

Endoscopic Surgery for Hemorrhagic Pineal Cyst Following Antiplatelet Therapy: Case Report

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Abstract

Pineal cysts of the third ventricle presenting with acute obstructive hydrocephalus due to internal cystic hemorrhage are a rare clinical entity. The authors report a case of a 61-year-old man taking antiplatelet medication who suffered from a hemorrhagic pineal cyst and was treated with endoscopic surgery. One month prior to treatment, the patient was diagnosed with a brainstem infarction and received clopidogrel in addition to aspirin. A small incidental pineal cyst was concurrently diagnosed using magnetic resonance (MR) imaging which was intended to be followed conservatively. The patient presented with a sudden onset of headache and diplopia. On admission, the neurological examination revealed clouding of consciousness and Parinaud syndrome. Computerized tomography (CT) scans demonstrated a hemorrhagic mass lesion in the posterior third ventricle. The patient underwent emergency external ventricular drainage with staged endoscopic biopsy and third ventriculostomy using a flexible videoscope. Histological examination revealed pineal tissue with necrotic change and no evidence of tumor cells. One year later MR imaging demonstrated no evidence of cystic lesion and a flow void between third ventricle and prepontine cistern. In patients with asymptomatic pineal cysts who are treated with antiplatelet therapy, it is important to be aware of the risk of pineal apoplexy. Endoscopic management can be effective for treatment of hemorrhagic pineal cyst with obstructive hydrocephalus.

Key words: pineal cyst, pineal apoplexy, endoscopic surgery, endoscopic third ventriculostomy

Introduction

Pineal cysts are often asymptomatic and encountered incidentally on computerized tomography (CT) scans or magnetic resonance (MR) imaging.^{1,2,22)} However, infrequently these cysts can cause obstruction of cerebrospinal fluid (CSF) flow at the entrance of aqueduct due to cystic expansion and/or intracystic hemorrhage.^{3–6,8–11,13–21,23,24,26)} The authors report a patient undergoing antiplatelet therapy for brainstem infarction who was presented with a symptomatic hemorrhagic pineal cyst.

Case Report

A 61-year-old man was presented with sudden onset of headache and diplopia. The patient had a prior history of uncontrollable diabetes mellitus. One month prior to admission, the patient was diagnosed with a brainstem infarction (Fig. 1A) and treated with clopidogrel (75 mg)

in addition to his regular medications including aspirin (100 mg). At that time, MR images demonstrated an incidental small cyst in the pineal region without ventriculomegaly (Fig. 1B). The patient was treated conservatively with the intention of outpatient follow-up.

On admission, he complained of headache and the neurological examination revealed clouding of consciousness and Parinaud syndrome. Initial CT scans demonstrated a high density mass lesion in the pineal region and dilatation of the lateral/third ventricles with intraventricular hemorrhage (Fig. 2A). MR imaging suggested acute hemorrhage with iso intensity on T₁-weighted (WI), low intensity on T₂-WI without enhancement on Gd-enhanced T₁-WI sequence (Fig. 2B, C). On Day 2, the patient's consciousness level declined and CT scans demonstrated acute obstructive hydrocephalus which was emergently treated with external ventricular drainage.

Three days later, the patient underwent endoscopic surgery via a right frontal burr hole using a flexible videoscope (VEF-V; Olympus Co., Tokyo). The cyst wall was observed after removal of clot of third ventricle (Fig. 3A).

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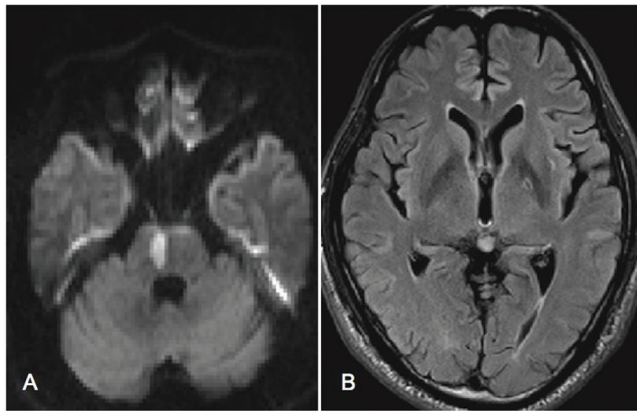


Fig. 1 Magnetic resonance (MR) image 1 month before onset. **A:** Axial diffusion-weighted MR image showing a high intensity lesion in the brainstem. **B:** Axial fluid attenuated inversion recovery MR image showing a small high intensity lesion in the pineal region.

