Endonasal Ethmoidectomy and Bifrontal Craniotomy with Craniofacial Approach for Resection of Frontoethmoidal Osteoma Causing Tension Pneumocephalus

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ABSTRACT

Tension pneumocephalus is an unusual, potentially life-threatening complication of frontal fossa tumors. We present an uncommon case of a frontoethmoidal osteoma causing a tension pneumocephalus and neurological deterioration prompting a combined endonasal ethmoidectomy and bifrontal craniotomy with craniofacial approach for resection. A 68-year-old man presented with a 1-week history of worsening headache, slowness of speech, and increasing confusion. Standard computed tomography scan revealed a marked tension pneumocephalus with ventricular air and 1-cm midline shift to the right. Further studies showed a calcified left ethmoid mass and a left anterior cranial-base defect. A team composed of neurosurgery and otolaryngology performed a combined endonasal ethmoidectomy and bifrontal craniotomy with craniofacial approach to resect a large frontoethmoid bony tumor. No abscess or mucocele was identified. The skull base defect was repaired with the aid of a transnasal endoscopy, a titanium mesh, and a pedunculated pericranial flap. Postoperatively, the pneumocephalus and the patient's symptoms completely resolved. Pathology was consistent with a benign osteoma. This is an uncommon case of a frontoethmoidal osteoma associated with tension pneumocephalus. Recognition of this entity and timely diagnosis and treatment, consisting of an endonasal ethmoidectomy and a bifrontal craniotomy with craniofacial approach, may prevent potential life-threatening complications.

KEYWORDS: Bifrontal craniotomy, craniofacial, endonasal ethmoidectomy, frontoethmoidal osteoma, tension pneumocephalus

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Osteomas are benign neoplasms of the paranasal sinuses, which are slow growing and are frequently diagnosed incidentally. Their incidence has been reported to be from 0.01% to 0.43%.¹⁻⁵ As many as 50% of patients with osteomas develop mucocoele, however, intracranial extension is very uncommon.⁶⁻¹⁶ Therefore, patients rarely present with neurologic manifestations. Associated brain abscesses have been reported to occur in less than 1% of the cases.^{4,15,17-20} Numerous cases of pneumocephalus associated with osteomas also have been reported.^{16,21} Although frontoethmoidal sinus osteoma presenting with tension pneumocephalus has been previously described,²²⁻²⁶ a combined endonasal and craniofacial approach has hitherto not been described.

In this article we describe an uncommon case of a frontoethmoid osteoma presenting with a tension pneumocephalus. It is important to recognize that a tension pneumocephalus, a potentially lethal yet highly treatable phenomenon, can develop from benign extracranial anterior skull base lesions and can be successfully treated with an endonasal ethmoidectomy and a bifrontal craniotomy with craniofacial approach.

CASE REPORT

A 68-year-old man with a past medical history of hypertension and prostate carcinoma presented with a 1-week history of worsening headache, progressive slowness of speech, and increasing confusion. He denied fevers, chills, nausea, photophobia, nuchal rigidity, rhinorrhea, or traumatic injury. On presentation, the patient had become lethargic but arousable. His speech was slow but fluent. Repetition, naming, and memory testing were intact. There were no cranial nerve deficits. Reflexes, sensation, motor strength, and coordination were normal. Lateral skull radiograph showed air in the lateral ventricle, air beneath the frontal lobe, and a small mass in the posterior ethmoid sinus (Fig. 1A). Noncontrast computed tomography (CT) of his

brain showed pneumocephalus with a disproportionate amount of air beneath the left frontal lobe and ventricular air with midline shift from left to right (Fig. 1B, 1C). Coronal reconstruction CT revealed a partially calcified mass in the left middle and posterior ethmoid sinus. Magnetic resonance imaging (MRI) confirmed these findings, and coronal images showed contiguity between the slightly enhancing paranasal sinus mass and the subfrontal air (Fig. 1D). The patient was admitted to the intensive care unit for fear of potential deterioration and was treated with intravenous steroids and diuretics resulting in improvement of his condition.

