

## Case Report

# An unusual palatal mass in a 54-year-old male: report of a rare case

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(Received: 4 June 2022, accepted: 12 September 2022)

**Keywords:**  
Rhinospordiosis /  
palate /  
Rhinospordium  
seeberi / dapson

**Abstract** – Rhinospordiosis is a chronic, granulomatous, muco-cutaneous infection caused by *Rhinospordium seeberi*. The infection is non-contagious and sporadic in humans, affecting predominantly the mucous membrane of the upper respiratory tract. It is quite an uncommon condition and seldom affects the oral mucosa. The clinical presentation of this condition in the oral cavity can mimic many other conditions. Here, we report a unique case of recurrent rhinospordiosis affecting the palate in a 54-year-old male.

## Introduction

Rhinospordiosis is an enigmatic chronic infection that is often perceived to be either fungal or protist in origin. The nature of the organism is debatable to date. It is endemic to South East parts of Asia and Sri Lanka. The disease spreads through contaminated water. It has a high recurrence rate with a rare tendency to disseminate from the original site to other areas of the body [1]. Clinically, the lesions present as painless, polypoid soft tissue masses that bleed readily. The clinical presentation of the condition in the oral cavity is a rare occurrence and very few cases are reported in literature. Hence, oral health care professionals need to identify, understand the etiology, clinical nature and management of this condition. Here we present a rare case of recalcitrant rhinospordiosis secondarily affecting the palate in a 54-year-old otherwise healthy male.

## Case report

A 54-year-old male reported to the Department of Oral Medicine and Radiology with a complaint of a painless growth in the palate of one year duration. Medical history revealed that patient had undergone surgical excision of recurrent

rhinospordiosis of sino-nasal tract multiple times in past last 22 years. Most recent surgery was performed one year back, a few months after which, he noticed a small growth in the palate. The growth was initially pea-sized which gradually increased to the present size. Patient did not give any history of similar oral lesions in the past. He did not have any other systemic comorbidities and/or immunocompromised state. The oral abusive habit history was non-contributory.

General and extra-oral examination revealed no abnormalities. Intra-orally, a large, irregular fleshy mass was seen on the midline of the hard palate extending posteriorly to the junction of hard and soft palate, anteriorly up to alveolus and upper incisors (Fig. 1). The surface of the growth had a nodular appearance and covered with greyish-yellow slough. On palpation, the growth was non-tender, mobile, firm in consistency with smooth texture and a broad base. There was no evidence of bleeding. Maxillary teeth were not mobile and there were no signs of oro-antral communication.

Taking into account the past history of sinonasal rhinospordiosis of refractory nature, we got a strong clinical suspicion of palatal involvement of the same. A differential diagnosis of palatal malignancy or a sino-nasal carcinoma invading into palate was also considered. The possible differential diagnosis of fleshy masses on the palate are discussed in Table I.

Panoramic radiograph revealed bilateral obliteration of the maxillary sinuses and sinus floor (Fig. 2).

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**Fig. 1.** Photograph showing the fleshy mass in the palatal mucosa on the left side and mirror image of the palatal mass on the right side.

