Synchronous Diagnosis of Intradiploic Epidermoid Cyst and Anatomically Close Associated Chronic Epidural Hematoma

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Key words
- Chronic
- Concurrent
- Epidural hematoma
- Epidermoid cyst
- Intradiploic

Abbreviations and Acronyms
CEDH: Chronic epidural hematoma
CT: Computed tomography
DWI: Diffusion weighted imaging
IEC: Intradiploic epidermoid cyst
MRI: Magnetic resonance imaging
T1WI: T1-weighted imaging
T2WI: T2-weighted imaging

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BACKGROUND: **Intradiploic epidermoid cyst (IEC) concurrent with chronic epidural hematoma (CEDH) has never been reported in the literature. We report a unique case of IEC concurrent with anatomically close associated CEDH.**

CASE DESCRIPTION: **A 54-year-old man presented with a 1-week history of headache, confusion, and drowsiness. Computed tomography exposed large, extraaxial, mixed-density lesions with bone destruction and substantial mass effect in the left frontotemporal region. Magnetic resonance imaging revealed 2 adjacent extraaxial lesions. The frontal lesion was predominantly hyperintense on both T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI), with an enhancement rim of the thickened dura mater. The frontotemporal lesion was mainly hyperintense on both TIWI and T2WI, with some areas of hypointensity on both T1WI and T2WI. No enhancement in the lesion was found. The patient underwent left frontotemporal craniotomy for resection of the lesion. At the time of the craniotomy, a copious amount of yellow liquid outflowed. After the bone flap was removed, an irregular tumor was found epidurally located and composed of fragile and laminated yellow materials. The tumor was totally removed. Histologic examination revealed an epidermoid cyst. Postoperative computed tomography showed mild bleeding in the operation area. His symptoms improved significantly after surgery, and he was discharged 1 week later. At 2 weeks' follow-up, the patient was well without neurologic deficits.**

CONCLUSIONS: **Intradiploic epidermoid cyst concurrent with chronic epidural hematoma is extremely rare. We present the first case of intradiploic epidermoid cyst concurrent with anatomically close associated chronic epidural hematoma.**

INTRODUCTION

Intradiploic epidermoid cyst (IEC) is a benign, rare, slow-growing lesion that arises between the 2 tables of cranial bones.[1,2] It accounts for about 25% of all intracranial epidermoid cysts.[3-5] IEC commonly occurs at the frontal bone, parietal bone, and junction of the calvarium and skull base.[6] Chronic epidural hematoma (CEDH) is a rare disease, and the exact definition is not established yet.[7,8] In this article, we report an unusual case of IEC concurrent with CEDH in the left frontotemporal region. The clinical, radiologic, origin, and mechanism of this rare disease is discussed. To the best of our knowledge, this is the first case of IEC concurrent with anatomically close associated CEDH described in the literature.

CASE PRESENTATION

A 54-year-old man presented with 1-week history of headache, confusion, and drowsiness. He was sent to a local hospital. Computed tomography (CT) exposed large, extraaxial, mixed-density lesions with bone destruction and substantial mass effect in the left frontotemporal region, and the inner table of the skull was destroyed more than the outer table (Figure 1A and B). The patient was transferred to our hospital. On admission, physical examination revealed a scar over his left frontal scalp. No focal neurologic deficits were present. His routine laboratory tests were normal. He experienced a horseback riding accident (kicking injury), with an open scalp wound and suspected skull fracture in the left frontal region, at 24 years of age. The wound had been treated surgically with lavage and sutures. Unfortunately, neither medical records nor a brain CT related to the head trauma were available.

Magnetic resonance imaging (MRI) revealed 2 adjacent extraaxial lesions. The frontal lesion was predominantly hyperintense on both T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI), with an enhancement rim of the thickened dura mater (Figure 2A–E). The frontotemporal lesion was mainly hyperintense on both TlWI and T2WI, with some areas of hypointensity on both TlWI and T2WI within the lesion (Figure 2A and B). No enhancement in the lesion was found after contrast administration (Figure 2C–E).

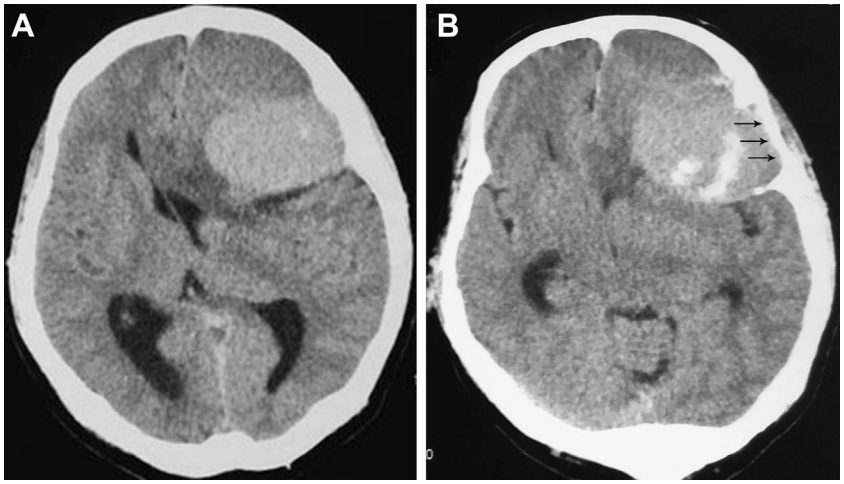

Figure 1. (**A**) Computed tomography exposed large extraaxial mixed density lesions and substantial mass effect in the left frontotemporal region. (**B**) The inner table of the skull was destroyed more than the outer table (*black arrows*).

