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A Case of Subdural Hygroma due to a Ruptured Arachnoid Cyst in the Middle Cranial Fossa That Improved after Long-term Subdural Drainage: A Case Report and Review of the Literature

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Abstract

Arachnoid cysts have the potential to rupture, leading to the development of a subdural hygroma following minor trauma. Although surgery may be considered in cases of increased intracranial pressure (ICP) or regional neurological symptoms, the optimal approach remains unclear. We report a case of subdural hygroma due to a ruptured arachnoid cyst (SHrAC) with elevated ICP successfully treated with long-term subdural drainage for over 1 month. A 26-year-old man with persistent headache was admitted to our hospital. Magnetic resonance imaging revealed an arachnoid cyst within the left middle cranial fossa and a subdural hygroma in the left frontotemporal region. He was referred to our neurosurgery department for surgical intervention due to elevated ICP. Although burr hole surgery was initially performed, subsequent recurrence of elevated ICP necessitated the insertion of a subdural peritoneal shunt. However, the shunt was then removed following the development of postoperative meningitis, and a subdural drain was placed to control ICP. Cerebrospinal fluid (CSF) drainage gradually decreased, and the elevated ICP improved. The subdural drain was removed approximately one and a half months after drain placement. The subdural hygroma progressively reduced and completely disappeared 4 months after drain removal. The gradual reduction in the pressure difference between the arachnoid cyst and the subdural hygroma due to long-term CSF drainage and inflammation caused by meningitis may have contributed to close arachnoid membrane laceration. Although alternative approaches, such as shunt insertion and basal fenestration, should always be considered in SHrAC treatment, long-term subdural drainage can be an option.

Keywords: arachnoid cyst, head trauma, traumatic rupture, subdural hygroma, subdural drainage

Introduction

Although arachnoid cysts represent approximately 1% of all intracranial occupational lesions,[1)] most are asymptomatic and do not require therapeutic intervention.[2)] Traumatic or spontaneous rupture of the cyst wall may result in a subdural hematoma or hygroma.[3)] Surgical treatment of a subdural hygroma due to a ruptured arachnoid cyst (SHrAC) is considered when intracranial hypertension or regional neurological symptoms occur. Various approaches, such as burr hole surgery and temporary subdural drainage,[1)] shunt insertion,[4)] and basal fenestration,[5)] have been reported; however, the appropriate treatment strategy remains unclear. First, we report here a case of long-term subdural drainage of SHrAC within the middle cranial fossa and postoperative meningitis.

Case Report

A 26-year-old man with a chronic headache after head trauma visited the neurology department of our hospital. He had an orthostatic headache and diplopia in the left