**Table I.** Table depicting lesions that can present as palatal masses with differentiating features [2–9].

Lesions	Differentiating features	Histopathological features
<u>Benign &amp; malignant tumors</u> Epithelial origin: Squamous Cell Carcinoma (SCC)	Exophytic, proliferative growth with irregular borders and indurated base	Crystal violet and Feulgen stains are superior in identifying the mitotic figures than H&E [2]
<u>Salivary origin:</u> Mucoepidermoid carcinoma Pleomorphic adenoma	Firm, painless swelling in the palate that is compressible Sometimes it has a blue/ red hue on the surface mimicking a mucocele [4]	Alcian blue and Mucicarmine stains are positive [3] Pleomorphic adenoma stains positive for mucicarmine [3]
<u>Naso-maxillary origin:</u> Malignancy of maxillary sinus	Rubbery submucosal mass in the palate without surface ulceration The condition can present as palatal swelling/mass when there is infiltration of malignancy into the oral cavity with probable clinical symptoms (nasal obstruction, paraesthesia of zygomatic region) [5]	
<u>Infections</u> Actinomycosis Mucormycosis	More common in immunocompromised individuals. Acute, painful palatal abscess with characteristic sulphur granules [6] Chronic ulcer with raised margins, exposing the underlying bone [8]	Gomorimethenamine silver stain is useful in identifying the filaments of the organism [7] Gomorimethamine silver identifies the fungus. Further gram stain can be done to differentiate from Actinomycosis, which is a bacterial infection [9]

High Resolution Computed Tomography (HRCT) scan of paranasal sinuses revealed a large mass measuring about 6 cm in the nasal cavity, causing destruction of septum, nasal turbinates and extends into the maxillary antrum. There was expansion and rarefaction of the hard palate on both sides with cortical erosion Lesion infiltrated the underlying soft tissue and caused cortical erosion of the maxillary arch (Fig. 3).

Patient was advised incisional biopsy of the lesion and was referred to Department of ENT for further management.

Histopathological examination of the lesion revealed keratotic, acanthotic hyperplastic stratified squamous epithelium overlying the fibro-collagenous stroma showing numerous variable sized sporangia with endospores and inflammatory response composed of lymphocytes, plasma cells, neutrophils, eosinophils, clusters of foreign body giant cells with engulfed spores suggestive of rhinosporidiosis (Fig. 4).

Patient was started on Dapsone therapy 100 mg once daily for a month and is on regular follow-up.



**Fig. 2.** Panoramic radiograph showing complete obliteration of bilateral maxillary sinuses.



**Fig. 3.** High Resolution Computed Tomography (HRCT) scan of paranasal sinuses showing a large mass measuring 6cm in the nasal cavity in axial and sagittal sections.

## Discussion

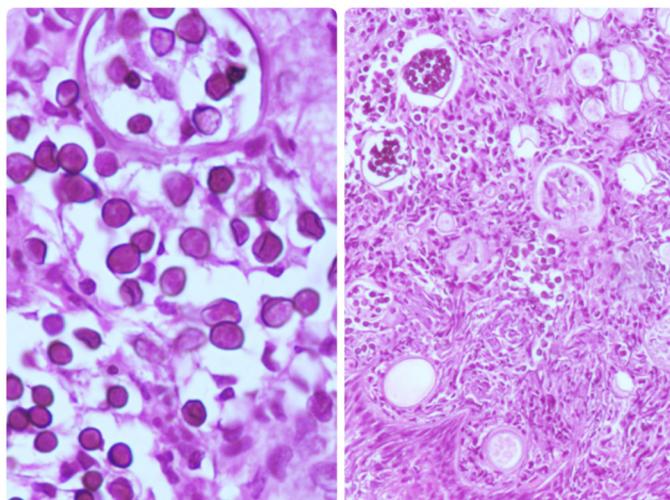
Rhinosporidiosis is a chronic infection caused by *R. seeberi* chiefly affecting the upper respiratory mucosa followed by oropharynx, eyes and genital mucosa. The disease has been known for over a century, after its first incidence in Argentina [1]. It is a chronic granulomatous disease endemic in South India, Srilanka, and some areas of the African continent, common in the tropics [2,10]. The disease affects all ages, but it is more common in third and fourth decades of life.

The causative organism is *R. seeberi*, generally known as 'water mould' [3,11]. Recent molecular studies have shown that the organism may be either a Cyanobacterium or a Protist [4,12]. However, the exact nature of the organism is still not established precisely. The infection spreads through ingestion of contaminated water. Water and soil are likely to be the pool of infection, thus frequently occurring in sand workers, farmers and people working in sluggish muddy water. It is suggested that endospores are infiltrated into the nasal mucosa which then mature into sporangia and burst out into the adjacent tissues [5,13]. Coincidentally, our patient happened to be a water supplier by

profession for over 15 years and the recurrence could be attributed to water infested with the organism.

