Pathogenesis of Delayed Tension Intraventricular Pneumocephalus in Shunted Patient: Possible Role of Nocturnal Positive Pressure Ventilation

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Key words
- Hydrocephalus
- Pneumoventricle
- Ventriculoperitoneal shunt
- Pneumocephalus
- Sleeping apnea
- Nocturnal positive pressure ventilation

Abbreviations and Acronyms
CPAP: Continuous positive airway pressure
CSF: Cerebrospinal fluid
CT: Computed tomography
VP: Ventriculoperitoneal

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BACKGROUND: **Delayed intraventricular pneumocephalus is a very rare and potentially serious complication of ventriculoperitoneal shunt. It can occur several months or years after shunting. Its pathogenesis is unclear. We herein discuss the underlying mechanisms and particularly the possible role of positive pressure ventilation.**

CASE DESCRIPTION: **A 60 year-old man presented with a lateral ventricle neurocytoma microsurgically resected complicated by a late-onset (15 months) postoperative hydrocephalus requiring an adjustable ventriculoperitoneal (VP) shunt. One month later, the patient was diagnosed with a sleep apnea and required a continuous positive airway pressure (CPAP) device. A few weeks afterward the patient presented with headaches and alteration of consciousness. CT-Scan revealed a massive intraventricular pneumocephalus associated with a millimetric left petrous bone defect. A transient breakout of the positive ventilation and a subtemporal surgical repair of the defect led to the rapid resolution of the pneumocephalus.**

DISCUSSION: **Delayed intraventricular pneumocephalus requires two conditions: a VP shunt and an osteodural defect. The CPAP may play an important trigger role in the pathogenesis of this complication through a ball valve mechanism. The management relies on transient suspension of the positive ventilation and the surgical repair of the identified defect with or without pressure adjustments of the valve.**

CONCLUSION: **Intraventricular pneumocephalus is a potentially serious complication of patients with a VP shunt and receiving positive pressure ventilation. The introduction of a CPAP device must be discussed with the neurosurgeon beforehand in shunted patients.**

INTRODUCTION

Intraventricular pneumocephalus after a ventriculoperitoneal (VP) shunt is rare, occurring when air enters through the shunt or comes into the cranial cavity through a skull base osteodural defect.[1] We report a rare case of delayed compressive intraventricular pneumocephalus occurring 15 months after the placement of a VP shunt in a patient treated for a sleeping apnea. The pathogenesis is discussed, as is the role of positive ventilation and the therapeutic strategy.

CASE DESCRIPTION

A 60-year-old man without any relevant medical history was operated on for a left lateral ventricle neurocytoma revealed by intracranial hypertension syndrome. He underwent a complete surgical removal through a left transcortical transfrontal approach. The postoperative course was uneventful, and he remained asymptomatic for a 15-months period, after which he developed a cognitive decline and gait disorders. The computed tomographic (CT) scan revealed an active communicating hydrocephalus; therefore, he required a VP adjustable shunt in the right ventricular atrium. All neurologic symptoms resolved, and he was discharged home on postoperative day 7, with an opening pressure of the shunt set at 150 mm H_2O. The immediate postoperative CT scan confirmed the good position of the ventricular catheter and a slight decrease in the ventricular size.

A few weeks later, sleep apnea was diagnosed with nocturnal recordings. The patient required a CPAP during his night sleep. Shortly afterward, a deterioration of his gait disorders was observed, in addition to a gradual psychomotor worsening and a fluctuant alteration of consciousness consistent with valve dysfunction. A brain CT scan showed a large expansive intraventricular pneumocephalus in the frontal and temporal horns (Figure 1). The pressure of the shunt was therefore adjusted from 150 to 200 mm H_2O, providing a slight improvement in neurologic status. A whole-body CT scan was performed to identify the origin of air penetration (lung, abdomen, pelvis). No abnormality was identified along the VP tube or in the abdominal cavity. The skull base thin-sliced CT scan confirmed a bone defect at the roof of a left petrous air cell

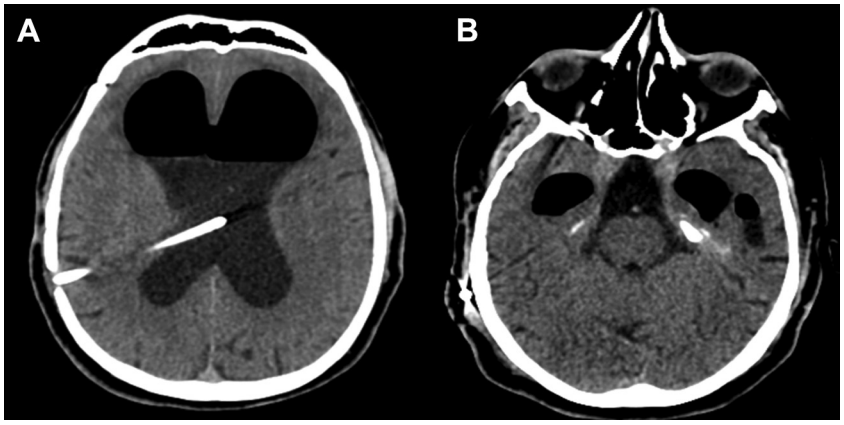

Figure 1. Brain axial computed tomographic scan without contrast: (**A**) major intraventricular frontal and (**B**) temporal pneumocephalus. An air bubble is noted in a porencephalic cavity against the lateral aspect of the left temporal horn.

