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Postoperative Symptomatic Cerebral Vasospasm: Requiring Attention Following an Uneventful Resection of an Epidermoid Cyst - A Case Report and Literature Review

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Abstract

Cerebral vasospasm associated with epidermoid cyst can be caused by tumor content spillage, such as spontaneous rupture and postsurgical resection. Symptomatic cerebral vasospasm following the resection of an intracranial epidermoid cyst is a rare but serious complication that lacks a consensus on treatment. Case presentation: A 10-year-old girl underwent an uneventful complete resection of a left cerebellopontine angle epidermoid cyst. On the second postoperative day (POD 2), she exhibited reduced speech, confusion, and hyperventilation followed by hypocapnia. On POD 4, she developed right hemiparesis and dysphasia. Cerebral magnetic resonance imaging showed restricted diffusion areas in her left temporal and parietal lobes and the dorsal thalamus. Magnetic resonance angiograms confirmed narrowing of the proximal middle cerebral arteries, consistent with vasospasm. Conservative management, consisting of intravenous hydration and corticosteroid administration, proved effective in resolving her symptoms and radiologic vasospasm. On POD 8, the extensive restricted diffusion areas notably decreased in size. Her right hemiparesis was completely resolved, and her dysphasia gradually improved over time. At the 1-year follow-up, she exhibited moderate transcortical sensory dysphasia. To our knowledge, this study is the first to report on a pediatric case of symptomatic cerebral vasospasm following an epidermoid cyst resection. The combination of tumor content spillage and hyperventilation may contribute to the occurrence of cerebral vasospasm and subsequent ischemia. This complication should be acknowledged after a complete and uneventful resection.

Keywords: epidermoid cyst, vasospasm, delayed ischemic deficit, hyperventilation

Introduction

An intracranial epidermoid cyst is a benign congenital tumor of ectodermal origin. Chemical meningitis is a well-known complication of resection caused by the spillage of epidermoid cyst contents into the subarachnoid space.[1)] Cerebral vasospasm can also be caused by subarachnoid spillage, such as spontaneous rupture and postsurgical resection.[2-8)] We present a patient who developed symptomatic vasospasm after an uneventful resection of an intracranial epidermoid cyst. To our knowledge, only three such adult cases have been previously reported.[9,10)] A consensus for the treatment of this rare but serious complication has not been established.

Case Report

A 10-year-old right-handed girl with no past medical histories presented with chronic headache. Brain magnetic resonance (MR) images showed a mass lesion in the left cerebellopontine angle. Restricted diffusion on diffusion-weighted images suggested epidermoid cyst (Fig. 1A and B). MR angiograms performed four times during the preoperative follow-up period showed normality. Resection was performed via a left subtemporal transtentorial approach due to slow growth observed over a period of 20

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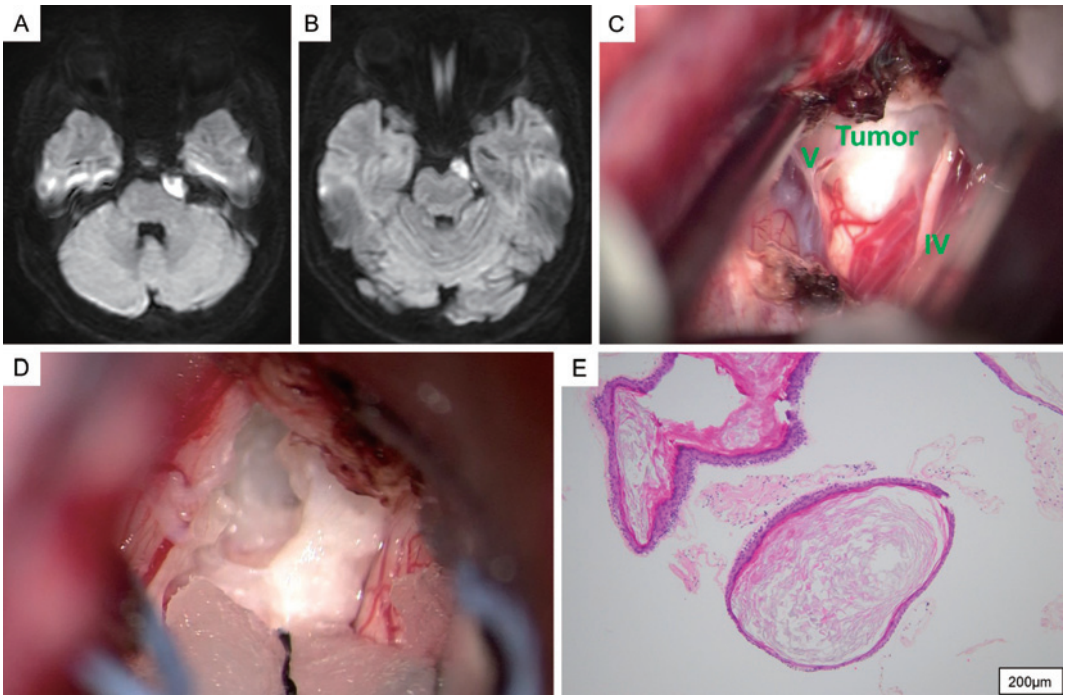

