

Neuroradiology

Arachnoid cysts with spontaneous intracystic hemorrhage and associated subdural hematoma: Report of management and follow-up of 2 cases

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ABSTRACT

Arachnoid cysts are one of the most frequently encountered intracranial space-occupying lesions in daily neurosurgery and neuroradiology practice. Majority of arachnoid cysts, particularly those of smaller sizes, have a benign uneventful lifetime course. Certain symptoms may indicate serious complications related to underlying arachnoid cysts. Hemorrhage is one of the most fearsome complications of arachnoid cysts and almost all reported cases in the literature have undergone surgical correction. In this study, we aimed to present clinical and radiologic follow-up findings in two adult cases of intracranial arachnoid cyst with spontaneous intracystic hemorrhage and associated subdural hematoma, one of which was successfully treated conservatively. In addition, we broadly summarized and discussed pertinent studies in the English literature.

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Introduction

Arachnoid cysts (ACs) constitute 1% of intracranial spaceoccupying lesions and are most commonly found in middle cranial fossa [1]. Most ACs are asymptomatic indolent lesions, and their prevalence has increased in the last decades as a result of frequent brain imaging in routine clinical practice. Symptoms are usually related to an increased intracranial pressure, size, or location of the ACs. Although most ACs remain stable in a life period, complete disappearance, intracystic hemorrhage, enlargement, and rupture causing subdural hematoma

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Fig. 1 – Hyperdense extra-axial cyst in the right anterior middle cranial fossa consistent with a blood-filled arachnoid cyst is seen on initial CT scan at admission (A). A companion right parietal SDH isodense to adjacent brain parenchyma with mild mass effect on the right hemicerebrum was also evident (B). Both the arachnoid cyst and ipsilateral SDH demonstrate high signal intensity on T1W MRI (C and D). CT, computed tomography; MRI, magnetic resonance imaging; SDH, subdural hematoma.

(SDH) may occur spontaneously over their natural course or after exertion, physical activity, or a traumatic insult [1–30].

Studies related to intracystic hemorrhage of ACs with or without associated SDH are scarce and mostly limited to case reports [2–6]. In this article, we presented and discussed the clinical, radiologic, and follow-up findings in two cases of intracranial AC with spontaneous intracystic hemorrhage and associated SDH. In addition, we summarized and discussed relevant cases in the literature.

Case presentations

Case 1

A 36-year-old male teacher with 1-week history of increasing headaches accompanied by vomiting and decreased visual acuity was admitted to our emergency department. The patient denied any history of trauma before his admission. The Glasgow Coma Score (GCS) was 15/15 upon arrival and strength of the muscles was scored as 5/5. A laboratory workup was unremarkable with no evidence of coagulopathy. A head computed tomography (CT) performed upon admission revealed a round extra-axial mass lesion in the right middle cranial fossa causing mild scalloping of the overlying bone, consistent with AC. The lesion was iso to slightly hyperdense to the cerebral cortex (Fig. 1A). There was an associated SDH layering over the ipsilateral parietal lobe that was isodense to the brain cortex (Fig. 1B). The AC and SDH both caused a mass effect against the brain parenchyma. On magnetic resonance imaging (MRI), both lesions showed high signal intensity on T1WI (Fig. 1C & D) and fluid-attenuated inversion recovery (FLAIR), and iso to hypointensity to cerebral cortex on T2WI. On gadoliniumenhanced brain MRI, the cystic lesion demonstrated no enhancement, in keeping with hemorrhagic cyst. A CT angiography showed no vascular abnormality, precluding any vascular etiology of the hemorrhage. The patient underwent surgical evacuation of SDH achieved after right craniotomy. Dark blood-filled cystic lesion with yellowish wall mouthed into sylvian fissure was noted at surgery, during which blood clot was evacuated and the underlying AC was decompressed and fenestrated. The cystic structure was proved to be AC at histopathologic examination of the cyst wall. A year after surgery, the patient's clinical condition and MRI findings were stable.



Fig. 2 – CT image performed about 2 months before the hemorrhagic event shows a left-sided arachnoid cyst extending to the sylvian fissure with remodeling of adjacent inner skull table (A and B). Intracystic hemorrhage confined to the cyst border (C). A slightly hyperdense ipsilateral subdural hematoma is also evident (D). CT, computed tomography.

