



Case Report

Dual infection of maxillary sinus masquerading as a malignant lesion: A rare presentation

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ABSTRACT

Primary sinonasal tuberculosis is a rare form of TB even in developing countries where TB is prevalent and invasive mycotic infection is more common in immunocompromised patients. It is unusual to have a dual infection with tuberculosis and mycosis in the maxillary region mimicking a neoplastic process and causing bone erosion. A 49-year old male presented with 2-year history of left-sided facial swelling. On microscopic examination, epithelioid cell granulomas, chronic inflammatory infiltrate, macrophages, giant cells, and focal necrosis were seen. Ziehl Neelsen staining for acid-fast bacilli with 20% H₂SO₄ was positive. Fungal hyphae were also seen in the sections examined. Special stains (Periodic acid-Schiff and Grocott's methenamine silver stain) for fungal hyphae were positive. Mycosis and tuberculosis should always be considered in the differential diagnosis of maxillary lesions or sinusitis.

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INTRODUCTION

Tuberculosis, a multi-system disease, is still a major health problem in most parts of the developing world. Pulmonary tuberculosis is the most common form of primary tuberculosis, although primary tuberculosis can occur at any other site.¹ Extrapulmonary TB constitutes approximately 25% of all cases of TB, and of this, 10–35% occurs in the head and neck region where nodal involvement is the most common. The extranodal TB of the head and neck region constitutes <1%.² Tubercular infection of the nose, paranasal sinuses, and maxillary region is extremely rare

even in developing countries like India where the incidence of pulmonary tuberculosis is very high. The mechanical protection offered by the unidirectional ciliary clearance of the mucosal conveyor belt and the effective bactericidal properties of nasal secretions can explain why this entity is not a usual diagnosis in routine clinical practice.³

Fungal infections of the paranasal sinuses can be invasive or non invasive. The non invasive form is more common and causes chronic rhinosinusitis. In contrast, the invasive form is rapidly progressive and occurs in immunocompromised patients such as those with diabetes, prolonged steroid use, and neutropenia. Patients with invasive fungal disease frequently present with more severe symptoms of rhinosinusitis, including fever, facial edema, nasal stuffiness, and discolored nasal discharge. Invasive fungal rhinosinusitis (IFRS) was recently found to be more common than in the past, with high mortality rates ranging from 50% to 80%.⁴ Fungal infection of maxillary sinuses usually affects immunocompromised patients with predisposing factors like HIV infection, uncontrolled diabetes mellitus, neutropenia, and prolonged use of corticosteroids. Occasionally, it may also be seen in immunocompetent patients. Mycosis of maxillary sinuses can be very invasive can cause bone destruction and can mimic malignancy as well as granulomatous diseases like sarcoidosis and Wegener's granulomatosis.⁵

Dual infection with tuberculosis and mycosis in maxillary sinuses is rarest of rare, especially in immunocompetent patients.

CASE REPORT

A 49-year-old male, chronic smoker and alcoholic presented with swelling on the left side of his face for 2 years. There was no history of cough, hemoptysis, chest pain, night sweats, malaise, anorexia, weight loss or fever. On examination, a non-tender diffuse swelling was present on the left cheek. It involved the left infraorbital and maxillary region with an oroantral fistula and depressed nasal bridge plate. NCCT PNS revealed a lobulated soft tissue mass lesion in the left maxillary region causing localized surrounding bone erosions and destruction suggestive of locally aggressive neoplastic soft tissue mass lesion. The swelling was removed in toto and sent to histopathology. We received multiple grey-white soft tissue pieces measuring 1.5×1.5×0.6cm. On microscopic examination, epithelioid cell granulomas, chronic inflammatory infiltrate, macrophages, giant cells, and focal necrosis were seen. Ziehl Neelsen staining for Acid-fast bacilli with 20% H₂SO₄ was positive. Fungal hyphae were also seen in the macrophages in the sections examined. Special stains (Periodic acid-Schiff and Grocott's methenamine silver stain) for fungal hyphae were positive (fig.1 and 2). Antitubercular and antifungal drugs were started and the patient responded well.

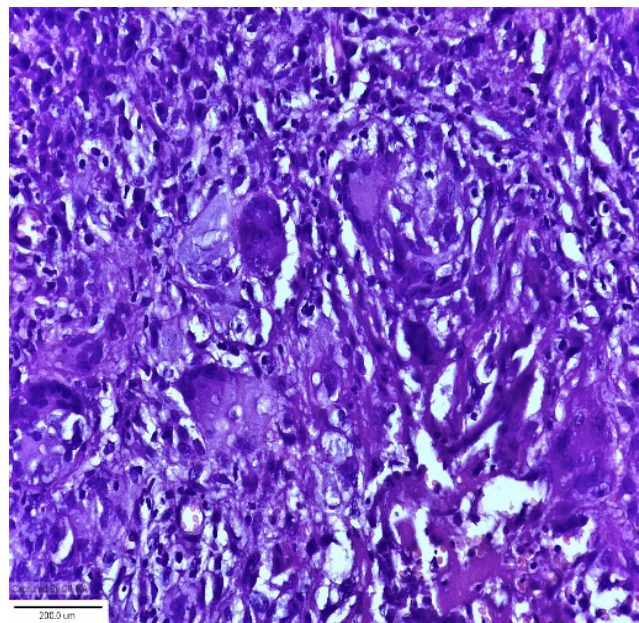


Figure 1 : Photomicrograph showing multinucleated giant cells (HE stain X 200)

