



Case Report

Spontaneous chronic subdural hematoma associated with arachnoid cyst in a child: A case report and critical review of the literature

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ABSTRACT

Background: Arachnoid cysts (ACs) are benign, congenital, fluid-filled collection between two layers of the arachnoid membrane accounting for about 1% of all the intracranial space occupying lesions. These lesions are usually asymptomatic and detected incidentally by magnetic resonance imaging (MRI) or computed tomography scan imaging (CT). However, these lesions can present as spontaneous chronic subdural hematoma (CSDH) causing neurological deficits that require neurosurgical intervention.

Case Description: We report a case of CSDH associated with AC in a 14-year-old Kuwaiti boy who presented with a 2 weeks history of headache, which was worsening over the time. Brain CT scan demonstrated a left frontotemporal large CSDH in contact with an underlying temporal AC that appeared isodense to the CSF. The patient underwent an emergency surgery to evacuate the CSDH through a burr hole, while the AC was left intact. During the postoperative period, the patient showed good recovery in terms of neurological symptoms. Follow-up MRI showed stable size of the AC with no recurrence of the CSDH.

Conclusion: Literature's review was done to determine the best surgical approach in treating spontaneous CSDH associated with AC. Burr hole evacuation of CSDH with irrigation only, leaving the AC intact, is a successful surgical approach for treatment and was associated with good outcome.

Keywords: Arachnoid cysts, Burr hole, Craniotomy, Rupture, Spontaneous chronic subdural hematoma

INTRODUCTION

Arachnoid cysts (ACs) are benign, congenital, fluid-filled compartments between two layers of the arachnoid membrane, comprising 1% of all the intracranial space-occupying lesions.^[3] In the majority of cases, ACs are asymptomatic and have been increasingly detected as an incidental finding with the advent of computed tomography and magnetic resonance imaging (MRI).^[4,18,49] They are sporadically recognized as a cause of chronic subdural hematoma (CSDH) after minor head injury in young people. However, there are several controversies regarding the incidence of ACs in association with CSDH, how does an AC favor the development of CSDH, and what is the best surgical approach to treat patients with a CSDH in association with an AC.

We report a case of AC in association with a spontaneous CSDH successfully treated by burr holes and irrigation. We also reviewed the available literature to recommend a surgical approach

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based on the proposed underlying pathophysiology and the outcome of the different surgical approaches as reported.

CASE REPORT

A 14-year-old Kuwaiti boy presented with 2 weeks history of headache, which was worsening over the time. There was no history of head trauma and physical examination was unremarkable for neurological deficits. Brain computed tomography (CT) scan demonstrated a left frontotemporal large CSDH [Figure 1] in contact with an underlying Type 2 Galassi temporal AC [Figure 2]. The patient underwent an emergency surgery to evacuate the CSDH through a burr hole while the AC was left intact. During the postoperative period, the patient's conditions gradually improved and ultimately his headache disappeared. The immediate postoperative CT head showed good evacuation of the CSDH [Figure 3]. One month after surgery, the left temporal CSDH was diminished significantly in size and the patient was asymptomatic. On postoperative MR imaging, the AC appeared as a markedly hyperintense area on T1- and T2-weighted MR images and was unchanged in size as compared to preoperative images [Figure 4]. A 2-year follow-up with MRI brain showed no change in the size of the AC nor recurrence of the CSDH.

DISCUSSION

The ACs are most often found in children with a male-to-female ratio of 3:1 and involve the middle cranial fossa in 50% of cases with a slight predilection for the left side.^[62,65] Although mostly asymptomatic, ACs can present in the form of headache and other features of high intracranial pressure due an increase in their volume due to unidirectional flow through a ball valve opening in the wall with resultant trapping of CSF within the cyst.^[37] Another form of presentation is in association with CSDH [Table 1], with an estimated annual risk of hemorrhage within or around the cyst of <0.1%.^[44] This figure seems to be more accurate than the reported 16% incidence of CSDH in patients with middle fossa AC^[18] or that of 2.43% of patients with CSDH would have an associated AC.^[44]

