Primitive Trigeminal Artery Variant Associated with Intracranial Ruptured Aneurysm and Cerebral Arteriovenous Malformation

—Case Report—

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Abstract

A 48-year-old female presented with a unique combination of ruptured aneurysm of the right middle cerebral artery (MCA) manifesting as sudden loss of consciousness associated with arteriovenous malformation (AVM) in the right parietal lobe and left primitive trigeminal artery (PTA) variant. Angiography revealed the right MCA aneurysm, AVM fed by the right anterior cerebral artery and MCA, and contralateral PTA variant. The PTA variant was an anomalous posterior cerebral artery originating from the ipsilateral cavernous internal carotid artery. The neck of the aneurysm was successfully clipped and the AVM was totally removed.

Key words: cerebral aneurysm, arteriovenous malformation, primitive trigeminal artery variant

Introduction

Primitive trigeminal artery (PTA) is the most common anomaly of carotid-basilar anastomoses with an angiographic incidence of 0.2%.1) Associated cerebrovascular anomalies, such as aneurysm7,8,15,19,21,22,28 or arteriovenous malformation (AVM)10,25,26 are very common.1) However, persistent primitive artery associated with cerebral AVM and aneurysm is very rare.4,6,11,24 Here, we report a patient with PTA variant of the left internal carotid artery (ICA), AVM fed by the right anterior cerebral artery (ACA) and middle cerebral artery (MCA), and ruptured aneurysm of the right MCA.

Case Report

A 48-year-old female was admitted to our hospital after sudden loss of consciousness followed by severe headache and vomiting on August 16, 1991. There was no family history of neurological disease. On admission, her consciousness level was 13 on the Glasgow Coma Scale (GCS). Neurological examination revealed mild motor weakness on the left and moderate neck stiffness. Funduscopic examination disclosed a blurred disc margin. Blood pressure was elevated at 180/100 mmHg. There were no other neurological or physical abnormalities. The clinical diagnosis was subarachnoid hemorrhage (SAH).

Computed tomographic (CT) scans demonstrated diffuse SAH in the basal cistern and considerable subarachnoid blood clots in the right Sylvian fissure.

Fig. 1 CT scans on admission, demonstrating diffuse SAH in the basal cistern and remarkable subarachnoid blood clots in the right Sylvian fissure.

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Right carotid angiograms showed an aneurysm of the MCA (Fig. 2) and a small AVM nidus in the related medial portion of the right parietal lobe (Fig. 3). The small nidus was fed by the right ACA and MCA, and drained into the superior sagittal sinus. Left carotid angiograms demonstrated PTA variant and a small nidus in the right parietal lobe with cross flow via the anterior communicating artery (Fig. 4). The right posterior cerebral artery (PCA), bilateral superior cerebellar arteries (SCAs), and the basilar artery (BA) were filled via the left posterior communicating artery (PcomA). The left PCA was supplied via the PTA variant from the left cavernous ICA. Left vertebral angiograms did not demonstrate the left PCA. SAH was considered to be due to a ruptured aneurysm of the right MCA.

An emergency operation was carried out. The neck of the aneurysm was successfully clipped through the right pterional approach. Dense SAH was noted in the right Sylvian fissure. Postoperatively, her consciousness disturbance improved to 14 on the GCS. Marked consciousness disturbance of 8 on the GCS occurred 7 days after the operation, which was considered due to late vasospasm. Hypertension-hypervolemic therapy was given. The consciousness disturbance and left motor weakness improved on the 15th postoperative day. On September 11, 1991, right carotid angiograms demonstrated that the aneurysm of the right MCA was completely clipped and the AVM in the right parietal lobe had not significantly changed. On September 18, 1991, the AVM was totally removed via a right parietal craniotomy. Cerebral angiograms 2 weeks after the second operation showed no AVM. She was discharged without neurological deficit on October 18, 1991.

**Discussion**

Carotid-basilar anastomoses occur during normal embryonic development. In the 4 mm human embryo, the ICA is anastomosed with the longitudinal neural artery, which later becomes the BA, by four vessels: the trigeminal, otic, hypoglossal, and proatlantal intersegmental arteries. Generally, after the development of the PcomA, BA, and vertebral artery when the embryo reaches a length of 14 mm, these primitive anastomotic vessels disappear. However, the anastomoses sometimes persist and...
become anomalies of the carotid-basilar anastomoses. PTA is the most common anomaly of these, with an angiographic incidence of 0.2%. Most PTAs are detected incidentally during carotid angiography, and are usually of minimal clinical significance. In our patient, the PTA variant was detected incidentally with the ruptured aneurysm of the contralateral MCA during left carotid angiography.