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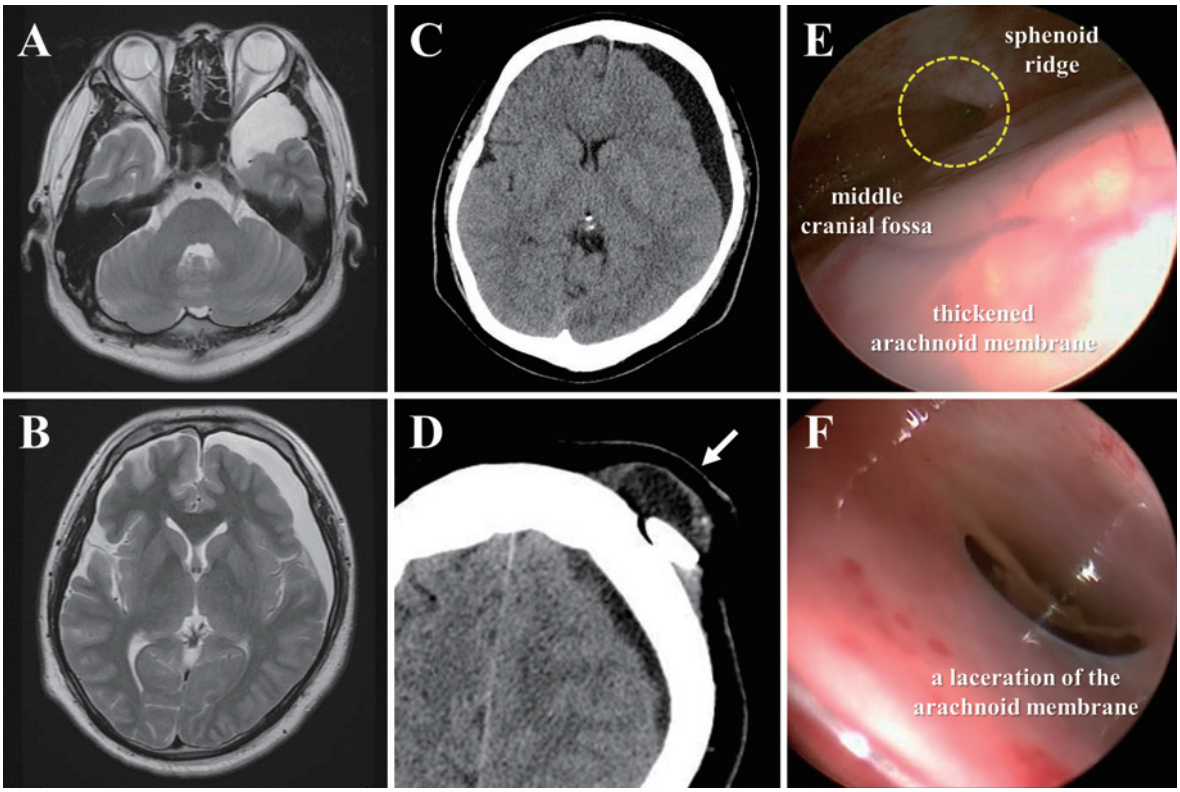
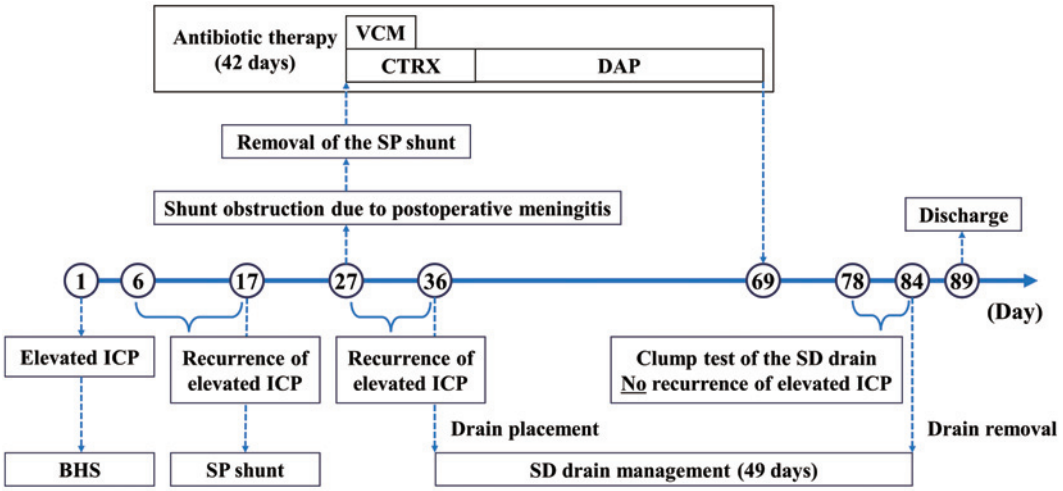


Fig. 1 Magnetic resonance imaging of the head conducted prior to the initial surgery showing an arachnoid cyst in the middle cranial fossa (A), along with accumulation of cerebrospinal fluid in the left subdural space (B). Computed tomography of the head prior to the second operation showing subdural hygroma in the frontotemporal convexity (C) and skin protrusion above the burr hole (D). Rigid neuroendoscopy reveals the presence of an arachnoid cyst in the middle cranial fossa (E, dotted circle) and a laceration of the arachnoid membrane (F).