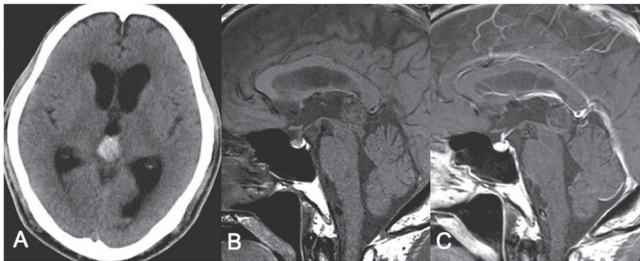


Fig. 2 **A:** Initial computerized tomography (CT) scan showing a high density mass lesion in the posterior third ventricle with evidence of intraventricular hemorrhage. **B, C:** Sagittal T₁-weighted (**B**) and Gd-enhanced (**C**) magnetic resonance (MR) images showing an iso intensity mass lesion without enhancement.

A tissue biopsy of the cyst wall was performed using grasping forceps. Although minor bleeding from the vascularized parts of the cyst occurred during removal of intracystic hematoma, complete hemostasis was obtained following artificial CSF irrigation (Fig. 3B). Finally, endoscopic third ventriculostomy (ETV) was performed in the standard manner.

Histological assessment revealed pineal tissue with necrotic change and dispersed localized sections of vessel wall thickening. The typical three-layered pineal cystic structure was not observed. Inflammatory reaction was detected adjacent to the hemorrhagic area. There was no evidence of tumor cells in any of the specimens (Fig. 3C). One year after surgery, MR imaging demonstrated membranous tissue obstruction at the entrance of the aqueduct, a post-ETV CSF flow void between the third ventricle and prepontine cistern, and evidence of any cystic lesion in the posterior third ventricle (Fig. 4A, B). During the follow-up period, the patient did not experience further

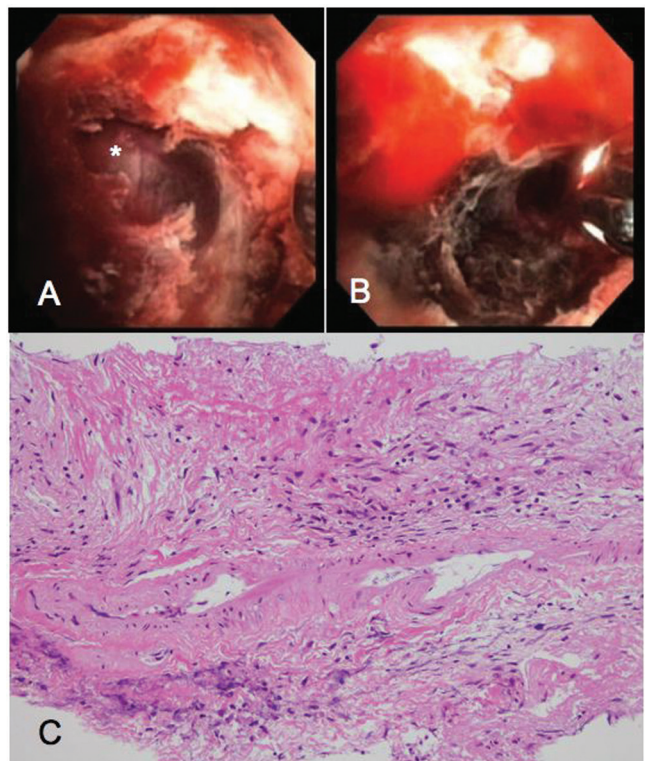


Fig. 3 Intraoperative photograph of the endoscopic surgery for hemorrhagic pineal cyst. **A:** The cyst wall (asterisk) was exposed by removal of clot around cyst. **B:** After resection of the cyst wall, intracystic hematoma was removed with grasping forceps. **C:** Photomicrograph of cyst specimen revealing pineal tissue with necrotic change. H & E, $\times 400$.

headache or neurological deficits. Postoperatively antiplatelet medication was reduced to a single drug (clopidogrel) for prevention of brainstem infarction.

