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## **Atypical Epidermoid Cyst With Repetitive Hemorrhages in the Supracallosal Region**

### **—Case Report—**

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### **Abstract**

A 63-year-old man presented with an unusual supracallosal epidermoid cyst with repetitive hemorrhages that initially manifested as severe headache in 2003. Physical examination found no neurological deficit. Computed tomography demonstrated a homogeneously high density mass lesion measuring  $3 \times 3$  cm in the supracallosal area, and the clinical diagnosis was cavernous malformation. No change occurred in neurological findings except for headache, but the lesion underwent hemorrhages several times over a 4-year period and increased in size to  $4 \times 5$  cm. Surgery revealed a pearly cyst filled with old blood and debris. Histological examination disclosed keratinizing, stratified squamous epithelium without hair follicles or sebaceous glands, consistent with the diagnosis of epidermoid cyst. Surgery was successfully performed without complications such as akinetic mutism, motor weakness, or chemical meningitis. The present case suggests that epidermoid cyst should be considered in the differential diagnosis of suspected cavernous malformation in any location.

Key words: epidermoid cyst, hemorrhage, supracallosal region

## Introduction

Epidermoid cyst is a rare tumor which accounts for approximately 1% of intracranial tumors.<sup>11,12</sup> Epidermoid cysts are usually congenital tumors, and are thought to arise from misplaced epithelial elements that have become trapped in the meninges, ventricles, or brain parenchyma. Rarely, epidermoid tumors are caused by iatrogenic puncture.<sup>5,8,9</sup> Epidermoid cysts tend to arise in paramedian locations such as the cerebellopontine angle, suprasellar, and ventricular regions, and more rarely in the thalamus, middle cranial fossa, brain stem, and callosal region.<sup>2,4,7,12,13</sup> Spontaneous hemorrhage is rare due to the avascular nature, and reported cases of intracranial epidermoid cyst with hemorrhage were preferentially located in the infratentorial compartment.<sup>3,10</sup>

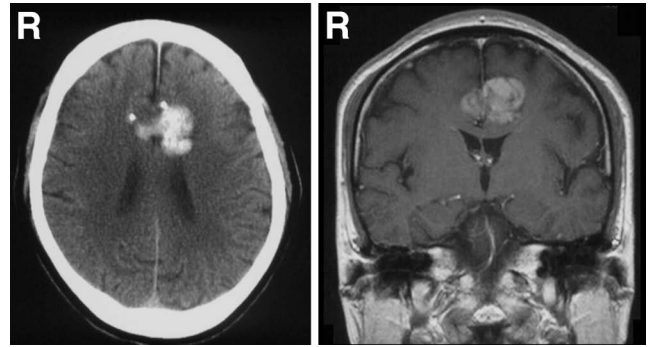
We describe a case of supracallosal epidermoid cyst presenting with repetitive hemorrhages.

## Case Report

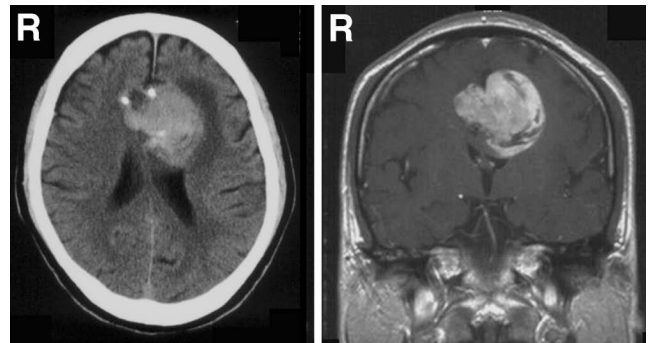
A 63-year-old man presented with continuous headache as the only symptom in April 2003. Physical examination found no neurological deficit. Computed tomography (CT) demonstrated a hyperdense multiple nodular mass of 3 × 3 cm in the supracallosal area, containing two calcification spots (Fig. 1 left). T<sub>1</sub>-weighted magnetic resonance (MR) imaging showed the lesion as a well-defined heterogeneously hyperintense intra-axial mass, and T<sub>2</sub>-weighted MR imaging as a homogeneously hypointense mass. The lesion was not enhanced with contrast medium (Fig. 1 right). The most likely clinical diagnosis at this point was considered to be cavernous malformation, although the possibility of extra-axial tumors such as meningioma could not be completely ruled out. The patient was conservatively observed because he refused consent to undergo surgical treatment. However, the mass became larger with repetitive hemorrhages over a 4-year period. Therefore, he was admitted for further evaluation and treatment in August 2007.

On admission, CT demonstrated a heterogeneously hyperdense mass of 4 × 5 cm in the same area, and associated with calcification at the bottom (Fig. 2 left). Most of the mass was homogeneously hyperintense on T<sub>1</sub>-weighted imaging, and hypointense on T<sub>2</sub>-weighted imaging. There were spotty areas which were hypointense on T<sub>1</sub>-weighted imaging and hyperintense on T<sub>2</sub>-weighted imaging. The mass was not enhanced with contrast medium (Fig. 2 right). Angiography showed no evidence of vascularity in the lesion.

