10.5005/jp-journals-10013-1117

CASE REPORT

A Rare Cause of Frontal Sinusitis

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ABSTRACT

Actinomycosis is a granulomatous and fibrosing disease caused by a Gram-positive, anaerobic, nonacid fast bacterium of the genus *Actinomyces*.

We report an unusual presentation of actinomycosis that involved the frontal sinus. Clinical features, diagnostic criteria, treatment and follow-up are presented.

Keywords: Actinomycosis, Frontal sinus, Pathology.

How to cite this article: Kumar M, Sadasivan A, Viswam V, Menon U. A Rare Cause of Frontal Sinusitis. Clin Rhinol Int J 2012;5(1):41-43.

Source of support: Nil

Conflict of interest: None declared

INTRODUCTION

Frontal sinusitis usually, though not always, occurs in association with chronic rhinosinusitis. Actinomycosis is commonly seen in the neck or face region in a background of immunocompromised status. We report a rare combination of these with its diagnostic dilemma and successful management protocol.

CASE REPORT

An otherwise healthy 35-year-old male presented to our hospital with 2 weeks history of headache and swelling above his left eyebrow. A computed tomography imaging of the brain was done elsewhere and reported as a case of frontal encephalocele. There was no history of fever, periorbital swelling, nasal discharge or proptosis.

The patient gave a history of sustaining a head injury 6 months prior. He had undergone surgery, apparently for sinusitis, 2 years earlier. No details were available.

On examination, there was a diffuse, soft, nontender swelling superior to the medial aspect of left eyebrow. Palpation revealed left frontal sinus tenderness. Nasal endoscopy revealed mild fullness anterior to left middle meatus. A repeat computed tomography showed bone erosion of medial wall of left orbit and posterior wall of left frontal sinus with soft tissue density within the frontal sinus (Fig. 1). MRI showed intermittent signal density in T1 and T2 weighted images, suggestive of thick fluid rather than soft tissue. This was more in favor of chronic infection and ruled out the possibility of encephalocele. In contrast enhanced MRI, there was mucosal enhancement, which was contradicting the initial suspicion of mucocele (Fig. 2).