Case 2

A 21-year-old nonsmoker male college student with a previously known AC in the left sylvian fissure was admitted to our emergency department for severe headache. He denied any traumatic event immediately before his admission. During a regular daytime while not performing any physical activity, he experienced a severe headache following "a strange burning sensation" at left half of his head. His GCS was 15/15 upon admission to our emergency room. Although he described a power deficient in his left upper extremities during transport to hospital, his muscle strength was 5/5 upon arrival, and there was no additional neurologic deficit by then. A laboratory workup was unremarkable with no evidence of coagulopathy. A head CT performed at admission revealed a hyperdense cystic lesion along the left middle cranial fossa, causing scalloping of the overlying bone (Fig. 2A and 2B). There was an associated hyperdense SDH layering over the left ipsilateral cerebral hemisphere (Fig. 2C). As compared to the previous head CT performed about two months before admission, the cystic lesion was unchanged in size (Fig. 2D). However, its internal density was increased indicating intracystic hemorrhage. Additionaly, there was an ipsilateral SDH with slightly hyperdense appearance compared to adjacent brain parenchyma. A CT angiography showed no vascular abnormality, precluding any vascular etiology of the hemorrhage. The patient's condition improved after appropriate analgesic and supportive therapy. Conservative treatment was opted due to unwillingness of the patient and his immediate family to accept potential risks of surgical intervention. His physical examination and laboratory studies including complete blood count were otherwise unremarkable. Cautious of potential recurrence risks, the patient was discharged from hospital upon the second week of initial admission in good clinical condition. The patient was examined at first, third, and sixth month of the incident, and his clinical condition remains stable.

Discussion

ACs constitute approximately 1% of all intracranial spaceoccupying lesions, and are histologically defined as accumulation of clear cerebrospinal fluid between the two thin layers of arachnoid mater [7]. Most ACs are asymptomatic, and they often go clinically unnoticed. However, more asymptomatic cysts are incidentally discovered these days in daily clinical practice, as a result of widespread performance of crosssectional imaging studies [1,8–11]. Rarely, ACs can dissipate after a head trauma or spontaneously vanish during the follow-up in asymptomatic cases [10]. The most common location of ACs is middle cranial fossa followed by retrocerebellar and convexity locations [8]. Middle cranial fossa ACs are more frequently seen in the vicinity of sylvian fissure where they cause scalloping and thinning of the overlying bone. These findings were also noted in the two cases we report in the present article.

