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Delayed Intraventricular Tension Pneumocephalus Due to Scalp-Ventricle Fistula: A very Rare Complication of Shunt Surgery

Skalp-ventrikul Fistülüne Bağlı Gelişen Gecikmiş Tansiyon Hidrosefalisi: Şant Cerrahisinin Nadir bir Komplikasyonu

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ABSTRACT

BACKGROUND: Although pneumocephalus and pneumoventricle are well known entities in neurosurgery practice, delayed intraventricular tension pneumocephalus following shunt surgery is extremely rare.

CASE DESCRIPTION: A 60-year-old man presented with vomiting, drowsiness, walking difficulty, urinary incontinence and headache one month after shunt placement for communicant hydrocephalus developing secondary to aneurysmal subarachnoid hemorrhage. Skull X- Rays and Computed Tomography (CT) revealed marked air in both lateral ventricles. Neither physical and neurological examinations nor laboratory studies and cerebrospinal fluid evaluations suggested central nervous system infection. He gradually improved after repairing the scalp incision defect above the previously opened burr-hole and bed rest.

CONCLUSION: In contrast to reported cases with delayed pneumocephalus developing after shunting, air entry was in skull base, air source was the scalp-ventricular fistula in the present case.

KEY WORDS: Pneumocephalus, Pneumoventricle, Ventriculoperitoneal shunt, Hydrocephalus

ÖZ

GİRİŞ: Nöroşirürji pratiğinde pnömosefali ve pnömoventrikül iyi bilinen antiteler olmakla birlikte, şant cerrahisi sonrası gelişen gecikmiş intraventriküler tansiyon pnöomosefalisi son derece nadirdir.

OLGU SUNUMU: 60 yaşında erkek hasta, subaraknoid kanamayı takip eden hidrosefali nedeni ile ventrikülоperitoneal şant operasyonundan 1 ay sonra, kusma, halsizlik, yürüme bozukluğu, başağrısı ve idrar kaçırma yakınmaları ile başvurdu. Direkt kranyal grafi ve bilgisayarlı beyin tomografisinde, her iki lateral ventrikülde belirgin hava izlendi. Fizik ve nörolojik muayenesinde, laboratuvar tetkiklerinde ve beyin omurilik sıvısı analizinde infeksiyon bulgusu saptanmadı. Şant operasyonu sırasında açılmış olan Burr-hole üzerindeki cilt insizyonunun primer tamiri ve yatak istirahati sonrası hastanın klinik tablosu hızla düzeldi.

SONUÇ: Şant cerrahisi sonrası gelişen gecikmiş pnömosefali olgularında havanın intrakranyal alana girişi genelde kafa tabanındaki bir defektten olur. Sunulan olguda ise hava girişi skalp-ventrikül arası fistül vasıtası ile olmuştur.

ANAHTAR SOZCÜKLER: Pnömosefali, Pnömoventrikül, Ventrikülоperitoneal şant, Hidrosefali

INTRODUCTION

Pneumocephalus is the presence of air within the intracranial space. Pneumoventricle is the occurrence of air in the ventricle. Pneumocephalus is a well-known condition in neurosurgery practice and often accompanies a surgical procedure at the postoperative period (6,8,10). Head injury and accompanying skull base fractures may often cause pneumocephalus (5,7,15). Delayed pneumocephalus and pneumoventricle developing after a cerebrospinal fluid diversion procedure for hydrocephalus is extremely rare and less than 50 cases have been described in the literature (1,3,4,6,9,11,12,13,14).

We present a unique case of delayed pneumoventricle due to a skin defect resulting from previous ventriculoperitoneal shunt surgery.

CASE DESCRIPTION

A 60-year-old man was admitted to hospital with severe headache, nausea and vomiting. Physical and neurological examinations were normal except for moderate neck stiffness. Cranial Computed Tomography (CT) was normal. A subarachnoid hemorrhage was confirmed after cerebrospinal fluid (CSF) evaluation. A left posterior communicating aneurysm was diagnosed after digital subtraction angiography. The aneurysm was coiled successfully and the patient was discharged from hospital two days later.

