



Case Report

A unique case of Sylvian arachnoid cyst complicated by chronic subdural hematoma

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ABSTRACT

Background: Arachnoid cysts (ACs) complicated by chronic subdural hematoma (CSDH) are a rare but distinct entity.

Case Description: A 27-year-old man previously diagnosed with Sylvian AC presented to the hospital with a persistent headache. He was not aware of any preceding head trauma. However, he frequently performed bench presses at the gymnasium, especially 4 weeks prior. The patient did not exhibit any neurological deficits at presentation. Computed tomography revealed slightly low-density areas in the right cerebral convexity. Magnetic resonance imaging revealed compressive masses in the right middle fossa and cerebral convexity. The patient underwent a craniotomy. Reflection of the dura mater exposed thickened arachnoid membrane. Making an incision resulted in the egress of fluid hematoma. The membrane separating the subdural hematoma and inner AC possessed fine vasculature and adjacent holes. Furthermore, there were fragile clots adhered to the inner wall of the cyst. Microscopic findings of the separating membrane were consistent with inflammatory granulation tissue, similar to those of the outer membrane of CSDH.

Conclusion: Exertional hypertension associated with the bench press may result in the rupture of fine arteries distributed over the AC wall. Under certain circumstances, the AC wall may transform into the outer membrane of CSDH.

Keywords: Arachnoid cyst, Chronic subdural hematoma, Exertional hypertension, Rupture of arachnoid cyst

INTRODUCTION

The human brain contains two types of arachnoid membranes: the outer and inner membranes. The former surrounds the whole brain, whereas the latter divides the subarachnoid spaces into nine cisterns.^[4] An arachnoid cyst (AC) generally arises as a congenital anomaly of the developing arachnoid cisterns in early intrauterine life caused by the anomalous splitting of the arachnoid membranes.^[1,9] ACs may be complicated by subdural hygroma and chronic subdural hematoma (CSDH), resulting from AC rupture frequently associated with contact sports, although it may occur spontaneously in rare instances. For such ACs, burr-hole drainage is thought to be an effective first-choice management.^[1,3,8,10,12] Sylvian ACs are a distinct entity that has been shown to be more likely to rupture than AC in other intracranial locations.^[7,13]

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Microscopically, CSDH can be classified into four types, with inflammatory and hemorrhagic inflammation types being the most frequent.^[2] Recent investigation has documented that compared to systemic blood, an increased number of immune blood cells are present in CSDH.^[5]

An exertional hypertension caused by acute high-intensity resistance exercises, such as the bench press, is reported to increase the intravascular pressure of the cerebral arteries.^[6]

Herein, we report a case of Sylvian AC complicated by CSDH presenting with peculiar intraoperative and microscopic findings. In this patient, exertional hypertension caused by bench press was assumed to be the underlying cause.

CASE PRESENTATION

A 27-year-old man presented to the hospital with a 3-week history of headaches. The patient was an office worker who was unaware of any preceding head trauma. He had previously been diagnosed with an asymptomatic Sylvian AC at the age of 10 years when he suffered a simple head injury and underwent cranial computed tomography (CT) at a local hospital. Since then, the AC remained asymptomatic. He did not play any contact sport but frequently performed bench presses at the gymnasium, especially 4 weeks prior. At presentation, he was well-oriented and did not exhibit any neurological deficits. Cranial CT revealed slightly low-density areas in the right temporal, frontal, and parietal convexities, with the cerebellopontine angle and ambient cisterns dilated on the right side. In addition, smooth-contoured erosive changes were observed in the right temporal skull [Figure 1]. Cerebral magnetic resonance imaging (MRI) identified compressive masses in the right middle fossa and the cerebral convexity. The former lesion presented as more hypointense than the latter on both T1- and T2-weighted sequences. Linear structures with varying thicknesses and hypointensity on both sequences were observed in and around the former lesion [Figure 2]. Magnetic resonance (MR) angiography revealed unidentifiable distal parts of the right middle meningeal artery and elevated M1 portion of the right middle cerebral artery [Figure 3]. The patient underwent frontotemporal craniotomy with a presumptive diagnosis of hemorrhagic AC. Intraoperatively, a reflection of the apparently intact dura mater revealed an unusually thick arachnoid membrane. On making an incision in the arachnoid resulted in an egress of dark red, fluid hematoma. The lower surface of the arachnoid was smooth, and after the evacuation of the fluid hematoma, soft and fragile clots were observed to scatter on the membrane, separating the subdural hematoma from the inner AC. The separating membrane possessed fine vasculature and two adjacent smooth-contoured, round holes. Resection of the membrane exposed fragile clots loosely adhered to the entire inner cyst wall. Circumferential removal of these clots revealed an intact frontal cortex [Figure 4]. Photomicrographs of the

