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CASE REPORT/CAS CLINIQUE

Maxillary rhinosinusitis due to *Fusarium* species leading to cavernous sinus thrombosis

Rhinosinusite maxillaire à Fusarium *sp. conduisant à une thrombose caverneuse du sinus*

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Summary Fungal rhinosinusitis is being recognized and reported with increasing frequency over the last two decades worldwide. Intracranial extension is the most dreaded complication of fungal sinusitis with high mortality rates. We report a case of chronic rhinosinusitis in a 55-year-old diabetic male, caused by *Fusarium* species. The patient was diagnosed as a case of chronic left maxillary sinusitis with cavernous sinus thrombosis. The sinus lavage showed fungal elements on direct microscopic examination and culture revealed growth of *Fusarium* species within 4 days of incubation. Conservative therapy with IV amphotericin B resulted in favorable outcome of the patient. This is an extremely rare case where cavernous sinus thrombosis occurred as a complication secondary to *Fusarium* species rhinosinusitis.

Résumé La rhinosinusite fongique est rapportée avec une fréquence accrue dans le monde au cours de ces deux dernières décades. L'extension intracrânienne est la complication la plus redoutée de ces sinusites fongiques avec un taux élevé de mortalité. Nous rapportons ici un cas de rhinosinusite chronique à *Fusarium* sp. chez un homme diabétique âgé de 55 ans. Le diagnostic de sinusite chronique du maxillaire gauche avec une thrombose caverneuse du sinus est alors porté. Le lavage du sinus montre des éléments fongiques à l'examen microscopique direct et sa culture révèle un *Fusarium* sp. après quatre jours de culture. Une thérapeutique conservatoire par amphotéricine B par voie IV a donné un résultat favorable. Cela est un cas

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extrêmement rare alors que la thrombose caverneuse du sinus peut survenir comme complication secondaire de ces rhinosinusites à *Fusarium*.

Introduction

Rhinosinusitis is a common disorder affecting approximately 20% of the population and chronic rhinosinusitis accounts for more than 90% of all the cases [1]. Fungal rhinosinusitis is most commonly caused by *Aspergillus* species followed by *Rhizopus*, *Mucor*, *Cladosporium*, *Candida*, *Cryptococcus* species, etc. The non-invasive ones are generally demateacious molds like *Curvularia*, *Bipolaris*, *Alternaria*, *Fusarium* species, etc. These cause intracranial complications in about 20% of the patients [12]. Chronic invasive fungal rhinosinusitis has a chronic course, often in subtly immunocompromised patients, such as those with diabetes mellitus and corticosteroid treatment [1].

Fusarium species may cause allergic sinusitis or chronic non-invasive or invasive sinusitis in immunocompetent host. In immunocompromised host it is always the invasive type. The clinical manifestations of fusarial sinusitis are indistinguishable from those caused by *Aspergillus* species. Necrosis of the mucosa is a hallmark and is a consequence of the angioinvasive nature of these mycoses [7]. Cavernous sinus thrombosis is a rare consequence of invasive fungal sinusitis. In invasive form, immunocompromised patients can allow the organism easy access into mucosal structures, infiltrating orbital and intracranial structures via hematogenous spread [2]. We present a unique case of cavernous sinus thrombosis complicating maxillary sinusitis caused by *Fusarium* species, which has not been reported previously.

Case report

A 55-year-old male with type-2 diabetes mellitus of 7 years duration presented with history of fever since 8 days. Two days later he started with headache, which was throbbing in nature, associated with projectile vomiting. This was followed by diplopia, photophobia and redness of both eyes. Patient gave history of recurrent maxillary and frontal sinusitis since last 1 year, which is indicative of chronic nature of the disease. According to the previous reports, it was noticed that blood sugar level was not under optimal control, ranging from 250 to 360 mg/dl. The patient was clinically assessed and admitted to intensive care unit. On general examination, patient was averagely built and nourished with mild pallor. Pulse rate was 110/min; blood pressure was 130/70 mmHg. Other vital parameters were within normal limit. Patient was conscious, oriented and febrile with terminal neck stiffness. On local examination, patient had tenderness over left maxillary sinus. There was bilateral proptosis with conjunctival injection and external ophthalmoplegia with normal size pupils. There was involvement of III, IV and VI cranial nerves. Deep tendon reflexes, motor and sensory system was normal and plantar response was flexor. On fundoscopy, there was bilateral papilloedema showing early changes. Other systemic examination (CVS, RS, and PA) was unremarkable. Blood investigations were as follows:

- Hb: 12.3 gm%;
- total WBC count: 18500 mm^3;
- platelet count: 2.5 lac/mm^3;
- ESR: 30 mm/hr;
- c-reactive protein: 3.8 mg/dL;
- BSL: 375 mg%;
- HbA_1C: 12.7%;
- BUL: 29 mg%;
- serum creatinine: 1.2 mg%;
- Na^+: 138 meq/L;
- K^+: 4.2 meq/L.

Urine analysis report revealed urine sugar: 4+ and urine acetone was negative. X-ray paranasal sinus: left maxillary sinus haziness with fluid level. In view of history of uncontrolled diabetes mellitus, recurrent maxillary sinusitis with clinical features of bilateral cavernous sinus thrombosis, patient was referred to MRI imaging of brain. In the next visit, the patient returned with only the reports of MRI findings, as the images were misplaced in the radiology department of the tertiary care centre. As per the reports, MRI was suggestive of bilateral cavernous sinus thrombosis with mild cerebral edema. At the same time, MRI imaging of paranasal sinuses was also done which revealed fluid level in left maxillary sinus. Clinically, the case was diagnosed as chronic invasive rhinosinusitis. Considering left maxillary sinus as a source of infection causing bilateral cavernous sinus thrombosis, diagnostic as well as therapeutic tapping was done under local anaesthesia and sinonasal tissue taken for histopathology and culture.

Laboratory findings

The specimen was sent to histopathology and microbiology laboratory in a sterile test tube for further investigations. Histopathology revealed filamentous fungi invading the sinonasal mucosa and angio-invasion with few inflammatory cells. Gram staining showed presence of inflammatory cells and bacterial culture was sterile. Potassium hydroxide mount of the specimen revealed slender, branched septate hyphae on microscopy (Fig. 1). The specimen was cultured on Sabouraud dextrose agar with and without antibiotics and incubated at room temperature at 37 °C. After 4 days of incubation, fungal culture showed cottony, white to cream coloured colony (Fig. 2). Lactophenol cotton blue mount of the growth showed hyaline, septate hyphae with acute angled branching with macroconidia and microconidia. Macroconidia were multicellular, sickle-shaped with three to five septae and microconidia with one to three septae (Fig. 3). Depending on the gross and microscopic findings, the fungal isolate was identified to be *Fusarium* species. The isolate was confirmed to be *Fusarium* species by the Department of Medical Microbiology, Postgraduate Institute of Medical Education and Research (PGIMER), Chandigarh,