

## Case Report

# Spontaneous Pneumocephalus Associated with Pneumosinus Dilatans

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The majority of cases of pneumocephalus are secondary to trauma or medical intervention. Spontaneous, non-traumatic pneumocephalus is an uncommon condition. Most cases of spontaneous pneumocephalus require surgery. However, if there is no evidence of infection or cerebrospinal fluid leak, bed rest and follow-up imaging is an alternative treatment. Herein, we report a 31-year-old man with spontaneous pneumocephalus associated with pneumosinus dilatans.

**KEY WORDS :** Spontaneous pneumocephalus · Hyperpneumatization · Pneumosinus dilatans.

## INTRODUCTION

Pneumocephalus is defined as an intracranial gas collection. Air can be localized in the epidural, subdural, subarachnoid, intraventricular or intraparenchymal spaces<sup>3</sup>. The majority of pneumocephalus cases are traumatic or iatrogenic. Spontaneous, non-traumatic pneumocephalus is an uncommon condition<sup>20</sup> and the typical causes of spontaneous intracranial air are barotraumas<sup>6-8</sup>, valsalva maneuvers<sup>3</sup>, adjacent air sinus infections<sup>5,16</sup>, bacteremia<sup>17</sup> and air cell hyperpneumatization<sup>15,18</sup>. Pneumosinus dilatans (PSD) refers to an abnormally enlarged, air-filled paranasal sinus without radiological evidence of localized bone destruction, hyperostosis, or mucous membrane thickening<sup>13</sup>. We report a case of spontaneous pneumocephalus associated with PSD.

## CASE REPORT

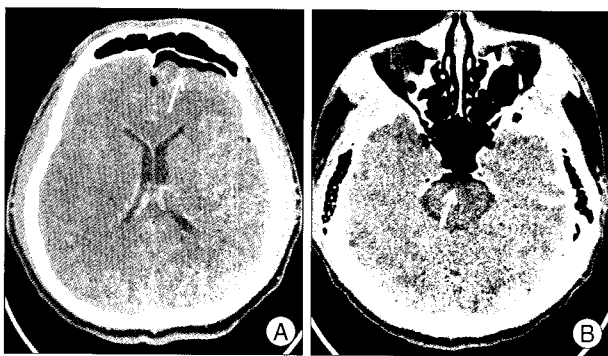
A 31-year-old man visited the emergency room because of a sudden headache and nausea that started with a "bang" sound in his head while he was tying a necktie. He had no previous history of trauma or medical illness. Computed

tomography (CT) scanning at a local hospital revealed pneumocephalus (Fig. 1). He was then transferred to our hospital for further evaluation and management. A repeat CT scan of his brain at the emergency room in our hospital showed multifocal pneumocephalus on the prepontine cistern, basal cistern, frontal area and anterior interhemispheric fissure (Fig. 2). The mastoid, sphenoid, and ethmoid air cells and air sinuses were extremely pneumatized, and the findings were consistent with pneumosinus dilatans (Fig. 3). There was no rhinorrhea, otorrhea or current sinus infection, and we consulted an otorhinolaryngologist who confirmed that both nasal cavities were clear. The possible site of air leak into the brain was thought to be the post-

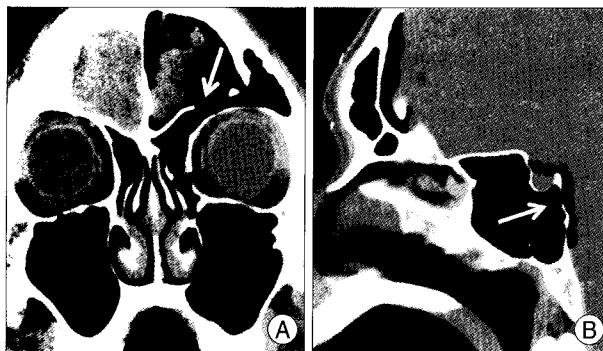


Fig. 1. Computed tomography of brain showing pneumocephalus.

