

Sporotrichosis of Maxillary Sinus

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ABSTRACT

Sporotrichosis or Gardener's disease is a chronic nodular subcutaneous mycotic disease, causative organism being *Sporotrichum schenckii*. The specific mode of entry of the fungus into the body, being mainly through small cuts and punctures and rarely by inhalation; which usually manifest as cutaneous and subcutaneous infections.

Here we report a rare case of sporotrichosis of maxillary sinus in an immunocompetent patient. Our case of sporotrichosis, a deep fungal infection involving maxillary sinus was confirmed by fungal culture and successfully managed by antifungal agents combined with surgical debridement.

Keywords: Sporotrichosis, Gardener's disease, Invasive fungal sinusitis.

INTRODUCTION

During the last two decades, the incidence of fungal infections has increased dramatically. The types of fungal diseases occurring in any particular patient depend upon the complex interplay of host immune system and local tissue environment.¹ Sporotrichosis is a chronic nodular subcutaneous mycotic disease with a worldwide distribution. The causative agent is *Sporotrichum schenckii*. The fungus gains access to the subcutaneous tissue via traumatic lesion, and proliferation of the fungus leads to the appearance of a nodule or small ulcer as soon as 5 days or as long as 6 months after inoculation. The regional lymph nodes become infected, and the spread of the fungus can be recognized by inflammation of the lymph vessels draining the initial lesion.⁶ On rare occasions, the nose and sinuses may be involved with the extension into orbit. This article reports a rare case of sporotrichosis involving the maxillary sinus in a farmer.

CASE HISTORY

A 66-year-old female patient, farmer by occupation, had reported to us with a complaint of swelling and pain in the right middle third of the face of duration of 3 months. Patient was giving a history of trauma to the same region, 6 months back. The swelling was gradual in onset and was diffuse in the first month for which she was on medication and found no relief. After one month, patient noticed multiple cutaneous nodules over the diffuse swelling with occasional fluid discharge and pain.

Her past medical and family history was noncontributory. On general physical examination no abnormality was detected.

Extraoral examination revealed a diffuse swelling of right middle-third of the face, superiorly it extended 1 cm behind the lateral canthus of eye with puffing of the lower eyelid. Inferiorly, swelling was 0.3 cm below the ala tragus line.

Anteriorly, up to the midline with deviation of nasal septum and posteriorly extended to the zygomatic region. The swelling was firm to hard in consistency and nontender on palpation. There were multiple cutaneous nodules and fluid discharge was noted from one of the nodules (Fig. 1).

Submandibular, submental, cervical lymph nodes on the right side were enlarged, mobile, firm in consistency, and was nontender on palpation.

Intraorally, a firm mass was palpable in the buccal vestibule extending around 3 mm below, in the right buccal mucosa, which was nontender (Fig. 2). All the teeth in first quadrant were tender on percussion.

Based on the clinical findings of a diffuse soft tissue swelling in the region of maxillary sinus with multiple cutaneous nodules and fluid discharge, the case was provisionally diagnosed as deep fungal infection of right maxillary sinus. Differential diagnosis of subcutaneous fungal infection, cervicofacial actinomycosis, malignancy of maxillary sinus, foreign body granuloma and lupus vulgaris were also considered.

Aspiration and biopsy of the cutaneous nodule were carried out. Hematological examination revealed normal parameters except for the ESR which was 120 mm/hr. HIV and HBV statuses were negative.

Radiological Investigation

PNS view revealed a radiopaque area overlapping over the middle-third of the face on the right side. A well-defined radiopaque mass was also noted in the sinus, which was extending from the floor to the superior-third of the sinus (Fig. 3).

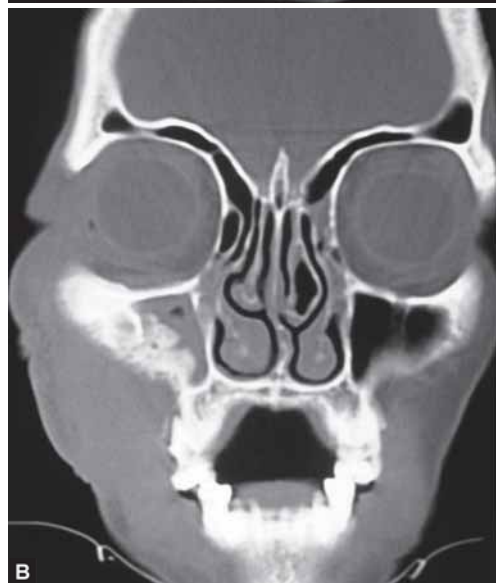
Chest X-ray did not reveal any foci of infection. CT of PNS showed irregularity and thickening of anterior wall of the right maxillary sinus, along with subcutaneous tissue thickening outside the sinus. A dome-shaped mass completely filling the sinus in the anterior wall which was reducing in size in the more posterior sections (Figs 4A and B).



