

CASE REPORT

A case of organized arachnoid cyst with repeated hemorrhage

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Introduction

Hemorrhage sometimes occurs within arachnoid cysts and consequently increases the size of the cyst. In cases where there is raised intracranial pressure or epileptic seizure, opening the cyst membrane or a cyst–peritoneal shunt will be used to treat the disorder. Chronic subdural hematomas accompanied by arachnoid cysts have been reported. However, to our knowledge organized arachnoid cysts with repeated intracystic hemorrhage have not yet been reported [1–9]. We experienced a case of organized arachnoid cyst after repeated intracystic hemorrhage, and report it here with some histological consideration for its pathogenesis.

Case History

A 36-year-old man was treated for chronic subdural hematoma associated with arachnoid cyst diagnosed

Key Clinical Message

Hemorrhage sometimes occurs within arachnoid cysts, however, organized arachnoid cysts has not been reported. We speculate the previous invasive stress or some kind of infection may have triggered the formation of the organized membrane, which may have formed via a similar mechanism to that for organized chronic subdural hematomas.

Keywords

Arachnoid cyst, endovascular surgery, organized, repeated hemorrhage.

6 years earlier. One year after diagnosis, cyst wall fenestration and a cyst-subarachnoid shunt were performed (Fig. 1A and B). After subsequent repeated hemorrhage, shunt obstruction had led to increased intracranial pressure, and the patient was referred to our hospital (Fig. 1C). Roentgenographic findings revealed a thickened cyst wall and abundant fibrous tissues within the cyst, and cyst fluid density appeared similar to that in chronic subdural hematomas. The cyst wall was adjacent to the middle cerebral artery branch in the Sylvian fissure (Fig. 1D). Cyst wall resection was performed after extending the previous craniotomy (Fig. 2). However, even after the surgical intervention, fresh hemorrhage continued within the cyst, and progressively increased intracranial pressure (Fig. 1E and F). Obstruction of the fenestrated cyst wall was suspected. On angiography, we observed a slight stain from the middle meningeal artery at the base of the arachnoid cyst. Therefore, after embolization of the middle meningeal artery with n-butyl cyanoacrylate,

endoscopic irrigation was performed through the burr hole (Fig. 3). Communicating fenestration within the fresh coagulated blood was confirmed endoscopically (Fig. 4). Six weeks after the surgical intervention, regrowth of the arachnoid cyst has not been observed (Fig. 5).

Histologically, a hyalinized membrane was formed under one layer of arachnoid membrane, and within the hyalinized membrane we observed a vascular rich layer. These findings are distinct from the sinusoidal vascular pattern observed in organized chronic subdural hematoma membranes. The differences may have originated from the anatomical and histological differences between the arachnoid membrane and dura matter (Fig. 6).

Discussion

It is well-known that chronic subdural hematomas develop into an organized type after repeated hemorrhage

[10]. However, this is not the case for arachnoid cysts. An organized membrane is believed to be formed by multiple hemorrhages from various sites of neomembrane, which is gradually transformed into fibrous material, and leads to the formation of multiple organized structures in the hematoma cavity [1]. The recurrence rate of chronic subdural hematoma varies from 3.1% to 33.3%, and in some cases, membranectomy under craniotomy may be necessary [11–14]. Expression of tissue-type plasminogen activator within the outer membrane may be involved in enlargement of chronic subdural hematoma; however, it is difficult to predict its recurrence [15]. A septum in the cavity, localization in the frontal base, a midline shift of more than 5 mm, and recognition of fresh hemorrhage within 4 days after surgery, are thought to be correlated with recurrence of chronic subdural hematomas. Wakui et al. [10] reported that not only enhanced MRI findings, but also endoscopic findings may be useful to predict the recurrence, and to decide appropriate strategies to treat

