Severe Vertebrobasilar Vasospasm After Iatrogenic Rupture of a Posterior Fossa Epidermoid Cyst: A Case Report of a Rare Complication Managed With Endovascular Intervention

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BACKGROUND AND IMPORTANCE: Aseptic meningitis and hydrocephalus have been reported after intracranial epidermoid cyst rupture. We present a rare case of clinically symptomatic vasospasm after iatrogenic rupture.

CLINICAL PRESENTATION: A middle-aged woman presenting with headache, facial paresthesia, and dizziness was found to have a 5-cm posterior fossa epidermoid cyst on magnetic resonance imaging. Resection was achieved through suboccipital craniectomy and C1 laminectomy. On postoperative day (POD) 1, the patient became unresponsive. After ventriculostomy placement for developing hydrocephalus, she failed to improve. Digital subtraction angiography revealed severe vertebrobasilar vasospasm, which was treated successfully with intra-arterial verapamil and milrinone. She experienced multiple episodes of recurrent vasospasm, all successfully treated with verapamil-milrinone. After ventriculoperitoneal shunt placement on POD 31, her condition stabilized; she was discharged to a rehabilitation center on POD 38.

CONCLUSION: This successful treatment of rare, clinically symptomatic vasospasm postiatrogenic epidermoid cyst rupture may help guide treatment in similar scenarios.

KEY WORDS: Endoscopic third ventriculostomy, Endovascular treatment, Intracranial epidermoid cyst, Vertebrobasilar vasospasm

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Epidermoid cysts contain epidermal tissue, keratin debris, and lipid deposits and can occur nearly anywhere in the body, including the central nervous system. The prevalence of epidermoid and dermoid cysts in the head and neck regions ranges from 1.6% to 7%, comprising approximately 1% of all brain tumors.[1,2] When located intracranially, they are typically found in the anterior or middle fossa, rather than the posterior fossa. Chemical meningitis and acute hydrocephalus after iatrogenic or spontaneous epidermoid cyst rupture have been well-reported[3,4]; however, to our knowledge, only 1 case of cyst rupture–associated intracranial vasospasm has been documented.[5] Interestingly, in that report, the patient presented approximately 2 weeks after cyst resection with ischemic changes evident on MRI.[5] Here, we present the case of a middle-aged woman who developed acute hydrocephalus and severe vertebrobasilar vasospasm on postoperative day (POD) 1 that required multiple endovascular interventions.

CLINICAL PRESENTATION

On the day of admission and surgery, the patient provided informed consent for treatment and HIPAA-compliant publication. Our local institutional review board does not require approval for the report of a single case. Additional data that support the findings of this report are available from the corresponding author on reasonable request.

Patient History and Clinical Findings

A middle-aged woman presented with right-sided headache, ear fullness, facial paresthesia, and dizziness, all of which had been progressively worsening for approximately 1 year. More recently, she had begun to experience intermittent confusion and gait instability. She was neurologically intact on examination and had no remarkable medical history. MRI with and without contrast

