

# Rhinosporidiosis: an unusual cause of nasal masses gains prominence

S S Chao, K S Loh

## ABSTRACT

**Introduction:** Rhinosporidiosis is a rare cause of nasal masses locally, with only two cases reported over a 35-year period.

**Methods:** Four patients with rhinosporidiosis, all from the Indian subcontinent, were managed at our tertiary referral centre over a recent five-year period. They presented with nasal masses and the diagnosis was confirmed by histological examination.

**Results:** All patients were treated by local excision of the nasal masses, and two also received dapsone therapy after surgery. During follow-up, local recurrence was found in two patients, one of whom had received dapsone.

**Conclusion:** With a significant number of foreign workers from endemic regions, this uncommon disease may be observed more frequently in the future. It is thus important to consider the diagnosis of rhinosporidiosis in patients from endemic regions presenting with nasal masses. The mainstay of treatment should be wide surgical excision.

**Keywords:** dapsone therapy, nasal mass, nasal obstruction, rhinosporidiosis, surgical excision

*Singapore Med J 2004 Vol 45(5):224-226*

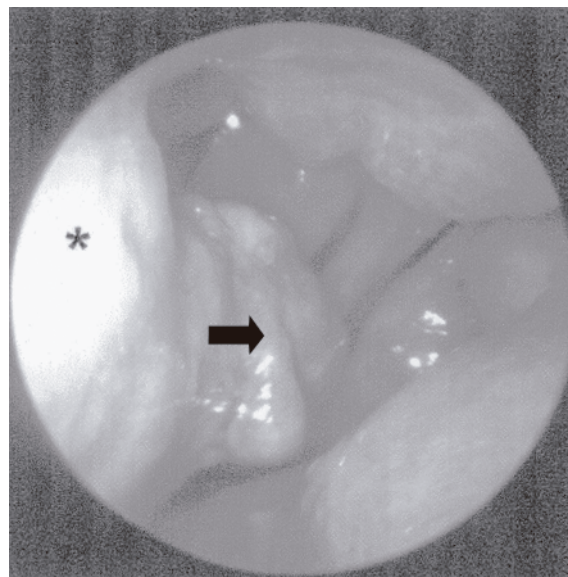
## INTRODUCTION

Rhinosporidiosis is an infectious disease endemic in southern India and Sri Lanka. The causative agent has been attributed to *Rhinosporidium seeberi*. In Singapore, three cases had been reported to date, with the first two cases in 1966 and 1986, respectively<sup>(1,2)</sup>. In 2001, we described another case with unique clinical features<sup>(3)</sup>. Over a recent 5-year period, four cases of rhinosporidiosis have been diagnosed and managed at our centre.

## CASE MATERIAL

### Case 1

A 25-year-old Bangladeshi man presented with right-sided nasal obstruction and yellowish nasal



**Fig. 1** Endoscopic photograph shows rhinosporidiosis lesions (arrow) arising from the edge of a large septal perforation (asterisk).

discharge for seven months. Examination showed a papillomatous mass attached by a stalk to the right inferior turbinate. Local excision was performed by excising the stalk from the inferior turbinate. Histological examination confirmed rhinosporidiosis. He was well until two years later when a recurrence was detected at the original site. The recurrent mass was excised together with the right inferior turbinate. He was subsequently followed-up for a year, during which no further recurrence was detected.

### Case 2

A 35-year-old Indian man from Madras presented with progressive bilateral nasal obstruction of 12 months duration. There was neither epistaxis nor rhinorrhoea, and his sense of smell was normal. He had a nasal operation in Madras five years earlier but was not told of the diagnosis. Physical examination showed erythematous papillomatous masses arising from the edges of a large septal perforation (Fig. 1). These masses were studded with white spots on the surface. Biopsy of the mass confirmed rhinosporidiosis. The masses were endoscopically resected with clear margins. He was started on dapsone 100mg daily

Department of  
Otolaryngology  
Head and  
Neck Surgery  
National University  
Hospital  
5 Lower Kent  
Ridge Road  
Singapore 119074

S S Chao, FRCS  
Associate Consultant

K S Loh, FRCS  
Consultant

Correspondence to:  
Dr Chao Siew Shuen  
Tel: (65) 6772 5370  
Fax: (65) 6775 3820  
Email: entv6@  
nus.edu.sg

post-operatively for one month, and returned to India six months later. There was no recurrence during the period of follow-up.

### Case 3

A 24-year-old Bangladeshi man working locally in the construction industry presented with a painful mass obstructing his right nasal cavity. It was associated with intermittent epistaxis. He gave a history of swimming in a lake in his hometown when he was 14 years old. Physical examination showed a 0.5cm granulomatous lesion attached to the floor of the right nasal cavity by a narrow pedicle. Excisional biopsy confirmed the diagnosis of rhinosporidiosis. A follow-up review six months after surgery showed no evidence of recurrence.

### Case 4

A 28-year-old man from southern India who had been working in the construction industry in Singapore for two years presented with nasal obstruction and throat discomfort. While in India some years ago, he had undergone nasal surgery. When he was younger, he used to swim in the rivers of his village. Physical examination revealed a mass in the left nasal cavity that extended posteriorly into the nasopharynx. A large 4cm by 8cm mass was also seen hanging freely from the nasopharynx into the oropharynx. He was able to voluntarily push the mass forwards from the oropharynx into the oral cavity. Macroscopically, the fleshy mass was studded with whitish spots on its surface.