lateral vision but no other neurological abnormalities. Cerebrospinal fluid (CSF) examination revealed elevated intracranial pressure (ICP) with an initial pressure of >300 mmH_2O, with a normal cell count (2/μL), protein concentration (27 mg/dL), and glucose level (67 mg/dL). T2-weighted head imaging revealed a Galassi type 2 arachnoid cyst within the left middle cranial fossa and a hyperintense area in the left frontotemporal subdural space (Fig. 1 A-B). Cerebral angiography showed no evidence of venous sinus obstruction/stenosis or any abnormal vascular network around the arachnoid cyst. He was diagnosed with a subdural hygroma and elevated ICP secondary to a ruptured arachnoid cyst. The elevated ICP persisted despite conservative treatment with mannitol and acetazolamide. He was then transferred to the neurosurgery department for surgical intervention. Figure 2 shows the clinical course of the present case. On day 1, burr hole surgery under local anesthesia was performed to improve the elevated ICP. Vigorous CSF drainage occurred upon incision of the subdural capsule. No hematoma was observed, and the findings indicated a subdural hygroma. Postoperatively, subjective symptoms such as headaches improved. On day 6, re-stitching was required due to CSF leakage from the surgical wound. On day 8, CSF examination revealed the recurrence of intracranial hypertension with an initial pressure of 280 mmH_2O. Conservative treatment was continued, but wound re-stitching due to CSF leakage was performed multiple times. On day 14, the patient's headache recurred. On day 15, computed tomography (CT) of the head revealed accumulation of a subdural hygroma, poor delineation of the cerebral sulcus, and skin protrusion above the burr hole (Fig. 1C-D). CSF examination revealed an initial pressure of >300 mmH_2O, but there were no signs of infection. On day 17, a subdural peritoneal (SP) shunt was inserted to regulate the elevated ICP. The symptoms of intracranial hypertension postoperatively improved, and a CT scan of the head revealed no worsening of the subdural hygroma. However, on day 27, ophthalmalgia and recurrence of the skin protrusion above the burr hole were observed. Shuntography revealed obstruction of the ventricular catheter, and the shunt system was subsequently removed. During surgery, the pale-yellow CSF was extravasated on opening the wound. The CSF examination revealed an elevated cell count (2477/μL), polynuclear cell count (1720/μL), protein concentration (290 mg/dL), and normal glucose level (53 mg/dL). The patient was diagnosed with shunt obstruction due to postoperative meningitis. After meningitis improved, reconstruction of the SP



BHS: burr hole surgery, CTRX: Ceftriaxone, DAP: Daptomycin, ICP: intracranial pressure, SD: subdural, SP: subdural peritoneal, VCM: Vancomycin

Fig. 2 Clinical course of the present case. On day 1, burr hole surgery was performed. On day 6, elevated intracranial pressure (ICP) recurred. On day 17, a subdural peritoneal shunt was inserted. On day 27, the shunt was removed due to postoperative meningitis. On day 36, a subdural drain was placed to control the ICP. From day 78, although the subdural drain was clamped, there was no recurrence of the elevated ICP. On day 84, the subdural drain was removed. On day 89, the patient was discharged. Antibiotic therapy was performed from days 27 to 69.

