Case Report

Subdural Empyema in the Adjacent Cortex of an Arteriovenous Malformation

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Summary

Background: The coexistence of arteriovenous malformation and subdural empyema is extremely rare.

Case Report: A 40-year-old man was admitted with a severe headache. His cranial computerized tomography findings showed a right-sided temporal intracerebral hematoma with a midline shift. During surgery, an organized capsule and subdural empyema, which contained pus and trabeculae, were seen as the dura was elevated. After resection of the infectious tissue, the intracerebral hematoma was reached and evacuated, and the nidus of the arteriovenous malformation (AVM) was resected. The recovery was uneventful. Herein, we discuss the formation mechanism of empyema, together with the AVM in the same hemisphere, and the possible role of the hemodynamic and ischemic processes associated with the steal phenomenon. Additionally, we present the proliferative and inflammatory roles of the molecular biological factors; vascular endothelial growth factor (VEGF) and basic fibroblastic growth factor (bFGF) promoting angiogenesis in AVM.

Conclusion: Additionally, we present the proliferative and inflammatory roles of the molecular biological factors; vascular endothelial growth factor and basic fibroblastic growth factor promoting angiogenesis in AVM.

Key words: Arteriovenous malformations, bFGF, intracerebral hemorrhage, steal phenomenon, subdural empyema, VEGF

Arteriovenöz Malformasyona Komşu Beyin Korteksi Üzerinde Subdural Ampiyem

Özet

Arteriovenöz malformasyon ile subdural ampiyemin birlikte görülmesi çok nadirdir.


Sonuç: Vasküler endotelyal büyüme faktörü ve basic fibroblastic büyüme faktörü gibi moleküler biyojik faktörlerin anjiyojene uyarmadaki proliferative ve enflamatuar rollerinin etkisi tartışıldı.

Anahtar Kelimeler: Arteriovenöz malformasyon, bFGF, intraserebral kanama, çalma fenomeni, subdural ampiyem, VEGF
INTRODUCTION
An arteriovenous malformation (AVM) is a vascular malformation characterized by an arteriovenous shunt through a collection of dilated vessels, without an intervening capillary network. It contains at least one enlarged feeding artery and at least one enlarged early draining vein. Associated pathologies are also described as aneurysms, Sturge-Weber syndrome, Osler-Weber-Rendu syndrome, Wyburn-Mason syndrome and Klippel-Trenaunay-Weber syndrome(5). Although they are thought to be congenital in origin, with an initiating event occurring in early embryological development, some authors suggest a more dynamic developmental process in the interrupted malformation and resorption of superficial veins(2,5).

Subdural empyema is primarily an intracranial suppurative infection located between the arachnoid and the overlying dura. It has a tendency to spread rapidly through the subdural space until being limited by specific boundaries (e.g. falx cerebri, tentorium cerebelli, base of the brain, foramen magnum, venous sinuses, etc.)(1). Subdural empyema is usually unilateral, mostly involves the frontal lobe, and occurs as a complication of meningitis, otitis, mastoiditis or frontal/ethmoidal sinusitis. It is usually seen in children and young adults(1), and early recognition and intervention is mandatory. Successful management of subdural empyema depends on early diagnosis, prompt evacuation of the pus, and appropriate antibiotic therapy. The coexistence of arteriovenous malformation and subdural empyema is extremely rare.

Here, we present the case of a 40 year-old man who was admitted to the emergency unit after a loss of conscience due to a right temporal intracerebral hematoma and underlying AVM. The clinical presentation was left hemiparesis and surgery was carried out.

The formation mechanism of empyema in association with AVM in the same hemispheric side, and possible role of the hemodynamic and ischemic processes associated with the steal phenomenon, will be described. Additionally, the proliferative and inflammatory role of molecular biological factors; vascular endothelial growth factor (VEGF) and basic fibroblastic growth factor (bFGF) promoting angiogenesis in AVM, and accompanying ischemia in the area will be explained(3).