Fig. 1 A and B: Preoperative axial diffusion-weighted images showed a hyperintense tumor in the left cerebellopontine angle, suggesting epidermoid cyst. C: An intraoperative photograph taken after dissection of the tentorium shows a pearly white tumor between the trochlear (IV) and trigeminal nerves (V). D: An intraoperative photograph taken during intracapsular debulking. E: The histopathological diagnosis (hematoxylin and eosin staining) showed the presence of squamous epithelial cells containing keratohyalin granules.

months during imaging surveillance. The tumor had a pearly white appearance (Fig. 1C, D). After performing piece-meal resection of the tumor within the capsule, the capsule was completely removed. The capsule did not adhere to the brainstem or any cerebral nerves. Irrigating the tumor cavity was intentionally avoided to prevent the spread of tumor contents. Postoperative brain computed tomography (CT) scans showed no intracranial hemorrhage or other abnormalities. The final pathological diagnosis was epidermoid cyst (Fig. 1E).

After recovering from anesthesia, she was neurologically intact and was transferred to the intensive care unit for monitoring. On the first postoperative day (POD), she developed a fever without signs of meningeal irritation and was transferred to a general ward. Her fever lasted for 5 days, up to 39.0°C. On POD 2, she exhibited reduced speech, confusion, and hyperventilation. Venous blood gas analysis showed a carbon dioxide partial pressure and pH of 23.9 mm Hg and 7.576, respectively. Brain CT scans were normal. On POD 4, she developed right hemiparesis and motor dysphasia. Delayed venous infarction due to the surgical manipulation was suspected and brain MR imaging was performed. Diffusion-weighted images showed extensive restricted diffusion areas in her left temporal and parietal lobes, as well as the dorsal thalamus (Fig. 2A-D). There was no residual tumor. Cerebral MR angiograms showed narrowing of the distal internal carotid arteries and proximal middle cerebral arteries (Fig. 2E-G). Cerebral MR venograms showed no evidence of venous infarction comparing pre and postimages (Fig. 2H-K). Blood testing for vasculitis and thrombosis disorders was negative. Therefore, cerebral vasospasm by the spillage of the tumor contents was speculated. Lumbar puncture, a psychologically highly invasive procedure, was avoided due to consideration of hyperventilation as a potential cause of ischemia. Conservative management, such as intravenous hydration and administration of corticosteroids and free radical scavengers, was initiated immediately. The following day, her right hemiparesis disappeared. On POD 8, repeat brain MR angiograms showed that the extensive restricted diffusion areas notably decreased in size (Fig. 3A-E). Her right hemiparesis was completely resolved, and her motor dysphasia gradually improved over time. Systemic corticosteroids were gradually tapered over a period of 1 week. On POD 20, brain MR angiograms showed complete resolution of vasospasm (Fig. 3F). She experienced minor reading and speech problems. She was transferred to a rehabilitation institution. At the 1-year follow-up, no radiological recurrence of the tumor or vasospasm was observed. However, the difficulty of understanding, suggestive of transcortical sensory dysphasia, persisted, and she needed additional support for her hearing impairment while attending