Fig. 1: Clinical photograph showing diffuse swelling in the right middle-third of the face with multiple cutaneous nodules



Fig. 2: A mass in the buccal mucosal region on right side



Figs 4A and B: CT of PNS showing irregularity and thickening of anterior wall of the right maxillary sinus, along with subcutaneous tissue thickening outside the sinus. A dome-shaped mass completely filling the sinus



Fig. 3: PNS view showing diffuse radiopaque area in the right middle-third with a well-defined dome-shaped radiopacity in the right maxillary sinus

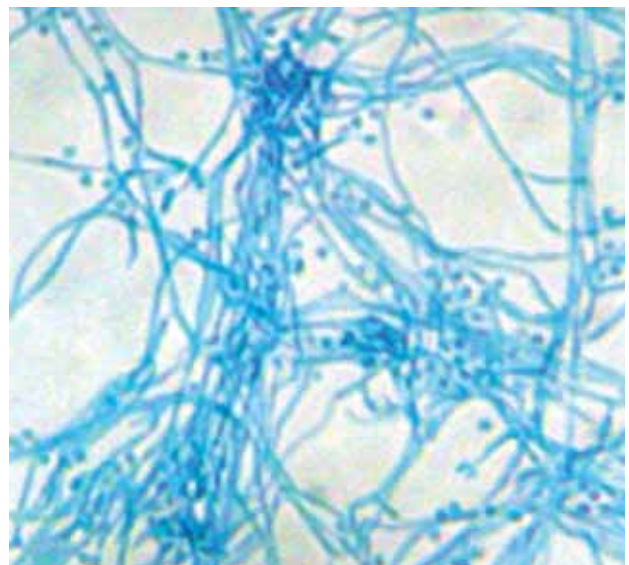


Fig. 5: Photomicrograph showing thin long fungal hyphae



Fig. 6: Resolution of the swelling and multiple lesions on the middle-third of the face

Radiological impression after both conventional and advanced imaging modality was that of invasive fungal sinusitis, with other possibilities of chronic osteomyelitis, malignancy of bone, subcutaneous lesion with underlying bone involvement.

Cytology report revealed the presence of tiny fungal yeast. PAS staining of the biopsy specimen revealed features of granulomatous inflammation and the presence of fungus. Fungal culture showed the presence of thin, long hyphae, which was confirmed to be *Sporothrix schenckii* (Fig. 5).

Treatment given to the patient was itraconazole 200 mg twice daily and surgical debridement of maxillary sinus. Patient showed drastic improvement in her condition on oral antifungal therapy with resolution of the skin lesion. Patient had undergone surgical debridement of the maxillary sinus and her condition has improved with it (Fig. 6). The oral antifungal agents were continued for three more months.

DISCUSSION

Sporotrichosis, first described by Schenck in 1898, is a fungal infection caused by *Sporothrix schenckii* which is naturally found in soil, hay, decaying vegetations, rosebushes, etc. It is thought to be a sporadic infection and enters the body by a local injury or by inhalation. It can present as cutaneous, lymphocutaneous, osteoarticular, pulmonary and disseminated forms, usual mode being cutaneous inoculation.^{5,6}

Cutaneous variety remains localized to the skin only or to the skin and subcutaneous tissues with 5 days to 6 months of incubation period.⁴ Osteoarticular sporotrichosis can involve single or multiple joints. They become infected secondary to hematogenous spread or through local inoculation. Pulmonary type acquired by inhalation, a rare manifestation, present as

chronic cavitary fibronodular disease. Disseminated infection is unusual, most likely to occur in immunosuppressed patients.⁶

Advanced imaging has an important role to evaluate invasive fungal sinusitis in support of the clinical suspicion of invasive disease. Periantral soft tissue infiltration of the fat planes surrounding the maxillary sinus in CT may represent the earliest imaging manifestation of an aggressive process.^{2,3} Bone destruction has been a prime determinant of aggressive disease, and information about bone involvement as well as involvement of contiguous structures, such as the orbit and brain, is useful in surgical planning. But fortunately, in our case being a chronic granulomatous variety, lesion was not extended to vital structures.

Reported cases of fungal paranasal sinus diseases are diverse over the past decades. In our case, patient was immunocompetent and presented with granulomatous invasive fungal sinusitis which was proved to be sporotrichosis; which usually presents as cutaneous and subcutaneous fungal infection.

In India, there are seldom case reports of sporotrichosis involving maxillary sinus. Most commonly as it enters the body through the local cuts or punctures and due to its less aggressive nature, most of the cases reported are of cutaneous and subcutaneous infections. In this case, the inoculation would have occurred either by a local route as the patient was giving a history of trauma to the same region on the face or by inhalation. In this case, prognosis is good as the vital structures are spared and the organism being less aggressive; as recurrence is a common finding in case of chronic granulomatous invasive fungal sinusitis; patients require a long-term follow-up.

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