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Clinical Record

Pneumocephalus as a delayed mastoidectomy complication

F Jégoux, B Godey, L Riffaud*, X Morandi*

Abstract

We report the case of a 43-year-old woman who presented with a spontaneous pneumocephalus, 37 years after a mastoidectomy. Clinical examination showed a cerebrospinal fluid leak, meningeal herniation in the superior part of the middle ear, and an audible noise from her ear when she stood up due to the entrance of air into the cranium. A computed tomography scan and magnetic resonance imaging showed the complete destruction of the tegmen tympani and the pneumocephalus in the temporal lobe. The patient underwent an emergency operation via a double middle-ear and subtemporal approach. The meningoencephalocoele and pneumocephalus were probably due to long term pressure upon too thin a tegmen tympani.

Pneumocephalus should be considered as a potential delayed post-operative complication of middle-ear surgery. Computed tomography and magnetic resonance imaging scanning supply accurate information and enable a planned surgical approach; they also allow a pathophysiological understanding and a correlation between the clinical signs and the radiological and peri-operative findings.

Key words: Pneumocephalus; Meningocoele; Encephalocoele; Mastoid; Surgical Procedures, Operative; Treatment Complications; Diagnostic Imaging

Introduction

The presence of air in the cranium is a frequent occurrence after head trauma or neurosurgery.[1] Pneumocephalus can also be due to a tumoral or infectious disease of the skull base.[2] Several cases have been described after otologic surgery; in these cases, pneumocephalus was diagnosed a few days after the surgical procedure and considered to be an acute complication.[3] Spontaneous cases were also described in patients with congenital bone defects of the tegmen tympani.[1,4–8]

We report the case of a woman who presented with a spontaneous pneumocephalus in the temporal lobe, 37 years after a mastoidectomy. To the best of our knowledge, this is the first case of pneumocephalus occurring such a long time after a mastoidectomy. We describe the clinical, computed tomography (CT) and magnetic resonance imaging (MRI) signs, their correlation with peri-operative findings, and the mechanisms that could have led to the clinical presentation.

Case report

A 43-year-old woman was admitted for headache and vomiting. She had undergone a mastoidectomy for mastoiditis when she was six years old; she had not experienced any otological sequelae except for moderate hearing loss on the right. She had no other medical history and, in particular, no recent head trauma or middle-ear infection. One week previously, she had felt faint and was found to have otalgia, clear otorrhoea and headache, without fever. Remaining upright was difficult because of an intense headache.

On presentation, the patient was fully alert. No facial palsy or vestibular or cerebellar syndrome was noted. During clinical examination, an objective, strange 'bubbling' sound was heard at the patient's ear when she stood up, followed seconds later by acute headache and loss of consciousness for a few seconds. When the patient was supine, profuse cerebrospinal fluid (CSF) otorrhoea was evident before she regained consciousness.

Ear examination showed that the patient had undergone an open surgical procedure. The inferior part of the tympanic membrane was retracted on the promontory, and the hypotympanum and the superior part of the tympanic cavity were filled with a soft, whitish mass.

An audiogram showed a 40 dB conductive hearing loss on the right, while hearing was normal on the left.

A right petrous bone CT scan showed that the roof of the tympanic cavity was completely eroded from the squamous part of the temporal bone to the inner ear. The superior part of the right middle ear was occupied by an opacity. There was an intracerebral air pouch in the right temporal lobe (Figure 1). An MRI scan confirmed these findings, showing pneumocephalus in the temporal lobe (Figure 2a), which was in contact with the inferior horn of the lateral ventricle (Figure 2b). The MRI also revealed continuity between the temporal cerebral tissue and the middle-ear opacity.

The working diagnosis was a pneumocephalus secondary to bone destruction by a middle-ear cholesteatoma.

Emergency surgery was performed. A combined middle-ear and middle cranial fossa approach provided good exposure of the tympanic cavity and the temporal lobe. Middle-ear exploration showed dura and cerebral tissue under a thin squamous layer without cholesteatoma.

