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Case Report

A rare presentation of rhinosporidiosis on buccal mucosa – a case report

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Abstract – Introduction: Rhinosporidiosis is a chronic mucocutaneous infection caused by *Rhinosporidium seeberi*, which mainly affects the mucus membranes of the nose, oropharynx, skin, eyes, and genital mucosa. The purpose of this case report was to illustrate the importance of clinicians in understanding the unusual incidence of this fungal infection, clinical presentation, treatment modalities, and recurrence rate. **Observations:** A clinical case report showed rare occurrence of rhinosporidiosis in the lower third of the face. The lesion was surgically excised by electrocauterization at its base. **Commentaries:** The patient showed no signs of recurrence even after 3 years of follow-up. Further research should be carried out to analyze the role of genetic predisposition in causing zoonotic infection. **Conclusion:** Rhinosporidiosis should also be considered as the differential diagnosis when facial swelling is encountered.

Introduction

Rhinosporidiosis is a chronic infection of the mucous membrane caused by the mesomycetozoan *Rhinosporidium seeberi* [1]. It is usually found in endemic areas, especially in tropical and subtropical regions such as India and Sri Lanka (88% of total cases), followed by South American and African countries [2–5]. However, it is rarely observed in European and American populations [6–8]. The lesions present as a polypoidal strawberry-like mass, pink to purple in color; sometimes pedunculated, in the nose (70%), eye (15%), throat, ear, and respiratory tract and rarely seen in the penis, lips, skin, and uvula [9–11]. It is a highly vascular lesion that bleeds upon touch. Rhinosporidiosis can occur at any age but commonly occurs between 20 and 35 years; males are four times more commonly affected than females. We report a case focusing on the need for clinicians to be aware of the unusual presentation of rhinosporidiosis in a clinical entity with comprehensive facts regarding the incidence, clinical presentation, and treatments followed by the recurrence rate.

Observations

A 30-year-old male patient presented with a chief complaint of swelling on the right side of the face for 3 months. The swelling was initially small and gradually progressed to the present size. On extraoral examination, facial asymmetry was noted with a well-defined unilateral swelling on the right lower third of the face, measuring approximately 2×2.5 cm, extending anteroposteriorly 2.5cm behind the commissure of the lip to 2 cm ahead of the tragus and superoinferiorly it extends from a line joining the tragus to the inferior border of the mandible. Sialography of right parotid gland shows irregular filling of parotid duct in distal part with pooling of the contrast in the area of the lesion suggestive of a cystic cavity. Intra-oral examination showed a fair oral hygiene status with swelling extending, anteriorly from the right buccal mucosa to the distal aspect of the upper right 1st molar tooth. The surface of the lesion appeared smooth and glossy (Fig. 1). On palpation, the swelling was soft and tender, compressible, and was not fixed to the underlying structures with no evidence of sinus opening or discharge. Examination of the ear, nose, nasopharynx and other systemic examinations did not reveal any other abnormalities. The patient was asymptomatic with no other contributory medical history or habit history such as consumption of unprocessed well water or pond bath to confirm

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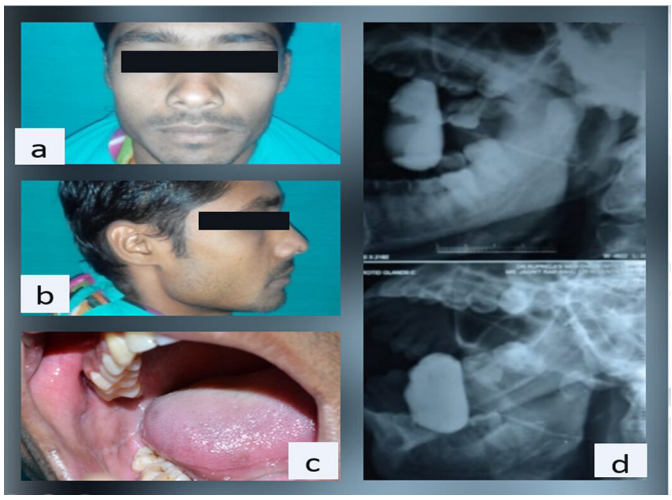

Fig. 1. depicts preoperative (a) Extra-oral front view, (b) Extra-oral side view, (c) Intraoral view of the lesion, (d) Sialography of right parotid gland shows irregular filling of parotid duct in distal part with pooling of the contrast in the area of the lesion suggestive of a cystic cavity.

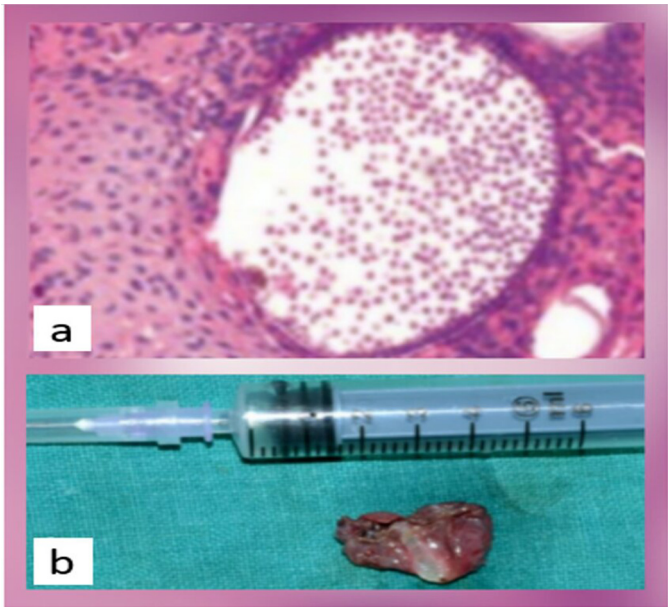

Fig. 2. (a) H& E stained section viewed under magnification of 40× shows sporangia containing numerous endospores, with chronic inflammatory cells & plenty of eosinophilis. (b) Aspirated serous fluid and excised surgical specimen measuring 2 × 1.5 cm.

