DOI: 10.5137/1019-5149.JTN.20885-17.2
Received: 20.06.2017 / Accepted: 20.09.2017
Published Online: 31.10.2017

Turk Neurosurg 29(6):940-944, 2019

Case Report

Spontaneous Intracystic Haemorrhage of an Arachnoid Cyst Associated with a Subacute Subdural Haematoma: A Case Report and Literature Review

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ABSTRACT

Arachnoid cysts (ACs) are congenital malformations that may develop anywhere in the subarachnoid space along the cerebrospinal axis, but are mostly observed in the temporal fossa and Sylvian fissure, predominantly on the left side. ACs account for 1% of all intracranial space-occupying lesions. ACs are potential risk factors for subdural haematoma in all age groups following a traumatic head injury. Although an intracystic haemorrhage of AC without evidence of a head trauma is very rare, it may particularly develop in children and young adults who spend much more time engaged in games and forced physical exercises. Here we present a rare case of spontaneous intracystic haemorrhage of AC with a subacute subdural haematoma and provide a review of the literature.

KEYWORDS: Arachnoid cyst, Spontaneous intracystic haemorrhage, Subacute subdural haematoma

INTRODUCTION

Arachnoid cysts (ACs) are extra-parenchymal and intra-arachnoidal cerebrospinal fluid (CSF) collections that do not communicate with the ventricular system (5,13,17). ACs are mostly observed in the temporal fossa and Sylvian fissure and account for 1% of all intracranial space-occupying lesions (22). They are usually asymptomatic, but may present with headache, nausea, vomiting, seizure and mass effects because of cyst enlargement and haemorrhage.

Because of the wide application of computed tomography (CT) and magnetic resonance imaging (MRI), there has been a relative increase in the diagnosis of asymptomatic ACs in recent years (22).

Head trauma is assumed to be one of the most important risk factors in the development of intracystic haemorrhage of AC and accompanying subdural haematoma (4,8,10). Less than 30 cases of spontaneous intracystic haemorrhage have been reported in the literature (10,13). We present a case of a 15-year-old patient who had spontaneous intracystic haemorrhage of AC with a subdural haematoma and review the relevant literature.

CASE REPORT

A 15-year-old boy presented with a 2-week history of recurrent left frontal-temporal headache and nausea episodes to the emergency room. There was no history of head trauma. He was conscious and oriented with a Glasgow Coma Scale score of 15/15. The neurological examination and laboratory test results were normal.

CT revealed a left frontal-temporal subacute subdural haematoma (SASDH) with a significant midline shift, which was accompanied by a Sylvian fissure-localised AC with an intracystic hyperdense image that suggested a haematoma (Figure 1). The patient underwent a large burr hole drainage of SASDH. During the surgery, it was observed that SASDH separated from the haematoma in the AC by the cyst membrane. Following SASDH drainage, the AC membrane was fenestrated, and intracystic blood clots were evacuated with CSF.

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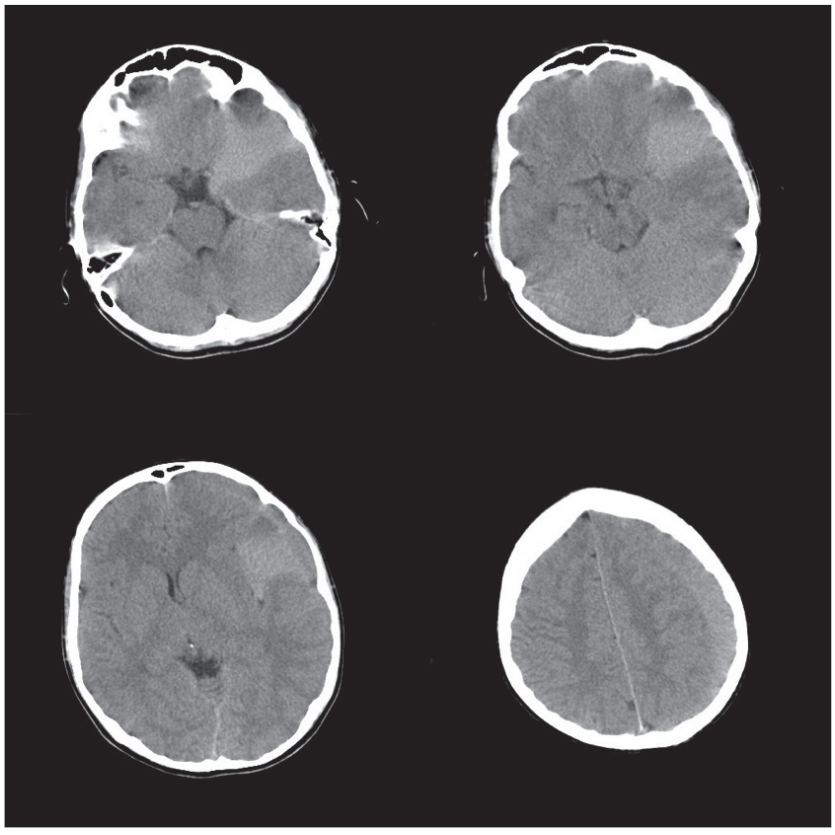

