Disseminated rhinosporidiosis destroying the talus: a case report

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ABSTRACT

Rhinosporidiosis is a chronic granulomatous fungal disease caused by Rhinosporidiosis seeberi. It usually affects the mucocutaneous tissue of the nose; bone involvement is rare. We report the clinical features, diagnosis, and management of rhinosporidiosis involving the face, forehead, and right foot. As antimicrobial therapy was ineffective, a below-knee amputation was performed.

Key words: rhinosporidiosis; talus

INTRODUCTION

Rhinosporidiosis is a chronic granulomatous fungal disease caused by Rhinosporidiosis seeberi. It is endemic in India and Sri Lanka, and may occur in the Americas, Europe, and Africa. It is more common in men and is transmitted by direct contact with spores through dust, infected clothing or fingers, and swimming in stagnant water. It usually affects the mucocutaneous tissue of the nose; bone involvement is rare. We report the clinical features, diagnosis, and management of rhinosporidiosis involving the face, forehead, and right foot.

CASE REPORT

In September 2006, a 53-year-old man presented with a one-year history of pain in his right ankle and multiple progressive painless warts on the upper lip and forehead. 10 years earlier, a swelling in his chin secondary to a fungal infection had been excised.

The patient had a painful gait and circumferential swelling around his right ankle with no scars or tracking sinuses. The swelling was warm and there was synovial hypertrophy. All movements of the ankle and subtalar joints were painfully restricted. He had 2 warty, pedunculated swellings of the forehead and upper lip (Fig. 1), which were not bleeding or friable. Examination of the nose was normal.
incised through an anterolateral approach. The whole right talus was found to be soft, friable, and collapsed, leaving no bone stock for tibiotalar arthrodesis. As the aetiology was in question, ankle arthrodesis was deferred until the diagnosis was confirmed.

Histopathologically, the chronically inflamed skin showed many sporangia of varying sizes filled with spores. The epidermis covering the lesion was hyperplastic (Fig. 4a). The talus showed trabeculae of cancellous bone with a lesion composed of closely arranged small to large sporangia with intact or ruptured walls. These contained spores of varying sizes (Fig. 4b). The intervening stroma contained moderate infiltrates of lymphocytes, plasma cells, histiocytes, and neutrophils. The diagnosis of rhinosporidiosis was established.

Attempts at limb salvage were abandoned in view of the likelihood of persistent infection, poor function, and the need for prolonged treatment. The option of a below-knee amputation was suggested.

Radiograph of the ankle revealed a grossly reduced joint space (Fig. 2). There were lytic areas in the talus with some new bone formation and areas of osteolysis in the lower end of tibia, fibula, and parts of the calcaneus. After curettage of the talus, a computed tomographic scan showed involvement of the calcaneus, tarsals, and the bases of the first and second metatarsals (Fig. 3).

The white cell count was 6700/mm$^3$; the differential count was 64% neutrophils, 6% eosinophils, 24% lymphocytes, and 6% monocytes. The erythrocyte sedimentation rate was 45 mm/hour. He was human immunodeficiency virus-negative by enzyme-linked immunosorbert assay.

An initial diagnosis of infective arthritis of tuberculous origin was made. An open biopsy and ankle arthrodesis were planned. The right ankle was

Figure 1  A sessile swelling of the upper lip.

Figure 2  Plain radiographs of the right ankle showing grossly reduced joint spaces, and lytic areas in the talus and calcaneus with new bone formation.

Figure 3  Computed tomography after initial curettage of the talus demonstrates involvement of the calcaneus, tarsals, and metatarsals.

Figure 4  Photomicrographs of (a) the skin and (b) bone showing sporangia of various shapes and sizes representing different stages of maturation (H&E, x100).
and the patient agreed. The lesions on the upper lip and forehead were excised at the same time.

**DISCUSSION**

*R. seeberi* is a fungus of the sporozoan classification. Since the first report in 1900, more than 200 cases have been reported, of which more than 80% were from India and Sri Lanka. Over 70% of the cases were in the nasal mucosa and about 15% were in the eye. Involvement of skin, bone, and other visceral is rare.

*R. seeberi* cannot be isolated in synthetic media, though it grows well in cell culture. Its diagnosis depends on the recognition of its distinctive morphology: numerous characteristic sporangia and spores in different stages of maturation. The differential diagnosis includes giant cell tumour, chondrosarcoma, and coccidioidomycosis, but the sporangia of *R. seeberi* should not be confused with the spherules of *Coccidioides immitis*. The sporangium of *R. seeberi* is larger and has thicker walls. The size and number of endospores on sporangia of *R. seeberi* is greater than those of *C. immitis*.

Rhinosporidiosis is primarily an infection of the nose. Occasional involvement of lips, palate, epiglottis, pharynx, larynx, trachea, bronchus, ears, conjunctivae, lacrimal sac, skin, vulva, vagina, penis, and scalp have also been reported. Osteolytic lesions involving the tibia, lateral condyle femur, hand and feet, and calcaneus have also been reported (Table).

Dissemination to anatomically unrelated sites is mainly attributed to haematogenous spread. However, some consider haematogenous and lymphatic spread unlikely and propose direct implantation. Scratching of the skin contaminated with nasal secretions may implant the spores and cause secondary lesions.

Dapsone is the recommended antimicrobial agent to prevent recurrence, though it may be ineffective. Therefore, in this case, a below-knee amputation was performed instead of limb salvage.

**REFERENCES**