Case Report

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A chronic subdural hematoma complicating an arachnoid cyst in a juvenile boxer: a rare case report with comprehensive literature review

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Introduction and importance: Arachnoid cyst (AC) is the most frequently founded lesion reported in adolescent patients suffering from chronic subdural haematoma (CSDH). Association between these two distinct clinical entities is known for a long time. However, in the literature there are numerous clinical cases that reflect this relationship and few large series that analyze them in detail. Paediatric population is more rarely affected with this association.

Case presentation: The authors report the case of AC of incidental discovery complicated with CSDH in 15-year-old male recreational boxer presented with progressive onset of holocranial drug-resistant throbbing headache with favourable clinical course after conservative treatment.

Clinical discussion: ACs are a well-known predisposing cause for CSDH after head trauma. In all cases of CSDH in children, the diagnosis of ruptured AC should be considered. Rupture may be spontaneous or following even mild head trauma with rupture of bridging veins causing subdural bleeding as it was seen in our patient who was practicing a full-contact free-sparring sport like boxing sustaining repeated and direct mild head traumas. MRI is recommended to detect small cysts in adolescents with CSDH. The management of these patients remains controversial.

Conclusion: This is a rare reported case of CSDH complicating an AC in a juvenile recreational male boxer. This association remains extremely rare in children and adolescents, as evidenced by the rare cases reported in the literature.

Key words: arachnoid cyst, boxer, case report, chronic subdural haematoma, head trauma

Introduction and importance

Arachnoid cyst (AC) is a benign intracranial lesion that is formed by an arachnoid membrane containing a fluid similar in its chemical characteristics to cerebrospinal fluid (CSF). It represents up to 1% of all space-occupying intracranial lesions and approximately half are located in the temporal fossa[1,2]. AC is the most frequently founded lesion reported in adolescent patients suffering from chronic subdural haematoma (CSDH). CSDH is, in brief, a haemorrhagic lesion, usually of traumatic nature, characteristic of the elderly or people with bleeding disorders[3]. Paediatric population is more rarely affected with this pathology[3,4].

Association between CSDH and AC is known for a long time; however, in the literature there are numerous clinical cases that reflect this relationship and few large series that analyze them in detail[5].

HIGHLIGHTS

- Arachnoid cyst is the most frequently founded lesion reported in adolescent patients suffering from chronic subdural haematoma.
- Rupture may be spontaneous or following even mild head trauma with rupture of bridging veins causing subdural bleeding.
- Computed tomography scan is the first tool of choice. Magnetic resonance imaging is more efficient to distinguish between haemorrhage within the arachnoid cyst and subdural haematoma.
- The management of patients remains controversial; some authors defend the exclusive treatment of subdural haematoma (burr hole irrigation and drainage) based on the fact that after evacuation, many cysts tend to remit spontaneously, especially if the arachnoid cyst was asymptomatic.

Here, and to the best of our knowledge, we report, through a comprehensive review of the relevant literature, the case of AC complicated with CSDH in 15-year-old male recreational boxer with favourable clinical course after conservative treatment.

This case report has been reported in line with the SCARE Criteria[6].

Case presentation

A 16-year-old male adolescent with no particular medical or surgical history was admitted to our neurosurgical

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department with the complaint of progressive onset of holocranial drug-resistant throbbing headache for about 2 months, without neither vomiting nor epileptic seizures or blurred vision. In fact, the patient was a recreational boxer in a non-professional martial arts school sustaining repeated and direct mild head traumas. On admission, the boy was alert and well oriented; physical examination showed normal pupil reactivity with no positive neurological signs. He was also free from any motor or sensory neurological deficit Fundus examination did not find any papillary oedema. A Non-enhanced computed tomography (CT) head revealed a right frontal-parietal-temporal high density subdural haematoma with 9 mm of thickness exerting a midline shift estimated at 6 mm (Fig. 1). As part of the search for any aetiology, a brain CT angiography was performed and did not find any cerebrovascular malformations including arteriovenous malformation or dural arteriovenous fistula (Fig. 2).