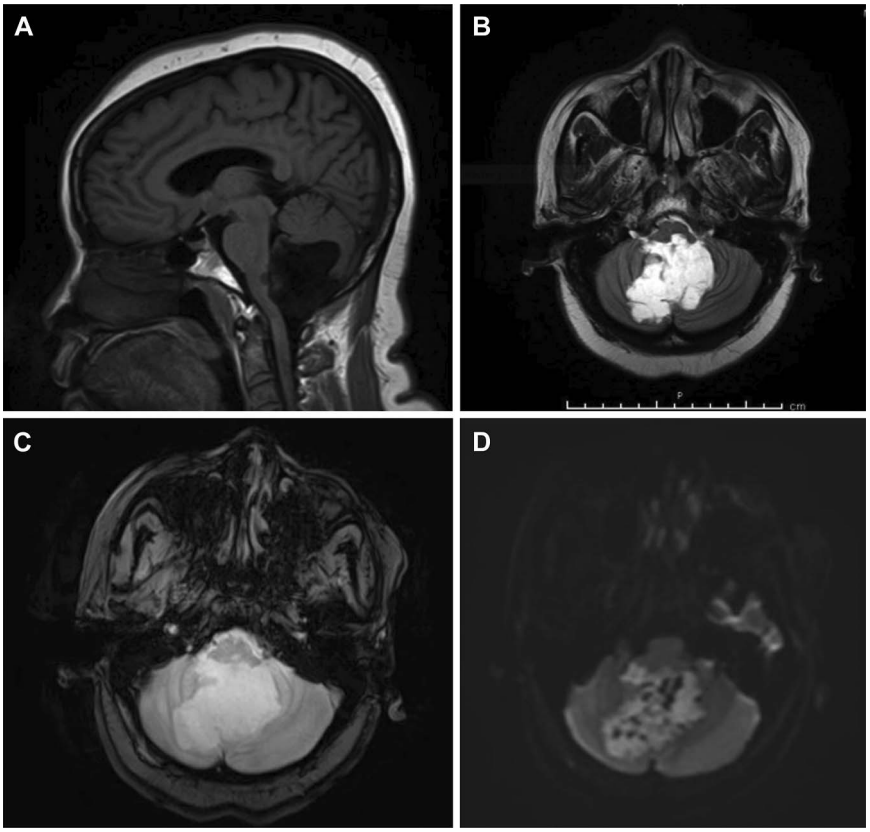

FIGURE 1. *MRI demonstrating the lesion characteristics, suggestive of an epidermoid cyst:* **A**, *sagittal T1-weighted image showing tumor hypointensity; axial T2-weighted images* **B**, *without contrast and* **C**, *with contrast showing tumor hyperintensity and contrast enhancement and* **D**, *diffusion-weighted image showing mild diffusion restriction with scattered hypointensities within the mass.*

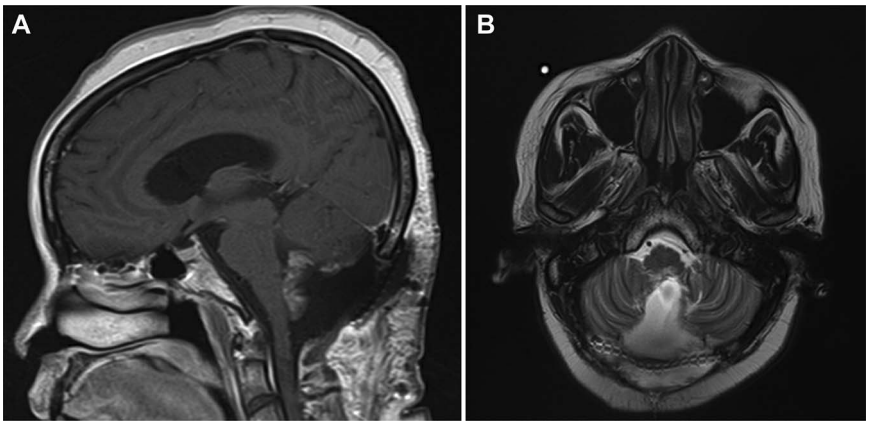

FIGURE 2. *Postoperative MRI revealing near-complete resection of the tumor with expected residual along the posterior pontomedullary junction in* **A**, *sagittal and* **B**, *axial planes.*

demonstrated a 5-cm, nonenhancing fourth ventricular mass that was subsequently confirmed on pathological analysis to be an epidermoid cyst (Figure 1A-1D).

Surgical Procedure

A suboccipital craniotomy with C1 laminectomy was performed with the patient in a prone concorde position to allow adequate access to the caudal end of the posterior fossa between the cerebellar hemispheres. Owing to the large size of the tumor, it was resected in a piecemeal fashion, which unfortunately led to spillage of tumor contents into the subarachnoid space. Attempts were made to minimize the spread of the contents outside the surgical field by packing cottonoids and using copious amounts of irrigation; however, admittedly, microscopic debris was still dispersed. Near the posterior pontomedullary junction, no clear plane was found, and a small amount of the tumor capsule remained attached to the brainstem (Figure 2A and 2B). Of note, no direct manipulation of the vertebral or basilar arteries occurred during the procedure. After additional copious irrigation using Ringer solution containing dexamethasone, the wound was closed in a standard fashion. The patient was successfully extubated and taken to the neurological intensive care unit; shortly thereafter, she was observed at her preoperative neurologically intact baseline.

