Case Report

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

Santiago G. Abbati1,2, Rafael R. Torino2

1Department of Neurosurgery, Hospital de Clínicas de Buenos Aires, Ciudad Autónoma de Buenos Aires, Argentina, 2Department of Neurosurgery, Hospital Británico de Buenos Aires, Ciudad Autónoma de Buenos Aires, Argentina

E-mail: *Santiago G. Abbati - sgabbati@gmail.com; Rafael R. Torino - rrtorino@gmail.com
*Corresponding author

Received: 11 December 11 Accepted: 1 February 12 Published: 14 March 12

This article may be cited as:
Abbati SG, Torino RR. Spontaneous intraparenchymal otogenic pneumocephalus: A case report and review of literature. Surg Neurol Int 2012;3:32.
Available FREE in open access from: http://www.surgicalneurologyint.com/text.asp?2012/3/1/32/93861

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

INTRODUCTION

Pneumocephalus is defined as the presence of air inside the skull. Its association with craniofacial trauma, neurosurgical or otological procedures, otomastoiditis, or skull base tumors 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]. Moreover, five cases of exclusively intraparenchymal location of the pneumocephalus have been described in the literature.

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 Right 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 [27] 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 | Venticulostomy 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 temporoparietal | 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 temporoparietal | 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...
| 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[16] 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 |
| 19   | Pennings[25] 2009 | M   | 43  | Extradural with pneumatocele Right temporo-occipital | Occipital swelling, crepitations in his right ear | Spontaneous | Mastoidectomy and epitympanotomy |
| 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].

DISCUSSION

The first report of a pneumocephalus was described in 1741 by Lecat. In 1884, Chiari first reported pneumocephalus on autopsy of a patient with
Only in the case described by Jelsma, 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, meningoitis, infectious sinus disease, or skull base tumors. The term pneumatocele is defined as subperiosteal accumulation of air and usually causes subcutaneous emphysema or a palpable mass.

Spontaneous otogenic pneumocephalus is very infrequent. There have been 23 cases reported up to date and just 5 with purely intraparenchymal location.

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. Six of these cases had an association with a pneumatocele, due to cranial bone thinning.

Locations of the remaining cases were intradural: five intraparenchymal, four intraventricular, and two subdural. Only in the case described by Markham, pneumocephalus had an intraparenchymal and intraventricular combination.

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. 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, ear noise, ear crepitations, and otalgia. The case reported here presented with a sudden otalgia prior to aphasia, probably due to excessive distension of tympanic membrane.

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

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. 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.

In the second possibility, the mechanism by which the air is trapped is known as “ball valve” described by Dandy. 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. 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.[7,15,19,20,31,34,37] This could be observed in 9 out of 13 cases of extradural location of the pneumocephalus.[15,16,17,24,26,27,33,34] 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.[31]

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 pneumocephalus 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.

REFERENCES

1. Ahren C, Thulin CA. Lethal intracranial complication following inflation in the external auditory canal in treatment of serous otitis media and due to defect in the petrous bone. Acta Otolaryngol 1965;60:407-21.
2. Andrews JC, Canalis RF. Otogenic pneumocephalus. Laryngoscope 1986;96:521-8.
3. Añorbe E, Ais P, Saenz de Ormijana J. Spontaneous pneumatocele associated with mastoid hyperpneumatization. Eur J Radiol 2000;36:158-60.
4. Barada W, Najjar M, Beydoun A. Early onset tension pneumocephalus following ventriculoperitoneal shunt insertion for normal pressure hydrocephalus: A case report. Clin Neurol Neurosurg 2009;111:300-2.
5. Chiari H. A case of accumulation of air in the ventricles of human brain. Zschr F Heilk 1884; 5.383-90.
6. Dowd GC, Molony TB, Voorhies RM. Spontaneous otogenic pneumocephalus: Case report and review of the literature. J Neurosurg 1998;89:1036-9.
7. Goldman RW. Pneumocephalus as a consequence of barotraumas. JAMA 1986;255:3154-6.
8. Guedes Bde V, da Rocha AJ, da Silva CJ, dos Santos AR, Lazarini PR. A rare association of tension pneumocephalus and a large frontoethmoidal osteoma: Imaging features and surgical treatment. J Craniofac Surg 2011;22:212-3.
10. Hyam JA, Morgan L, Mendoza ND. Coma caused by spontaneous otogenic pneumocephalus. Clin Neurol Neurosurg 2008;110:62-4.

11. Jelsma F, Moore DF. Cranial aerocele. Am J Surg 1954;87:437-51.

12. Kamide T, Nakada M, Hayashi Y, Hayashi Y, Uchiyama N, Hamada J. Intraparenchymal pneumocephalus caused by ethmoid sinus osteoma. J Clin Neurosci 2009;16:1487-9.