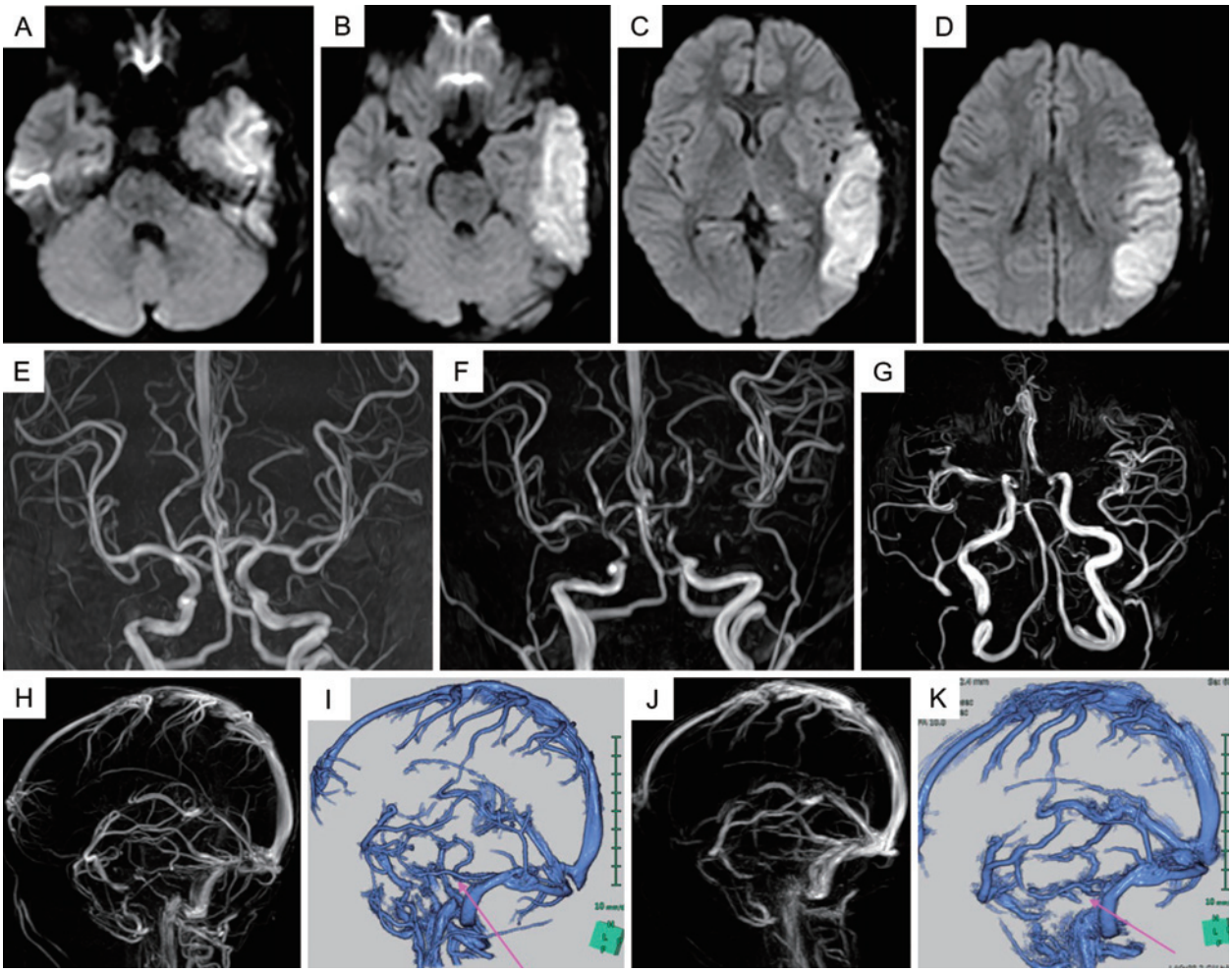

Fig. 2 A-D: Axial diffusion-weighted images 4 days after surgery showed areas of hyperintense signal in her left temporal and parietal lobes and the dorsal thalamus. There was no residual epidermoid cyst. E: Preoperative magnetic resonance (MR) angiogram. F and G: MR angiograms 4 days after surgery showed narrowing of the distal internal carotid and proximal middle cerebral arteries, with the degree of narrowing being more severe on the left side. Posterior cerebral arteries were intact. H and I: Preoperative MR venograms. An arrow indicates the left vein of Labbe by three-dimensional reconstruction. J and K: MR venograms 4 days after surgery showed no significant changes. An arrow indicates the left vein of Labbe by three-dimensional reconstruction.

school.

Discussion

To our knowledge, this study is the first to report on a pediatric case of symptomatic cerebral vasospasm following an epidermoid cyst resection, demonstrating the effectiveness of conservative medical treatment. Symptomatic cerebral vasospasm following epidermoid cyst resection[9,10] and transient vasospasm due to epidermoid or dermoid cyst rupture[2-8] are rare occurrences. Intracranial epidermoid cyst contains lipid metabolism.[2,6] The pathogenesis is presumed to involve the free radicals produced from lipid peroxides within the tumor.[11] Experimental injection of a lipid peroxide into the canine cisterna magna causes chemical meningitis and induces cerebral vasospasm.[12] Manipulation of the cerebral arteries during epidermoid cyst resection may also lead to vasospasm. However, in our case, the affected arteries (internal carotid and middle cerebral arteries) were not exposed in the surgical field. Therefore, arterial manipulation cannot explain the occurrence of vasospasm. The affected arteries and tumor cavity were in basal cistern. Aw et al. reported a case of a left cerebellopontine angle epidermoid that developed cerebral infarction due to cerebral vasospasm 2 weeks postsurgery.[10] Cerebrospinal fluid circulation might explain the delayed occurrence of symptomatic cerebral vasospasm following surgery. In the two previous reports on postoperative vasospasm, the tumors were consistently large, suggesting that the larger tumors present a significant risk of scattering lipid peroxides.[9,10] However, cases of epidermoid rupture with ischemic attacks have been reported even in small tumors, suggesting that this phenomenon may not be solely related to tumor size.[2-8] This complication should