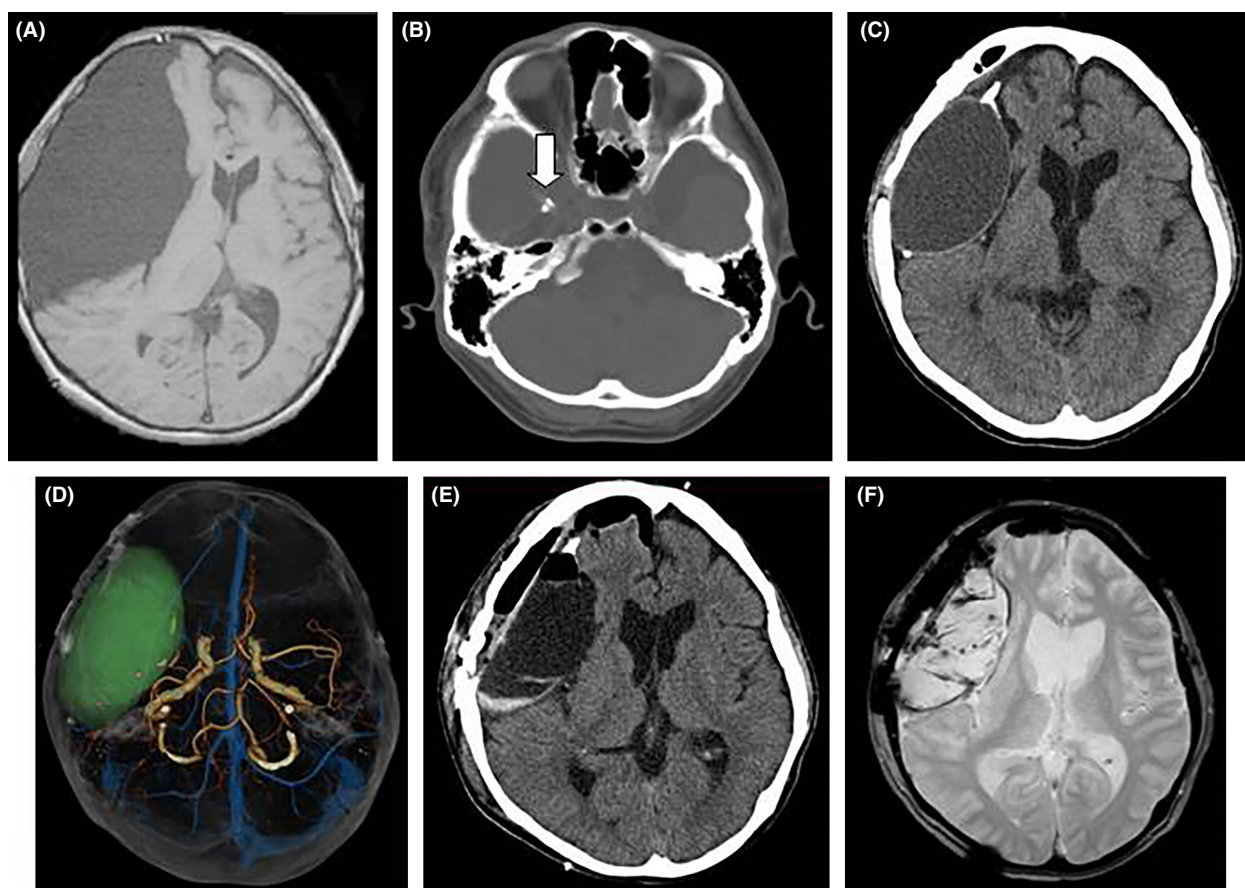


Figure 1. (A) The patient was previously treated under the diagnosis of arachnoid cyst by partial membranectomy and cyst–subarachnoid shunt. (B) The tube previously inserted is indicated by the white arrow. (C) The patient was referred to our hospital because of the increased size of the arachnoid cyst with chronic hemorrhage. (D) Right MCA branch was adherent to the cyst wall on 3D-CTA. (E, F) Repeated intracystic hemorrhage was observed even after the large area of membranectomy on CT and MRI.

