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CASE REPORT

Progressively enlarging childhood chronic subdural hematoma surgically treated 26 years after diagnosis

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Abstract

Pediatric chronic subdural hematoma (CSDH) is a relatively common disorder. Treatment often requires burr hole drainage or subduroperitoneal shunt placement; some patients are managed conservatively. However, the long-term outcome of untreated pediatric CSDH is unknown. The authors report a case of a huge, progressively enlarging subdural granuloma that was surgically treated 26 years after the initial diagnosis of CSDH. This 30-year-old male patient presented with worsening intermittent atonic seizure-like movements, which had been noted since he was 4 years old. At that time, the patient was diagnosed with CSDH at an outside hospital, but an operation was refused by the parents. Magnetic resonance imaging (MRI) performed at 23 years of age showed a huge subdural mass on the right frontoparietal region and a smaller mass on the left side with a significant midline shift. Upon presentation at the age of 30, MRI revealed worsening of the right subdural mass and midline shift. Subsequently, the patient underwent craniotomy and subtotal removal of the mass and capsule. The volume of the content was approximately 430 cc. Untreated pediatric CSDH can grow progressively, even over several decades. Patients with CSDH, especially those managed conservatively, should be closely monitored for worsening symptoms over a long-term follow-up period.

Keywords chronic subdural hematoma · pediatric · calcification · long-term follow-up

Introduction

Pediatric chronic subdural hematoma (CSDH) is a relatively common disorder. It is usually found in infants, whose skull is vulnerable to expansion, or in children harboring an arachnoid cyst [6]. Ventriculoperitoneal shunting is one of the most important predisposing factors of CSDH [8]. The most common treatments for CSDH are burr hole drainage or subduroperitoneal shunting, though some patients are managed conservatively. There are reports of calcified CSDH after a prolonged period of time in untreated or unrecognized cases [5, 8, 10]. In some cases, calcification covers an extensive surface of the brain, leading to a condition termed "armored brain" [2, 9, 11]. Most of these reports are not the consequence of conservative management with long-term follow-up, but rather they are recognized at the time of radiological diagnosis of a calcified CSDH. Thus, the long-term progression and outcome of untreated, unresolved pediatric CSDH is unknown. Growth of the calcified CSDH is rarely reported [5, 7].

Herein, the authors report a case of a huge and partially calcified CSDH that was progressively enlarging during the 26 years following the initial diagnosis.

Case

A 30-year-old male patient presented with worsening intermittent atonic seizure-like movements, which had been noted since he was 4 years of age. At age 4, the patient was diagnosed with CSDH at an outside hospital, but his parents refused the operation. At 8 years of age, he presented at our hospital with seizures and underwent subduroperitoneal shunt surgery and began treatment with an antiepileptic drug. Though magnetic resonance imaging (MRI) was performed, his MR images from this time were not available; however, we found a schematic drawing of the MRI from the patient's record (Fig. 1a). When he presented to us again at 23 years of age due to seizures, MRI showed a huge subdural mass on the right frontoparietal region and a smaller one on the left side