The patient' parents were informed about his diagnosis and surgery through a burr hole trepanation and irrigation was thus suggested, however, it was completely refused. Therefore the patient was put under clinical follow-up. Further radiological investigations with standard MRI associated to a full study MR angiography confirmed the right collection with an isosignal on T1 weighted-sequence compared to grey matter and T1 hypersignal compared to cerebrospinal fluid. Both T2 and fluid attenuated inversion recovery sequences showed hypersignal without restricted signal on diffusion weighted image. This collection appears to be in continuity with an ipsilateral temporopolar Galassi I arachnoid cyst having the same signal as the above haematoma and measuring 37 $\times$26 mm in diameter. It is associated with haemorrhagic sediment in discreet hypersignal on T1 weighted-sequence and hyposignal on both T2 and fluid attenuated inversion recovery weighted images. The three-dimensional time of flight angiogram showed no abnormalities (Fig. 3). Complete blood count, haemostasis assessment, and serum electrolytes test were all in normal range.

Regarding the stability of patient's neurological condition as well as the significant improvement of headache, he was discharged from our department under enteral analgesics (1000 mg of paracetamol every 6 h) and hyperhydratation with appointment at the outpatient clinic in 30 days for a clinical and radiological assessment. The patient and his parents were warned at discharge that this sporting activity is risky for their child's condition and that the risk of recurrence of bleeding and collection of a new chronic subdural haematoma is high. This is why leaving this activity seems reasonable. However, the patient was lost to follow-up.

Clinical discussion

ACs are a well-known predisposing cause for CSDH after head trauma[7]. Association between these two clinical entities was first described by J. Chan in 1971[7]. As a result, around 20 cases have been reported in the literature[8]. In all cases of CSDH in children, the diagnosis of ruptured AC should be considered.

ACs are space-occupying lesions of congenital origin that can be located at any point in the central nervous system, although in general the middle cranial fossa is the most common site, especially in the sylvian fissure[9]. These cysts are characteristic of the paediatric age where they represent 1% of all intracranial extensive lesions. Galassi[3] classified sylvian arachnoid cysts into three types: type I: small, spindle-shaped cyst limited to the anterior portion of the middle cranial fossa, below the sphenoid ridge with free communication of subarachnoid space; type II: superior extent along the Sylvian fissure with displacement of the temporal lobe and slow communication with subarachnoid space; type III: the cyst is large and fills the whole middle cranial fossa with displacement of not only the temporal lobe but also the frontal and parietal lobes and often results in midline shift with little communication with subarachnoid space.

Although many of ACs are symptoms free during childhood and may be diagnosed in adulthood, either because they

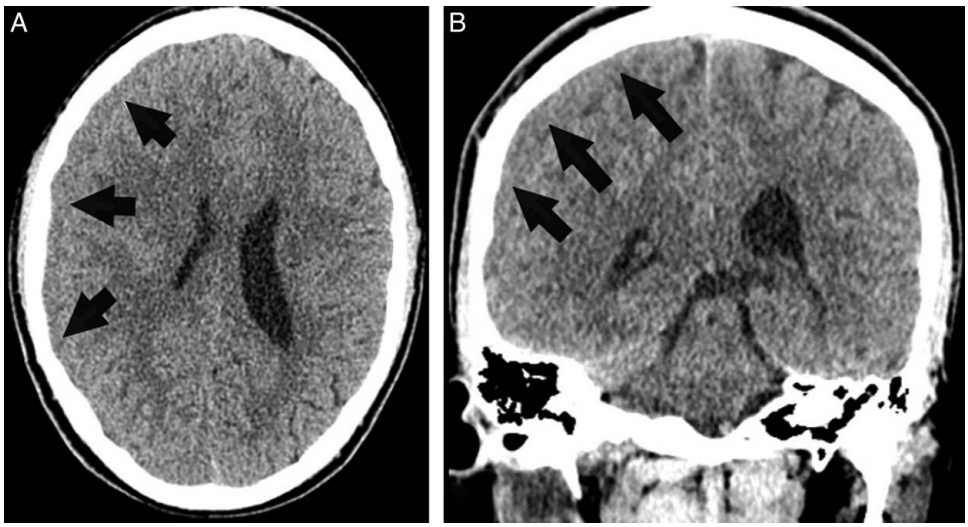

Figure 1. A non-enhanced brain computed tomography in axial (A) and coronal plane (B) and parenchymal window showing a right frontal-parietal-temporal high density subdural haematoma (black arrows) with 9 mm of thickness with a midline shift. Note the ventricular asymmetry due to the mass effect.

