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Rhinosporidiosis of Parotid Duct Region: A Rare Case Report

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Abstract

Rhinosporidiosis is an age old disease known to man for over a hundred years. Although it presents itself worldwide, it is indigenous in South Asia with highest occurrence in Southern India and Sri Lanka. Usually it affects the mucocuteneous tissues of the nose and conjuctival mucosa. Extramucosal expression of rhinosporidiosis is relatively rare. Only few cases of parotid rhinosporiodosis has been reported so far in the literature. Therefore, this is possibly the fourth case to be reported.

Keywords: Rhinosporodiosis, Parotid Duct, R. Seeberi

Introduction

After its first description in Argentina, Rhinosporidiosis was reviewed in 1998 and described as a chronic granulomatous disease mostly of fungal origin caused by *Rhinosporidiosis seeberi*.[1] However, Herr et al in 1999, showed that this organism is a "protist" named mesomycetozoa and not a classical "fungus" based on 18SrRNA from infected tissue[2]. Usually affecting the nasal mucosa, extra mucosal manifestations of rhinosporidiosis is relatively uncommon. Although it is endemic in Southern India and Sri Lanka, it has also been reported in America, Europe and Africa.[3] Some non-endemic cases have been reported in areas like in Delhi as

well[4]. Besides affecting the humans, it has also known to affect several species of farm, domestic and wild animals. Men are more affected than women. A predilection is seen for the O type blood group followed by AB blood group.[5] The presumed mode of infection is through traumatized epithelium [5] and known to be transmitted by direct contact with spores of species through dust, infected clothing or fingers and swimming or working in stagnant water.[6] In humans, a single outbreak of ocular and nasal sporidiosis was reported in Serbia because of bathing in same infected lake. CT scanning can be used to assess the site and extent of the lesion[7]. We present here, a case of parotid rhinosporidiosis. Only a few cases have been reported so far. [11,12]

Case Report

A 65 year old male patient presented to our department with a 4 months history of recurrent painless swelling on the right side of the face. Patient was a rice farmer native to Himachal Pradesh. Extra oral examination revealed swelling of 10x6cm in dimensions (Fig.1) which was soft on palpation. Overlying skin was smooth, stretched and fairly glossy in appearance with no local rise in temperature. Intraorally the mucosa appeared to be normal (Fig. 2) and aspiration of the lesion was negative. Routine blood investigations were carried out. Patient's blood Group was found to be of O^{-ve}. Preoperative OPG revealed no significant findings and the patient refused to undergo further radiographical examination due to financial issues (Fig. 3). After thorough clinical and systemic examination (RBS, BT,CT, TLC, DLC, Hb%, RBS, HIV-I &II, HbsAg), the lesion was surgically excised sparing the parotid duct, through an intraoral incision on the buccal mucosa under local anaesthesia; the excised tissue mass was sent for histopathological examination.

Clinical diagnosis was based on histopathological examination which revealed multiple globular cyst of varying sizes containing thick-walled spherical sporangia with daughter cells of 20-80µm.The connective tissue stroma was highly infiltrated with mixed inflammatory infiltrate of lymphocytes, plasma cells, histiocytes and neutrophils. The cystic cavities were lined by parakeratinized epithelium and every cyst represented a thick-walled sporangium containing numerous daughter spores in different stages of development. (Fig.4). The confirmation of Rhinosporidiosis was made based on histopathological findings; therefore, we gave a provisional diagnosis of cysts, sialolithiasis, sialocele. As Dapsone is a known effective therapy for Rhinosporidiosis, the patient was kept on dapsone therapy (100mg daily for 6 months) and no recurrence was reported at 3 months follow up.

Discussion

Since its first report, around 70% of the documented cases were involving the nasal mucosa and 15% involving eyes.[8] The natural habitat of R. Seeberi has not been clearly demonstrated. Kumara Kaluarachchi et al performed an in situ hybridization for the purpose of identifying the microbe with the help of PCR.[9] They were able to make a definitive identification of R. Seeberi spores in the deposition of ground water samples. It is infective as the lesion will always be accompanied with a pathogen but not infectious as no case has ever been documented of cross infection among the same family of humans or animals. The first case of lymphatic spread in disseminated rhinosporidiosis was reported in 2002.[10]

Majority of cases reported occur in upper respiratory tract, mostly nasal mucosa, nasopharynx, larynx and soft palate.Rhinosporidiosis has been associated with people working in marshy lands and stagnant water.

The present patient is a rice farmer in Himachal Pradesh, which is concurrent with the literature.[9] Rhinosporidiosis of parotid region is rare, but has been reported.[10]. Capoor

et al have reported 3 cases, where the patients belonged to non endemic areas like Bihar and Uttar Pradesh respectively[4]. In our patient, the lesion was a subcutaneous one with normal appearing overlying skin and mucous membrane. We suspect the portal of entry was either nasal, oral, or traumatized epithelial route. As we were not sure about the diagnosis, to proceeded conservatively, using an intraoral incision before using a definitive extraoral surgical approach. As no recurrence was noted for a 3 month follow up; no further surgical intervention was done and Dapsone therapy was continued. Diagnosis is usually made with histopathology and by cytodiagnosis. In literature, rhinosporidial histopathology was described by Karunaratne[12] as polypoidal, granular, grape like sessile or pedunculated mass on the mucosal surface. Spontaneous regression have been reported in few cases but the ultimate treatment remains surgical.