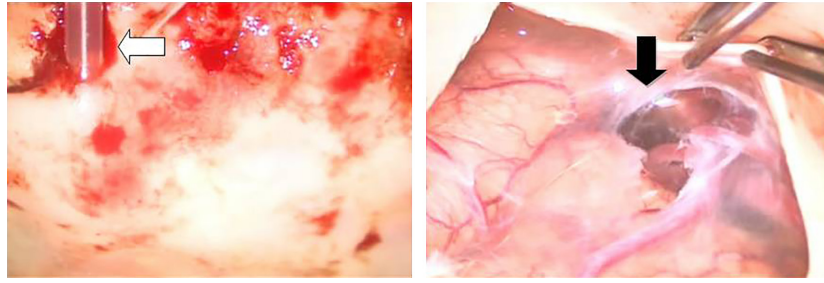


Figure 2. Photograph shows the organized arachnoid membrane. The membrane was hard, but bled easily. The white arrow indicates the previously placed shunt tube, and the black arrow indicates the arachnoid membrane beneath the organized membrane.

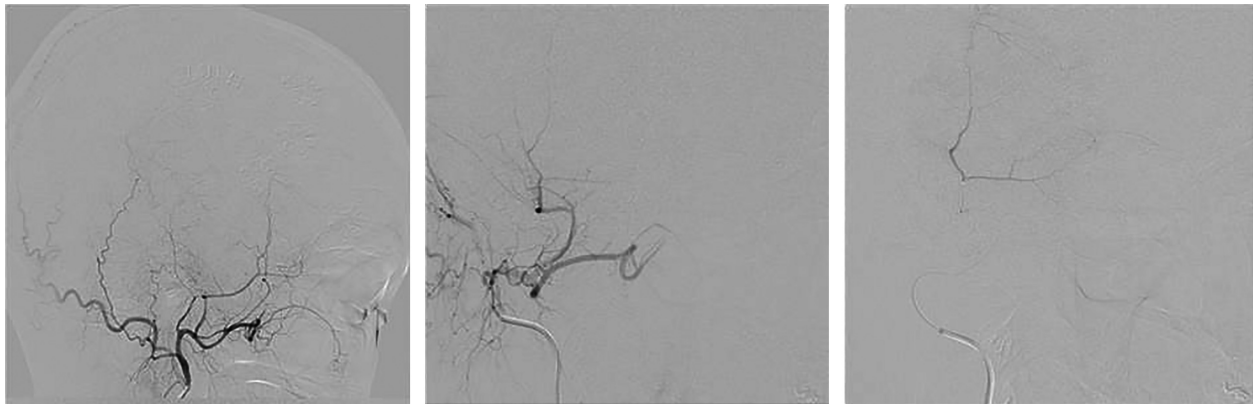


Figure 3. Arteriography revealed a distinct blush to the arachnoid membrane from branches of MMA. Intra-arterial embolization was performed using n-butyl cyanoacrylate against one of the feeding arteries.

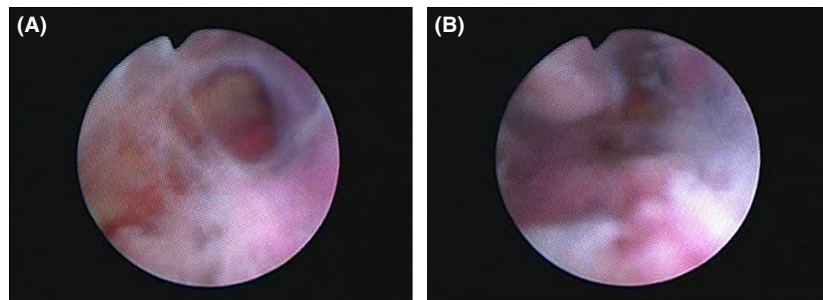


Figure 4. (A) Endoscopic appearance of the second operation revealed that the fenestration of arachnoid cyst membrane was still patent, and irrigation was performed through the burr hole. (B) Coagulated blood was observed endoscopically.

organized chronic subdural hematoma. Treatment strategies for organized chronic subdural hematoma can be applied to cases of organized arachnoid cysts.

