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Case Report

Intracranial parenchymal capillary hemangioma: A case report

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ABSTRACT

Background: Capillary hemangioma is a rare benign hemangioma that occurs in the soft tissues of the skin, orbit, head, and neck. Intracranial cases, especially intraparenchymal cases, are extremely rare. In this study, we report the course of an intracranial parenchymal capillary hemangioma with left mild motor paresis and involuntary movements of the left upper extremity and was successfully treated by surgical resection, including radiological and pathological examinations.

Case Description: This is a case of a 60-year-old woman who presented with motor weakness and involuntary movement of the left upper extremity. Computed tomography and magnetic resonance imaging revealed the right frontal hemorrhagic mass lesion without enhancement of contrast medium. Cerebral digital subtraction angiography showed no vascular stain and abnormal arteriovenous shunt. Preoperatively, we diagnosed cavernous hemangioma with a hemorrhagic component located in the right motor cortex. Because this case was symptomatic, we performed a craniotomy and gross total resection of the right frontal lesion. The diagnosis of capillary hemangioma was made by histological examination, including immunohistological study.

Conclusion: Because intraparenchymal capillary hemangiomas are difficult to diagnose with preoperative imaging, surgical treatment, and histopathological examination are important.

Keywords: Capillary hemangioma, Intraparenchymal, Pathological findings, Radiological findings

INTRODUCTION

Capillary hemangioma is an uncommon, benign vascular neoplasm that manifests within the subcutaneous tissues of the integumentary system, orbit, cranium, and cervicocephalic region. The reported incidence of this condition in neonates ranges from 1.1% to 2.6%, but intracranial presentations are exceedingly rare occurrences.[1,6] In this instance, we present an exceptional case of intracranial parenchymal capillary hemangioma in a patient exhibiting weakness and involuntary movement of the left upper extremity who underwent surgical intervention. We hereby provide a comprehensive analysis of the radiological and pathological findings associated with this case, in addition to a thorough review of the literature.

CASE REPORT

A 60-year-old female patient with no remarkable medical history was admitted to our hospital due to the presence of involuntary movements and weakness in her left upper extremity. Upon examination, her level of consciousness was Glasgow Coma Scale: E4V5M6, and she exhibited weakness, specifically in her left upper extremity. A computed tomography (CT) scan revealed a high-density area (3 mL) located in the right motor cortex, suggesting subcortical hemorrhage [Figure 1a]. Contrast-enhanced CT revealed no enhanced lesion [Figure 1b]. Magnetic resonance imaging (MRI), T1 weighted and T2 weighted images displayed a mass with mixed intensity and the abscense of surrounding flow void [Figures 1c and d], and T1 weighted images wigh Gadolinium contrast exhibited a heterogenous intensity mass with surrounding with surrounding contrast enhancement [Figures 1e-g]. No other abnormalities were observed in other regions. Cerebral digital subtraction angiography (DSA) indicated the absence of vascular staining, abnormal arteriovenous shunts, or developmental venous malformations adjacent to the subcortical hemorrhagic lesion [Figure 2]. Based on these findings, we established a diagnosis of cavernous hemangioma with hemorrhagic change. Despite the lesion being small and located within an eloquent area, it was symptomatic, and the presence of prolonged cerebral edema on imaging led us to decide on surgical intervention. The patient underwent a right frontal craniotomy under general anesthesia. Upon exposure of the cortex, edema, and swelling were observed, and the brain surface was pale above the lesion [Figure 3a]. A 15-mm cortical incision was made, revealing a reddish, nonencapsulated mass lesion with hemorrhagic change. The border between the white matter and the lesion was well-defined, allowing for complete tumor resection [Figures 3b-e]. The excised specimen exhibited a reddish-black color [Figures 3e and f]. Upon inspection of the lesion, no mulberry formations were identified [Figure 3f], suggesting that this lesion might differ from cavernous hemangioma. Postoperatively, the patient experienced no complications, and her left upper extremity function recovered to a level of practical usefulness. She was discharged from our hospital on the 17th day following the surgery. Histopathological examination of the lesion revealed the presence of capillary-like vascular structures with thin-walled endothelial cells displaying irregular anastomoses and areas of hemorrhage [Figures 4a and b]. There was no blood filling within the vessels, as seen in the histopathology of cavernous hemangiomas, and the extravascular area consisted of thick connective tissue. Immunostaining reveals numerous CD31 and CD34-positive capillary endothelial cells. Perivascular cells were

