CASE REPORT

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Palatal rhinosporidiosis: an unusual case report and review of the literature

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Abstract (J Korean Assoc Oral Maxillofac Surg 2018;44:293-297)

Rhinosporidiosis is a chronic, granulomatous, mucocutaneous infection caused by *Rhinosporidium seeberi*. The infection is non-contagious and sporadic in humans. The site most commonly affected is the mucous membrane of the nose and nasopharynx, followed by the oropharynx, trachea, bronchi, ear, eye, and genitourinary tract. It can also spread to other areas through blood and lymph. Here, we report a case of rhinosporidiosis affecting the palate in a 60-year-old female patient.

Key words: Rhinosporidiosis, *Rhinosporidium seeberi*, Sporangiospores, Granulomatous infection

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I. Introduction

Rhinosporidiosis is a rare granulomatous infection caused by *Rhinosporidium seeberi*[1]. *R. seeberi* was discovered by Guillermo Seeberi in Argentina in 1990[2]. It is widely considered to be a fungus as its morphological features are similar to those of fungi and protozoa[1]. Cases of this disease have been documented for over 100 years, long before the causative agent was described in 1990[3]. It affects both humans and domestic animals endemic to India and Sri-Lanka[4]. The disease commonly affects the mucous membrane of the nose and nasopharynx, along with some reported cases involving the eye, ear, trachea, bronchi, genitourinary tract, larynx, and parotid gland[5]. Clinically, the disease presents as a polypoid, soft mass of the nose (most common site), eye, conjunctiva, or urethra. Osteolytic bone infiltration is another identifying feature[6]. Antifungal and/or antimicrobial drugs are not effective against *R. seeberi*, and the only treatment for rhinosporidiosis is surgical excision of the affected area with deep cauterization to prevent reoccurrence. Here, we describe a case in which a 60-year-old female presented with a large palatal ulcer.

II. Case Report

A 60-year-old female reported to an outpatient department complaining of pain and an ulcer in the upper right back tooth region for the past 18 to 20 days. Medical history revealed that extraction of the right maxillary 1st and 2nd molars had occurred 22 days prior. Two days following the extraction she began experiencing severe pain at the site of extraction concurrent with the appearance of a white patch/necrosed area on the right palate which gradually increased to the size observed in our department. She reported severe radiating pain throughout her head region which was aggravated by chewing and relieved by medication. There was no other significant family or surgical history.

Upon extra-oral examination, a solitary, soft swelling was noted on right side of the face extending superior-inferiorly from the infraorbital region to the corner of the mouth and antero-posteriorly from the ala of the nose to 1 cm anterior to the tragus of the ear, measuring about 3 cm×3 cm at its largest point. Upon palpation, the area was soft to the touch.

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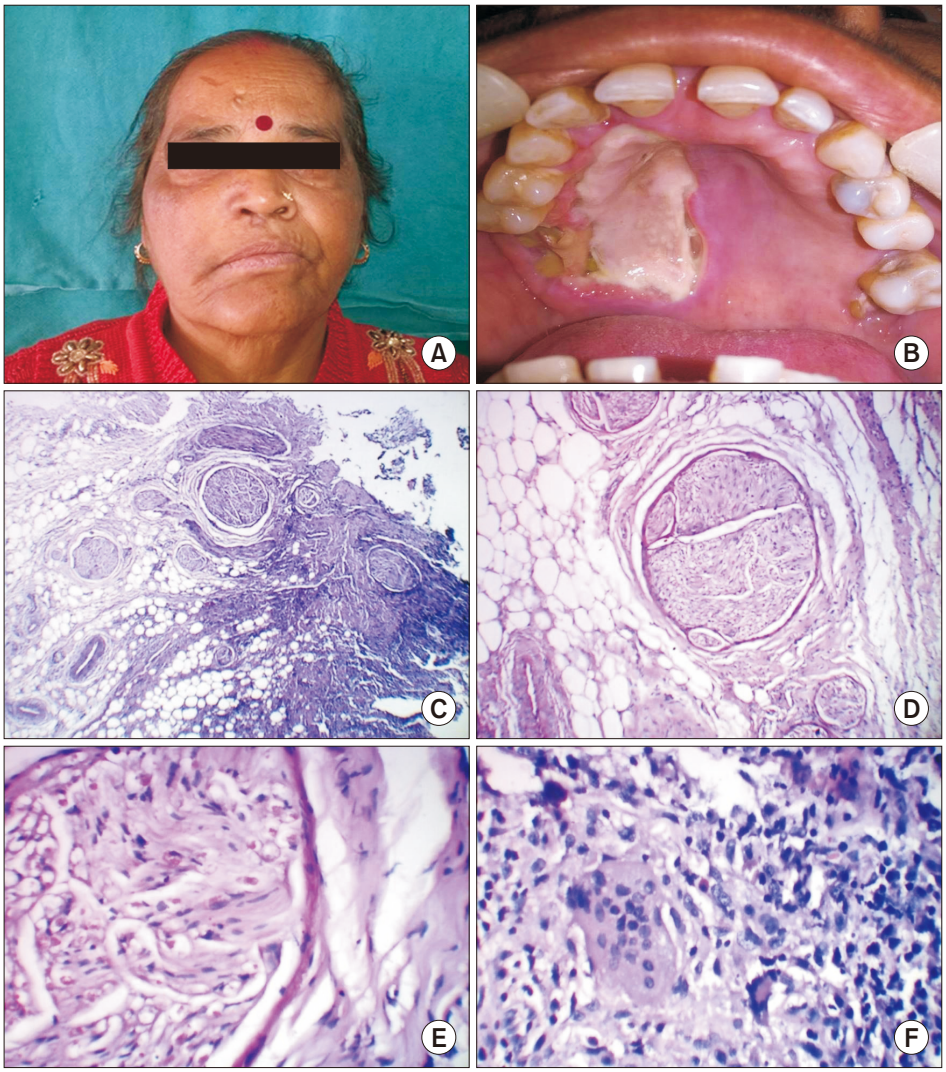

