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Case Report

Spontaneous intraparenchymal otogenic pneumocephalus: A case report and review of literature

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Abstract

Background: Pneumocephalus is commonly associated with head and facial trauma, ear infection, or surgical interventions. Spontaneous pneumocephalus caused by a primary defect at the temporal bone level without association with pathological conditions is very rare. Few cases have been published with purely intraparenchymal involvement. We describe a rare case of spontaneous pneumocephalus arising from the mastoid cells with intraparenchymal location and present an extensive review of the existing literature.

Case Description: A 57-year-old woman presented a brief episode of sudden otalgia in her left ear that was followed by a motor aphasia. Imaging revealed a left temporal intraparenchymal pneumocephalus in a close relationship with a highly pneumatized temporal bone. Left temporal craniotomy and decompression were performed. Further subtemporal exploration confirmed a dural defect and other osseous defects in the tegmen tympani, which were both consequently closed watertight.

Conclusion: Although extremely rare, a spontaneous intraparenchymal pneumocephalus with mastoidal origin should be considered as a possible diagnosis in patients with suggestive otological symptoms and other non-specific neurological manifestations. Surgery is indicated to repair bone and dural defects.

Key Words: Intraparenchymal, pneumocele, spontaneous pneumocephalus, tegmen tympani, temporal bone

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INTRODUCTION

Pneumocephalus is defined as the presence of air inside the skull.[20,23,33,37] Its association with craniofacial trauma, neurosurgical or otological procedures,[5,13,20,28,30,35] otomastoiditis,[2] or skull base tumors[9,12] is well known. By contrast, spontaneous pneumocephalus caused by a primary defect at the temporal bone level without association with pathological conditions, as those mentioned above, is very rare and so far 23 cases have been reported [Table 1].[3,4,7,8,10,11,15-18,20,24-27,29,31-34,36,37] Moreover, five cases of exclusively intraparenchymal location of the pneumocephalus have been described in the literature.[4,15,18,29,32]

We report a patient with a large spontaneous otogenic pneumocephalus in the left temporal lobe which was manifested initially by ear pain followed by a sudden

Table 1: Review of spontaneous pneumocephalus in the literature by year

Case	Author/Year	Sex	Age	Location	Clinical presentation	Mechanism	Treatment
1	Jelsma[11] 1954	M	39	Extradural with pneumatocele Right parieto-occipital	Palpable emphysema on back of head, hemianopsia, weakness of left leg and arm, headache, nausea and vomiting, syncope	Spontaneous	Needle aspiration
2	Markham[18] 1967	F	64	Intraventricular and intraparenchymal Left temporal	Headache, nausea, vomiting, expressive aphasia, weakness of right arm	Change in altitude	Repair of dura
3	Madeira[16] 1977	M	57	Extradural Right parieto-occipital	Left homonymous hemianopsia, right ear fullness, headache	Nose blowing	Mastoidectomy, temporalis muscle was rotated into the defect
4	Goldmann[8] 1986	M	26	Subdural Left frontoparietal	Headache after ascent during diving	Scuba diving	Observation Abstain from diving
5	Stavas[32] 1987	M	64	Intraparenchymal Left temporal	Expressive aphasia	Brain irradiation for metastatic small cell carcinoma of the lung, chronic coughing	Mastoidectomy and mastoid air cells were packed Dural repair
6	Spar[31] 1994	F	27	Intraventricular right lateral ventricle	Vomiting, headache, and loss of balance	Spontaneous	Closure with dural patch
7	Maier[17] 1996	M	24	Extradural Right parieto-occipital	Bilateral scotoma, headache	Valsalva's maneuver	Mastoidectomy and obliteration with bone wax and collagen
8	Dowd[7] 1998	F	78	Intraventricular Right and left lateral ventricles	Left hemiparesis, expressive aphasia, headache, ear noise	Spontaneous	Ventriculostomy Dural repair Eustachian tube closure
9	Park[24] 1998	M	49	Extradural Right parieto-temporal	Headache	Air pressure changes during flying, nose blowing	Middle cranial fossa floor blocked with a large pericranial flap
10	Vallejo[34] 1999	M	20	Extradural Right temporo-occipital	Headache, hemianopsia	Valsalva's maneuver	Repair of defect with bone dust and fibrin glue
11	Añorbe[3] 2000	M	27	Extradural with pneumatocele Right parieto-occipital	Retroauricular mass which increased in size on Valsalva's maneuver	Valsalva's maneuver	Communication between antrum and aditus was sealed by hydroxyapatite
12	Bahloul[4] 2003	M	47	Intraparenchymal Left temporal	Aphasia	Spontaneous	Needle puncture of the air collection and duroplasty
13	Schrijver[27] 2003	F	30	Extradural Left temporo-occipital	Asymptomatic	Valsalva's maneuver, chronic coughing	Observation Stop Valsalva's maneuver
14	Richards[26] 2004 *Case 1*	M	17	Extradural with pneumatocele Right temporal	Retroauricular mass, otalgia, bloody discharge	Nose blowing	Closure of defect with bone pate and fibrin glue, obliteration of mastoid with abdominal fat and fascia
15	Richards[26] 2004 *Case 2*	F	50	Extradural Left temporal	Burning pain, paraesthesia of left cheek and lower jaw, reduced sensation of 2nd and 3rd branches of trigeminal nerve	Vigorous nose blowing due allergic rhinitis and nasal polyps	Stop sneezing/blowing Bone defects obliteration with bone pate and fibrin glue, mastoid filled with abdominal fat

