Cerebral Venous Sinus Thrombosis Associated with Systemic Multiple Hemangiomas Manifesting as Chronic Subdural Hematoma

—Case Report—

Yukio TAKAMURA, Shigefumi MORIMOTO, Teiji UEDE, Toshiaki YAMAKI*, Yoshihiro MINAMIDA*, Akinori YAMAMURA*, and Toshio NAKAGAWA*

Department of Neurosurgery, Kushiro City General Hospital, Kushiro, Hokkaido; *Division of Neurosurgery, Shinsapporo Neurosurgical Hospital, Sapporo

Abstract

A 35-year-old male was admitted with headache, nausea, and vomiting persisting for 2 days. Computed tomography (CT) revealed a left chronic subdural hematoma. Cerebral angiography demonstrated cerebral venous sinus thrombosis (CVST). He had presented with a subcutaneous mass involving the neck at age 2 years, which was shown to be a cavernous angioma, and thereafter shown signs of consumptive coagulopathy with systemic multiple hemangiomas. Burr hole aspiration of the hematoma was performed. Seventy-two hours later, he developed clouding of consciousness and right hemiparesis. CT revealed a fresh hematoma in the operated subdural cavity and hemorrhagic diathesis manifested. A frontotemporoparietal large craniotomy was performed to remove the hematoma. Extensive electrocauterization was required. He had a satisfactory postoperative course. Collateral venous pathways, resulting from the CVST due to systemic multiple hemangiomas, may have caused hemodynamic stress in the bridging veins which subsequently induced chronic subdural hematoma.

Key words: cerebral venous sinus thrombosis, hemangioma, chronic subdural hematoma, consumptive coagulopathy, disseminated intravascular coagulation

Introduction

Cerebral venous sinus thrombosis (CVST) may be promoted by various pathological conditions, such as infectious processes, closed head injury, neoplasm, metabolic disorder, dehydration, pregnancy, and disseminated intravascular coagulation (DIC). We describe a rare case of CVST associated with systemic multiple hemangiomas manifesting as chronic subdural hematoma.

Case Report

A 35-year-old male presented with headache, nausea, and vomiting persisting for 2 days in February 1992. He had developed a subcutaneous mass in the neck when aged 2 years which had gradually enlarged. The mass was totally removed at age 5 years. Histological examination revealed cavernous angioma. Another angioma in the lower lip was removed at age 20 years. Multiple subcutaneous masses had been found throughout his body over a 30-year period.

Neurological examination on admission found no definite abnormalities. Physical examination revealed multiple hemangiomas in the head, neck, axilla, flank, throat, and scrotum. There was no apparent history of seizure or head trauma. Computed tomography (CT) revealed a left chronic subdural hematoma (Fig. 1). Magnetic resonance (MR) imaging showed a chronic subdural hematoma over the left cerebral convexity with midline shift. MR imaging with gadolinium demonstrated multiple enhanced small masses adjacent to the dura mater, which were thought to be dilated veins (Fig. 2). Cerebral angiography showed complete occlu-
sion of the anterior segment of the superior sagittal sinus and the right cavernous sinus, and segmental occlusion of the left transverse sinus. Multiple tortuous collateral vessels had developed, mostly through the inferior sagittal sinus which passed posteriorly to empty into the dilated straight sinus. The right Trolard’s vein, the right basal vein, and the superior and inferior vermian veins were enlarged and tortuous (Figs. 3-5). Laboratory data at admission are shown in Table 1.

The hematoma was evacuated through a burr hole. Seventy-two hours later, he developed conscious disturbance and right hemiparesis. CT showed a fresh hematoma in the operated subdural cavity with marked midline shift. His platelet count fell to 125,000/mm³ and he manifested hemorrhagic diathesis. His fibrinogen level was 54 mg/dl and fibrin degradation products 20 μg/ml. The hematoma was removed through a frontotemporoparietal craniotomy. Electrocauterization was used extensively to achieve hemostasis in the scalp, pericranium, dura mater, and outer membrane of the hematoma. He completely recovered within 20 days. No recurrence of the subdural hematoma has since occurred.

