Severe Epistaxis from Rhinosporidiosis : A Case Report

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Abstract

Rhinosporidiosis is quite rare in Thailand, but the actual incidence may be higher than the numbers reflect. This is thought to be due to misdiagnosis by physicians who are unfamiliar with this disease entity or physicians who found a case but did not describe its presence. The authors report a case in a 17 year old male with the lesion involving both nasal cavities, nasopharynx and left maxillary sinus presenting with severe epistaxis, which was treated by complete surgical removal with electric cauterization of its base. We warn physicians who face this problem to be aware of this disease entity.

The sources of epistaxis may locate in the nasal cavities or surrounding structures such as paranasal sinuses, nasopharynx and/or the middle ear cavity. The etiologies include trauma, infection, tumor, hypertension, bleeding disorders and other systemic diseases. The teenaged-male patient presenting with nasal masses and severe epistaxis is likely to have nasopharyngeal angiofibroma. But contact bleeding nasal masses in some patients may result from rhinosporidiosis that bring the patients to the ENT clinic with severe epistaxis. The disease is quite rare in Thailand. There were 5 Thai patients in previous 3 reports by Chalermkumpeewej (1939)[1], Cutchavaree (1980)[2] and Pumhirun (1983)[3]. Since then there have been no additional reports. The authors present a Thai patient with severe epistaxis and the disease was proved to be rhinosporidiosis.

CASE REPORT

A 17-year-old Thai male patient living in Utumpornpisai, Srisakes, the northeast province, was referred to the ENT clinic from Srisakes Hospital on February 4, 1995 because of severe bleeding from his right nasal cavity for 3 days. He had some degree of nasal obstruction and watery dis-

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charge, often blood tinged in the right nasal cavity for 6 months. The bleeding was not controlled by conservative measures in the local hospital. Combined anterior and posterior nasal packings were left in place during referral. Other medical history was unremarkable. He had never been abroad. He had habitually dived and swum in stagnant water since childhood. There was no history of tuberculosis among the members of the family.

On examination, a polypoid reddish mass was found in the nasopharynx extending into both posterior choanae (Fig. 1). The mass was soft, friable, irregular surface and contact bleeding. Rigid endoscope revealed an extensive mass involving the left maxillary sinus, floor of the left nasal cavity, nasopharynx, posterior aspect of the right inferior turbinate and the right nasal cavity. Right maxillary sinus appeared normal. Biopsies were undertaken from the left maxillary sinus, nasal cavities and nasopharynx.

The pathological reports of all specimens were consistent with rhinosporidiosis (Fig. 2). The mass was completely removed together with Rt. partial inferior turbinectomy and left partial maxillectomy. The base of the lesion in the nasopharynx was electrically cauterized. The recovery was uneventful, without any antifungal administration.

The patient was regularly followed-up in the ENT clinic up to 1 year without any evidence of recurrence.

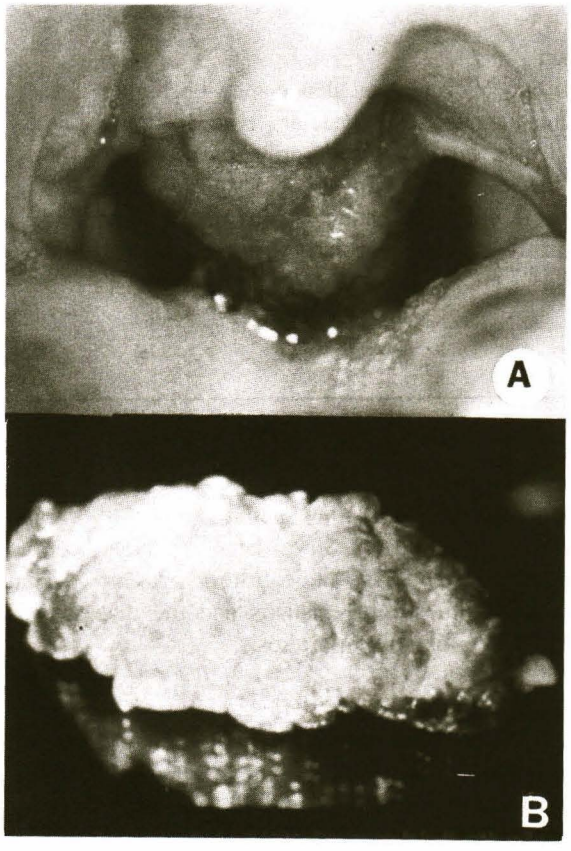

Fig. 1. **A. The red, strawberry like, studded with white flecks, nasopharyngeal mass extending beyond the uvula.**
B. The specimen from surgical removal.

