Surgical Evacuation of a Pontine Hematoma due to Rupture of a Capillary Telangiectasis in a Young Patient

—Case Report—

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Abstract

A 17-year-old female presented with a small pontine hematoma considered as having arisen from rupture of a capillary telangiectasis. Vertebral angiography showed fenestration of the left distal vertebral artery without any vascular anomaly in the pons. Histological examination of the surgical specimen verified the presence of a capillary telangiectasis adjacent to the hematoma. Rupture of a capillary telangiectasis as the source of pontine hematoma is very rarely documented histologically in surviving patients.

Key words: capillary telangiectasis, pons, cerebral hemorrhage, vertebral artery, cerebral angiography

Introduction

Surgical evacuation of spontaneous pontine hematomas in children and young adults has been reported by a number of authors.^{1,2,6,10,11,16,18,21,24,29,30)} However, evidence of a vascular malformation having ruptured into the pons has rarely been demonstrated in operated cases.^{8,9,19,21,22,24,30)} We treated a juvenile patient for subependymal hematoma of the pontine tegmentum, in which a capillary telangiectasis was present in the surgical specimen.

Case Report

A 17-year-old female experienced gradual onset of unsteady gait for 2 months preceding hospital admission. Vertigo, vomiting, and headache appeared suddenly, 2 weeks prior to admission. This vertigo was

induced on the left lateral position. The degree of these symptoms were gradually improved.

On admission the patient was alert, although headache and vertigo remained. Neurological examination revealed lateral horizontal nystagmus on both sides and ataxic gait. A lumbar puncture produced clear, watery cerebrospinal fluid. Roentgenograms of the skull were normal. A computerized tomographic (CT) scan demonstrated a small, high-density mass, approximately 1 cm in diameter, adjacent to the right side of the compressed fourth ventricle. Contrast enhancement was slight (Fig. 1). Left vertebral angiography revealed fenestration of the left distal vertebral artery, but other vascular anomalies and mass effect were not evident at the posterior fossa (Fig. 2).

We performed a bilateral suboccipital craniectomy. After lateral retraction of the cerebellar tonsils the vermis was split and the rhomboid fossa of the

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Fig. 1 Enhanced CT scan, showing a round, high-density mass adjacent to the fourth ventricle.

Fig. 3 Histological appearance of the surgical specimen, demonstrating abundant capillaries of varying diameter separated by inflammatory cells. HE stain, $\times 100$.

Fig. 2 Left vertebral angiogram, anteroposterior view in the arterial phase, revealing fenestration of the left distal vertebral artery.

fourth ventricle came into view. A small, reddish, convex mass covered by the ventricular ependyma was visible at the right ventral corner of the fourth ventricle within the right pontine tegmentum. No abnormal vessels were seen around the mass. The ependymal layer was pulled away and a portion of the hemorrhagic tissue was removed.

The surgical specimen consisted of a massive hemorrhage with many small vessels, compatible with a capillary telangiectasis (Fig. 3). The capillary walls, which were thin and of varying diameter, were composed of typical, well-differentiated endothelial cells. The capillaries were separated by inflammatory, hemosiderin-laden cells.

The postoperative course was uneventful. Nystagmus, vertigo, and unsteady gait gradually improved, although these symptoms persisted to a mild degree. Two months after the operation the patient was able to attend school. A postoperative CT scan, on infusion of contrast material, demonstrated a residual high-density region, and a repeat CT scan revealed its persistence. However, by 8 months

postoperatively it had disappeared completely.

Discussion

In the reported cases of surgically treated pontine hematomas, hemangiomatous tissue,³⁰⁾ a cryptic angioma,²¹⁾ a vascular malformation,²²⁾ and arteriovenous malformations^{9,19,24)} were histologically confirmed as the source of hemorrhage. However, a capillary telangiectasis has not previously been reported as a histologically documented cause of spontaneous pontine hematoma. Although it is known that capillary telangiectases are the most common vascular malformations in the pons,²³⁾ their role in pontine hematoma is difficult to demonstrate because they are usually small and stable and the wall of the hematoma cavity may be destroyed by bleeding. In our case a specimen obtained from the clot and fibrin-containing tissue on the floor of the fourth ventricle was found to be a capillary telangiectasis that happened, by chance, to be adjacent to the clot.

There has been no report of a pontine capillary telangiectasis associated with fenestration of a vertebral artery. The incidence of fenestration in the distal vertebral artery varies between 0.23 and 1.90%,^{5,12)} and is more frequent in Japan than in America and Europe.⁵⁾ There are some reports^{12,17)} of fenestration of the vertebral artery associated with congenital vascular anomalies, in particular with aneurysms and angiomas. It is interesting that in our case a capillary telangiectasis in the pons was associated with fenestration of the left distal vertebral artery.

Surgery for pontine hematomas is not performed routinely. In most of the operated cases of juvenile pontine hemorrhage,^{1,2,6,10,16,18,29,30)} brain tumor could not be ruled out before surgery. There are several reasons for the difficulty in diagnosis. The onset is not only sudden but also gradually progressive and symptoms tend to be recurrent. Vertebral angiography rarely demonstrates a vascular malformation. CT scans occasionally, over several months, reveal a high-density or heterogeneously dense mass in the pons. In cases of brain stem hematoma, Papo et al.²⁰⁾ mentioned that surgery is necessary in cases of intracranial hypertension caused by subependymal hematoma but that surgery is inappropriate for intra-axial hematomas. We operated on our patient because the CT findings were inconclusive; the high-density area seen on the CT scan might have been a brain tumor with a hematoma located in the right pontine tegmentum adjacent to the fourth ventricle.

Concerning the prognosis of capillary telangiectases within the pons, MacCormick et al.¹³⁾ stated that these appear to be the most innocuous type of angioma, for of 27 cases of pontine lesions, none was found to be associated with significant hemorrhage at autopsy. Russell and Rubinstein²³⁾ pointed out that examination of quiescent capillary telangiectases provided no evidence of small antecedent hemorrhages, which is not the case with cavernous angiomas. On the other hand, there have been several reports^{3,14,15,25,26,31)} of postmortem findings of pontine hematomas resulting from rupture of capillary telangiectases. Prolonged and progressive neurological symptoms were present in these cases. According to these reports, capillary telangiectases apparently do not bleed easily. However, when a small hemorrhage arises from this type of angioma, as in the case reported here, it is uncertain whether or not rebleeding will occur in future which was reported in cases^{2,4,21)} with juvenile pontine hematoma secondary to rupture from other angiomas. In our patient, the contrast-enhanced, high-density area in the pons disappeared from the CT scan 8 months after surgery. It has not reappeared, nor have there been any new symptoms. The vertigo this patient experience initially on the left lateral position is typical MPPV (malignant persistent positional vertigo),^{27,28)} due to hemorrhage in the right juxtarestiform body.⁷⁾

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