Figure 1: Preoperative CT scan images showing left temporal Sylvian intracystic haematoma of an arachnoid cyst and accompanying subacute subdural haematoma.

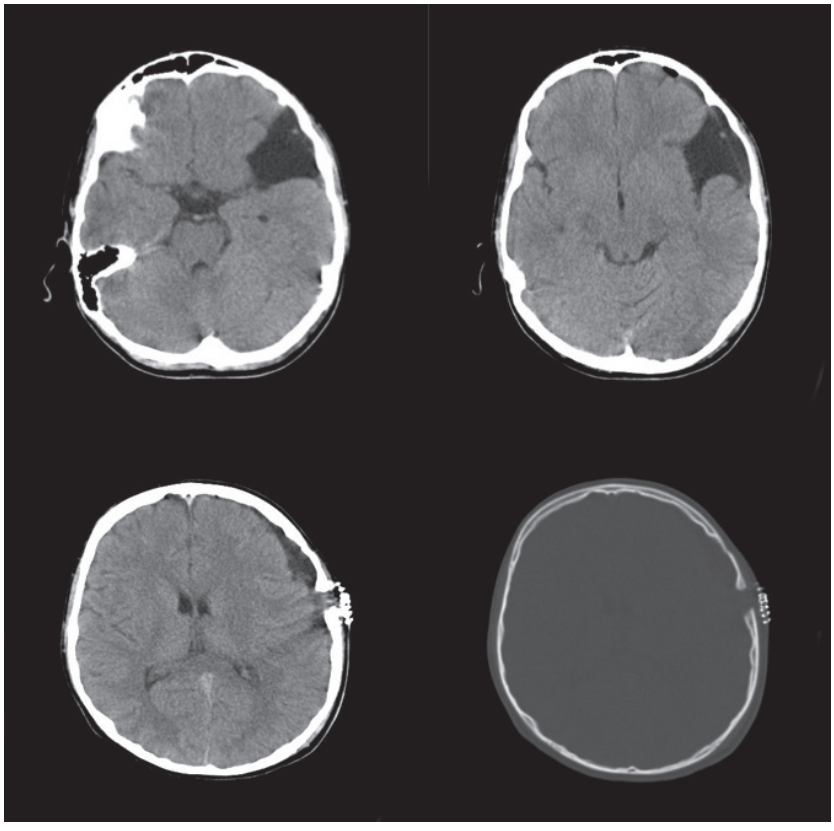

Figure 2: Postoperative CT scan images of the existing left temporal Sylvian arachnoid cyst with the resolution of the subacute subdural haematoma and the intracystic haematoma.

The postoperative period was uneventful, and the patient completely recovered. Follow-up CT (Figure 2) and MRI (Figure 3) revealed the resolution of SASDH and the intracystic haematoma.

■ DISCUSSION

Two types of congenital supratentorial AC have been described. The more common type is a dilated and circumscribed cyst. In this type, the adjacent subarachnoid space is lined by the arachnoid membrane and external arachnoid cells on the external surface, whereas the inner surface is usually lined by the pia and internal arachnoid cells (22). Starkman et al. described the less common type of AC as a concept of an intra-arachnoid cyst, which is considered to be formed by splitting and duplication of the arachnoid membrane (24). Robinson defined AC to be related to a primary embryological malformation of the meninges that resulted in the compression of the underlying temporal lobe instead of spontaneous temporal agenesis (20). Similarly, in a post-mortem study of two brains with temporal AC, Shaw revealed that there was no volume or weight difference between the left and right sides of the brains (22).