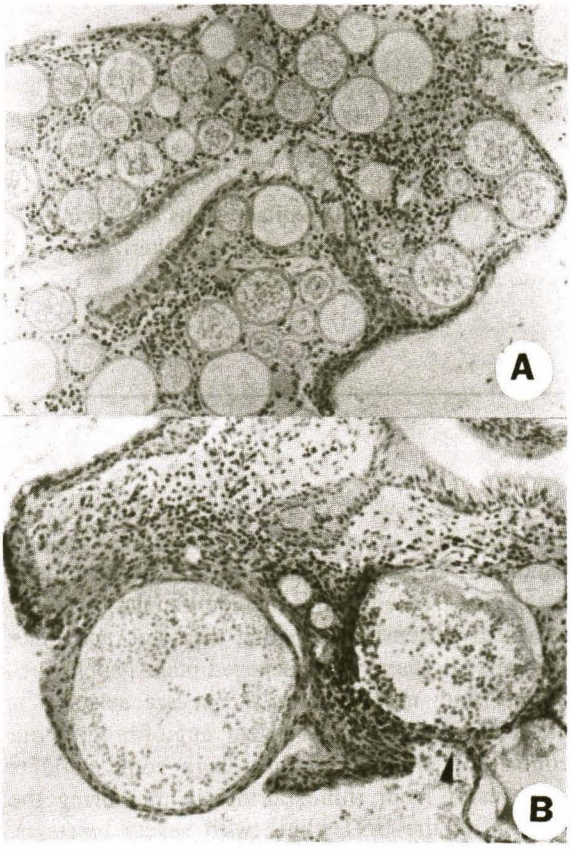

Fig. 2. **A. Multiple sporangia in varying stages.**
B. Discharge of spores from the mature sporangium through a weak spot (arrow head).

DISCUSSION

Rhinosporidiosis is a chronic infectious disease caused by *Rhinosporidium seeberi*, a fungus from the class of *Phycomycetes*, family *Coccidiodaceae*. The disease was first reported in 1900 by Seeber[4]. Initially it was considered to be a protozoa. In 1923 Ashworth & Logan demonstrated that it was a fungus[5]. For years, attempts to culture the organism were unsuccessful, but in 1989, a group at Trivandrum Medical College reported some success[6].

The organism is dimorphic consisting of two phases, the saprophytic in the soil, and the parasitic phase in the tissue of animals such as cattle, horses, mules, dogs, flocks and humans[2]. Trauma to the nasal mucous membrane is an essential predisposing factor. Impairment of ciliary action, irritation and stagnation of secretion from anatomic obstruction may also play a role[2].

Although a definite course of transmission has yet to be demonstrated, many authors agree that water is a necessary medium of transmission[7,8]. Ocular infection is airborne[9]. Transmission of rhinosporidiosis from animals to humans, which is theoretically possible, has yet to be demonstrated[7]. Attempts to reproduce the chronic infection in animals have failed also[10].

The infective stage of the fungus is the functional spore. The spore after implantation on the traumatized mucocutaneous tissue enters the deep tissue and leads a parasitic life. The nucleus undergoes a series of mitotic divisions to 128 nuclei. The envelope of the trophocyte is thickened by a deposit of cellulose. At the end of this process the sporangium reaches a diameter of about 200-250 microns in diameter containing more than 16,000 young spores. A weak spot in the membrane called "pore" serves for the rupture and discharge of spores from the mature sporangium. Enormous numbers of spores escape into the nasal secretions and produce a granulomatous reaction[7,10,11].

