

Hematoma within the Outer Membrane of the Arachnoid Cyst Located in the Middle Fossa: A Mechanism of Development of Chronic Subdural Hematoma Associated with Arachnoid Cysts

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Abstract

It is well known that chronic subdural hematoma (CSDH) occasionally arises in patients harboring an arachnoid cyst (AC) located in the middle fossa. Although the pathogenesis of CSDH associated with ACs remains unknown, several possible mechanisms have been proposed. In many reported cases, the patients with ACs in the middle fossa would be symptomatic according to development of CSDH. A 9-year-old girl presenting with a headache, nausea, and diplopia was referred to our department. Magnetic resonance imaging showed an AC in the left-side middle fossa and ipsilateral CSDH with a remarkable mass effect. Irrigation of the CSDH, partial removal of the outer membrane of the AC and CSDH, and endoscopic cystocisternostomy were performed to relieve her symptoms. Postoperative clinical course was excellent. Histopathological examination of the outer membrane of the CSDH demonstrated an arachnoid cell layer and hemorrhage from the granulation inside the membrane, and collagen fibers outside the membrane. These findings strongly suggested that the membrane and the content of the CSDH were derived from the outer membrane of the AC, and the CSF including the hemorrhage within the membrane, respectively. Laceration and hemorrhage from granulation tissue within the outer membrane of the AC are considered as one of the developmental mechanisms of the CSDH associated with AC in the middle fossa.

Keywords

Arachnoid Cyst, Cyst Wall, Chronic Subdural Hematoma, Laceration, Arachnoid Membrane

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1. Introduction

Arachnoid cysts (ACs) located in the middle fossa infrequently present with chronic subdural hematoma (CSDH) [1]-[7]. It has been estimated that the occurrence rate of CSDH constitutes 15% - 30% of the patients with an AC in the middle fossa. On the contrary, it is reported that AC can be detected in only 2.4% of the population with CSDH [8]. Although most of the patients with ACs located in the middle fossa are asymptomatic, CSDH can render these patients to become symptomatic [9] [10].

Although the precise mechanism of the development of CSDH associated with AC remains unknown, several possible theories have been advocated as follows. First, fluctuating movement of the fluid within AC transfers the shearing force to the outer membrane to tear the small vessels on the cyst wall [5] [11]. Second, the pressure through the cyst induces injury of the bridging vein or vessels running in the cyst wall [3] [7] [12]. Third, laceration of the cyst wall leads to the formation of granulation and the subsequent hemorrhage of the cyst wall [13] [14]. These possible theories of CSDH associated with AC pathogenesis have undergone long speculation, but none have been demonstrated on pathological examination. The mechanism of CSDH development in our case is discussed.

2. Case Report

A 9-year-old girl who presented with a headache, nausea, and diplopia consulted with our department. Neurological examination revealed only left-sided abducens palsy, suggesting the etiology of her diplopia. She had a history of a fall off a bicycle ride and had hit the left temporal region of her head on the ground 2 months ago. At that time, a computed tomography (CT) scan of the head displayed a cystic lesion located in the middle cranial fossa, suggesting an AC (**Figure 1(a)**). A CT scan was performed in our hospital again, and it showed an AC in the middle fossa and subdural fluid collection, spreading in the left convexity, and compressing ipsilateral hemisphere remarkably (**Figure 1(b)**). Her symptoms and signs seemed to be attributable to the intracranial hypertension. Magnetic resonance imaging (MRI) demonstrated the cyst in the middle fossa presenting with isointensity compared to the cerebrospinal fluid (CSF), confirming it as AC. Furthermore, the subdural fluid collection at the left convexity showed a slight hyperintensity on a T1-weighted image (T1-WI) and hyperintensity on T2-WI, suggesting hematoma in the chronic phase (**Figure 2**).

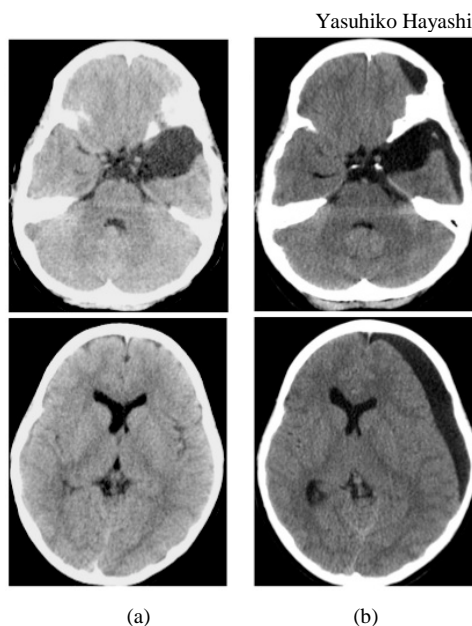


Figure 1. (a) A computed tomography (CT) scan of the head obtained when the patient encountered minor head trauma showed the arachnoid cyst located in the left middle fossa; no subdural hematoma was detected; (b) A CT scan of the head obtained at admission, 2 months after minor head trauma, showed a previously detected arachnoid cyst and subdural fluid collection with remarkable mass effect.