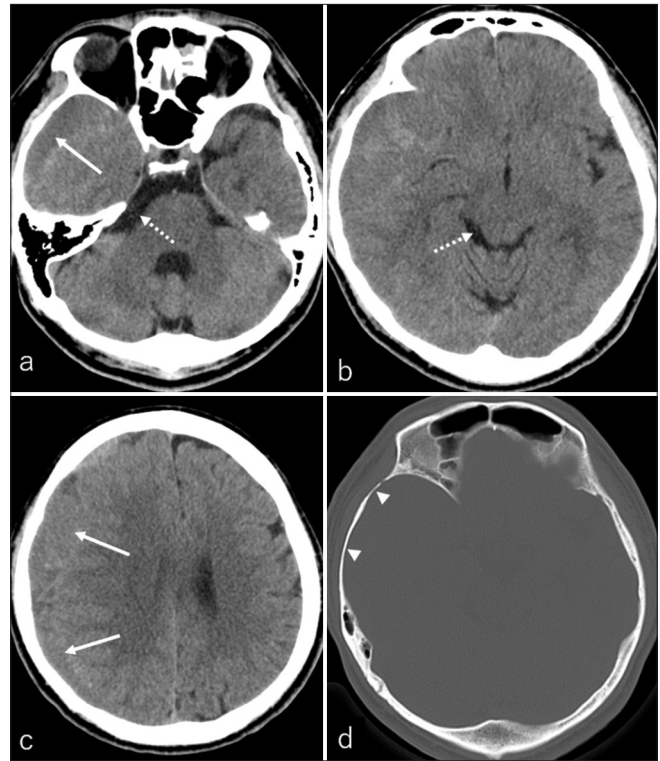


Figure 1: Axial computed tomography scans at presentation, taken (a) at the level of the inferior horn of the lateral ventricle, (b) third ventricle, and (c) upper lateral ventricle, showing slightly low-density areas in the right temporal, frontal, and parietal convexities (a and c, arrows). The cerebellopontine angle and ambient cisterns show dilation on the right side (a and b, dashed arrow). (d) A bone-target image at the same level as Figure (a) shows smooth-contoured erosive changes in the right temporal skull (arrowheads).

resected separating membrane further showed lymphocytic and eosinophilic infiltration, predominantly in the inner part of the membrane [Figure 5a]. Immunohistochemical examination revealed positive staining for CD68 and CD34, predominantly in the inner part of the membrane, suggesting monocytic and microvascular proliferation [Figures 5b and c]. Stains for glial fibrillary acidic protein and AE1/3 were negative. These findings were consistent with those of inflammatory granulation tissue or the inflammatory type of the outer membrane of CSDH.^[2] The patient's postoperative course was uneventful. MRI performed on postoperative day 5, at the level of the third ventricle, revealed a less compressive, extra-axial cyst in the right Sylvian fissure and satisfactory evacuation of the CSDH [Figure 6]. At present, the patient has been followed-up without recurrence of CSDH or hemorrhage in the AC for 6 months.

DISCUSSION

The present patient was previously diagnosed with Sylvian AC and had been asymptomatic for a long period prior to the

