**Intracranial, Intra-parenchymal Capillary Hemangioma – Case Report –**

Yuichiro Koga,¹ Saori Hamada,¹ Hisayasu Saito,¹ Takuya Akai,¹ and Satoshi Kuroda¹

We report a very rare case of intracranial capillary hemangioma. This 15-year-old girl complained of pulsating headache in the temple area that aggravated with change of body positions. This headache usually lasted for 5 min and resolved without any treatment. Preoperative computed tomography (CT) and magnetic resonance imaging (MRI) strongly suggested cavernous hemangioma in the right deep parietal lobe. She underwent complete resection of the tumor through right parietal craniotomy. Postoperative course was uneventful. Histologic examinations demonstrated a densely grown numerous capillary-like vascular structure with endothelial cells, hemosiderin deposition, and hemorrhage. Intracranial, intra-parenchymal capillary hemangioma is a very rare vascular tumor or tumor-like lesions. Only four cases with intracranial, intra-parenchymal capillary hemangioma were reported previously. Differential diagnosis includes other vascular tumors such as cavernous hemangioma, but it is not so easy to differentiate capillary hemangioma from other lesions. Therefore, surgical excision and histologic diagnosis would be important to diagnose it if possible.

**Keywords:** intracranial capillary hemangioma, vascular tumor, surgery

**Introduction**

Capillary hemangiomas are benign vascular lesions or vascular tumors that are commonly found on the skin or soft tissue of neonates or infants. They are reported to occur in 1.1–2.6% of full-term neonates especially in their face, scalp, chest, or back.¹,² On the other hand, intracranial capillary hemangiomas are rare, because only 34 cases have been reported previously. However, a majority of them (30/34; 88.2%) originate from the dura mater and are found as an extra-axial mass.³ In this report, the authors present a quite rare case of intracranial, intra-parenchymal capillary hemangioma in the right parietal lobe.

**Case Report**

A 15-year-old girl with a past history of asthma and allergic rhinitis complained of intermittent headache in the right temporal area, which lasted for 5 min after body motion. One month later, she experienced severe headache followed by blurred vision, and was referred to prior hospital. She was diagnoses as having intracranial bleeding in the right parietal lobe and was transferred to our hospital. Neurological examinations on admission revealed no definite abnormality. Her symptoms gradually improved after admission. Plain computed tomography (CT) scan demonstrated a high-density mass with a low-density lesion in the right parietal lobe (Fig. 1A). Both T1- and T2-weighted magnetic resonance imaging (MRI) revealed a mixed-intensity mass with a subacute stage hematoma was located in the right parietal white matter that was very close to the lateral ventricle. The mass was partially enhanced. No other lesions were observed (Figs. 1B–1D). She was diagnoses as having cavernous hemangioma with subacute stage hematoma.

She underwent the resection of mass lesion. Following the induction of general anesthesia, she was placed on a lateral position and a right parietal craniotomy was made. Cerebral cortex was very edematous and swollen. The hematoma was not capsulated and was completely evacuated through a 1-cm corticotomy. Then, the vascular lesion could be identified in the white matter. The mass lesion was reddish in color and was associated with many small feeders and drainers. The mass was completely resected (Fig. 2). Postoperative course was uneventful and she was discharged without any neurological deficits. She is completely free from any neurological events for 8 months after surgery.

Histologic examinations demonstrated a densely grown numerous capillary-like vascular structure with endothelial cells, hemosiderin deposition, and hemorrhage. This histologic finding was consistent with capillary hemangioma (Fig. 3).