The first description of an AC in association with CSDH was by Davidoff and Dyke in 1938.^[44] Reviewing the literature, we found 57 reports published during 1976–2019, with a total number of 183 cases of CSDH in association with AC with age ranging between 3 years and 58 years [Table 1].^[1,7,8,11-17,20-22,25,26,29,30,33,34,36,39,40,42,45,46,48,54-60,63,64,66-71] There were 31 single case reports, 12 publications reporting on 2–3 patients, and 14 case series reporting on 5–16 patients. There are some data to support that the coexistence of CSDH and AC is not incidental, and two pathogenic mechanisms were proposed to explain such association. First, electron microscopic studies showed that dural border cells extend from the dura and adhere tightly to the arachnoid membrane, delimiting a “potential space” – namely, the “subdural compartment.” The presence of an AC, due to an increase in its size or fluid motion within the cyst, weakens the subdural compartment at the level of the junction of the two compartments and causes bleeding with no or minimal trauma.^[15] Second, the unsupported vessels around the AC wall and abnormal veins bridging the widened Sylvian fissure are fragile and could bleed spontaneously or after a relatively minor trauma resulting in CSDH.^[10,18,35,38,43] These two proposed mechanisms should be put in consideration when approaching this disease entity.

Although there are a few published cases in which a CSDH associated with an AC was treated conservatively,^[2,6,19,44] most patients were managed surgically. There is no general agreement on the best surgical approach, craniotomy,^[5,31,32,43,51,52,61] or burr hole, and irrigation.^[15,19,23,24,38,41,44] The second pathogenic mechanism would suggest that evacuation of the CSDH alone is not sufficient, and that removal of the membrane of the AC and coagulation of the associated abnormal vessels by craniotomy are required to prevent or reduce the risk of rebleeding. However, irrigation of the CSDH through burr holes was shown to be adequate in most of the cases.^[23,24] Oka *et al.* (1994) reviewed the literature and found that out of 18 patients treated with burr hole and irrigation, only four required a craniotomy because of rebleeding or fluid retention in the subdural space or under the scalp. The other patients achieved a good outcome with only the irrigation procedure.

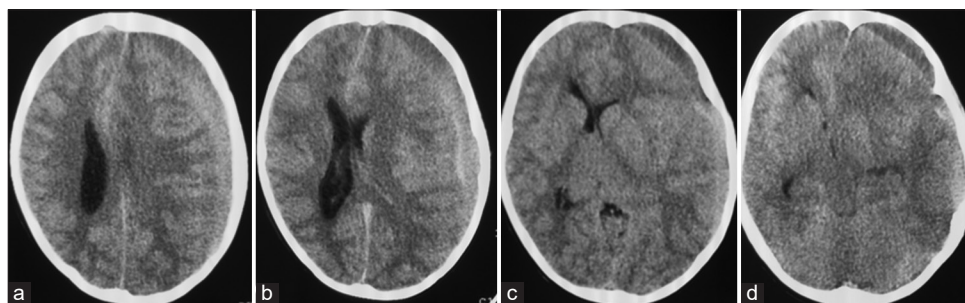


Figure 1: (a and b) Brain CT scan demonstrated a left frontotemporal large CSDH.

Table 1: The case series published in the English language literature of CSDH in association with AC.

Author (s)	Number of cases
Kim <i>et al.</i> , 2019	2
Kim <i>et al.</i> , 2019	1
Furtado <i>et al.</i> , 2019	1
Kaszuba <i>et al.</i> , 2018	1
Johnson <i>et al.</i> , 2018	1
Kode <i>et al.</i> , 2018	1
Adin <i>et al.</i> , 2017	2
Yüksel <i>et al.</i> , 2016	1
Kobayashi <i>et al.</i> , 2016	1
Chandra <i>et al.</i> , 2015	1
Patel <i>et al.</i> , 2015	1
Bora <i>et al.</i> , 2015	1
Kwak <i>et al.</i> , 2013	11
Takayasu <i>et al.</i> , 2012	2
Kertmen <i>et al.</i> , 2012	1
Zeng <i>et al.</i> , 2011	2
Wang <i>et al.</i> , 2011	11
Gelabert-González, 2010	12
Dilvesi <i>et al.</i> , 2009	1
Hong <i>et al.</i> , 2008	1
Domenicucci <i>et al.</i> , 2009	8
Ziaka <i>et al.</i> , 2008	1
Kocaeli and Korfali, 2008	1
Chan <i>et al.</i> , 2008	1
Zhang <i>et al.</i> , 2007	2
Wester and Helland, 2008	11
Leung and Fan, 2005	1
Czernicki <i>et al.</i> , 2005	3
Abderrahim <i>et al.</i> , 2004	2
Demetriades <i>et al.</i> , 2004	1
Tsuzuki <i>et al.</i> , 2003	1
Mori <i>et al.</i> , 2002	12
Ulmer <i>et al.</i> , 2002	1
Prabhu and Bailes, 2002	1
Wijdicks <i>et al.</i> , 2000	5
Parsch <i>et al.</i> , 1997	16
Cole and Lam, 1996	1
Okura <i>et al.</i> , 1994	1
Oka <i>et al.</i> , 1994	3
Servadei <i>et al.</i> , 1993	3
van Burken <i>et al.</i> , 1992	1
Kamiura and Oka, 1990	1
Endo <i>et al.</i> , 1988	7
Munk <i>et al.</i> , 1988	1
Page <i>et al.</i> , 1987	7
Saito <i>et al.</i> , 1987	3
Suzuki <i>et al.</i> , 1986	1
Takanohashi <i>et al.</i> , 1986	3
Cappelen and Unsgaard, 1986	1

(Contd...)

