Spontaneous bilateral epidural hematomas caused by chronic sinusitis: illustrative case

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BACKGROUND Spontaneous bilateral epidural hematomas (EDHs) are rare. The aim of this study was to report a 21-year-old male with spontaneous bilateral EDHs to discuss the pathogenesis of spontaneous bilateral EDHs caused by chronic sinusitis.

OBSERVATIONS A 21-year-old male with no history of head trauma was admitted to the hospital for headache and unconsciousness. The patient had bilateral nasal bleeding on the day before admission and had chronic sinusitis since childhood. The head computed tomography examination after admission showed bilateral EDHs and bilateral sinusitis, the head magnetic resonance imaging showed chronic sinusitis, and the endoscopic examination during surgery further confirmed that the patient had severe sinusitis with erosion of the bilateral nasal mucosae. The patient underwent emergent surgical treatment. The cerebral vascular malformation, autoimmune diseases, low intracranial pressure, blood system diseases (such as sickle cell disease), abnormal blood coagulation, and skull or meningeal lesions were all excluded after operation.

LESSONS Chronic sinusitis may lead to EDHs through causing vascular degeneration, and abruption of the dura mater and skull. For young patients with spontaneous EDHs, neurosurgeons should carefully ask patients whether they have a history of chronic sinusitis to exclude the possibility of bleeding caused by chronic sinusitis.

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KEYWORDS spontaneous; bilateral epidural hematomas; chronic sinusitis; case report

Epidural hematoma (EDH) is mostly caused by head trauma, and most patients with EDH also have skull fracture and suture diastasis. Spontaneous EDH also exists but rarely occurs, and the underlying causes for spontaneous EDH include cerebral vascular malformation, blood coagulation abnormalities, frontal sinusitis, blood diseases, etc. Bilateral EDHs are very rare and comprise only approximately 2% of all intracranial hematomas.1 Spontaneous bilateral EDHs are extremely rare, with only a few reported cases without relevant epidemiological data (Table 1). This article presents the case of a 21-year-old male patient with bilateral EDHs caused by chronic sinusitis, the first of its kind.

Illustrative Case

A 21-year-old male was admitted to a community hospital because he had headache, nausea, and vomiting for 1 day, as well as coma for 6 hours. One day before admission, the patient had a headache accompanied by vomiting and bilateral nasal bleeding at home, but the patient remained conscious. His grandmother denied that he had a history of strenuous physical activities and head trauma. His family and the patient himself did not pay much attention to the previously described symptoms and signs, and the patient went to sleep. Six hours before admission, one of his family members called him to get up for breakfast, only to find that the patient was unconscious. The patient was rushed to a community hospital. The head computed tomography (CT) examination showed bilateral frontal EDHs and bilateral sinusitis (Fig. 1). The patient was then urgently transferred to our hospital for further evaluation and treatment. On physical examination, the patient was still unconscious, the diameter of the right pupil and the left pupil was 5 mm and 3 mm, respectively, and the pupillary light reflex was disappeared, the

ABBREVIATIONS CT = computed tomography; DSA = digital subtraction angiogram; EDH = epidural hematoma; GCS = Glasgow Coma Scale; RF = rheumatoid factor; SLE = systemic lupus erythematosus; vWF = von Willebrand factor.

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Glasgow Coma Scale (GCS) score was E1V1M2, body temperature was 36.6°C, heart rate was 95 beats per minute, blood pressure was 128/57 mm Hg, respiratory rate was 20 breaths per minute, and oxygen saturation was 98% while he was breathing ambient air. The results of the various laboratory tests are as follows. The white cell count was $14.5 \times 10^9/L$, with 90.1% neutrophils; the platelet count was $277 \times 10^9/L$; the red blood cell count was $5.93 \times 10^{12}/L$; the hemoglobin count was 162 g/L; the international normalized ratio was 1.09; the

