Chronic subdural hematoma associated with sylvian arachnoid cyst in juvenile athletes: report of two cases and literature review

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Abstract
The association of chronic subdural hematoma (CSDH) and arachnoid cyst (AC) is uncommon. We reported 2 juvenile athletes with CSDH associated with AC which occurred in their daily sports activities and reviewed the literature. Both of them were treated surgically, with satisfactory outcome. AC is a common predisposing factor in young patients with CSDH. The complication of intracranial bleeding is an indication for surgical management. Though there are still controversies in the treatment of asymptomatic AC, it is the consensus that the patients with AC should avoid violent sports so as to reduce the incidence of intracranial hemorrhage resulted from head injuries.

Key words: Hematoma, subdural, chronic; Arachnoid cysts; Adolescent; Athletes

Both chronic subdural hematoma (CSDH) and arachnoid cyst (AC) are common clinical entities in daily neurosurgical practice, whereas the association of CSDH and AC is uncommon. CSDH is generally believed to occur in elderly patients, occasionally in pediatric or young patients with certain predisposing factors. AC is the most frequently detected lesion reported in young population suffering CSDH.

AC is believed to be extracerebral, intra-arachnoidal cerebrospinal fluid (CSF) collection, comprising 1% of all the intracranial mass. With easier access to modern neuroimaging, an increasing number of ACs are diagnosed. They are thought to be congenital and microscopically formed by mesothelial cells. The precise etiology is still not completely understood. They may occur at any age but often affect young population with a male-to-female ratio of 3:1. AC usually involves the middle cranial fossa with a slight predilection for the left side. The mass in the middle fossa, namely sylvian AC, represents more than half of intracranial AC, half of which are asymptomatic and detected incidentally. However, they may become symptomatic due to intracystic or subdural hemorrhage or enlargement of the cyst. Occasionally, head injuries causing subdural hematomas may be the first presentation of a previously undiagnosed AC. Here we presented 2 cases of sports-related CSDH associated with left sylvian fissure AC in juvenile athletes and discussed the pertinent data in the literature.

Case 1
A 14-year-old boy was admitted with the complaint of progressive headache for about a week, accompanied by projectile vomiting twice before his attendance to our unit. He was an athlete in an amateur sport school. Twenty-eight days before admission, he attended to outpatient department because of an incidental head injury due to an inappropriate falling posture in jump training. Although he did not regard it a serious injury, CT scan was arranged, which showed a left sylvian AC (Figures 1A, 1B), confirmed by a subsequent MRI (Figures 1C, 1D). There were no signs of intracranial subdural or intracystic bleeding. On admission, the patient was drowsy with orientation intact, and physical examination showed a mild hemiparesis in right limb. CT scan found intracystic bleed-
ing and left frontal-parietal-temporal CSDH with significant midline shift. An emergent craniectomy was then performed to evacuate the subdural hematoma and fenestration of the cyst wall was carried out. During operation, no evidence of other pathological changes was detected. Postoperative pathological examination confirmed the diagnosis of epithelial cyst. The patient recovered completely without headache or other postoperative complications. He was discharged after improvement and presented no symptoms since then.

Case 2
A 16-year-old male student who accepted regular training in a football school was admitted and he complained of increasing headache and nausea for 4 weeks, especially in the morning. He denied the history of head trauma other than frequent heading of the football in competition and training. On admission, he was in a relatively normal status without any significant neurological deficit. CT scan (Figure 2A) and subsequent MRI (Figures 2B-2E) revealed a temporal-frontal-parietal CSDH associated with a left middle fossa AC, and midline shift. At operation via a burr hole craniotomy, a large CSDH was confirmed and evacuated. Clinical recovery was satisfactory with no persistent neurological deficit. Postoperative CT scan showed the outline of a sylvian AC in the left middle cranial fossa (Figure 2F). The patient recovered rapidly without complications, and lived with good neurological condition since then.