From the Departments of Otorhinolaryngology and *Neurosurgery, Centre Hospitalier et Universitaire Pontchaillou, Rennes, France.
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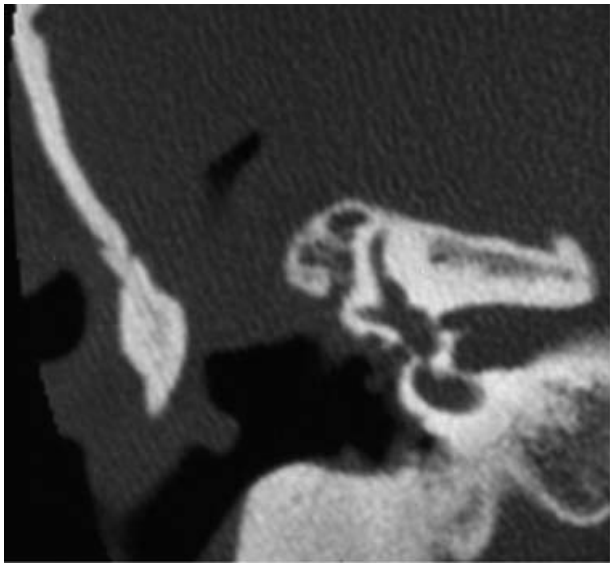

FIG. 1

Temporal bone computed tomography scan in coronal section. Extensive tegmental destruction is seen, with opacity of the superior part of the mastoid cavity. Pneumocephalus is clearly shown in the temporal lobe.

The subtemporal approach allowed this encephalocoele to be replaced above the petrous bone. A dural defect was noted and an intracerebral fistulous tract was found extending from the meningeal defect to the lateral horn of the ventricle. The pneumocephalus was drained, the fistulous tract was closed, the defect in the dura was sutured, and a bone fragment was placed on the superior face of the temporal bone upon the middle ear to support the temporal lobe and to avoid recurrence of the encephalocoele.

The patient's neurological status improved. The day after the surgical procedure, her level of consciousness was normal. No recurrence of headache or otorrhoea was noted. She was discharged home after seven days.

Two months after the surgical procedure, a cerebral MRI confirmed the absence of encephalocoele and pneumocephalus. One year later, the otologic and neurologic examinations were satisfactory and the patient was free of symptoms.

Discussion

To our knowledge, this is the first case of delayed, acquired, post-operative pneumocephalus related to middle-ear surgery. Markham[1] has extensively reviewed this topic and published a series of 295 cases. Pneumocephalus can be due to trauma, neoplasm, infection, surgical intervention or idiopathic causes. The bone barrier between the dura and the middle ear, the thickness of the tegmen tympani, and the number and the size of the temporal bone air cells vary greatly between individuals.[9] Spontaneous otogenic intracerebral pneumocephalus is often due to a congenital defect,[1,4–8,10,11] but several cases without a defect have been described. Richards *et al.*[12] reported two cases in which spontaneous entry of air into the head appeared to have occurred via a hypercellular mastoid air cell system, without any congenital tegmen defect. In both these cases, forceful sneezing and nose-blowing were considered to be contributory factors.

Otogenic pneumocephalus can be considered as an immediate complication in middle-ear surgery when the dura is opened, whether accidentally or deliberately. Several cases have been reported following mastoidectomy, all occurring a short time after the intervention.[1–4,13–15]