the diagnosis of rhinosporidiosis. Routine laboratory investigations were within normal limits except for elevated ESR (Erythrocyte sedimentation rate). Histpathological examination of the surgical specimen stained with haematoxylin & eosin (H& E) was further viewed under the electron microscopy of magnification 40× showed sporangia containing numerous endospores with an intense chronic inflammatory cells infiltrate mainly lymphocytes and few plasma cell with plenty of eosinophilis. On aspiration using a 22 gauge 5 ml syringe yielded a serous fluid (Fig. 2). The culture showed fungal spores

Fig. 3. The intraoperative pictures of (a) incision placement, (b) cystic bag appearance of the lesion, (c) curettage after surgical excision.

along with inflammatory cells. The lesion was surgically excised with electrocauterization at its base (Fig. 3). There was no recurrence within a follow-up period of 3 years.

Commentary

Rhinosporidiosis is commonly seen in young males who are occupationally active outdoor workers such as agriculturists, sand workers, divers *etc*. The causative agent of the rhinosporidiosis is a mesomycetozoan known as *Rhinosporidium seeberi*. The nasal cavity was the most common site of rhinosporidiosis followed by the conjunctiva, upper eyelid, pharynx, trachea, cheek, parotid duct, skin, and genitalia. Rhinosporidiosis manifests as a tumour-like friable, vascular pedunculated or sessile polyp mass, with tiny white dots, often present in the nasal mucosa or ocular, conjunctivae, of humans and animals. It is a slow-growing waterborne disease, often present as a dormant stage before the patients become symptomatic. It enters through the traumatized epithelium and the disease is more prevalent in rural settings, particularly among individuals working or in contact with contaminated or stagnant pond water [5]. As the causative agent enters the host's body, it forms a round and thick-walled sporangia in the submucosa of the affected site, and appears as white dots producing an endosporulating cells known as sporangiospores of size ranging from 10 to 200 mm. Current study reported a rare presentation of rhinosporidiosis on the lower third of face. Histopathological examination showed connective tissue composed of numerous bilamellar spaces resembling sporangia with an increase in eosinophils. In addition, aspiration for cytology and culture showed yeast forms/spores of fungi with acute inflammatory cells, suggesting fungal infection. Therefore, rhinosporidiosis should be considered as one of the differential diagnosis of the facial swelling. Spontaneous regression of rhinosporidiosis in animals and humans is rare and surgical intervention is the main treatment modality. Current clinical case of rinosporidiosis showed a successful outcome with surgical

excision along with electrocauterization of the base. There was no recurrence even after 3 years of follow-up. In- contrast, few studies [12–14] have shown recurrence with surgical excision alone. Therefore these studies used surgical excision along with dapsone as the main treatment modality to arrest the maturation of sporangia and to promote fibrosis in the stroma. A study conducted by Deepa *et al.* [15] reported a recurrence rate in immunocompromised patients despite using dapsone. This could be due to genetic variation in the host and their altered responses to certain drugs. However, a combination of dapsone with antifungal drugs such as griseofulvin, amphotericin B, trimethoprim sulphadiazine, and sodium stibogluconate have been proven to reduce the rate of recurrence of rhinosporidiosis [16,17].

In summation, the pathogenesis and recurrence rate of rhinosporidiosis are still considered a mysterious factors. Thus, constant effort from the clinicians as well as microbiologists should be put to identify an environmental niche and to measure the ecological density of the organism in various water bodies. Few studies have stated whether that nonspecific immune reactivity in the host, blood group and HLA types play important role in initiating the focus of infection of rhinosporidium seeberi [5]. However, further research should be carried out to analyze the role of genetic predisposition or increased exposure in the environment in causing this mesomycetozoan infection.

Conclusion

Rhinosporidiosis is a rare granulomatous mucocutaneous infection caused by *Rhinosporidium seeberi*. The transmission of an organism is most commonly mediated by exposure to spores from contaminated dust, soil and water. Surgical intervention is the treatment of choice. However, further studies should be carried out firstly, regarding etiopathogenesis and the role of genetics in the recurrence rhinosporidiosis. Secondly, clinicians should focus on minimally invasive treatment modalities for rhinosporidiosis which can be easily practiced in rural settings.

Authors contributions

B.S. Santosh: Concepts & Design; A. Harish Kumar: Helped with search of clinical studies; Rajdeep Singh: Data Acquisition; Jerin Jose: Manuscripit preparation, editing; D.M. Shivamrthy: Helped with editing; Diana Daniel: Helped with review.

Informed Consent

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Ethical committee approval

Institutional ethical committee approval was taken. Informed consent from the patient obtained.

Conflict of interest

The authors declare that they have no conflicts of interest in relation to this article.

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