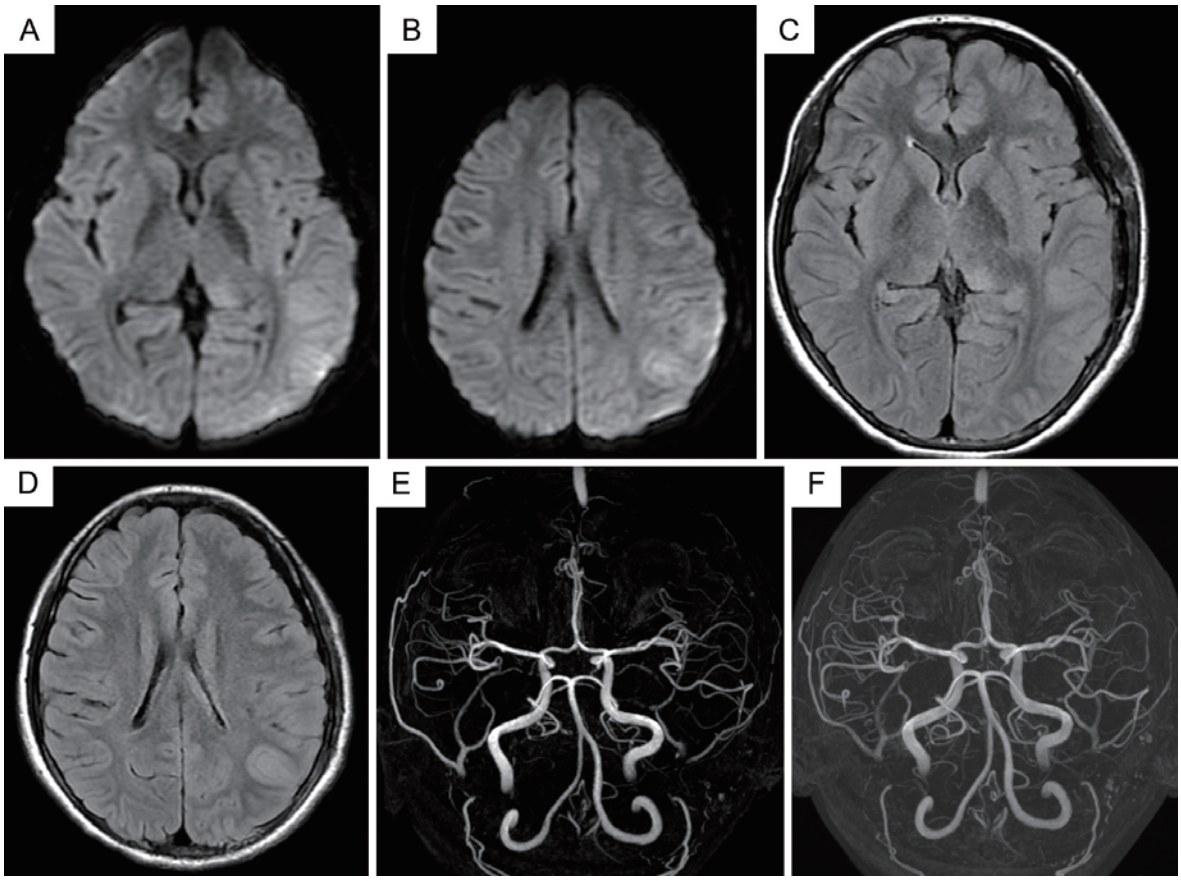

Fig. 3 A and B: Axial diffusion-weighted images. C and D: Fluid-attenuated inversion recovery images 8 days after surgery showed only small, slightly hyperintense areas in the left temporoparietal region and dorsal thalamus. E: Magnetic resonance angiograms showed significant improvement in cerebral vasospasm. F: Twenty days after surgery, the vasospasm had resolved.

