Brain Tumors Complicated by Pneumocephalus Following Cerebrospinal Fluid Shunting

—Two Case Reports—

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Abstract

A 16-year-old male with cerebellar astrocytoma and a 29-year-old male with convexity meningioma, both complicated by hydrocephalus, developed pneumocephalus following tumor removal and shunt emplacement. Both patients underwent ventriculoperitoneal shunt emplacement and total tumor removal, and were discharged from hospital in good condition. After 1 year or 10 months, respectively, pneumocephalus and porencephalic cysts in the temporal lobes developed. Since neuroradiological examination could not locate the bone defects in the skull base, the dura mater covering the anterior cranial fossa was repaired. Fistulae were observed in the cribriform plate in both patients. Postoperatively, one patient showed persistent cerebrospinal fluid rhinorrhea. Intratympanic cerebrospinal fluid retention was demonstrated by tympanic puncture ipsilateral to a porencephalic cyst. The other patient again developed pneumocephalus. Magnetic resonance imaging indicated that the base of the porencephalic cyst was in contact with the tegmen tympani. The dura mater covering the floor of the middle cranial fossa was repaired in both patients, and the symptoms ameliorated. The bone defects in the skull base were apparently deeply involved in the development of the porencephalic cysts, indicating that such cysts provide useful information for locating cranial bone defects.

Key words: hydrocephalus, ventriculoperitoneal shunt, pneumocephalus

Introduction

Cerebrospinal fluid leakage and pneumocephalus generally occur after trauma, but may also accompany other diseases. Bone and dural defects in the skull base always underlie these conditions, but the sites are occasionally very difficult to identify. We describe two patients with brain tumors and long-standing hydrocephalus who suffered cerebrospinal fluid rhinorrhea and pneumocephalus following tumor removal and ventriculoperitoneal (VP) shunt procedures.

Case Reports

Case 1: A 16-year-old male had suffered from severe headaches precipitated in the morning for 1 year prior to presentation. Gait disorder had devel-

oped in the month previously. Neurological examination revealed marked cerebellar ataxia. Computed tomographic (CT) scans demonstrated a low-density area accompanied by a mass effect in the right cerebellar hemisphere, suggesting cerebellar astrocytoma. Hydrocephalus was also present. A VP shunt was emplaced and the tumor removed. He was discharged in good condition without cerebellar ataxia.

One year later, he became aware of an abnormal sound originating within his head, and revisited our hospital. There was no new neurological deficit. CT scans revealed air in both lateral ventricles, and a porencephalic cyst in the left temporal lobe (Fig. 1). The air probably entered the lateral ventricles from the air cells through the bone and dural defects because the VP shunt had decreased the intracranial pressure. Therefore, an anti-siphon device was added to the shunt system. Air no longer entered, and the pneumocephalus disappeared. Ten days later, cerebrospinal fluid rhinorrhea developed.

Fig. 1 Case 1. *left*: CT scan, showing a porencephalic cyst (arrow). *right*: CT scan, showing air in both lateral ventricles.

Fig. 2 Case 2. *left*: CT scan, showing air in both lateral ventricles. *right*: CT scan, showing a porencephalic cyst (arrow).

Neuroradiological examination could not locate the fistula, so we repaired the dura mater covering the floor of the anterior cranial fossa which was the most probable site of the fistula. During surgery, three fistulae about 2 mm in diameter were found in the cribriform plate and the dura mater in contact with the plate. The flow of cerebrospinal fluid rhinorrhea, however, decreased only by half after surgery.

Four months later, tympanic puncture revealed cerebrospinal fluid accumulation in the left tympanic cavity. Cerebrospinal fluid leakage *via* the left petrous bone was suspected. We performed left temporal craniotomy to examine the floor of the middle cranial fossa, but no site responsible for such leakage could be found, although it may have been too small to identify even with the aid of an operating microscope. After the floor of the relevant area of the middle fossa was repaired with temporal fascia, cerebrospinal fluid rhinorrhea disappeared. There has been no recurrence during the following 7 years.

Case 2: A 29-year-old male had shown abnormalities in speech and behavior, and changes in personality during the month before presentation. CT scans revealed hydrocephalus and a meningioma affecting the right lateral convexity of the skull. The tumor was totally removed and a VP shunt emplaced. He was discharged in good condition.