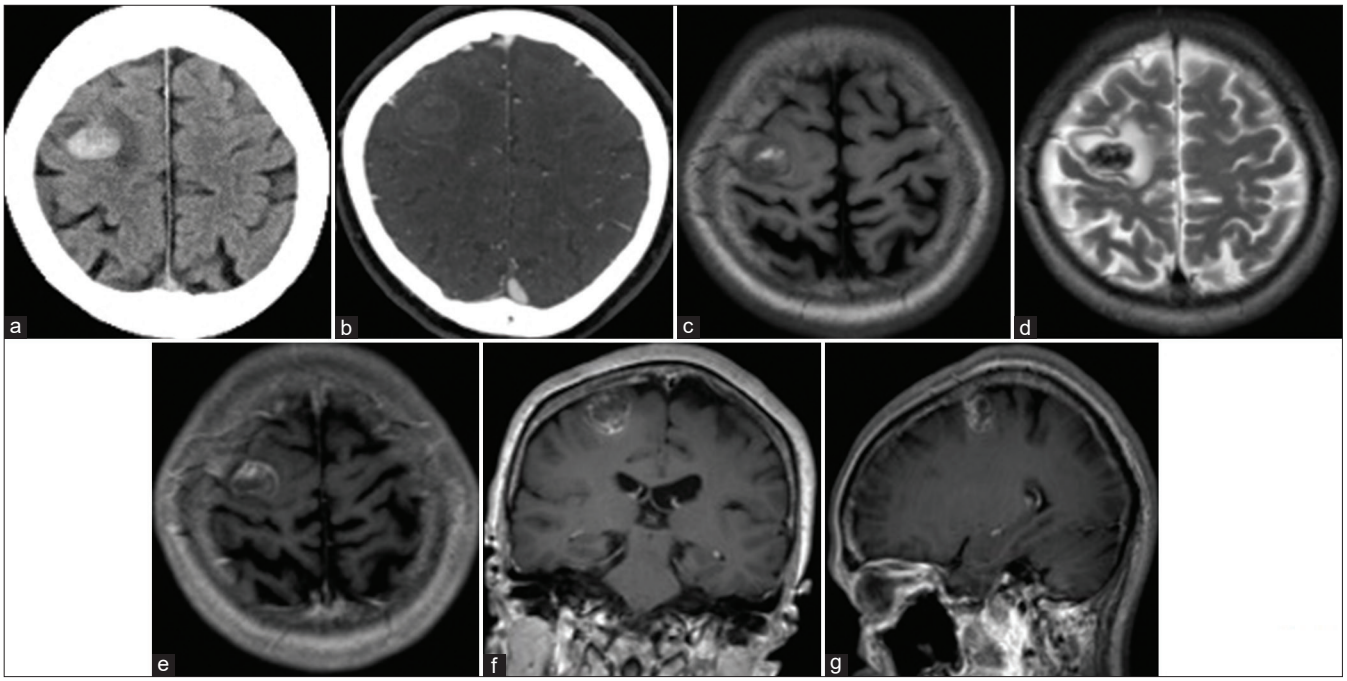

Figure 1: Head computed tomography (CT) and brain magnetic resonance imaging (MRI). (a and b) CT showed that a high-density lesion in the right precentral gyrus indicates intraparenchymal hemorrhage without contrast enhancement. (c and d) MRI T1 and T2-weighted images showed that heterogeneous intensity mass lesion at the precentral gyrus consists of hemorrhagic change. (e-g) MRI T1-weighted image with gadolinium (e: axial view, f: coronal view, g: sagittal view) exhibited a heterogeneous intensity mass with surrounding contrast enhancement. There was no flow void around the lesion.

positive for smooth muscle actin (SMA). The p53 was negative, and the Ki67 positivity was low (<3%), consistent with a benign hemangioma [Figures 4c-j]. The epithelial cell marker, cytokeratin (AE1/AE3), is negative, ruling out metastatic clear cell carcinoma. The intercapillary cells were negative for inhibin, epithelial membrane antigen (EMA), and glucose transporter-1 (GLUT1) (red blood cells were positive), excluding hemangioblastoma, meningioma, and hemangiopericytoma [Figures 5a-h].

