

## Case Report

## Delayed Pneumoventricle Following Endonasal Cerebrospinal Fluid Rhinorrhea Repair with Thecoperitoneal Shunt

### Abstract

Pneumocephalus and pneumoventricle are well-documented in neurosurgical practice. Although both are common posttraumatic sequelae, iatrogenic causes are also well recognized. Iatrogenic causes may be seen after intracranial surgical procedures or cerebrospinal fluid (CSF) diversion procedures. Small amount of pneumoventricle postshunt procedure is usually a self-limiting condition. Rarely, the patient may develop tension pneumoventricle which requires emergency intervention. The occurrence of delayed tension pneumoventricle/pneumatocele following surgery for CSF rhinorrhea with CSF diversion procedures is very rare. We report one case of late presentation of delayed tension pneumoventricle with temporal pneumatocele in a patient who underwent transnasal endoscopic repair of CSF fistula followed by thecoperitoneal shunt. This condition is potentially lethal that requires prompt recognition and surgical treatment.

**Keywords:** Cerebrospinal fluid rhinorrhea, pneumatocele, pneumocephalus, tension pneumoventricle, thecoperitoneal shunt

### Introduction

The occurrence of pneumoventricle as a delayed complication of cerebrospinal fluid (CSF), rhinorrhea repair with thecoperitoneal shunt is a rare presentation. Tension pneumocephalus is a known and common entity as compared to tension pneumoventricle. The presence of pneumatocele in the temporal lobe in association with the above condition makes it a unique clinical presentation.

### Case Report

This 48-year-old female presented with complaints of CSF rhinorrhea since 2 months. There was no history or clinical finding suggestive of trauma or meningitis. Her neurological examination otherwise was unremarkable. Computed tomography (CT) face/skull base followed by magnetic resonance imaging (MRI) brain and diagnostic nasal endoscopy were done which showed the defect in the cribriform plate and left a lateral wall of the sphenoid sinus [Figure 1]. Lumbar puncture done showed the CSF opening pressure of 35 cm of water and no evidence of infection.

She underwent transnasal endoscopic repair of CSF fistula along with placement of

thecoperitoneal shunt with no anti-siphon device. The bath-plug technique was used to seal the defects by introducing a fat plug with a specifically secured vicryl suture into the intradural space, followed by applying traction on the suture to seal the defect much like a bathplug seals a bath. Rectus abdominis fascia graft was harvested from the same abdominal wound used for shunt placement. The defect was further reinforced by fascia, fat and surgical, and fibrin sealant. It was decided to place thecoperitoneal shunt as the CSF opening pressure was very high to prevent the recurrent CSF leak. Postoperatively, she was symptom-free and discharged to home.

After 1 month, she presented with memory disturbances, multiple episodes of vomiting and headache. There was no recurrence of CSF rhinorrhea or postnasal drip. MRI brain showed pneumoventricle with right temporal pneumatocele [Figure 2]. Diagnostic endoscopy was done which showed dislodged fascia graft. She underwent emergency repacking of the CSF fistula with the removal of thecoperitoneal shunt and aspiration of pneumoventricle underwater seal which was under high pressure. Fascia graft was repositioned to cover the defect after sealing it with fat

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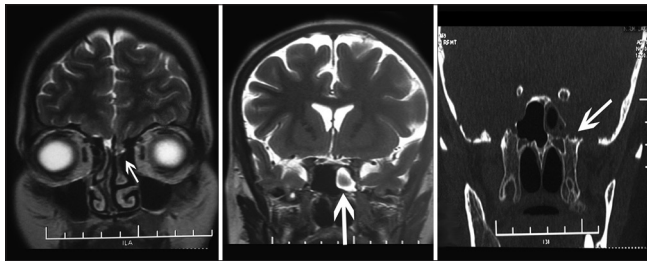
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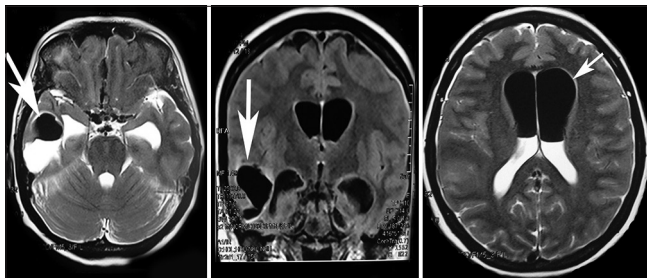
using the bath-plug technique as in the previous surgery. Fat and fascia packing was reinforced with a pedicled Hadad flap and fibrin sealant. She improved in her symptoms postoperatively. Postoperative CT brain showed good resolution of pneumoventricle with reduced size of ventricular system [Figure 3]. She remained symptom free at 6-month follow-up.