DISCUSSION
AC is generally believed to be congenital and usually occurs in young patients whereas CSDH tends to occur in elderly people. The association between these two distinct clinical entities was first noted in 1971 and has been reported occasionally since then. Although no explanation for this predisposition has been proven, some theories have been suggested. As AC is less compliant than normal brain tissue, the effect of intracranial "cushioning" during trauma is reduced with an increased likelihood of bleeding in bridging veins, unsupported blood vessels around the cyst wall, and leptomeningeal vessels in the base of the cyst. Additionally, AC is occasionally associated with subdural effusion, which is generally thought to result from tearing of the outer wall of AC after head injury. CSF with or without blood may leak into the subdural space and then facilitates the formation of "outer membrane" under the dura mater, which forms fragile neocapillaries or sinusoids. These blood vessels in the outer membrane are fenestrated and allow plasma fluid leakage and resultant enlargement of subdural effusion. Then bleeding occurs repeatedly from the capillaries with degenerating endothelium accompanied by local hyperfibrinolysis, assembling the development of effusions into CSDH. It might promote the formation of CSDH in the patients as case 1 since there were no signs of bleeding in the...
The cases of subdural hematoma in patients with AC of the middle fossa have increased since the first report. Tamburrini et al. studied the actual incidence of intracranial subdural, epidural and intracisternal bleeding in patients with temporal AC (132 cases). The observed risk of associated subdural hematoma was 2.27% (3/132). Mori et al. reported that the rate was 1.5% in a series of 541 patients (12/541). The actual rate of cyst-related intracranial hemorrhage after head trauma caused by sports is unknown. Some sports related subdural hematomas associated with AC have been reported in contact sports, such as boxing, and non-contact sports, such as basketball, even racing. Herewith, we reported two cases of intracranial bleeding in so-called non-contact sports: jumping and football.

Except some non-specific syndromes, AC is usually asymptomatic until it has complications which may lead to subdural hematoma or intracisternal hemorrhage or subdural hygroma, which may occur spontaneously or after minor head trauma. Patients with CSDH associated with AC often presented with symptoms of increased intracranial pressure. In young patients, headache is more common because they tend to suffer increased intracranial pressure caused by CSDH. If the patients with asymptomatic AC show signs of increased intracranial pressure after head injury, CSDH should be considered. With few exceptions, CT is diagnostic. If CT diagnosis is impaired by an isodense AC due to the presence of intracisternal hematoma, MRI is the radiological investigation of choice, as it is hard to distinguish the hemorrhage within the AC from subdural hematoma. Furthermore, even small AC in the middle fossa and also the convexity may be a predisposing factor for CSDH, MRI is recommended to detect small AC in young patients with CSDH.

There are some controversies on the treatment of CSDH associated with AC. Uncertainties for the clinical management is brought about by the indefinite natural case history. It is believed that if AC is complicated with subdural hematoma and/or intracisternal hemorrhage, surgical treatment is necessary. But which is the best option? Immediate removal of the membrane of AC and evacuation of subdural fluid by craniotomy was recommended previously. Nowadays, with the recognition that most symptoms of CSDH associated with AC are due to increased intracranial pressure and the possibility of spontaneous resolution after the evacuation of hematoma, a single burr hole irrigation of CSDH without cyst removal or fenestration is recommended by various authors in asymptomatic AC and if AC is symptomatic or preoperative symptoms persist, additional surgery to treat AC should be considered. The management is still a matter of debate. It seems that the surgical decision depends on the surgeon’s attitude more than on a rational base due to the lack of the evidence-based research.

In asymptomatic cases admitted into hospital by incidental recognition of the malformation, a significantly high percentage of neurosurgeons in a recent survey were in favor of a “prophylactic” operation which was motivated by the life-long risk of intracranial bleeding whereas some argued that the risk was so high to justify a preventive operation. The rate of hemorrhage within or around the cysts is reported to not exceed 40/100 000 (0.04%) per year. Risk-to-benefit ratios concerning substantial benefit and possible complications in young population should be carefully calculated in deciding surgical management, no matter with shunting procedures, craniotomy and fenestration, pure or endoscopic-assisted surgery. Although the best management of sylvian fissure AC will remain controversial in the near future, it is the consensus that the AC patients and their families should be informed to avoid so-called contact sports such as boxing. Actually, since complicating hemorrhage may be caused by a minor trauma and there are some reports about the associated CSDH occurring in football games and even competitive race walking, or jumping, the patients with diagnosed AC should avoid violent sports so as to reduce the incidence of intracranial hemorrhage caused by head injuries.

REFERENCES


(Received December 7, 2010)
Edited by LIU Jun-lan