He was readmitted to hospital 15 days later with a two-day history of urinary incontinence and drowsiness. On examination, the patient had altered consciousness and could not recognize his relatives. The cranial CT scan showed marked dilatation of the lateral and third ventricles with transependymal fluid passage (Figure 1). He underwent a shunt operation for communicant hydrocephalus and a normal pressure ventriculoperitoneal shunt was implanted. The postoperative period was uneventful and he was discharged after three days with a normal neurological examination.

The patient was readmitted a month later with a two-day history of vomiting, drowsiness, incontinence, walking difficulty and headache. The physical examination revealed an incision scar at the left frontotemporal region and at the right koher point. There was a necrotic area and scalp defect on the suture line at the koher point. Neurological examination demonstrated confusion and gait disturbance. There was no fever or neck stiffness. Routine laboratory studies were within normal limits. There were no symptoms or findings suggestive of a central nervous system infection. Lumbar puncture and cerebrospinal fluid study showed no abnormality. Skull X-Ray showed massive air in both lateral ventricles. CT scans also revealed air in both lateral ventricles and just under the previous burr-hole (Figure 2A,B). Further assessment with high-resolution coronal CT did not demonstrate any defect at the skull base. Neck, chest

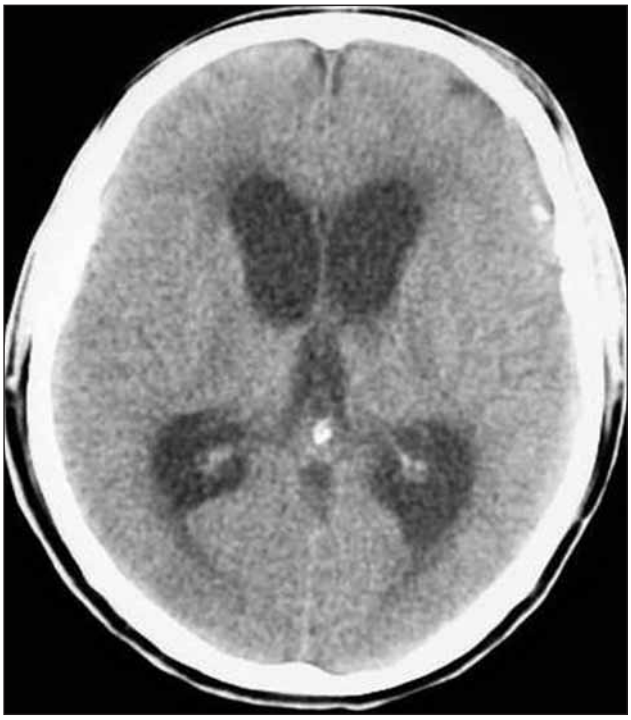

Figure 1: *CT scan reveals hydrocephalic dilatation of both lateral and third ventricles.*

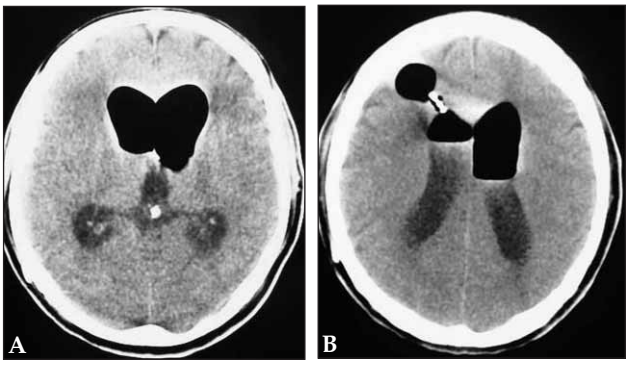

Figure 2A: *CT scan shows marked air in both lateral ventricles*
Figure 2B: *CT scan also demonstrates a porencephalic cyst just beneath the entrance of ventricular catheter to intracranial cavity.*

and abdominal X-rays showed no disconnection along the shunt system. The necrotic tissue was debrided and the skin defect was closed primarily with vicryl material under local anesthesia. The patient was ordered complete bed rest and his symptoms and findings gradually improved within a week. Closure of the skin defect and bed rest resulted in gradual disappearance of the intraventricular air (Figure 3A,B). No more air inflow was observed in clinical follow-up. A prophylactic antibiotic was given during hospitalization. The patient was discharged from hospital 1 week after admission without any deterioration of neurological status. He was clinically asymptomatic and the air in ventricles was totally disappeared on CT scan at 1-month follow-up (Figure 4).

