Aspergillosis of the Petrous Apex and Meckel's Cave

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ABSTRACT

Cranial cerebral aspergillosis is a rare entity in immunocompetent patients. Invasive disease involving the petrous apex and Meckel's cave has rarely been described. We present a case of localized invasive petrous apical and Meckel's cave disease in an immunocompetent patient who presented with hemicranial neuralgic pain.

KEYWORDS: Aspergillosis, petrous apex, Meckel's cave, computed tomography, MRI

CASE REPORT

Cranial cerebral aspergillosis is rare, usually seen in immunocompromised patients. Invasive disease involving Meckel's cave and the petrous apex has been rarely described. Diseases in both areas have been associated with direct extension from chronic otitis media, sinusitis, or following petrous apex cholesterol granuloma drainage.¹⁻⁴

A 75-year-old woman presented with a 6-month history of severe right-sided facial pain diagnosed as atypical trigeminal neuralgia. The pain also extended to the right ear and the periauricular and temporal regions, and was characterized by a paroxysm of dull to sharp pain for a few minutes before gradually subsiding. Over time, the pain also involved her jaw and tongue, leading to a loss of lingual sensation. She was treated over this period with numerous drug regimens for idiopathic trigeminal neuralgia without complete relief. There was no alteration in balance, diplopia, facial weakness, hearing loss,

or vertigo. She described occasional fever and night sweats. During the course of her illness, she lost 6 kg. Apart from hypertension and hepatitis C of unknown source, she had no other significant medical history. Laboratory testing was normal, and she was immunocompetent. Physical examination revealed hypesthesia in the right trigeminal distribution, particularly V2. There was also right sided hemilingual loss of sensation and a reduced right corneal reflex. Extraocular movements and papillary responses were normal.

Petrous temporal bone computed tomography (CT; Fig. 1) demonstrated widening of the right Vidian canal and foramen lacerum, soft tissue in the lateral right sphenoid sinus, and apparent erosion of the lateral wall of the right sphenoid sinus. Magnetic resonance demonstrated a small amount of T2 hyperintense soft tissue in the lateral right sphenoid sinus, erosion of the right Vidian and carotid canals, and enhancing soft tissue in Meckel's cave and the petrous apex (Fig. 2). The soft

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Figure 1 Axial high-resolution computed tomography of the skull base without contrast. There is expansion of the right Vidian canal (white arrow) and foramen lacerum (white arrowhead). There is apparent erosion of the lateral wall of the right sphenoid sinus (RSS) with soft tissue within the sinus cavity. The normal left Vidian canal (black arrow) and foramen lacerum (black arrowhead) are seen.

tissue surrounded the horizontal petrous right internal carotid artery and there was absence of blood flow in the artery on magnetic resonance angiography. The center of the mass was restricted on diffusion imaging, suggesting an abscess.

The patient underwent transsphenoidal endoscopic biopsy under stereoscopic navigation. The sphenoid sinus was widely exposed to visualize the posterior and lateral wall, including the sella, carotid indentation, optic-carotid recess, and midclivus. The lateral sphenoid mucosa was inflamed; this was stripped. The underlying bony wall was intact, contrary to what was suggested by the CT. Entering the petrous apex using a 4-mm diamond burr, granulation tissue was dissected and then frank pus extruded. Further dissection into Meckel's cave resulted in a second extrusion of frank pus. Frozen section revealed fungal disease. Samples were sent for pathology and microbiology; the region was irrigated and filled with fat and Gelfoam (Pharmacia & Upjohn Company, Kalamazoo, MI) to prevent cerebrospinal fluid leak. There were no postoperative complications.

Periodic acid-Schiff staining (Fig. 3) showed multiple small fragments of granulation tissue with occasional inflamed sphenoidal tissue. Aggregates of plasma cells were noted. In the inflamed areas, there were collections of fungal hyphae with septations at acute angles, compatible with *Aspergillus* infection. Microbiology was positive for *Aspergillus fumigatus*. Appropriate intravenous medical therapy was instituted.

Figure 2 Coronal T2 (A, B), coronal T1 with gadolinium (C, D), axial T1 with gadolinium (E, F), axial diffusion (G), and magnetic resonance angiography (H). There is abnormal mass like soft tissue in the petrous apex (A, C, E, arrows) and Meckel's cave (B, D, F, arrows), which enhances following gadolinium. The lesion demonstrates central restricted diffusion (G, arrow). There is no flow in the right internal carotid artery on magnetic resonance angiography (H, arrow), indicating occlusion.