| Table 1 – Review of the English literature. | | | | | | | | | |
|--|----------------------|--------------------|---------------|---|---|---|--|-----------------------------------|---|
| Authors' name. | Years of publication | Number of patients | Age/sex | Trauma history | Symptoms at admission | Location of the AC in the brain | Location and laterality of SDH with respect to AC | Intracystic hemorrhage | Clinical management |
| Hall et al. [12] | 2016 | 1 | 34/M | Spontaneous | Progressive Headache, vomiting | Right temporal | Right/ipsilateral | None | Surgery |
| Rashid et al. [24] | 2016 | 1 | 2 mo/M | Mild trauma | Episodic headache, photophobia, paresthesias, agitation | Right temporal | Bilateral SDH | None | Surgery |
| Pascoe et al. [25]. | 2014 | 1 | 34/M | Sports injury | Headache, diplopia, conscious loss | Left middle cranial fossa | A miniscule right SDH; mainly left SDH | None | Surgery |
| Shrestha et al. [7] | 2014 | 4 | 21/M 15/M | 3 Spontaneous, 1 unknown | Headache Dizziness vomiting | Left frontotemporal (n = 3) Right frontotemporal | Ipsilateral in all cases | None | Surgery |
| | | | 16/F 5/F | | | (n = 1) | | | |
| Liu et al. [3] | 2014 | 3 | 10/M | Minor trauma (n = 1) Spontaneous (n = 3) | Headache, dizziness, hallucination, vomiting, seizure, and tinnitus | Right middle cranial fossa (n = 2) | Ipsilateral in all | Present in one of the cases | Surgery (n = 3) |
| | | | 5/F | | , | Left convexity (n = 1) | Three cases | | Follow-up then surgery (n = 1) |
| Henriques et al. [17] | 2007 | 1 | 2/M 10/M | Spontaneous | Headache, ocular deviation | Left middle cranial fossa | Contralateral | None | Conservative |
| Krishnan and Kartikueyan [26] | 2013 | 1 | 15/M | Minor trauma | Headache, vomiting. | Right middle cranial fossa | Ipsilateral | None | Surgery |
| Patel et al. [16] | 2013 | 1 | 9/M | Spontaneous | Headache, vomiting. | Bilateral temporal | Right SDH | None | Surgery |
| Kwak et al. [4] | 2013 | 9 | 8 to 61 years | Not detailed | Headache, dizziness, | Mainly middle cranial | Ipsilateral. | Present | Surgery |
| | | | | | and vomiting | fossa | | (n = 3) | |
| Seddighi et al. [11] Takayasu et al. [20] | 2012 2012 | 1 2 | 23/M 8/M | Moderate trauma Mild to moderate | Headache and vomiting Headache, nausea, | Left middle cranial fossa Left middle cranial fossa | Ipsilateral Ipsilateral | None None | Surgery Surgery (n = 1) |
| | | | | trauma (n = 2) | fatigue, and amnesia | | | | |
| | | | 3/M | | - | | | | Conservative FU then surgery (n = 1) |
| Bilginer et al. [27] | 2009 | 3 | 15/M | Mild to Moderate trauma | Headache, vomiting, double vision | Left middle cranial fossa | Ipsilateral | None | Surgery |
| | | | 28/F 12/M | | | | | | |
| Iaconetta et al. [5] | 2006 | 1 | 13/M | Spontaneous | Headache, vomiting | Left middle cranial fossa | Ipsilateral | Present | Surgery |
| Maeda et al. [6] | 1993 | 1 | 14/M | Physical activity, no | Severe headache | Left temporal fossa | Ipsilateral | Present | Surgery |
| | | - | -, | trauma. | | r titir r tobba | 1 | | |
| Mori et al. [29] | 2002 | 12 | 5 to 71 years | Trauma (n = 8), | Most frequently | Middle cranial fossa | Ipsilateral | Present | Surgery |
| | | | | surgery (n = 4) | headache and vomiting | (n = 8), convexity $(n = 2)$, posterior fossa $(n = 2)$ | | | |
| Ibarra and Kesava [30] | 2000 | 1 | 11/M | Spontaneous | Headache, nausea, and vision problems | Left middle cranial fossa | Ipsilateral | Present | Surgery |

AC, arachnoid cyst; FU, follow-up; mo, month old; SDH, subdural hematoma.

SDHs are among the common observations in neurosurgery practice, and their prevalence is reported to be 13-14 cases per 100,000 subjects, with a high prevalence of unnoticed incidents [12]. The majority of SDHs occur after trauma-induced rupture of the bridging veins [13]. Less than 5% of SDHs may occur spontaneously [14]. In the two cases that we report here, there was no history of significant head trauma, and the hemorrhagic event requiring emergency hospital admission occurred in rest state without any physical activity. Vascular malformations, aneurysms, and fistulae formations may underlie a small percentage of spontaneous SDHs [15]. Any of these potential etiologies were excluded by angiographic studies in both cases we reported here.