DISCUSSION

Most cases with pneumocephalus occur after head trauma, especially after skull base and sinus fractures. This phenomenon is also usually associated with neurosurgical procedures, especially in the sitting surgical position. Neoplasms, gas producing-anaerobic central nervous system infections, mucoceles, congenital neuroenteric cysts, dural defects, lumbar drain insertion, congenital skull defects and disorders of otogenic origin are other causes of pneumocephalus. Tension pneumocephalus is a rare form of pneumocephalus in which the air is under pressure; it is generally due to communication between the atmosphere and the intracranial cavity.

Cerebrospinal fluid (CSF) shunting has been associated with a significant number of complications. Infections, shunt malfunction, slit ventricle, subdural hematomas are among the most common. Although pneumoventricle is common immediately afte ther shunting procedure, delayed tension pneumocephalus or pneumoventricle following CSF diversion is an extremely rare complication.

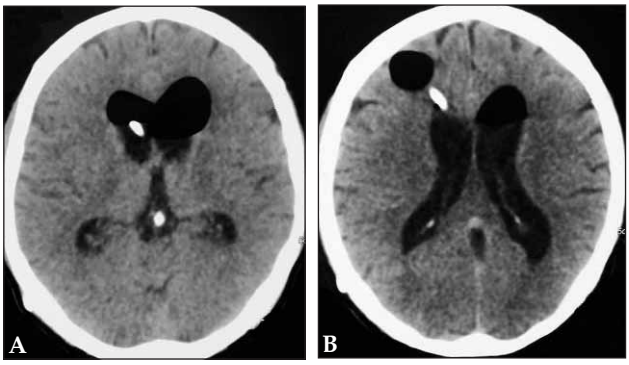

Figure 3A: *CT scan demonstrates the marked reduction of air in both lateral ventricles. Tip of the ventricular catheter is in cerebrospinal fluid.*
Figure 3B: *CT scan demonstrated a little reducing of air beneath the burrhole.*

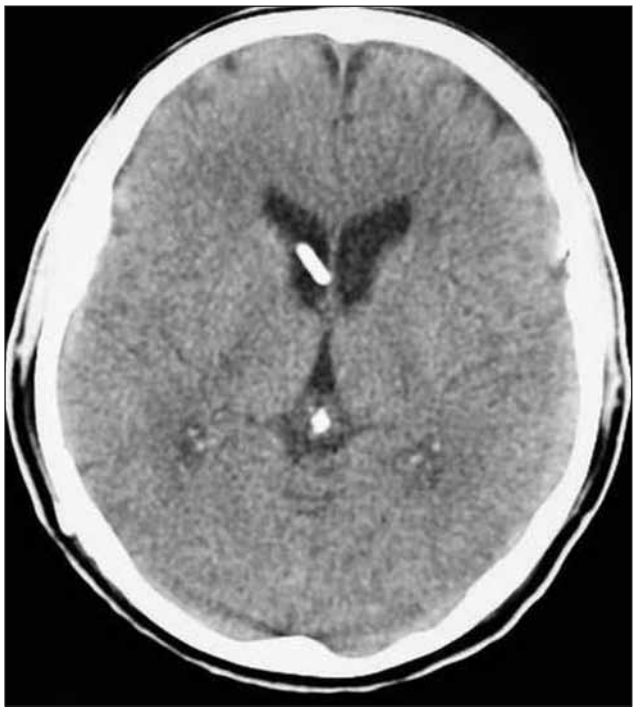

Figure 4: *Air in ventricles totally disappeared on 1-month follow-up CT scan.*

We believe that the possible mechanism of pneumocephalus development is based mainly on two factors;

1) The presence of a defect in the dura and skull causing air inflow with a "one-way ball valve mechanism".

2) A decrease in intracranial pressure (ICP) causing a pressure imbalance between intracranial and extracranial spaces.

