

## Case Report

# Disseminated rhinosporidiosis following spontaneous regression of the possible primary lesion

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## Introduction

Rhinosporidiosis is a chronic disease with a tendency to recur and occasional dissemination, caused by the organism *Rhinosporidium seeberi*. The taxonomy of this organism is controversial and is still evolving. *R. seeberi* is currently identified as a protistan parasite (broad group of eukaryotic unicellular organisms) and has been included in a novel class named *Mesomycetozoea* (1). The first case of rhinosporidiosis was reported from Argentina by Guillermo Seeber in 1900 and since then the disease has been reported from 70 countries around the world (1). This disease is highly endemic in India and Sri Lanka, particularly South India and the dry zone of Sri Lanka (1). In addition to humans, rhinosporidiosis has also been reported in domestic and farm animals (1). The natural habitat of *R. seeberi* is believed to be natural stagnant ground water and presumed mode of infection is through the traumatised epithelium ("transepithelial infection"). The most commonly affected site is nasal mucosa and nasopharynx (70%) followed by conjunctiva and lacrimal sac (15%); however, other sites such as lips, oral cavity, upper respiratory tract, perineum, rectum and skin are also occasionally affected (1).

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Disseminated rhinosporidiosis is rare and only a few cases have been reported to date. We report a case of disseminated rhinosporidiosis in the skin and subcutaneous tissue in a middle aged male.

## Case report

### Clinical history

A 48-year-old male patient presented with multiple pink and skin coloured polypoidal nodules over the scalp, trunk and limbs of 3 years duration (figure 1A). About 2 months back he had also noticed diffuse swellings of both thighs (figure 1B). Inquiry into the past medical history revealed a polyp in the nasal mucosa which had healed spontaneously several years back. Apart from this he did not have any significant illness. He made his living as an unskilled manual labourer and lived in a rural village in the district of Anuradhapura, Sri Lanka. For washing and bathing purposes he used a nearby natural water way or a river.

### Pathological features

Excision biopsies of scalp and trunk nodules and tru cut biopsies of the thigh swelling were performed. Histology of all biopsies revealed numerous sporangia of *Rhinosporidium*, in different stages of development, containing endospores. Some degenerated empty sporangia were also present. The dermis contained a mixed inflammatory infiltrate, whereas the inflammation was minimal in the

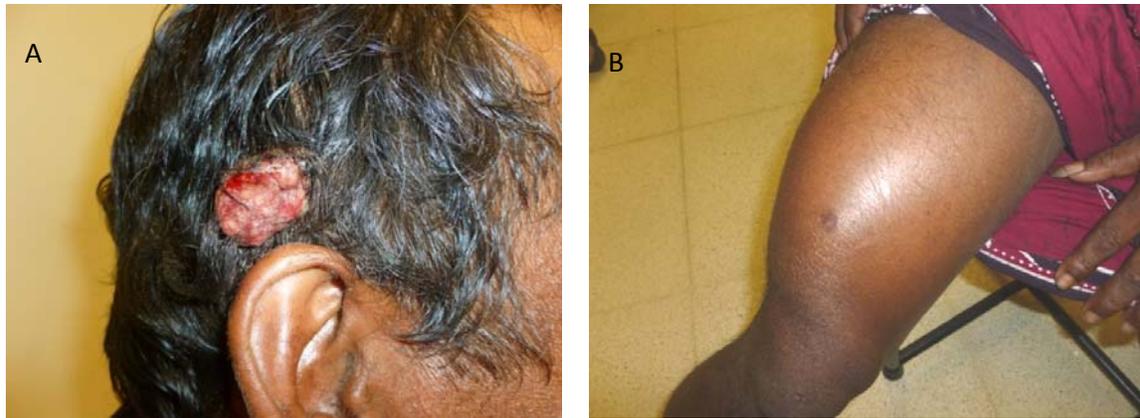


Figure 1. Skin and subcutaneous lesions. A) Rhinosporidiosis in the scalp skin forming a polypoidal lesion. Note the nodular surface and reddish fleshy consistency. B) Rhinosporidiosis involving the subcutaneous tissue of the thigh presenting as a diffuse swelling. Note that the skin is intact.

subcutaneous lesion of the thigh (figures 2A-C). Accordingly a diagnosis of disseminated cutaneous and subcutaneous rhinosporidiosis was made.