ACs are reported to be symptomatic in 5% of cases [8]. Symptoms could be related to the mass effect from the larger cysts or an increase in intracranial pressure either resulting from intracystic fluid communication with the subarachnoid space due to a ball-valve mechanism or secretory activity of arachnoid cells within the cyst lumen [2]. Complications such as intracystic hemorrhage or cystic rupture are very rarely seen in ACs and could be preceded by a minor head trauma, more likely in larger cysts [9]. However, spontaneous intracystic hemorrhage with or without associated SDH as well as ACs with no intracystic hemorrhage but with associated SDHs or chronic subdural hygromas have been previously reported [2,9,12,16]. Although in most of reported cases the AC and the associated SDH were ipsilateral, a case of SDH contralateral to the AC has also been reported [17]. To better demonstrate the current state of knowledge regarding such cases, we conducted a review of "Pubmed" and "Medline" using the terms "arachnoid cyst," "subdural hematoma," and "arachnoid cyst AND subdural hematoma" using English language filter. We also used the "similar articles" and "studies citing these articles" functions of Pubmed to expand our review. We then included studies reporting AC-induced acute or subacute SDH or intracystic hemorrhage. We tabulated the main findings of included studies and demographics of the subjects (Table 1). According to this review, all reported cases experienced headache. However, other symptoms, including, but not limited to vomiting, decreased visual acuity, amenorrhea, rapid head growth, precocious puberty, and developmental delay have also been reported [2,18,19]. Concordant with demographics of the cases we reported here, Parsch et al. reported a higher risk of hemorrhage in young male subjects with middle cranial fossa ACs in their retrospective study conducted on 16 cases of ACs complicated with SDH [10]. This finding was also evident in the studies we included in our review of the literature, a composition of 30 cases in total (Table 1). Corollary to this finding, all reported cases of intracystic hemorrhage that were included in our literature review also occurred in young men. Nevertheless, a conclusion merely based on these observational findings is beyond the scope of this article. Although surgical evacuation of the SDH along with neuro-endoscopic fenestration, shunting, and microsurgical treatment were the treatment of choice and were performed in the vast majority of cases as noticed in 29 out of 30 cases found in the literature (Table 1), conservative follow-up has also been described in extremely rare cases [1,10,17]. One of our cases is being conservatively followed up and has been doing well at sixth month from the hemorrhagic event. According to our review of the

literature, successful conservative treatment has been only fully reported in 1 case [17]. Along with aforementioned report, our findings may raise the chance of conservative treatment consideration in certain clinical scenarios of amenable subjects. However, more studies are needed to reach a conclusion and prove scientific merit of such an approach.

In conclusion, the present report illustrates previous, initial, and follow-up findings in two patients with rare intracystic hemorrhagic complications of ACs associated with SDH, without known previous head trauma. One patient was successfully treated conservatively, whereas the other had to undergo emergency surgical intervention. More studies are needed to prove such an approach in analogous clinical conditions.

REFERENCES

- Chen Y, Fang HJ, Li ZF, Yu SY, Li CZ, Wu ZB, et al. Treatment of middle cranial fossa arachnoid cysts: a systematic review and meta-analysis. World Neurosurg 2016;92:480–90.
- [2] Ildan F, Cetinalp E, Bağdatoğlu H, Boyar B, Uzuneyüoglu Z. Arachnoid cyst with traumatic intracystic hemorrhage unassociated with subdural hematoma. Neurosurg Rev 1994;17:229–32.
- [3] Liu Z1, Xu P, Li Q, Liu H, Chen N, Xu J. Arachnoid cysts with subdural hematoma or intracystic hemorrhage in children. Pediatr Emerg Care 2014;30:345–51.
- [4] Kwak YS, Hwang SK, Park SH, Park JY. Chronic subdural hematoma associated with the middle fossa arachnoid cyst: pathogenesis and review of its management. Childs Nerv Syst 2013;29:77–82.
- [5] Iaconetta G, Esposito M, Maiuri F, Cappabianca P. Arachnoid cyst with intracystic haemorrhage and subdural haematoma: case report and literature review. Neurol Sci 2006;26:451–5.
- [6] Maeda M, Kawamura Y, Handa Y, Kubota T, Ishii Y. Value of MR imaging in middle fossa arachnoid cyst with intracystic and subdural hematoma. Eur J Radiol 1993;17:145–7.
- [7] Shrestha R, You C. Spontaneous chronic subdural hematoma associated with arachnoid cyst in children and young adults. Asian J Neurosurg 2014;9:168.
- [8] Al-Holou WN, Terman S, Kilburg C, Garton HJ, Muraszko KM, Maher CO. Prevalence and natural history of arachnoid cysts in adults: clinical article. J Neurosurg 2013;118:222–31.
- [9] Cress M, Kestle JR, Holubkov R, Riva-Cambrin J. Risk factors for pediatric arachnoid cyst rupture/hemorrhage: a casecontrol study. Neurosurgery 2013;72:716–22.
- [10] Parsch CS, Krau J, Hofmann E, Meixensberger J, Roosen K. Arachnoid cysts associated with subdural hematomas and hygromas: analysis of 16 cases, long-term follow-up, and review of the literature. Neurosurgery 1997;40:483–90.
- [11] Seddighi A, Seddighi AS, Baqdashti HR. Asymptomatic presentation of huge extradural hematoma in a patient with arachnoid cyst. Br J Neurosurg 2012;26:917–8.
- [12] Hall A, White MA, Myles L. Spontaneous subdural haemorrhage from an arachnoid cyst: a case report. Br J Neurosurg 2016;8:1–4.
- [13] Miller JD, Nader R. Acute subdural hematoma from bridging vein rupture: a potential mechanism for growth: clinical article. J Neurosurg 2014;120:1378–84.
- [14] Avis SP. Nontraumatic acute subdural hematoma: a case report and review of the literature. Am J Forensic Med Pathol 1993;14:130–4.
- [15] Barton E, Tudor J. Subdural haematoma in association with intracranial aneurysm. Neuroradiology 1982;23: 157–60.