shunt was planned. On day 36, a subdural drain was placed to control the elevated ICP temporarily (Fig. 3A-B). Rigid neuroendoscopy revealed a thickened cerebral capsule and an arachnoid cyst with a significant laceration within the middle cranial fossa (Fig. 1E-F). CSF drainage from the cyst was also observed. The trajectory of the rigid neuroendoscopy was not aligned, and the inside of the arachnoid cyst could not be clearly observed. For postoperative management, the drip chamber of the subdural drain was positioned 10 cm above the external ear canal, and the amount of CSF drainage was 400-600 mL/day. The drip chamber height was gradually increased and was maintained at 20 cm from day 39. Symptoms of elevated ICP, such as a headache and imaging findings, remained unchanged (Fig. 3C-D). Although antibiotic therapy with ceftriaxone and vancomycin was initiated on day 27, vancomycin was discontinued on day 34 due to drug-induced renal dysfunction. However, on day 39, fever and increased C-reactive protein (6.88 mg/dL) were identified, indicating that ceftriaxone was ineffective. On day 41, daptomycin treatment was initiated to target methicillin-resistant coagulase-negative *Staphylococci* and methicillin-resistant *Staphylococcus aureus*. Thereafter, the patient's fever and blood and CSF findings improved, and antibiotic therapy was terminated on day 69 (6 weeks). During this period, CSF culture examinations were performed several times, but all were negative; the causative bacteria could not be identified. As meningitis did not recur, the subdural drain was clamped and removed on days 78 and 84, respectively. The duration of the subdural drain placement was extended to 49 days. After the subdural drain was removed, there was no recurrence of elevated ICP or deterioration in imaging findings. Consequently, the patient was discharged on day 89. After discharge, a follow-up head magnetic resonance imaging was performed. The subdural hygroma gradually reduced in size (Fig. 3E-F) and completely disappeared approximately 4 months after the subdural drain removal (Fig. 3G-H).

Discussion

In this case, the surgical management emphasized two crucial clinical issues: Long-term subdural drainage helped improve SHrAC with elevated ICP and the timing of additional surgery for drainage procedures, such as burr hole surgery or temporary subdural drainage for SHrAC, should be noted.

We believe that long-term subdural drainage is advantageous for improving SHrAC. Arachnoid cysts associated with subdural hygroma are rare, and treatment methods remain controversial.[4] We searched for the keywords "arachnoid cyst" and "subdural hygroma" in PubMed. We identified 77 cases of arachnoid cysts complicated by subdural hygroma, for which the treatment strategy was described.[1,3-42] Conservative treatment was selected as the initial treatment for 21 patients.[7,8,10,12,13,15,33,35-37,40] However, several patients undergo surgical interventions. A total of 60 patients comprising the 4 who had conservative treatment underwent surgery: basal fenestration was performed in 27,[5,9,15-17,21-23,30,31,34,39,42,43] drainage procedures (burr hole surgery