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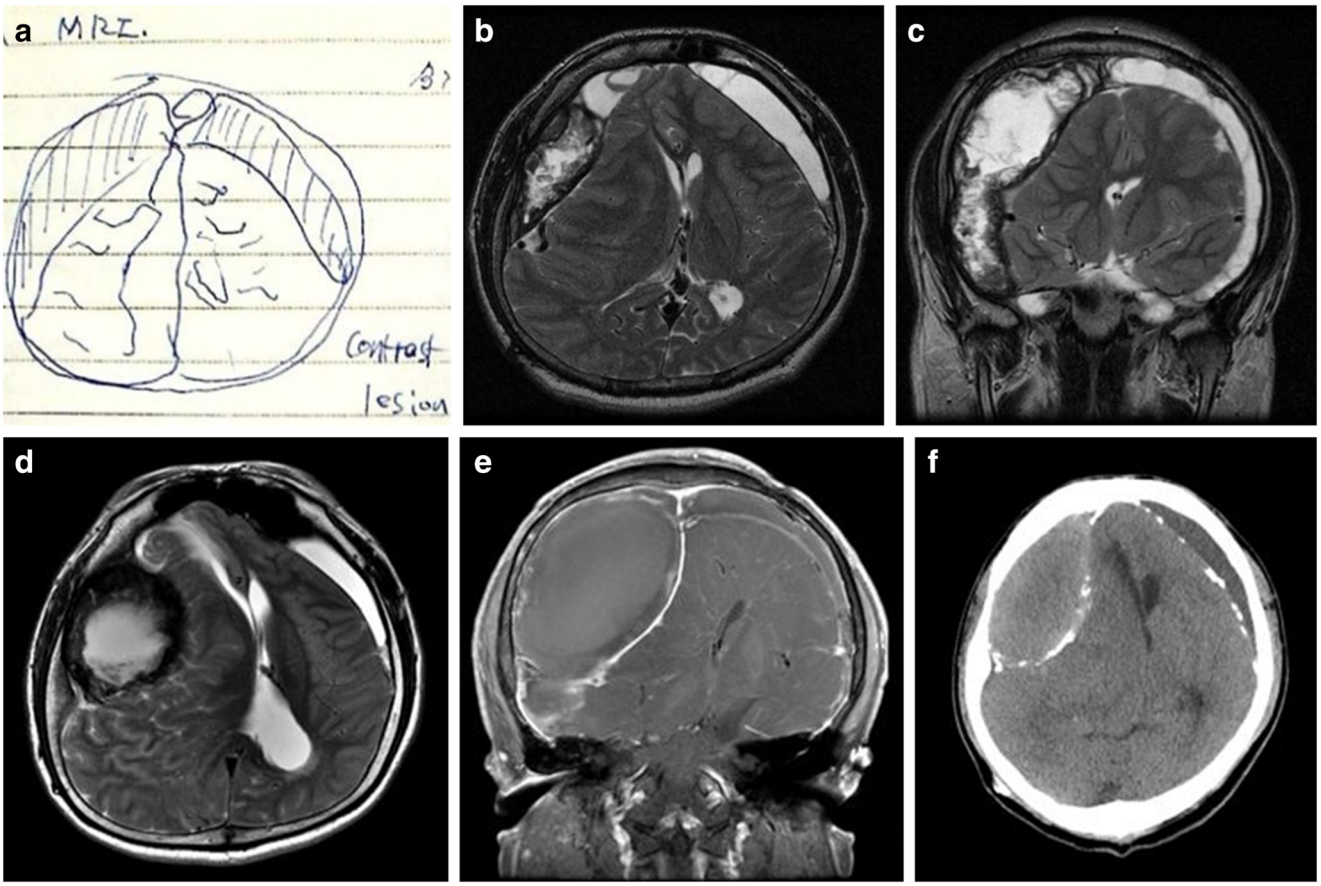

Fig. 1 Sequential radiological findings of the case. **a** Schematic drawing from the patient's medical record, dated January 19, 1994 (8 years old), mentioning subdural fluid collection with slightly high signal on T1-weighted imaging (T1WI) and high signal on T2WI without modifications of the lesions with gadolinium enhancement (radiological films could not be made available). **b**, **c** The magnetic resonance images taken at 23 years of age revealed a large subdural hematoma with a significant midline shift and ventricular collapse in the right frontoparietal region. **d**, **e** The magnetic resonance imaging performed at 30 years of age revealed an enlarged subdural hematoma and worsened midline shift. **f** A computed tomography scan showed partial calcification of the hematoma capsule

with significant midline shifting (Fig. 1b, c). He had a relatively large head with a circumference of 59.5 cm. He had mild intellectual deficits but a normal social life and no focal neurological deficits. We recommended an operation, but he refused the procedure. Seven years later, at the current presentation, follow-up MRI revealed an enlarged right subdural mass and worsening of the midline shift (Fig. 1d, e). CT scan showed partial calcification of the hematoma capsule (Fig. 1f). Surgery was recommended, and the patient agreed. The operation consisted of a craniotomy and subtotal removal of the mass and capsule. The subdural mass was covered by a thick, fibrous capsule containing a yellowish mud-like material and fluid (Fig. 2). The volume of the mass was approximately 430 cc. Biopsy revealed partially calcified fibrous membranous tissue and a granuloma (Fig. 3). Because the patient's brain expansion was unsatisfactory, subduroperitoneal shunt surgery was performed 1 month later. Follow-up MRI obtained 1 year after surgery revealed a greatly improved midline shift and ventricular collapse, but the expansion of the brain was still unsatisfactory (Fig. 4).

Discussion

This report describes the long-term course of untreated pediatric CSDH. Our patient was first diagnosed at 4 years of age; however, treatment was refused by the parents at the time. Subsequent MR imaging was performed at 8, 23, and 30 years of age. By age 30, the CSDH had progressed to a large and granulomatous mass, and an operation was performed.

