Rhinosporidiosis: Its varied presentations

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Abstract

Rhinosporidiosis is the fungal infection generally affecting the nasal mucosa. In this article we are presenting two unusual cases of rhinosporidiosis. First case presented with nasopharyngeal mass, without any past history of bathing in stagnant water or epistaxis. Mass was excised using nasal endoscope. Second case presented with recurrence of rhinosporidiosis even after nine times surgical intervention with systemic Amphotericin-B. In this patient, surgical excision of mass was assisted with Amphotericin-B nasal douching and oral Itraconazole. Both patients are doing well with no recurrence till date.

Keywords Rhinosporidiosis varied presentation endoscope Treatment

Introduction

Rhinosporidiosis is a chronic granulomatous lesion caused by *Rhinosporidium Seeberi*, a fungus from the class of phycomycetes of family *coccidioidaceae*^{1,3}. It generally affects the mucous membrane of nose and nasopharynx but occasionally involves conjunctiva, lacrimal sac, lips, palate, uvula, maxillary antrum, epiglottis, larynx, trachea, bronchus, ear, scalp, skin, penis, vulva and vagina^{1,2}. Osteolytic lesions in the bones of hands and feet are also reported. It is usually

limited to surface epithelium but may present as widespread visceral involvement^{1,2}.

It presents clinically as reddish polypoidal mass with friable irregular surface and history of recurrent epistaxis. Viscid nasal discharge is also reported in early stages^[3,6,7]. Diagnosis is usually made following histological examination of biopsy specimens from polypoid lesions. Giemsa, Gridley and Toluidine blue are used as permanent stains for detecting the fungus.

The most successful treatment reported is wide excision with a cutting diathermy and cauterization of the base^{2,3,5}. Medical treatment in form of Amphotericin B and Dapsone may be given in addition^{2,3,8}.

Here we are presenting two cases of rhinosporidiosis with unusual presentation and use of endoscopes in surgical management.

Case Report

Case 1: A 48-year-old male, resident of UttarPradesh, presented to outpatient department with complaints of nasal obstruction for six months, mass in oral cavity since two months and difficulty in swallowing since one month. He gave no history of anosmia, bathing in ponds or epistaxis.

Fig. 1 Mass hanging in oropharynx in case 1.

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Fig. 2 specimen of the excised mass in case 1.

Fig. 3 histopathological picture of excised specimen in case 1 showing the sporangia with spores.

Fig. 4 photograph of case 2 showing stenosed right anterior nares and mass in left side

Fig. 5 cytology of nasal smear of case 2 showing the sporangia with spores.

On examination the nasal cavities appeared normal but patency was found to be decreased both sides. A reddish polypoidal mass of about 5x3cm was seen hanging in oropharynx, pushing soft palate anteriorly (Fig 1). Diagnostic endoscopy revealed a mass in nasopharynx arising from the lateral wall of nasopharynx. Noncontrast Computed Tomography (NCCT) revealed enhancing soft tissue mass in nasopharynx and oropharynx with patent nasal cavities. Patient underwent routine haematological and urine examination. Blood sugar, liver function and kidney function tests were within normal range. Chest x-ray was normal. There was no evidence of any systemic diseases. Patient was taken up for endoscopic excision of mass under general anaesthesia. Specimen was sent for histopathological examination (Fig. 2), which confirmed the diagnosis of rhinosporidiosis (Fig. 3). Postoperatively patient was started on alkaline nasal douching and oral Itraconazole (100mg twice daily). Patient is under follow up by regular diagnostic nasal endoscopic examination. Three months postoperatively, the patient is doing fine with no recurrence.

Case 2: A 25-year-old patient, resident of Nepal, presented with complaints of recurrent nasal mass with epistaxis. He gave past history of being operated nine times for similar complaints. He also gave history of receiving a systemic course of Amphotericin B. Previous biopsy reports of excised mass gave diagnosis of rhinosporidiosis. In addition to this he complained of difficulty in swallowing and occasional oral bleeding. On examination there was a friable mass protruding from left nostril and vestibular stenosis of right nostril. Oral examination showed similar mass with multiple attachments to soft palate, posterior pharyngeal wall, anterior and posterior tonsillar pillars (Fig 4). Smear of nasal discharge itself showed the sporangia confirming the diagnosis of recurrence of rhinosporidiosis (Fig 5). Patient was planned for revision surgery after a repeat CT scan of nose and paranasal sinuses. NCCT showed soft tissue enhancing mass

in nasal cavities, nasopharynx and oropharynx. Under general anaesthesia mass was excised from left nasal cavity using nasal endoscope. Moure incision was given on right side to gain access to nasal cavity. Disease was removed and nasal vestibular stenosis was corrected. Oropharyngeal mass was also excised and its multiple attachments were cauterized using electro-cautery. Histopathology of the excised mass also confirmed our diagnosis of rhinosporidiosis. Postoperatively, patient was started on Amphotericin-B nasal douching and oral Itraconazole. Patient is followed up by regular endoscopic examination and repeated kidney and liver function tests.