AC-related symptoms mainly appear because of cyst enlargement, which is mainly described by a ball valve mechanism. Several authors defined this mechanism as the diffusion of fluid into the cyst because of the osmotic gradient between the cyst and adjacent subarachnoid space, and they also revealed the role of fluid-secreting cells that lined the inner cyst wall during enlargement (6,19).

Davidoff and Dyke were the first to report a case of intracystic haemorrhage of AC associated with a subdural haematoma in 1938 (1). In 2006, Iaconetta et al. published a literature review regarding AC together with an intracystic haemorrhage and subdural haematoma (10). Among over 37 cases, 23 had no history of head trauma. Since then, just two more cases of such a combination have been reported, one in 2008 by Hong et al. and in 2013 by Kahilogullari et al. (8,11).

An intracystic haemorrhage and subdural haematoma may occur spontaneously or after a head trauma as a complication of AC. Several authors have described this mechanism on the basis of the rupture of intracystic or bridging vessels, particularly veins (4,10). The presence of unsupported veins that surround the cyst wall and a lower compliance of the cyst wall compared with that in normal brain tissue may be responsible for this rupture (18). Furthermore, the fragile, supporting stroma predisposes one to rupture and bleeding even after a minor injury (4).

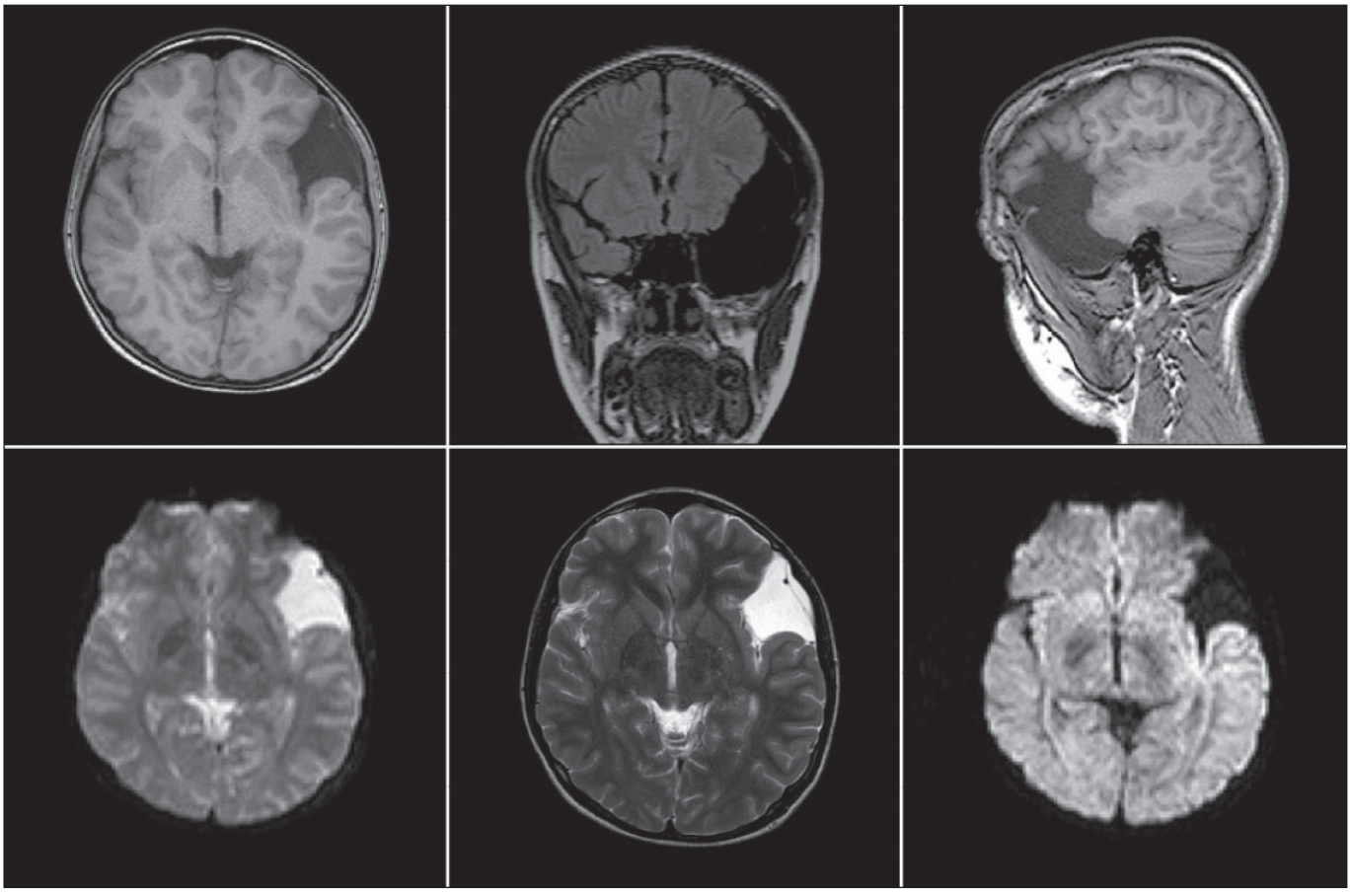

Figure 3: Postoperative MRI of the existing left temporal Sylvian arachnoid cyst with the resolution of the subacute subdural haematoma and the intracystic haematoma.

Table I: Review of the Treatment of SDH Accompanying with AC in the Literature

	Author	Age(years)/Gender of Patient	Treatment
1	Domenicucci et al., (2)	7/M, 21/M, 28/F	Burr hole drainage
2	Endo et al., (3)	22/F, 17/M, 16/M 16/F, 7/F	Cystectomy, Craniotomy Burr hole
3	Hara et al., (7)	13/M	Cystectomy, Craniotomy
4	Yamakawa et al., (26)	3/M	Cystectomy, Craniotomy
5	Kushida et al., (12)	19/M, 17/M	Cystectomy, Craniotomy
6	Maeda et al., (14)	14/M	Cystectomy, Craniotomy
7	Oka et al., (16)	17/M, 24/M	Burr hole drainage
8	Page et al., (18)	15/M, 17/M 11/M, 23/M, 17/F, 12/F	Burr hole drainage Cystectomy, Craniotomy
9	Takayasu et al., (25)	8/M 3/M	Burr hole drainage Small craniotomy
10	Sakai et al., (21)	6/F 15/M	Cystectomy, Craniotomy None

