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Case Report

Intracranial, Intra-parenchymal Capillary Hemangioma – Case Report –

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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

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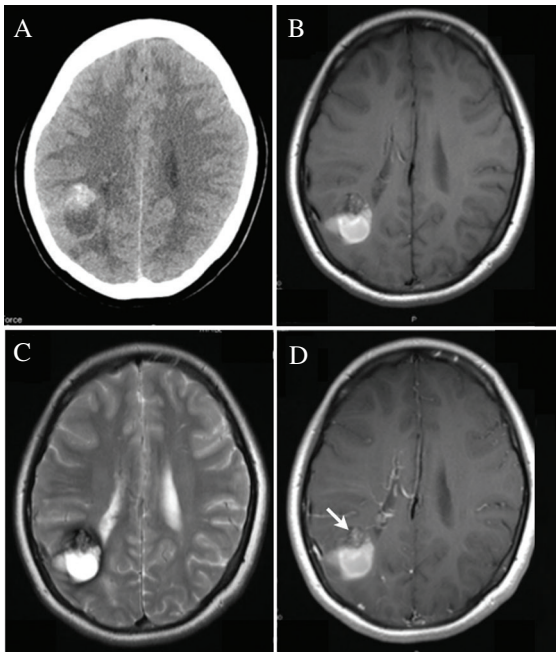

Fig. 1 Preoperative findings on CT and MRI. (A) Plain CT scan showed a high-density mass with low-density cyst in the right deep parietal lobe. The mass demonstrated a mixed signal intensity on both T1- (B) and T2-weighted MRI (C). Adjacent cystic lesion showed high signal intensity on both T1- (B) and T2-weighted MRI (C), suggesting the hematoma in subacute stage. (D) Gadolinium-enhanced T1-weighted MRI revealed a partial enhancement of the mixed signal intensity mass (arrow).

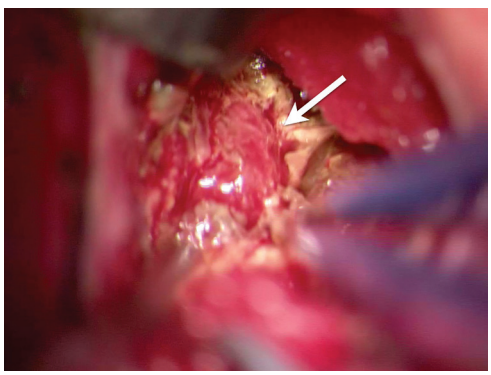

Fig. 2 Intraoperative finding. After the removal of hematoma, the vascular lesion could be identified in the white matter of the right parietal lobe (arrow). The mass lesion was reddish in color and was associated with many small feeders and drainers.

itself usually has mixed intensity signal on these sequences. Furthermore, the hematoma was located beside the tumor in this case, but is usually located within the tumor.[5)]

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 intraosseous 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.[3)] 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.[3)] 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

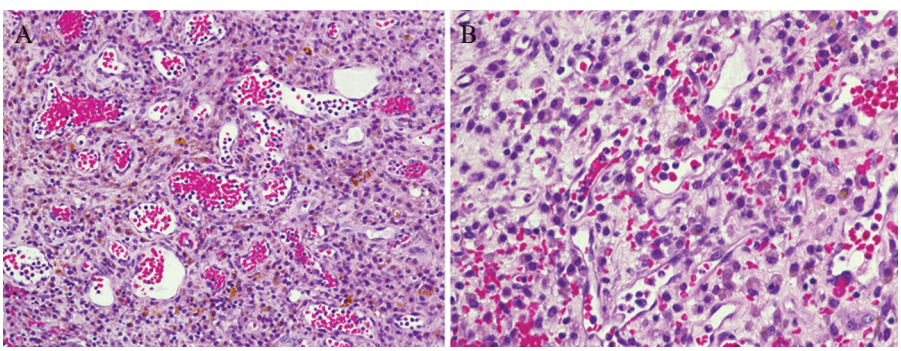

Fig. 3 HE staining of surgical specimen. A densely grown numerous capillary-like vascular structure with endothelial cells, hemosiderin deposition, and hemorrhage was identified, strongly suggesting capillary hemangioma. A) original magnification ×40, B) original magnification ×100.

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

Conflicts of Interest Disclosure

None.

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