On endoscopy, the large lesion was found to arise from a narrow stalk on the mucosa of the lateral aspect of the left inferior turbinate close to the roof of the inferior meatus. No other abnormality was seen in the nasal cavity or nasopharynx. The mass was resected endoscopically, and was confirmed on histology to be rhinosporidiosis. The patient was prescribed dapsone 100mg daily for three months after the operation. At four months post-resection, recurrent lesions were detected on the posterior wall of the nasopharynx. He returned to India shortly after the recurrence was detected, before any further treatment could be instituted.

## DISCUSSION

Rhinosporidiosis is a chronic granulomatous disease endemic in South India and Sri Lanka. Areas with the next highest prevalence are South America and Africa. Infrequently, isolated cases have been reported in other parts of the world. In Singapore, this condition had been extremely rare, with only two reported cases between 1966 and 2000. However, we have

recently managed four cases of rhinosporidiosis over a 5-year period, with three cases presenting within one year. All four patients were foreign workers who had spent most of their life in endemic areas, suggesting that these cases were imported.

The taxonomy of the causative organism is unclear, though most microbiologists had initially considered it a fungus. Fungal stains such as methanamine silver and periodic acid-Schiff could stain the wall of the organism. However, culture of *R. seeberi* had been unsuccessful in all artificial media, though it could be maintained through its life cycle in tissue cultures. Recently, Ahluwalia et al hypothesised that the causative organism was not a fungus but a prokaryotic cyanobacterium *Microcystis aeruginosa*, based on the findings that this organism was isolated from both the clinical specimens of patients and the pond water samples where they bathed<sup>(4,5)</sup>. Herr et al however, through analysis of the 18S small subunit ribosomal DNA groups, concluded that *R. seeberi* was related to a group of fish parasites referred to as the DRIP clade<sup>(6)</sup>.

The mode of transmission is also unclear but since the nose and eye are the most commonly affected sites, it has been suggested that the organism is air or water-borne. Water and soil are believed to be the reservoir of infection, given the increased incidence of disease found in sandworkers, paddy cultivators and people bathing in stagnant muddy waters. Two of our patients had a history of bathing in rivers in their hometowns years before the onset of disease, supporting the above observation.

Rhinosporidiosis primarily affects the mucus membrane of the nose and nasopharynx. The characteristic lesion is a friable, vascular polyp, which may be pedunculated or sessile. The surface is studded with tiny white dots from spores beneath the epithelium, giving it a "strawberry-like" appearance. They arise primarily from the nasal cavity and can spread backwards to the nasopharynx and oropharynx. Other areas in the head and neck region can be involved, including the conjunctiva, oral cavity, tonsils, epiglottis, larynx, trachea, and ethmoid or maxillary sinuses. Urethral, vaginal and rectal lesions have also been reported. Systemic disease is rare but can include multiple mucocutaneous, hepatic, renal, pulmonary, splenic or bone lesions, associated with fever, wasting, and even death<sup>(2)</sup>. In all our patients, the disease was localised to the nasal cavity and the main presenting symptom was nasal obstruction. Other common symptoms include epistaxis and viscid nasal discharge. As the disease runs a slow course, lesions may be present for many years before the patients become symptomatic. Indeed, the nasal mass

may be large enough to hang into the oropharynx, as in Case 4.

The gross appearance, though distinctive, is not diagnostic. Being friable with a tendency to bleed, they can be mistaken for neoplastic lesions. Another differential diagnosis to consider in our local context is tuberculous infection. Diagnosis is confirmed by histology. Round or oval cells with thick walls containing the sporangium are seen in a fibrovascular stroma infiltrated by chronic inflammatory cells. Various stages of the life-cycle can also be seen in the histological sections.

Surgical excision is the mainstay of treatment. It has been advocated that a wide surgical margin is necessary to reduce the risk of recurrence, though this may be associated with significant morbidity like haemorrhage and nasal septal perforation. Because of this, limited surgical excision with cautery of the base of the lesion has been attempted, and to further reduce the risk of recurrence, various adjuvant medical therapies, including anti-fungals such as griseofluvin and amphotericin B<sup>(7)</sup>, have been tried without much success.

The only drug appearing to have clinical promise is dapsone<sup>(8-10)</sup>. All our patients had complete excision without wide surgical margins and two of them were treated subsequently with dapsone. Recurrences were detected in two patients, one of whom had three months of dapsone therapy. This suggests that a complete excision without wide surgical margins may be inadequate. The usefulness of dapsone in our patients is unclear because of the small numbers and the inability to follow-up the

patients for a longer period of time. As such, based on our experience, future patients should be treated primarily with wide surgical excision irrespective of whether dapsone is given post-operatively.

In conclusion, in a non-endemic area like Singapore, rhinosporidiosis is uncommon and the diagnosis is usually not apparent. However, with a significant proportion of the workforce in Singapore being from the Indian subcontinent, it is likely that this condition will be observed more frequently in future. It is thus prudent to keep this condition in mind when managing patients from endemic areas with nasal masses.

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