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Unilateral Retroorbital Pain Secondary to Isolated Sphenoid Sinus Aspergillosis

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A 75-year-old man with type 2 diabetes mellitus presented with a one-day history of general fatigue, vomiting, and headache. He noted severe pain in the left retroorbital region, not increasing on eye movement. He was awake and alert, and febrile (38.6 °C). No meningeal signs, diplopia, or visual disturbance were noted. Laboratory examination revealed a white blood cell count of 7.23×10^3 cells/μL (normal range, 3.58×10^3 to 8.15×10^3 cells/μL) with 90.1% neutrophils (normal range, 39.6% to 67.0%) and C-reactive protein level of 27.1 mg/L (normal value < 2.6 mg/L). His nasal endoscopic examination findings were unremarkable. Plain cranial computed tomography revealed a soft tissue density with high-density calcifications on the left sphenoid sinus (Figures 1A and B; blue arrowheads), suggestive of fungal sinusitis. Bone destruction and other sinus lesions were not noted.

The patient underwent trans-nasal endoscopic sphenoidotomy. Examination of the resected tissue revealed mycetoma with longitudinal septate hyphae. These branched at acute angles and stained black on Grocott's methenamine silver stain under bright-field microscopy (Figure 2). These findings were indicative of aspergillosis. The resected tissue culture was positive for *Aspergillus fumigatus*. The patient was prescribed 400 mg oral voriconazole twice daily on the first day, followed by 200 mg twice daily for eight weeks. His headache eventually subsided, and the postoperative course was uneventful. He was discharged three weeks after admission. No recurrence was found on computed tomography performed at the 12-week follow-up visit.

Patient consent for publication has been obtained.

Comment

Isolated sphenoid sinus aspergillosis is rare due to the preferential seeding of fungal spores in the ethmoid and maxillary sinuses.[1,2] The incidence of sphenoid sinus aspergillosis is low, between 0.5 and 1.2% per year.[1] The most common symptom is retroorbital and occipital headaches, followed by nasal blockage or discharge and recurrent mild epistaxis.[1] However, the diagnosis of aspergillosis in an isolated sphenoid sinus is challenging because of its non-specific symptoms. Therefore, this condition is often diagnosed at the time of operation or postoperatively and perhaps is even underdiagnosed.

Fortunately, chronic non-invasive sphenoid sinus aspergillosis is a benign disease. However, the sphenoid sinus is adjacent to vital structures, such as the cranial nerves, including the optic nerve, internal carotid artery, and cavernous sinus. Therefore, a prompt diagnosis is essential for patients with sphenoid sinus aspergillosis because its delayed diagnosis may lead to serious complications such as cerebral nerve involvement and cavernous sinus thrombosis due to bone destruction and invasion to adjacent organs.

Imaging modalities are essential in the diagnosis of sphenoid sinus aspergillosis. With the prevalent use of computed tomography for the evaluation of headaches, the identification rate of sphenoid sinus aspergillosis has increased considerably. The central high density in sphenoid sinus lesions on computed tomography, caused by the accumulation of calcium salts towards the necrotic area of central mycetoma, is considered characteristic of aspergillosis and is useful in the diagnosis of this condition.[3] It has been reported that the sensitivity of computed tomography for the diagnosis of this disease is 53%[1] based on the presence of calcification in the sphenoid sinus, as seen in this case. Moreover, magnetic resonance imaging is useful due to its high sensitivity and specificity, especially in differentiating sphenoid sinus tumours.[1] Therefore, if computed tomography findings are suggestive of a tumour, magnetic resonance imaging may be useful to more clearly delineate the lesion.[4] Histological examination using Grocott's methenamine silver stain is sufficient to confirm the diagnosis of sinus aspergillosis. However, because the fungal ball is composed of dead spores, it is estimated that culture identifies the pathogen in only 30% of the cases.[1] Moreover, invasive aspergillosis requires prompt diagnosis and treatment, and the presence of invasion of the mucosa by the pathogen should be confirmed.

The sphenoid sinus aspergillosis in this case was of a non-invasive type and was considered to be aspergilloma of the sphenoid sinus.[5] Since this condition has a chronic course, an acute course of fever and elevated C-reactive protein, an inflammatory marker, as observed in this case were findings suggestive of a bacterial infection. Therefore, although culture of the resected tissue and pus were negative for bacteria, it cannot to be ruled out that complications from an acute bacterial sinusitis with aspergilloma increased the pressure in the sphenoid sinus, leading to retroorbital pain. Although no antibiotics were prescribed in this case, drainage, a basic surgical principle for abscesses, may have been sufficiently effective for bacterial sinusitis. Medical therapy alone is insufficient for aspergilloma, and a surgical intervention is always recommended.

Isolated sphenoid sinus aspergillosis is a rare but crucial disease that should be considered as a cause of sphenoid sinusitis. Moreover, surgical resection of the aspergilloma may prevent further complications, such as acute bacterial sinusitis, and more invasive sinus involvement, , especially in patients with diabetes and other immunocompromised conditions.

Authors' Contribution

All authors had access to the patient's data and played a role in writing the manuscript. All authors approved the final version of the manuscript.

