

Radiological Study of Two Extensive Cases of Rhinosporidiosis

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Case Report

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Article History

Received: 23.07.2018

Accepted: 05.08.2018

Published: 30.08.2018

DOI:

10.36347/sjams.2018.v06i08.012



Abstract: To illustrate the significance of imaging studies in case of extensive rhinosporidiosis. Two cases presented with swelling over the left side of face and bilateral/unilateral nasal mass. Case 1: Patient underwent contrast-enhanced CT scan of nose and paranasal sinus which revealed bilateral nasal mass, mass in the lacrimal sac, left nasolacrimal duct dilatation, maxillary sinus involvement in the left side & nasopharynx. Case 2: Patient underwent contrast – enhanced CT scan of head and neck which revealed the involvement of left nasal cavity, left lacrimal sac, left nasolacrimal duct, left maxillary sinus, nasopharynx, oropharynx and laryngopharynx upto the thyroid cartilage. Both patients underwent excision of the mass, dacryocystectomy, endoscopic excision of nasal mass, endoscopic left maxillary clearance and endoscopic excision of the mass from oropharynx and laryngopharynx in case of the 2nd patient. Histology of nasal & facial masses was suggestive of rhinosporidiosis. There was no recurrence of disease in 6 months follow up. Rhinosporidiosis can be extensive and can present as huge facial swelling as evident in these cases. CT scan is mandatory to delineate the extent of the disease and surrounding structure involvement.

Keywords: Nasolacrimal rhinosporidiosis, maxillary rhinosporidiosis, CT scan, endoscopic excision, dacryocystectomy.

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous inflammatory disease of the mucous membrane caused by *Rhinosporidium seeberi*. It most commonly involves the nasal cavity and the nasopharynx.

Other less common sites of involvement are the conjunctiva, lips, nasolacrimal system, oropharynx, larynx, epiglottis, trachea, bronchus etc. Satyanarayna in his series of 255 cases of rhinosporidiosis found only 2 case of maxillary sinus involvement [1]. In our previous series of 98 cases of rhinosporidiosis only one documented case of maxillary swelling observed [2].

CASE REPORT

Case 1

A 22 yrs male patient presented to the outpatient department with a large swelling arising from the area beneath the inner canthus of the left eye (fig 1(a)). He also complained of nasal obstruction with occasional epistaxis and watering from the left eye. The patient underwent two intranasal operations for similar complaints in the last two years. On anterior rhinoscopy a pink fleshy mass was seen occupying both the nasal cavities and scar marks was seen on the dependent part

of the swelling indicating previous surgery (fig 1(b)). CT scan of paranasal sinuses demonstrated an opacity occupying the left nasal cavity with extension to left maxillary sinus along with dilatation of the left nasolacrimal duct with opacity in the right nasal cavity; the bone of the maxillary antrum was thickened and sclerosed. (fig 2). It was clinically diagnosed as a case of recurrent Rhinosporidiosis occupying both nasal cavities with involvement of left maxillary sinus and left nasolacrimal system resulting in a protruding mass from the skin medial to the inner canthus of the left eye.

The patient was treated by dacryocystectomy along with endoscopic excision of the intranasal mass. Maxillary sinus mass was also removed endoscopically. Postoperatively patient was put on dapsone 100mg. Once daily for 3 month and on 6th month follow up there was no recurrence.



Fig-1(a): Large swelling from area beneath the inner canthus of left eye. **1(b)** Mass in both the nasal cavities and scar over the swelling

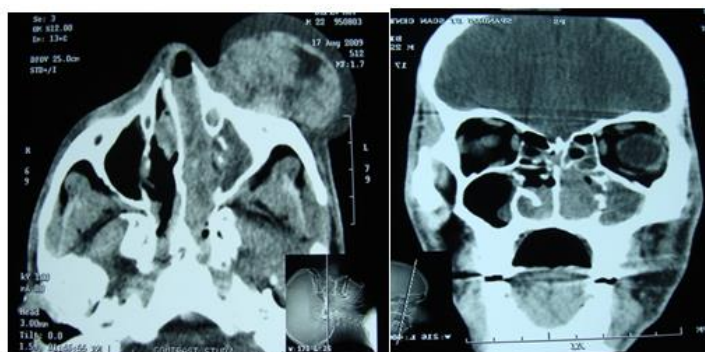


Fig-2: CT scan of PNS (axial & coronal sections) showing heterogeneous opacity in the both nasal cavities and left maxillary sinus along with dilatation of the left nasolacrimal duct