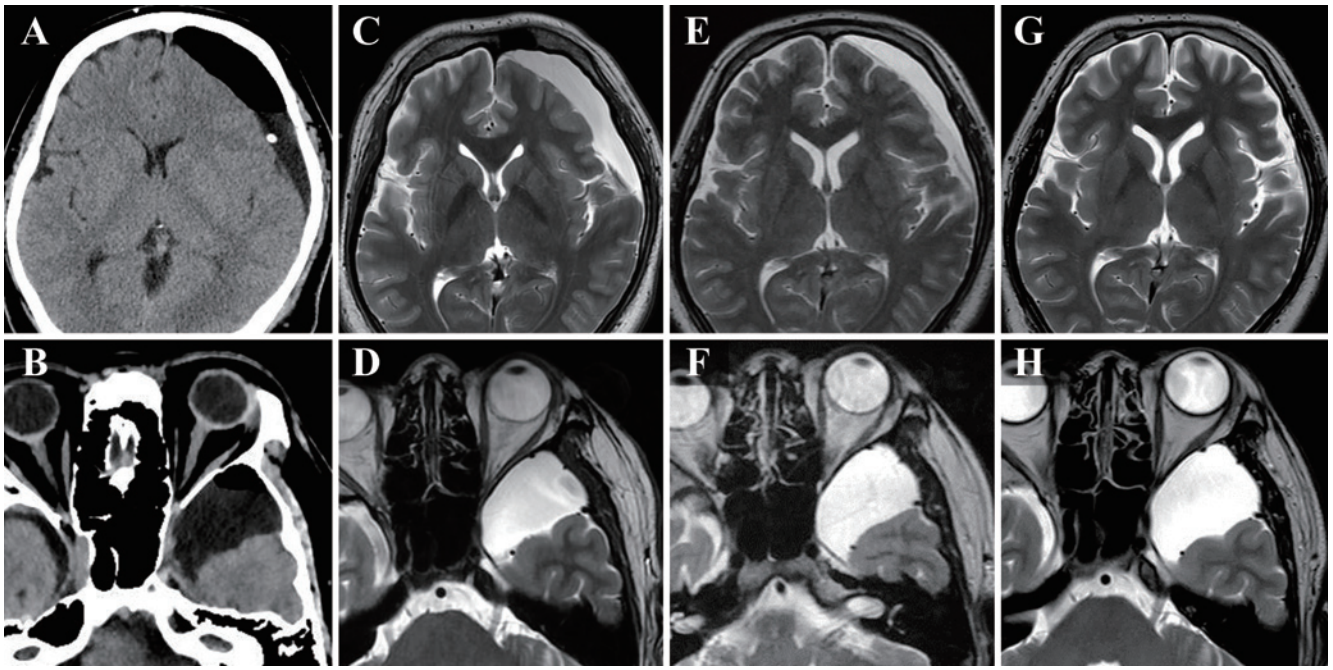


Fig. 3 Computed tomography of the head immediately following subdural drain placement (A, B) and magnetic resonance imaging (MRI) of the head 37 days (C, D) after subdural drain placement reveals no changes in the subdural hygroma and arachnoid cyst. Subsequent MRI of the head on 10 days (E, F) and approximately 4 months (G, H) after subdural drain removal demonstrates a gradual decrease and disappearance of cerebrospinal fluid within the left subdural space, with no change of the arachnoid cyst.

and temporary subdural drainage) in 18,[1,3,6,18,24,26-30,35,38] shunt insertion (SP shunt or cyst peritoneal shunt) in 14,[1,4,11,14,19,20,41] and others in one case.[5] To our knowledge, there has never been a case of SHrAC in a patient who had undergone long-term subdural drainage for more than 1 month, as in this case. SHrAC is caused by a laceration of the arachnoid membrane, forming a one-way valve mechanism and CSF outflow through the laceration.[11] Persistent CSF drainage from the arachnoid cyst may contribute to the failure to normalize the elevated ICP after subdural drainage.[28] Based on these mechanisms, the drainage procedure alone is unlikely to improve SHrAC because it does not effectively seal the laceration of the arachnoid membrane or establish novel CSF circulation. Therefore, we assume that shunt insertion and basal fenestration are more reliable methods for controlling elevated ICP in SHrACs. However, if infection occurs, the insertion of an artificial device or craniotomy becomes difficult, and treatment methods to control the elevated ICP in SHrAC are limited. Although the precise timing of laceration closure is unknown, a subdural drain must be placed to control elevated ICP. Shunt removal in patients who underwent shunt insertion for SHrAC has been reported. In these reports, the shunt was removed from seven cases.[4,24] The duration of shunt implantation ranged from 6 weeks to 3 years. The gradual reduction in pressure due to CSF drainage from the arachnoid cyst to the subdural hygroma may alleviate the strain on the laceration, thus leading to the disappearance of the check valve mechanism. Although one patient required shunt removal due to shunt infection, subsequent shunt replacement was not required.[11] It is possible that the inflammation caused by meningitis also contributes to laceration closure. In our case, a laceration was identified on the surface of the arachnoid cyst during the fourth surgery. The volume of subdural drainage decreased as meningitis improved, and continuing antibiotic therapy for 6 weeks sufficiently improved arachnoid membrane laceration in our case. Based on these clinical courses, we hypothesized that long-term subdural drainage for SHrAC with elevated ICP could be an option to improve arachnoid membrane laceration. However, the transition to shunt insertion or basal fenestration should always be considered.