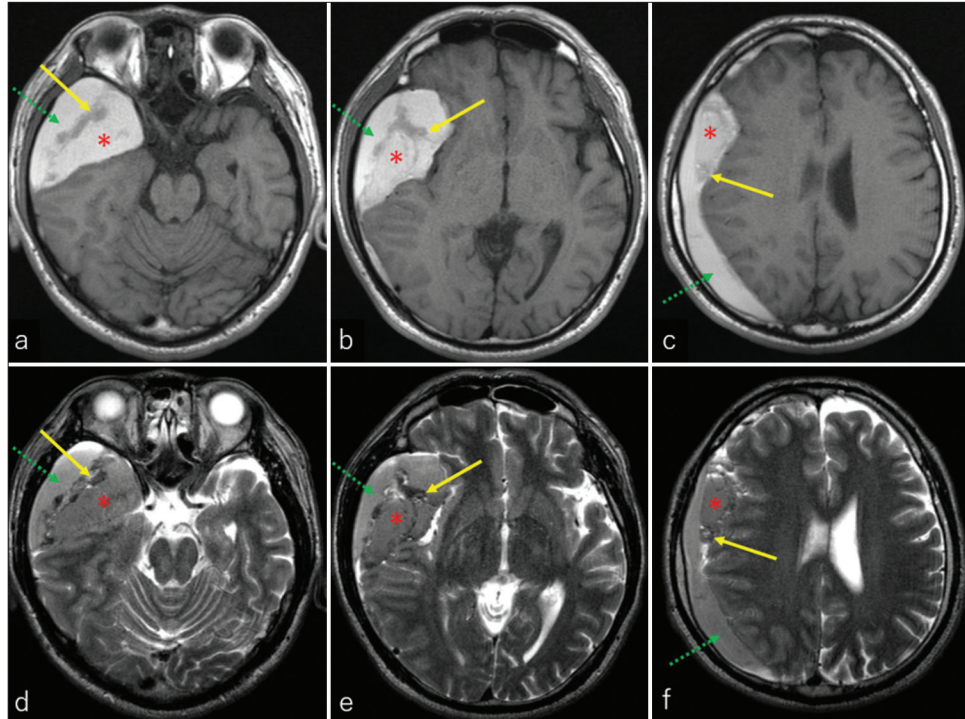


Figure 2: (a-c) Axial T1 and (d-f) T2-weighted magnetic resonance imaging taken at the level of the (a, d) inferior horn of the lateral ventricle, (b, e) third ventricle, and (c, f) upper lateral ventricle showing compressive masses in the right middle fossa (a-f, asterisk) and cerebral convexity (a-f, dashed arrow). The former lesion appears more hypointense, compared to the latter, on both T1- and T2-weighted sequences. Of note, linear structures with varying thicknesses and presenting hypointensity on both sequences can be observed in and around the former lesion (a-f, arrow).



Figure 3: Anteroposterior view of magnetic resonance angiography showing unidentifiable distal parts of the right middle meningeal artery, compared to the contralateral side (arrows), with elevated M1 portion of the right middle cerebral artery (dashed arrow).

development of the headache. Neuroimaging examinations at the presentation, as well as subsequent intraoperative findings, indicated a fluidy subdural hematoma and adjacent cyst accompanied by hemorrhagic changes. Moreover,

postoperative MRI revealed an extra-axial cyst within the Sylvian fissure. In addition, intraoperative and microscopic findings suggested the development of CSDH. Therefore, we hypothesized that the pre-existing Sylvian AC may have ruptured, followed by the formation of the CSDH. Given that the inflammatory changes, histologically similar to those of CSDH,^[2,5] were predominantly present in the inner part of the AC wall, and peculiar fragile clots were predominantly observed on the inner cyst wall, we thought that the CSDH might have developed from the AC and extended subdurally through holes found in the AC wall.

The present patient was unaware of the preceding head trauma and had frequently performed bench presses 4 weeks before the presentation. Prior studies have indicated that exertional hypertension associated with the bench press can increase the intravascular pressure in the cerebral arteries.^[6] Furthermore, in the present case, part of the AC wall separating from the CSDH was vascularized near the holes. Therefore, we assumed that transient but repetitive hypertension induced by bench press might have resulted in ruptures of fine vessels distributed over the AC wall, as well as adjacent arachnoids, followed by the formation of holes in the wall. In addition, consecutive bleedings predominantly into the AC cavity, hypothesized from the intraoperative