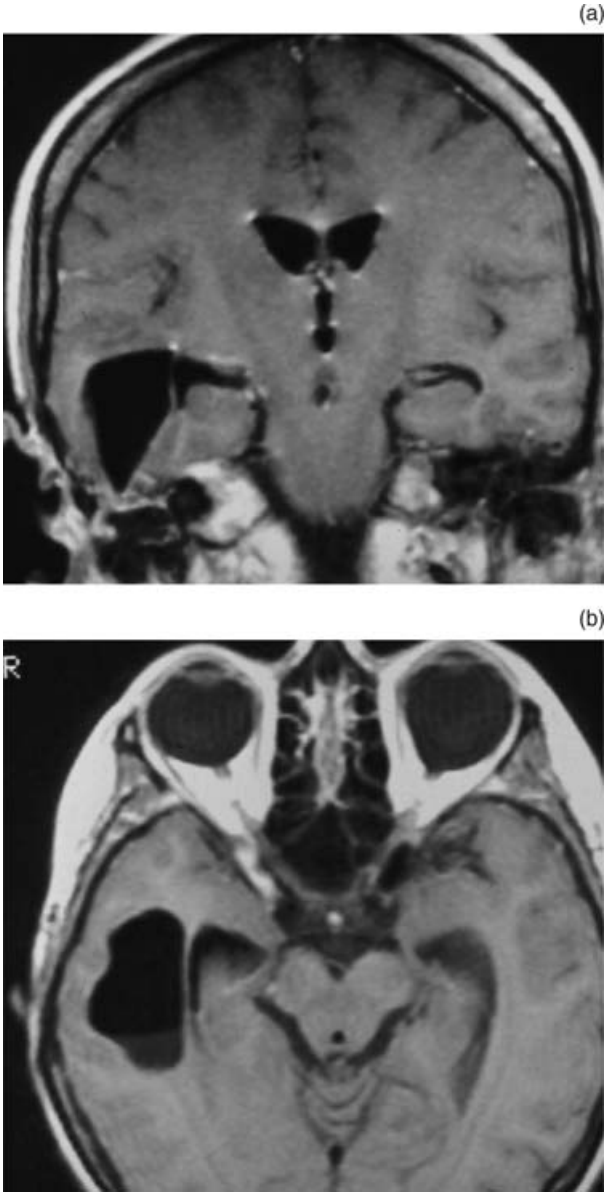

FIG. 2

(a) Magnetic resonance imaging, enhanced T1 coronal section, showing pneumocephalus and encephalocoele. (b) Magnetic resonance imaging, enhanced T1 axial section, showing pneumocephalus with fluid level close to inferior horn of lateral ventricle.

Delayed post-operative, otogenic pneumocephalus has not yet been described.

The development of pneumocephalus requires two conditions: firstly, a break in the bone and in the dura, and secondly, a pressure gradient that allows the entry of the air into the cranium.

In the presented case, the break in the dura was not due to a surgical injury but resulted from brain herniation. Post-surgical meningoencephalocoele into the middle ear is well known.[14,15] Bone injury occurs most often during initial entry into the mastoid, or in the tegmen tympani during drilling between the postero-superior external auditory canal and the middle fossa.[15] In the presented case, a congenital defect would have been diagnosed at the first procedure. It is unlikely that complete tegmental destruction was due to the first procedure drilling. This progressive lysis can be explained by contact with the

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pathological mucosa of the mastoid, and/or the weight of the temporal lobe upon the tegmen tympani thinned by the surgical procedure.

- **This is a case report of pneumocephalus occurring as a delayed complication of mastoidectomy**
- **The role of imaging is highlighted**
- **The pathophysiology and clinical and peri-operative findings are discussed**

The pressure gradient can be explained either by episodes of increased pressure in the middle ear, or by a negative intracranial pressure. In some spontaneous cases, an episode of elevation of the air pressure in the middle ear occurred, such as nose-blowing, sneezing, air travel, scuba diving, valsalva manoeuvre or bouts of coughing.[5,6,8,12] Pneumocephalus has also been described after use of continuous positive airway pressure in the treatment of sleep apnoea syndrome.[16] Several mechanisms can explain a decrease in the intracranial pressure. Horowitz[13] described a mechanism whereby the intracranial volume is an absolute constant, and variations in one of its components (brain, blood or CSF) must be compensated for by variation in another component. A decrease in intracranial pressure could result from adopting the upright position. In the presence of a fistulous connection across the dura to an aerated cavity, the loss of CSF could also provoke a low intracerebral pressure, and air may enter the intracranial space in response to the negative pressure gradient. The present case was particularly striking on this point. The patient was well when she was supine. When she stood up, the intracranial pressure decreased, allowing the audible entry of air into the cranium, with an objective bubbling noise. She then felt faint and became rapidly supine because of headache. In the supine position, the intracranial pressure increased and a profuse otorrhoea allowed a decrease in the intracerebral volume, equalising the pressures.

This complication must be treated as an emergency because of the neurological risks due to intracranial pressure variations and infectious complications. The surgical procedure consists of closing the intracerebral fistulous tract, the break in the dura and the bony defect.

Excessive drilling of the tegmen tympani and antri leads to reduced thickness, and this may be the first pathophysiological step in the development of otogenic pneumocephalus; it should therefore be avoided when performing a mastoidectomy.

Conclusion

Pneumocephalus should be considered as a potential delayed post-operative complication of middle-ear surgery. Imaging gives accurate information to inform the choice of surgical approach. In the case presented, imaging also enabled pathophysiological understanding and correlation between clinical signs and peri-operative findings.

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Address for correspondence:
Dr Franck Jégoux,
Service ORL et Chirurgie Maxillo-Faciale,
CHU Pontchaillou,
Rue Henri Le Guillou,
35037 Rennes, Cedex 9, France.

E-mail: franck.jegoux@chu-rennes.fr

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