(Figure 2A). The contact of this bone defect with an air bubble within a porencephalic cavity of the left temporal lobe (Figure 2B–D) led us to hypothesize that it was probably the site of entry of air into the brain. Magnetic resonance imaging confirmed this hypothesis, showing a communication between the small temporal porencephalic cavity and the left temporal horn (Figure 3). Because of the importance of this symptomatic pneumocephalus and the insufficient effect of shunt pressure adjustment, a surgical treatment of this defect was chosen. The middle fossa was explored extradurally through a left temporal craniotomy. The bone defect and two adjacent dural tears were identified. The dura mater was sutured, and a customized bone graft was taken from the bone flap (splitting of the bone flap) to close the defect at the skull base, secured with glue and fat tissue. The postoperative course was uneventful. Subsequent CT scans showed a gradual resolution of the intraventricular pneumocephalus (Figure 4). The valve was readjusted to 140 mm H_2O. The clinical symptoms finally resolved in a few days, and the patient was discharged home. The CPAP was reintroduced 3 months later, and the patient remained asymptomatic at the last follow-up, 3 years after the skull base repair.

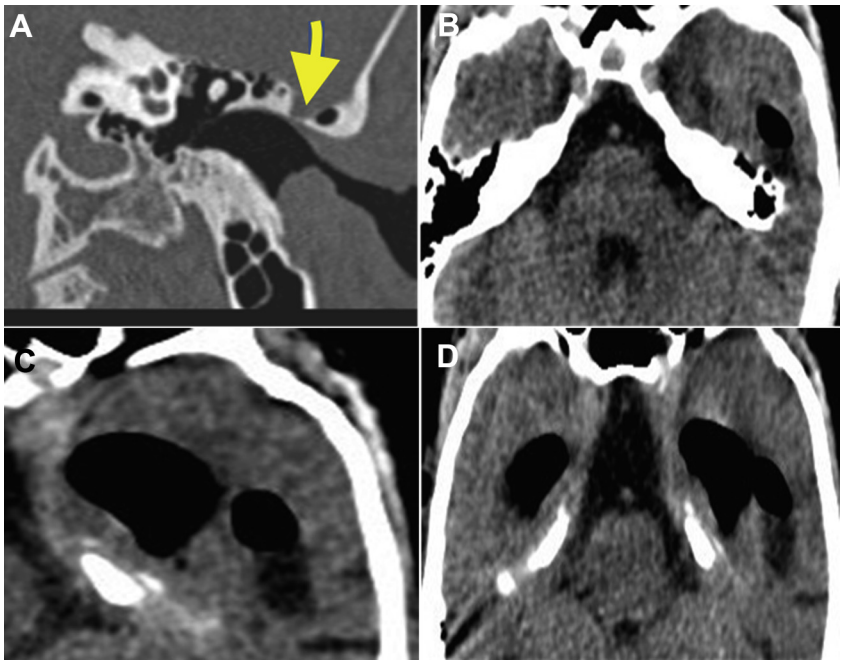

Figure 2. Cerebral computed tomographic scan without contrast. (**A**) Thin coronal section (bone window) showing the petrous defect (*arrow*). (**B**–**D**) Axial sections (parenchymal window) showing the porencephalic cavity with an air-liquid level close to the temporal horn of the lateral ventricle and adjacent to the underlying petrous air cells.

DISCUSSION

Intraventricular pneumocephalus is a rare complication of VP shunts. To our knowledge, the first intraventricular pneumocephalus case in a patient with a VP shunt and coexisting osteodural defect was published in 1975 by Pitts et al.[2] This complication can be immediate, occurring early in the postoperative course, or delayed several months or even years after the VP shunt placement.[1,3]

The pathogenesis of spontaneous pneumocephalus associated with ventricular shunting is based on the coexistence of two conditions: 1) a reduced intracranial pressure because of ventricular CSF depletion and 2) a skull base osteodural defect, allowing air to enter the cranial cavity. In rare cases, air can ascend into the cranial cavity through the distal catheter, therefore representing a differential diagnosis.[1] The skull base defect may be congenital, but frequently it is linked to thinning and erosion of the meninges and the bone[2] as a consequence of chronic hydrocephalus.[4] This condition is most often observed in the middle cranial fossa, which is explained by the thinness of the bone of the petrous air cells.[5,6] Bone dehiscence or abnormally thin bone at the level of the skull base may also result from embryologic anatomic variations, particularly at the level of the facial canal.[7]

As patients with Sylvius aqueduct stenosis can have thinning of the skull base and particularly dorsum sellae,[6] they are at risk for the development of