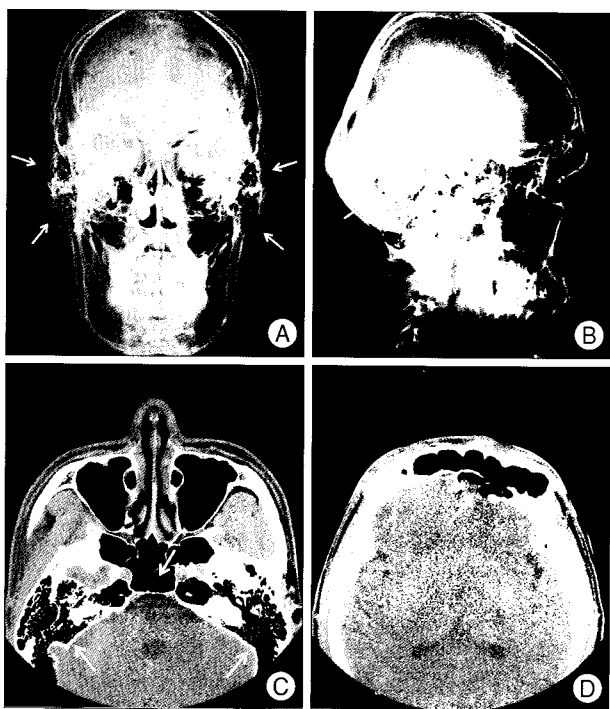
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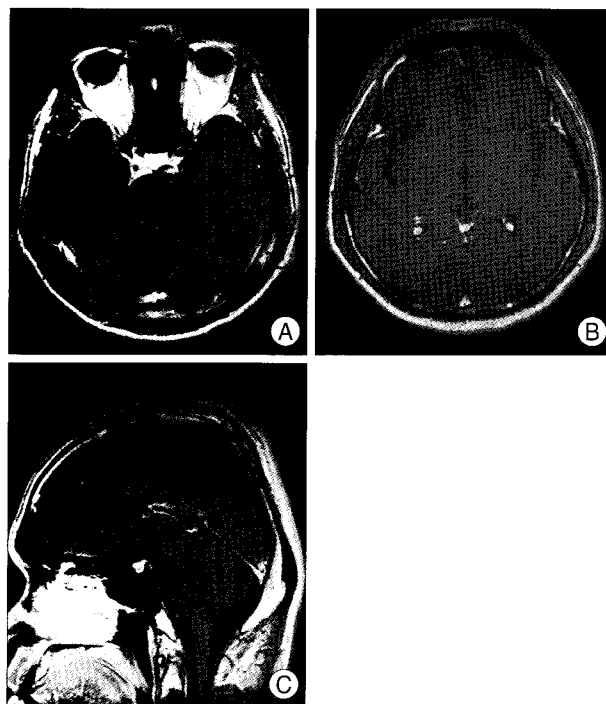
**Fig. 2.** Computed tomography of brain on admission. Axial plane demonstrates pneumocephalus in the frontal convexity (A) and prepontine cistern (B).



**Fig. 4.** Computed tomography scan of the brain images. Coronal and sagittal reconstruction planes demonstrate a localized thinning of the frontal sinus posterior wall (A) and of the sphenoidal sinus posterior wall (B).



**Fig. 3.** Radiologic examinations demonstrate extreme expansion of the sinus. Simple skull X-ray (A and B) and computed tomographic scan (C and D) shows the extent of frontal sinus and mastoid air cells and sphenoidal sinuses.



**Fig. 5.** Enhanced magnetic resonance imaging scan of the brain demonstrates no evidence of infection and residual fluid collection in the sinus and the resolution of pneumocephalus shown on the previous computed tomography scan. Axial planes (A and B) and sagittal reconstruction planes (C).

erior table of the frontal sinus or the sphenoid posterior wall (Fig. 4).

