Case Report

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Rhinosporidiosis of Parotid Duct: A Rare Case Report

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ABSTRACT

Background: Rhinosporidiosis is a non-contagious, sporadic, chronic granulomatous disease of man and large domestic animals caused by a fungus called Rhinosporidium seeberi. Rhinosporidiosis has been reported from many countries but is endemic in certain parts of India, Sri Lanka and South Asia. It affects chiefly nose and nasopharynx followed by ocular tissue. It commonly presents as a vascular polypoid mass of nasal mucosa. The lesions appear pink with minute white dots, which give it a strawberry-like appearance. Rhinosporidiosis is also known to involve many rare sites and may become disseminated to occur in a generalized form. Rhinosporidiosis of the parotid duct is rare and only five reported cases could be found in the literature. Here, we report a case of Rhinosporidiosis, which clinically presented as a facial swelling indicating that fungal infections should also be considered as one of the differential diagnoses whenever facial swellings are encountered. This patient presented no signs and symptoms of Rhinosporidiosis at any other site.

Keywords: Rhinosporidiosis, Periodic acid–Schiff stain, Endospores, sporangium .

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous fungal infection caused by R. Seeberi; which affects humans, horses, dogs, and to a lesser extent cattle, cats, foxes and birds. It has been reported from many countries especially tropical areas such as parts of India and Sri Lanka. In India most commonly affected areas are Chhattisgarh, Kerala, Tamilnadu & Orissa. Most common sites of involvement are nose and nasopharynx (70%) followed by ocular involvement; conjunctiva and lacrimal apparatus (15%) and rarely Mouth, upper airway, skin, genitals, rectum (15%).[1, 2]

The etiologic agent of Rhinosporiosis, R. Seeberi, has traditionally been considered a fungus. Recent 18S rRNA gene analysis has placed R. seeberi into a novel group of aquatic parasites of the class Mesomycetozoea, some of which cause similar diseases in amphibians and fish. The first case from India was reported by O'Kineay in 1903. [3, 4] This disease most commonly occurs in children and young adults aged 15-40 years with a definite male predilection. It is possibly transmitted to humans by direct contact with spores through dust, through infected clothing or fingers, and through swimming in stagnant waters.[4] We present a case of Rhinosporidiosis of parotid duct who presented with cystic swelling in parotid duct.

CASE REPORT

22 year old male patient visited to Outpatient Department of Chhattisgarh Dental

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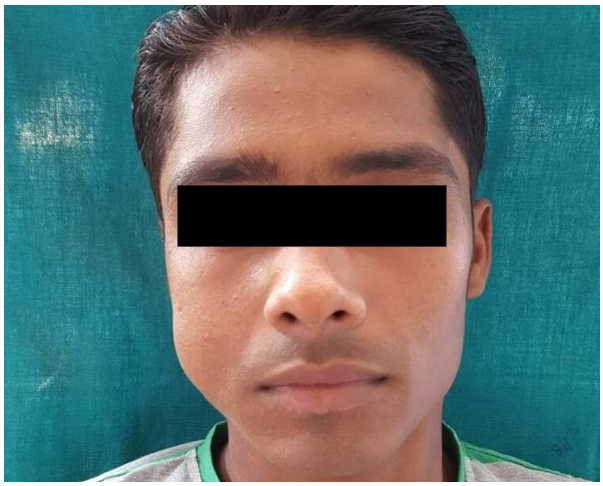

Fig 1: Extraoral clinical photograph showing diffuse swelling on right side of face.

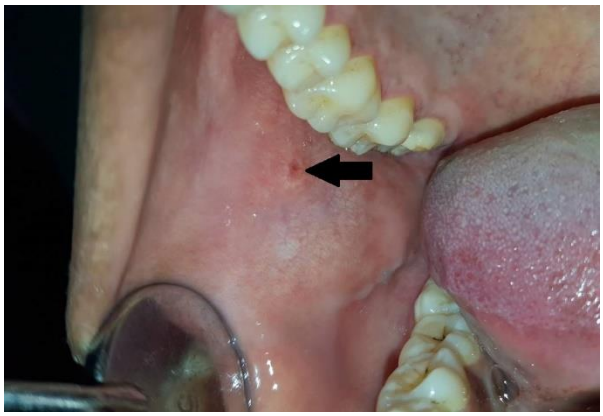

Fig 2: Intraoral clinical photograph showing presence of inflamed parotid duct orifice (arrow).