Table 1 Description of cases with symptomatic vasospasm following a resection of an intracranial epidermoid cyst resection in the literature

References	Age (years)	Sex	Onset	Tumor location	Procedure	Spastic vessel	Treatment	Sequelae
Ecker et al.	43	Male	2 weeks	Cerebellopontine angle	Debulk	Ipsilateral M1	Conservative	Minor cognitive and speech problems
Ecker et al.	23	Female	36 hours	Frontal and temporal lobe	Total removal	Ipsilateral M1	Balloon angioplasty	Mild hemiparesis
Aw et al.	44	Female	A few days	Parasellum	Total removal	M1	Papaverine	Not mentioned
Our case	10	Female	4 days	Cerebellopontine angle	Total removal	Bilateral M1	Conservative	Moderate transcortical sensory aphasia

be noted after surgery, irrespective of tumor size. In our case, since the size of the tumor is relatively small, it is difficult to determine whether the pathogenesis is vasospasm due to tumor contents. However, it is challenging to consider other etiologies.

The means of preventing and treating epidermoid cyst-related cerebral vasospasm have not been established. The incidence of chemical meningitis is higher after incomplete resection[13] and complete resection might reduce the risk of vasospasm.[10] Nonetheless, our patient developed symptomatic vasospasm despite achieving a complete resection. Three adult cases of symptomatic cerebral vasospasm following an epidermoid cyst resection were reported.[9,10] In three of the four cases, including ours (Table 1), a total removal procedure was performed. Thus, it is likely that the incidence of vasospasm cannot be solely reduced through

total resection. When performing a resection, it is important to avoid spilling tumor materials into the subarachnoid space to prevent meningeal irritation.[13] In our case, despite efforts to contain the tumor contents within the capsule, a piece-meal resection could still potentially lead to the spillage of the tumor contents. While en bloc resection is the preferred approach, its feasibility can be challenging depending on factors such as tumor size and adhesion degree. The use of corticosteroids might be beneficial in preventing chemical meningitis. A study involving 28 patients who underwent an epidermoid cyst resection and received perioperative systemic corticosteroids reported that only three patients developed postoperative chemical meningitis.[14] In our case, irrigation of the tumor cavity was intentionally avoided to prevent the spread of tumor contents. While the controversy surrounding irrigation of the tumor cavity persists, it is noteworthy that postoperative vasospasm occurred in a report that did not support irrigation. This suggests that irrigation might prevent this complication.[9] Another report suggested that irrigating the surgical site with a hydrocortisone solution may reduce the risk of postoperative chemical meningitis.[10] Regarding treatment, in our patient, the vasospasm was diffuse; therefore, focal endovascular treatment was not indicated. Ecker et al. reported the endovascular treatment of vasospasm after the resection of an intracranial dermoid cyst using balloon angioplasty.[9] However, procedural complications have been mentioned. The absence of a neurovascular treatment specialist at our hospital posed challenges in promptly conducting cerebral angiography and endovascular treatment. Establishment of a system for rapid cerebral angiography would expand options for accurate diagnosis and treatment. There is no standard treatment for the pediatric vasospasm. However, several literature reports have shown that medical treatment has proven effective, like our case.[15]

Our patient exhibited hyperventilation on POD 2, which may have attributed ischemia due to the vasoconstrictive effects of hypocapnia on cerebral arteries.[16] What differentiates this case from previous cases is the patient's hypersensitivity to postoperative physical and psychological stress, which led to the development of hyperventilation syndrome. In a study involving 470 patients who underwent cranial base tumor resection, the incidence of angiographic vasospasm was 1.9%.[17] Asymptomatic vasospasm, which might have otherwise gone unnoticed, could have become symptomatic. Our patient was particularly susceptible to perioperative stress, which can lead to hyperventilation. Considering hyperventilation as a potential cause of ischemia, we avoided performing a lumbar puncture and angiography, which are psychologically invasive for children. The limitation of this report is that these inspections were not conducted. Managing various psychological and physiological stressors such as pain and nausea is important in preventing hyperventilation; however, this can be difficult in pediatric cases. Therefore, prophylactic medical treatment, such as corticosteroids, may be warranted.

In conclusion, neurosurgeons should be aware that symptomatic vasospasm may occur following the resection of an intracranial epidermoid cyst, even when the size of the tumor is relatively small and the tumor resection is complete and uneventful. The combination of tumor content spillage and hyperventilation may contribute to the occurrence of cerebral vasospasm and subsequent ischemia. It is difficult to determine whether the pathogenesis is vasospasm due to tumor contents from our case due to lack of inspections. Therefore, further study is needed to explore the pathogenesis.

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Conflicts of Interest Disclosure

The authors have no conflicts of interest to declare.

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