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*Case Report*

# Tension pneumoventricle in a patient with a ventriculoperitoneal shunt and an ethmoidal meningoencephalocele

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## ABSTRACT

**Background:** Tension pneumoventricle is a rare, life-threatening complication. It has been rarely described in patients with ventriculoperitoneal (VP) shunts.

**Case Description:** A 28-year-old male patient with a VP shunt became progressively lethargic after falling from his wheelchair. Skull X-rays and head CT scan showed abundant air inside the ventricles. He was taken to the operating room, and the shunt was revised without improvement. Two days later, a frontal external ventricular drain was placed to remove the air. In the investigation toward the etiology of the pneumoventricle, a review of previous head CT scans and brain MRIs showed that the patient had a small left frontonasal meningoencephalocele extending into the ethmoid, which had been unnoticed. He underwent repair of the defect with adequate sealing of the frontal skull base.

**Conclusion:** In a shunted patient with moderate or severe symptoms from a tension pneumoventricle, external ventricular drainage is required to remove the air as the shunt is inadequate.

**Keywords:** Ethmoid, Meningoencephalocele, Pneumoventricle, Tension, Ventriculoperitoneal shunt

## BACKGROUND

Intraventricular pneumocephalus, also known as pneumoventricle, is usually asymptomatic and does not need treatment if present in small amounts. However, tension pneumoventricle is a rare, life-threatening complication that can cause a decline in the neurological status.[8,10,13,19] Symptoms of tension pneumoventricle may include amnesia, headache, aphasia, mutism, gait deterioration, hearing loss, dementia, decline in mental status, agitation, delirium, seizures, focal neurological deficit, progressive loss of consciousness, dilation of pupils, and cardiac arrest.[1,5,11-13,15,16,18-21,24] Tension pneumoventricle is usually a neurosurgical emergency requiring urgent treatment.[1,5,8,10] The etiology should be adequately managed to prevent subsequent episodes.

Tension pneumoventricle has been rarely described after placement of ventriculoperitoneal (VP) shunts.[6,7,12,13,15,17,18,22-24] It had also been reported in patients with lumboperitoneal shunts.[11] Tension pneumoventricle can develop after a skull base fracture.[2,5] Lumbar spinal drainage performed to treat rhinorrhea after head trauma may precipitate the development of

tension pneumoventricle.[8] Spontaneous cerebrospinal fluid (CSF) fistulas had been associated with tension pneumoventricle.[9] The rupture of a basal encephalocele may be involved in the pathophysiology.[4] However, sometimes, the etiology cannot be identified.[3,15,24] When the source of the pneumoventricle remains unknown, a temporary external ventricular drain (EVD) may be necessary to remove intracranial air and improve the patient's neurologic status.[17]

## CASE DESCRIPTION

A 28-year-old male patient with a VP shunt suffered a fall from his wheelchair, for which he became progressively lethargic during the following 2 days. On evaluation, his neurological examination showed a Glasgow Coma Scale of 12. Eight years prior, he was diagnosed with a left cerebellar arteriovenous malformation during workup for headaches [Figure 1]. The patient had multiple embolization with N-butyl cyanoacrylate and coils followed by staged Gamma Knife radiosurgery. He developed symptomatic radionecrosis with acute obstructive hydrocephalus, and a VP shunt had to be placed in addition to chronic steroids and hyperbaric chamber treatment.

As the patient had a VP shunt, it was believed that the acute lethargic presentation was due to malfunctioning of the VP shunt or a traumatic brain injury secondary to the fall. Manual palpation of the shunt valve showed good pumping and refill. A radiological workup for the shunt malfunction was ordered. The skull X-rays showed abundant air inside the ventricles, and we thought that it could be the source of his symptoms [Figure 2]. The axial head CT scan confirmed the initial radiographic impression of tension pneumoventricle without evidence of an acute traumatic brain injury. Coronal and sagittal head CT reconstructions were performed later by the neuroradiologist and showed a left ethmoidal meningoencephalocele with a fistulous air tract extending into the ventricle, which was not identified on the initial axial images [Figure 3]. We thought that the air inside the ventricle might have produced malfunctioning of the valve mechanism and brought the patient to the operating room to inspect the shunt system and, at the same time, remove some of the air inside the ventricles.

