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Case Report

Iatrogenic pseudoaneurysms associated with cerebrospinal fluid diversion procedures

Alan Chalil[1], Michael D. Staudt[1], Stephen P. Lownie[1,2]

[1]Departments of Clinical Neurological Sciences, [2]Medical Imaging, London Health Sciences Centre, Western University, London, Ontario, Canada

E-mail: Alan Chalil - Alan.Chalil@lhsc.on.ca; *Michael D. Staudt - Michael.Staudt@londonhospitals.ca; Stephen P. Lownie - Steve.Lownie@lhsc.on.ca
*Corresponding author

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Abstract

Background: Cerebrospinal fluid diversion procedures, including ventriculoperitoneal (VP) shunt and external ventricular drain insertion, are common treatments for hydrocephalus. Common complications include obstruction, infection, and hemorrhage. Pseudoaneurysm formation secondary to catheter insertion is a distinctly rare complication, and usually involves the anterior cerebral artery or branches of the external carotid artery (superficial temporal artery or middle meningeal artery).

Case Description: We present the case of a fusiform pseudoaneurysm in a 36-year-old female, which arose from a branch of the middle cerebral artery following VP shunt insertion. Parenchymal and intraventricular hemorrhage at the catheter insertion site developed 15 days postoperatively. The VP shunt was removed, and the aneurysmal segment was coagulated and occluded. Use of a limited dural opening during ventricular catheter placement may have been a factor in pseudoaneurysm formation.

Conclusions: The literature regarding this rare complication is reviewed. Careful consideration should be given to vascular anatomy when planning shunt insertions, and a cruciate dural opening for cortical visualization and coagulation may help avoid this complication. Prompt identification and management of iatrogenic pseudoaneurysms is essential to avoid re-bleeding and associated hemorrhagic complications.

Key Words: Complications, external ventricular drain, hemorrhage, pseudoaneurysm, traumatic aneurysm, ventriculoperitoneal shunt

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INTRODUCTION

Cerebrospinal fluid (CSF) diversion procedures, including external ventricular drainage (EVD) and ventriculoperitoneal shunt (VP) insertion, are quite common in neurosurgery. These procedures are indicated for treatment and monitoring of increased intracranial pressure secondary to hydrocephalus, trauma, intraventricular hemorrhage, subarachnoid hemorrhage, or shunt failure.[2,7,9,11,22] Common associated risks include infection, hemorrhage and

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obstruction, and less commonly seizures, premature suture closure (in pediatric patients), and seeding of metastases.[34]

Hemorrhage rates following EVD catheter insertion reportedly average around 10%,[10] and include intraventricular, intraparenchymal, or subarachnoid hemorrhage. Hemorrhage can be attributed to multiple factors including a pre-existing coagulopathy, anticoagulant or antiplatelet use, overdrainage of CSF causing injury to bridging veins, the presence of a pre-existing lesion such as a tumor or a vascular malformation, and aneurysmal re-bleeding.[24,26] There are only rare reported cases in which insertion of an EVD or VP shunt was associated with development of an iatrogenic pseudoaneurysm. Traumatic aneurysms are a rare entity themselves, accounting for less than 1% of all intracranial aneurysms.[27]

The literature and management strategies regarding this rare complication are reviewed. The current article describes the illustrative case of an adult female who developed a multicompartmental hemorrhage due to an iatrogenic pseudoaneurysm following a VP shunt insertion. Etiology, investigations, and management are discussed. Prompt diagnosis and treatment are essential in avoiding the high morbidity associated with these rare complications.

CASE DESCRIPTION

A 36-year-old female with a history of untreated hypertension presented to the emergency department with severe headache and a decreased level of consciousness. Computed tomography (CT) of the head demonstrated a large cerebellar parenchymal hemorrhage [Figure 1a, b], but CT angiogram was negative for a vascular malformation, aneurysm, or venous sinus thrombosis. A right frontal EVD was inserted, and the patient underwent suboccipital decompression and clot evacuation, with clinical improvement. Subsequent vascular imaging, including magnetic resonance (MR) and digital subtraction angiography was negative for pathology [Figure 2a-c]. A thorough hematological workup was also negative. As such, this was considered a hypertensive hemorrhage.

