the lesion.<sup>8</sup> Histopathologically, the lesion reveals polypoid fibroconnective stroma containing globular cysts which represent thick-walled sporangia containing numerous “daughter spores” in different stages of development. The stroma reveals inflammatory infiltrate. Periodic acid-schiff (PAS) stain, Gomori's methenamine silver stain, and mucicarmine stain are used to demonstrate *Rhinosporidium seeberi*.<sup>9</sup>

The surgical excision of the polyp, preferably by electrocautery, is the treatment of choice. The radical removal of pedunculated polyps is easier than excision of sessile polyps which require removal of the adjacent mucosa as there may be spillage of endospores.<sup>2</sup>

Dapsone (4,4-diaminodiphenyl sulphone) is the only drug proven to be beneficial in rhinosporidiosis.<sup>10,11</sup>

## CONCLUSION

We present a rare case of rhinosporidiosis of the lacrimal sac presenting as infra-orbital swelling which can pose a diagnostic dilemma. A high index of suspicion is required for detecting this rare manifestation of the rare disease.

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