**Discussion**

In this adolescent case, preoperative diagnosis was cavernous hemangioma because of the findings on CT and MRI. However, visual inspection during surgery did not fit it, because the hematoma was not capsulated and the mass was reddish in color. Histologic examination strongly supported it. The finding was typical capillary hemangioma. Histologically, capillary hemangioma is unencapsulated mass that are lobular in shape. Microscopically, a single layer of endothelial cells without abnormalities line poorly defined capillary channels. They definitely differ from the more common intracranial cavernous hemangioma insofar as the large dilated, blood-filled vessels lined by flattened endothelium associated with wall thickening due to adventitial fibrosis are absent in capillary hemangioma.⁴ Retrospectively, the findings on MRI was a little bit atypical as cavernous hemangioma. Namely, most of tumor itself was low intensity on both T1- and T2-weighted MRI in this case, while cavernous hemangioma itself usually has mixed intensity signal on

¹Department of Neurosurgery, Graduate School of Medicine and Pharmaceutical Sciences, University of Toyama, Toyama, Toyama, Japan

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As aforementioned, intracranial capillary hemangioma is a very rare entity in the CNS. Only 35 cases have been reported previously. As shown in Table 1, their age was very widely varied from 0 to 82 years. More importantly, most of them (31/35; 88.6%) were found as the extra-axial mass on CT or MRI. Intraoperative findings in 33/35 surgically treated cases proved it. Their origin includes the dura mater in the convexity (n = 9), middle cranial fossa (n = 10), cerebellar tentorium (n = 5), cavernous sinus (n = 3), and petrous bone (n = 1). All of them were well-demarcated and were homogeneously enhanced by the contrast material. On the basis of their locations and, most of them were diagnosed as meningioma before surgery.1,4,6–19 Interestingly, intracranial capillary hemangioma may arise from the ethmoid and sphenoid sinuses, infundibular recess, fourth ventricle, and anterior choroidal artery as an extra-axial mass.20–23 In fact, capillary hemangioma is only presented as an intrasosseous form in the skull in WHO Classification of Tumours of the Central Nervous System Revised 4th Edition.24 On the other hand, intracranial, intra-parenchymal capillary hemangioma is extremely rare, and only four cases have been reported. Thus, Abe et al.31 reported two cases with intracranial intra-parenchymal capillary hemangioma. They had multiple small lesions in the subcortical area. Their age was 16 and 20 years old, respectively, which was very similar to that of present case. They concluded that the capillary hemangioma in the CNS significantly differs from other vascular neoplasms, including hemangiopericytoma and hemangioblastoma, and is benign lesions that can be surgically removed and cured without adjuvant therapy.31 Younas et al.25 (2011) also reported a 69-year-old case with multiple capillary hemangiomas in the brain. All five lesions were hemorrhagic and preoperative diagnosis included metastatic brain tumor and cerebral amyloid angiopathy, but open biopsy revealed capillary hemangioma. John et al.26 reported a 59-year-old case that had a large cystic lesion with enhancing mural nodule in the right temporal lobe. Histological findings were consistent to capillary hemangioma (see Table 1).25 Therefore, radiological findings of intracranial, intra-parenchymal capillary hemangioma may widely vary and be difficult to differentiate capillary hemangioma from other lesions in the brain. This is quite different from these sequences. Furthermore, the hematoma was located beside the tumor in this case, but is usually located within the tumor.25

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Table 1  Summary of previously reported cases of intracranial capillary hemangioma