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Age (yrs), Sex</th>
<th>Cause of Bilat EDHs</th>
<th>EDHs Site</th>
<th>Surgery</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Kuwayama et al., 1985</td>
<td>21, F</td>
<td>Hypofibrinogenemia</td>
<td>Temporal</td>
<td>Done</td>
<td>Recovered</td>
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<tr>
<td>Ishige et al., 1985</td>
<td>34, M</td>
<td>SLE</td>
<td>Parietal &amp; occipital</td>
<td>Done</td>
<td>Recovered</td>
</tr>
<tr>
<td>Charles et al., 1987</td>
<td>34, M</td>
<td>Alcoholic brain atrophy</td>
<td>Occipital</td>
<td>Done</td>
<td>Recovered</td>
</tr>
<tr>
<td>Grabel et al., 1989</td>
<td>2, M</td>
<td>Unknown</td>
<td>Temporal</td>
<td>Done</td>
<td>Recovered</td>
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<tr>
<td>Dufour et al., 2001</td>
<td>36, F</td>
<td>Cavernous hemangioma</td>
<td>Parietal</td>
<td>No</td>
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<tr>
<td>Melike et al., 2004</td>
<td>9, M</td>
<td>Eosinophilic granuloma of skull</td>
<td>Parietal</td>
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<td>Recovered</td>
</tr>
<tr>
<td>Ng et al., 2004</td>
<td>23, F</td>
<td>Unknown</td>
<td>Frontal</td>
<td>Done</td>
<td>Recovered</td>
</tr>
<tr>
<td>Aslam et al., 2006</td>
<td>Unknown</td>
<td>Extradural metastatic Ewing’s sarcoma</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Unknown</td>
</tr>
<tr>
<td>Verma et al., 2007</td>
<td>3, M</td>
<td>Scurvy</td>
<td>Frontal</td>
<td>No</td>
<td>Recovered</td>
</tr>
<tr>
<td>Li et al., 2012</td>
<td>14, F</td>
<td>Low intracranial pressure</td>
<td>Temporal &amp; parietal</td>
<td>Done</td>
<td>Recovered</td>
</tr>
<tr>
<td>Hettige et al., 2015</td>
<td>7, F</td>
<td>Sickle cell disease</td>
<td>Temporal</td>
<td>Done</td>
<td>Recovered</td>
</tr>
</tbody>
</table>

FIG. 1. Head CT on admission showed a high-density shadow in the right nasal cavity (A) and bilateral frontal EDHs (B). Head MRI showed bilateral sinusitis, the inflammation emitting high signals on T2-weighted images (C). Head DSA on admission (D and I) did not show any vascular abnormalities. Arterial phases: left internal carotid arteriography (D), left external carotid arteriography (E), right internal carotid arteriography (G), and right external carotid arteriography (H). Venous phases: left internal jugular venography (F), right internal jugular venography (I).
Discussion

Observations

Spontaneous bilateral EDHs are extremely rare, so the incidence is unknown. Here, for the first time, we report a 21-year-old male patient with bilateral EDHs caused by chronic sinusitis. The underlying causes of spontaneous bilateral EDHs are not well established. It is generally accepted that spontaneous EDHs are induced by various causes, such as cerebral vascular malformation, autoimmune system diseases, low intracranial pressure, blood diseases, coagulation disorders, skull tumor, and dural metastasis of malignant tumor, sinusitis and otitis media.2,3

Ishige et al.4 reported a 14-year-old girl with SLE who had bilateral EDHs due to epidural vascular degeneration. In contrast, the autoantigens associated with vasculitis in systemic lupus erythematosus (SLE), the antinuclear antibodies, and the rheumatoid factor (RF) were all negative. One week after admission, bone marrow aspiration was performed on the patient. The morphologies of the bone marrow cells were normal and the immunotyping of hematological tumor cells were also normal, so blood diseases were excluded (Fig. 2). The clotting factors II, V, VII, VIII, IX, X, XI, XII, and the von Willebrand factor (vWF) antigen were all examined, all of which were within normal limits (Table 2). Two weeks after admission, the patient was successfully weaned from the ventilator; the patient then underwent head magnetic resonance imaging (MRI), which showed chronic sinusitis. Next, we performed nasal endoscopy at 4 weeks after admission, and we found the erosion and adhesion of the patient’s nasal septum mucosae. One month after the operation, the patient’s consciousness level was improved, and the GCS score was increased to 13.

Our patient; therefore, the SLE was not considered as the cause of bleeding in this patient.

Low intracranial pressure is also a cause of spontaneous EDH. Low intracranial pressure syndrome is generally caused by the decrease of brain volume, clinical symptoms include orthostatic headache, dizziness, mental retardation, and behavioral changes. Low intracranial pressure includes spontaneous and secondary types (such as craniocerebral surgery, spinal surgery, lumbar puncture, spinal anesthesia).5 Li et al.6 reported a 14-year-old girl who had developed bilateral EDHs due to low cranial pressure caused by massive cerebrospinal fluid loss during cervical spine surgery. Our patient had no orthostatic headache and no history of spinal surgery or lumbar puncture, and no significant dural enhancement was observed on head MRI. Therefore, the patient’s bleeding was not considered to be caused by low cranial pressure.

