CLINICAL REPORT

A Rare Presentation of Rhinosporidiosis

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Abstract Rhinosporidiosis is a common condition seen in India specially in endemic areas and in low socio economic areas. Here we present with a rare presentation of cheek swelling caused by obstruction of stenson's duct with a short history of 5 months. Sialolith of parotid duct is uncommon presentation but not unheard of, the subsequent treatment and further histopathology of the ductocele revealed the causative factor for the obstruction of parotid duct was Rhinosporidiosis which a very rare presentation with very few cases reported in literature. On further investigations it was found to be primary Rhinosporidiosis. Hence it is eminent that Rhinosporidiosis should also be kept in mind while looking for a cause for salivary duct obstruction specially in endemic areas.

Keywords Parotid duct · Rhinosporidiosis · Ductocele · Sporangium

Introduction

Rhinosporidiosis is a benign granulomatous disease caused by *Rhinosporidium seeberi* (*R. seeberi*). (1) Rhinosporidiosis has been reported from about 70 countries with diverse geographical features although the highest incidence has been from India and Sri Lanka. In India, the

disease is endemic in the states of Chhattisgarh, Kerala, Tamil Nadu, Orissa and West Bengal. (2) Rhinosporidiosis caused by the organism *R. seeberi* is a chronic granulomatous infection involving the mucous membranes.

The most common site of involvement is the nose and nasopharynx where the lesion presents as a polypoidal mass with irregular surfaces. The next common site is the eye. Lesions of rare sites have been reported such as bone, vagina, vulva, oropharynx, buccal mucosa, tongue, paranasal sinuses, skin, subcutaneous tissue, penis, urethra, trachea, larynx and bronchus. Most of the patients present with Rhinosporidiosis of the nasal cavity, nasopharynx and oropharynx. Molecular biological techniques have more recently demonstrated this organism to be an aquatic protistan parasite, and it has been placed into a new class, the Mesomycetozoa, along with organisms that cause similar infections in amphibians and fish [1, 2]. This reclassification is not without controversy, as other researchers have presented data that *R. seeberi* is a cyanobacterium, further demonstrating the difficulties that arise when working with pathogens that cannot be maintained in the laboratory setting. Other molecular work has demonstrated evidence that *R. seeberi* may have host-specific strains (e.g., human vs. dog vs. swan). The involvement of Rhinosporidiosis in parotid duct extremely rare, and at present there have been a handful of cases reported worldwide. An extra nasal involvement is rare, it hinders diagnosis at the initial examination stage because of lack of familiarity of site and symptoms [4].

Case Report

A 16-year-old male patient presented with cheek swelling of the left side since past 5 months. There was no history of previous trauma or recurrent parotitis in the past. The

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swelling increases during the time of meals or on consumption of any food, it is not associated with pain or tenderness over the swelling. The swelling persists even after the time of meal until the saliva is expressed out to oral cavity by compression. Past history revealed patient had history of pond bathing since past 3–4 years (Fig. 1).

On clinical examination there was a soft cystic swelling over the left cheek which measured to 3×2 cm. On palpation the mass was soft, non-tender, unilocular, fluctuant swelling with smooth surface. It was situated anterior to masseter and was not fixed to the skin or underlying tissue. Intra oral examination shows inflamed and dilated Stensen's duct opening. On compression on the swelling expulsion of thick seromucinous fluid (saliva) is seen draining through the duct opening. Rest of the oral cavity examination was normal. Both nasal endoscopy and otoscopy of both sides revealed no relevant findings. Patient was subjected to radiological examination of CT scan with dynamic contrast sialography which revealed a cystic dilated part of Stensen's duct (left side) in the anterior part of the gland measuring 2.6 cm (AP) $\times$ 2.7 cm (TR) $\times$ 3.9 cm (CC) with the posterior part of the duct seen in continuity with the posterolateral aspect of this lesion. The right side had normal contrast filling in the Stensen's duct (Fig. 2).