Discussion

Pineal cysts are typically asymptomatic and incidentally detected on neuroimaging studies. Sawamura et al. reported that asymptomatic pineal cysts more than 5 mm diameter account for 1.3% on MR imaging studies.²²⁾ These epidemiologic features were predominant in younger females. Recently Al-Holou et al. also reported similar findings of 2% and 1% prevalence in younger and adult patients, respectively.^{1,2)}

These cysts usually become symptomatic due to compression of brain tissue or blockage of CSF flow from cystic expansion. Intracystic hemorrhage leading to pineal apoplexy has been reported only occasionally. Previous reported cases with progressive symptoms due to hemorrhage are summarized in Table 1.^{3,4,6,9-11,13-21,23)} Hemorrhagic pineal cysts have been found in a broad spectrum in the age group from newborn infants to senile

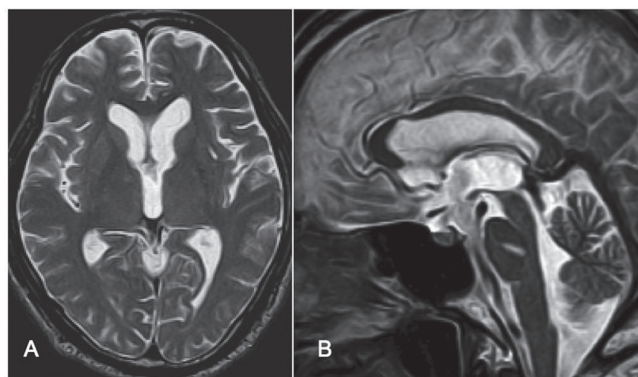


Fig. 4 Axial (A) and sagittal (B) T₂-weighted magnetic resonance (MR) images 1 year after surgery. There was no evidence of residual cystic lesion in the posterior third ventricle. The entrance of the aqueduct was obstructed by membrane and a flow void was demonstrated between the third ventricle and the prepontine cistern.

patients. The grade or extent of hemorrhage is also variable, ranging from minor intracystic xanthochromic fluid levels to intraventricular hemorrhage.

In 1976, Apuzzo et al. reported one case of pineal apoplexy due to hemorrhagic pineal cyst under anticoagulant therapy.³⁾ Sarikaya-Seiwert et al. also suggested a potentially increased risk of anticoagulation-induced hemorrhage in pineal cysts.²¹⁾ However, in the majority of reported cases to date, the precise etiology of bleeding has been unclear. In our case, the patient received dual antiplatelet therapy (clopidogrel in addition to aspirin) for brainstem infarction. In a multi-center trial of antiplatelet therapy for cerebral stroke, intracranial hemorrhage was more frequent in patients treated with both aspirin and clopidogrel than clopidogrel alone.⁷⁾ Based on these rare cases, it is advisable to inform patients with pineal cysts treated with anticoagulant or antiplatelet therapy of the possible risk of intracystic

Table 1 Summary of reported cases of hemorrhagic pineal cyst with progressive symptoms