The patient underwent surgery for hematoma removal via an anterior interhemispheric approach. The hematoma was visible through the parenchyma of the cingulate gyrus, and the tumor was located intra-axially in the cingulated gyrus. The boundary of the tumor and the brain parenchyma could not be clearly identified in the upper part of the tumor, whereas the mass was surrounded by a characteristic pearly colored capsule at the bottom. The capsule and the old clot and debris inside were totally removed except for the bottom part which was firmly adher-



**Fig. 1** Computed tomography scan (left) showing a hyperdense multiple nodular mass of 3 × 3 cm in the supracallosal area and two calcified spots. Coronal T<sub>1</sub>-weighted magnetic resonance image with contrast medium (right).



**Fig. 2** Computed tomography scan (left) and coronal T<sub>1</sub>-weighted magnetic resonance image with contrast medium (right) showing enlargement of the mass lesion to 4 × 5 cm.

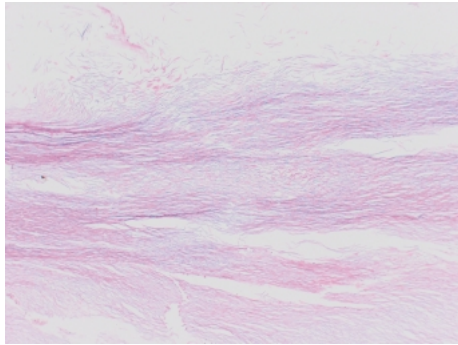
ent to the anterior cerebral arteries (ACAs) with calcification.

The histological specimen was obtained from the bottom part of the tumor containing calcification and with firm adhesion to the ACAs. Histological examination revealed that the cyst wall consisted of keratinizing, stratified squamous epithelium without hair follicles or sebaceous glands (Fig. 3). The fibrous nodule consisted of hyalinized fibrous tissue accompanied by hemosiderin pigmentation. Based on these findings, the diagnosis was epidermoid cyst. Fresh red blood cells, hemosiderin, and other breakdown products of hemoglobin were found in the specimen. In addition, the specimen was rich in vessels (Fig. 4). Interestingly, capillaries and arteries were seen within the specimen.

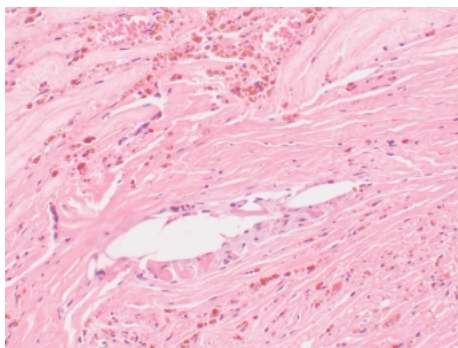
The patient's postoperative course was uneventful. He was discharged home without neurological deficits. At the follow-up examination at 1 year after surgery, the patient was free from recurrence or hemorrhage, as confirmed by CT and MR imaging.

## Discussion

Epidermoid cysts are well-defined lesions surrounded by a



**Fig. 3** Photomicrograph showing the cyst wall consisting of keratinizing, stratified squamous epithelium. Hematoxylin and eosin stain, original magnification  $\times 100$ .



**Fig. 4** Photomicrograph showing hemosiderin and capillary vessels surrounded by granulation tissue. Hematoxylin and eosin stain, original magnification  $\times 100$ .

pearly colored capsule. The cyst wall consists of a layer of stratified squamous epithelium. The cysts are filled with desquamated epithelial cells consisting mainly of keratin in concentric layers and cholesterol. Epidermoid cysts appear as well-circumscribed lobulated hypodense masses which resemble arachnoid cysts in most cases on CT. Epidermoid cysts typically appear as hypointense on  $T_1$ -weighted imaging, and hyperintense on  $T_2$ -weighted imaging similar to cerebrospinal fluid, with no enhancement by gadolinium. In addition, epidermoid cysts typically appear as hyperintense on diffusion-weighted images, and the apparent diffusion coefficient value is high.<sup>6)</sup> In our case, the mass was hyperintense on  $T_1$ -weighted imaging and hypointense on  $T_2$ -weighted imaging, which might represent a hematoma with methemoglobin.

In addition to the atypical radiographical features of the present epidermoid cyst, the clinical course over 4 years was unusual. The growth rate of an epidermoid tumor may be one generation per month, essentially the same as the turnover time of normal human skin.<sup>1,12)</sup> In contrast, the present case demonstrated rapid enlargement of the tumor associated with hemorrhage. Histological examination showed hemosiderin and breakdown products of hemoglobin in the tumor, coinciding with the findings of

the CT and MR imaging. Our case had moderate vascularity with capillaries and arteries, as found in a previous case.<sup>3)</sup> Such vascularity may have contributed to the repetitive hemorrhages and growth of the tumor. In addition, epidermoid tumors usually tend to adhere to neurovascular structures including the cranial nerves, perforating arteries, veins, and brain stem.<sup>11)</sup> We speculate that the tumor in our case arose from the brain parenchyma of the cingulate gyrus, extended outward to subarachnoid space, and resulted in the calcification and the firm adhesion at the bottom of the tumor.

The present case was unequivocally shown to be supracallosal epidermoid cyst with repetitive hemorrhages. Surgeons should consider that epidermoid cysts can cause hemorrhage and exhibit atypical radiological features, as demonstrated in our case, to avoid surgical complications such as chemical meningitis and to increase the success rate.

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