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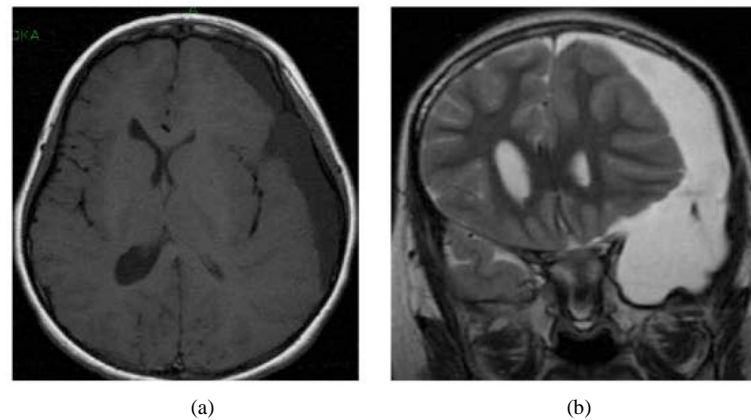


Figure 2. Magnetic resonance imaging (MRI) of the head at admission revealed that the intensity of the subdural hematoma was iso-hyperintense on a T1-weighted image (T1-WI) (a) and hyperintense on T2-WI (b), suggesting a hematoma in the chronic phase.

Irrigation of the subdural hematoma, partial resection of the outer membrane of the AC, and endoscopic cystocisternostomy were planned to relieve her symptoms and signs. A small temporal craniotomy was placed just above the CSDH. After the dural incision, a small resection of the thin outer membrane of the CSDH beneath the dura (**Figure 3(a)**) allowed the drainage of the CSF-like fluid within the hemorrhage, which was completely irrigated with the saline. No vessels could be found on the surface of the outer membrane connecting with the dura. After the partial resection of the outer membrane of the AC (**Figure 3(b)**), a rigid endoscope (Karl Storz, Germany) was advanced into the cyst cavity. The internal carotid artery (ICA), posterior communicating artery, and oculomotor nerve were identified through the transparent cyst wall. We made two small perforations on the cyst wall with a forceps between the ICA and the oculomotor nerve and one more perforation was added just above the ICA to make a stable communication between the cyst and the carotid cistern. Postoperative clinical course was excellent, and her preoperative symptoms and signs immediately completely disappeared. A CT scan obtained 6 months after the operation showed a remarkable reduction of the AC and the disappearance of the subdural space (**Figure 4**).

Histopathological examination of the outer membrane of the AC showed that the membrane was composed of arachnoid cells, which was confirmed with epithelial membrane antigen (EMA) staining (**Figure 5(a)**). The diagnosis of the arachnoid cyst was made based on this pathological finding. The outer membrane of the CSDH also contained an arachnoid cell layer and hemorrhaging from the granulation tissue on the inside of the outer membrane (facing the cyst cavity), and collagen fibers covered the outside of the membrane (facing the convexity dura) (**Figure 5(b)**, **Figure 5(c)**). These pathological findings, that the membrane of the CSDH consisted of an arachnoid cell layer and granulation tissues, including inflammatory cells, suggested that the lacerated outer membrane of the AC might transform into the membrane of the CSDH.

3. Discussion

Since Davidhoff and Dyke first reported the co-existence of arachnoid cysts and chronic subdural hematomas [15], it has been well known that the ACs located in the middle fossa occasionally associated with the development of CSDH [1]-[8]. However, the exact developmental mechanism of the CSDH remains unknown.