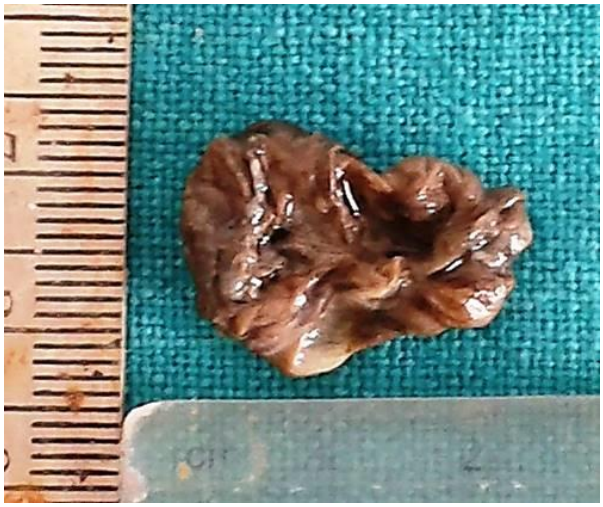

Fig 3: Gross specimen.

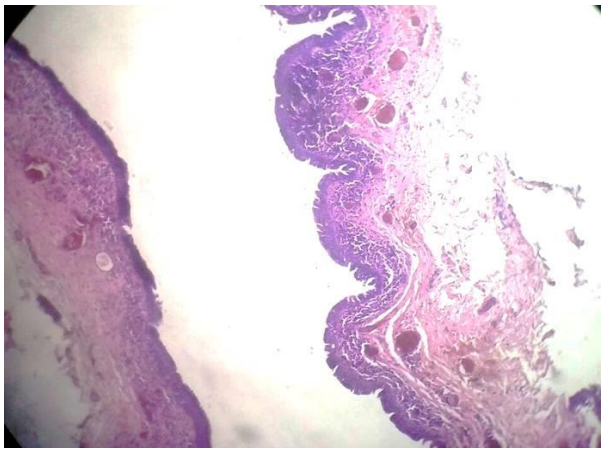

Fig 4: Low power photomicrograph showing numerous papillary projections in the cystic space (H & E Stain, 40X magnification).

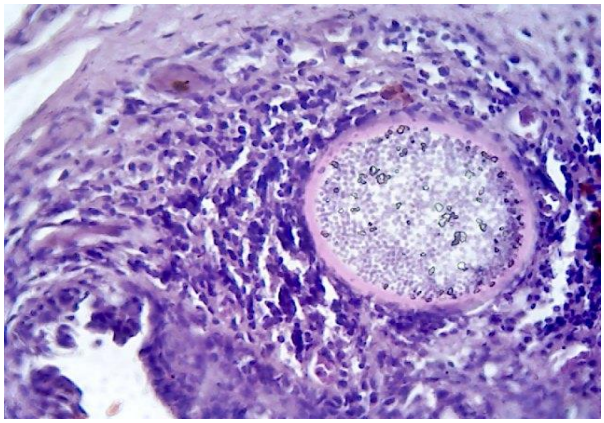

Fig 5: High power photomicrograph showing Sporangium and sporangiospores with numerous chronic inflammatory cells (H & E Stain, 400X magnification).

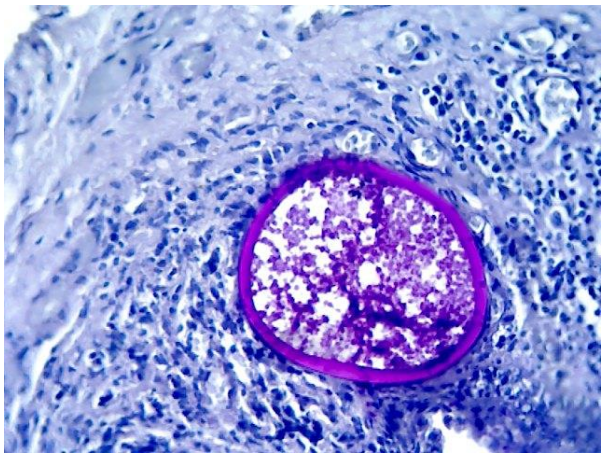

Fig 6: High power Photomicrograph showing Sporangium and sporangiospores showing PAS positivity (PAS Stain, 400X magnification).

College & Research Institute, Sundra, Rajnandgaon; with a chief complaint of swelling on the right side of face since 1 month. Patient noticed swelling on the right side of cheek, which increases while eating and talking. Patient used to drool out saliva by himself after which swelling regresses. Patient experienced dull and intermittent pain while eating and relieved on drooling of saliva. The patient's physical status was fair and both his medical history and habit history were non-contributory. There was no history of pus discharge and bleeding from swelling.

Extra-oral examination revealed presence of diffuse swelling on the right side of face measuring approx. 3×3 cm. extending anterio-posteriorly, 1.5 cm from nasolabial fold to 2.5cm in front of tragus; Superoinferiorly 1.0cm from infra-orbital ridge to 1.5cm above the lower border of mandible .Swelling regresses after drooling of saliva. After giving lime juice, swelling increased in size. Swelling was non-tender, compressible, reducible on palpation and not fixed to the underlying structures. No palpable regional lymph nodes were noticed. Overlying skin appears to be normal. Intra-orally diffuse swelling present on the right buccal mucosa with prominent inflamed ductal orifice of parotid associated with drooling of saliva.

