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Spontaneous pneumocephalus of an otogenic origin

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Presence of air in the cranial cavity may be caused by a number of factors (Table 1), most of which are related to cranial or iatrogenic trauma. According to Stendel and Heckel,[1] 7% to 9% of cranial traumas studied by CT showed intracranial air. When pneumatic cavities of the skull base (middle ear, nasal fossae, and paranasal sinuses) are involved in trauma, intracranial air is more frequent than shown in previous studies.[2] Certain trauma at a distance from the skull may provoke pneumocephalus, as when the thoracic cage and spine are involved.

Whenever there is leakage of cerebrospinal fluid (CSF) from the endocranium toward other cavities, air may enter through the same pathway. Less often, after surgery air is found in the intracranial compartments of air-containing structures anatomically placed far away from the endocranium, as has been described in thoracotomies.[3]

In any case, it seems that cases of pneumocephalus originating from the mastoid bone occur less often than those originating from the nasosinusal region.[4] Another series of possible causes is less common. Some of them are related to traumas, although not directly precipitated by them. Thus pneumocephalus has been reported after placement of a nasogastric tube[5] or after facemask ventilation in cranioencephalic trauma.[6] It has also been described after use of continuous positive airway pressure in treatment of sleep apnea syndrome,[7] as well as after epidural anesthesia or after nasotracheal intubation.

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In some cases pneumocephalus may occur without a history of trauma (such as that occurring in relation to expansive processes originating in or around the pneumatic cavities). In these cases either physical therapies[4] or bone erosion may be responsible for allowing communication between the pneumatic cavities and the endocranium. Thus we have pneumocephalus associated with paranasal sinus tumors, which histologically may be either malignant or benign. Other cases are caused by processes associated with anatomic malformations located in the pneumatic cavities.[8,9] Another type of pneumocephalus is formed by acute intracranial infectious processes caused by anaerobic micro-organisms or chronic infectious processes of neighboring areas—mostly chronic otitis media.[10] Finally, in rare cases air appears in the endocranium with none of the previously described predisposing factors. These cases are properly termed *spontaneous pneumocephalus* and include those of both otogenic[11,12] and systemic origin (barotrauma).

CASE REPORT

A 20-year-old man was referred from another center to the neurosurgery department of our hospital for treatment of pneumocephalus of unknown cause. Symptoms included continuous headaches, fronto-occipitally located, with a paroxysmic course and no specific time pattern (ie, occurring predominantly during the day or night). No chronic infectious processes or previous cranial traumas were reported. Notably, sinusitis had been diagnosed in this patient, and frequent Valsalva maneuvers had been prescribed.

Neurologic examination revealed absence of venous pulse and blurring of the nasal margin optic disc of his left eye. No CSF rhinorrhea or otorrhea was observed, nor was spontaneous nystagmus with provoking postural or Valsalva maneuvers. Findings during the remainder of the physical examination were normal.

In the first imaging study, a simple skull radiograph in the anteroposterior and lateral views showed an important air

content in the right temporo-occipital area that did not change with posture. Also noted was a well-developed pneumatization of the mastoid bone involving the squamous part of the occipital bone. A CT scan showed the intimate relationship between air content and the posterosuperior facet of right petrous bone and homolateral hyperpneumatized mastoid cells (Fig 1). MRI showed the extradural location of air content with detachment and medial displacement of the sigmoid sinus with respect to the internal bone table.

After being studied in the neurosurgery department, the patient was referred to the ENT department for otologic assessment and study of a possible otic fistula. Routine ear, nose, and throat examination was normal, including liminal-tone audiometry, impedancimetry, and bilateral thresholds of stapes reflex. Hyperpressure maneuvers to demonstrated CSF leakage did not demonstrate CSF otorrhea or rhinorrhea. Findings of other complementary studies were all in the normal range.

The patient remained in the hospital for 9 days so that the course of his pneumocephalus could be monitored. Far from resorbing, the pneumocephalus maintained its initial size and location; therefore surgical treatment was indicated.