- [16] Patel RA, Levy ML, Crawford JR. Spontaneous subdural haemorrhage in a child with bilateral middle cranial fossa arachnoid cysts. BMJ Case Rep 2013;28:bcr2013200802.
- [17] Henriques JG, PianettiFilho G, Henriques KS, Fonseca LF, Melo RP, Silva MC, et al. Spontaneous acute subdural hematoma contralateral to an arachnoid cyst. Arq Neuropsiquiatr 2007;65:1034–6.
- [18] Wang Y, Wang F, Yu M, Wang W. Clinical and radiological outcomes of surgical treatment for symptomatic arachnoid cysts in adults. J Clin Neurosci 2015;30:1456–61.
- [19] Greenfield JP, Souweidane MM. Endoscopic management of intracranial cysts. Neurosurg Focus 2005;19:1–9.
- [20] Takayasu T, Harada K, Nishimura S, Onda J, Nishi T, Takagaki H. Chronic subdural hematoma associated with arachnoid cyst. Two case histories with pathological observations. Neurol Med Chir (Tokyo) 2012;52:113–7.
- [21] Sonnet MH, Joud A, Marchal JC, Klein O. Suprasellar arachnoid cyst after subdural haemorrhage in an infant. A case based update. Neurochirurgie 2014;60:55–8.
- [22] Kawanishi A, Nakayama M, Kadota K. Heading injury precipitating subdural hematoma associated with arachnoid cysts. Neurol Med Chir (Tokyo) 1999;39:231–3.
- [23] Page AC, Mohan D, Paxton RM. Arachnoid cysts of the middle fossa predispose to subdural haematoma

formation: fact or fiction? Acta Neurochir Suppl (Wien) 1988;42:210-5.

- [24] Rashid S, Watson C, Agarwal R. Episodic headache and arachnoid cyst related subdural hematoma. Headache 2016;56:1354–5.
- [25] Pascoe HM, Phal PM, King JA. Progressive post traumatic tearing of an arachnoid cyst membrane resulting in intracystic and subdural haemorrhage. J Clin Neurosci 2015;22:897–9.
- [26] Krishnan P, Kartikueyan R. Arachnoid cyst with ipsilateral subdural hematoma in an adolescent—causative or coincidental: case report and review of literature. J Pediatr Neurosci 2013;8:177–9.
- [27] Bilginer B, Onal MB, Oguz KK, Akalan N. Arachnoid cyst associated with subdural hematoma: report of three cases and review of the literature. Childs Nerv Syst 2009;25:119–24.
- [28] Ochi M, Morikawa M, Ogino A, Nagaoki K, Hayashi K. Supratentorial arachnoid cyst and associated subdural hematoma: neuroradiologic studies. Eur Radiol 1996;6:640–4.
- [29] Mori K, Yamamoto T, Horinaka N, Maeda M. Arachnoid cyst is a risk factor for chronic subdural hematoma in juveniles: twelve cases of chronic subdural hematoma associated with arachnoid cyst. J Neurotrauma 2002;19:1017–27.
- [30] Ibarra R, Kesava PP. Role of MR imaging in the diagnosis of complicated arachnoid cyst. Pediatr Radiol 2000;30:329–31.