| Authors          | Year | Age  | Sex | Origin              | Treatment         | Pathology |
|------------------|------|------|-----|---------------------|-------------------|-----------|
| Extra-axial mass |      |      |     |                     |                   |           |
| 1 Willing et al. | 1993 | 1    | M   | Convexity           | Resection         | Yes       |
| 2 Watanabe et al.| 2001 | 8    | M   | Middle cranial fossa| Resection         | Yes       |
| 3 Tsao et al.    | 2003 | 15   | F   | Middle cranial fossa| Radiosurgery      | No        |
| 4 Tsao et al.    | 2003 | 19   | F   | Middle cranial fossa| Radiosurgery      | No        |
| 5 Abe et al.     | 2004 | 8    | M   | Middle cranial fossa| Resection         | Yes       |
| 6 Simon et al.   | 2005 | 31   | M   | Cerebellar tentorium| Resection         | Yes       |
| 7 Le Bihannc et al.| 2005 | 1.5 months | M | Anterior choroidal artery | None         | Yes       |
| 8 Brotchi et al. | 2005 | 10   | F   | Convexity           | Resection         | Yes       |
| 9 Karikari et al.| 2006 | 3    | M   | Fourth ventricle    | Resection         | Yes       |
| 10 Smith et al.  | 2007 | 26   | F   | Middle cranial fossa| Resection         | Yes       |
| 11 Uyama et al.  | 2008 | 4    | F   | Convexity           | Resection         | Yes       |
| 12 Daenekindt et al.| 2008 | 2   | M   | Middle cranial fossa| Resection         | Yes       |
| 3 Maure et al.   | 2010 | 44   | F   | Convexity and middle fossa | Resection         | Yes       |
| 14 Lee et al.    | 2010 | 59   | F   | Infundibular recess | Biopsy            | Yes       |
| 15 Phi et al.    | 2012 | 8    | M   | Convexity           | Resection         | Yes       |
| 16 Ph et al.     | 2012 | 13   | M   | Cerebellar tentorium| Resection         | Yes       |
| 17 Phi et al.    | 2012 | 30   | F   | Cerebellar tentorium| Resection         | Yes       |
| 18 Phi et al.    | 2012 | 44   | F   | Ethmoid and sphenoid sinuses | Resection         | Yes       |
| 19 Morace et al. | 2012 | 26   | F   | Cavernous sinus     | Resection/radiation| Yes       |
| 20 Morace et al. | 2012 | 61   | F   | Cavernous sinus     | Resection/radiation| Yes       |
| 21 Morace et al. | 2012 | 14   | M   | Middle cranial fossa| Resection/radiation| Yes       |
| 22 Morace et al. | 2012 | 42   | M   | Convexity           | Resection         | Yes       |
| 23 Zheng et al.  | 2012 | 3    | M   | Middle cranial fossa| Resection         | Yes       |
| 24 Mirza et al.  | 2013 | 28   | F   | Cerebellar tentorium| Resection         | Yes       |
| 25 Mirza et al.  | 2013 | 41   | F   | Convexity           | Resection         | Yes       |
| 26 Jalloh et al. | 2014 | 0.5 months | M | Middle cranial fossa| Resection         | Yes       |
| 27 Okamoto et al.| 2015 | 82   | F   | Convexity           | Resection         | Yes       |
| 28 Nepute et al. | 2016 | 40   | M   | Petrous bone        | Resection         | Yes       |
| 29 Xia et al.    | 2017 | 33   | F   | Cerebellar tentorium| Resection         | Yes       |
| 30 Low et al.    | 2017 | 64   | F   | Cavernous sinus     | Biopsy            | Yes       |
| 31 Almaghrabi et al.| 2017 | 59 | F  | Convexity           | Resection         | Yes       |
| Intra-axial mass |      |      |     |                     |                   |           |
| 1 Abe et al.     | 2004 | 20   | M   | Subcortical         | Resection         | Yes       |
| 2 Abe et al.     | 2004 | 16   | F   | Subcortical         | Resection         | Yes       |
| 3 Younas et al.  | 2011 | 69   | M   | Subcortical and basal ganglia | Resection         | Yes       |
| 4 John et al.    | 2012 | 59   | M   | Subcortical         | Resection         | Yes       |
| 5 Present case   | 2019 | 15   | F   | Subcortical         | Resection         | Yes       |

intracranial, extra-axial capillary hemangioma that has almost similar findings on CT and MRI.

In the present case, the lesion was completely resected, resulting in complete resolution of the symptoms. As suggested previously, surgical resection would be the best treatment option for intracranial capillary hemangioma presenting with neurological deficits (Table 1).

In conclusion, the authors reported a very rare adolescent case with intracranial, intra-parenchymal capillary hemangioma presenting with repeated headache probably because of increased intracranial pressure after bleeding. To the best of our knowledge, there are only four reported cases with intracranial, intra-parenchymal capillary hemangioma. Differential diagnosis includes other vascular tumors such as cavernous hemangioma, but it is not so easy to differentiate capillary hemangioma from other lesions. Therefore, surgical excision and histologic diagnosis would be important to diagnose it if possible.
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Corresponding author:
Satoshi Kuroda, MD, PhD, Department of Neurosurgery, Graduate School of Medicine and Pharmaceutical Sciences, University of Toyama, 2630 Sugitani, Toyama, Toyama 930-0194, Japan.

*skuroda@med.u-toyama.ac.jp*