Fig. 1. A. Extraoral photograph of a patient depicting swelling on the right side of the face. B. Intraoral photograph depicting a non-healing ulcer on the lateral part of palate. C. Photomicrograph of a H&E stained section viewed at low magnification (×40) showing multiple mature and immature sporangia with numerous adipocytes. D. Photomicrograph of a periodic acid-Schiff (PAS) stained section viewed at low magnification (×100) showing magenta, thick walled sporangia. E. Photomicrograph of a PAS stained section viewed at high magnification (×400) showing magenta, thick walled sporangia. F. Photomicrograph of H&E stained section viewed at high magnification (×100) showing inflammatory infiltrate with giant cells.
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(Fig. 1. A) Intraoral examination revealed a solitary ulcer on the right side of the palate which was roughly oval in shape, measured about 2 cm×3 cm, and extended antero-posteriorly from the palatal aspect of the right maxillary incisor to the anterior region to the soft palate and mesiodistally from the mid-palatine raphe to the alveolar bone. The margins were well-defined and surrounded by an erythematous area with a surface covered by slough. Upon palpation the lesion was tender, non-scrapable, and associated with oro-antral communication along with occasional epistaxis.(Fig. 1. B) All vital signs were within normal limits. The patient had a history of diabetes over the last four years and was taking regular diabetes medication. After clinical examination a provisional diagnosis of mucormycosis was made, and surgical excision of the whole lesion was planned. Upon haematological investigation, the complete blood count was under the normal limit except for the diabetic profile, where HbA1c was 11.8%.

Gross examination of the surgically excised tissue revealed that it was cream/brown in colour and soft-firm to the touch. There were five total pieces of tissue, the largest of which measured 3 cm×3 cm. Four representative pieces were sectioned, processed, and stained with H&E stain and periodic acid-Schiff (PAS) stain.

Microscopic examination of H&E stained sections at low magnification revealed highly inflamed connective tissue in the stroma with multiple small (immature) to large (mature) sporangia. Numerous adipocytes were also seen in the stroma.(Fig. 1. C) In PAS stained sections a large sporangium was present with a thick magenta-coloured wall along with multiple spores, suggesting different stages of development. (Fig. 1. D, 1. E) At high power magnification the presence of a mixed inflammatory infiltrate predominantly comprised of lymphocytes, histiocytes, and neutrophils was evident. A few giant cells were also noted.(Fig. 1. F) The overall histopatho-

logical features were suggestive of rhinosporidiosis following a clinic-pathological review.

When the patient was re-examined, she did not show any evidence of involvement of extraoral sites. The patient did not give any history of trauma or bathing in stagnant water. She reported back to the outpatient department for follow-up three months after surgery, and there were no signs of recurrence of disease.

III. Discussion

Rhinosporidiosis is a distinctive granulomatous infection of the mucocutaneous zone[6]. It has been known since ancient time and was first noted in Argentina over 100 years ago[4]. The causative organism, *R. seeberi*, was originally thought to be a parasite commonly referred to as water mould. It was first proposed that the organism was a cyanobacterium[7], though it was later suggested to be a fungus[8]. However, molecular techniques have recently identified the organism as an aquatic protistan parasite in the Mesomycetozoea class[9]. This disease has global occurrence, is widely reported in India, and can cause delayed, painless disease with confined morbidity. There are reports of the disease lasting 30 years. Secondary bacterial infection can cause increased morbidity. In addition to humans, rhinosporidiosis can affect several species of farm, domestic, and wild animals including cattle, buffalos, dogs, cats, goats, horses, mules, and several species of ducks, swans and water fowl[10].

The mechanisms of transmission are not well understood, but the pathogen is predicated to enter through traumatised epithelium. For example, in river workers, rhinosporidiosis is caused by abrasions from sand and contact with ground water, which is a suggested reservoir for *R. seeberi*. Use of contaminated stones for soaking up residual urine has been connected to urethral rhinosporidiosis in men. The spores of the fungus are also found in animal faeces and urine which can affect people who bathe in contaminated ponds[11].