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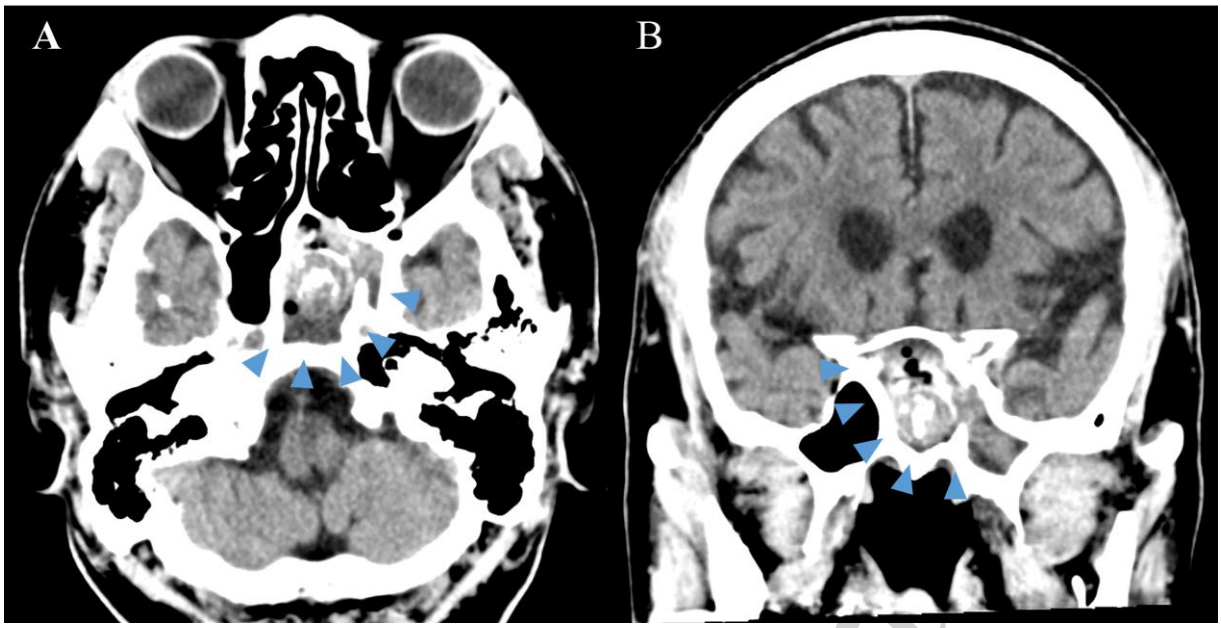

Figure 1: Axial (A) and coronal (B) planes of plain cranial computed tomography demonstrating a soft tissue mass with abnormal high-density calcifications in the left sphenoid sinus (blue arrowheads).

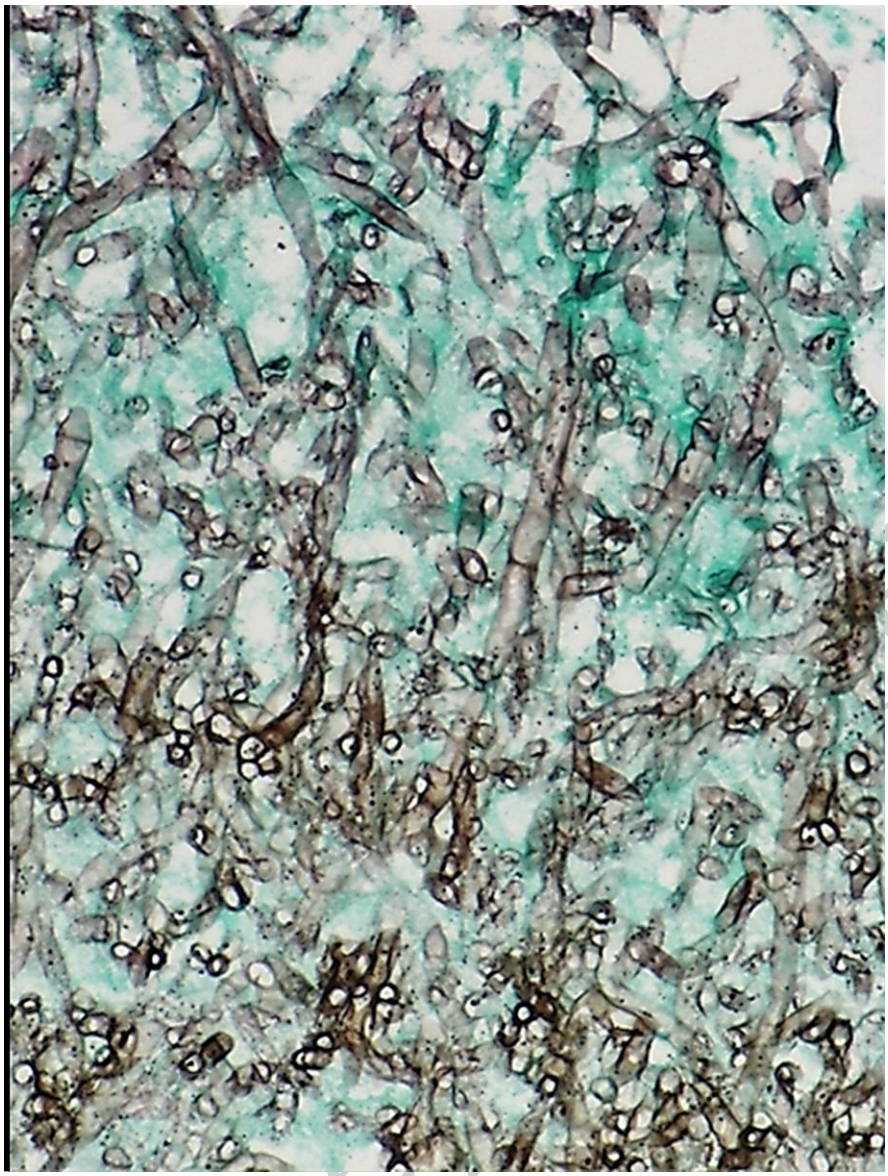

Figure 2: Bright-field microscopy of the resected tissue showing septate hyphae branching at acute angles and producing the characteristic black stain on Grocott's methenamine silver staining, indicative of aspergillosis.