Contd...

Table 1: Contd....

Case	Author/Year	Sex	Age	Location	Clinical presentation	Mechanism	Treatment
16	Krayenbühl[15] 2005	M	48	Intraparenchymal Left temporal	Ear noise, expressive aphasia, headache, homonymous hemianopsia	Spontaneous	Aspiration, closure of defects with bone wax, temporalis muscle fascia flap, and fibrin glue
17	Hyam[10] 2008	F	69	Intraventricular Right and left lateral ventricles	Loss of consciousness	Spontaneous	Mastoid air cells were packed with bone wax and dural defects patched with a fascia lata graft
18	Tucker[33] 2008	M	19	Extradural Right temporo-occipital	Headache, nausea	Excessive nose blowing habit	Stop excessive nose blowing habit Pyramid bone defect repaired with vascularized galeal flap
19	Pennings[25] 2009	M	43	Extradural with pneumatocele Right temporo-occipital	Occipital swelling, crepitations in his right ear	Spontaneous	Mastoidectomy and epitympanotomy Bony defects were covered with autologous bone pate and fibrin glue Mastoid was filled with abdominal fat
20	Singh[29] 2010	M	77	Intraparenchymal Left temporal	Sensation of ear congestion, motor aphasia, diminished sensation in his feet	Single Valsalva's maneuver	Craniotomy with surgical decompression and obliteration of the dural fistula
21	Wanna[36] 2010	F	74	Subdural Right posterior fossa	Gradual decline in neurologic status	Spontaneous	Transmastoid retrolabyrinthine approach, evacuation of air collection, repair of the defects with temporalis muscle, bone paste, fascia, abdominal fat and fibrin sealant
22	Mohammed[20] 2011	M	23	Extradural with pneumatocele Left occipito-parietal	Headache, emphysema increased in size by forceful sneezing and nose blowing	Spontaneous	Mastoidectomy, bone defect was sealed with tragal perichondrial–cartilage graft and a temporalis muscle plug
23	Zhao[37] 2011	M	62	Extradural with pneumatocele Left occipital supra-infratentorial	Occipital palpable mass	Spontaneous	Intraaural approach, bone defect was sealed with wax, temporalis fascia flap, and fibrin glue
24	Present Case 2012	F	57	Intraparenchymal Left temporal	Otalgia, headache, motor aphasia	Spontaneous	Temporal craniotomy Aspiration of the air collection Closure of the dura with temporalis muscle fascia and fibrin glue Tegmen tympani bone defects were closed with autologous bone and fibrin glue

motor aphasia. A left temporal craniotomy with decompression of pneumocephalus and obliteration of the fistulous communication between the dura and the temporal bone was performed.

CASE REPORT

A 57-year-old woman was admitted to the hospital with sudden otalgia in her left ear that was noticed by her daughter. Five minutes later, the patient had an acute onset of speaking difficulties and left-sided headache. These symptoms were not precipitated by coughing, straining, or Valsalva's maneuver. There was no history of head trauma, surgery, or ear infection. Neurological examinations revealed solely a motor aphasia and the other neurological functions were normal. The otological examination was completely normal.

The computed tomography (CT) showed a hypodense space-occupying lesion (−1000 Hounsfield units) compatible with air in the white matter of the left temporal lobe, of approximately 4 cm diameter [Figure 1a]. Similar findings were found on plain radiographs of the skull [Figure 1b].

