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Case report

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Abstract

We report a case of intraparenchymal pneumocephalus caused by ethmoid sinus osteoma in a 57-year-old man who had suffered from severe allergic rhinitis because of which he had to frequently blow his nose. He was referred to our hospital for headache and mild left hemiparesis. A computed tomography scan revealed a large intraparenchymal air entrapment in the right frontal lobe related to an osteoma in the ethmoid sinus. The osteoma eroded the upper wall of the sinus and extended into the anterior cranial fossa. During the operation, we noticed that the osteoma had protruded intracranially through the skull base, disrupted the dura, and extended toward the frontal lobe. To the best of our knowledge, this is the first case of intraparenchymal pneumocephalus caused by an ethmoid sinus osteoma.

KEY WORDS · pneumocephalus · osteoma · ethmoid sinus · intraparenchymal
1. Introduction

Osteomas are benign, slow-growing bone tumors. They are often asymptomatic in their early stage, but may occasionally cause serious complications. In this report, we present a case of intraparenchymal pneumocephalus caused by an ethmoid sinus osteoma.

2. Case report

A 57-year-old man was suffering from headache and mild left hemiparesis 2 days before he was admitted to our hospital. Computed tomography (CT) scans revealed a large air entrapment in the right frontal lobe (Fig. 1A) and a bony mass in the right ethmoid sinus (Fig. 1B). The osseous mass (diameter, approximately 2 cm) that extended into the right anterior cranial fossa was located close to the intraparenchymal air entrapment (Fig. 1C). The patient had no history of craniofacial trauma or surgery. He suffered from severe allergic rhinitis because of which he had to frequently blow his nose for over 2 weeks. The patient was prohibited from blowing his nose after hospitalization. X-ray images obtained repeatedly over a period of time revealed a gradual decrease in the size of the air entrapment; however, accidental blowing of the nose by the patient resulted in an influx of air, and as a result, the size of air entrapment increased to that observed on admission.

A right frontal craniotomy was carried out using the subfrontal approach. During the operation, we noticed that the bony mass in the ethmoid sinus eroded the upper sinus wall, breached the dura, and had extended to the frontal lobe. Small holes were observed in the sinus wall surrounding the mass. After the partial removal of the ossifying tumor tissue, which was located above the sinus wall, the dural opening was primarily sutured and covered by a periosteal graft. The defect in the upper wall of the ethmoid sinus was repaired with a polyglactin acid sheet (PAS; Neoveil®; Gunze Corp., Kyoto, Japan), fibrin glue, and a vascularized periosteal graft. Histological examinations confirmed the growth of compact bone, a finding consistent with the diagnosis of osteoma (Fig. 2).

The postoperative course was uneventful. A day after the operation, the headache and mild left hemiparesis disappeared. A CT scan obtained 1 month postoperatively showed little residual air in the right frontal lobe (Fig. 1D).
3. Discussion

Osteomas are relatively common, benign, and slow-growing tumors occurring mainly in the paranasal sinuses, particularly the frontal sinus. Osteomas are usually asymptomatic, but may sometimes cause complications such as headache, cerebrospinal fluid (CSF) fistula, meningitis, ptosis, diplopia, and pneumocephalus. Pneumocephalus is a rare but frequently reported complication of frontal sinus osteomas. A majority of these reports have described air entrapment within the subdural cavity. However, intraparenchymal pneumocephalus caused by an ethmoid sinus osteoma has not been reported thus far.

Two theories have proposed the mechanism underlying intracranial pneumocephalus caused by an osteoma. One of these theories—the inverted pop-bottle mechanism—states that leakage of the CSF through a dural or an arachnoid defect leads to a siphoning effect wherein the air is drawn up into the low-pressure intracranial space. The other theory—the ball-valve mechanism—states that the breached dura acts as a simple flap valve when the extracranial pressure exceeds the intracranial pressure. Our case conforms to the latter theory. In our patient, blowing of the nose increased the extracranial pressure and led to an influx of air into the brain. We presume that air was further injected into the brain parenchyma without any air leakage into the subdural space since the dura was firmly adherent to the pia mater owing to the gravity of the brain.

The management of paranasal osteoma is controversial; however, most agree that surgical intervention is indicated in symptomatic cases or in the case of rapidly growing osteomas. The surgical approaches followed are external craniotomy, endoscopic transnasal craniotomy, or combined craniotomy. We chose craniotomy in order to accomplish complete restoration of the dural defect. Whether osteomas should be resected totally or partially is debatable. We resected only the part of the osteoma that was localized above the sinus wall. No recurrence has been reported in cases of partial and total resection. However, Koivunen and colleagues reported that the average growth rate of paranasal sinus osteomas was 1.61 mm/yr. We have been regularly monitoring the size of the residual osteoma in our patient.

Previous reports have mentioned the use of galea, fascia, muscle, fat, periosteum, and bone wax for the repair of the dura and bone defect, but we used fibrin glue, periosteal graft, and PAS, which is an absorptive, non-woven, loose, and highly elastic sheet. The efficacy of using PAS in combination with fibrin glue to repair the surgical defect has been evaluated in some reports.
Johnson and colleagues reported a case of frontal sinus osteoma causing intraparenchymal pneumocephalus. They observed air leakage from the surface of the frontal cortex while operating. During our surgery, neither did we consciously look out for air leakage nor did we observe the same. The patient’s clinical course during which a reduction in the size of air entrapment was noticed when he was prohibited from blowing his nose, indicated that closure of communication between the ethmoid sinus and the brain parenchyma is a primary step for curing the pneumocephalus. CT scans of the patient obtained during follow-ups revealed a gradual decrease in the intracranial air, and the CT scan obtained 1 month postoperatively revealed that only some air remained in the frontal lobe. We think that venting the intraparenchymal air is not essential to prevent the recurrence of pneumocephalus.

In this report, we have presented a rare case of intraparenchymal pneumocephalus caused by an ethmoid sinus osteoma. Surgical intervention was found to be very useful, and the patient was cured. It is important to be aware of the possibility of an osteoma to occasionally lead to intracranial complications.
Reference

Figure Legends

Fig. 1. A: A computed tomography (CT) scan demonstrating an enormous air entrapment in the right frontal lobe. B: A CT scan showing a bone window with the osteoma (diameter, approximately 2 cm) in the right ethmoid sinus. C: A sagittal reconstructed CT scan demonstrating the ethmoid sinus osteoma, located adjacent to the large air entrapment and penetrating the upper sinus wall. D: A CT scan obtained 1 month after the operation showing only some residual air in the right frontal lobe.

Fig. 2. A photomicrograph showing a section of the ossifying tumor tissue. The lesion was diagnosed as an osteoma. Hematoxylin and eosin (H&E) staining.

Fig. 3. A picture showing the mechanism of air influx into the brain parenchyma. The pia mater and dura mater are firmly adherent owing to the weight of the brain. The air escaped through the dura and pia mater into the brain parenchyma.