Blood diseases and abnormal blood coagulation are also the causes of spontaneous bleeding of EDH. Kuwayama et al.7 reported a 21-year-old female patient with spontaneous EDHs due to hypofibrinogenemia. Hettige et al.8 reported a 7-year-old girl with spontaneous EDHs due to sickle cell disease. Ntantos et al.9 reported a 44-year-old female patient with refractory immune thrombocytopenic purpura complicated with cerebral venous thrombosis, the patient developed spontaneous EDHs and subdural hematomas.

Cranial lesions and meningeal metastases of tumor can also cause spontaneous EDH. Melike et al.10 reported a 7-year-old boy

![FIG. 2. The bone marrow morphology test (A, original magnification × 100; B, original magnification ×1000, Wright's-Giemsa staining) at 1 week after admission shows that the granulocyte was slightly increased, the mature erythrocytes were generally normal, and there was hyperplastic anemia. The white cell count was 12.5 × 10⁹/L, the platelet count was 128 × 10⁹/L, the red blood cell count was 3.2 × 10¹²/L, and the hemoglobin count was 82 g/L on that day.](image-url)

<table>
<thead>
<tr>
<th>TABLE 2. Result of screening of blood examinations</th>
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<tbody>
<tr>
<td><strong>Examination</strong></td>
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<tr>
<td>---</td>
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<tr>
<td>White cell count</td>
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<tr>
<td>Red blood cell count</td>
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<tr>
<td>Platelet count</td>
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<tr>
<td>Hemoglobin count</td>
</tr>
<tr>
<td>Neutrophil percentage</td>
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<tr>
<td>Hemagglutination test*</td>
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<tr>
<td>Autoimmune system antibody test†</td>
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</tbody>
</table>

*aCL = Anti-Cardiolipin; APTT = activated partial thromboplastin time; β2-GP1 = β2-glycoprotein 1; ds-DNA = double-stranded deoxyribonucleic acid; Fib = fibrinogen; INR = international normalized ratio; PT = prothrombin time; RF = rheumatoid factor; Sm = smooth muscle; SSA = Sjogren syndrome A; vWF = von Willebrand factor. |

† Two weeks postoperative data.
with eosinophilic granuloma of skull, who developed spontaneous bilateral EDHs. The rupture of the tumor cyst was the possible cause for EDHs in this case. There are also reports of EDH caused by rupture of meningeal metastatic tumor, but those EDHs are unilateral and occur generally in middle-aged or elderly patients.9,10

Long-term sinusitis can erode intracranial blood vessels, which may rupture when patients forcefully cough or blow their noses. We noticed that the patient had bilateral nasal bleeding on the day of admission, history of chronic sinusitis, signs of chronic sinusitis on MRI after surgery, and bilateral nasal bleeding, so we speculated that bilateral frontal EDHs may be caused by chronic sinusitis. In addition, we found that the patient had erosions of the bilateral nasal mucosae and severe sinusitis during endoscopic examination, which further verified our hypothesis. Furthermore, we found that the patient’s dura did not adhere tightly to the skull and extensive venous bleeding occurred during the operation. Taken together, we excluded cerebral vascular malformation, autoimmune diseases, low intracranial pressure, blood diseases, abnormal blood coagulation, and skull or meningeal lesions in our case and we speculated that before the onset of the bilateral EDHs, the patient might habitually blow his nose forcefully, resulting in a sudden increase in the pressure of the frontal sinus cavity with chronic inflammation to repeatedly force the accumulated inflammatory exudate and air into the space between the skull and the dura mater through the concealed fistula, causing an erosion of intracranial blood vessels and a progressive detachment of the dura mater from the inner table of the cranial bones.

Lessons

Bilateral spontaneous EDHs caused by sinusitis are extremely rare. For patient with spontaneous EDHs, the patient’s past history should be carefully inquired. The history includes runny nose, nose bleed, nose blow, and other symptoms for a long time. If possible, imaging exams, such as CT, MRI, or nasal endoscopy should be performed when sinusitis is suspected. EDH caused by chronic sinusitis may be accompanied with extensive venous hemorrhage, so that neurosurgeons should make adequate preparations, such as suspension of red cells or autologous blood before craniotomy in case that hemorrhagic shock occurs.

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References

Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Yu, Yao, Huang, Fang, Jiang, Acquisition of data: all authors. Analysis and interpretation of data: Yu, Luo, Zhou, Huang, Jiang. Drafting the article: Yu, Yao, Luo, Zhou, Fang. Critically revising the article: Yu, Huang, Jiang. Reviewed submitted version of manuscript: Yu, Fang. Approved the final version of the manuscript on behalf of all authors: Yu. Statistical analysis: Huang. Administrative/technical/material support: Yu. Study supervision: Yu, Yao.

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