Five weeks following admission, the development of communicating hydrocephalus required the insertion of a VP shunt [Figure 1c, d]. Following burr hole placement, the dura was coagulated and monopolar cautery along a forceps was used to puncture the dura and cauterize the pia. After a single pass, good CSF egress was observed, and the ventricular catheter was gently advanced to approximately 10 cm. The surgery was well tolerated and the hydrocephalus and level of consciousness improved.

On day 15 following VP shunt insertion, the patient developed a dilated and fixed right pupil. An emergent CT scan demonstrated acute intraparenchymal hemorrhage into the right parietal lobe along the ventriculostomy tract with extensive intraventricular hemorrhage, acute hydrocephalus, and midline shift [Figure 1e, f]. An EVD was inserted through the previous right frontal burr hole, and a CT angiogram followed by a four-vessel angiogram revealed a pseudoaneurysm of a right distal middle cerebral artery (MCA) branch immediately adjacent to the entry point of the ventricular catheter in the right parietal region [Figure 2d-f]. A right parietal mini-craniotomy was performed for coagulation of the

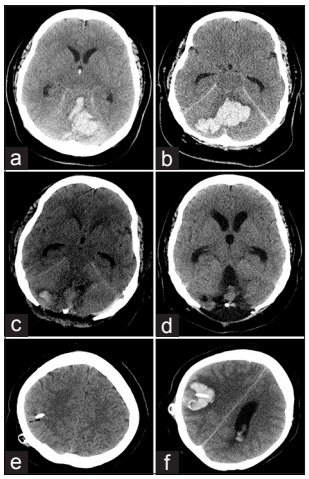

Figure 1: Axial CT imaging demonstrates initial cerebellar hemorrhage with extension into the third and fourth ventricles and effacement of the basal cisterns (a and b). Immediate postoperative imaging following suboccipital decompression demonstrates good hematoma evacuation (c), with delayed development of communicating hydrocephalus (d). Immediate postoperative imaging following VP shunt insertion does not demonstrate any adverse findings (e), and delayed imaging is consistent with hemorrhage along the shunt tract (f)

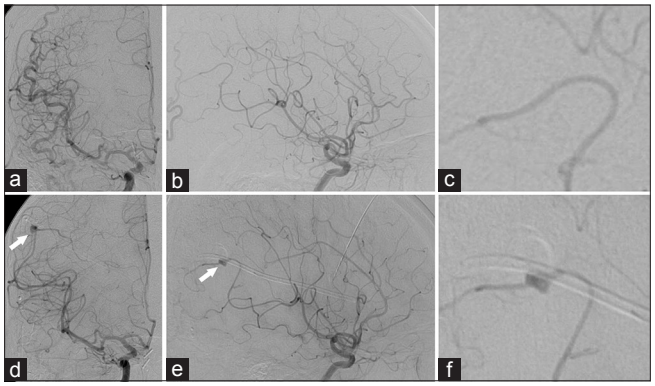

Figure 2: Diagnostic cerebral angiography performed via a right common carotid injection (a: AP; b: lateral; c: lateral zoom) does not demonstrate any aneurysm, arteriovenous malformation, or arteriovenous fistula related to right middle cerebral artery and anterior cerebral artery. Following delayed hemorrhage along shunt tract, right internal carotid injection (d-f) identifies a pseudoaneurysm along the M4 segment of the right middle cerebral artery immediately adjacent to the right parietal ventriculostomy catheter (arrows)

right distal MCA pseudoaneurysm and removal of the VP shunt. Repeat VP shunt placement was performed 2 months following pseudoaneurysm resection. The patient was discharged to a rehabilitation facility and continued to make cognitive and functional improvement. She has since been discharged home with in-home nursing and physiotherapy assistance.