Histologically, a hyalinized membrane was formed under the arachnoid membrane, and we observed a vascular rich layer within the hyalinized membrane. Initial surgical intervention, or some kind of infection, may have triggered the formation of the organized membrane [16]. Several factors are reported to be correlated with the

growth of chronic subdural hematoma, such as tissue-type plasminogen activator, hypoxia-inducible factor-1 alpha, vascular endothelial growth factor, cyclooxygenase-2, PI3 kinase/Akt signaling, chemokines, Ras/MEK/ERK signaling, PIGF, JAK-STAT3 signaling pathway, and an effect of corticosteroid has also been reported [15, 17–25]. However, to our knowledge there is no report regarding organized arachnoid cysts. Shuangshoti *et al.* reported two cases of calcified arachnoid cyst, and Papavaslopoulos

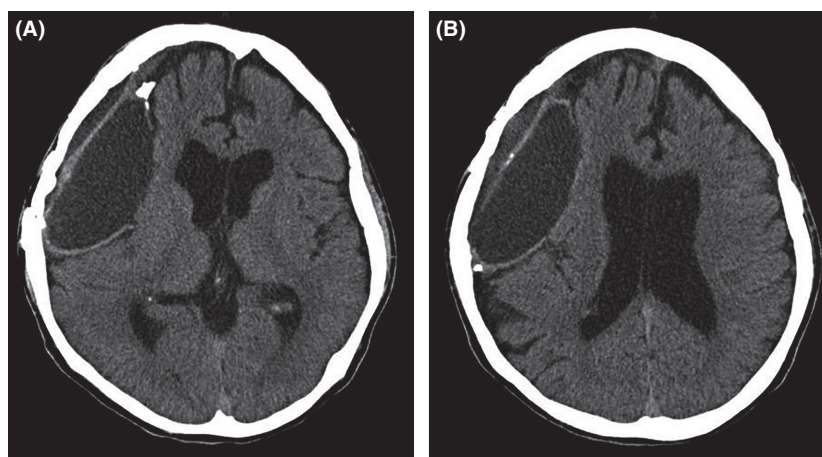


Figure 5. CT image obtained 6 weeks after the surgery demonstrated a good clinical course and no recurrent hemorrhage.

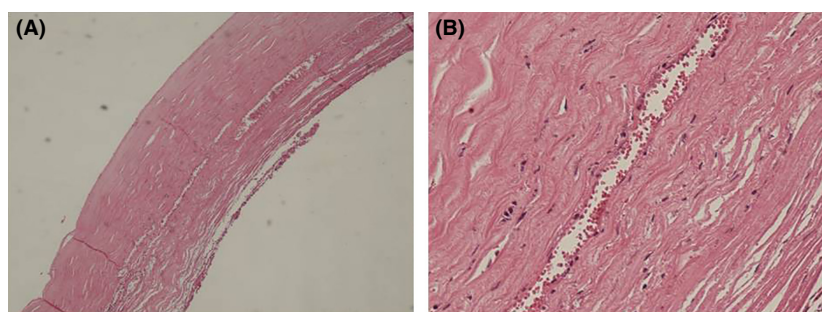


Figure 6. Microscopically, the membrane was formed with a thin arachnoid membrane as an outer layer. Under the arachnoid membrane, a thick hyalinized and organized membrane is seen, and distinct neovascularization is observed within the layer of the organized membrane.

et al. reported a case of ossified arachnoiditis accompanied by syringomyelia, but these are not cases of repeated hemorrhage within arachnoid cysts [26]. Cases must be accumulated and analyzed to elucidate the mechanism of the neovascularization of organized arachnoid cysts. Repeated irrigation or resection of the organized membrane, or both, under craniotomy are the possible treatment strategies. Endovascular embolization has recently been reported to be effective against chronic subdural hematoma. However, this embolization has not yet been reported for organized arachnoid cysts. Careful long-term follow-up of the effect of endovascular surgery for organized arachnoid cysts is warranted.

Conclusion

We describe the case of a 36-year-old male patient who had been treated for a growing arachnoid cyst with organized cyst wall. We successfully treated the case with membranectomy under craniotomy, endovascular embolization, and endoscopic cyst wall fenestration. We

speculate that invasive stress or some kind of infection at the initial surgery may have triggered the formation of the organized membrane with rich vasculature, which may have formed via a similar mechanism to that for organized chronic subdural hematomas.

Conflict of Interest

None. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-Reported COI Disclosure Statement Forms through the website for JNS members.

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