On admission, he was prescribed bed rest and prophylactic intravenous antibiotics for a week and he was given laxatives and advised against valsalva maneuvers. Initial laboratory examinations showed a white blood cell (WBC) count of  $12,700 \times 10^6/L$ , red blood cell (RBC) count  $4.92 \times 10^{12}/L$ , fasting glucose 115 mg/dL, sodium (Na) 138 mEq/L, potassium (K) 3.4 mEq/L, HS-CRP [0-1.0] 0.13 mg/L, and ESR [0-10] 5 mm/h. Four days after admission, repeated laboratory analysis revealed WBC  $3740 \times 10^6/L$ , RBC  $4.86 \times 10^{12}/L$ , fasting glucose 139 mg/dL, Na 141

mEq/L, K 4.3 mEq/L, HS-CRP [0-1.0] 0.38 mg/L, and ESR [0-10] 10 mm/h. His headache resolved gradually over several days without surgical intervention. During bed rest, his vital signs were maintained within normal ranges.

Because there were no symptoms or signs suggestive of dural defect or intracranial or sinus infection, cisternography or other evaluations were not performed. Enhanced brain magnetic resonance imaging (MRI) was performed on the ninth day of admission to rule out sinus infection or fluid collection (Fig. 5). The scanned image was clear, and the pneumocephalus had been completely absorbed. He was discharged ten days after the development of pneumocephalus.

After one month of follow-up, he did not complain of any symptoms, and the pneumocephalus did not recur.

## DISCUSSION

In 1967, Markham published an extensive review of pneumocephalus, which consisted of 284 cases previously reported in the literature and 11 additional cases from his own experience. Among these 295 cases of pneumocephalus, trauma was the etiological factor in 218 cases (73.9%), neoplasm in 38 cases (12.9%), infection in 26 cases (8.8%), surgical complication in 11 cases (3.7%) and there were two cases in which no apparent etiology was found (0.6%)<sup>10</sup>. Except for the case of neoplasm, spontaneous pneumocephalus including infection numbered 28 cases (9.4%). Other authors have reported rare cases of spontaneous pneumocephalus. Babl et al.<sup>3</sup> reported a ten-year-girl who complained of several days of headache. Her symptoms had begun with a "popping feeling" in her occipital area after several episodes of forceful sneezing. She was treated conservatively at first but sphenoid sinus obliteration was required on her second admission because of CSF rhinorrhea. Anorbe et al.<sup>2</sup> reported a 27-year-old man who complained of a retroauricular mass that grew larger on performing valsalva's maneuver. An axial CT scan confirmed a mastoid and temporal hyperpneumatization, pneumocephalus and a pneumatocele. He needed antrotomy and sealing of the aditus and tympanic antrum because of increased pneumatocele. Schrijver and Berendse<sup>14</sup> reported a 30-year-old woman with epidural air collection and extensive hyperpneumatization of the left mastoid and erosion of the adjacent bone. She had frequently performed valsalva's maneuver. She was treated conservatively and

advised to cease performing valsalva's maneuver. Tucker et al.<sup>19</sup> reported a 19-year-old male with a dull, diffuse headache associated with nausea. He did report a habit of excessive nose blowing. Axial CT scan showed hyperpneumatization, and he was treated conservatively at first. However, one year later he underwent craniotomy and repair of a defect using a large vascularized galeal flap. There was a case of frequent nose blowing precipitating pneumocephalus<sup>12</sup> and cases of hyperpneumatization of air cell and tumor-related spontaneous pneumocephalus<sup>13</sup>. These cases were treated with surgical sealing of the bony defects.

Considering the previous reports (Table 1), spontaneous pneumocephalus has been commonly caused by barotraumas and can be accompanied by hyperpneumatization of air cell or PSD. High pressures well beyond intracranial pressure are generated in the sinuses by actions including nose blowing, coughing, straining, sneezing, and valsalva maneuver and by environmental pressures including those encountered in mountain climbing, during flights and during scuba diving. With high pressure, air may be driven into the intracranial cavity. Disease in the paranasal sinuses or middle-ear cleft may be responsible for causing the breach in the skull. However, extreme pneumatization of air cells could be an important underlying condition for spontaneous pneumocephalus even without outside forces.