A high-resolution CT using a bone algorithm demonstrated a close relationship of the air bubble with air cells of the mastoid, which were highly pneumatized [Figure 1c]. The temporal bone was fluid free and showed no signs of fracture, acute or chronic inflammatory changes.

Magnetic resonance (MR) showed the large intraparenchymal pneumocephalus in the white matter of the left inferior and middle temporal gyrus with minimal perilesional edema [Figure 1d]. No pathological gadolinium enhancement indicative of an underlying tumor was present.

We performed a left temporal craniotomy for decompression and to seal the presumed temporal bone and dura defects. After opening the dura, the cortex was pressed out and a puncture of pneumocephalus was carried out at the level of the second temporal sulcus, with high-pressure evacuation of the air collection and consequent brain relaxing. Later, an extradural approach to the tegmen tympani showed a 5-mm bone defect in close relationship with an underlying dural hole, through which output of cerebrospinal fluid was observed [Video 1 and Figure 2]. Other six smaller bone defects (2 mm) were also found at the level of tegmen tympani, but without any dural defects.

A watertight closure of the dura with temporalis muscle fascia and fibrin glue was performed. All bone defects were closed with autologous bone and fibrin glue.

After the operation, the patient's symptoms gradually improved, and the further postoperative course was uneventful. The patient was discharged home 5 days after the operation with minimal speech problems, which were treated with speech therapy. The follow-up examination 1 month later revealed normal speech and absence of pneumocephalus on CT and MR [Figure 3].

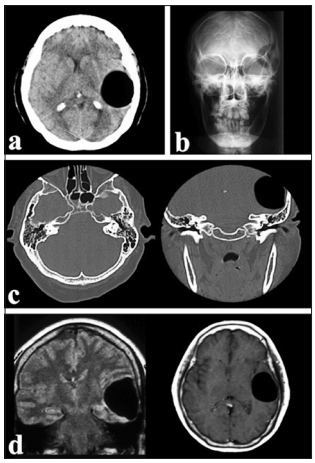

Figure 1: Pre-operative imaging. (a) CT shows a single hypodense lesion in the left temporal lobe with mild mass effect. (b) Plain skull radiograph demonstrating a bubble air. (c) Axial and coronal CT with bone window shows the close relationship of pneumocephalus with a highly pneumatized temporal bone. (d) Coronal FLAIR MR shows the pneumocele in the left inferior and middle temporal gyrus and post-gadolinium T1-weighted axial MR shows no pathological enhancement

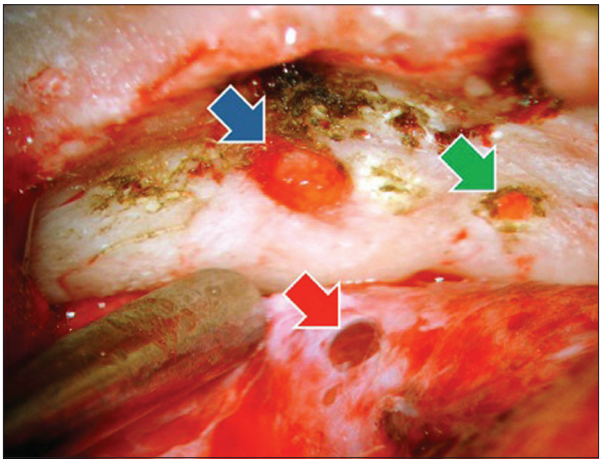

Figure 2: Operative findings. Extradural subtemporal approach to the tegmen tympani area showed a 5-mm bone defect (blue arrow) in close relationship with an adjacent dural defect (red arrow), through which cerebrospinal fluid leak was observed. Other smaller bony defects of about 2 mm (green arrow) were also found, but without any relationship with dural hole

DISCUSSION

The first report of a pneumocephalus was described in 1741 by Lecat.[11,37] In 1884, Chiari first reported pneumocephalus on autopsy of a patient with