During the surgery, it was found that the ventricular catheter was patent, and the valve had good distal flow, although a substantial amount of air inside the valve chamber. Air bubbles were observed draining together with a clear CSF when the valve was disconnected from the ventricular catheter. A new valve was placed, and the patient was admitted to the intensive care unit. No improvement was observed during the following 2 days, and a repeated head CT scan still showed a significant amount of pneumoventricle. He was returned to the operating room, and a frontal EVD was placed to remove the air trapped inside the ventricles. After 1 day, the EVD was kept closed to monitor the intracranial pressure. A subsequent head CT scan 6 days later showed resolution of the pneumoventricle and the EVD was removed when the second CSF culture was negative. No active evidence of a CSF leak was observed during the hospitalization.

In our investigation toward the etiology of the pneumoventricle, a review of previous head CT scans and MRIs showed that the patient had a small left frontonasal meningoencephalocele extending into the ethmoid which had been previously unnoticed [Figure 4]. A small left frontal skull base defect produced migration of the meningoencephalocele into the ethmoid sinus. He did not present symptoms related to the meningoencephalocele on any of the prior admissions.

The otolaryngology department was consulted to repair the meningoencephalocele and frontal skull base defect. The patient underwent endoscopic endonasal removal of the meningoencephalocele with a reconstruction of the

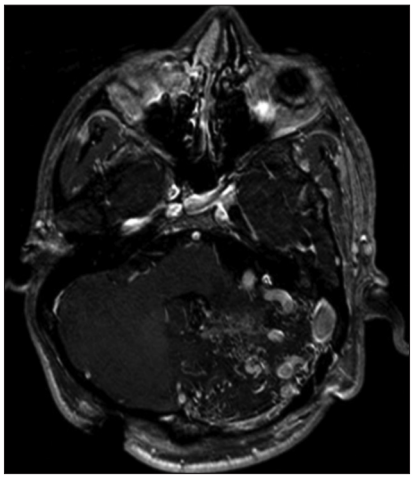

**Figure 1:** Axial T1-weighted brain MRI with contrast shows a large left cerebellar arteriovenous malformation.

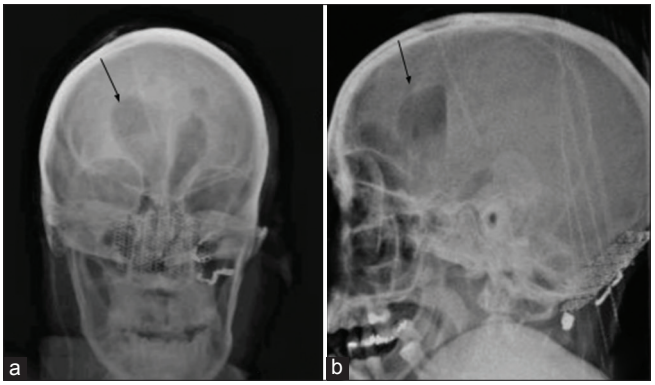


**Figure 2:** Skull X-rays films (a) anteroposterior view and (b) lateral view showing abundant air inside the ventricles (black arrow).

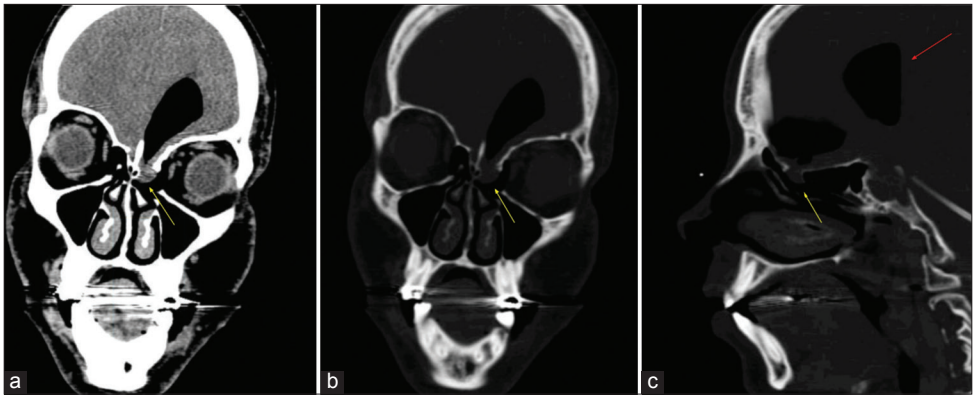


**Figure 3:** Head CT scan reconstruction images (a) coronal brain window view, (b) coronal bone window view, and (c) sagittal bone window view showing the left ethmoidal meningoencephalocele (yellow arrow) and the fistulous air tract extending into the ventricle (red arrow).

