Rhinosporidiosis of the left wrist joint: a case report

Surojit Mondal,1 Aniket Chowdhury2
1 Department of Orthopaedics, BS Medical College, West Bengal, India
2 Department of Community Medicine, BS Medical College, West Bengal, India

Address correspondence and reprint requests to: Dr Surojit Mondal, Department of Orthopaedics, BS Medical College, West Bengal, India. Email: sm3036@gmail.com

ABSTRACT
Rhinosporidiosis is a chronic granulomatous disease caused by *Rhinosporidium seeberi*. It usually occurs in the mucous membranes of nose, nasopharynx, and eyes, and less commonly in extra nasal sites such as skin, bones, genitalia, and even the internal organs. Rhinosporidiosis occurs in the wrist joint with isolated bony involvement is rare. We report one such case in a 50-year-old man who presented with a non-tender, fixed swelling over his anterolateral aspect of left forearm. Radiography and computed tomography showed a lytic destructive lesion involving the distal radius, ulna, carpals, and base of metacarpals. Biopsy revealed features of rhinosporidiosis. The patient underwent below-elbow amputation.

Key words: rhinosporidiosis; *Rhinosporidium*

INTRODUCTION
Rhinosporidiosis is a chronic granulomatous infection caused by *Rhinosporidium seeberi*. It usually occurs in young adults in the mucous membrane of nose, nasopharynx, and eyes, and less commonly in extra nasal sites such as skin, bones, genitalia, and even internal organs. Rhinosporidiosis occurs in the wrist joint with isolated bony involvement is rare.

CASE REPORT
In March 2012, a 50-year-old man presented with a 5-month history of an ulcerating and fungating mass in the anterolateral aspect of the left forearm (Fig. 1). There was serosanguinous foul smelling discharge from the swelling, which bled on touch. The mass was 9x7x5 cm in dimension, firm, friable, and multilobulated, with a delineated margin. There was no pain over the swelling, and the patient had no fever. The nose and oral cavity did not reveal any lesions. The serological test for human immunodeficiency virus was negative.

Radiography and computed tomography revealed an expansile, lytic, destructive lesion involving the anterolateral end of the left distal radius, distal ulna,
all carpals, and bases of the metacarpals, as well as a soft-tissue mass with calcification within (Fig. 2). There was no periosteal reaction or area of sclerosis. The mass mimicked a bone tumour, but biopsy revealed features of rhinosporidiosis.

The patient underwent below-elbow amputation (Fig. 3). The excised mass was pink, with a fleshy consistency and was studded with whitish spots on its surface (Fig. 4). Histological and microscopic examination of the mass using H&E, PAS, PAP,

Figure 1  A fungating and ulcerating mass over the anterolateral aspect of the left forearm.

Figure 2  Three-dimensional computed tomography shows a lytic destructive lesion involving the anterolateral end of the left distal radius, distal ulna, all carpals, and bases of metacarpals.

Figure 3  The excised left forearm after below-elbow amputation.

Figure 4  A 9x7x5 cm, firm, fixed, and friable mass originating from the distal end of the left radius.
MGG, and mucicarmine stains showed features of rhinosporidiosis. The stroma contained vascular fibroconnective tissues with fibroblasts and myofibroblasts and an inflammatory infiltrate (neutrophil, granulocytes, lymphocytes, plasma cells, and histiocytes). The stroma contained many globular cysts of varying size containing innumerable spores, which was also noted within the marrow space of the radius. Each of these cysts represented a thick-walled sporangium containing numerous ‘daughter spores’ in different stages of development (Fig. 5).

**Morphological criteria** were based on the diameter of the endospores (5–10 μm) and sporangia (50–1000 μm) to distinguish *R. seeberi* from *Coccidioides immitis*, another common nasal mycosis fungus.

**DISCUSSION**

*R. seeberi* was initially considered a fungus of the sporozoan classification. However, electron microscopic, histopathological, and molecular studies indicate that it is a eukaryote pathogen rather than a fungus. More than 80% of such cases have been reported from India and Sri Lanka. Over 70% occurred in the nasal mucosa, and about 15% in the eye. *R. seeberi* cannot be isolated in synthetic media, although it grows well in cell culture. Its diagnosis depends on the recognition of sporangia and spores in different stages of maturation. Its differential diagnosis includes plasmocytoma, metastasis, and coccidiomycosis. *R. seeberi* should be distinguished from *C. immitis*. In cases where occasional spherules are seen in the sputum, cytological differentiation from *C. immitis* may be required. *C. immitis* has similar mature stages represented by large, thick-walled, spherical structures containing endospores, but its spherules are smaller (diameter of 20–80 μm) and contain smaller endospores (diameter of 2–4 μm), compared to those of *R. seeberi*. Moreover, *C. immitis* does not stain with mucicarmine.

Rhinosporidiosis is associated with rural and aquatic environments and is transmitted by direct contact with spores through dust, infected clothing or fingers, swimming in stagnant water, trauma, followed by autoinoculation. Haematogenous dissemination was reported to result in painless, firm-to-hard, subcutaneous nodules that remain unattached to skin. Visceral involvement, particularly of the liver, lung, and brain has been reported in disseminated disease. Rhinosporidiosis involving the bone is rare and can manifest in 3 ways: (1) local invasion of skull by nasal rhinosporidiosis, (2) as a part of disseminated rhinosporidiosis, and (3) primary manifestation, without any evidence of lesions elsewhere.

Dapsone is the only drug that is regarded as useful for treating rhinosporidiosis. Nonetheless, in cases of isolated bone involvement, excision with clear margins is the mainstay of treatment.

**DISCLOSURE**

No conflicts of interest were declared by the authors.

**REFERENCES**