The goal of surgery was to seal the communication between the mastoid bone and the endocranium. A right mastoidectomy with preservation of the posterior wall of the external auditory canal was performed. This approach provided the necessary exposure while saving the middle ear so as to preserve the patient's hearing. Drilling was guided by classic structures, by numerous bubbles produced by pneumocephalus content when in contact with cleaning saline solution, and by imaging study. Imaging studies demonstrated a direct relationship between air content and the medial wall of the mastoid cells (and not with tegmen tympani or with tegmen antri). Thus communication between the mastoid bone and endocranium was located in the theoretic point of Citelli's angle. This done, a seal was made with a mixture of bone powder and Tissucol (Immuno G., Vienna, Austria), and the closure was reinforced by a sheet of external cortical bone (2.5 × 1 cm) obtained before drilling of the mastoid bone.

A week after surgery, a control CT study was done and showed absence of air, expansion of the encephalon, and the very small old cavity filled with liquid (Fig 2). A month after surgery, this finding had disappeared, revealing normal endocranial architecture. The patient was asymptomatic, and his hearing was similar to that before surgery.

DISCUSSION

In 1884 Chiari made the first diagnosis (postmortem) of pneumocephalus occurring in a patient with ethmoiditis, caused by doing a forced Valsalva maneuver. In 1913 Luckett became the first person to radiologically demonstrate the pathologic existence of intracranial air in an alive human being. The first case report of otogenic pneumocephalus was thanks to Duken, who in 1915 reported the presence of air in the encephalon of a patient with a bullet injury in the mastoid region. Since then, many authors have reported different events that may cause pneumocephalus (middle ear chronic otitis, 30%; petrous bone fractures, 36%; middle ear surgery, 30%).[13] All of them have in common the existence of some traumatic, iatrogenic, or pathologic antecedent justifying such an entity.

Table 1. Etiologic classification of pneumocephalus

Traumatic pneumocephalus
1. After cranial fractures (sinusal, orbital, and temporal fractures)
2. Iatrogenic
• With preceding trauma (intubation, resuscitation maneuvers, placement of nasogastric tubes, etc)
• With no preceding trauma (after radiation, after locoregional surgery [otic or nasosinusal], after surgery at a distance [thoracotomies, lumbar punctures], with use of CPAP devices)
Tumoral pneumocephalus
1. Malignant tumors (nasosinusal, otic, or orbital)
2. Benign tumors (nasosinusal, otic, or orbital)
Infectious pneumocephalus
1. Infection of pneumatized cavities of the skull base
2. Intracranial infection by gas-producing micro-organisms
Spontaneous pneumocephalus
1. Associated with malformations (meningoencephalocele)
2. Not associated with malformations
• Otogenic origin
• Sinusal origin

CPAP, Continuous positive airway pressure.

In this case no pathologic antecedent explained the presence of air in the intracranial compartment except for doing Valsalva maneuvers in a forced and maintained way. For that reason, although the origin is otogenic, we consider the genesis of this clinical picture to be a spontaneous one. This makes it different from most cases of pneumocephalus reported in literature, in which the main cause is trauma, followed by locoregional or distant surgical management and local expansive or infectious processes.

Some unusual cases of spontaneous pneumocephalus of otogenic origin have been described,[4,13] but in the literature to date, we have found only a single case report of pneumocephalus provoked by Valsalva maneuvers in the same manner as ours.[11]

Anatomic Justification

In 1[4] of 4 reported cases of spontaneous pneumocephalus of otogenic origin, the patient was treated with radiotherapy (30 Gy) for a solid small-cell tumor, which could have caused radionecrosis of the temporal bone with posterior development of pneumatocele; another case[13] was caused by a sudden change in atmospheric pressure because the patient practiced acrobatic free fall with rapid pressure changes, which pro-

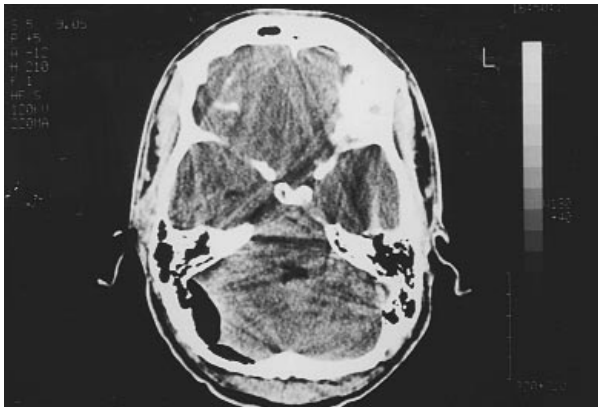

Fig 1. CT scan showing the intimate relationship between air content and posterosuperior facet of right petrous bone; homolateral hyperpneumatized mastoid cells are also shown.