### Discussion

The natural history and prognosis of CVST are highly variable. In recent series, the mortality was only 10%, possibly due to recent developments allowing the diagnosis of benign forms of CVST with minimal symptoms, or to spontaneous recovery. Our patient had no predisposing factors for CVST and presented with chronic subdural hematoma. The pathogenesis

<table>
<thead>
<tr>
<th>Table 1 Laboratory data</th>
<th>Our patient</th>
<th>Reference range</th>
</tr>
</thead>
<tbody>
<tr>
<td>WBC (×10³/mm³)</td>
<td>6500</td>
<td>4000–8000</td>
</tr>
<tr>
<td>RBC (×10⁹/mm³)</td>
<td>5.7</td>
<td>4.1–5.4</td>
</tr>
<tr>
<td>Hematocrit (%)</td>
<td>46.4</td>
<td>39–50</td>
</tr>
<tr>
<td>Platelet (×10⁹/mm³)</td>
<td>25.4</td>
<td>12–40</td>
</tr>
<tr>
<td>Prothrombin time (sec)</td>
<td>12.4</td>
<td>control 10.5</td>
</tr>
<tr>
<td>Partial thromboplastia time (sec)</td>
<td>38.6</td>
<td>control 32.0</td>
</tr>
<tr>
<td>Plasma fibrinogen (mg/dl)</td>
<td>97</td>
<td>200–400</td>
</tr>
<tr>
<td>FDP (µg/dl)</td>
<td>10</td>
<td>&lt; 10</td>
</tr>
<tr>
<td>FDP-D dimer (µg/dl)</td>
<td>19.1</td>
<td>&lt; 1</td>
</tr>
<tr>
<td>Antithrombin III (%)</td>
<td>97</td>
<td>&gt; 80</td>
</tr>
</tbody>
</table>

FDP: fibrin degradation products, RBC: red blood cell count, WBC: white blood cell count.
of CVST was not established, but a possible mechanism is his hypercoagulable state. Complications of hemangiomas include hemorrhage, pain, heart failure, and DIC. Giant or multiple hemangiomas are known to be a variant of DIC. The cause of his DIC is not clear, but possibly the blood was static in the venous sinusoids of the tumor itself and the abnormal endothelium had activated both platelets and contact factors. Coagulation analysis revealed prolongation of prothrombin time and partial thromboplastin time, increased levels of serum fibrin degradation products and fibrin degradation products—D dimer, and decreased level of plasma fibrinogen, which are compatible with chronic DIC. Patients with nonmetastatic superior sagittal sinus thrombosis as a complication of systemic cancer have also suffered DIC.

Head trauma or alcoholism, a common cause of chronic subdural hematoma, was not present in our patient. Nontraumatic chronic subdural hematomas are occasionally associated with hemorrhagic diathesis secondary to advanced cancer, hemodialysis, liver cirrhosis, and hemophilia. Our patient may have developed collateral venous channels as a result of asymptomatic repeated occlusion and reperfusion of the main cerebral veins and dural sinuses due to his abnormal coagulation state. We therefore postulate that the collateral venous pathways caused hemodynamic stress in the bridging veins which subsequently induced the chronic subdural hematoma. In addition, hemorrhagic diathesis secondary to blood coagulation abnormalities might facilitate the formation of chronic subdural hematoma. This case demonstrates the unique concurrence of systemic multiple hemangiomas and CVST, manifesting as chronic subdural hematoma.

**Acknowledgment**

The authors thank Prof. K. Miyasaka, Department of Radiology, Hokkaido University Medical School, for his critical review of this manuscript and for his valuable advice.

**References**

5) George JN, Aster RH: Thrombocytopenia due to enhanced platelet destruction by nonimmunologic


Address reprint requests to: Y. Takamura, M.D., Department of Neurosurgery, Kushiro City General Hospital, 1-12 Shunkodai, Kushiro, Hokkaido 085, Japan.