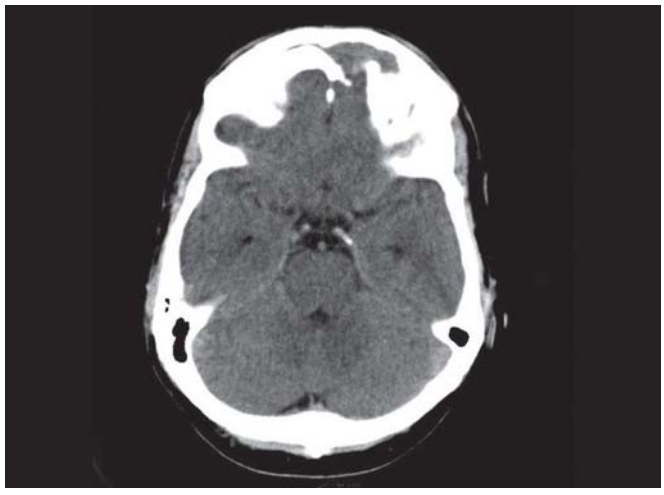


Fig. 1: Axial cut of CT PNS showing dehiscent posterior wall of frontal sinus

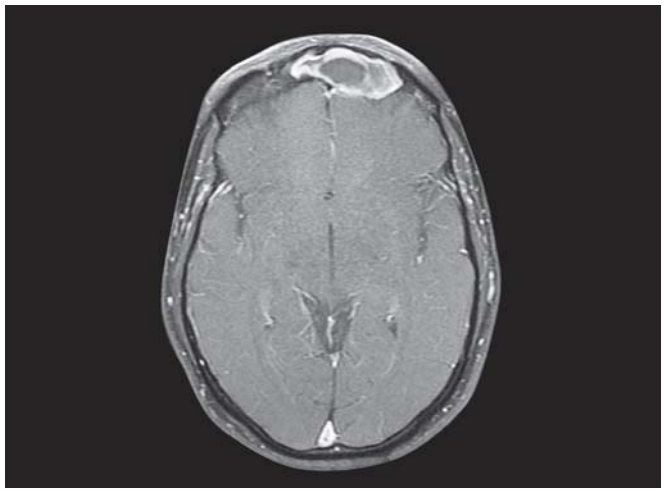


Fig. 2: Axial cut of contrast MRI showing mucosal enhancement in fontal sinus

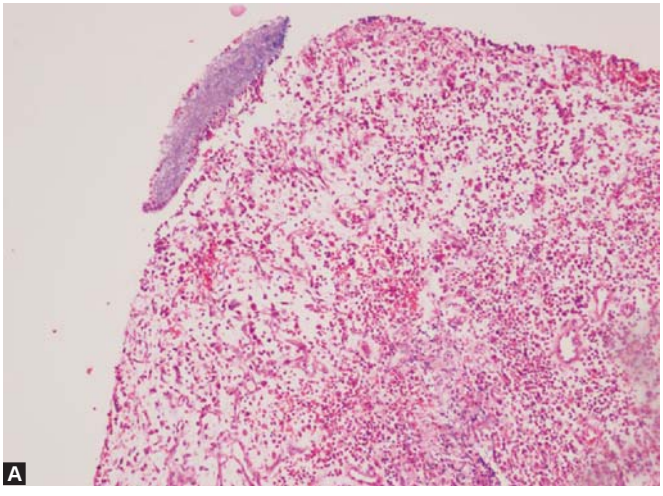
Patient was taken up for frontal sinus surgery with a combined external (Lynch Howarth) incision and endoscopic approach. The sinus was filled with brown clay like material. This was completely evacuated and sent for histopathological examination, fungal smear test and Gram staining. Mycology report came first citing the presence of long slender branching bacilli and no evidence of yeast or filamentous fungi. Gram staining revealed variable numerous branching filamentous bacteria and Gram-positive coccoid forms, suggestive of actinomycosis. The initial histopathological report was suggestive merely of acute on chronic sinusitis with no identifiable fungus. However, a review following the Gram stain report revealed mixed

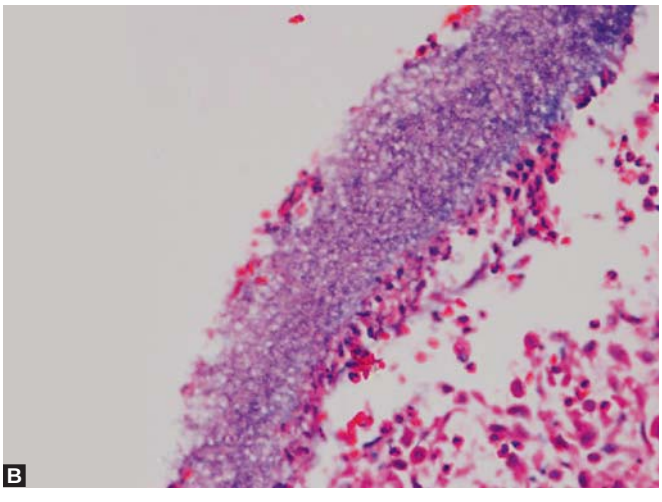
inflammatory infiltration with a colony showing clumps of hemophilic filaments with neutrophils sticking to it (Splendore-Hoeppli phenomenon) (Figs 3A and B), latter of higher magnification. A repeat Gram staining of this section of the slide was done for the purpose of documentation (Fig. 4).

The patient was started on intravenous crystalline penicillin 20 lakh IU/dose 6th hourly for 4 weeks, followed by oral penicillin (Kaypen) 500 mg 6 hourly for 5 months. He has been asymptomatic for the subsequent 1 year with no evidence of recurrence.

DISCUSSION

In humans, the causative agent for actinomycocis is *Actinomyces israelii*. Other *Actinomyces*, such as *A. naeslundi*, *A. odontolyticus* and *A. viscosus* are also involved in some cases. All these species may be found in the oral cavity, respiratory and digestive tracts as normal commensals.