Saltzman has described three angiographic conditions related to PTA: in type 1, the distal BA, SCA, and PCA are filled via the PTA, and the PcomA is hypoplastic or anaplastic; in type 2, filling is similar except that the ipsilateral PCA is filled by the ICA via the PcomA; and in type 3, there is poor filling of the distal BA and SCA, with each PCA filled by the respective PcomA. In addition, a variant of PTA called PTA variant has also been described recently. In this variant, one cerebellar artery, usually the anterior inferior cerebellar artery or SCA, is supplied by the cavernous ICA. This is thought to arise when a PTA is associated with incomplete fusion of the longitudinal neural arteries. In our patient, the PTA variant represents the anomalous PCA which originated from the ipsilateral cavernous ICA.

PTA is frequently associated with intracranial vascular anomalies (25%), especially with cerebral aneurysm (14%). The major sites of aneurysms associated with PTA are the circle of Willis or the PTA. The combination of ruptured aneurysm of the contralateral MCA and PTA is rare. PTA associated with AVM is relatively common (4.5%). Jayaraman et al. reviewed 11 similar cases of AVM associated with PTA, finding most clinical symptoms included intracranial hemorrhage or SAH. Many similar cases of AVM with PTA have since been described. However, the combination of persistent primitive artery, cerebral AVM, and cerebral aneurysm is extremely rare, with only four such cases reported (Table 1). Our patient presented with the unique combination of PTA variant of the left ICA, AVM fed by the right ACA and MCA, and ruptured aneurysm of the MCA.

Embryonic maldevelopment may result in cerebrovascular malformation. The nature of this embryonic disorder is not known. Cerebral AVMs and PTA are congenital, while the origin of aneurysms is controversial, with acquired and congenital factors being implicated. Hemodynamic stress is emphasized as the cause of aneurysm today. The occurrence of arterial aneurysms and AVM is usually characterized by a more proximal position of the AVM, believed to be associated with increased blood flow in the arteries supplying the AVM. In our patient, the AVM nidus was so small that it is difficult to postulate an increased blood flow was large enough to induce hemodynamic stress. Therefore, it is unlikely that hemodynamic stress is the only cause of arterial aneurysm. Trauma or perinatal infection, a cause of congenital intracranial aneurysms, was not present in this case. Possibly maldevelopment had caused structural insufficiency of the arterial wall, especially of the media, inducing massive arterial ectasias and later saccular aneurysm. Common congenital factors may be important in the development of arterial aneurysm.

References


Table 1 Summary of five cases of persistent primitive artery (PPA), AVM, and aneurysm

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Author (Year)</th>
<th>Age/ Sex</th>
<th>Symptom</th>
<th>Side of PPA</th>
<th>Location of AVM</th>
<th>Location of aneurysm</th>
<th>Treatment</th>
<th>Outcome*</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Karasawa et al. (1972)</td>
<td>28/F</td>
<td>SAH</td>
<td>lt POA</td>
<td>lt temporal lobe</td>
<td>extracranial portion of lt VA</td>
<td>uncertain</td>
<td>died</td>
</tr>
<tr>
<td>2</td>
<td>Tomchick et al. (1979)</td>
<td>uncertain</td>
<td>SAH</td>
<td>rt PTA</td>
<td>rt cavernous sinus</td>
<td>A1–A2 junction of rt ACA</td>
<td>uncertain</td>
<td>uncertain</td>
</tr>
<tr>
<td>3</td>
<td>Brick and Roberts (1987)</td>
<td>41/F</td>
<td>ICH</td>
<td>rt PTA</td>
<td>rt occipital lobe</td>
<td>cavernous portion of rt ICA</td>
<td>total removal of AVM</td>
<td>good recovery</td>
</tr>
<tr>
<td>4</td>
<td>Franz et al. (1985)</td>
<td>30/M</td>
<td>SAH</td>
<td>rt PAA</td>
<td>lt supratentorial</td>
<td>supracallosal portion of rt ICA</td>
<td>clipping of aneurysm</td>
<td>died</td>
</tr>
<tr>
<td>5</td>
<td>Present case</td>
<td>48/F</td>
<td>SAH</td>
<td>lt PTA variant</td>
<td>rt parietal lobe</td>
<td>rt MCA</td>
<td>clipping of aneurysm, total removal of AVM</td>
<td>good recovery</td>
</tr>
</tbody>
</table>


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