DISCUSSION

Capillary hemangioma represents the most prevalent benign vascular neoplasm affecting the integumentary system and mucous membranes. Typically observed in neonates, the majority of these lesions undergo spontaneous regression by the age of 5 years.[3] On the other hand, intracranial cases are extremely rare, and according to our survey, a total of 48 cases have been reported in adults and pediatrics.[7,8] Of these, only five adult cases have occurred within the brain parenchyma.[7] No obvious gender difference was found. While capillary hemangiomas are commonly observed in neonates and progress from childhood to puberty, intracranial capillary hemangiomas have been reported across a broad age range from 0 to 82 years, with an average age of 22.0 years. In the case of intraparenchymal lesions in adults, patients range in age from 20 to 82 years old, with a mean age of 45.2 years. Symptoms commonly reported include cranial nerve impairment, intracranial hypertension, and epileptic seizures. In our case, the patient experienced motor paresis and epileptic involuntary movements. CT and MRI examinations often lack specific findings but frequently reveal cyst formation, multiple signal voids, and intratumoral hemorrhage accompanied by surrounding cerebral edema. Contrast-enhanced MRI typically demonstrates homogeneous enhancement, although our case exhibited only a very faint enhancement. Cerebral DSA often reveals tumor staining. However, our case did not exhibit the typical imaging findings described above. Notably, none of the previously reported cases of intraparenchymal brain lesions displayed the typical imaging findings observed in the present case. In all such cases, as in our case, the preoperative diagnosis was cavernous hemangioma, metastatic brain tumor, or hemangioblastoma, which made it very difficult to differentiate them from other intraparenchymal lesions.[1,5,10]

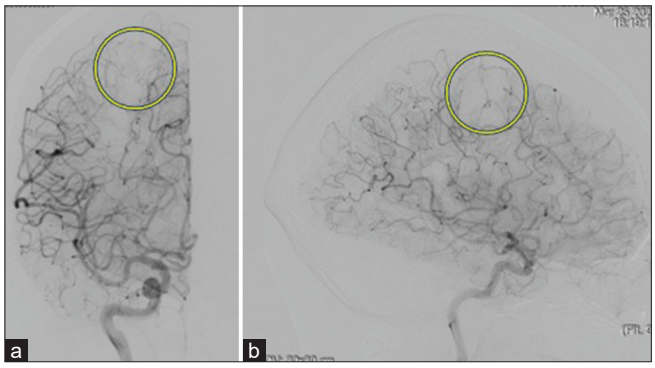

Figure 2: Digital subtraction angiography (DSA). (a) Anteroposterior view of the right internal carotid artery (ICA), (b) Lateral view of right ICA, DSA showed a vascular area consisting of a lesion (yellow circle) and no abnormal vascular structure or tumor stain.

Although regression of residual lesions has been reported following systemic administration of steroids and