The histological appearance reveal infiltration with mostly lymphocytes around the spores. The sporangia and spores can be found in subepithelial areas and in the epithelial lining. The stroma is infiltrated with lymphocytes, plasma cells, polymorphs and eosinophils. Giant cells of the foreign body type appear around sporangia. Neovascularization can be found as well[7,10,11].

Rhinosporidosis usually affects the nasal mucous membrane and appears as a pink or red papillomatous growth which is soft, friable, pedunculated or sessile, very vascular and bleeds on manipulation. It may resemble a strawberry or ripe raspberry, as the surface is studded with white flecks, which are the mature sporangia[2,7].

Most of the cases are farmers, low economical groups, especially those living in crowded and unsanitary conditions[10]. A high incidence has also been observed in those who dive and swim in stagnant water or ponds[2,9]. The lack of familial incidence rules out interhuman transmission[9].

Nasal obstruction and epistaxis are the most common presenting symptoms[2,7,9]. The bleeding is due to great vascularity of the mass[10]. Watery discharge, often tinged with blood, is the third symptom and may become mucopurulent with subsequent infection. Other symptoms include sneezing, coryza, a sensation of the presence of a foreign body and pain in the nose. Most cases are unilateral lesions but the patient in this report had bilateral nasal lesions and unilateral maxillary sinus lesion simultaneously. Diagnosis is based on clinical evaluation and pathological examination. Recently, Jaiswal recommended the cytologic smear as a routine preliminary screening technique in the diagnosis of suspected cases. A smear demonstrates the typical spores both in isolated cases and in groups. This technique has many advantages in being simple, easy to perform, cost effective, less time consuming and does not need technical expertise[12].

The most successful treatment, with the lowest incidence of recurrence, is surgical extirpation and cauterization of the base of the lesions. Satyanarayana reported a recurrence rate of 11 per cent after surgical treatment[8]. Khan followed with 22 patients of whom 18 were treated by cutting diathermy. The latter had no recurrence, whereas, in the 4 cases in which the lesions were removed with forceps and snares all had recurrent disease[10]. Our patient was treated by extensive surgical removal of the lesion, and cauterization of its base. Without any additional treatment, the patient was free of disease at 1 year-follow-up.

Rhinosporidiosis is generally considered to be insensitive to systemic antimycotics. The use of intravenous amphotericin B is advised when surgical removal is incomplete[13]. Nair applied

diaminophenylsulfone to 32 patients with nasopharyngeal rhinosporidiosis, 71.4 per cent of the patients did not show recurrence in a three-year period and none of them needed additional surgery during that period[14]. The prognosis of rhinosporidiosis is usually good, and is rarely fatal. Hemorrhage and generalized dissemination may however lead to death[7].

In Thailand, all 5 patients reported in 1939, 1980 and 1983 were farmers. Each patient lived in a different province, namely Pathum Thani, Chon Buri and Bangkok including Srisakes for the patient in this report. The endemic area in Thailand still cannot be concluded.

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รายงานผู้ป่วยเลือดกำเดาไหลรุนแรงจากภาวะติดเชื้อไรโนสปอริเดียม

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รายงานผู้ป่วยชายไทย อายุ 17 ปี เป็นโรคติดเชื้อไรโนสปอริเดียม ของโพรงจมูกทั้งสองข้าง และ maxillary sinus ข้างซ้าย ซึ่งมาโรงพยาบาลจุฬาลงกรณ์ด้วยอาการเลือดกำเดาไหลรุนแรง ผู้ป่วยได้รับการรักษาโดยการผ่าตัดเอาก้อนเนื้อออกทั้งหมด เมื่อติดตามผลการรักษาเป็นระยะเวลา 1 ปี ไม่ปรากฏโรคขึ้นมาอีก ผู้ป่วยรายนี้นับเป็นผู้ป่วยชาวไทย รายที่ 6 ที่บันทึกไว้ในประเทศไทย และเป็นผู้ป่วยรายแรกที่มีการติดเชื้อภายในโพรง maxillary sinus ด้วย

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