

Epidural Empyema Caused By Frontal Sinus Aspergillosis

Epidural Ampiyeme Neden Olan Frontal Sinüs Aspergillomu

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ABSTRACT

Aspergillosis infection located primarily in the frontal sinus and invading into the dura is rare. A 74-year-old male patient was admitted with headache of 3-month duration that did not respond to analgesics. Cranial MRI showed an irregular hyperintense lesion in T2 FLAIR and T1 FLAIR, arising from the right frontal sinus and advancing to the intracranial space. A collection, hyperintense on T2 FLAIR and isointense on T1 FLAIR, adjacent to the left frontal sinus was observed, consistent with epidural empyema. The lesions were drained surgically, and subsequently treated with antifungal therapy using Amphotericin B. The headache resolved completely after surgical drainage. The patient has been followed-up for a year and is currently asymptomatic. Aspergillosis infections lodging primarily in the frontal sinus and spreading to the dura are very rare, therefore, we report this case in the present paper. (*Archives of Neuropsychiatry* 2012;49: 83-5)

Key words: Frontal, sinus, epidural, aspergillosis, empyema

ÖZET

Primer olarak frontal sinüse yerleşerek duraya yayılım gösteren aspergillosis enfeksiyonu oldukça nadirdir. Yetmiş dört yaşında erkek hasta 3 aydır olan analjezik tedaviye cevap vermeyen baş ağrısı yakınmasıyla başvurdu. Kranyal MR'ında; sağ frontal sinüsten intrakranyal alana doğru uzanan T2 FLAIR ve T1 FLAIR'de hiperintens irregüler lezyon olduğu, intraserebral alana doğru ilerlediği görüldü. Sol frontal bölgede ise T2 FLAIR'de hiperintens, T1 FLAIR'de ve GRE (ECHO) incelemesinde izointens görünen epidural ampiyem ile uyumlu kolleksiyon saptandı. Cerrahi olarak lezyonlar drene edildi, amfoterisin B tedavisi başlandı. Cerrahi drenaj sonrası bir yıllık takibi boyunca benzer baş ağrısı tekrarlamadı. Primer olarak frontal sinüse yerleşerek duraya doğru yayılım gösteren ve epidural ampiyeme neden olan aspergillosis enfeksiyonlarının çok nadir olması nedeniyle bu olgu sunulmaktadır. (*Nöropsikiyatri Arşivi* 2012;49: 83-5)

Anahtar kelimeler: Frontal, sinus, epidural, aspergillosis, ampiyem

Introduction

Epidural empyema is mostly caused by sinusitis, trauma associated with skull fracture and craniotomy as well as orbital cellulitis and cranial osteomyelitis. The most common infectious agents are staphylococci, and aspergillus species have also been reported, even though rarely. Primary frontal sinus aspergilloma was first reported by Gupta in 1973, after that, a few cases of aspergilloma located in the frontal sinus were reported (1-9).

Primary frontal sinus aspergilloma is a rare clinical condition that is difficult to diagnose. In this paper, we present a case of primary frontal sinus aspergilloma which eroded the posterior wall of the frontal sinus, reaching the epidural space and resulting in empyema.

Case

A 74-year-old male patient with type 2 diabetes mellitus presented with a progressive headache of 3-month duration. The pain was initially located to the right side of the head and later disseminated to the whole cranium and had a throbbing character. Although it was decreased by analgesics, it did not disappear. Neurologic examination showed mild paresis in the left lower extremity and extensor response of the plantar reflex.

Cranial MRI studies demonstrated an irregular hyperintense lesion in T2 FLAIR and T1 FLAIR which originated from the right frontal sinus and advanced to the intracranial space, did not invade the intracerebral space, and had contrast enhancement (Figure 1). A collection neighboring the left frontal sinus that was

hyperintense on T2 FLAIR and isointense on T1 FLAIR and GRE (ECHO) was observed, which was consistent with epidural empyema. It was considered that the mass lesion in the right frontal sinus which showed contrast enhancement progressed to the intracranial space and resulted in dural thickening and left frontal empyema. Biochemistry revealed sedimentation rate of 76 mm/ hour, WBC 10300, and CRP 10 mg / dl. Computed tomography (CT) of the chest and ultrasound of the abdomen were normal.

The patient underwent bifrontal craniotomy that included superior part of the frontal sinuses. The left side of the frontal sinus was filled with black-green soft tissue. This pathologic material was removed along with sinus mucosa. The posterior wall of the sinus was eroded by the lesion and the epidural space was invaded by a harder and more elastic granulation tissue tightly adherent to the dura mater. After resection of the epidural granulation tissue, the resultant bone defect was repaired with autologous calvarial bone grafts. Bilateral total obliteration of the frontal sinuses was carried out using subcutaneous fat tissue obtained from the abdominal wall. Next, a frontal galeal flap was turned over to the sinus and fixed to the frontal base by using sutures and fibrin glue. Histopathological examination revealed fungal microorganisms that showed septation, had a hyphal

diameter of 4-5 microns, bifurcated with 45 degrees, and stained strongly with GMS and PAF stains, all consistent with *Aspergillus* spp. Culture showed growth of *Aspergillus flavus* (Figure 2). Intravenous liposomal amphotericin B treatment was initiated and continued for 3 months. The headache resolved completely after surgical drainage. The patient has been followed-up for 12 months and is currently asymptomatic. Control MRIs performed on the third and sixth months showed no residual or recurring lesions.

Discussion

Invasive *Aspergillus* infections are often reported in immunocompromised patients with neutrophil defects or under steroid therapy (10). On the other hand, *Aspergillus* spp. may rarely cause invasive infections in immunocompetent individuals (11,12). HIV infections, diabetes mellitus, prosthetic devices, trauma and advanced age are other predisposing factors (10,13). Our patient had diabetes mellitus and advanced age as predisposing factors.