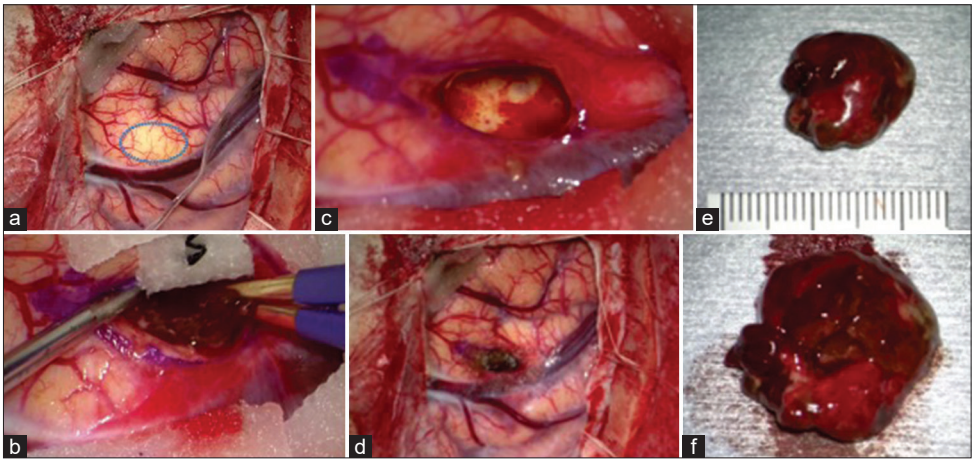

Figure 3: Intraoperative findings. (a) Upon exposure of the cortex, edema, and swelling were observed, and the brain surface was pale above the lesion (blue circle). (b-d) *En-bloc* resection was performed. We could see a nonencapsulated mass lesion with hemorrhagic change (b), and the border between the white matter and the lesion was well-defined (c). (e and f) The specimen exhibited a reddish-black color; upon inspection of the lesion, no mulberry formations were identified, suggesting that this lesion might differ from cavernous hemangioma.