Table 1: (Continued).

Author (s)	Number of cases
Tajima <i>et al.</i> , 1984	6
Kusuno <i>et al.</i> , 1984	1
Kushida <i>et al.</i> , 1983	5
Kadowaki <i>et al.</i> , 1983	1
Nabeshima <i>et al.</i> , 1982	3
Auer <i>et al.</i> , 1981	5
Varma <i>et al.</i> , 1981	6
Smith and Smith, 1976	1
Total	183

Table 2: The case series published 1981–1993, where the majority of patients underwent craniotomy for evacuation of CSDH and fenestration of the AC.

	n=	Burr hole	Craniotomy	Conservative
Servadei <i>et al.</i> , 1993	3	0	3	0
Page <i>et al.</i> , 1987	7	3	4	0
Kushida <i>et al.</i> , 1983	5	0	5	0
Auer <i>et al.</i> , 1981	5	0	5	0
Varma <i>et al.</i> , 1981	6	2 (failed)	6	0
Total	26	5/3	23/21	0

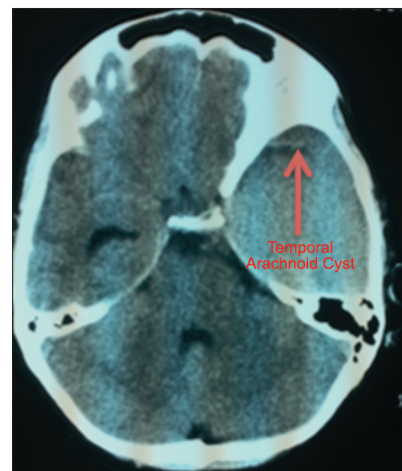


Figure 2: Brain CT scan demonstrated a left frontotemporal large CSDH in contact with an underlying temporal AC that appeared isodense to CSF (red arrow).

Our review of the literature revealed two interesting findings. First, there was a clear change in the surgical approach for treating CSDH associated with AC over time. Case series published 1981–1993 included a total of 26 patients, of which 23 (88.5%) underwent a craniotomy (21 as first option and two after failed burr hole irrigation)

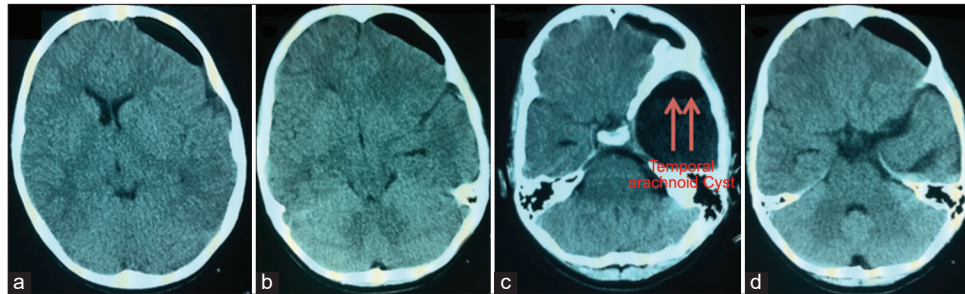


Figure 3: (a-d) The immediate postoperative CT head showed good evacuation of the CSDH.

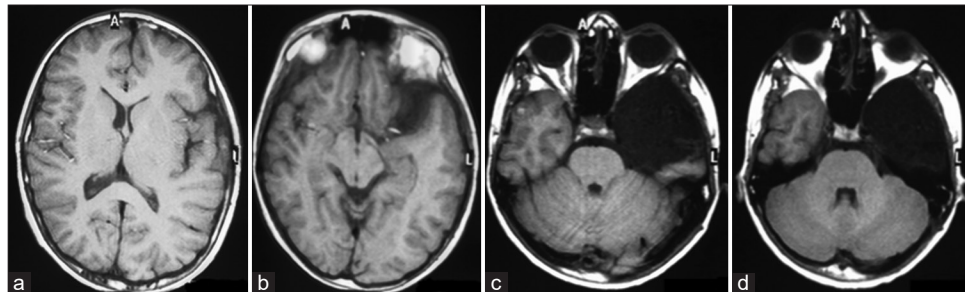


Figure 4: (a-d) Postoperative MR imaging showing unchanged size of the AC as compared to preoperative images.

