Giant Aneurysm of the Azygos Anterior Cerebral Artery
Associated with Acute Subdural Hematoma
—Case Report—

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Abstract
A ruptured giant aneurysm of the azygos anterior cerebral artery (ACA) associated with an acute subdural hematoma (SDH) occurred in a 67-year-old male with two episodes of sudden severe headache and transient loss of consciousness. Neurologically, he had mild weakness of the left lower extremity. Computed tomography showed an elliptical heterogeneous hyperdense mass in the interhemispheric fissure in front of the corpus callosum and an acute SDH on the right. Angiography disclosed a giant aneurysm (2.8 × 2.0 cm) at the distal end of the azygos ACA. Removal of the SDH and aneurysmal neck clipping achieved a good outcome. Successive small bleedings may allow the aneurysmal dome to develop adhesions to the arachnoid membrane, and the final rupture will occur into the subdural space, resulting in a SDH.

Key words: azygos artery, anterior cerebral artery, giant aneurysm, subdural hematoma

Introduction
The azygos anterior cerebral artery (ACA) is a comparatively rare variation of the ACA, defined as a single unpaired ACA supplying the medial surface of both cerebral hemispheres. The incidence is 1.1% in human autopsy cases.3 Aneurysms of the azygos ACA have been reported, but giant aneurysms are extremely rare, with only three such cases in the literature.3,13,31 Ruptured intracranial aneurysms associated with acute subdural hematoma (SDH) have an incidence of 0.5-7.9%.4,5,7,8,27,29 SDHs due to ruptured distal ACA aneurysms are rare.2,4,10,12,14

Here, we report a ruptured giant aneurysm of the azygos ACA presenting as acute SDH without subarachnoid hemorrhage, and discuss the mechanism of formation of the SDH.

Case Report
A 67-year-old male suddenly complained of severe headache and became unconscious for about 10 minutes. He had been diagnosed as hypertensive about 8 months before, but had received no medication. Computed tomographic (CT) scans at a local hospital disclosed an elliptical heterogeneous hyperdense mass (2.0 × 2.5 cm) in the interhemispheric fissure in front of the genu of the corpus callosum, a crescent hyperdense area (0.8 cm maximum thickness) over the right cerebral convexity, and a linear hyperdense area along the falx, with the midline displaced to the left.

He was transferred to our institute about 2 hours later. On admission, his blood pressure was 138/76 mmHg, and his Glasgow Coma Scale score 14. He complained of headache and weakness of the left lower extremity. Soon after admission, he suddenly lost consciousness for a short period, accompanied by severe headache, slight vomiting, and urinary incontinence.

Plain skull x-ray films showed no abnormalities. CT scans revealed the hyperdense interhemispheric mass growing to 2.5 × 3.0 cm, and the crescent hyperdense area to 1.5 cm maximum thickness. However, the scans showed no hyperdense area in the subarachnoid space (Fig. 1). Right carotid angiograms showed that the proximal ACA after the
horizontal (A,) portion was single, with a giant aneurysm at the division into two pericallosal arteries. Left carotid angiograms with a contralateral carotid compression confirmed the azygos ACA, although the A, portion of the left ACA was hypoplastic. The aneurysmal shadow (2.8 × 2.0 cm) remained until the early venous phase, although the aneurysmal neck was unclear. Therefore, the neck was thought to be broad. Blood flow into the distal ACA was good. An avascular area of 1.0 cm thick was observed over the right cerebral convexity, and the azygos artery was shifted 1.0 cm to the left (Fig. 2).

A ruptured giant aneurysm of the azygos ACA with an acute SDH was diagnosed. The aneurysm was approached through the interhemispheric fissure via a paramedian frontal craniotomy. There was considerable hematoma in the subdural space over the right frontal and temporal lobes, but no subarachnoid hemorrhage. The pulsating aneurysmal dome, surrounded by much clotting, adhered firmly to the free edge of the falx. The aneurysm projected from the parent artery to the right and upward to the edge of the falx. The aneurysmal neck was clipped with a Sugita straight clip, and the dome was resected. The azygos artery was clipped totally for 13 minutes and 20 seconds.

The postoperative clinical course was generally good, and the paresis of the left lower extremity vanished. However, on the 3rd postoperative day, mild confabulation and disorientation appeared. The former continued for only a few days but the latter persisted for about 2 weeks. Right carotid angiograms 3 weeks after the operation revealed that the aneurysm was completely clipped and blood flow in the parent and the distal pericallosal arteries was good (Fig. 3). Postoperative CT scans showed that the SDH and the midline shift had vanished. He was discharged on the 44th postoperative day without neurological deficits.

**Discussion**

The azygos ACA is found in 0.5–3.2% of adult brains, and is often associated with anomalies of the central nervous system or cerebral aneurysms. The incidence of associated aneurysms is 13–71%. Therefore, the presence of the azygos ACA is deeply related to the formation of aneurysms.

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There are two mechanisms proposed for the formation of such aneurysms: the aneurysms develop accompanying the congenital anomalous artery; and the aneurysms result from the hemodynamic stress caused by the azygos artery.\textsuperscript{18} The latter is supported by the frequent occurrence of aneurysms at the peripheral end of the azygos artery.\textsuperscript{9,21,24} However, giant aneurysms, more than 2.5 cm in diameter, are not so frequent with an incidence of 7.4–13\% of all intracranial aneurysms.\textsuperscript{22,23,28}

Giant aneurysms of the azygos ACA are extremely rare, with only three previous cases reported (Table 1).\textsuperscript{13,31} These aneurysms and ours all occurred at the distal end of the azygos ACA. The previous three cases had symptoms caused by mass effect, gait disturbance, generalized convulsion, or transient ischemic attack. In our case, the symptoms were caused by aneurysm rupture. CT findings in all four cases were large, globoïd, hyperdense masses in the interhemispheric fissure, with enhancement around the periphery of the lesion postcontrast in the previous three cases.