The possible pathogenesis of the CSDHs associated with ACs has been described as follows. First, ACs have less compliance than a normal brain, resulting in reduced intracranial cushioning following trauma. The fluctuating movement of the cyst transfers shearing movements to the outer membrane thus tearing the small vessels between the outer membrane and the dura mater and causing small bleeding into the subdural space [5] [11]. Second, injury of the bridging veins or vessels running in the AC wall due to the easy transfer of pressure through the cyst facilitates the influx of the blood into the subdural space [3] [7] [12]. Third, laceration of the cyst wall due to trauma leads to the influx of CSF and blood within the membrane, and formation of granulation

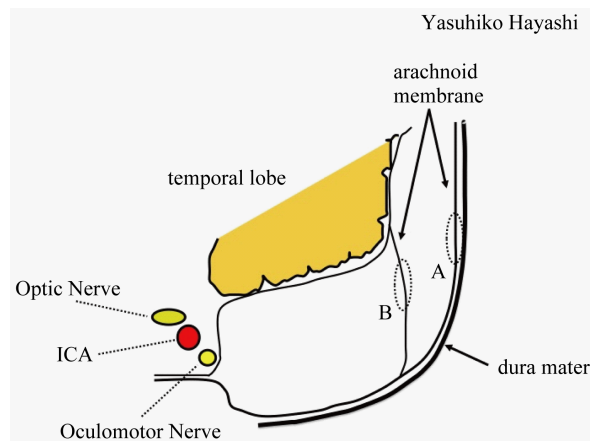


Figure 3. This scheme shows the location of the arachnoid cyst in the left middle fossa and the hematoma in the outer membrane of the cystic wall (anterior-posterior view). Dotted circles indicate the site to perforate and resect the outer membrane of AC and CSDH.

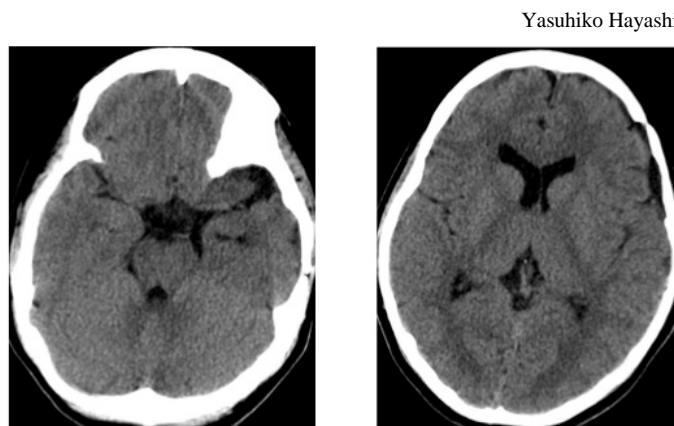


Figure 4. A CT scan of the head obtained 6 months after the operation showed a remarkably reduced arachnoid cyst and a completely diminished subdural hematoma.

tissue with an inflammatory reaction. Subsequent repeated hemorrhages from the organized granulation tissue facilitate the development of hematoma within, inside, or outside the cyst wall [13] [14]. Regarding the granulation formation, it was assumed that the vessels' ingress occurs around the cyst wall after its formation, and the injury of vessels can cause an inflammatory response to lead to the formation of the granulation tissue and subsequent hematoma [16].

Servadei *et al.* suggested that the source of the bleeding causing a subdural hematoma could be identified at the interface between the dura mater and the outer membrane of the AC at the temporal skull base. Therefore, they recommended careful coagulation of the membrane at the skull base [6]. Mori *et al.* speculated that a subdural hygroma caused by the rupture of an arachnoid cyst preceded the formation of the CSDH about the mechanism of CSDH development [4]. Wester *et al.* discussed the possible mechanism of CSDH formation and made an emphasis on the loose attachment of the AC membrane to the dura in the middle fossa, and its possible role as an “extra wall” covering easily bleeding vascular structures in the dura [7]. Kwak *et al.* focused on small bridging vessels between the dura and the outer membrane of the AC and suggested that these small vessels are the source of the initial bleeding leading to CSDH from an AC [3]. Many authors have emphasized the importance of the structures between the dura and the arachnoid membrane.

In our cases, both the inner and outer membranes of the CSDH contained an arachnoid cell layer demon