Hospital Course

On the morning of POD 1, the patient remained at her neurologically intact baseline; however, approximately 24 hours after the procedure, she began to experience right facial anesthesia. A previously planned MRI with and without contrast demonstrated near-complete resection. No ischemic changes were observed. Within 2–3 hours, the patient's neurological and mental status declined, and she became acutely unresponsive, prompting emergent head computed

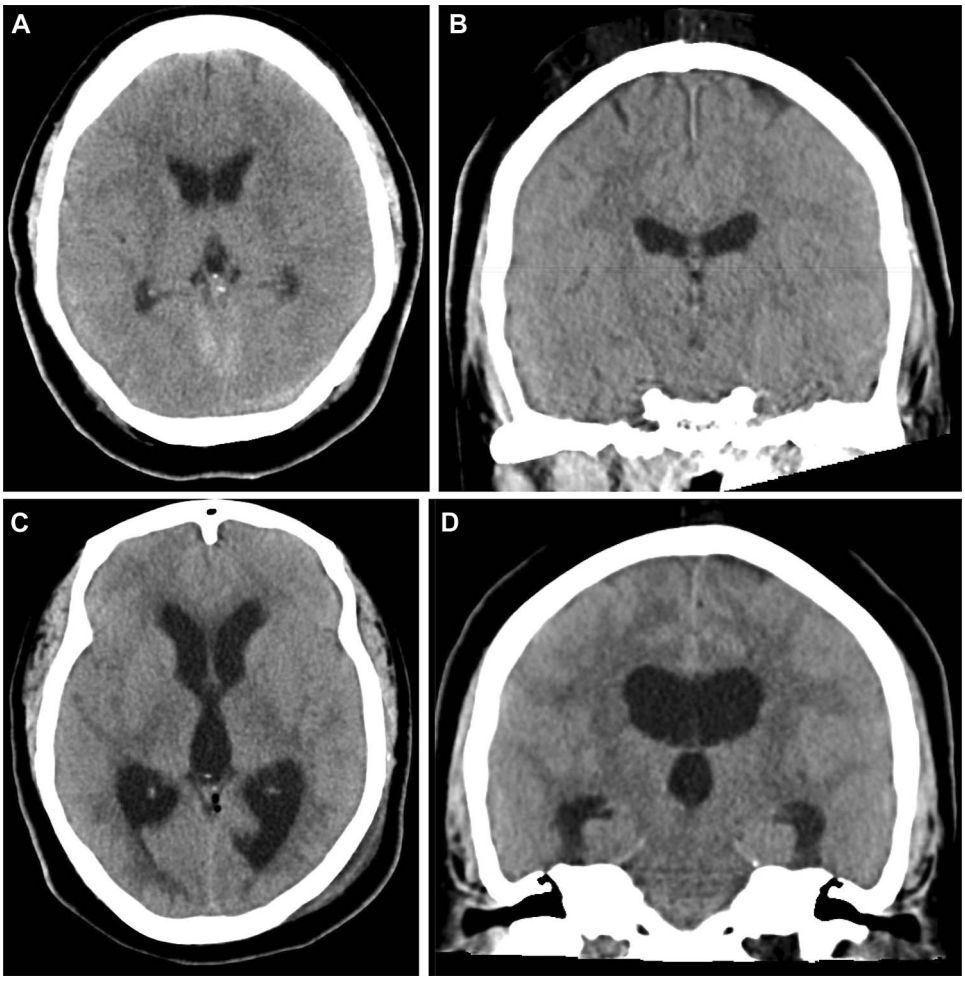

FIGURE 3. *Initial postoperative computed tomography images demonstrating normal ventricular caliber in the* **A**, *axial and* **B**, *coronal planes. Developing hydrocephalus is observed in the* **C**, *axial and* **D**, *coronal planes.*