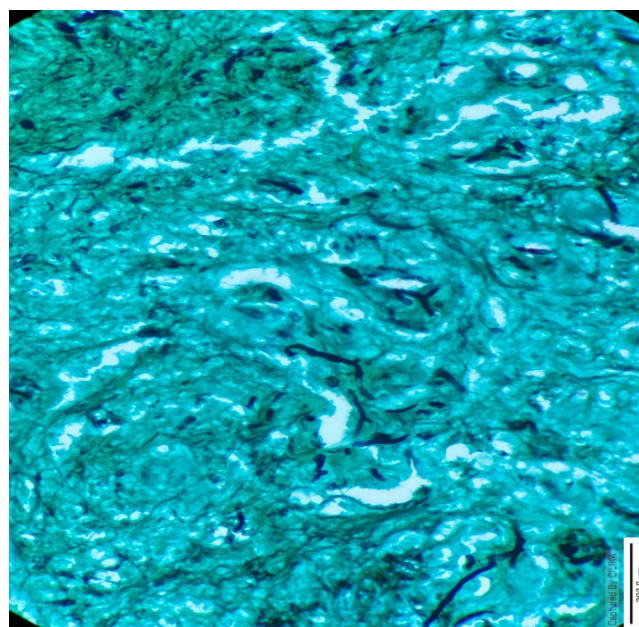


Figure 2: Broad aseptate hyphae showing 90-degree branching angle (GMS, X200)

DISCUSSION

Tubercular and fungal infections of the paranasal sinuses need to be recognized early to avoid complications like bone erosions and spread to orbit. Early diagnosis can prevent significant mortality and morbidity.⁶

Sinonasal TB is an uncommon lesion and extremely rare as a primary presentation but important since, if overlooked, may result in a fatal outcome. Gleitsmann reported the first case of TB in the sinonasal area.² Any of the sinuses may be affected by tuberculosis but maxillary and ethmoid are the

most susceptible. It is more frequent in females, and common symptoms are nasal obstruction, nasal discharge, crusting, and epistaxis. It commonly involves the nasal septum and the inferior turbinate. Sometimes, septal perforation, cleft of the nasal ala, or facial abscess may be present.⁷ In most cases, the bacilli reach the paranasal sinuses from the lungs by way of the bloodstream or by direct extension. In our case, the patient did not have pulmonary tuberculosis. Paranasal sinus tuberculosis without the involvement of the nasal cavity as in the present case is extremely rare.⁸

Mycotic infections usually occur in patients with chronic sinusitis with predisposing causes like trauma, prolonged corticosteroid therapy, immunosuppressive drugs, uncontrolled diabetes mellitus, neutropenia, HIV infection, radiation therapy, and burns. Our patient was immunocompetent and there was no history of corticosteroid or any other immunosuppressive therapy. The fungal agents usually isolated from paranasal sinuses include *Aspergillus*, *Mucor*, *Histoplasma*, *Coccidioides*, *Candida*, *Fusarium* spp, *Curvulariaspp*, *Pseudallescheria boydii* and *Alternaria* spp. *Aspergillus* is the most common fungal pathogen causing fungal sinusitis. The highest mortality rate (33.3%) was reported among patients with zygomycotic infection.⁹ A variety of different causative organisms can cause mycotic infection of the maxillary region including *Aspergillus*, *Candida*, and *Mucor*, and can cause invasive or non-invasive disease. Invasive mycoses can cause intraorbital and intracranial growth causing symptoms like blurred vision, gradual loss of vision, chemosis, and proptosis. If the brain is involved, it can lead to cavernous sinus thrombosis and various CNS manifestations. In our case, the infection had eroded the anterior and medial wall of the left maxillary sinus, the left lateral wall of the nasal cavity, and the left half of the maxillary bone and also extended into the right side of the maxilla crossing midline.¹⁰ Early and correct diagnosis combined with proper treatment can reduce the fatality rate associated with his disease. Symptoms easily overlap with other diseases and it is difficult for the physician to recognize them.⁹ Differential diagnoses of fungal and tubercular infection of the maxillary region including benign lesions like mucocele, polyps, other infective and non-infective granulomatous diseases like rhinoscleritis, sarcoidosis and Wegener's granulomatosis, and malignant lesions tend to have irregular intracranial surface with expansion of cavity and focal bone destruction.¹¹

Our case mimicked malignant lesions clinically and radiologically due to the erosion caused by the organisms. Dual infection of the maxillary region by tuberculosis and mycosis is extremely rare. It is obligatory to identify both infections for early management as missing even one of these can lead to poor therapeutic outcomes and increased morbidity due to the invasive nature of the organisms. Aggressive surgical debridement and antifungal and antitubercular drug therapy are necessary for satisfactory management and for complete eradication of the disease. The World Health Organization guidelines consist of a

6-month regimen: rifampicin, isoniazid, and pyrazinamide for the first 2 months, followed by 4 months of rifampicin and isoniazid. The most common cause of treatment failure is poor drug compliance which may lead to the emergence of drug-resistant organisms. Prolonged alternative therapy is required to treat this form of TB, often for up to 2 years. If TB is treated properly and consistently, these resistant forms are much less likely to develop.² Patients with IFRS historically received amphotericin B after pathological examination demonstrated tissue invasion by a fungus. The new drug voriconazole was reserved only for patients with a positive *Aspergillus* culture.⁴ Injection amphotericin B (dose 1–1.5 mg/kg/ day) and its lipid formulation (3–5 mg/kg/day). Lipid formulations of amphotericin B are considered superior over amphotericin B deoxycholate in the treatment of mucormycosis, as the former has less nephrotoxic effects, better penetration to the brain, and superior immunomodulatory effects.¹²

CONCLUSIONS

Mycosis and tuberculosis should always be considered in the differential diagnosis of maxillary lesions or sinusitis which do not respond to conservative therapy with antibiotics. Dual infections should always be kept in mind and ruled out to prevent unnecessary delay and morbidity for diseases that can be cured effectively.

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