Takayasu et al. emphasised that the pathological findings in a case of chronic subdural haematoma associated with AC comprised a typical outer membrane with tissue granulation and macrocapillaries owing to inflammatory infiltration (25).

Particularly in children and young adult patients, in the absence of a head trauma, spontaneous tearing of the AC wall during games and forced physical exercises leads to the leaking of CSF and blood into the subdural space (15,23). The amount of subdural accumulation increases over time, changing the osmotic gradient owing to the effect of subdural degradation products and fenestrated immature vessels in the subdural outer membrane (9).

In the presence of progressive symptoms such as seizure, motor deficits, or severe headache that is unresponsive to medical therapy, several authors suggest that burr hole drainage of the subdural haematoma should be the first choice of treatment (Table I) (2,3,7,12-14,16,18,21,25). Cystectomy via a craniotomy is the preferred treatment in cases of recurrence after evaluating the burr hole (10).

A subdural haematoma is a rare but an important complication of AC that is mostly observed in children and young adult patients, with a male predominance (23). Because ACs generally do not have specific symptoms, neurological examination, CT and MRI are helpful in the differential diagnosis of patients who present with headache, nausea, vomiting and dizziness following forced physical exercise, even without a history of a head trauma.

The most widely accepted treatment choices for subdural haematoma that company AC are burr hole evacuation and craniotomy with cystectomy in descending order. There was no significant difference between the outcomes of these treatments according to the literature.

■ CONCLUSION

Assuming the possibility of the development of SDH as a complication of AC with or without a head trauma, incidentally diagnosed AC patients should be further evaluated with clinical follow-ups and periodical radiological assessments with a cranial MRI on an annual basis, even if the patients have no complaints. In addition, children and young adults with AC should be informed regarding the potential risks of contact sports and forced physical exercises.

■ ACKNOWLEDGEMENT

Preparation for publication of this article is partly supported by Turkish Neurosurgical Society.

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