tomography angiography. Interestingly, computed tomography angiography demonstrated developing hydrocephalus for which a ventriculostomy was placed (Figure 3A-3D) and a small-caliber vertebrobasilar circulation, consistent with potential vasospasm. Despite ventriculostomy drainage and return-to-normal intracranial pressures, no improvement was observed. Diagnostic digital subtraction angiography (DSA) demonstrated severe vasospasm of the vertebrobasilar tree (Figure 4A). An intra-arterial infusion of 30-mg verapamil and 5-mg milrinone was administered through the right vertebral artery, achieving significant improvement in the radiographic vasospasm and the patient's neurological examination (Figure 4B).

On PODs 2 and 3, the patient underwent additional vasospasm treatments in the form of intra-arterial verapamil and milrinone. There were no complications during any of these treatments, and continued improvement in the degree of radiographic vasospasm and the patient's clinical examination was observed at each intervention.

At the beginning of POD 4, progressive weaning of the ventriculostomy was attempted; however, multiple attempts failed because the patient become neurologically depressed when the ventricular drainage was reduced or eliminated, and her neurological status improved on drainage. On PODs 12 and 13, the patient's neurological examination deteriorated, despite additional ventriculostomy drainage. DSA again demonstrated significant vasospasm requiring intra-arterial verapamil and milrinone treatment resulting in radiographic and clinical improvement.

Despite additional weaning attempts, the patient was dependent on ventriculostomy drainage and ultimately underwent an endoscopic third ventriculostomy on POD 17 in accordance with her wishes before ventriculoperitoneal shunt placement. Postprocedure, the external drainage catheter was kept clamped, and the patient's neurological status became depressed. The catheter was opened to drain, but she did not return to her neurological baseline. She underwent another DSA, which revealed persistent vasospasm that was successfully treated with verapamil and milrinone, after which she again improved.

On POD 31, a ventriculoperitoneal shunt was placed, with serial follow-up computed tomography scans showing decreasing ventricular caliber. Although diffusely deconditioned from her prolonged hospital course, the patient recovered, was neurologically intact, and was discharged to a short-term rehabilitation facility on POD 38. At the 6-month follow-up evaluation, the patient had made a full recovery and remained neurologically intact.

DISCUSSION

We report a case of an iatrogenically ruptured epidermoid cyst in the posterior fossa that became complicated by hydrocephalus that ultimately required ventriculoperitoneal shunting and severe vasospasm requiring multiple rounds of endovascular spasmolytic therapy.

The only other instance of vasospasm after resection of an epidermoid cyst was reported by Aw et al,[5] in a middle-aged man with a left temporal epidermoid cyst. After undergoing uncomplicated surgical debulking, this patient presented 2 weeks later with severe vasospasm and was treated conservatively. Our case is similar, except for the very early onset of vasospasm and the endovascular management that was performed. In addition, our patient developed concurrent hydrocephalus that was ultimately treated with cerebrospinal fluid (CSF) diversion in the form of a ventriculoperitoneal shunt. Interestingly, it is difficult to ascertain whether the patient's vasospasm and hydrocephalus are inter-related or coincidental occurrences. However, it has been postulated that a reduction in intracranial pressure through ventriculostomy drainage is beneficial for vasospasm after subarachnoid hemorrhage.[6,7]