DISCUSSION

Iatrogenic aneurysms occur mostly in conjunction with injury to a cortical vessel, often presenting as acute blood noted upon the "blind" insertion of a catheter through a burr hole. In the current case, however, no blood was observed on initial dural puncture or during catheter passage; the thin tip of a Harris forceps was used in conjunction with monopolar cautery to puncture the dura and coagulate the pia. As with other reported cases (summarized in Table 1), the cortex was not visualized upon completion of the durotomy and prior to catheter insertion. We hypothesize either the monopolar cautery puncture or catheter insertion to have caused the vessel injury.

Etiology and pathophysiology

Traumatic aneurysms are rare, with an incidence of 0.09–0.4% of all intracranial aneurysms;[6,23] iatrogenic aneurysms presumably account for even a smaller portion

Table 1: Pseudoaneurysms in patients undergoing CSF shunting procedures

Author	Age, Sex	Iatrogenic injury	Associated Hemorrhage	Aneurysm/ Pseudoaneurysm Location	Duration to Diagnosis	Management	Outcome
Shirane *et al*. 1999[31]	4 mo, F	VP shunt removal	IVH, SAH	Pseudoaneurysm: left anterior choroidal avulsion	21 weeks	Surgical resection and reconstruction of ICA	N/A
Horowitz *et al*. 2005[15]	7 d, M	VP shunt insertion	IVH, ICH	Pseudoaneurysm: left ACA (A3 segment)	3 weeks	Endovascular coiling	Neurologically intact at 1 year
Jenkinson *et al*. 2006[16]	15 y, F	VP shunt insertion or removal	ICH	Traumatic aneurysm: fusiform left MCA (M4 segment)	6 weeks	Surgical resection	Mild hemiparesis, mild expressive aphasia, right visual neglect, right homonymous lower quadranopsia
Tubbs *et al*. 2006[34]	10 y, M	VP shunt insertion	Bilateral IVH	Pseudoaneurysm: right pericallosal artery (A3 segment)	1 week	Surgical trapping	Mild leg weakness
Chen *et al*. 2012[3]	39 y, M	VP shunt insertion	IVH, ICH	Pseudoaneurysm: left distal callosomarginal artery (A3 segment)	13 days	Glue embolization	No complications
Kosty *et al*. 2013[18]	62 y, M	EVD insertion converted to VP shunt	None	Pseudoaneurysm: right Callosomarginal artery (A3 segment)	12 weeks	Onyx embolization	No complications
Grandhi *et al*. 2013[12]	64 y, M	EVD insertion	None	Pseudoaneurysm: right distal MMA	1 day (lesion noted during diagnostic angiogram)	No treatment	Death
Choudhri *et al*., 2014[4]	40-75 y, one F & two M	EVD insertion	ICH, SAH	Pseudoaneurysm: middle inferior branch of right pericallosal artery (A3 segment)	1 day (lesion noted during endovascular coiling of primary aneurysm)	Endovascular coiling	No complications
		EVD insertion	SAH	Pseudoaneurysm: left PComm/PCA (P1 segment)	1 day	Endovascular coiling	Death
		EVD insertion converted to VP shunt	EDH	Pseudoaneurysm: right MMA	1 day (lesion noted during endovascular coiling of primary aneurysm)	Endovascular coiling + glue embolization	No complications
Raygor *et al*. 2015[28]	58 y, F	EVD Insertion	None	Pseudoaneurysm: right middle frontal artery/ ACA (A3 segment)	10 days	Open surgical resection	Neurologically intact
Current case	36 y, F	VP shunt insertion	IVH, ICH	Pseudoaneurysm: right distal MCA (M4 segment)	15 days	Open surgical resection	Residual cognitive impairment; ongoing rehabilitation