Figs 3A and B: Photomicrograph of the evacuated material showing mixed inflammatory infiltration with a colony of hemophilic filaments with neutrophils sticking to it (Splendore-Hoeppli phenomenon) (A) H & E stain; x100 (B) H & E stain; x400

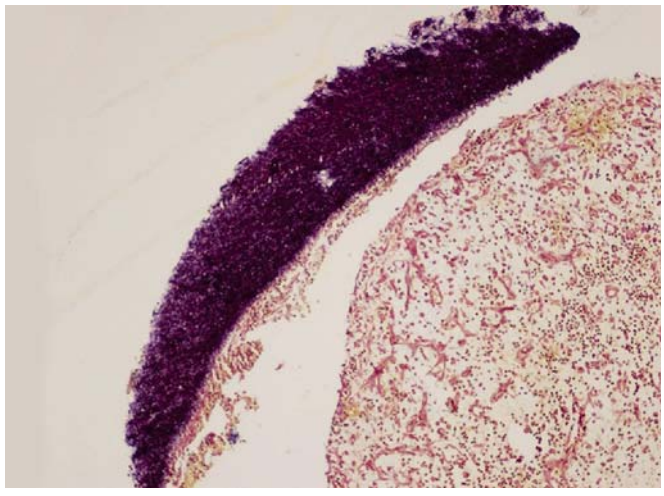


Fig. 4: Gram stain showing Gram-positive filaments (x400)

When the integrity of the mucosal barrier is compromised and access to the tissue is gained, these organisms become pathogenic. Trauma, surgery and dental extractions are common conditions thought to facilitate such an access.[1] Diabetes, immune suppression, long term steroid use and malnutrition have also been implicated as predisposing factors.

Tissue pathology in actinomycosis consists of a prolonged chronic inflammatory process creating a tumor like mass tissue destruction, osteolysis and multiple sinus tracts. It has both granulomatous and suppurative features.[2] It can manifest either as an acute fast progressing form which presents as a soft swelling associated with suppuration or as a chronic slowly progressing form which produces a woody hard swelling with multiple fistulae or sinus tracts.[3]

Anatomically, the actinomycosis infections are classified into cervicofacial, thoracic and abdominal types. The former is the commonest form comprising up to 50% of cases in some series, but reports that involve the nose and sinuses are rare.[1,4,5]

The usual site of this rare infection in the sinonasal region is the maxillary antrum.[6] Very rarely, it can involve the ethmoid or sphenoid sinus. However, its occurrence in the frontal sinus is extremely rare. In our literature search, we could find only one such case.[4]

The diagnosis of actinomycosis largely depends on the presence of sulfur granules in the exudates on culture and on the histologic examination of the biopsy specimen. The cultures must be brought directly to the laboratory under anaerobic conditions and plated on brain, heart or blood agar. Colonies take approximately 2 weeks of strict anaerobic incubation to grow.[7] Computed tomography scans can be helpful in the identification of bone involvement and in the evaluation of the extent of the lesion.

In the present case, diagnosis was made on the basis of histologic examination of the biopsy specimen obtained after sinus surgery. However, radiological investigations were needed to clear the initial dilemma regarding possibility of mucocele.

Treatment consists of surgery for the lesion and administration of a long course of antibiotic for 6 to 12 months. *Actinomyces* tends to invade locally and the avascular fibrotic wall of the lesion makes treatment with antibiotics difficult.[8] Therefore, surgical removal of the involved tissues is essential for successful treatment of paranasal sinus actinomycosis.

Actinomyces species are susceptible to most antimicrobials. Penicillin administration remains the norm in dealing with actinomycosis. The prescribed schedule is intravenous treatment for 2 to 6 weeks, followed by long term oral administration of penicillin V for a period of 6 to 12 months.

Our case represents an extremely uncommon manifestation of actinomycosis, with successful management protocol. We report it for bringing to light the high index of suspicion required to make an accurate diagnosis and initiate the appropriate antibiotic therapy.

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