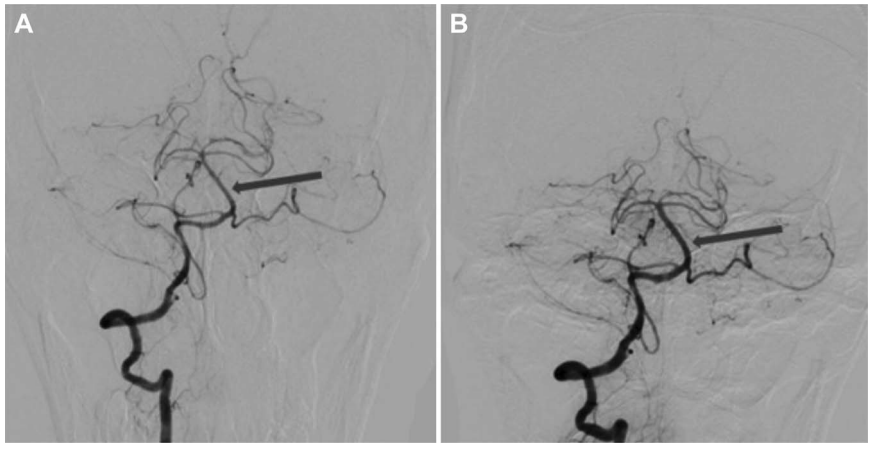

FIGURE 4. **A** *and* **B**, *Illustration of one of the spasmolytic therapeutic angiograms performed: digital subtraction angiogram of a right vertebral artery contrast injection showing severe vasospasm in the basilar artery (arrow in* **A**), *with improvement subsequent to intra-arterial infusion of 30 mg of verapamil and 5 mg of milrinone (arrow in* **B**).

Several mechanisms have been proposed to explain the pathogenesis of vasospasm during tumor excision or rupture. These include accumulation of blood breakdown products in basal cisterns, hypothalamic dysfunction because of direct irritation, direct manipulation and damage to the blood vessel walls, release of chemical substances from a resected tumor, and autoimmune or paraneoplastic mechanisms.[8-11] It is our opinion that direct irritation of the patient's vessels (ie, from tumor contents, manipulation, and breakdown of postsurgical blood products) in combination with the increased external pressure through hydrocephalus led to the patient's precipitous decline, which resolved after CSF drainage and vasospasm treatment. This is further evidenced by the concurrent declines requiring increased CSF drainage and vasospasm treatment. Regarding the patient's hydrocephalus, we assume that iatrogenic intraoperative rupture of the cyst with release of contents, despite our attempts to reduce spread, led to inflammation of the subarachnoid spaces and cisterns that in turn caused impaired CSF reabsorption.

The milrinone administered to the patient in this report has not been approved for intraarterial use by the US Food and Drug Administration. However, this application has been previously reported.[12]

CONCLUSION

We present a rare case of severe postoperative vertebrobasilar vasospasm and hydrocephalus after resection of a posterior fossa epidermoid cyst. The vasospasm was successfully managed with serial intra-arterial spasmolytic treatments, and fortunately, the patient had no residual effects observed at the 6-month follow-up. It is the authors' goal that this case report raises awareness and stimulates discussion regarding a rare, but real and potentially devastating, consequence of posterior fossa epidermoid resection when en bloc resection is not possible.

Author Contributions

Conception and design: Housley, Levy; Acquisition of data: All authors; Formal analysis and investigation: All authors; Statistical analysis: Not applicable; Drafting the manuscript: All authors; Critically revising the manuscript: All authors; Reviewed the submitted version of the manuscript: All authors.

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COMMENTS

This case report provides a unique insight into a rare complication following the iatrogenic rupture of a posterior fossa epidermoid cyst. In addition, the authors detail the successful management of severe vertebrobasilar vasospasm using intra-arterial verapamil and milrinone, which is a valuable contribution to the scarce literature on this topic.

However, several questions could have been addressed to understand this uncommon clinical scenario better. The authors could provide more

operative details, including whether the tumor was easily dissected and if precautions were taken to avoid the development of aseptic meningitis, vasospasm, and hydrocephalus.

The relationship between hydrocephalus, ventriculostomy, and vasospasm remains less than completely clear. The clinical deterioration despite CSF drainage and the later requirement of a ventriculoperitoneal shunt needs further clarification. Additionally, the choice of an endoscopic third ventriculostomy in the context of "chemical meningitis" deserves further clarification.

Knowing if the patient had preoperative hydrocephalus due to the sizeable 5 cm posterior fossa lesion would also be informative. In addition, the presentation of preoperative and postoperative imaging could have been beneficial for readers to visualize the exact scenario.

The authors navigated a challenging complication well, and while a more comprehensive account of preventative measures would have been helpful, the account may prove imminently useful to a reader facing a similar case or circumstances.

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