CASE PRESENTATION
A 40 year-old man with a loss of consciousness after severe headaches and left-sided hemiparesis was admitted to the emergency unit. On cranial computed tomography (CT), a large temporal intracerebral hematoma was found (Fig. 1). He had no fever, and there was no sign of scalp, dermal, ear, nose or sinus infection. The complete blood count and coagulation tests were normal. The white blood count was 10000/mm³. Since the case was urgent, the C-reactive protein and sedimentation rate tests were not carried out. The patient underwent surgery immediately, and a right frontotemporal craniotomy was performed. When the dura was opened, a subdural cavitation with yellowish-white membranes, containing pus and trabeculae, was encountered and resected. Then, the hematoma was removed with the underlying arteriovenous malformation, and an associated saccular aneurysm was clipped. Pathological examination confirmed AVM and an encapsulated infection (Fig. 2).

The microbiological examination of the subdural pus material was negative for any microorganisms. Still, antibiotherapy of a third generation cephalosporin was begun for 2 weeks, and oral antibiotherapy was continued for 6 weeks. Retrospective examination of the preoperative cranial CT images revealed the presence of a frontal hypodense subdural collection ending near
the sagittal sinus. The post-operative course was uneventful, other than mild left hemiparesis which recovered in the lower extremity, but did not resolve in the upper extremity.

Figure 1: A) Preoperative CT showing the right temporal intracerebral hematoma with midline shift B) In upper section hypodense collection which was proved to be encapsulated infection on histopathological examination

Figure 2: A) H&E stain: Capsule formation B) the subarachnoid space contains scattered inflammatory cell showing infection C) Abnormal vascular structures D) Brain parenchyma between the abnormal vasculature showing arteriovenous malformation
DISCUSSION

The prevalence of cerebral AVMs in the western population is < 0.01%, and the detection rate is 1 in 100,000 people per year. The mean age at diagnosis is 27±13 years. Hemorrhage (52%), headache (18%) and seizures (13%) are the most common presentations. Associated intranidal, arterial or venous aneurysms can also be found(2,5).

The pathogenesis of AVM is not completely understood, and several mechanisms have been proposed: primarily congenital at the embryonic stage (40-80 mm in length), primarily an abnormality of the primordial capillary or venous formation(2,5), or a continuing process by interrupted malformation and resorption of the superficial cerebral veins(2). The pathogenesis of AVM is thought to begin with mechanical, hormonal, thrombotic, hemodynamic, thermal, ischemic/hypoxic or inflammatory triggering factors(2,3,5). Steal phenomenon, where the blood supply is preferentially delivered to the AVM at the cost of normal brain parenchyma, can lead to focal atrophy and focal neurological symptoms(2,3,5). Accelerated collateral blood flow from surrounding vascular territories, and decreased blood flow to the parenchyma, results in the steal phenomenon (steal peripheral to AVM, steal beyond AVM borders, massive steal beyond AVM borders)(2,3,5). This aggravates ischemia in the normal brain tissue adjacent to the AVM, thus promoting angiogenesis and/or an infectious process.

Sharma et al.(4) reports two cases of brain abscess after the embolization of arteriovenous malformation. And again, only after embolization, Mourier et al.(1) reports a pyogenic infection of the nidus and the overlying parenchyma. Hypoxia is known to stimulate the expression of VEGF(3). The proliferative and inflammatory role of molecular biological factors; vascular endothelial growth factor and basic fibroblastic growth factor promoting angiogenesis, are well known entities(5). Hemorrhage and thrombogenesis can result in the release of VEGF from the platelets(5), and infection can lead to the release of VEGF at the source of infection. Thus, in theory, insults such as stroke, brain contusion and encephalitis could potentially induce the development of AVM(2,3).

This rare case of AVM, together with subdural empyema, underwent emergency surgery as a result of a large temporal intracerebral hematoma. Before surgery, no detailed imaging (e.g. contrast-CT, MRI, MR angiography) was carried out. Only after the craniotomy was performed, and opening the dura, did we recognize the subdural empyema.

CONCLUSION

This case reports an interaction between AVM and the infectious process, not exactly revealing the reason-cause modality or the time impact, which is thought to be imperative and must be further investigated in experimental models.

AVM is a dynamic lesion, and the ischemic effects in a large fistulous AVM due to the steal phenomenon can be the promoting factor in infectious and inflammatory processes near the same localization. The infectious dynamics can induce the development or enlargement of AVMs, since they are not purely congenital lesions.

Though hematoma evacuation is an emergency, one must still keep in mind that different lesions can accompany the situation. As the surgical approach may be different in the presence of co-existing pathologies, the careful detection of the CT findings preoperatively is mandatory.
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