<table>
<thead>
<tr>
<th>Author (Year)</th>
<th>Age/ Sex</th>
<th>Rupture</th>
<th>Signs and symptoms</th>
<th>Size of aneurysm (cm)</th>
<th>Thrombosis</th>
<th>Operation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Hayashi et al. (1985)\textsuperscript{13}</td>
<td>59/M</td>
<td>–</td>
<td>GC attack, anosmia</td>
<td>4 × 4</td>
<td>+</td>
<td>clipping</td>
</tr>
<tr>
<td></td>
<td>57/M</td>
<td>–</td>
<td>TIA, lt hemiparesis</td>
<td>4 × 4</td>
<td>+</td>
<td>none</td>
</tr>
<tr>
<td>Yamagami et al. (1986)\textsuperscript{31}</td>
<td>51/M</td>
<td>–</td>
<td>gait disturbance</td>
<td>2.7 × 2</td>
<td>+</td>
<td>clipping</td>
</tr>
<tr>
<td>Present case</td>
<td>67/M</td>
<td>+</td>
<td>headache, loss of consciousness</td>
<td>2.8 × 2</td>
<td>–</td>
<td>clipping</td>
</tr>
</tbody>
</table>

GC: generalized convulsion, TIA: transient ischemic attack.

Table 2  Reported cases of SDH due to ruptured distal ACA aneurysm

<table>
<thead>
<tr>
<th>Author (Year)</th>
<th>Age/ Sex</th>
<th>Site of aneurysm</th>
<th>Size of aneurysm (mm)</th>
<th>SAH</th>
<th>ICH</th>
<th>Operation</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Strass et al. (1932)\textsuperscript{9}</td>
<td>31/M</td>
<td>small branch of ACA</td>
<td>not described</td>
<td>?</td>
<td>+</td>
<td>clipping</td>
<td>death</td>
</tr>
<tr>
<td>Clarke and Walton (1953)\textsuperscript{3}</td>
<td>42/F</td>
<td>rt ACA</td>
<td>6</td>
<td>+</td>
<td>+</td>
<td>clipping, removal of SDH</td>
<td>death</td>
</tr>
<tr>
<td>Handel et al. (1978)\textsuperscript{10}</td>
<td>43/M</td>
<td>lt ACA</td>
<td>not described</td>
<td>+</td>
<td>–</td>
<td>trapping, removal of SDH</td>
<td>death</td>
</tr>
<tr>
<td>Honda et al. (1984)\textsuperscript{14}</td>
<td>47/M</td>
<td>lt ACA</td>
<td>20</td>
<td>+</td>
<td>–</td>
<td>clipping, removal of SDH</td>
<td>good</td>
</tr>
<tr>
<td>Ban et al. (1985)\textsuperscript{2}</td>
<td>56/F</td>
<td>lt ACA</td>
<td>6 × 3</td>
<td>+</td>
<td>–</td>
<td>clipping, removal of SDH</td>
<td>MD</td>
</tr>
<tr>
<td>Hasegawa et al. (1986)\textsuperscript{12}</td>
<td>50/F</td>
<td>rt ACA</td>
<td>12.5 × 7 × 6</td>
<td>+</td>
<td>+</td>
<td>clipping, removal of SDH</td>
<td>SD</td>
</tr>
<tr>
<td>Present case</td>
<td>67/M</td>
<td>azygos ACA</td>
<td>28 × 20</td>
<td>–</td>
<td>–</td>
<td>clipping, removal of SDH</td>
<td>good</td>
</tr>
</tbody>
</table>

dle cerebral artery (27%), the ACA and anterior communicating artery (17%), and the vertebrobasilar artery (3%). SDH associated with ruptured aneurysm of the distal ACA is extremely rare, with only seven reported cases, none due to a giant aneurysm (Table 2). Most cases also demonstrated intracerebral hematoma and/or subarachnoid hemorrhage.

Four mechanisms of formation of SDH have been proposed. 1) Successive small bleedings allow the aneurysmal dome to develop adhesions to the arachnoid membrane, and the final rupture occurs into the subdural space. 2) The arachnoid membrane is torn by the rapid accumulation of blood under pressure from the leaking aneurysm. This was apparently the cause in most cases. 3) Massive intracerebral bleeding ruptures the cortex and lacerates the arachnoid membrane. 4) An aneurysm arising from the carotid artery in the subdural space ruptures and directly causes a SDH. Mechanism 1) may be supplemented by enlargement of the aneurysm and continuation of the water hammer effect causing firmer adhesion of the aneurysm and the arachnoid membrane before the second bleeding, so laceration of the arachnoid membrane covering the aneurysm allows exposure of the dome in the subdural space. A series of nine ruptured aneurysms with acute SDH included seven cases (78%) with big aneurysms and seven (78%) with blebs, suggesting a tendency to easy rupture and bleeding into the intracerebral and subdural spaces adjoining the subarachnoid space.

Our case had a SDH without subarachnoid hemorrhage. The dome of the giant aneurysm had adhered firmly to the falx, and therefore the dome had also adhered to the arachnoid membrane, probably followed by direct rupture into the subdural space. The mechanism of development was therefore that of 1) above.

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