The optimal timing for additional surgery, such as shunt insertion or basal fenestration for drainage, is unknown. Of the 60 patients with SHrAC who underwent surgery, 15 required additional surgery. Of the 27 patients who underwent shunt insertion, 2 also underwent basal fenestration (7.7%).[15,39] Although 2 of the 14 patients who underwent shunt insertion additionally received basal fenestration (15.4%),[4] it was associated with shunt removal. In contrast, 11 of the 18 patients who initially underwent drainage procedures also underwent shunt insertion or basal fenestration (61.1%).[1,3,18,24,27,28,30,32,35] Table 1 shows the 18 cases in which drainage procedures were performed.[1,3,6,18,24,26-30,32,35,38] Although subdural drainage was performed in 10 of the 18

Table 1 Eighteen reported cases of extracranial drainage for subdural hygroma due to arachnoid cyst

Year	Author	Age, sex	Cause of rupture	First surgery	Duration of SD drainage	Additional surgery	Timing of additional surgery
2023	Almousa	11 F	Spontaneous rupture	Burr hole	—	—	—
2004	Gupta	22 M	Trauma	Burr hole	—	—	—
2003	Huang	33 M	Trauma	Burr hole	—	—	—
1997	Parsch	18	Trauma	Burr hole	—	—	—
1992	Minamori	60 F	Not described	Burr hole	—	—	—
2006	Offiah	8 M	Trauma	Burr hole	—	SP shunt	Shortly after
2004	Cakir	9 M	Spontaneous rupture	Burr hole	—	SP shunt	Subsequent conversion
1998	Vigil	16 M	Trauma	Burr hole	—	Cyst fenestration	1 week
1997	Albuquerque	25 M	Trauma	SD drain	1 day	—	—
1997	Albuquerque	9	Trauma	SD drain	Unknown	—	—
2002	Gelabert-González	6 M	Trauma	SD drain	3 days	SP shunt	3 days later
2010	Hasegawa	5 M	Trauma	SD drain	4 days	Arachnoidplasty	1 month later
1997	Albuquerque	6 M	Trauma	SD drain	Over 3 days	SP shunt	Subsequent conversion
1997	Albuquerque	25	Trauma	SD drain	Over 3 days	SP shunt	Unknown
2006	Klein	14 M	Trauma	SD drain	Unknown	SP shunt	Unknown
2004	Poirrier	15 M	Spontaneous rupture	SD drain	Unknown	SP shunt	Unknown
1997	Parsch	5	Trauma	SD drain	Unknown	SP shunt	Unknown
1997	Parsch	12	Trauma	SD drain	Unknown	Cyst fenestration	Unknown

SD, subdural; SP, subdural peritoneal

cases, most reports did not clearly describe the sufficient duration for subdural drainage and the optimal timing of additional surgery. In some of these reports, the timing of additional surgery was expressed as "3 days later,[30]" "1 week,[32]" "shortly after,[3]" or "subsequent conversion.[1,27]" There have been reports of cases of subdural drainage that evaluated potential shunt insertion based on ICP monitoring.[1,28] In these cases, postoperative ICP monitoring at 24 h showed a sustained ICP of 300 mmH_2O[28] or persistently elevated ICP beyond 72 h,[1] and shunt insertion was performed. These reports suggest the possibility of early recurrence of elevated ICP in the postoperative period. In our case, conservative treatment failed to improve the symptoms and elevated ICP, thus indicating surgery. After no improvement with burr hole surgery, an SP shunt was inserted to control the elevated ICP. Although our treatment approach up to SP shunt insertion aligned with previous reports, we thought that the timing of the additional surgery was too late. During this period, recurrent CSF leakage from the wound due to increased ICP was observed. It is thought that the delayed wound healing contributed to subsequent infections. Although direct ICP monitoring was not conducted in our case, the presence of CSF leakage on postoperative day 5 and the abnormal CSF examination results on postoperative day 7 strongly suggested elevated ICP. When a drainage procedure is performed for SHrACs, additional surgery should be considered during the early postoperative phase.

Conclusions

The patient had difficulty controlling the elevated ICP caused by SHrAC in the middle cranial fossa, and multiple operations were performed. Long-term subdural drainage for more than 1 month contributed to the control of elevated ICP and arachnoid membrane laceration improvement. Additional treatments, such as shunt insertion and basal fenestration, should be considered in the early postoperative period with the initial performance of burr hole surgery or temporary subdural drainage for SHrAC.

Informed Consent

Informed consent was obtained from the patient.

Conflicts of Interest Disclosure

All authors declare no conflict of interest.

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