Following the histological diagnosis patient was investigated to exclude immunosuppression. He was found to be immunocompetent and free of diabetes mellitus.

### Discussion

Rhinosporidiosis is a chronic infection caused by *Rhinosporidium seeberi*. The disease is highly endemic in the Dry zone of Sri Lanka and Southern India (1). The infection is common in adult males and most patients give a history of exposure to stagnant water sources. In Sri Lanka, higher incidence of rhinosporidiosis have been reported among paddy cultivators and river sand workers whose occupation is related to natural aquatic environments (1,2). In the present case, patient is from a rural area in the district of Anuradhapura, which is a highly endemic region for rhinosporidiosis. He may have acquired the initial infection from the natural water sources he uses for daily washing purposes or cultivation work he engaged in as a hired labourer.

Rhinosporidiosis typically produces a localised infection in mucosal tissue presenting as a mass or a polyp, commonly in the nose; primary skin infection is rare. Therefore, the initial infection in this patient may have been the nasal polyp which had regressed spontaneously several years back. Occasional cases of spontaneous regression of the nasal rhinosporidiosis have been reported (1). Furthermore, one case of disseminated cutaneous rhinosporidiosis following successful surgical excision of the initial nasal infection has also been reported (3).

There is only limited knowledge on the host immune response to the rhinosporidial infection and the mode of distant spread in cases of disseminated disease. Local spread is believed to be due to auto-innoculation of endospores and disseminated rhinosporidiosis is believed to be due to haematogenous spread. Lymphatic spread of the disease is thought to be rare due to rarity of presence of lymphadenitis even in the disseminated disease (1). Therefore, it is possible that this patient had the disseminated disease in the skin and subcutaneous tissue due to haematogenous spread following initial nasal infection which had regressed spontaneously. Favouring the haematological spread theory, the thigh lesions did not have any skin ulcerations and the disease was largely confined to the subcutaneous tissue.