## Discussion

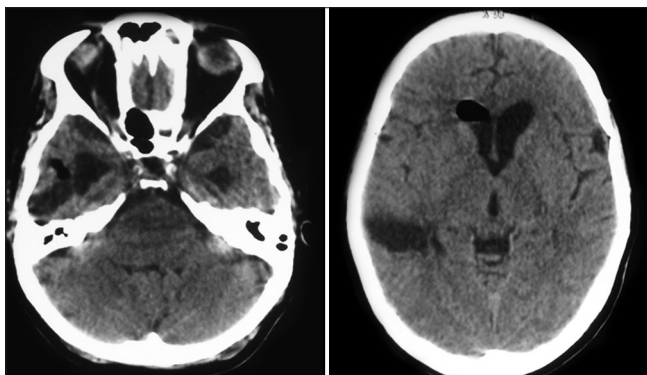
Pneumocephalus is defined as the presence of air in the intracranial compartment due to communication between intracranial and extracranial compartments.<sup>[1-3]</sup> Tension pneumocephalus is a rarer form of pneumocephalus in which the air is under high pressure.<sup>[1,2,4-7]</sup> Pneumocephalus occur most commonly in head injuries.<sup>[1-3]</sup> Intra- and post-operative pneumocephalus/pneumoventricle is well-documented,



**Figure 1:** Magnetic resonance images of the patient showing cerebrospinal fluid fistula in cribriform plate (short arrow) and lateral wall of sphenoid sinus (long arrow) and computed tomography paranasal sinuses bone window showing defect (long arrow) in the sphenoid bone with cerebrospinal fluid filling into the left sphenoid sinus (long arrow)



**Figure 2:** Magnetic resonance imaging brain showing pneumatocele (long arrow) in the right temporal lobe and pneumoventricle (short arrow)



**Figure 3:** Postoperative computed tomography brain showing good resolution of tension pneumoventricle and right temporal pneumatocele

especially, in sitting position surgeries, nitrous oxide anesthesia, and CSF diversion surgeries.<sup>[1-3]</sup> Other conditions causing pneumocephalus are CNS infections caused by gas-producing organisms, congenital neurenteric cysts, and postradiotherapy for nasopharyngeal carcinoma.<sup>[1,2,4-7]</sup> Small amounts of pneumoventricle alone are common after shunt surgeries, ventricular tumor surgeries.<sup>[1-3,8]</sup> Sometimes, wound breakdown following the shunt surgeries may cause influx of air peritubally and cause pneumoventricle.<sup>[9]</sup> Delayed tension pneumoventricle is an extremely rare complication and <50 cases have been described in the literature.<sup>[1]</sup> Pneumocephalus/pneumoventricle is usually benign which does not require any treatment, and it decreases at a rate of 25% per week.<sup>[4,9,10-12]</sup>

Two different mechanisms have been proposed in the development of delayed tension pneumoventricle/pneumocephalus.<sup>[1-3,13,14]</sup>

1. Dandy's theory of ball valve mechanism: one-way ball valve mechanism causing air to flow into the skull through dural defect where the exit is prevented by brain or meninges sealing the leak site
2. Horowitz inverted soda-bottle effect: negative pressure develops inside the cranial cavity as a result of excessive loss of CSF. This drop in intracranial pressure (ICP) causes air to flow from the extra to the intracranial space across the pressure gradient.

In our case, the patient presented with spontaneous CSF rhinorrhea and was treated by endoscopic skull base defect repair with thecoperitoneal shunt placement. CSF rhinorrhea can be due to traumatic or nontraumatic causes. Traumatic can be either due to head injuries causing skull base fractures or due to iatrogenic causes. Spontaneous leaks could be associated with or without raised ICP. High-pressure leaks could account up to 45% of the nontraumatic CSF rhinorrhea.<sup>[15]</sup> Sustained increase in ICP causes bony erosion and creation of an osteodural defect in pneumatized parts of the skull base such as cribriform plate, craniopharyngeal canal, sella, and sphenoid-occipital synchondrosis leading to CSF leak.<sup>[7,15]</sup> CSF leaks in these cases have been postulated to represent a manifestation of benign intracranial hypertension or pseudotumor cerebri.<sup>[16-19]</sup> In our case also CSF leak was associated with raised ICP with no evidence of trauma or infection. This could be an underlying benign ICP with or without congenital defect.