The preoperative diagnosis was an intradiploic mass lesion concurrent with a chronic epidural hematoma. The patient underwent left frontotemporal craniotomy for resection of the lesion. At the time of the craniotomy, a copious amount of yellow liquid outflowed. After the bone flap was removed, an irregular tumor was found epidurally located and composed of fragile and laminated yellow materials (Figure 3A). The dura mater was thickened but intact throughout (Figure 3B). The tumor was detached easily from underlying dura, and the underlying dura was not breached. The exposed frontotemporal bone was widely destroyed, thinner, and partially perforated by the tumor (Figure 3C). Peritumoral bone tissue was excised by high-speed drill until the healthy, normal bone was reached circumferentially. Surgical findings confirmed that the tumor originated from the diploe, while the source of the hemorrhage was not identified. The tumor was totally removed, and the cavity was washed repeatedly with 0.9% saline. The dura was tacked up because there was a large epidural space remaining.

Histologic examination showed that the cystic structure was lined with squamous epithelium containing laminated keratin material and cholesterol crystals (Figure 4). Pathologic findings were consistent with the diagnosis of an epidermoid cyst.

His symptoms improved significantly after surgery. A CT scan taken on the first day post operation showed mild bleeding in the operation area (Figure 5). We planned close observation and a serial checkup of CT scans of the bleeding. The patient was discharged from the hospital without complications 1 week later. At 2 weeks' follow-up, the patient was well without neurologic deficits.

DISCUSSION

Epidermoid cysts are rare, congenital, slow-growing lesions that develop from ectodermal remnants during neuroembryogenesis.[9,10] They are commonly located in the cerebellopontine angle, juxtasellar areas, diploe, and spinal canal.[11] Intradiploic epidermoid cysts are frequently located in the occipital, frontal, and parietal bones.[12] Chronic epidural hematoma is a rare disease in the literature. There is no exact definition of the interval between the acute trauma and the moment of diagnosis to classify a CEDH.[7,8] Iwakuma et al[13] defined a "CEDH" when the interval between trauma and CT scan was $\geq$13 days, according to their histologic findings of calcification of the capsule. In medical practice, an epidural hematoma is classified as chronic when it appears as a mixed-density or lucent hematoma with a contrast-enhanced membrane.[7,8] Chronic epidural hematoma and delayed epidural hematoma are 2 different diseases. Delayed epidural hematoma is defined as a hematoma that is not present or insignificant on the initial CT scan done within the first 24 hours after trauma, and a subsequent CT scan shows a sizeable EDH.[14,15] The pathogenesis of IEC has been postulated as congenital and acquired mechanisms. Congenital acquisition may develop from ectodermal remnants that remain within the cranial bones during the third to fifth weeks of embryogenesis,[6,16] while the acquired type develops following head trauma or iatrogenic reasons with epidermal cells being implanted into the diploe of the skull.[2,17] Arko et al[18] reported that IEC has sex predilection toward men from their review, and they inferred that this may be because of a higher incidence of head injury and skull fractures in men. The etiology and pathogenesis of a CEDH remain controversial, but several possible mechanisms have been proposed: 1) venous origin and slow accumulation of the blood[19,20]—the source of bleeding may be from the emissary veins, meningeal venous sinuses, and diploic marrow[21,22]; 2) immediate dural stripping from the calvarium at head trauma, producing a space to receive low-pressure venous bleeding—this may initiate the formation of a CEDH[22,23]; 3) repeated bleeding and progressive dural stripping, leading to the expansion of a CEDH.[20-22]

Our patient experienced a horseback riding accident, with an open scalp injury and suspected skull fracture in the left frontal region, at 24 years of age. The wound had been treated surgically with lavage and sutures. Because of the lack of data related to the previous surgery in our patient, we cannot establish whether the IEC was congenital or acquired. However, as the IEC grew slowly and progressively, the dura mater was stripped from the skull and venous bleeding filled the epidural space. Repeated bleeding and progressive dural stripping led to the expansion of the CEDH.

Intradiploic epidermoid cysts are slow-growing lesions, and the symptoms and signs are related to the tumor size. Although both diploic tables are usually involved, many giant IECs are associated with extensive destruction of the inner table.[24] Patients with CEDHs can be asymptomatic or have persistent symptoms of headache, nausea, lethargy,