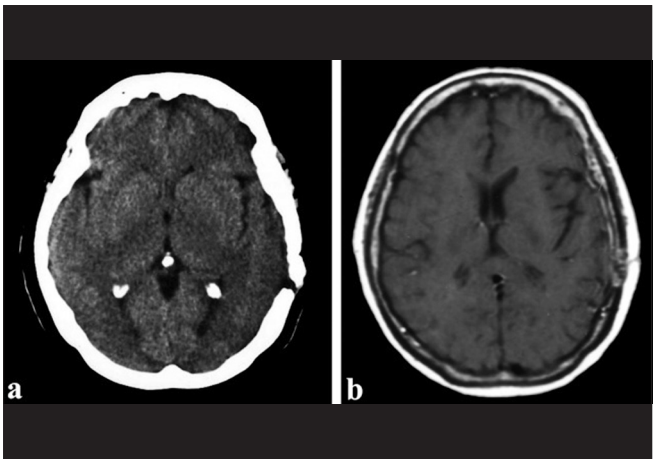

Figure 3: Postoperative imaging. (a) Axial CT and (b) axial post-gadolinium T1-weighted MR show a total resolution of the pneumocephalus

ethmoiditis.[6,15,20,33] The initial evidence of intracranial air was performed by Luckett in 1913 through a skull radiograph in a patient with cranial vault fracture.[15,37] Jelsma[7,11,15] was the first to give a description of subdural spontaneous pneumocephalus in 1954.

Spontaneous pneumocephalus is defined as accumulation of air intracranially (epidural, subdural, subarachnoid, intraventricular, and/or intraparenchymal) without association with craniofacial trauma, otological, or neurological surgery, meningitis, infectious sinus disease, or skull base tumors.[15,25,33,37] The term pneumatocele is defined as subperiosteal accumulation of air and usually causes subcutaneous emphysema or a palpable mass.[25]

Spontaneous otogenic pneumocephalus is very infrequent.[20,25,37] There have been 23 cases reported up to date and just 5 with purely intraparenchymal location.[3,4,7,8,10,11,15-18,20,24-27,29,31-34,36,37]

Although a spontaneous otogenic extradural pneumocephalus is rare, since the dura is tightly adherent to the calvarium, 13 of all the published cases had extradural location.[3,11,16,17,20,24-27,33,34,37] Six of these cases had an association with a pneumatocele, due to cranium bone thinning.[3,11,20,25,26,37]

Locations of the remaining cases were intradural: five intraparenchymal,[4,15,18,29,32] four intraventricular,[7,10,18,31] and two subdural.[8,36] Only in the case described by Markham, pneumocephalus had an intraparenchymal and intraventricular combination.[18]

According to previous reports, headache is the symptom most commonly present, followed by otorrhea, meningeal signs, aphasia, vertigo, facial paralysis, visual changes, vomiting, fainting, paralysis, seizures, and so on.[15,25,33,37]

Left ear otalgia was the first symptom in the present case, prior to the onset of motor aphasia. Only six cases with spontaneous otogenic pneumocephalus manifesting with acoustic phenomena have been reported in the literature. The symptoms were ear fullness sensation,[16,29] ear noise,[7,15] ear crepitations,[25] and otalgia.[26] The case reported here presented with a sudden otalgia prior to aphasia, probably due to excessive distension of tympanic membrane.

Overall, 13 patients had an increase in middle ear pressure evoked by repeated Valsalva's maneuvers, nose blowing, altitude change, chronic coughing, or diving, which were postulated as the mechanisms for developing the pneumocephalus.[3,8,16-18,24,26,27,29,32-34] Medical history revealed that only one of them had an additional possible condition: a whole-brain irradiation for metastasized small cell carcinoma of the lung.[32] The remaining 11 cases did not experience any triggering events.

In principle, for pneumocephalus to occur, a persistent negative intracranial pressure gradient or an extracranial positive pressure source is necessary.[33]

The first possibility is known as the "inverted soda bottle" or "siphon effect" and could be observed in the cases of low intracranial pressure by dural leak or shunt placement, where the CSF is replaced by air.[7,20,27,33] In the absence of these factors, low or negative intracranial pressure occurs due to an excessive loss of CSF either through settling into the distendable spinal subarachnoid space or by simple drainage via normal pathways. However, in the presence of a fistulous connection across the dura to an aerated sinus, air may enter the intracranial space in response to the negative pressure gradient. If the brain substance was tightly adherent to the dura at the fistula site, the air could bypass the extracerebral spaces and penetrate the brain directly in the path of least resistance.[7]

In the second possibility, the mechanism by which the air is trapped is known as "ball valve" described by Dandy.[7,15,20,33,34,37] In this case, in order to produce a spontaneous pneumocephalus, two pathological conditions must coexist. First, a defect in the temporal bone needs to be present to allow communication of air from the mastoid cells to the intracranial compartment. Second, there should be a gradient of pressure between the middle ear and the intracranial space to allow the air to enter the cranium. Valsalva's maneuvers or changes in ambient pressure result in the passage of high pressure air through the fistula. This increases intracranial pressure which leads to the dura and/or brain to quickly obliterate the fistula, allowing the air collected to become trapped.