Headache and cerebrospinal fluid rhinorrhea developed 10 months later, followed by consciousness disturbance. CT scans demonstrated a large volume of air in both lateral ventricles, but the fistula could not be located. An operation to repair the dura mater covering the floor of the anterior cranial fossa discovered fistulae in the right cribriform plate and the dura mater in contact with the plate. Postoperatively, cerebrospinal fluid rhinorrhea and

Fig. 3 Case 2. MR image, showing a porencephalic cyst in contact with the tegmen tympani (arrow).

pneumocephalus disappeared, and he was discharged.

He was readmitted 1 month later complaining of a "splashing sound" inside his head. There was no cerebrospinal fluid rhinorrhea. CT scans again demonstrated a large volume of air in the ventricles (Fig. 2 *left*). A porencephalic cyst was identified in the left temporal lobe (Fig. 2 *right*). Magnetic resonance (MR) images indicated that the base of the porencephalic cyst was in contact with the tegmen tympani (Fig. 3). Therefore, the inferior horn of the lateral ventricle, porencephalic cyst, and tympanic cavity probably communicated through this route.

Through a left frontotemporal craniotomy, the dura mater was incised to expose the base of the temporal lobe. There was a small fistula about 3 mm in

Fig. 4 Case 2. *upper*: Intraoperative photograph, showing brain tissue invaginating into a fistula in the petrous bone. D: dura mater, I: invaginating brain tissue, M: middle cranial fossa, T: temporal lobe. *lower*: Intraoperative photograph, showing a porencephalic cyst communicating with the inferior horn. P: porencephalic cyst, T: temporal lobe, V: inferior horn of the lateral ventricle.

diameter in the dura mater and petrous bone, and the brain tissue had invaginated into the fistula (Fig. 4 *upper*). The porencephalic cyst was opened by an approach from the brain surface. Air was released and the brain inflation disappeared. The inferior horn of the temporal lobe was visualized through the fistula deep in the porencephalic cyst (Fig. 4 *lower*). The brain tissue invaginating into the fistula was removed, and a muscle segment 4 cm in diameter positioned firmly over the floor of the middle cranial fossa, and fixed into place using fibrin glue. Pneumocephalus disappeared postoperatively. There has been no recurrence during the following 2 years.

Discussion

Pneumocephalus has developed after cerebrospinal fluid shunt for chronic hydrocephalus in only 18

cases^{2-8,10-15)} including ours (Table 1). The patients ranged widely in age from 12 to 77 years. The diseases underlying hydrocephalus were brain tumors in nine patients (including 7 of posterior cranial fossa tumors), and aqueductal stenosis in seven. The other two patients had communicating hydrocephalus of unknown causes. In all patients, the hydrocephalus had persisted for a fairly long period. Air was retained subdurally in two cases, and intracerebrally and intraventricularly in the other 16. Surgery demonstrated direct communication between the air cells and ventricles in almost all patients.

The delay from cerebrospinal fluid shunting to the discovery of pneumocephalus was fairly long, *i.e.* 1 week to 5 years, possibly due to the time required to form a pathway on the skull base for communication between the ventricles and air cells. Symptoms of pneumocephalus were headache in eight patients, consciousness disturbance in eight, motor paralysis in six, gait disorder in two, reduced intelligence in three, recent memory disturbance in two, cerebrospinal fluid rhinorrhea in two, and convulsion in one. Intracranial "splashing sounds" were the primary complaint in seven cases. This is also called "bruit hydroaerique," and is a pathognomonic symptom of pneumocephalus.⁷⁾

Pneumocephalus usually occurs as a result of head injury, with air entering from the fractured base of the skull. In non-traumatic pneumocephalus, however, the source of air entry was fistulae in the tegmen tympani or petrous bone in nine patients (bilaterally in 2), the wall of the frontal sinus in three, the upper wall of the ethmoidal sinuses (ethmoidal plate and cribriform plate) in six, the sella turcica in one, and the crista galli in one. The fistula site was unknown in one case. Three patients including ours had multiple fistulae. The structures in the middle cranial fossa were most frequently affected. In particular, all nine patients with brain tumors had fistulae in the tegmen tympani or petrous bone. Furthermore, porencephalic cysts occurred just above the fistulae in seven of these patients (bilaterally in 2). The other two cases were not well described. Interestingly, these cysts were connected with the fistulae in the middle cranial fossa, and also communicated with the lateral ventricles. Porencephalic cysts were also found in the frontal lobe in three non-tumor cases, and all communicated with the fistulae and lateral ventricles. Therefore, porencephalic cysts indicate that the bone and dura mater beneath have defects, through which pneumocephalus develops. Porencephalic cysts are then very helpful in identifying the site of the fistulae