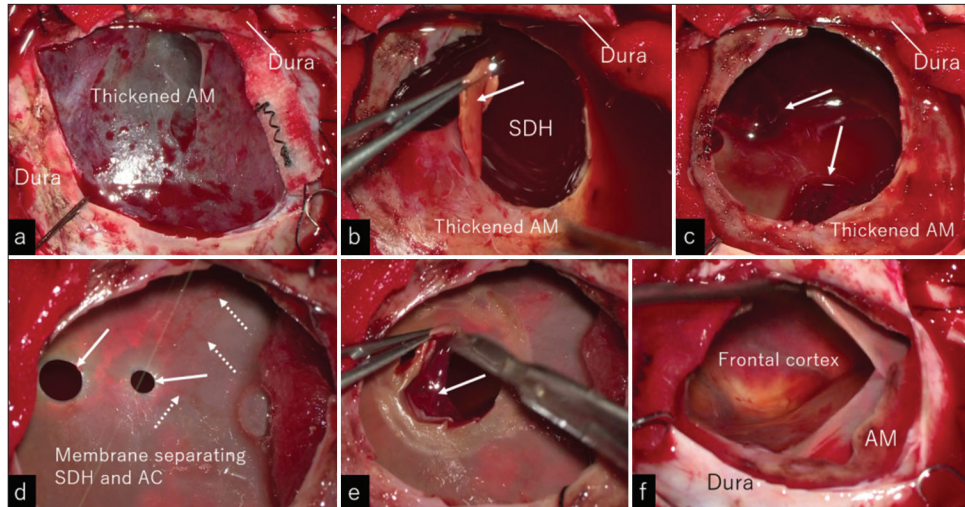


Figure 4: (a-f) Intraoperative photos showing the procedures in a step-by-step manner. (a) Reflection of the dura mater exposed unusually thickened arachnoid membrane. (b) Making an incision to the arachnoid resulted in the egress of dark red, fluid hematoma. The lower surface of the arachnoid was smooth (arrow). (c) After the evacuation of the fluid hematoma, soft and fragile clots were observed to scatter on the membrane separating the fluid hematoma and inner arachnoid cyst (arrows). (d) The separating membrane possessed fine vessels (dashed arrows) and two adjacent smooth-contoured, round holes (arrows). (e) Resection of the membrane exposed fragile clots loosely adhered to the entire inner cyst wall (arrow). (f) Circumferential removal of the clots revealed the intact frontal cortex. AC: Arachnoid cyst; AM: Arachnoid membrane; and SDH: Subdural hematoma.

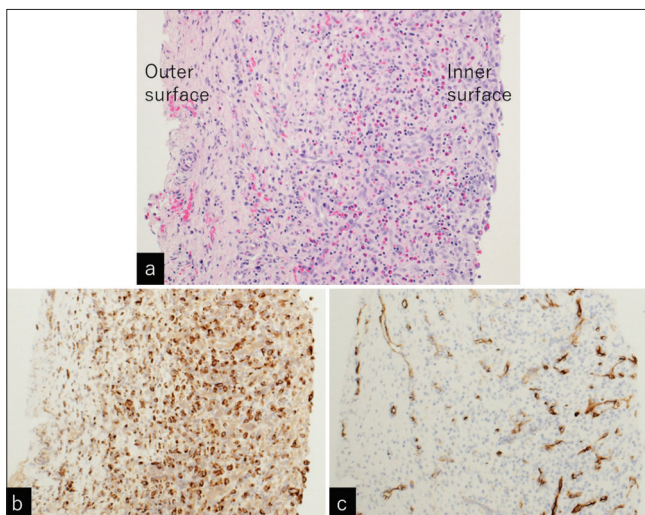


Figure 5: (a) Photomicrographs of the resected separating membrane showing lymphocytic and eosinophilic infiltrations, predominantly in the inner part of the membrane. Immunohistochemical examination showed positive staining for (b) CD68 ($\times 20$) and (c) CD34 ($\times 20$) predominantly in the inner part of the membrane, suggesting monocytic and microvascular proliferation. (a) Hematoxylin and eosin staining, $\times 20$.

findings, may have triggered the development of CSDH. The co-occurrence of CSDH and AC is not unique. However, to our knowledge, the hypothesis on the development of CSDH in association with a cerebral AC has not been

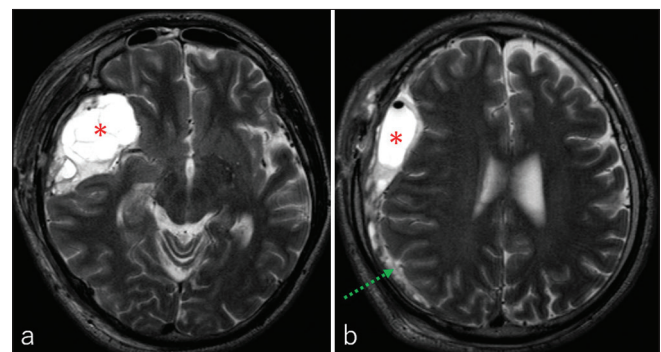


Figure 6: Axial T2-weighted, magnetic resonance imaging, performed on postoperative day 5, at the level of the (a) third ventricle and (b) upper lateral ventricle showing less compressive, extra-axial cyst in the right Sylvian fissure (asterisk), and satisfactory evacuation of chronic subdural hematoma (dashed arrow).

described.^[1-3,10,12,13] Under certain circumstances, the AC wall may transform into granulation tissue, histologically similar to the outer membrane of CSDH. It may be recommended that patients with ACs avoid performing strenuous bench presses, as this action can place a heavy burden on the body and significantly increase the intravascular pressure of the cerebral arteries.

The diameter of the middle meningeal arteries has been reported to increase in patients with mature CSDHs.^[11] However, in our case, MR angiography did not delineate the

distal parts of the artery on the affected side, instead of an increased flow of the artery. This finding suggests that the etiology of the present CSDH is different from that of a typical, growing CSDH with a mature outer membrane supplied by the artery.

CONCLUSION

Exertional hypertension associated with the bench press may result in the rupture of fine arteries distributed over the AC wall. Under certain circumstances, the AC wall may transform into the outer membrane of CSDH.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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