Case was taken up for surgery under general anesthesia. The openings of the Stensen's duct on the affected side was

cannulated with epidural catheter. A horizontal incision of approximately 2 cm incision was made on the most prominent part of the swelling which revealed a thin walled cystic swelling deep to subcutaneous layer of skin which was traced both proximally and distally and were found to be in continuity with the Stensen's duct posteriorly thereafter it was excised where the duct appeared normal. An epidural catheter was used to stent both the distal and proximal ends of excised duct and an end to end anastomosis was done with sutures, to maintain the patency of the duct. The cyst was excised in toto without rupture of the walls (Fig. 3). The excised specimen was submitted for histopathologic examination which showed cyst wall lined predominantly by fattened epithelium and focally by metaplastic squamous epithelium. The cyst wall shows many thick walled sporangia containing endospores surrounded by chronic inflammatory infiltrate comprised predominantly of lymphocytes and focally by foreign body type multinucleated giant cell reaction. Implying that the cyst walls contained *R. seeberi* sporangia in them (Fig. 4).

Retrospective pan endoscopy is done for nasal cavity, nasopharynx, pharynx and larynx to look for evidence of rhinosporidiosis. No morphological lesions were seen in any other sites. Post operatively the epidural catheter was left in situ for 3 weeks and prophylactic antibiotics for a week and anti-inflammatory for 3 weeks. After 3 weeks the epidural catheter is removed and regular milking of parotid duct is advised. The patient is being under regular follow up for 1 year with no recurrence of the disease or symptoms of parotid duct obstruction.

Discussion

In 1923, Ashworth was the first person who described the life cycle of the organism. Recently it has been postulated that the causative organism is not a fungus but a Cyanobacterium, *Microcystis aeruginosa*. The mode of infection from the natural aquatic habitat is through the traumatized epithelium and can spread by auto-inoculation, hematogenous route or lymphatics. A review of articles shows the predominance of diseases more in South Asia, with male to female ratio of 4:1. All ages of patients are affected along with this high possibility of recurrence even after prevention of source of infection. Recurrences can occur due to spillage of endospores into adjacent epithelium or because of incomplete removal of base of the stalk during excision. Present case of Rhinosporidiosis of the parotid duct is a very rare occurrence with as low as 5 cases reported in literature [5]. The first case of rhinosporidiosis of the parotid duct was reported by Topazian in the year 1967. Subsequently, there are only four published cases in the literature; all of them are from India. In all these cases,

Fig. 1 Pre OP

Fig. 2 Pre OP radiologic imaging

Fig. 3 Intra OP images with specimen

Fig. 4 (HE 10×) (PAS 20×)

the patients presented with a cystic swelling in the parotid region and were clinically diagnosed as parotid duct cyst. Rhinosporidiosis was not suspected in any of these cases pre-operatively [6–10]. Rhinosporidiosis of nasal cavity, nasopharynx, oral cavity and larynx could easily be justified with history of pond bathing and drinking of contaminated water.

Mandalik et al. suggested the possibility of natural patulous and everted anatomy of the orifice of the parotid duct in the buccal cavity could have promoted a quick passage of the spores into the duct during mouth washing with pond water resulting in the development of the disease [3].

In this particular case the options of superficial parotidectomy or excision of the effected part of the duct with end to end anastomosis of the duct with stenting was

available. Excision the duct was considered and was carried out taking special consideration age of the patient and with reassurance of regular follow up.

Conclusion

The prevalence of Rhinosporidiosis is high in India, mostly in south and central India, the commonest sites effected being nasal cavity, oropharynx and nasopharynx. With India being an endemic area, Rhinosporidiosis should be kept in mind when thinking of a differential diagnosis for a mass in any part of head and neck region. The incidence of Rhinosporidiosis contributing to a ductocoele is quiet rare with less than 10 cases being reported in literature. This case report is in view of Rhinosporidiosis in parotid duct

leading to ductocoel. Hence concluding that a Rhinosporidiosis could also be one of the causative factor for formation of ductocoel and should always be looked for in histopathology of specimen to rule out Rhinosporidiosis.

Compliance with Ethical Standards

Conflict of interest None.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from the individual participants included in the study.

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