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Case Report

Notorious Case of Rhinosporidiosis Presenting as Sinonasal Polyposis

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Abstract

Rhinosporidiosis is a rare chronic granulomatous disease, caused by acquatic protozoan *Rhinosporidium seeberi*. Characterised by polypous lesion of mucosa of nose, conjunctiva and urethra. Rhinosporidiosis is endemic in Chhatisgarh South India, Srilanka, South America and Africa. Male preponderance seen. more common in age group 15-40. We hereby present a case description of 46 yr old male native of Rajasthan with polypoidal mass rhinosporidiosis. Patient presented with long standing history of nasal obstruction and intermittent epistaxis for 2-3 yrs. Diagnosis was confirmed by HPE examination and he was successfully treated by complete surgical excision.

Keywords: Rhinosporidiosis; Granulomatous Diseases; Epistaxis; Nasal Obstruction

Introduction

Rhinosporodiosis is a rare chronic granulomatous disease caused by *Rhinosporodium seeberi* [2]. It is prevalent in south Asian countries affecting mainly southern parts of India and Srilanka [1,6]. Disease exhibits no racial predilection although its pattern displays male gender preponderance. Besides humans the disease has been reported in other animals such as horses and bovines [3]. Most common source of acquiring the disease is frequently bathing in stagnant water ponds [6]. Originally *Rhinosporodium seeberi* was considered to be an a protozoan and subsequently a fungus, but currently it is classified as an aquatic mesomycetazoan (In between fungi and animals) on the basis of phylogenetic analysis of 18S rDNA [5].

Rhinosporodiosis is a chronically localised infection of mucous membrane mainly limited to surface epithelium but sometimes present also as widespread visceral involvement. Clinically lesion presents as a reddish polypoidal friable mass mainly affecting the nose, throat, conjuctiva, ear, genitalia and even osteolytic bony lesions of hands and feet has been reported [1,4]. Clinical symptomatology include nasal obstruction and epistaxis, viscid nasal discharge is seen in early stages of the disease [2,4]. Diagnosis is usually confirmed following histological examination of biopsy specimens from polypoid lesions which shows round and thick walled sporangia in the submucosa of the affected site varying from 10-200 mm in size which are visible as white dots in the mucosa containing smaller daughter cells (called sporangiospores). It can be visualized with fungal stains such as Gomori methenamine silver (GMS) and periodic acid-Schiff (PAS), as well as with standard haematoxylin and eosin (H&E) staining [1,4]. Extension of disease is evaluated through NCCT of nose and PNS.

The only curative approach is the surgical excision combined with electrocoagulation of the base followed by medical treatment of Dapsone or Amphotericin B [1,4]. Often recurrence and operated site bacterial infection are most common complication [1]. This report therefore is aimed at documenting a case of 46 year old native male with a polypoid sino nasal rhinosporidiosis.

Case Report

A 46 year old male, resident of Bharatpur, India presented to outpatient department of Otolaryngology at a tertiary care centre with complaints of long standing bilateral nasal obstruction, occasional nasal discharge, mass in oral cavity and intermittent epistaxis, symptoms gradually progressed in last 2-3 years. Patient had history of nasal trauma because of RTA approx. 8 years back. Patient had history of one prior surgery in private hospital 4 years back for similar complaints. There were no history of any constitutional symptoms, anosmia, bathing in ponds. On anterior rhinoscopic examination there was an erythematous papillomatous mass present in bilateral nose completely obliterating the nasal cavity. The mass was friable and had a few whitish dots over it. On oral cavity examination reddish polypoidal mass 2x3 cm was seen hanging in oropharynx pushing the uvula anteriorly.

Preoperative examination comprise of biochemical and hematological profile as well as punch biopsy from the mass. Biochemical and hematological indices were with in normal limit. However the histopathological examination of tissue reveals soft tissue covered with respiratory epithelium and showing non-specific inflammation. Large number of sporangia of rhinosporidiosis are present. Non contrast CT of paranasal sinus with gem stone spectral imaging shows ill defined soft tissue density lesion with few air foci noted in left nasal cavity in anterior part extending in rt nasal cavity and left maxillary sinus causing irregular destruction of nasal septum in mid part extending along the anterior wall of nasopharynx causing narrowing of nasopharyngeal airway, size of lesion ~ 45 x 28 x 31 mm. These findings were pathognomic of rhinosporidiosis, therefore patient was planned for definitive surgery in which mulberry like mass endoscopically excised from bilateral nasal cavity, left maxillary sinus and oropharynx. mass was also attached to inferior turbinate for which partial inferior turbinate reduction was done. After excising the mass on direct nasal endoscopy a large posterior septal perforation was seen. Gross resected specimen depicted an intact polypoidal mass measuring 4 x 2 cm soft and friable tissue which was sent for histopathological examination. Postoperatively patient was started on alkaline nasal douching and oral dapsone 100 mg per day for 6 months. HPE of the excise specimen showed features consistent with Rhinosporidiosis, associated with chronic inflammatory cell infiltrate and foreign body giant cell reaction. Patient is under surveillance through regular outpatient visits (Figure 1).