The outcome of untreated CSDH is variable and can include spontaneous resorption, organization, or calcification. However, the exact pathogenesis, mechanism of improvement after surgery, and outcomes are unknown. One of the main reasons for these unresolved questions is that we do not have a relevant experimental model [1].

The incidence of organization or calcification is reported to be 0.5% to 10% [4, 5]. The development of computed tomography scanning has improved the ability to detect CSDH and its calcification. The reported time interval of calcification after the development of CSDH is variable, from several months to many years. Papanilokaou et al. [8] reported a case

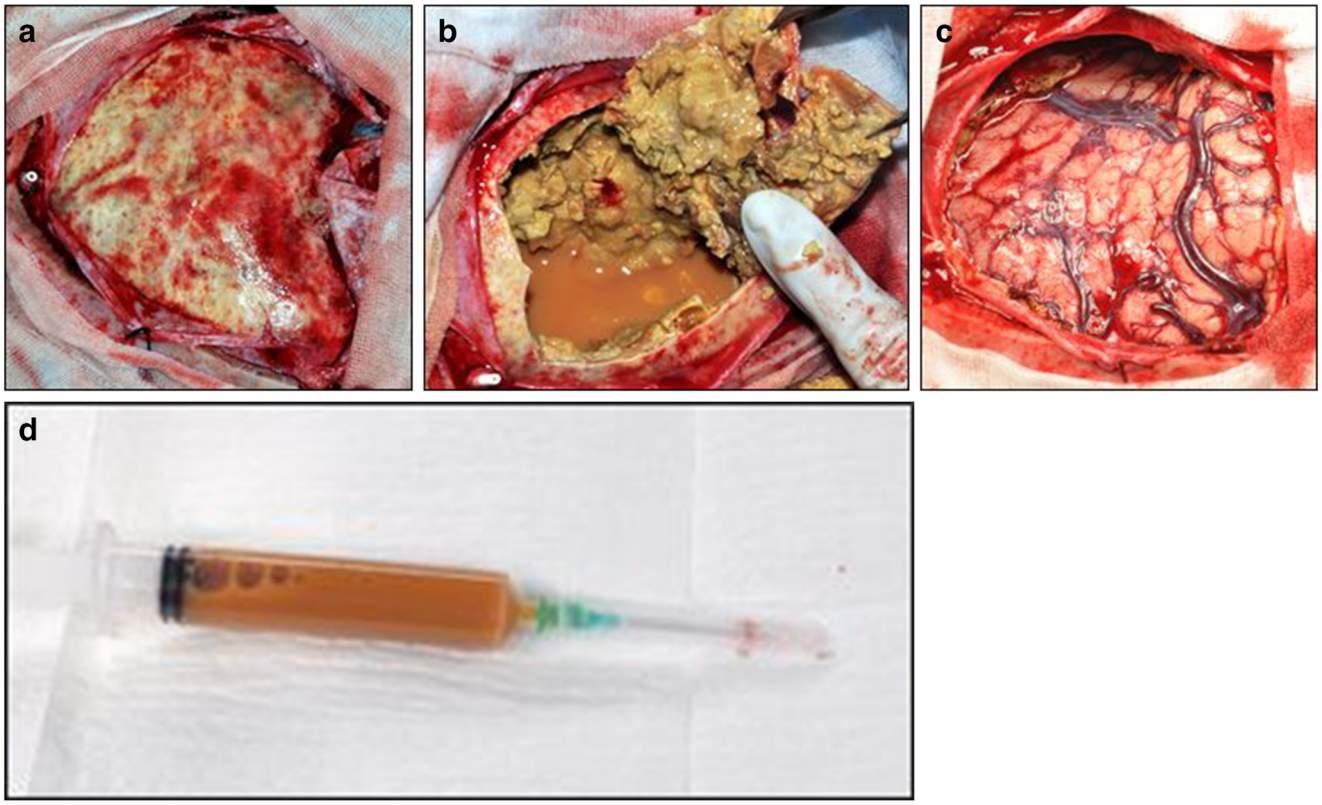

Fig. 2 Right frontoparietal craniotomy with subtotal removal of the thick capsule and granulomatous content was performed (**a–c**). Mud-like content and yellowish fluid (**d**) were identified. The volume of the content was estimated to be 430 cc

of shunt revision after 33 years in a patient with bilateral calcified CSDH.