ACA=anterior cerebral artery, EDH=epidural hematoma, EVD=external ventricular drain, ICA=internal carotid artery, ICH=intraparenchymal hemorrhage, IVH=intraventricular hemorrhage, MCA=middle cerebral artery, MMA=middle meningeal artery, PCA=posterior cerebral artery, SAH=subarachnoid hemorrhage, VP=ventriculoperitoneal

of those numbers. Traumatic aneurysms can be divided into two groups: true aneurysms and pseudoaneurysms. True aneurysms occur after an incomplete injury of the arterial vessel wall which involves the inner lamina and media layers. Once the layers are healed, a focal weakness in the arterial wall persists which forms a true aneurysm.[27]

Pseudoaneurysms occur after a focal injury to the vessel wall which produces a hematoma in-between the adventitia and muscularis layers at the site of injury. Subsequent resolution of the hematoma forms the outer layer of the newly formed pseudoaneurysm.[27] White *et al.* demonstrated that inducing puncture injuries in canine arterial walls produced focal defects and thus weakening in the intima and fibrotic changes in the muscularis layer.[35] Histological analysis from the pseudoaneurysm in the current case was not available; however, similar findings were demonstrated in histological analysis of iatrogenic pseudoaneurysms secondary to ICP monitor insertion[20] and stereotactic biopsy.[27]

The delay between the initial vessel insult and pseudoaneurysm formation is highly variable, and ranges from 14 to 21 days or more, with some authors reporting delays of many years.[1,19] In the current case, the pseudoaneurysm was discovered during repeat vessel imaging obtained on day 15 following VP shunt insertion. Pseudoaneurysms tend to be inherently fragile, and carry a mortality rate up to 50%[19] with a recurrent hemorrhage rate of up to 40%; specifically, pseudoaneurysms resulting from puncture injuries are associated with intracerebral hemorrhage rates of up to 80%, if left untreated.[13] Horiuchi *et al.* reported a rupture rate of 72.4% in traumatic MCA aneurysms, with favorable outcomes in 75% of treated patients compared to 37.5% managed conservatively.[14]

Pseudoaneurysms in the literature

Pseudoaneurysms secondary to EVD or VP shunt insertion occur in both pediatric and adult patients [Table 1]. The age at diagnosis ranged from 7 days to 75 years, with 7 males and 5 females in reported cases. However, considering the limited number of cases, these data carry little if any epidemiological significance. Most pseudoaneurysms presented as a combination of subarachnoid and intraventricular hemorrhage, while two pseudoaneurysms presented as local intraparenchymal hemorrhage.

The anterior cerebral artery (ACA) or one of its distal branches was involved in 6 cases of pseudoaneurysm formation following EVD or VP shunt insertion,[3,4,15,18,28,34] with only one case involving a fusiform traumatic aneurysm of the MCA,[16] and one pseudoaneurysm involving the posterior communicating artery.[4] A pseudoaneurysm involving the middle meningeal artery (MMA) presented as an epidural hematoma owing to the anatomical location of the injured vessel.[4] ACA pseudoaneurysms were associated with a medial catheter trajectory; the entry point in these cases was typically located in the middle or posterior cerebral artery territory, suggesting that vessel injury occurred during passage of the catheter with the guiding stylet, rather than during drilling or opening the dura. The one reported case of a fusiform traumatic aneurysm involving the distal MCA likely formed due to an inflammatory reaction from the adjacent shunt catheter over years.[16] Removal of the catheter was hypothesized to be the inciting event for aneurysm rupture. However, the lack of pre-operative vessel imaging makes it hard to determine when the insult to the vessel wall occurred.

Shirane *et al.* reported intraventricular and diffuse subarachnoid hemorrhage secondary to a pseudoaneurysm of the proximal internal carotid artery,[31] which formed as a result of anterior choroidal artery avulsion during catheter removal and not as a direct injury from catheter insertion. Internal carotid artery involvement was often reported as a complication of endoscopic surgeries, such as transphenoidal resection of suprasellar tumors.[5,17,21] Not all pseudoaneurysms were a result of a direct trauma from the endoscope, and one suggested etiology was due to traction on the tumor capsule weakening the posterior cerebral artery vessel wall.