The patient was taken to the operating room for a combined craniofacial, subfrontal-transnasal approach by a neurosurgery-otolaryngology team. A bifrontal craniotomy was performed with extradural cranialization of the frontal sinus. Upon entering the cribriform plate, a large dural defect was seen with a large porencephalic cavity. No mucous lining, abscess capsule, or fluid-filled cyst was identified. In addition, a transnasal endoscopy was used to perform an ethmoidectomy. A firm tumor on the left side of the ethmoid encompassing the medial wall of the maxillary sinus was seen (Fig. 2A). Endoscopic maxillary antrostomy was also performed. The transnasal endoscopy was used to free the large base of the tumor from the posterior ethmoidal septum and the superior turbinate. However, the tumor could not be removed endoscopically due to the size and extent of the lesion. Therefore, osteotomes were used to liberate the tumor from the anterior cranial floor and the tumor was removed en bloc.

The floor of the frontal fossa was repaired using a titanium mesh layered with a pedunculated pericranial flap as well as fibrin glue. The dura was repaired primarily. Postoperatively, the tension pneumocephalus resolved, and a lumbar drain was used in the first 3 postoperative days to allow healing of the repair. The patient was treated with a 3-day course of perioperative antibiotics while the lumbar drain was in and he was discharged to home in stable condition after 15 days in the hospital. Pathology revealed a benign mixed cancellous and cortical-type osteoma (Fig. 2).

Figure 1 (A) Lateral skull radiograph shows the air in the lateral ventricle (large arrow) and beneath the frontal lobe (curved arrow). There is also a small mass in the posterior ethmoid sinus (small arrow). (B) Computed tomography (CT) image shows a mass (arrow) 3 cm $\times$ 1.5 cm in the left middle and posterior ethmoid sinus with a focus of dense calcification. (C) On a CT image through the base of frontal lobes (arrow), the upper part of the mass is contiguous with the pneumocephalus (curved arrow). (D) Enhanced coronal magnetic resonance imaging shows the paranasal sinus mass (arrow) and its relationship to the frontal air (curved arrow).

DISCUSSION

Osteoma is a common tumor of the skull and facial bones. Osteomas are very frequent in the paranasal sinuses, especially the frontal sinus, but they also are

found in the ethmoid sinus and more rarely in the sphenoid or maxillary sinuses.^{10,20} The true incidence of skull-based osteomas is not known, due to the indolent nature of these tumors.¹⁵ The reported incidence varies from 0.01% to 0.43%.¹⁻⁵ A history

Figure 2 (A) Intraoperative endoscopic image of the ethmoid sinus lesion. (B) Osteoma-bone interface. The interface between the osteoma (top) and normal bone (bottom) is indicated by the white arrow (hematoxylin and eosin [H & E] stain, $\times 100$). (C) Osteoma. The tumor is located directly beneath the sinus epithelium (white arrow) (H & E stain, $\times 100$). (D) Osteoma. Spicules of bone are seen within a meshwork of loose, fibrous tissue. The bone is rimmed by osteoblasts (white arrows) and an occasional, multinucleated osteoclast (black arrow) (H & E stain, $\times 400$).

of trauma is reported only in 28% of the cases.¹⁰ Up to half of patients with osteoma may develop secondary mucocele or pyocele.^{6–16} Because of their benign nature, intracranial manifestations of these tumors is uncommon. Occasionally, brain abscess and intracranial mucocele are seen together.¹⁵ Although the development of pneumocephalus^{16,21} as well as tension pneumocephalus,^{22–26} in cases of intracranial extension, treated with craniotomy and resection of the lesion has been previously described, a combined endonasal and craniofacial approach has not been described.

The lesion in this patient was composed of an admixture of trabecular and cortical-type bone that

had a composite architecture of both woven and lamellar bone (Fig. 2B–D). Cement lines were prominent focally, giving it a Paget-like appearance. However, the features diagnostic of Paget's disease were lacking. Only rare osteoclasts and focal osteoblastic rimming were seen (Fig. 2D). The latter also makes osteoid osteoma less likely. Also, the architecture of the trabecular bone was inconsistent with fibrous dysplasia. The bony spicules appeared thicker, and there was a focal, disorganized lamellar pattern. Hence, the lesion was classified as a mixed cancellous and cortical-type osteoma.