There are a few reports of calcified SDHs that are quite large, which is a long-term consequence of untreated SDH. Because most of the reports of calcified CSDH do not mention the patient's history before presentation, we do not know the long-term course or progression of organized or calcified CSDH. Among five cases of calcified CSDH described by Imaizumi et al. [5], two were found to be enlarged after 2 and 3 years of follow-up. Enlargement of the organized

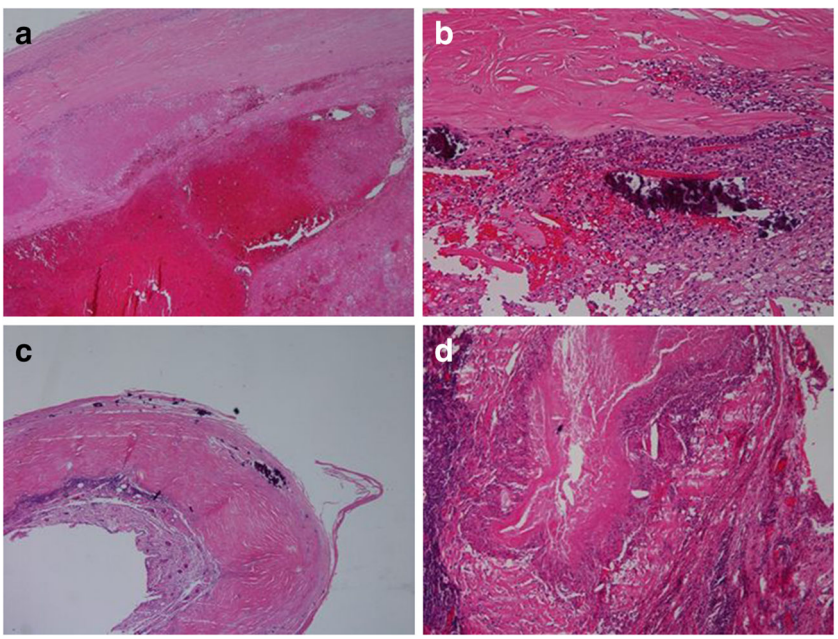

Fig. 3 Pathologic findings of the membrane. Outer membrane revealed thickened fibrous capsule with organized hematoma (**a**, × 40) and scattered calcification with inflammatory cells (**b**, × 100). Inner membrane revealed irregularly thickened wall with multi-scattered calcification (**c**, × 20) and organized hematoma with surrounding hyalinization, inflammation, and calcification (**d**, × 100)

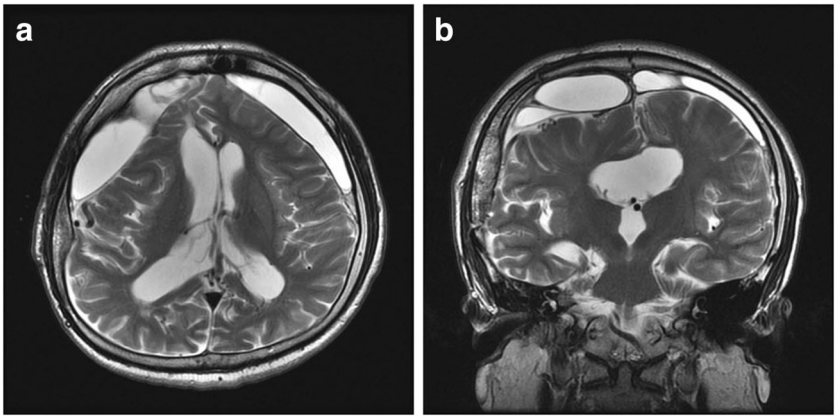

Fig. 4 Follow-up magnetic resonance imaging performed 1 year after surgery revealed remarkable improvement of the midline shift and ventricular collapse, though brain expansion was not satisfactory

CSDH 1 year after partial removal was reported by Mori et al. [7]. In infants, mild asymptomatic subdural hygroma can develop into CSDH [3]. In such cases, if we do not recognize the CSDH sufficiently early, it may enlarge progressively and result in permanent sequelae.

Our patient showed a slow progression of the subdural mass lesion for several decades, which rationalizes the performance of surgery and close follow-up for CSDH, especially in children. Our case was unique in its observation of the progressive growth of the CSDH, even for decades. The calcification in our case was relatively sparse compared to other reports of calcified CSDH; the hematoma may not have enlarged if the calcification was dense enough.

Conclusion

Herein we report a case of a progressively enlarging CSDH with partial calcification that was surgically treated 26 years after the initial diagnosis. This case highlights the importance of long-term follow-up for untreated CSDH, especially in children.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval For this type of study, formal consent is not required.

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