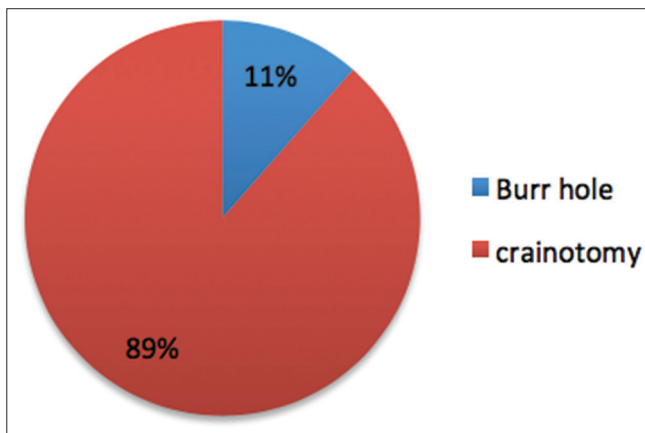


Figure 5: The case series published 1981–1993, where the majority of patients underwent craniotomy for evacuation of CSDH and fenestration of the AC.

[Table 2] [Figure 5]. On the other hand, case series published 1994–2019 included 79 patients, 71 (89.9%) were treated successfully with burr hole and irrigation and only four required a craniotomy [Table 3] [Figure 6]. Second, there was a high success rate for the burr hole and irrigation approach, where only a few required reoperations with same technique, but none of the 71 patients required a craniotomy on long-term follow-up.

Histological data from transmission electron microscopy studies indicated the presence of a membrane separating the AC from the CSDH, rendering them two separate a noncommunicating entity.^[47,50,53] Because a thin membrane

Table 3: The case series published 1994–2019, where the vast majority of patients treated with burr hole evacuation of CSDH.

	n=	Burr hole	Craniotomy	Conservative
Kim <i>et al.</i> , 2019	2	2	0	0
Adin <i>et al.</i> , 2017	2	0	1	1
Kwak <i>et al.</i> , 2013	11	11	0	0
Takayasu <i>et al.</i> , 2012	2	2	0	0
Wang <i>et al.</i> , 2011	11	9	2	0
Gelabert-González, 2010	12	11	0	1
Domenicucci <i>et al.</i> , 2009	8	8	0	0
Mori <i>et al.</i> , 2002	12	12	0	0
Parsch <i>et al.</i> , 1997	16	13	1	2
Oka <i>et al.</i> , 1994	3	3	0	0
Total	79	71	4	4

separates the two entities, the CSDH remains confined within the intradural compartment, whereas an AC remains exclusively intra-arachnoid. These anatomical and imaging features could explain the good outcome after evacuating the CSDH leaving the membrane separating it from the AC intact. Furthermore, several reports showed good surgical outcome in patients treated by CSDH drainage alone while the AC was left intact, and the postoperative course resembled that of patients with CSDH not associated with an AC.^[15,27,28,31,38,44]

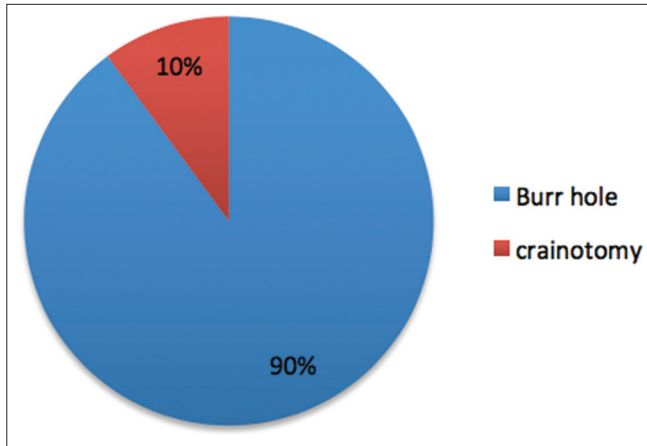


Figure 6: The case series published 1994–2019, where the vast majority of patients treated with burr hole evacuation of CSDH.

CONCLUSION

The available literature suggests that most patients presenting with CSDH and associated AC can be successfully treated with burr hole and irrigation, and only a minority would require craniotomy.

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.

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