Normal pressure leaks represent 55% of the nontraumatic cases of the CSF rhinorrhea.<sup>[20,21]</sup> It is hypothesized that the spontaneous leak is due to point erosions in the skull base which occur in normal person as a result of physiologic alterations in CSF pressure with transient increase in ICP up to 80 mm of water lasting for few seconds.<sup>[15]</sup> Other nontraumatic causes of CSF leak include congenital skull base defects, erosion of the skull base by tumors, infection, mucocele, and following radiation.

CSF diversion in patients with long-standing raised ICP may result in the pneumoventricle by air aspiration through a preexisting congenital or iatrogenic skull base erosion/fistula.<sup>[3]</sup> These fistulous sites/erosion points are plugged by scarred meninges or gliotic brain which open up due to a drop in ICP causing inward flow of air. This air is prevented from escaping by temporarily resealing of meningeal cicatrix and this cycle repeatedly happens, resulting in tension pneumoventricle (ball valve mechanism). Shunts by their siphon effect can create significant negative ICP drop which ranges from  $-30$  to  $-155$  mm of water, and sometimes as low as  $-440$  mm of water.<sup>[2,5]</sup> Pneumatocele is located close to the site of fistulae and more common in the temporal lobe. In our case also there might have been a sustained negative pressure caused by the thecoperitoneal shunt without anti-siphon device.

Pneumoventricle presents usually with symptoms and signs of raised ICP such as a recurrent headache with vomiting, impairment of consciousness, seizures, memory disturbances, and gait disturbance. Sometimes, patients present with acute or chronic meningitis. Intracranial splashing sounds called “bruit hydroaérique” are characteristic in some patients. Similarly, our patient also presented with memory disturbances and cognitive impairment during the second presentation.

The delay from CSF shunting to the development of pneumocephalus may vary from a week up to 5 years.<sup>[7,22]</sup> The usage of high-pressure shunts and antisiphon devices have been recommended by some authors to prevent this complication.<sup>[1,2,23]</sup> Our routine policy is to place Chhabra standard adult thecoperitoneal shunt with no anti-siphon device. However, anti-siphon device has advantages of preventing over-drainage of CSF, and the reservoir gives access to check the patency of the shunt system. We have seen an increased risk of shunt obstruction with anti-siphon device. We had 42 cases who underwent Lumbar-peritoneal (LP) shunts without anti-siphon device in the past 5 years and none of them presented with shunt obstruction or shunt-related morbidity other than shunt migration (five patients) and abdominal pseudocyst (two patients). Programmable shunt provides the benefit of adjusting the pressure setting according to the ventricular pressure. We routinely do not use programmable LP shunts as ours is a resource-limited center with most of the patients coming from low-socioeconomic strata.

Prevention of infection, treatment of raised ICP, aspiration of pneumoventricle, closure of fistula, and removal of shunt tube are the keys to successful management of tension pneumoventricle secondary to CSF fistula.<sup>[23]</sup> Broad-spectrum antibiotics are used after shunt removal, but its prophylactic usage is debated.<sup>[1,5,23]</sup> Removal of the shunt tube relieves the sustained negative pressure which may cause recurrent pneumoventricle/pneumocephalus. Postshunt removal a temporary CSF diversion is preferred

by some authors, especially, if the infection is doubted clinically or confirmed.<sup>[1,2,23]</sup>

In our case, as there was dislodgement of fat-fascia graft, repacking was done followed by aspiration of pneumoventricle underwater seal and thecoperitoneal shunt was removed. We feel the tension pneumoventricle caused air to dissect into the right temporal lobe region under pressure forming the temporal pneumatocele. The shunt was removed to alleviate negative pressure gradient as it was nonprogrammable shunt with no anti-siphon device. The patient had complete resolution of symptoms after the procedure. This makes us think that we should probably reconsider the usage of anti-siphon device and also a pedicled flap to repair the skull base defects. A programmable valve may be the best choice in financially affordable patients. In patients with normal/moderately high ICP (25 cm of water), a temporary lumbar drain can be considered for few days until the defects heal thus preventing the recurrent CSF leaks.

## Conclusion

Although it is a rare entity, tension pneumoventricle should be considered in patients who have undergone CSF diversion procedures along with anterior skull base repair. Sometimes, it can occur as a delayed complication which may lead to acute neurological deterioration and sudden death. Hence, prompt diagnosis is necessary for timely intervention and prevention.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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## Conflicts of interest

There are no conflicts of interest.

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