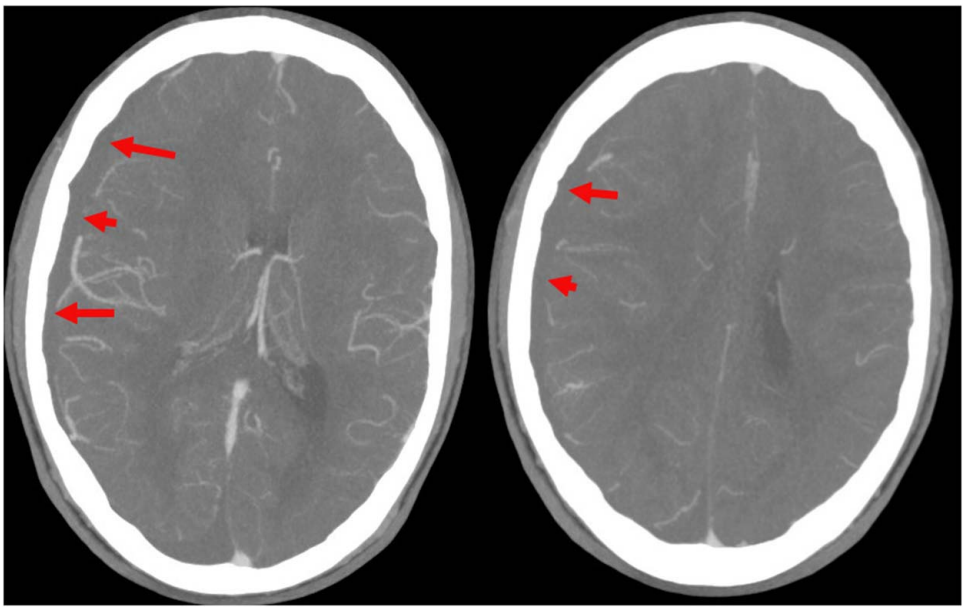

Figure 2. Axial brain computed tomography angiography in multiplanar reconstruction and maximum intensity projection showing no cerebrovascular malformations or abnormalities. Note the mass effect exerted by the haematoma on the ipsilateral cerebral vascular network (red arrows).

become symptomatic or because they are incidental findings[2] as it was seen in our patient where the cyst was classified Galassi I and it was therefore of incidental discovery. Symptomatology is directly related to the patients' age. In children, cysts usually cause skull deformities when settled in the convexity of the cerebral hemispheres or increased intracranial pressure related to obstruction in CSF flow when located in the midline[10]. In older children and adult patients, ACs may manifest in very different ways, usually related to the progress in their volume or with various associated complications such as spontaneous intracystic haemorrhage, spontaneous or traumatic rupture into the subdural space[1,11]. Rupture may be spontaneous or following even mild head trauma with rupture of bridging veins causing subdural bleeding[12,13] as it was seen in our patient who was practicing a full-contact free-sparring sport like boxing sustaining repeated and direct mild head traumas.

CSDH is a collection of blood in various stages of degradation located in the subdural space that grows slowly progressively causing symptoms. It is characteristic of the elderly population where its development is favored by senile involution of the brain, intake of drugs that alter blood coagulation, and a higher rate of accidents, usually mild, that condition the onset of a series of mechanisms that lead to haematoma formation[14]. CSDH is rarely found in children and adolescent, due to the fact that cerebral parenchyma occupies almost all of the subarachnoid spaces, unlike the elderly in whom cortical atrophy is found widening the subdural space[4,13]. The appearance of CSDH in young patients (< 50 years) should always make us think about the possibility of some factor added to its development, among which are ventricular bypass valves, spontaneous CSF fistulas[15] or an arachnoid cyst as it was seen in our case.