Traumatic aneurysms of the MMA were reported on two occasions in addition to multiple dural arteriovenous fistulas secondary to VP shunt or EVD catheter insertion;[4,12] interestingly, the MMA was involved in all reported cases of catheter-associated fistulas except one. The anatomical location of the MMA, specifically when the burr hole is made lateral to Kocher's point, was hypothesized to be responsible for this high correlation, especially when the procedure causes a dual insult to the MMA and one of the cortical veins, either during drilling, dural opening, or catheter insertion.

Traumatic pseudoaneurysms have also been reported secondary to other "blind" procedures besides EVD and VP shunt insertion. Le *et al.* reported a traumatic MCA aneurysm from the insertion of an ICP monitor after puncturing the dura with an 18-gauge needle.[20] The aneurysm presented as an ICH extending to the lateral ventricle and was managed with craniotomy and excision, similar to the case as described by Shah *et al.*[30] Cortical vessel traumatic aneurysms have also been described following stereotactic biopsy, with both reported cases incidentally identified on follow-up imaging.[27,29] Interestingly, both procedures were complicated by a small amount of bleeding during the initial biopsy. In pediatrics, Overton and Calvin published a report of an MCA pseduoaneurysm presumed to be secondary to multiple right parietal subdural taps in a 9-month-old child.[25] This aneurysm presented as recurrent subdural collections/hematoma, and was only

discovered intraoperatively during a large craniotomy, and was treated through excision and coagulation of the parent vessel.

Scalp vessel injuries commonly involved the superficial temporal artery or facial artery, and were associated with tunnelling of the catheter through the scalp after EVD insertion. Occipital artery pseudoaneurysms have also been reported following retrosigmoid craniotomy.[32,33]

Principles of management

Pseudoaneurysms carry a relatively high risk of rupture, and require prompt treatment once identified. The duration to identify the lesion ranged between immediate (same day) and up to 21 weeks following VP shunt or EVD insertion. Management consists of standard treatments for intracranial aneurysms, including open surgical clipping or coagulation, and endovascular embolization with coils or glue. Conservative management is not recommended in cases of traumatic aneurysms and pseudoaneurysms due to the high rates of catastrophic bleeds and mortality, which approach 50% in conservatively managed patients compared to 18% in treated patients.[1,8,19]

As with other intracranial aneurysms, several factors contribute to determining the treatment modality, including the location of the aneurysm, morphology, the presence or absence of hemorrhage and associated hematoma, and patient factors including age and comorbidities. Endovascular coiling was the preferred treatment modality in six of the cases, but was found to be of limited value when the pseudoaneurysm involved cortical vessels of small diameter, which limits catheter access.[3,4,15,18] Open surgical resection was the treatment modality in four cases.[16,28,31,34] In the case reported by Shirane *et al.*, open intervention was necessary to evacuate the associated hematoma and reconstruct the proximal ICA due to an anterior choroidal avulsion injury.[31] Once treated, most patients had minimal to no neurological deficits. However, these patients tend to have poor prognoses associated with their primary pathology; in one of the cases reported by Choudhri *et al.*, a patient suffered clinical vasospasm and subsequent reperfusion injury, which was fatal and unrelated to the treated pseudoaneurysm.[4] In another case, a distal MMA pseudoaneurysm was left untreated due to family preferences and withdrawal of care was initiated.[12]

CONCLUSION

Iatrogenic pseudoaneurysms are a rare but serious complication of blind procedures, including EVD, VP shunt or ICP monitor insertion, stereotactic brain biopsy, and subdural collection drainage. Attempting proper visualization of the cortex through a generous dural opening is recommended, although not always possible through a small burr hole. Considering the rarity of such complications, it is not necessary to obtain vessel imaging prior to every procedure. However, the suspicion for pseudoaneurysm formation should be high with new intraparenchymal or intraventricular hemorrhage following such procedures, and prompt surgical management is essential.

Declaration of patient consent

Informed consent was obtained for the use of patient history and radiographic images for teaching and research purposes.

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Conflicts of interest

There are no conflicts of interest.

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