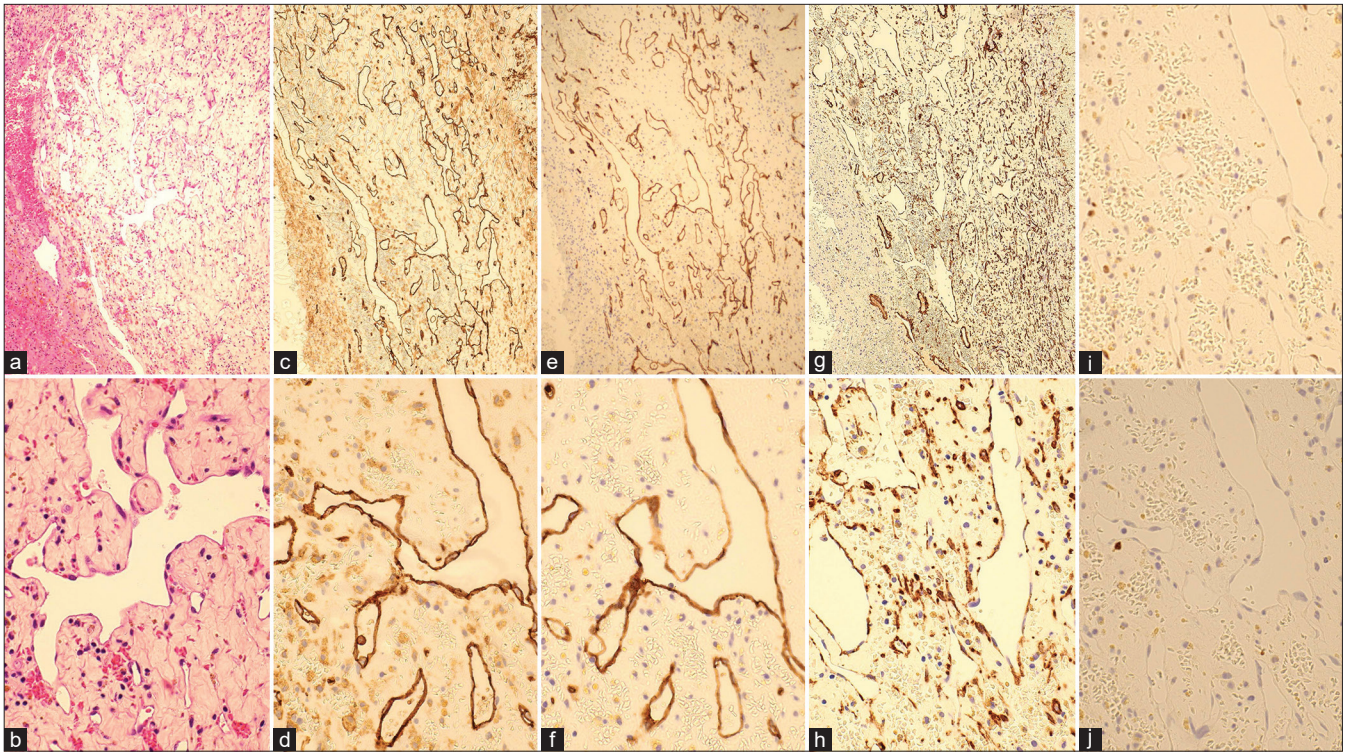

Figure 4: Micrographs of hematoxylin-eosin (HE) and immunohistochemical findings (CD 31, CD 34, SMA, Ki-67, p53). (a and b) HE staining ×10, ×40: capillary-like vascular structures with thin-walled endothelial cells displaying irregular anastomoses. There was no blood filling within the vessels as seen in the histopathology of cavernous hemangiomas, and the extravascular area consisted of thick connective tissue; (c and d) immunohistochemical staining: CD 31 ×10, ×40; (e and f) immunohistochemical staining: CD 34 ×10, ×40; (g and h) immunohistochemical staining: SMA ×10, ×40; (i) p53; (j) Ki-67. Immunostaining demonstrated positive expression of CD31 and CD34, indicative of endothelial cells forming irregular lumens.

interferon alpha, surgical excision represents the most common treatment approach. Hemorrhage from partially excised lesions has been documented in some cases, so total resection of the tumor was recommended.[9] As for the natural course of intracranial capillary hemangioma, there are limited reports, and to the best of our knowledge, no cases have been followed up for longer than 41 months.[4] Recurrence has not been reported among patients who underwent excisional surgery, and the favorable improvement of symptoms in many cases suggests a relatively positive prognosis. However, Abe *et al*. reported that the apoptosis index of intracranial capillary hemangioma is significantly lower than that of cutaneous capillary hemangioma, implying a low likelihood of spontaneous regression of intracranial capillary hemangioma.[2] Thus, whenever feasible, surgical excision of the capillary hemangioma might prevent unnecessary interventions and lead to a relatively favorable outcome. However, in cases such as the present one, where the capillary hemangioma develops within the brain parenchyma and lacks preoperative imaging findings suggestive of a capillary hemangioma, formulating a treatment strategy centered on complete surgical excision becomes exceedingly difficult. Histologically, the findings in our case strongly support the diagnosis of capillary hemangioma. Microscopically, the presence of a single layer of endothelial cells positive for CD31 and CD34 defines the capillary channels. Importantly, it is crucial to differentiate capillary hemangioma from intracranial cavernous hemangiomas, which are characterized by large dilated vessels filled with blood cells lined by flattened endothelium. In our case, there was no blood component filling the vascular endothelial structures as seen in cavernous hemangiomas, but rather a marked thickening of the interstitial tissue. This might be one of the histopathological findings of capillary hemangioma within the brain parenchyma. Due to the difficulties associated with preoperative diagnosis and the similarity of histopathologic findings, numerous cases of capillary hemangioma may be pathologically diagnosed as cavernous hemangiomas. There is no clear distinction in terms of histopathology or prognosis between capillary and cavernous hemangiomas. We anticipate the accumulation of further cases in the future.