Figure 1: A: Large (100-450 microns), thick-walled sporangia with 1000+ endospores each 6-10 microns. B: Surgical specimen showing rhinosporidiosis. C: pre op CT scan showing ill-defined mass in lest nasal cavity, D: post op DNE showing septal perforation.

Discussion

Rhinosporidiosis is a chronic granulomatous disease caused by a fungus *Rhinosporidium seeberi* from the class of phycomycetes and family coccidiomycetes [4]. Though it appears to occur universally, rhinosporidiosis remains largely endemic in Indian subcontinent [3]. The organism appears round or oval in early phase, and is 2 to 10u in diameter with repeated nuclear divisions. Approximately 16,000 spores are present in large sporangia. It is dimorphic fungi having two phases, the saprophytic in soil and the parasitic in tissue of animals such as cattle, horses, mules, dogs, flocks and humans [4]. Rhinosporidiosis is epidemiologically associated with exposure to water, the placement of *R. seeberi* in a clade of aquatic parasites by molecular analysis, and the fact that

watery substances facilitate the release of mature endoconidia from the sporangium suggest that the natural niche of *R. seeberi* is water [5]. It was initially consided by most microbiologist as a fungus just because of its property to be stained by fungal stains such as Gomori methenamine silver (GMS) and periodic acid Schiff (PAS). It is postulated recently by authors that etiological agent of the disease was not a fungus but a prokaryotic cyanobacterium called *Microcystis aeruginosa* [2]. Most commonly it involves nose characterised by single pedunculated polyp or multiple sessile polyps arising from mucosa. Manifestations like multiple cutaneous reddish polyps, bone lesions, and corneal mass are also seen in some cases [6]. In the nasal cavity, inferior meatus is most commonly involved because of its close proximity with the nasolacrimal duct. Lesion begin in nasal cavity and then extended into the orbit via the nasolacrimal duct [9]. Rhinosporidiosis is an infectious disease where the pathogen is always found in tissue of lesion. There is no documented evidence, however, that this disease is contagious. The majority of cases are sporadic. The presumed mode of transmission is from the natural aquatic habitat of *R. seeberi* through traumatized epithelium, most commonly via nasal sites, but also via the external urethral meatus, the conjunctiva or the skin [7]. *Rhinosporidium seeberi* should be distinguished from another microrganism, Coccidioides immitis. The latter one has similar mature stages represented by large, thick-walled, spherical structures containing endospores, but the spherules are smaller (diameter of 20-80 µm versus 50-1000 µm) and contain small endospores (diameter of 2-4 µm). Coccidiodes does not stain with the mucicarmine [8]. The treatment for rhinosporidiosis consists in surgical excision. Meticulous excision is treatment of choice, although rare cases of spontaneous regression have been reported. Main cause of recurrence is due to spillage of spores in adjacent mucosa. Recurrence of the disease after surgery is rare and can be prevented by cauterization of the base of the lesion or alternatively by cryotherapy [7]. Although no adequate medical therapy exists, dapsone that interferes with maturation of spores has been used most commonly to prevent the recurrence of disease [10].

Conclusion

Rhinosporidiosis is less frequently seen in Norther part of India. However, with several sporadic cases reported in our region, follow-up of index cases and epidemiological surveillance to ascertain prevalence and source of infection is necessary. For a surgeon, ophthalmologist and radiologist, this is a necessary differential to be kept in mind for sinonasal masses. Both clinical and radiological aspects are required to reach correct diagnosis. Endoscopic removal of mass with cauterisation of the base remains a gold standard approach for treatment.

Bibliography

1. Shukla Das., *et al*. "Nasal rhinosporidiosis in humans: new interpretations and a review of the literature of this enigmatic disease". *Medical Mycology: Official Publication of the International Society for Human and Animal Mycology* 49.3 (2010): 311-315.

2. Godwins O., *et al*. "Nasal rhinosporidiosis". *Journal of the National Medical Association* 100.6 (2008): 713-715.

3. Uledi Sefu., *et al*. "Human nasal rhinosporidiosis: a case report from Malawi". *The Pan African Medical Journal* 9 (2011): 27.

4. VP Venkatachalam., *et al*. "Rhinosporidiosis: its varied presentations". *Indian Journal of Otolaryngology and Head and Neck Surgery* 59.2 (2007): 142-144.

5. Annie I., *et al*. "Cluster of nasal rhinosporidiosis, eastern province, Rwanda". *Emerging Infectious Diseases* 25.9 (2019): 1727-1729.

6. Amit Kumar Dey., *et al*. "Rhinosporidiosis: a rare cause of proptosis and an imaging dilemma for sinonasal masse". *Case Reports in Otolaryngology* (2016).

7. Kaimbo Wa Kaimbo d., *et al*. "Conjunctival rhinosporidiosis: a case report from a congolese patient". *Bulletin de la Societe Belge d'Ophtalmologie* 309-310 (2008): 19-22.

8. Luca morelli., *et al*. "Human nasal rhinosporidiosis: an italian case report". *Diagnostic Pathology* 1 (2006): 25.

9. Payal Mukherji and Neha Shilpy. "Ocular rhinosporidiosis-a case report". *Indian Journal of Clinical and Experimental Ophthalmology* (2022).

10. Aswin Chandran., *et al*. "Pharyngeal Rhinosporidiosis". *BMJ Case Report* 13.6 (2020): e236404.