Clinically, the disease can manifest in 4 forms: nasal, ocular, cutaneous and disseminated types. Nasal form is the most often encountered manifestation and clinically presents with epistaxis and associated sessile pink-red polyp in the nose. Ocular form is often presented as a sessile friable pedunculated flat polyp in the eye. About 15% of the cases have ocular manifestations involving either palpebral or bulbar conjunctiva, lacrimal sac or nasolabial duct [6,14]. Skin involvement is seen as tiny papules with warty growth which may manifest as an infected ulcer. The disseminated type is seldom seen as spherules of *R. seeberi* in the lungs, bone, liver, spleen, limbs or brain [3,11]. Parotid involvement has been reported in literature by Topazian [7,15] and Sivapathasundaram [8,16]. Rhinosporidiosis involving the parotid duct causing bilateral facial swelling was reported by Santanu *et al.* [9,17]. The only case of rhinosporidiosis with palatal involvement was reported in a 60 year old woman by Harshita *et al.* [3,11].

The disease is characterised by the formation of vascular, friable lesions that are papillomatous or polypoidal with a granular surface [10,18]. The lesion is extremely vascular and



**Fig. 4.** Pictomicrograph ( $\times 40$ , hematoxylin and eosin stained) showing sporangia on the left side and pictomicrograph ( $\times 10$ , hematoxylin and eosin stained) showing chronic inflammatory infiltrates and giant cells with sporangia on the right side.

readily bleeds on touch. Histologically it is characterized by the presence of mucosal and submucosal cysts (sporangia). Sporangia contain numerous endospores seen with hematoxylin-eosin; organism [11,19]. Spores are better demonstrated with Verhoff's von Gieson, Mayer's mucicarmine, Grocott Gomori methamine silver stains [12,20]. Other laboratory investigations are not conclusive in nature.

Treatment is a complete surgical excision, but recurrence rates are high and is somewhere between 10% and 30%. According to a study done by Bhandary *et al.* (2012), the recurrence rate of the infection was 22.6% and about 4% had multiple recurrences [13,21]. The use of laser surgery, cryosurgery, coablation, harmonic scalpel and electrocautery reduce the recurrence. Medical management of rhinosporidiosis is still a viable area for research. The organism does not respond to antibiotics or antifungals. However, Dapsone is considered to be effective as it ceases the maturation of sporangia and causes fast degeneration, increases the granulomatous response with fibrosis and prevents recurrence by interfering with folic acid metabolism of the organism [11,19]. Considering the same, our patient was started on oral tablet Dapsone 100 milligrams once daily for one month with which he has shown significant improvement and patient now is on regular follow-up.

The disease is known for its notoriously high recurrence rate. The rationale behind the high recurrence of the infection is still not clear, though some school of thought believes that it is due to spillage of spores into the adjacent tissue during surgery; caution should be taken for this particular adversity of the lesion.

## Conclusion

Although presentation of rhinosporidiosis in the oral cavity is comparatively rare, one should consider it in the differential diagnosis when a fleshy mass is seen. As the infection

often mimic malignancy, prompt investigations along with histopathological diagnosis is mandated for early diagnosis. Patient has to be on lifetime follow-up due to the increased risk of multiple recurrences.

## Conflict of interest

The authors have no conflict of interest to disclose.

## Funding

This research did not receive any funding.

## Ethical statement and Informed consent

The consent was obtained from the patient for publication.

## Author contribution statement

Ranjane Srinivasan: Concepts, design and manuscript preparation; Mathangi Kumar: Design, manuscript preparation; Ravindranath Vineetha: Manuscript editing; Dipak Ranjan Nayak and Swati Sharma: Investigation and surgical management.

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