There are two theories to explain the pathophysiological basis of pneumocephalus<sup>9</sup>. One theory is the "ball valve mechanism". Nose blowing and sneezing in association with a dural tear operating as a ball valve seems the most likely explanation for pneumocephalus. Fragments of bone, dural flap, sinus mucosa or granulation tissue can all act as a unidirectional valve<sup>4</sup>. Another mechanism suggested to explain pneumocephalus is the "inverted bottle effect me-

**Table 1.** Summary of cases with spontaneous pneumocephalus

Author	Age	Sex	Precipitating factor	Symptom	HP or PSD	Complications	Associated condition	Treatment
Babl et al. <sup>3</sup>	10	F	Forceful sneezing	Headache	None	CSF rhinorrhea	None	Surgery
Añorbe et al. <sup>2</sup>	27	M	Valsalva's maneuver	Auricular mass	Yes	None	None	Surgery
Schrijver et al. <sup>14</sup>	30	F	Valsalva's maneuver	Asymptomatic	Yes	None	Bronchial asthma, pulmonary abscess	Conservative
Richards et al. <sup>12</sup>	17	M	Nose blowing	Auricular mass	Yes	Otalgia, bloody discharge	None	Surgery
	50	F	Cough, sneeze	Facial pain, paresthesia	Yes	None	Allergic rhinitis, nasal polyps	Surgery
Scholsem et al. <sup>13</sup>	74	M	None	Headache	Yes	None	Meningioma	Surgery
Tucker et al. <sup>19</sup>	19	M	Nose blowing	Headache, nausea	Yes	None	None	Surgery
Our case	31	M	None	Headache, nausea	Yes	None	None	Conservative

CSF : cerebrospinal fluid, HP : hyperpneumatization, PSD : pneumosinus dilatans

chanism” when there is an associated meningeal defect from which CSF leakage may occur. With CSF flow out of the cranium, compensatory air enters to equalize the pressure<sup>3,4,11</sup>.

The most common presenting complaint in pneumocephalus is headache<sup>10</sup>. Other symptoms or findings include CSF rhinorrhea, meningeal signs, hemiparesis, papilledema, and cranial nerve palsies<sup>3</sup>. However, the presentation of pneumocephalus is often vague and the diagnosis is frequently unsuspected<sup>1,9,10</sup>.

Either conservative therapy or surgical intervention may be indicated. The decision depends in part upon the type of pneumocephalus as well as its etiology and severity<sup>10</sup>. Surgical treatment for pneumocephalus is indicated when the defect does not heal satisfactorily, allowing recurrence with continued or recurrent cerebrospinal fluid rhinorrhea or otorrhea associated with pneumocephalus or signs of increased intracranial pressure due to intracranial air accumulation; recurrent pneumocephalus with or without meningitis; neoplasm that creates a pathway for ingress of air; infection that allows air to enter the skull or is gas producing; and presence of an intracerebral aerocele, since this implies adherence of the brain to a fistulous tract<sup>10</sup>.

In the case described here, there was no clearly defined etiological factor. However, our patient had PSD, and it is possible that he had some dehiscence of the frontal posterior table or sphenoid posterior wall. Because we cannot completely rule out the possibility that wearing a necktie can cause a valsalva effect and because he had pneumosinus dilatans, the ball valve mechanism may have played a key role this case. He had no additional signs of infection, we therefore tried conservative treatment with good response.

## CONCLUSION

Pneumocephalus of nontraumatic, spontaneous origin is rare. According to the available literature, most cases of spontaneous pneumocephalus result from nose blowing, sneezing or valsalva maneuver. In patients with PSD, pneumocephalus can occur spontaneously. If there are no signs of infection or symptoms of dural defect, we believe that observation and serial imaging would be an alternative treatment option.

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