Intracranial fungal sinus infections may cause fever, nasal congestion, headache, proptosis, visual disturbances (14) and, these symptoms often depend on the affected intracranial space. Our case complained of a mild headache for 3 months that had progressive loss of response to analgesics. Neurologic examination showed mild paresis in the left lower extremity and extensor plantar response.

Previous studies have shown that *Aspergillus* is the most common agent in fungal sinusitis, mycetomas lodge most frequently in the maxillary sinuses (15-17). *Aspergillus* infections can be invasive or noninvasive. Noninvasive infection causes mucosal destruction or bone expansion similar to allergic sinusitis or sinonasal fungus ball, and presents with symptoms of chronic sinusitis, however, without invasion of soft tissues or bone. Noninvasive aspergillosis usually affects immunologically competent individuals, and depends essentially on local host conditions. Invasive infection is localized and fulminant. Localized disease often begins in the sinuses, spreads to the neighboring structures, causes focal bone erosion, may advance to the vessel wall and finally, leads to stroke or death. Fulminant form is associated with multiple organ involvement. In our case, the presence of mucosal and bone invasion, and spread of the infection to the epidural space led us to the opinion that the infection was an invasive form.

Siddiqui et al. separated craniocerebral aspergillosis with sinonasal origin into 3 types, based on CT and MR findings: type 1: sinonasal disease with intracerebral aspergillosis without being contiguous, type 2: sinonasal disease contiguous with intracranial extradural extension and type 3: sinonasal disease with only orbital and/or cranial base bone invasion /destruction. Although the MRI findings of our patient were similar to type 2, there were some differences. The lack of a mass lesion and appearance of epidural empyema were the predominant features in our patient.

A case of epidural empyema, frontal bone infection and subcutaneous cellulitis caused by *Aspergillus fumigatus* and occurring 2 months after a frontal craniotomy performed after trauma is reported in the literature (18). Imaging studies showed

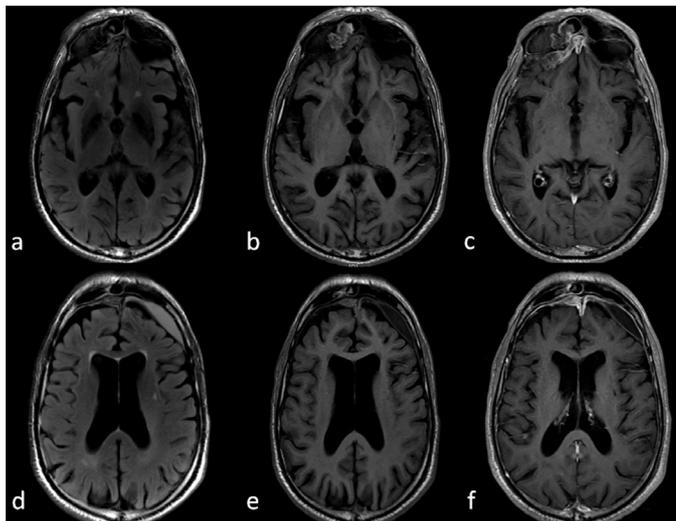


Figure 1. Cranial MRI studies showed left frontal empyema and an irregular hyperintense lesion originating from the right frontal sinus. (a,d) T1 FLAIR sequence, (b,e) T2 FLAIR sequence, (c, f) T1-weighted images with contrast

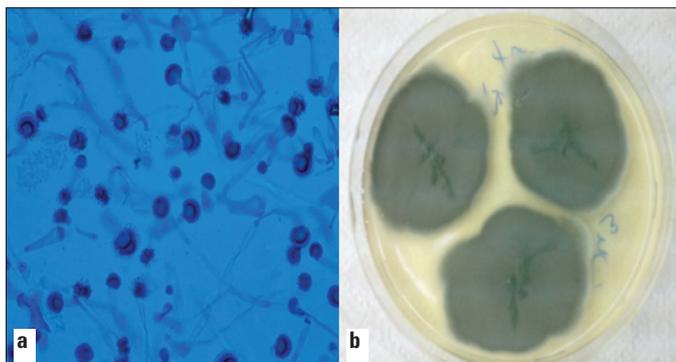


Figure 2. (a) Microscopic appearance of *Aspergillus flavus* with cotton blue lactofenol stain. (b) Macroscopic appearance in Sabouraud agar

coexistence of primary frontal sinus aspergilloma and epidural empyema. The case presented here had no history of cranial trauma or surgery. The etiologic factor was determined to be *Aspergillus flavus*.

The invasive form of aspergillosis possesses high morbidity and mortality rates even in immunocompetent patients. Therefore, early diagnosis, before the organism invades the central nervous system and vascular structures, is essential. Once the diagnosis is established, an adequate surgical resection and aggressive postoperative antifungal treatment are the cornerstones of treatment. Itraconazole per se may be curative in chronic invasive aspergillosis, however, fulminant forms require amphotericin B after radical surgery (19). The significance of combined antifungal treatment in skull base invasive aspergillosis is stressed (20). Regarding the duration of antifungal treatment, periods between 6-10 weeks or up to 6 months in skull base aspergillosis are reported (19-20). Our patient underwent complete surgical resection and later, antifungal treatment for 3 months. The patient is currently asymptomatic and has been followed-up for the last 12 months.

Conclusion

Paranasal sinus aspergillomas spreading to the intracranial cavity often have a mass pattern involvement, however, they may rarely extend to the dura and result in dural thickening. The case presented in this report is different with respect to frontal localization and presence of epidural empyema. Early diagnosis, complete surgical resection and antifungal treatment are necessary for successful infection control.

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