The relationship between CSDH and AC has been known for a long time in the neurosurgical literature, although there is no satisfactory explanation to justify this association. Mori *et al.*[15] reported that the incidence rate of both lesions was 1.5%. Page *et al.*[16] proposed two theories: (a) after a head trauma there would be changes in the CSF flow that would lead to facilitating the rupture of bridging veins or vessels in the cyst wall; (b) ACs have less compliance than brain parenchyma. As a result, and after minor trauma, the bridging veins break more easily, either ipso or contralaterally, thus producing a subdural haematoma. In both theories, it is assumed that both lesions have a neighbourhood relationship, as is the case in most of the published cases. Kwak *et al.*[17] found that small bridging veins located between the dura mater and the AC's outer wall were obviously the source of the primary CSDH bleeding.

In order to make diagnosis, CT scan is the first tool of choice. MRI is more efficient to distinguish between haemorrhage within the AC and subdural haematoma. Therefore, MRI is recommended to detect small cysts in adolescents with CSDH[18] as it was performed in our patient where the cyst was not seen on CT but discovered incidentally on MRI.

The management of these patients remains controversial; some authors defend the exclusive treatment of subdural haematoma (burr hole irrigation and drainage) based on the fact that after evacuation, many cysts tend to remit spontaneously, especially if the AC was asymptomatic[19,20]. On the contrary, there is a majority opinion of simultaneous or successive treatment of both lesions with craniotomy that allows evacuation of the haematoma and fenestration of the cyst membranes[21] or evacuation of this haematoma and insertion of a cystoperitoneal shunt[22,23]. In our case we opted for the conservative treatment given the refusal of the surgery as well as the clinical improvement of the patient. Mori *et al.*[15] operated on 12 CSDHs secondary to arachnoid cysts rupture. The outcomes reported were satisfactory with reduction in the