Conclusion

In conclusion, only a few cases of rhinosporidiosis have been reported in India. This case was interesting to us because of the fact that the patient belonged to a non-endemic region. Unfortunately, due to socioeconomic circumstances, we were unable to perform the standard diagnostic techniques, and a long term follow up was not possible as the patient did not report for the same.

Fig. 1: Frontal view of patient

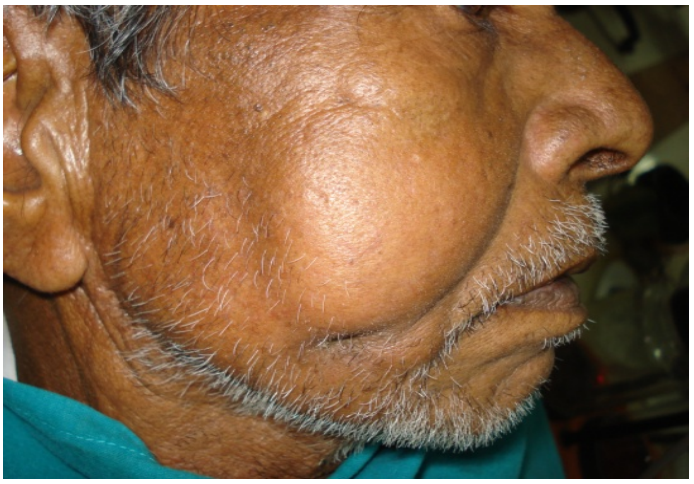

Fig. 2: Profile view of patient

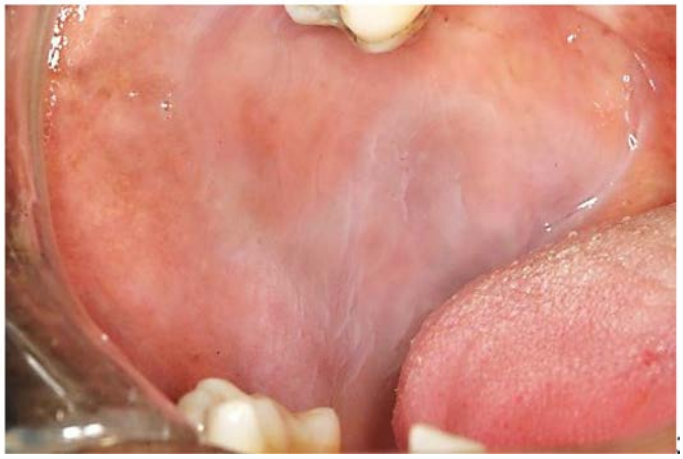

Fig. 3: Intraoral pre-operative photograph

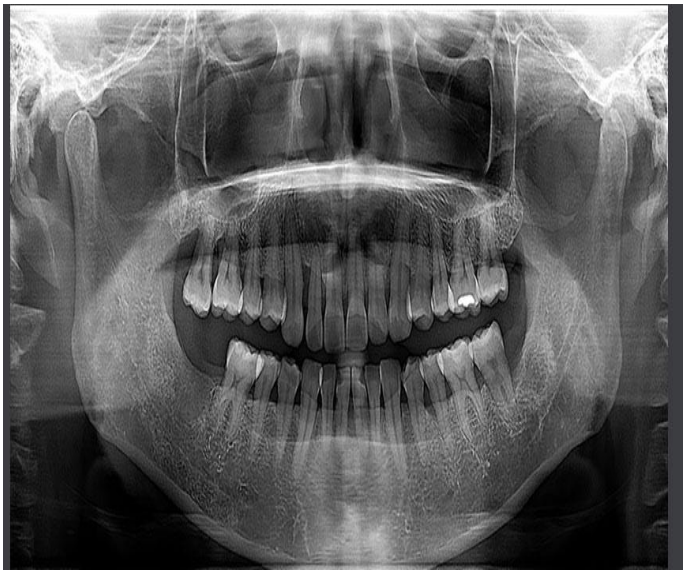

Fig. 4: Pre-operative radiograph

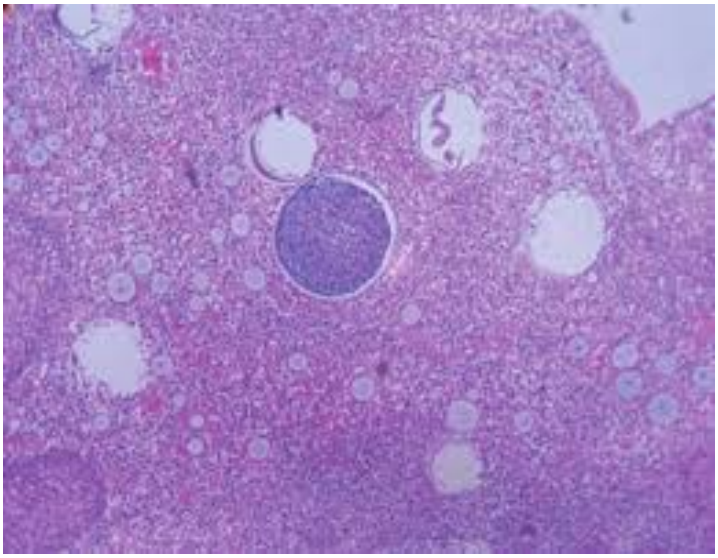

Fig. 5: Histopathological picture

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