13. Kanner A, Nageris B, Chaimoff M, Rappaport Z. Spontaneous pneumocephalus in the posterior fossa in a patient with a ventriculoperitoneal shunt: Case report. Neurosurgery 2000;46:1002-4.

14. Kim J, Song SW, Cho JH, Chang KH, Jun BC. Comparative study of the pneumatization of the mastoid air cells and paranasal sinuses using three-dimensional reconstruction of computed tomography scans. Surg Radiol Anat 2010;32:593-9.

15. Krayenbuhl N, Alkadhi H, Jung HH, Yonekawa Y. Spontaneous otogenic intracerebral pneumocephalus: Case report and review of the literature. Eur Arch Otorhinolaryngol 2005;262:135-8.

16. Madeira JT, Summers GW. Epidural mastoid pneumatocele. Radiology 1977;122:727-8.

17. Maier W, Fradis M, Schermet R. Spontaneous otogenic pneumocephalus. Ann Otol Rhinol Laryngol 1996;105:300-2.

18. Markham JW. The clinical features of pneumocephalus based upon a survey of 284 cases with report of 11 additional cases. Acta Neurochir (Wien) 1967;16:1-78.

19. Mirone G, Rotondo M, Scuotto A, Bocchetti A, D’Avanzo R, Natale M, et al. Spontaneous intraparenchymal tension pneumocephalus triggered by compulsive forceful nose blowing. Emerg Med J 2009;26:837-8.

20. Mohammed E, Profant M. Spontaneous otogenic pneumocephalus. Acta Otolaryngol 2011;131:670-4.

21. Nagai H, Moritake H. Otogenic tension pneumocephalus complicated by Eustachian tube insufflation in a patient with a ventriculoperitoneal shunt: Case report. J Neurosurg 2007;106:1098-101.

22. Nyrop M, Bjerke PK, Christensen J, Jorgensen KE. Extensive and symptomatic cranial pneumatization: Caused by frequent performance of Valsalva’s manoeuvre? J Laryngol Otol 1999;113:480-2.

23. Osborn AG, Daines JH, Wing SD, Anderson RE. Intracranial air on computerized tomography. J Neurosurg 1978;48:355-9.

24. Park P, Chandler WF, Telian SA, Doran S. Spontaneous chronic epidural pneumocephalus resulting from hyperpneumatization of the cranium causing mass effect: Case report. Neurosurgery 1998;42:1384-6.

25. Pennings R, Liao W, Cremers CW. A spontaneous otogenic extradural pneumocephalus. Otol Neurotol 2009;30:864-7.

26. Richards SD, Saeed SR, Laitt R, Ramsden RT. Hypercellularity of the mastoid as a cause of spontaneous pneumocephalus. J Laryngol Otol 2004;118:474-6.

27. Schrijver HM, Berendse HW. Pneumocephalus by Valsalva’s maneuver. Neurology 2003;60:345-6.

28. Shaikh N, Masood I, Hanssens Y, Louon A, Hafiz A. Tension pneumocephalus as complication of burr-hole drainage of chronic subdural hematoma: A case report. Surg Neurol Int 2010;1:27.

29. Singh A, Alvarez J. Spontaneous otogenic intracerebral pneumocephalus. West J Emerg Med 2010;11:107.

30. Sloan T. The incidence, volume, absorption, and timing of supratentorial pneumocephalus during posterior fossa neurosurgery conducted in the sitting position. J Neurosurg Anesthesiol 2010;1:59-66.

31. Spar JA. Spontaneous CFS communication to the middle ear and external auditory canal: A case report. Acta Radiol 1994;35:506-8.

32. Stavas J, McGachie RE, Turner DA, Nelson MJ. Symptomatic intracranial pneumatocele from mastoid sinus of spontaneous origin: Case report. J Neurosurg 1987;67:773-5.

33. Tucker A, Miyake H, Tsuji M, Ukit a T, Nishihara K, Ito S, et al. Spontaneous epidural pneumocephalus. Neurol Med Chir (Tokyo) 2008;48:474-8.

34. Vallejo LA, Gil-Carcedo LM, Borras JM, De Campos JM. Spontaneous pneumocephalus of otogenic origin. Otolaryngol Head Neck Surg 1999;121:662-5.

35. Villarejo F, Carceller F, Alvarez C, Bencomo J, Perez Diaz C, Goldman L, et al. Pneumocephalus after shunting for hydrocephalus. Child Nerv Syst 1998;14:333-7.

36. Wong G, Young IA, Swanson E, Haynes D. Spontaneous otogenic posterior fossa pneumocephalus. Otolaryngol Head Neck Surg 2010;143:593-4.

37. Zhao N, Wang DD, Huang X, Karri SK, Wu H, Zheng M. Spontaneous otogenic pneumocephalus presenting with occipital subcutaneous emphysema as primary symptom: Could tension gas cause the destruction of cranial bones? J Neurosurg 2011;115:679-83.