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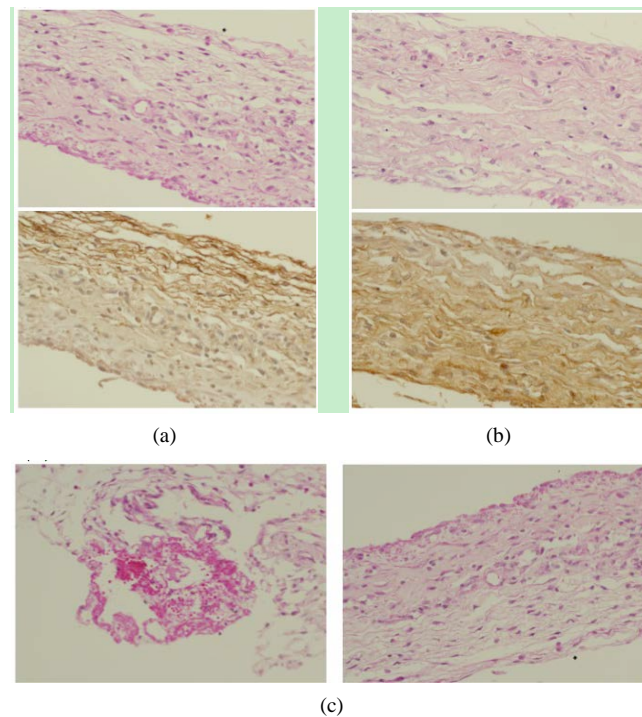


Figure 5. (a) Pathological examination of the outer membrane of the subdural hematoma demonstrated that the arachnoid cell layer on the inside of the outer membrane (facing the cyst cavity), and collagen fibers covered the outside of the membrane (facing the convexity dura) (upper: hematoxylin & eosin, lower: EMA, $\times 200$); (b) Pathological examination of the outer membrane of the arachnoid cyst demonstrated the arachnoid cell layer (upper: hematoxylin & eosin, lower: EMA, $\times 200$); (c) The outer membrane of the subdural hematoma showed the hemorrhage from the granulation tissue on the cyst wall and the inflammatory cells, including lymphocytes.

strated by the immunohistochemistry with EMA, which clearly indicated that the laceration of the arachnoid membrane had occurred in the development of the CSDH. The content of the CSDH was CSF-like fluid with blood, and a hemorrhage from the granulation tissue with the inflammatory response on the surrounding outer membrane was also found on the specimens. These histological findings corresponded to the third developmental mechanism of CSDH associated with AC described above. In the cases of CSDH without ACs, subdural hygroma is produced by a separation of the dura-arachnoid interface, when there is sufficient subdural space. Any pathological condition, inducing the cleavage of tissue within the dural border layer at the dural-arachnoid interface, can induce proliferation of dural border cells with the production of a neomembrane. In-growth of new vessels, especially along the outer membrane, can cause bleeding from these vessels. In our case, the newly developed granulation tissue arose in the intra-arachnoid space, which is quite different from the finding in traumatic CSDH derived from subdural hygroma [17] [18]. The histological features of the outer membrane in CSDH revealed several degrees and combinations of maturity and intensity of the inflammatory reaction and hemorrhage [19].

MRI findings of the CSDH in our case were slightly hyperintensity on T1-WI and hyperintensity on T2-WI, which is compatible with the operative findings of the cyst content with blood and CSF. Our case reasonably adjusts the third mechanism described above; the laceration of the arachnoid cell layer in AC allowed CSF to enter into the intra-membranous space and caused a hemorrhage from the granulation tissue with inflammatory cells in response. To the best of our knowledge, the histological verification of the hemorrhage, possibly leading to the hematoma formation within the cyst wall, could not be found.

Takahashi *et al.* presented a similar case of an AC with a hematoma within the cyst wall [13]. They supposed the pathogenesis of their case as follows; some forces induced by previous minor trauma had split the outer membrane of the cyst, and bloody CSF seeping through the cyst membrane, including many capillary vessels, may have collected in the wall itself.

Our report about the hematoma within the cyst wall of the AC is thought to be valuable, because no report with clear pathological demonstration about this pathogenesis was found before. However, we suspect that this hematoma within the cyst wall is actually not rare, and is simply underreported among the cases of the CSDHs associated with ACs. The reasons are speculated to be as follows; histological examinations have not been performed in many such cases, some ACs are multilobular, showing their property of lacerating in the membrane of the cyst wall, as well as the pathogenesis of a primary AC [10]. At the congenital period, the cyst is considered to be a result of an aberrant development of the subarachnoid space during the splitting of the primitive perimedullary mesh by CSF flow [10] [20]. Further pathological examinations of the walls of the CSDHs associated with ACs are necessary to elucidate hematoma formation within the cyst wall.

Conflict of Interests

All authors declare no conflict of interest.

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Abbreviations

AC: arachnoid cyst

CSF: cerebrospinal fluid

CSH: chronic subdural hematoma

CT: computed tomography

EMA: epithelial membrane antigen

MRI: magnetic resonance imaging

WI: weighted image