Discussion

Rhinosporidiosis is a chronic granulomatous disease caused by a fungus *Rhinosporidium seeberi* from the class of phycomycetes and family coccidiaceae. The disease was first reported in 1900 by Seeberi³. In the early phase of infection, the organism appears round or oval, and is 2 to 10u in diameter with repeated nuclear divisions. Large sporangia 300 to 350u in diameter are found containing nearly 16,000 spores. The organism is dimorphic consisting of two phases, the saprophytic in soil and the parasitic in the tissue of animals such as cattle, horses, mules, dogs, flocks and humans^{3,6}. The infective stage of the fungus is the functional spore³. The spores are 10 to 12u in diameter and contain storage materials arranged as spherules 1-2u in size¹.

Trauma to nasal mucous membrane, diving and swimming in stagnant water or ponds are proposed as the essential predisposing factors^{3,6}. Impairment of ciliary action, irritation and stagnation of secretion from anatomic obstruction to drainage may also play a role^{3,6}. A definite course of transmission by water is yet to be demonstrated^{7,8}. Ocular infection is generally airborne⁹. In our first case of rhinosporidiosis, there was no history of bathing in stagnant water or ponds.

Nasal mass, nasal obstruction and epistaxis have been reported as the most common presenting complaints^{3,7,9}. Other complaints may vary according to the site of the infection. Oral mass, haemoptysis, ocular mass and bony swellings are some of the other presentations. Rhinosporidiosis usually affects the nasal mucous membrane but cases have been reported of isolated nasopharyngeal involvement¹⁵. It appears as pink or red papillomatous mass, which is soft, friable and generally pedunculated. Being a very vascular swelling, it bleeds on touch or manipulation.

It may resemble a strawberry or a ripe raspberry, as the surface is studded with white flecks, which are the mature sporangia^{3,6,7}. In our first case, inspite of a mass of size 5 x 3cm, there was no history of epistaxis or oral bleed. Differential diagnosis of antrochoanal polyp with squamous metaplasia and inverted papilloma was taken into consideration in this case. The C.T. scan picture and histopathology of specimen ruled out both conditions and confirmed our diagnosis of rhinosporidiosis. Diagnosis depends largely on the histopathology of tissue⁴. Histopathology shows the sporangia with the stroma infiltrated with lymphocytes, plasma cells, polymorphs and eosinophils. Giant cells of foreign body type can also be seen around the sporangia. The degenerated sporangia with collapsed membranes and liberated spores may be seen^{1,3}. Recently nasal smear has been recommended as a routine preliminary screening technique in diagnosis^{3,10}. By direct examination of smear using Gridley stain, the chitinous wall appears bright purple and endospores as brown. In our second case we were able to demonstrate the sporangia in the nasal smear cytology.

Surgical excision of the mass forms the mainstay of the treatment^{2,3,5}. Surgical excision should be accompanied with the cauterization of the base to ensure postoperative haemostasis and complete removal of disease. Use of nasal endoscopes is of great help in surgical treatment of these patients. It helps in complete removal of disease under vision. It is also helpful in postoperative follow up of patients. It helps in monitoring the response of treatment and early detection of any recurrences. In our cases with nasopharyngeal attachments of mass, use of nasal endoscope was of great help in surgical excision.

Rhinosporidiosis is generally considered to be insensitive to the antimycotic or antibiotic treatment^{3,5}. Medical management in the form of local injection of depot corticosteroids into the polypoidal masses², systemic course of Amphotericin-B and dapsone may be given^{2,3}. The use of Amphotericin is usually recommended when either surgical excision is incomplete or there is recurrence¹². Nair published a study in which he gave Dapsone to 32 patients of rhinosporidiosis and reported no recurrence in 71.4% of patients in three years follow up¹¹. In our second case, inspite of nine surgical interventions and systemic course of Amphotericin-B, patient had recurrence. It has been postulated that locally applied Amphotericin may be of help in these patients by acting locally at the site of infection. The

systemic complications are also less in this mode of treatment. We have seen that this treatment given in addition to the oral Itraconazole to our second patient has controlled the disease process till date. There has been no recurrence at 3 months postoperative interval. With our experience of these two patients, we infer that rhinosporidiosis can present at unusual site without any history of epistaxis or bathing in ponds. As seen in second case, sometimes it may take such a aggressive course that no surgical or medical intervention help the patient.

Summary

Rhinosporidiosis is a chronic granulomatous fungal infection caused by *Rhinosporidium seeberi*. It is generally seen in those taking bath in stagnant waters. It is known to infect the nasal mucosa primarily but other areas like eyes, bones, oral cavity, nasopharynx, skin, trachea, bronchi etc are also reported to be affected. Diagnosis depends on the histological picture of the mass but the sporangia may be demonstrated in the nasal smear also. Treatment consists of surgical excision of the mass. Use of endoscopes has helped in ensuring complete excision of the mass under vision. Though the recurrence is rare but in cases of incomplete excision or recurrence, medical treatment in form of Amphotericin-B is given. Dapsone or intralesional steroids form the alternative modes of treatment.

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