DISCUSSION

Aspergillus, a spore-forming saprophytic thermotolerant fungus, is a ubiquitous airborne conidia. Of the 350 strains, *A. fumigatus*, *A. flavus*, and *A. niger* are associated with human disease. The clinical spectrum includes invasive and noninvasive forms, the former particularly associated with an immunocompromised state. It can, however, occasionally effect immunocompetent hosts, as in our case. The disease process usually involves the paranasal sinuses or mastoids with extension to contiguous orbital and intracranial compartments. Sinus aspergillosis can present in four ways: acute invasive, chronic invasive, chronic granulomatous, and aspergilloma.⁵ The acute invasive form is usually seen in severely immunocompromised patients, such as those with neutropenia or bone marrow transplant. There is sinus opacification and bony erosion. Pathologically,

Figure 3 Periodic acid-Schiff stain sections (low magnification A, high magnification B). There are multiple small fragments of granulation tissue with occasional inflamed sphenoidal tissue. Aggregates of plasma cells are noted. In the inflamed areas, there are collections of fungal hyphae with septations at acute angles and compatible with *Aspergillus* infection.

there is coagulative necrosis and invasion of contiguous structures with scant inflammatory infiltrate.⁵ The chronic form is often seen in less severely immunocompromised patients, such as diabetes, steroid therapy, and HIV/AIDS. Again sinusitis and bone erosion are seen. On pathology, there is an infiltrative mass with mixed inflammatory infiltrate, inflammatory necrosis, and invasion of contiguous structures.⁵ The chronic granulomatous form presents with sinusitis, bone erosion, and contiguous spread. There is an infiltrative mass on pathology with granulomatous inflammation. This form is seen in immunocompetent patients from Sudan, Middle East, and the Indian subcontinent.⁵ Sinus aspergilloma or fungal ball presents with maxillary sinusitis but no bone destruction or invasion. These aspergilloma form in older patients with prior sinus abnormalities.⁵ Our patient, although immunocompetent, had the chronic invasive form of Aspergillosis. Although she had chronic hepatitis C, she did not demonstrate any clinical or laboratory immunosuppression as a result of it. To escape viral clearance and develop chronic infection, the hepatitis C virus (HCV) likely alters the host immune response to the HCV itself.⁶ Whether this alteration in the host immune response to the HCV affects the host immune response to other unrelated pathogens is uncertain. In our patient, she appeared to have a normal immune system by laboratory testing.

Petroapical involvement, considered a form of central nervous system aspergillosis, is rare and has only previously been described in association with *Aspergillus otitis* media, sinusitis, and as a long-term complication of apical cholesterol granuloma.^{1,2,4} Petrous apex aspergillosis usually presents with cranial nerve palsy, trigeminal neuralgia, and orbital pain.^{7,8} Involvement of the petrous apex is thought to be due to movement of the fungus via air conduits between the apex and the base of the petrous bone, due to direct invasion from sphenoidal sinus disease, or via vascular channels and fascial planes. In this

case, it was thought to arise from sinus disease, although only minor soft tissue was ever imaged in the sinus and there was never any clinical episode of sinusitis. There is usually a history of and imaging findings of sinus disease in craniocerebral aspergillosis.⁹

Invasion of surrounding bone results in osteomyelitis. Vascular invasion results in cerebral vasculitis and aneurysms. In our case, there was vascular involvement as evident by absence of flow in the internal carotid artery. Vascular invasiveness is thought to be due to the ability of the fungus to breakdown elastin by elastase production. *Aspergillus* strains that produce elastase have been shown to be of greater virulence and result in greater mortality rates in animal studies. This may result in greater clinical aggressiveness as most effective barriers to spread, including arterial walls, contain significant elastin. Hyphal intramural growth undermines vascular structural integrity and typically results in fusiform aneurysm formation, with a tendency to affect longer, proximal segments of internal carotid artery. Rupture of these aneurysms may result in fatal subarachnoid hemorrhage.^{10,11}

Petrous apex and Meckel's cave aspergillosis is extremely rare in immunocompetent patients and potentially fatal. Vigilance is needed by clinicians because management should be aggressive with combined medical and surgical intervention.^{12,13}

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