Authors (Year)	Age/Sex	Symptom & Sign	Cause of hemorrhage	Treatment	Outcome
Apuzzo (1976)	56/M	HA, lethargy, ataxic gait	anticoagulant therapy	craniectomy, total excision	nystagmus, ataxia
Higashi et al. (1979)	51/F	HA, LOC	unknown	craniotomy, total excision, V-P shunt	gaze palsy
Osborn et al. (1989)	30/M	HA, Parinaud syn	unknown	shunt, craniectomy, subtotal excision	asymptomatic
Klein & Rubinstein (1989)	30/F	HA, gaze palsy	unknown	total excision	lost to follow-up
Turtz et al. (1995)	21/M	HA, Parinaud syn	unknown	stereotactic endoscopic fenestration	asymptomatic
Koenigsberg et al. (1996)	21/M	HA, Parinaud syn	drug abuse	endoscopic cyst incision	asymptomatic
Mena et al. (1997)	20/M	Parinaud syn, Papilledema	unknown	excision	12 years alive
Swaroop et al. (1998)	35/F	HA, papilledema, ataxia	unknown	craniotomy, total excision	transient gaze palsy
Mukherjee et al. (1999)	70/M	HA, hearing loss, LOC	unknown	V-P shunt, partial excision	hearing recovery
Di Chirico et al. (2001)	16/F	HA, papilledema	unknown	ETV	asymptomatic
Michielsen et al. (2002)	30/M	HA, visual deficit	unknown	stereotactic endoscopic subtotal excision	asymptomatic
McNeely et al. (2003)	4/F	HA, lethargy	unknown	endoscopic subtotal excision	asymptomatic
	12/F	HA, syncope	unknown	drainage, ETV, craniotomy total excision	asymptomatic
Avery et al. (2004)	71/F	Syncope	anticoagulant therapy	shunt	asymptomatic
Patel et al. (2005)	29/F	HA, visual disturbance	unknown	craniotomy, total excision	asymptomatic
Nimmagadda et al. (2006)	10D/F	macrocephaly	unknown	observation	asymptomatic
Majeed et al. (2007)	10/F	HA, gaze palsy	unknown	craniotomy, total excision	asymptomatic
Sarikaya-Seiwert et al. (2009)	16/F	HA, impaired concentration	unknown	craniotomy, total excision	asymptomatic
	16/F	HA, papilledema	unknown	craniotomy, total excision	asymptomatic
	38/F	HA, impaired concentration	anticoagulant therapy	craniotomy, total excision	malignant tumor
Present case	61/M	HA, Parinaud syn	antiplatelet therapy	endoscopic partial excision, ETV	symptoms due to brainstem infarction

ETV: endoscopic third ventriculostomy, HA: headache, LOC: loss of consciousness, V-P: ventriculoperitoneal.

hemorrhage and the potential associated complication of acute obstructive hydrocephalus. Furthermore, based on our case, we cannot ignore the possibility of the effect of uncontrolled diabetes as a contributing to the increased risk of hemorrhage.

Various surgical approaches, including microsurgical resection, stereotactic aspiration, and endoscopic approach have been employed for the treatment of symptomatic pineal cyst.^{5,6,9–17,20,21,23–26} Among these options, the cases of spontaneous cyst regression following only ventriculoperitoneal shunt or ETV have been reported.^{4,6} A detailed explanation of the mechanism of this phenomenon has been described as follows: a change in the pressure gradient between the cyst and the ventricle cavity as a result of treatment to normalize ventricular pressure leads to displacement of cyst fluid into the third ventricle space.⁶ However, in hemorrhagic cases, it is important to differentiate from neoplasma such as glioma, pineocytoma, pineoblastoma, and germ cell tumors. Therefore, histological diagnosis is crucial, and we believe that microsurgical or endoscopic resection is an ideal approach to hemorrhagic pineal cysts.

Several reports have shown that an endoscopic surgery can be useful for the treatment of symptomatic pineal cysts.^{5,11,16,24,25} Michielsen et al. reported 4 patients with pineal cyst who underwent endoscopic surgery via the ventricles.¹⁶ They described that the endoscope was an diagnostic and surgical tool, and even total cyst resection is possible by this method. In our case, only partial removal of the intracystic hematoma was performed because of the bleeding that encountered in vascularized parts of the cyst. Nevertheless, postoperatively, the clinical signs and symptoms of obstructive hydrocephalus resolve and almost complete radiological disappearance of the hematoma and cyst cavity was achieved. In this sense, hemorrhagic pineal cysts can be successfully treated only by partial endoscopic removal of the cyst wall.

Michielsen et al. suggested that ETV for obstructive hydrocephalus due to cyst expansion was not necessary because normal CSF flow recovered immediately after cyst reduction.¹⁶ However, Uschold et al. who reported use of a supracerebellar infratentorial endoscopic approach claimed that posterior ETV (between third ventricle and quadrigeminal cistern) was beneficial for treatment of cases of the incomplete cyst resection.²⁵ In our case, postoperative MR imaging revealed residual cyst wall adherent to the ependymal layer around the entrance of the aqueduct. ETV for persistent blockage of CSF flow in hemorrhagic pineal cysts can be an effective adjunctive treatment option.

Conflicts of Interest Disclosure

None declared.

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