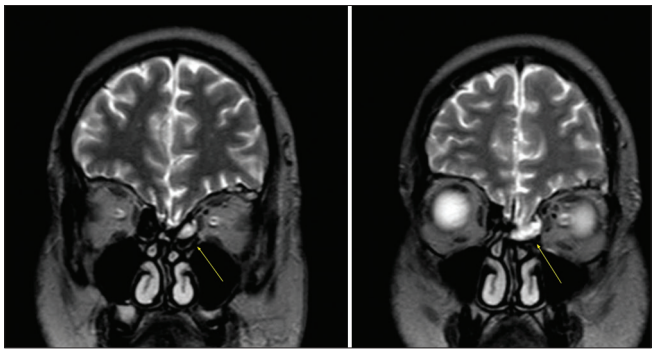

**Figure 4:** Coronal T2-weighted brain MRI (left and right) shows a small left frontobasal meningoencephalocele herniating into the ethmoid (yellow arrow).

frontal skull base defect using an underlay cartilage patch placed at the defect and a vascularized nasoseptal mucosal graft placed in an overlay fashion. No postoperative lumbar drainage was used. The patient did not present any CSF leak postoperatively, and the head CT scan showed small-sized ventricles with no pneumoventricle. Six months after the resection of the meningoencephalocele and reconstruction of the skull base defect, he continues asymptomatic without evidence of a CSF leak.

## DISCUSSION

In this case, the patient's deterioration was initially thought to be secondary to the malfunction of the VP shunt. However, the valve pumped well and refilled quickly. At surgery, the shunt system was found to work adequately, although the air inside the valve chamber. We believe that the fall he sustained from the wheelchair caused the rupture of the arachnoid membrane of the meningoencephalocele occupying the frontal skull base defect. Due to the negative pressure produced by the shunt, air was forced into the brain and ventricles, causing the patient's symptomatic presentation of tension pneumoventricle without evident rhinorrhea.

Cartwright and Eisenberg were the first to report on a tension pneumoventricle caused by the rupture of a basal encephalocele.[4] Ohkawa *et al.* reported the first case of a tension pneumoventricle in a patient with a VP shunt secondary to a temporal lobe encephalocele extending into the lateral sphenoid sinus recess.[14] To the best of our knowledge, our patient is the second case of tension pneumoventricle in a patient with a VP shunt and a meningoencephalocele. For patients with a basal skull defect, improvement is seen after placement of a temporary EVD or modification of the shunt system. Intracranial pressure alterations induced by a previously placed VP shunt might play a role in facilitating the development of tension pneumoventricle.[14] The shunt's possible reduction of intracranial pressure produces a one-way valve as air enters the ventricles but cannot escape from the intracranial compartment.[4,5,22] Long-standing intracranial hypertension can produce skull base defects by erosion of the anterior cranial fossa floor.[6] In those patients who develop a CSF leak, the negative pressure inside the ventricles allows air to enter the ventricles.[1,3,5,17] For the treatment of pneumoventricle in patients with VP shunts, modification of the valve pressure may be sufficient for mildly symptomatic cases.[23] However, for significantly symptomatic patients, the placement of an EVD is usually required.[2,20] Patients usually show an immediate improvement. If there is a CSF fistula, treatment by completely excluding the fistula is necessary because of the lethal risk of pneumoventricle and meningitis.[9,13,17,23] Based on our case and others in the literature, we think that a VP shunt is incapable of removing air in large enough volumes to improve symptoms.[13]

## CONCLUSION

Tension pneumoventricle is a rare but potentially life-threatening complication. In patients with moderate or severe symptoms, external ventricular drainage is required to remove the air. A skull base defect or meningoencephalocele can produce a tension pneumoventricle. Patients with VP

shunts can also develop tension pneumoventricle. In these patients, the VP shunt may be incapable of removing air in large volumes to improve symptoms.

### Declaration of the patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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