Case Report

Simultaneous Expansion of Pseudomeningocele and Development of Remote Subdural Hygroma: A New Phenomenon

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Summary

Development of subdural hygroma (SDG) distant from primary neurosurgery is uncommon. We present a rare relationship between remote supratentorial extension of a local SDG and expansion of an infratentorial pseudomeningocele following resection of a cerebellar hemangioblastoma in a 61-year-old female with von Hippel-Lindau disease. Initial Magnetic resonance imaging showed a cystic hemangioblastoma in the right cerebellum with mass effect on the brain stem. The patient undertook median suboccipital craniectomy with duraplasty followed by development of bilateral retrocerebellar SDG and a pseudomeningocele beneath the dural graft. Nineteen days after the operation, a remote SDG appeared supratentorially with concurrent interval growth of the pseudomeningocele. Early management of the growing pseudomeningocele may minimize the occurrence of remote SDG as a post-operative complication.

Key words: Craniotomy, Pseudomeningocele, Subdural hygroma, von Hippel-Lindau disease

INTRODUCTION

SDG is caused by acute or chronic accumulation of cerebrospinal fluid (CSF) in the subdural space. Risk factors for SDG include trauma, intracranial hypotension, cerebral atrophy, and iatrogenic disruption of the arachnoid membrane.
Pseudomeningoceles are subcutaneous fluid collections from the extravasation of the CSF through defects of arachnoid membrane and dura mater. Patients undertaking craniotomies have increased risks of developing both SDG and pseudomeningocele. However, SDG typically occurs at or near the surgical fields. Occurrence of a remote supratentorial SDG from an infratentorial surgery is rare. We present a patient with complications from a remote SDG related to a growing pseudomeningocele following resection of a von Hippel-Lindau (VHL)-associated cerebellar hemangioblastoma. Early interventions of growing pseudomeningoceles may minimize including subsequent complications.

CASE PRESENTATION

A 61-year-old female with VHL disease presented with progressive weakness of four limbs for months. In addition to VHL-associated cerebellar hemangioblastoma and multiple cysts in the liver, pancreas, and kidneys, she suffered from diabetes mellitus and hypertension. Neurological examinations showed unremarkable findings on cranial nerves. There was weakness of the upper and lower extremities with active movement against gravity. A series of lab examinations was significant only for elevated serum creatinine (1.9 mg/dL).

Magnetic resonance (MR) imaging showed a cystic hemangioblastoma in the right cerebellum with mass effect on the brain stem and fourth ventricle (Figure 1). In order to prevent hydrocephalus, she underwent a suboccipital craniectomy and C1 laminectomy to resect the tumor. Duraplasty was performed with use of a three-dimensional collagen matrix graft (DuraGen). Two days after the operation, MR imaging depicted bilateral retrocerebellar SDG and a cystic lesion, representing pseudomeningocele, beneath the dural graft (Figure 2). Two weeks later, her mental status declined with gravity-eliminated muscle power of the upper extremities and paralyzed lower ones. Follow-up MR images obtained 19 days after the operation demonstrated interval growth of the pseudomeningocele and extension of the infratentorial SDG to right supratentorial convexities (Figure 3). The pseudomeningocele showed no contrast enhancement or restricted diffusion. With concern of the complicated underlying diseases, she was transferred to a local nursing home without further surgical interventions.

![Figure 1: Sagittal (A) and axial (B) MR images show a 3.6-cm cystic lesion (thin arrow) in the right cerebellum with an enhancing nodule (thick arrow) and marked mass effect compressing the 4th ventricle (arrowhead) and distorting the brain stem.](image)
VHL is an inherited autosomal dominant syndrome with a spectrum of associated benign and malignant tumors, among which cerebellar hemangioblastoma is the most common one. The hemangioblastoma may remain quiescent or grow in accelerated rate. Surgical intervention is reserved for patients with neurologic symptoms, enlargement of the tumor or cyst, or hemorrhage. Following neurosurgical procedures, a watertight dural closure is considered helpful to reduce complications, such as fistulas, infections, herniation, and adhesion. However, CSF leakage may occur from pinholes made during suturing.\(^4\) In addition, well establishment of fibroblast activity in the matrix takes at least two weeks and incorporation of the graft into the dura needs four to six months to complete.\(^3\) In the posterior fossa, elevated CSF pressure from the dependent portion further increases the risk of
(2) On MR images, pseudomeningoceles usually follow signals of CSF and show communications with subarachnoid space. However, the communications may be too small to be seen.

In addition to the pseudomeningocele, differential diagnoses for the post-operative fluid collection beneath the dural graft include abscess, hematoma, and seroma. However, the rapid development of the lesion and lack of restricted diffusion and peripheral contrast enhancement exclude the diagnosis of abscess. Hematomas typically show sequential signal changes from evolution of hemoglobin breakdown. Seromas are transudate of fluid accumulating in the surgical field and usually spontaneously subside in weeks after surgery. A thin dark pseudocapsule is usually present around a seroma with surrounding soft tissue edema on MR images. With the combined clinical and imaging findings, the cystic lesion beneath the dural graft is most compatible with a pseudomeningocele.

SDG is a well-known post-operative complication caused by an arachnoid tear with formation of a one-way valve trapping CSF in the subdural space. However, the occurrence of supratentorial SDG after surgery of the posterior fossa was rarely reported. (1) The most popular proposed mechanism is an arachnoid tear in the surgical field with CSF tracking supratentorially to form the remote SDG. In patients with traumatic brain injury, remote SDG may develop contralateral to the side of decompressive surgery due to remote arachnoid tear from shearing stress, rapid decreased intracranial pressure, or disturbance of CSF circulation. (5) In addition to the proposed mechanisms, our patient demonstrates a new phenomenon for the development of remote SDG from simultaneous expansion of the pseudomeningocele. The enlarging pseudomeningocele may compress and torque the brain, causing a remote arachnoid tear and contributing to remote supratentorial extension of the local SDG.

In conclusion, postoperative pseudomeningoceles can grow and associate with the development of remote SDG. Early interventions of growing pseudomeningoceles may minimize subsequent complications.

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