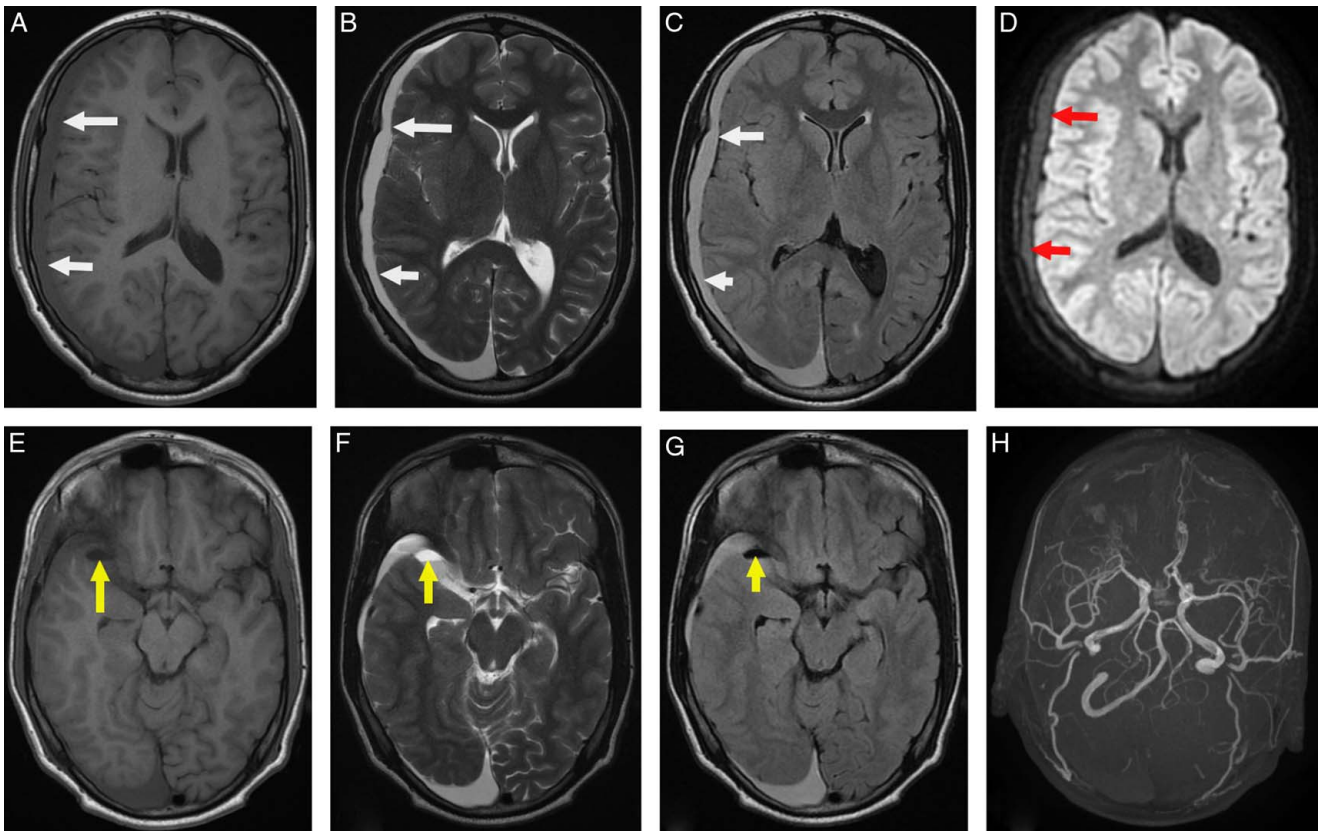

Figure 3. Brain MRI in axial plane showing a right collection with an isosignal on T1 weighted-sequence and T1 hypersignal compared to cerebrospinal fluid ((A); white arrows). On T2 ((B); white arrows) and fluid attenuated inversion recovery sequences ((C); white arrows) it showed hypersignal without restricted signal on diffusion weighted image ((D); red arrows). Note the ipsilateral temporopolar Galassi I arachnoid cyst ((E–G): yellow arrows). A three-dimensional time of flight angiogram showing no abnormalities (H).

cyst's size postoperatively. These finding was also made by Rakier and Feinsod[24], as well as Albuquerque and Giannotta[25]. Likewise, Parsch *et al.*[20] in their series of 16 cases including 12 operated patients by 2 trephinations and 2 were managed by craniectomy and membranectomy. Two patients received only simple monitoring. For these authors, fenestration of the cyst should only be performed when the cyst was symptomatic before its rupture. This was not the case in our reported case where our patient was clinically totally asymptomatic. It does not therefore seem necessary to fenestrate the cyst and thus, simple evacuation of the haematoma by trepanation is sufficient.

Conclusion

In the full-contact free-sparring sport like boxing, ACs may be a relative contraindication to practice. In children and adolescents participating in these activities, headache is sufficient to indicate neuroimaging. Once diagnosis is made, the practice must be forbidden. CSDH following the rupture of an AC remains an extremely rare pathology in children and adolescents, as evidenced by the rare observations reported in the literature. The treatment remains controversial. Evacuation of the haematoma by trepanation, with monitoring of the cyst's course is one of the admitted stategy. Surgical fenestration is thus indicated when this latter becomes symptomatic.

Ethics approval

None.

Consent for publication

A verbal consent was obtained Written informed consent for participation in the study was obtained from the patient's parents.

Consent

Written informed consent was obtained from the patient's legal guardian for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

M.B.: conceptualization, methodology, software. M.B., W.B.: data curation, writing—original draft preparation. M.B.: visualization,

supervision, writing—reviewing and editing M.Z.B.: validation. All authors have read and approved the manuscript, and ensure that this is the case.

Conflicts of interest disclosure

The authors declare that they have no competing interests.

Research registration unique identifying number (UIN)

None.

Guarantor

Mehdi Borni.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Provenance and peer review

Not commissioned, externally peer reviewed.

Patient perspective

During hospitalization and at the discharge, the patient and his legal guardian were given the opportunity to share their perspectives on the intervention the boy received and they were satisfied with the care.

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