Since they consist of trabeculae of mature bone, osteomas appears as a radiodense focus on

plain roentgenograms or CT that protrude into the sinus as a smooth, raised mass from the surface of a parent bone. Radiographically, these tumors have distinct characteristics making for a limited differential diagnosis. Pathologically related lesions, such as osteoid osteoma and osteochondroma, are uncommon in the paranasal sinuses and generally have a more complex appearance on cross-sectional imaging with greater heterogeneity. On MRI, these benign tumors may be difficult to detect because, like air within the paranasal sinus, the osteoma itself is hypointense on T1-weighted, T2-weighted, and enhanced T1-weighted images. Occasionally, inflammatory mucosal changes may be seen on T2-weighted or enhanced T1-weighted MRI as linear or globular areas of increased signal intensity around the osteoma. Pneumocephalus, reportedly as small as 0.5 mL of air, can be diagnosed by CT or MRI.

A few case reports describe the development of an intracranial mucocoele and a brain abscess secondary to frontoethmoidal osteoma.^{14,15,23} Brunori et al²³ described a mucocoele resulting from a frontoethmoidal osteoma leading to a hypertensive pneumocephalus during the initial hospitalization. In our case, however, no mucocoele was present. Intracranial air dissected in the left frontal lobe formed a porencephalic cavity. The slowly growing tumor and the defect in the dura contributed to the development of a ball-valve mechanism leading to intracranial air entrapment.^{23,26} We speculate that the air entrance is facilitated by inspiration and the tumor closes the fistula during expiration, coughing, or sneezing. The tumor also prevents cerebrospinal fluid (CSF) rhinorrhea. We assume that the increasing volume of intracranial air eventually dissects through the Virchow-Robin spaces over time creating a porencephalic cavity. Markedly elevated intracranial pressure occurs as air acts as a space-occupying lesion. It is also important to recognize that in our case the patient presented with global mental status changes without a focal neurologic deficit.

We advocate the aggressive endonasal and craniofacial approach for many tumors of the anterior skull base. The principle goal of surgery

is not only to safely resect the lesion, but also to restore integrity to the anterior cranial base with formal reconstruction. We have previously reported on technical issues that aid in managing postoperative complications such as CSF rhinorrhea or pneumocephalus.²⁷ In this case, the anterior cranial fossa was reconstructed, with the aid of endonasal approach, with a titanium mesh as well as a pedicled pericranial flap due to the sheer size of the defect. CSF diversion using lumbar drainage is important in the prevention of postoperative rhinorrhea. The potential for exacerbating a tension pneumocephalus exists with the use of lumbar drainage postoperatively if the cause of the tension pneumocephalus is not addressed by the surgical resection and the skull base reconstruction, especially if the ball-valve mechanism is not eliminated. However, once dura is repaired and the skull base is reconstructed properly, postoperative lumbar drainage for CSF diversion will allow proper healing of the repair and minimize its failure, i.e., CSF leakage.

Repair of the anterior skull base focuses on a watertight closure of dura, separating the nasosinusal space from the cranial contents, and re-establishing cosmetic integrity to the region. There are several methods described in the literature that are employed in cranial base reconstruction.²⁸ Smaller defects are often successfully addressed with local two-layer pericranial or three-layer myofascial galeopericranial flaps. However, larger cranial base defects also require supporting materials in addition to local flaps for a more rigid construct. Sinha et al²⁹ studied 20 patients with extensive defects involving the posterior table of the frontal sinus, cribriform plate, fovea ethmoidalis, planum sphenoidale, and medial orbital roof either unilaterally or bilaterally. These defects were repaired with titanium mesh against the dura with calvarial bone grafts covered with a pericranial flap that was reinforced with fibrin glue at the edges to seal the nasal cavity from the dural repair. With greater than 1-year follow up on 15 patients there were no CSF leaks, hematomas, infections, or exposure of grafts. Badie and associates³⁰ studied the use of titanium mesh

and a pericranial graft without fibrin glue to repair large anterior floor defects. The authors reported a CSF leak rate of 15%.

In conclusion, we report an uncommon case of a frontoethmoidal osteoma associated with a tension pneumocephalus in the absence of mucocoele or brain abscess. Timely diagnosis and aggressive treatment of a combined neurosurgery-otolaryngology approach may prevent potentially life-threatening complications.

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