Case 2

A 30 yrs male patient presented in the emergency department with respiratory distress for few days and a tracheostomy was performed on arrival. The patient had a large swelling arising from the conjunctiva of the left upper and lower eyelid which mimicked plexiform neurofibromatosis, a smaller swelling from the left lacrimal sac (fig,3(a)). On anterior rhinoscopy a mass was seen occupying the left nasal cavity. The mass was seen obstructing the oral cavity and involving the hard palate as well (fig 3(b)). CT scan demonstrated an opacity occupying the left nasal cavity with involvement of the osteomeatal complex and extension to the left maxillary sinus, left lacrimal sac, left nasolacrimal duct (fig,4(a), (b)). The coronal(fig 5(a)) and sagittal(fig 5(b)) section showed the extension of the heterogeneous enhancing mass from the base of the skull, involving the naso-oro-laryngopharynx upto the tracheostomy tube at the level below the thyroid

cartilage. It was clinically diagnosed as a case of rhinosporidiosis occupying the left nasal cavity with involvement of left maxillary sinus, left nasolacrimal system, left upper and lower eyelid, resulting in a protruding mass from the skin medial to the inner canthus of the left eye and a large disfiguring mass from the left lower eyelid hanging over the left cheek.

The patient was planned for staged treatment. Initially dacrycystectomy done along with drilling of nasolacrimal duct. Then lower lid blepharoplasty was performed. Excision of the conjunctival mass along with mucosal grafting was attempted with the involvement of ophthalmologist. The pharyngo-laryngeal mass was removed by coblation. Similar to the first case, postoperatively patient was put on dapsone 100mg. Once daily for 3 month and on 6th month follow up there was no recurrence observed.



Fig-3(a): Swelling from the left lacrimal sac and mass arising from the left upper and lower eyelid. **3(b)** mass in the left nasal and oral cavity

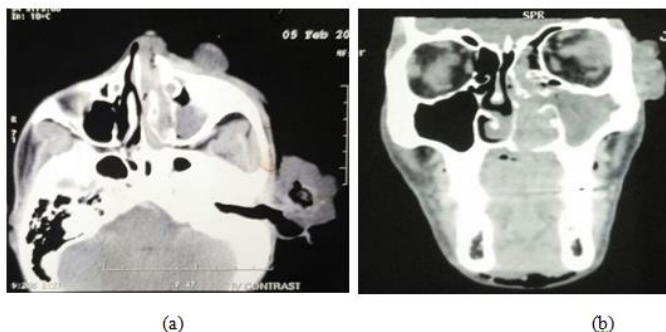


Fig-4: axial (a) & coronal (b) sections show mass occupying the left nasal cavity involving the osteomeatal complex, left maxillary sinus and left nasolacrimal duct

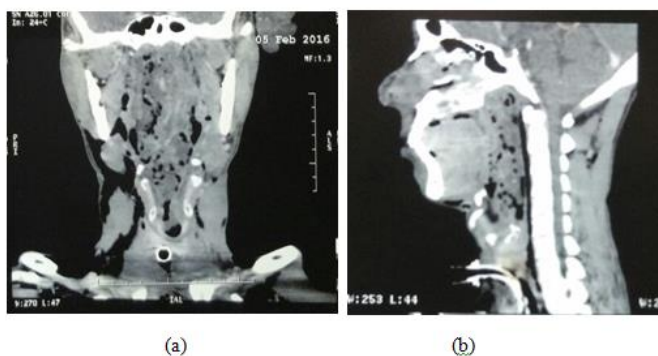


Fig-5: coronal (a) & sagittal (b) sections shows the mass extending from the skull base upto the tracheostomy tube at the level below the thyroid cartilage involving the naso-oro-laryngopharynx

DISCUSSION

Rhinosporidiosis is a chronic granulomatous disorder caused by *Rhinosporidium seeberi*, but its taxonomy is still debated as it shows morphological features resembling those of fungi and protozoa. Earlier it was classified as a fungus as fungal stains such as methanamine silver and PAS could stain the wall of the organism, but culture of *Rhinosporidium seeberi* had been unsuccessful in all artificial media. Molecular biological techniques have more recently demonstrated that this organism is an aquatic protistan parasite. Saha

et al. demonstrated by multiplex PCR that it belongs to the DRIPs clade of fish parasite [3].