Ultrasonography was done which ruled out possibilities of Sialolithiasis. Ultrasonography showed a cystic lesion measuring 3.0×3.0×2.5 cm, with internal echoes in the right side of face in the subcutaneous plane. Aspirate was collected and sent for histopathological examination. The aspirate showed turbidity. Protein content of the aspirate was also analyzed which came out to be approx.1gm /lit (100mg /dL). H & E and PAS staining of the smear revealed presence of few squamous epithelial cells, chronic inflammatory cells along with numerous PAS positive granules. With the presenting complaints, clinical examination, and ultrasonography as well as after analysis of aspirate the differential diagnosis included a mucous retention cyst and Parotid duct cyst. The cystic part of parotid duct was excised and specimen submitted for histopathological examination.

Histological examination of H & E stained section of the lesion revealed presence of a cystic cavity with numerous papillary projections into the cystic space. These papillary projections were lined predominantly by stratified squamous epithelium and in few areas by psuedostratified ciliated columnar epithelium. Numerous bilaminar, refractile, eosinophilic cystic spaces containing hematoxyphilic granules (endospores), resembling sporangia were seen in the wall of the cyst. Sporangia were in different phases of maturation. Many empty sporangia were seen due to discharge of endospores in the surrounding connective tissue.

The surrounding connective tissue was quite vascular and contained numerous dilated blood vessels engorged with RBCs. Intense chronic inflammatory cell infiltrate was seen immediately subjacent to epithelium as well as surrounding sporangia; chiefly in the form of lymphocytes, plasma cells as well as few eosinophils. On PAS staining of the tissue section Sporangium and sporangiospores showed positivity along with few other areas like psuedostratified ciliated columnar lining also showed positive reaction with PAS. The histopathology was suggestive of Rhinosporidiosis.

DISCUSSION

First case of Rhinosporidiosis was reported by Malbron of Buenas Aires in 1892. Seeber in 1990 first described in detail about the microorganism causing Rhinosporidiosis. The life cycle of the microorganism causing Rhinosporidiosis was given by Ashworth in 1923.[5,6] Morphology of the microorganism is similar to that of a fungus and protozoa. But Rhinosporidium is considered to be a fungus because of its periodic acid-Schiff positivity and taking up silver stains. [5, 6]

Rhinosporidiosis is endemic in India and Sri Lanka, but it is also reported in South America, US England, Egypt, and South Africa. In India, the disease is endemic in the states of Chhattisgarh, Kerala, Tamil Nadu, Orissa and West Bengal [7]. The microorganisms enter into the body via any traumatized epithelium; most commonly they enter through nasal mucosa. Most of cases occur in upper respiratory sites.[8] The first case of rhinosporidiosis of the parotid duct was reported by Topazian in the year 1967.[9] Till date very few cases of rhinosporidiosis involving parotid duct have been reported. By clinical examination only it was difficult to establish a diagnosis because clinical

presentation was similar to that of retention cyst. There was history of increase in the size of swelling during meal time. This patient presented no signs and symptoms of Rhinosporidiosis at any other site. Analysis of aspirate also showed increased protein content. Cytology also revealed presence of mucous .All these findings were leading towards a diagnosis of retention cyst caused by mucus plug.

Diagnosis in the present case was established after examining H & E stained section of the excised lesion which showed presence of large, thick-walled spherical structures called sporangia in the wall of the cyst (50-1000 µm). These sporangia are lined by thick homogeneous eosinophilic capsule. These sporangia contain numerous smaller daughter cells known as "sporangiospores" (20-80 µm). The surrounding connective tissue shows vascular granulation tissue and mixed inflammatory cells. These sporangia as well as sporangiospores can be visualized by routine Hematoxylin & Eosin stain as well as by PAS , mucicarmine & Gomori methenamine silver stain. [8, 10]

It is important to distinguish R. seeberi from Coccidioides immitis. C. immitis shows similar large, thick-walled, sporangia containing endospores, but the sporangia are of smaller diameter (20- 80 µm) when compared with R. Seeberi (50- 1000 µm) and contain small endospores (diameter of 2 to 4 µm). Moreover, C. immitis shows negativity for mucicarmine stain whereas R Seeberi shows positivity for the same.[8,11]

Differentiation of R. seeberi from C. immitis is important as rhinosporidiosis is refractory to antibiotic therapy. Dapsone is given for treating the cases of rhinosporidiosis. It arrests maturation of sporangia and enhances degenerative changes in them. Treatment of choice is surgical excision. Antifungal agents such as griseofulvin and amphotericin B are tried to reduce recurrences. But they do not appear to be promising. Complete excision with wide surgical margins is the important as recurrences occur because of spillage of endospores in the surrounding mucosa.[8]

CONCLUSION

Though Rhinosporidiosis is common in the nasal cavity and respiratory tract, but it can occur at unusual sites also. Hence in case of any facial swellings, one should think of Rhinosporidiosis as one of the differential diagnosis .it is evidenced from this case report that though fungal infections are rare to occur; still their importance as one of the differential diagnosis cannot be denied.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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