The life cycle of *R. seeberi* is also not well understood; its established reservoir has not been determined, and attempts to culture it *in vitro* have been unsuccessful. Histological sections have been used to determine the developmental stages of the pathogen. Its life cycle consists of two stages; an endospore that develops into a "trophocyte," which then develops into a mature "sporangium." A mature sporangium is a thick-walled cyst that contains 12,000 spores and ranges from 60 to 450 microns in size. Fully developed sporangia are a source of spores on external surfaces while developing or immature sporangia are present deep within the spores. Once the spores are released from sporangia they can enter nearby tissue and continue their life cycle[2].

Rhinosporidiosis is a deep-seated and confined infection of mucous membranes. It clinically presents as a polypoid, soft tissue mass most commonly located in the nose, throat, eye, or ear but can also be found in the oropharynx, larynx, trachea, bronchi, and genitourinary tract[6,12]. The nose and nasopharynx are affected in 70% of cases, while ocular conjunctiva and other sites are affected in 30% of cases. It is not contagious and is not transmitted through sexual contact[2,13].

The pathogen forms round and thick-walled sporangia in the sub-mucosal layer of the affected site which appears as white dots on the superficial skin[4,6]. Osteolytic bone destruction is also an important clinical sign[5]. The disease can spread systemically to cause lesions of the skin, urethra, anus, and penis[14]. The disease is prevalent in rural areas and can affect any race[4]. Men are more commonly affected with a ratio of 4:1 compared to women[15]. The incidence of the disease is surprisingly low given that a high number of people in the endemic areas bathe in stagnant waters. Because the disease has a slow progression, lesions may have a chronic history before symptoms present[4].

Clinically, the disease can manifest as one of four forms: nasal, ocular, cutaneous, or disseminated form[2].

• Nasal form: the most common form of rhinosporidiosis; clinically presents as epistaxis with a sessile red/pink polyp in the nose that can be unilateral or bilateral.

• Ocular form: sessile growth with friable, pedunculated polyps in the eye; polyps formed are mostly flat and primarily occur on the lacrimal sac and spread downwards towards the nasal passage.

• Cutaneous form: lesion occurs as tiny papules that grow with wart-like friable crenulated surfaces; can easily turn into an ulcer and become infected.

• Disseminated form: the rarest form; manifests as spherules of *R. seeberi* in the bone, lung, liver, viscera, spleen, trunk, limbs, or brain[2].

Various case reports have been published on rhinosporidiosis including those by Hussein and Rashad[16], Ngamdu et al.[17], Sivapathasundharam et al.[18], Rajeshwari et al.[19], Burgess et al.[20], Yadav and Shrestha[21], Fouzia et al.[22], Maruf Raza et al.[23], and John et al.[24].(Table 1)[1,3,5,12,16-24] In these reports, the most commonly affected site was the nasal cavity, and additional common features include male predominance and affected individuals that are 50 to 60 years old. However, this case report is unique given the occurrence of infection on the

Table 1. Details of published cases of rhinosporidiosis

Case no.	Author	Year	Age (yr)	Sex	Site
1	Hussein and Rashad[16]	2005	25	Male	Nasal polyp
2	Morelli et al.[3]	2006	26	Male	Nasal cavity
3	Nayak et al.[12]	2007	48	Male	Multiple polyps all over body
4	Sivapathasundharam et al.[18]	2009	73	Male	Parotid gland
5	Rajeshwari et al.[19]	2010	37	Male	Throat
6	Sefu and Fauzia[1]	2011	70	Male	Nasal cavity
7	Burgess et al.[20]	2012	12	Female	Arytenoid
8	Babu et al.[5]	2012	42	Male	Nasal cavity
9	Ngamdu et al.[17]	2014	18	Male	Nasal cavity
10	Yadav and Shrestha[21]	2014	18	Male	Stensen's duct
11	Fouzia et al.[22]	2015	60	Male	Nasal cavity
12	Maruf Raza et al.[23]	2016	40	Female	Parotid gland
13	John et al.[24]	2017	44	Male	Eyelid

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palate in a female patient.

In a non-endemic area like Bareilly, India, rhinosporidiosis is infrequent and therefore may present a diagnostic challenge. However, this report demonstrates that dentists and health care professionals should be more aware of this infection. Additionally, individuals who are regularly exposed to potentially contaminated areas, such as stagnant water and soil, should follow safety precautions.

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Authors' Contributions

H.P., S.T., and M.A. contributed to concepts and design; H.P. contributed to intellectual content; M.A. and H.P. contributed to literature search; S.T. and B.S.M. contributed to data acquisition and analysis; H.P., B.S.M., M.A., and S.T. contributed to manuscript preparation; H.P., B.S.M., and S.T. contributed to manuscript editing and review.

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Consent for Publishing Photographs

Written informed consent was obtained from the patient for publication of this article and accompanying images.

Conflict of Interest

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

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