Air entry from the skull base defect has been determined in almost all previously reported cases (4,6,7,8,12). Based on this knowledge, many authors have suggested the following mechanism for the development of pneumocephalus: Long-standing intracranial hypertension accompanying the hydrocephalus causes thinning and erosion of the dura mater and the skull base (6,11,12). Gliotic brain tissue and cicatrized meninx may possibly invaginate into this defect and the defect may then

not allow entry of air to the intracranial cavity during the long-lasting intracranial hypertension. After the shunting procedure, this fistula may act as a one-way valve and allow passing inward of air due to the drop in intracranial pressure. The negative pressure inside may "suck in" air from outside by a one-way valve mechanism in the duramater. Failure of air to leave the cranial vault leads to the development of tension pneumocephalus. Repetition of this process leads to accumulation of air in the intracranial compartment and consequently causes a porencephalic cyst and pneumoventricle after communication with the lateral ventricle during long-lasting inflammation. The intracranial pressure may fall to subatmospheric levels, especially in the erect position, if the shunting system does not include an antisiphon device (4). Medium pressure shunt systems are generally inserted in hydrocephalus following subarachnoid hemorrhage. The usage of low-pressure valves probably causes pneumoventricle more frequently in these patients. In the same way, we can speculate that adjustable valves may allow the control of intraventricular pressure and the management of pneumoventricle.

The improved neurological presentations and resolved pneumocephalus after closure of the skin defect under local anesthesia and simple bed rest may support this explanation. We reported here a unique case where the source of entry was in the cranial vault, and air entered from the scalp defect in contrast to all previously reported cases.

Management of pneumocephalus is based on the treatment of elevated intracranial pressure, treatment of meningitis, shunt management and finally closure of the main source of air inflow. Tension pneumocephalus causing acute elevation of intracranial pressure may lead to severe neurological deterioration. This potentially life-threatening condition requires immediate correction of ICP by simple aspiration or continuous drainage. Most cases of small pneumocephalus do not require any surgical management. Intracranial air also often resolves spontaneously. The treatment must be focused on direct surgical closure of the main site of air entry (6,7,8,12). Imaging modalities play a crucial role in identification of the cause for the pneumocephalus. In 1994, Kawajiri reported two cases and the reviewed another 16 cases in the literature with pneumocephalus after CSF shunting where the source of entry was fistulae in tegmen tympani, petrous bone, frontal sinus, ethmoid plate or sella turcica. He emphasized that surgery demonstrated a direct communication between the air cells and ventricles in almost all patients (6). Many authors believe that the treatment should mainly be focused on the surgical closure of the site on air entry. If the site of fistula cannot be established, dural repair in the most likely site of the fistula is recommended (1,4,12). The etiology of pneumocephalus has to be verified clearly in most cases with careful clinical evaluation and the air entrance site may be searched by thin slice CT. Porencephalic cysts may indicate the source as the cysts tend to locate beneath fistulae (4,6,12). CT scan is the diagnostic method of choice. It is sensitive for detecting intracerebral/intracranial air as small as 0,5 cc. Furthermore, 3D CT may show the defect of basis cranii as a possible site of air entry. We used thin coronal CT slices for determining the probable bony defect and could not find any skull base defect. The dehiscence of the wound just above the burr-hole was a possible entry point of air. CT scans demonstrated isolated air in addition to a pneumoventricle just under the burr-hole. We closed the wound to inhibit the air inflow as recommended. Some authors recommend modification of the shunt system with the use of high-pressure shunt or antisiphon devices would avoid pneumocephalus. Pneumocephalus has still been reported despite the use of antisiphon devices.

All patients with pneumocephalus should be closely monitored for occurrence of infection. CSF evaluation is recommended to evaluate possible infection in all these patients. Prophylactic use of antibiotics in these cases is still controversial (2). Broad-spectrum antibiotics are indicated and removal of the whole shunt system is strongly recommended although is patent if an infection is suspected clinically or detected (14). A sterile and well-functioning shunt system should not be revised (4,12). A CSF sample did not show infection in the present case and we decided it was unnecessary to revise the system.

We found only one reported case similar to present case in the literature. Sasani and al. reported a case with delayed and isolated intraventricular tension pneumocephalus after shunting due to unhealing wound (13). According to our knowledge this is the second report of an unusual complication of the shunt procedure.

In conclusion, the present case highlights the uncommon complication of a shunting procedure. Although tension pneumoventricle is a rare condition after CSF diversion procedures, a probable diagnosis of tension pneumocephalus was made as the subsequent CT scans after closure of scalp defect showed resolving pneumocephalus that coincided with an improvement in the neurological patient's condition.

We recommend that the skin incision should be carefully closed during primary shunt surgery to prevent air inflow with a "one way ball valve mechanism" via scalp-ventricle fistula.

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