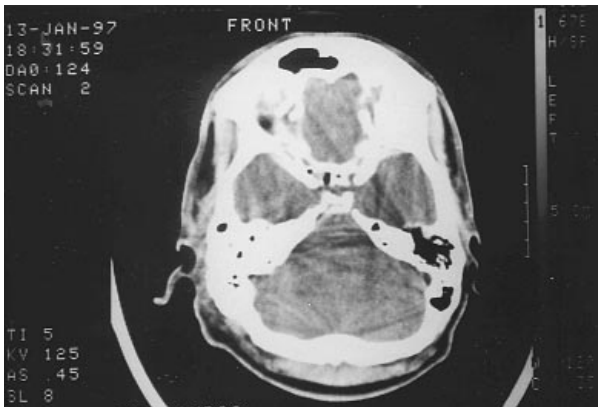

Fig 2. CT scan showing absence of air and expansion of encephalon 1 week after surgery.

voked entrance of air in the endocranium proceeding from the paranasal sinuses. Maier et al[11] described air inside the cranium caused by forced Valsalva maneuvers in a patient with a very well developed mastoid pneumatization. Madeira and Summers[14] described a patient with extradural pneumocephalus located in the sinodural angle region, for which the only antecedent was violent nose blowing. Finally, Keller et al[15] showed a case of spontaneous pneumocephalus resulting from an idiopathic bony defect in the mastoid cavity.

In these last 3 cases, similar to ours, air collection was located in the parieto-occipital region, and its placement was not dependent on head position because the air was not free but was instead contained in a space limited by dura and bone. The parieto-occipital extradural location of air is typical of pneumocephalus of an otogenic origin, usually being traumatic or caused by congenital defects of tegmen tympani.[8-10] This is because of the great adherence existing between bone and dura at that level.

In our case communication between the ear and endocranium was established in the retrosigmoid cells close to the occipital cells, the same as in cases described by Maier et al[11] and Madeira and Summers.[14] Hyperpneumatization predisposes a patient to pneumocephalus because it implies thinning (sometimes extreme) of intercellular septa, thus facilitating its rupture with maneuvers of endotympanic hyperpressure. The distribution and degree of pneumatization achieved by mastoid cells were studied by Dietzel,[16] who pointed to an increase in mastoid pneumatization in the last 80 years. During surgery we observed the thinness of cellulary septa and its extreme fragility.

Physiologic Justification

There are, in general, 2 mechanisms by which we may find air inside the cranium. The first requires a previous leakage of CSF with consequent development of a relatively negative intracranial pressure, which would suction air inside to balance pressures. Air collection is generally distributed along the cisternae and leptomeningeal space.

The second mechanism, corresponding to the case we are concerned with, consists of a valve mechanism that allows air passage from pneumatic cavities toward the endocranium in the presence of positive endotympanic pressures. The valve is then closed when endocranial pressure increases above the pneumatic cavity pressure, with air ending up trapped; this produces an intracranial but extradural tension air collection with a consequent raised intracranial pressure.

A substantial difference with the first described pathogenic mechanism is that air collection becomes located—in the absence of dura breakage—in the extradural space, a normally virtual one, because of the difficulty of detachment of this membrane from bone. This implies on the one hand that air collection locates contiguous to valvular communication and on the other hand that pneumocephalus does not change in its position with posture of the head.

Diagnosis

As in a more extensive series of otogenic pneumocephalus,[10] in our case the first reported symptom was headache. Diagnosis of this entity may go unnoticed by the otoneurologist during routine examination. Complementary image studies—mostly CT—and collaboration with neurosurgeons are required for diagnosis.[10]

Clinical features presented by spontaneous pneumocephalus of an otogenic origin are generally scarce, with headache being its main symptom. This contrasts with clinical symptoms in the remaining types of pneumocephalus, which are much more florid (agitation, delirium, clouding of consciousness, otorrhea, and otorrhagia).

Treatment

Treatment of tension pneumocephalus of whatever cause requires surgery in an attempt to relieve intracranial pressure. The aim of surgery is to identify communication between the pneumatic cavity (mastoid bone in our case) and endocranium and to seal it, generally by use of autologous materials (cartilage, free fascia, temporal muscle, and bone powder).

CONCLUSION

In patients with previous or current symptoms of raised intracranial pressure and known hyperpneumatization of mastoid air cells, physicians should consider spontaneous pneumocephalus of otogenic origin as a possible cause.

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