Table 1 Summary of reported cases of non-traumatic, shunt-associated pneumocephalus

Author (Year)	Age/ Sex	Cause of hydrocephalus	Period of shunting	Site of fistula	Porencephalic cyst
Pitts <i>et al.</i> (1975) ¹¹⁾	21/?	cerebellar astrocytoma	1 yr	tegmen tympani	
Little and Maccarty (1976) ⁸⁾	22/M	aqueductal stenosis	2 mos	frontal sinus	frontal lobe
Ikeda <i>et al.</i> (1978) ⁶⁾	22/F	aqueductal stenosis	3 mos	frontal sinus	frontal lobe
Steinberger <i>et al.</i> (1979) ¹³⁾	29/F	aqueductal stenosis	1 wk	frontal sinus	
Jooma and Grant (1983) ⁷⁾	12/M	aqueductal stenosis	9 mos	ethmoidal plate	frontal lobe
Sunada <i>et al.</i> (1984) ¹⁴⁾	15/M	aqueductal stenosis	2 mos		
Ruge <i>et al.</i> (1985) ¹²⁾	28/M	aqueductal stenosis	3 yrs	cribriform plate	
Goffin and Plets (1985) ⁵⁾	35/F	pineal tumor	8 mos	tegmen tympani	temporal lobe
Tanaka <i>et al.</i> (1986) ¹⁵⁾	47/M	CP angle tumor	2 wks	tegmen tympani	temporal lobe
	39/M	CP angle tumor	8 mos	petrous bone	temporal lobe
Ferrante <i>et al.</i> (1988) ⁴⁾	51/F	aqueductal stenosis	15 days	cribriform plate	
Coraddu <i>et al.</i> (1989) ³⁾	67/F	CP angle tumor	5 yrs	petrous bone	
Mylonas (1991) ¹⁰⁾	77/F	communicating hydrocephalus	6 wks	sella turcica	
	24/F	communicating hydrocephalus	2 yrs	crista galli, ethmoidal plate	
Aoyama <i>et al.</i> (1991) ²⁾	43/F	convexity meningioma	2 mos	petrous bone	temporal lobe
	55/M	cavernoma of the fourth ventricle	3 mos	petrous bone	temporal lobe
Present Case 1	16/M	cerebellar astrocytoma	1 yr	tegmen tympani, cribriform plate	temporal lobe
Present Case 2	29/M	convexity meningioma	10 mos	tegmen tympani, cribriform plate	temporal lobe

CP: cerebellopontine.

which may cause pneumocephalus and cerebrospinal fluid leakage.

What mechanism is involved in the formation of porencephalic cysts? Ahren and Thulin¹⁾ suggested the following mechanism, based on the tegmen tympani demonstrating a small defect in 21% of the population, and is very thin in 16%. Intracranial hypertension accompanying chronic hydrocephalus causes thinning and partial defects of the dura mater covering the skull base. A portion of the brain tissue invaginates into a pre-existing or pressure-induced fistula in the tegmen tympani, and necrotizes during the long-lasting intracranial hypertension. Usually the necrotized brain tissue or cicatrized meninx plugs the fistula. If intracranial pressure is lowered by shunting at this stage, air is introduced from the air cells into the brain parenchyma through a "ball

valve" mechanism involving the necrotized tissue. Repetition of this process increases the amount of air retained in the brain parenchyma, and consequently causes a porencephalic cyst. If this cyst is increasingly inflated, communication will be established with the inferior horn of the lateral ventricle, and air introduced into the ventricle.

In two previous cases,^{6,8)} porencephalic cysts were already present in the frontal lobe before the cerebrospinal fluid shunt was emplaced. In these cases, the possible mechanism of cyst formation was as follows. Hydrocephalus increasingly elevated the intracranial pressure, resulting in ventricular wall cracking in an area near where the brain tissue had invaginated into a fistula formed on the skull base. Cerebrospinal fluid entered through the cracks, gradually split the brain parenchyma to form a

porencephalic cyst, and reached the fistula on the skull base, causing cerebrospinal fluid rhinorrhea. This process may be similar to the mechanism of "puncture porencephaly,"⁹⁾ which occasionally occurs when shunt dysfunction following ventricular drainage or shunt surgery elevates intracranial pressure.

Bone defects in the skull base may cause pneumocephalus or cerebrospinal fluid leakage, but are difficult to identify. Non-traumatic defects are frequently observed in the floor of the middle cranial fossa as well. Porencephalic cysts indicate that the structure beneath the cyst is likely to have a defect.

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