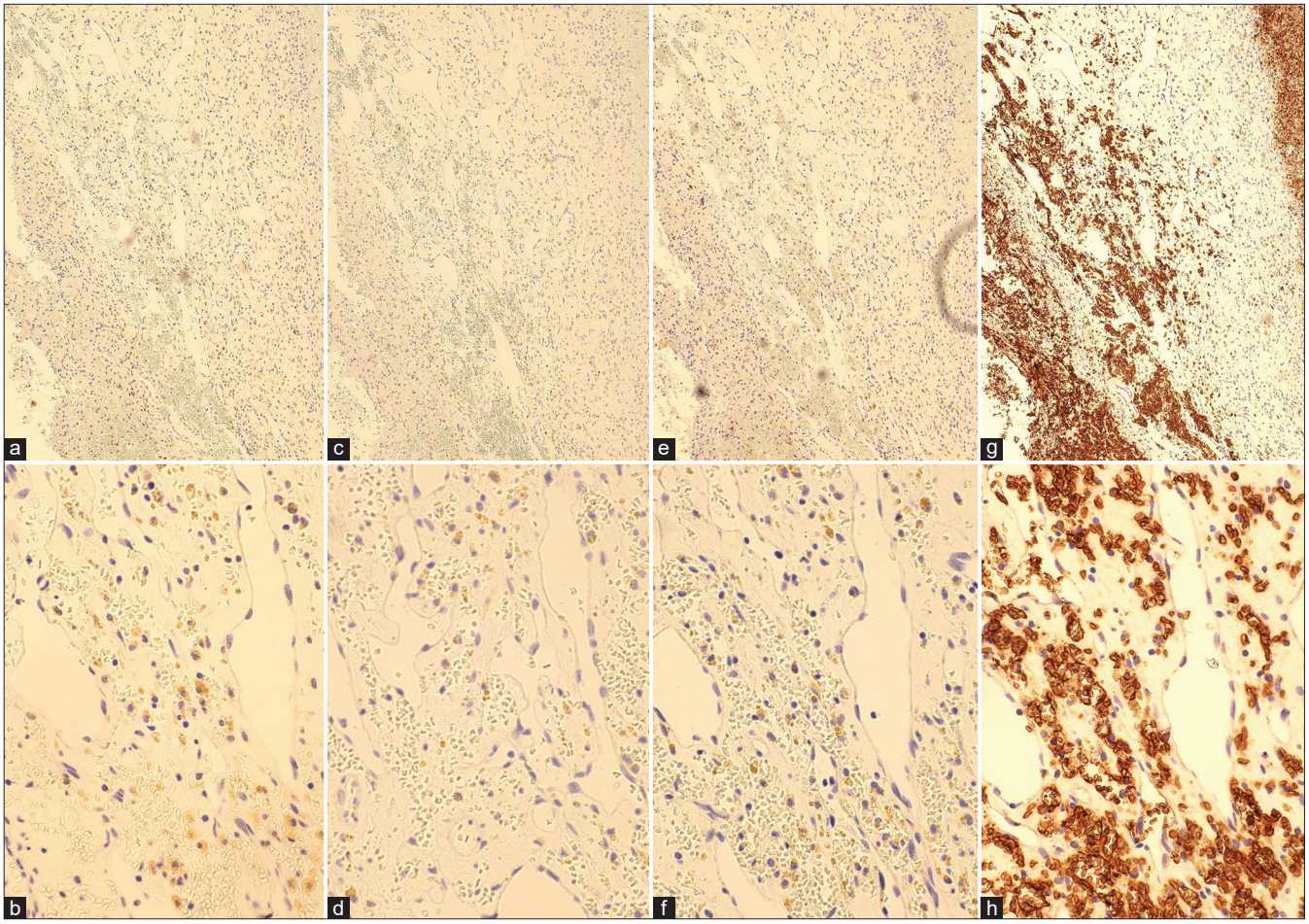

Figure 5: Immunohistochemical findings (CD 31, CD 34, SMA, Ki-67, p53). (a and b) Cytokeratin AE1/AE3 ×10, ×40; (c and d) Inhibin ×10, ×40; (e and f) EMA ×10, ×40; (g and h) GLUT-1 ×10, ×40.

CONCLUSION

Capillary hemangioma within cerebral parenchyma is very rare, especially in adults. For symptomatic lesions, surgical treatment is a valid option for histopathological characterization and neurological improvement. The etiology of intraparenchymal capillary hemangioma has not been established, and more cases need to be accumulated.

Authors contributions

Kosuke Sasaki: Writing and editing; Atsushi Kuge (Corresponding author): Conceptualization, editing; Yu Shimokawa: Data curation, investigation; Tetsu Yamaki : Data curation, investigation; Rei Kondo: Methodology, editing; Yukihiko Sonoda: Supervision.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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