Rhinosporidiosis is endemic in India and Srilanka but has also been reported from the United States, South America and Iran. It is more commonly seen in adult males and is possibly transmitted to humans by direct contact with spores through dust, infected clothing or fingers, and through swimming in stagnant waters. Endospores penetrate the nasal mucosa, mature into sporangium within the submucosal compartment, and after maturation burst with release of

sporangia into surrounding tissue. Clinically, the lesion presents as a soft, polypoid mass sometimes pedunculated in the nose, eye and its adnexa, or conjunctiva. Histologically, the infected tissue reveals granulomatous reaction, pseudocystic abscesses and fibrosis around the causative organism.

Lacrimal sac extension in rhinosporidiasis is not unusual one and nearly in all cases infection ascends from the nose [4]. Nasolacrimal duct dilation in the present case proves this theory. Bone erosion in rhinosporidiosis is rarely reported. Madhavan[5] reported a case of rhinosporidial infection of forehead with underline bone erosion.. Adiga *et al.* [6] reported a case of tibial rhinosporidiosis with erosions of the cortical bone. Prabhu *et al.* [7] in their series of rhinosporidiosis found bone involvement in the form of rarefaction, partial or complete erosion of inferior turbinate, thinning of medial maxillary wall and septal erosion. Keskin *et al.* [8] presented a case of rhinosporidiosis with involvement and erosion of the anterior wall of the maxillary sinus. A further unusual case has been described by Suryawanshi *et al.* [9] with involvement of the lateral end of the right clavicle mimicking a bone tumour. In the present case bone erosion was observed in the medial wall of left maxilla, left nasolacrimal duct system and a protruding mass arising from the skin of the inner canthus of left eye.

Prabhu *et al.* [7] studied imaging features of rhinosporidiasis in. In their study most common imaging features are well-defined lobulated, irregular moderately enhancing soft tissue masses with HU ranging from 70 to 160, primarily centred in the inferior nasal cavity. They found NLD involvement in four cases and considered it to be the route for lacrimal sac involvement. However, they didn't find a single case of maxillary sinus involvement [7]. MRI study of rhinosporidiasis was done in a single case by Rath *et al.* [10] and they observed intensely enhancing lesion in the affected area which may be due to rich vascularity of rhinosporidiasis.

CONCLUSION

Imaging study is important in recurrent rhinosporidiasis. It shows the extent of disease, its attachment, any bony erosion and extent into surrounding structure.

REFERENCES

1. Satyanarayana C. Rhinosporidiasis: With a record of 255 cases, Acta otolaryngological. 1960;3-4:348-366
2. Saha SN, Mondal AR, Bera SP, Das S, Banerjee AR. Rhinosporidial infection in West Bengal- a Calcutta based hospital study. Indian Journal of Otolaryngol Head Neck Surg. 2001;53(2):100-104
3. Saha S, Mondal D, Khetawat D, Roy A, Chakrabarti S, Bhattacharya B. A molecular approach (multiplex polymerase chain reaction) for

diagnosis of rhinosporidiosis. Indian Journal of otolaryngology and head and neck surgery. 2002 Oct 1;54(4):264.

4. Kameswaran S, Lakshmanan: Rhinosporidiasis. ENT disorder in atropical enviornment, 2nd Ed, MERF publication. 1999;19-34
5. Madhavan M, Ratnakar C, Mehdiratta K S. Rhinosporidial infection of the forehead –(report of a case).J Postgrad Med. 1978;24:235-6
6. Adiga BK, Singh N, Arora VK, Bhatia A, Jain AK, Acta Cytologica. 1997;41:889-891.
7. Prabhu SM, Irodi A, Khiangte HL, Rupa V, Naina P. Imaging feature of Rhinosporidiasis on contrast CT. Indian Journal of Radiol. Imaging 2013,23:212-8
8. Keskin G, Ustündağ E, Kaçarözkar S, Keskin S. An erosive mass involving the nasal cavity and maxillary sinus: rhinosporidiosis. Kulak burun bogaz ihtisas dergisi: KBB= Journal of ear, nose, and throat. 2004;13(5-6):143-7.
9. Suryawanshi PV, Rekhi B, Desai S, Desai SM, Juvekar SL, Jambhekar NA. Rhinosporidiosis isolated to the distal clavicle: A rare presentation clinicoradiologically mimicking a bone tumor. Skeletal radiology. 2011 Feb 1;40(2):225-8.
10. Rath R, Baig SA, Debata T. Rhinosporidiosis presenting as an oropharyngeal mass: A clinical predicament?. Journal of Natural Science, Biology, and Medicine. 2015 Jan;6(1):241.