There is a wide variation among individuals in number, size, and distribution of the air cells in the temporal bone.[14] The middle ear pressure is presumed to play an important role in the extension of the pneumatization. Constantly increased pressure because of Eustachian

tube dysfunction or the habit of performing Valsalva's maneuvers may lead to hyperpneumatization.[22] In addition, defects in the petrous bone might be common in the general population. Ahren[1] described an autopsy series of 94 bony defects at the tympanic roof. Twenty-one percent of the specimens had a single defect and 6% had more than five defects. Our case had a single larger defect (5 mm) and six smaller defects (2 mm).

In order for an extradural pneumocephalus to occur, a condition that allows progressive separation between the dura and the bone must be present, as these structures are often attached. This condition is represented by repeated Valsalva's maneuvers, excessive nose blowing habit, or chronic coughing.[7,15,19,20,33,34,37] This could be observed in 9 out of 13 cases of extradural location of the pneumocephalus.[3,16,17,24,26,27,33,34] In the remaining four cases with similar location, these conditions were not found.[11,20,25,37] Since no defect is present at the dura, the air remains trapped in the epidural space. The pathological adherences are probably responsible for closing the dura at the bony defect, allowing the air to be trapped.

The pneumocephalus with intradural location (subdural, subarachnoid, intraparenchymal, or intraventricular) needs a dural defect in addition to the bony defect. In these cases, arachnoid and the brain closed the fistula by a "ball valve" effect. In none of these cases, a sustained over time condition was found, unlike the extradural location, that could predispose the appearance of pneumocephalus. Only in 3 out of the 10 cases of intradural location, a trigger condition for a sudden increase of pressure was found. These were the case reported by Markham in which the pneumocephalus was associated with an altitude change, the case reported by Goldman which was associated with scuba diving, and the case reported by Singh which was associated with a unique Valsalva's maneuver.[7,18,29] In the remaining seven cases with intradural location and in the one presented here, no suggestive episodes of elevated middle ear pressure could be documented, thus being classified as spontaneous.

In the present case, the absence of extra-axial air can be explained by abrupt direct passage of the air through the point of lowest resistance at the dura–bone interface. Adherences between bone and dura could be explained in some circumstances. Minor head trauma in childhood, which the patient may not recall, could have resulted in bone microfractures, gradual defects in the pyramidal bone, and lacerations of the dura. Other plausible contributing cause of adherences is a history of inflammatory process of the middle ear. Infrequently, faulty embryogenesis may lead to a meningoencephalocele that produces arachnoid granulations and adherences to the tegmen tympani.[33]

In our case, there was no clear anamnestic evidence and the precise underlying mechanism for both abrupt pressure changes and the development of the dural–bone adhesions remains unknown.

The management of spontaneous penumocephalus is the surgical evacuation of the air collection when it is accompanied by signs of intracranial hypertension. In addition, to avoid recurrence and to prevent further infections, it is mandatory to repair the bone defects of the temporal bone (more often at the tegmen tympani area) and the underlying dural holes, if present. Most of the cases were treated by closing the existing fistula using muscle fascia flap, cartilage, and bone wax to seal the communication from the extracranial to the intracranial compartment.[15-17,20,25,26,29,32,33,37] An alternative described by Dowd for cases of recurrence is the closure of the Eustachian tube in order to reduce the pressure inside the middle ear.[7] Only two cases were managed conservatively without surgery.[4,27] In addition, patient education to avoid Valsalva's maneuvers or nose blowing habit can possibly contribute to reduce recurrence.

CONCLUSIONS

Spontaneous otogenic pneumocephalus is a very rare entity. Often, there is a temporal bone defect that allows communication between the middle ear and the cranial cavity. Spontaneous otogenic pneumocephalus should be suspected in patients with sudden onset of otological symptoms and other non-specific neurological manifestations. The diagnosis is even more likely in cases of a highly pneumatized temporal bone and without signs of ear infection or tumor growth. The surgery aims to relieve intracranial pressure and repair the fistula at the temporal bone roof and dura.

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