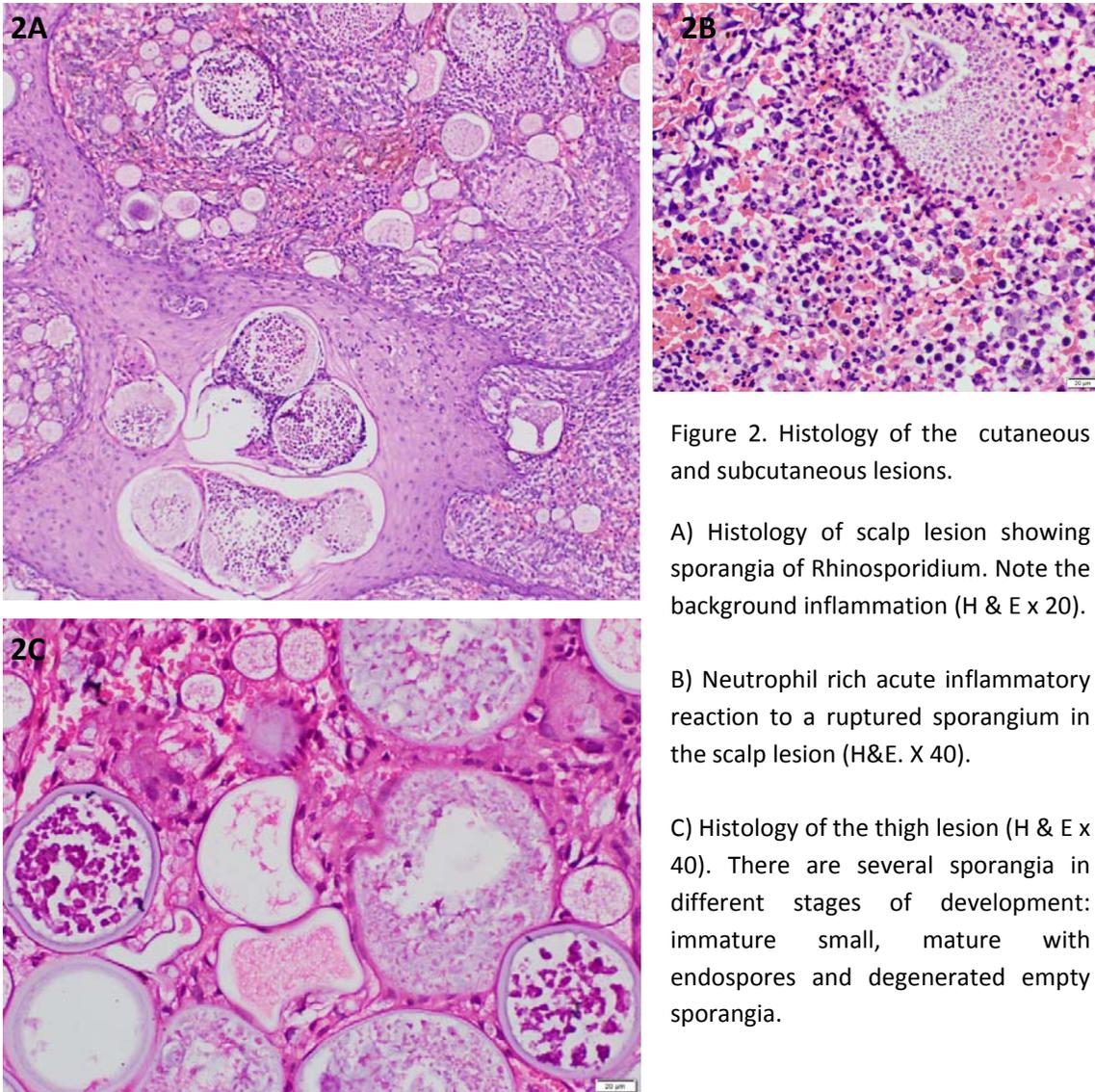


Figure 2. Histology of the cutaneous and subcutaneous lesions.

A) Histology of scalp lesion showing sporangia of *Rhinosporidium*. Note the background inflammation (H & E x 20).

B) Neutrophil rich acute inflammatory reaction to a ruptured sporangium in the scalp lesion (H&E. X 40).

C) Histology of the thigh lesion (H & E x 40). There are several sporangia in different stages of development: immature small, mature with endospores and degenerated empty sporangia.

Disseminated rhinosporidiosis is rare; when present most cases tend to spread to cutaneous and subcutaneous tissue and visceral dissemination is extremely rare (3-8). In this patient the disease was confined to the skin and subcutaneous tissue and there was no evidence of visceral involvement. There is no knowledge on the host and organism factors which predispose the dissemination of the disease. Following the diagnosis of disseminated rhinosporidiosis he was investigated to exclude an underlying immunodeficiency and was found to be immunocompetent. Disseminated disease has been reported both among

immunocompetent and immunocompromised hosts (4 – 9).

The diagnosis of rhinosporidiosis is by histopathological analysis of tissue by demonstration of rhinosporidial bodies. A detail description of histopathological features of rhinosporidiosis has been done by Karunaratne (1,10). The characteristic pathological features include presence of sporangia with endospores in varying stages of development. Some sporangia may be degenerated and devoid of endospores. Typically sporangia have a thick bilamellar wall (Figure 2). Endospores demonstrate deep

magenta staining with Periodic acid-Schiff stain, a feature which may be useful when analysing cytology smears. The stroma can contain a variable inflammatory reaction even in the same patient. The predominant inflammatory cells present are lymphocytes and histiocytes and free endospores may be surrounded by neutrophils (Figure 2C). A foreign body type giant cell reaction may be seen within sporangia in some cases (1). In the present case subcutaneous lesions in the thigh had minimal inflammatory infiltrate, whereas the scalp lesion had lymphocyte predominant mixed dermal inflammatory infiltrate (Figure 2A and B).

Surgical removal and electrocauterization of the lesions are the treatments of choice. The only drug that is found to have an anti rhinosporidial effect is Dapsone which may arrest the maturation of sporangia and accelerate degenerative changes. Nodular lesions in the head and upper trunk in this patient were surgically removed and excision sites were electrocauterized and Dapsone therapy was commenced.

In summary, this is a rare presentation of rhinosporidiosis with cutaneous and subcutaneous dissemination. The primary focus of infection could have been the nasal polyp which had spontaneously regressed some years back. Spontaneous regression and dissemination following such regression are extremely rare in rhinosporidiosis.

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