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Case report

Capillary hemangioma of the sphenoid sinus with intrasellar and parasellar extensions [☆]

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ABSTRACT

Capillary hemangiomas are benign vascular tumors usually diagnosed in infancy and involving skin and soft tissues. We report a rare case of an adult capillary hemangioma of the left sphenoid sinus extending into the intrasellar and parasellar regions. A 71-year-old woman presented with complaints of headaches and diplopia. Magnetic resonance imaging revealed a T2-hyperintense mass-like lesion involving the left posterior ethmoid and sphenoid sinuses and left cavernous sinus, that had extended into the intrasellar and parasellar regions. Dynamic contrast-enhanced computed tomography revealed enhancing vessel-like structures within the lesion during the arterial phase, which were observed to expand further in the late phase of the examination. This characteristic feature indicated increased vascularity, suggesting the presence of a capillary hemangioma. The patient underwent endoscopic resection via the endonasal transpterygoid approach, and the lesion was diagnosed histopathologically as a capillary hemangioma. To aid diagnosis and clinical management, radiologists should be aware of the imaging findings related to capillary hemangiomas.

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Introduction

Capillary hemangiomas are benign vascular tumors that involve the skin and soft tissues and are usually diagnosed in in-

fancy [1]. Some rare cases may display central nervous system involvement, and the most common locations are the spinal cord and cauda equina [2]. Contrary to cavernous hemangiomas, intracranial capillary hemangiomas are extremely rare; only 53 cases have been reported in the literature to date.

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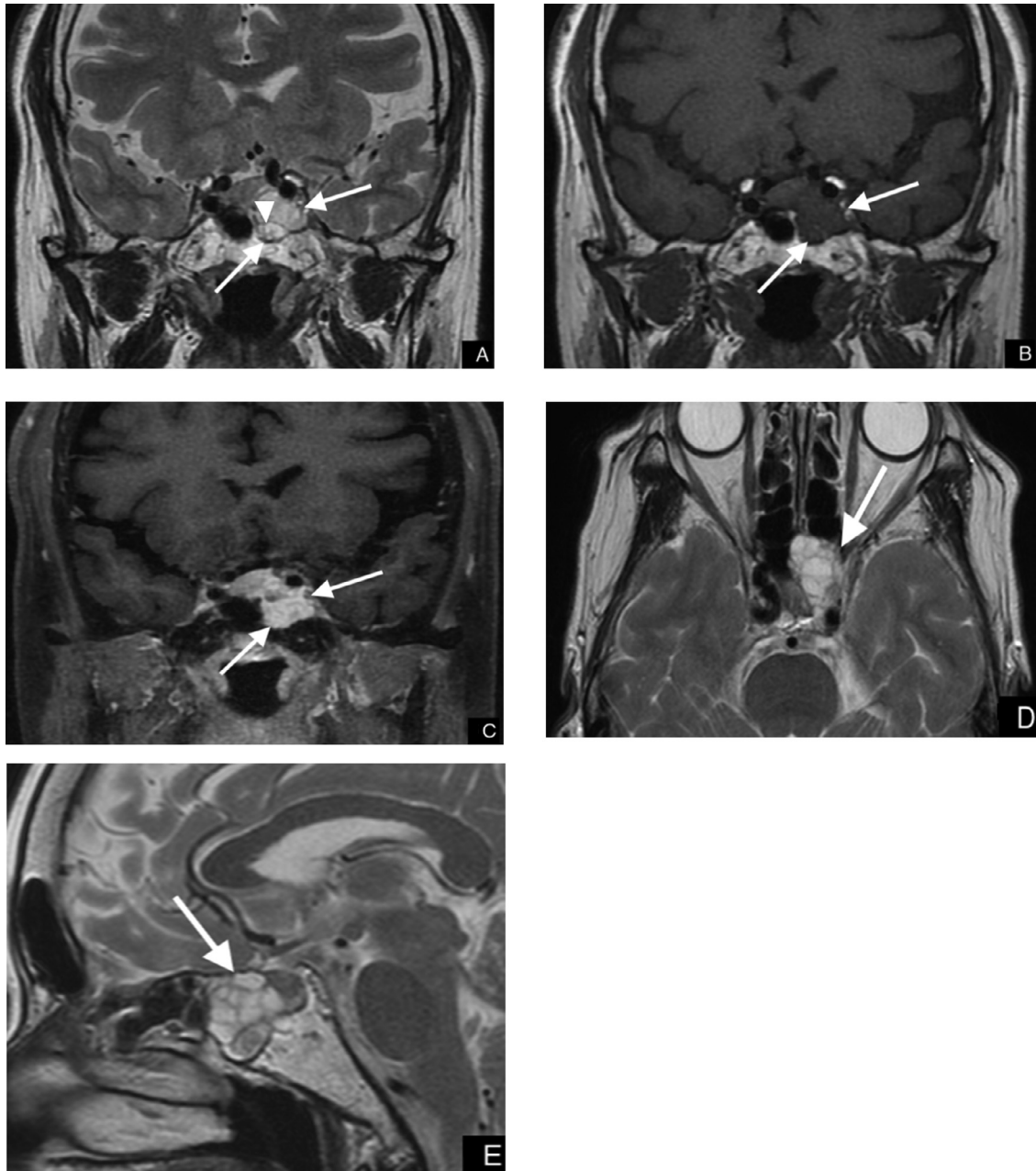


Fig. 1 – On magnetic resonance imaging (MRI) scan, the tumor (arrows) displayed marked hyperintensity and hypointense, thin internal septations (arrowhead) on T2-weighted images (1A). T1-weighted images revealed a uniformly hypointense mass (1B). Almost the entire lesion was homogeneously enhanced after gadolinium administration on fat-suppressed T1-weighted images (1C). The relationship of the tumor (arrow) with the pituitary gland as well as surrounding sinuses and structures is revealed on transverse and sagittal images (1D and 1E respectively).

We found a total of 8 previous studies of ten patients that reported lesions involving the cavernous sinus [3–10]. Among them, Phi et al. [7] reported a capillary hemangioma that involved the ethmoid and sphenoid sinuses and extended into the cavernous sinus. Here, we present and discuss the radiographic features of a rare case of an adult capillary hemangioma of the left sphenoid sinus with extensions into the intrasellar and parasellar regions that were detected using multimodality imaging.

Case report

A 71-year-old woman with complaints of headaches and diplopia was referred to our hospital after the detection of a lesion in the parasellar region on computed tomography (CT). The patient's medical history included hypertension, diabetes mellitus, colon cancer, and episodes of depression. Physical and neurological examinations confirmed

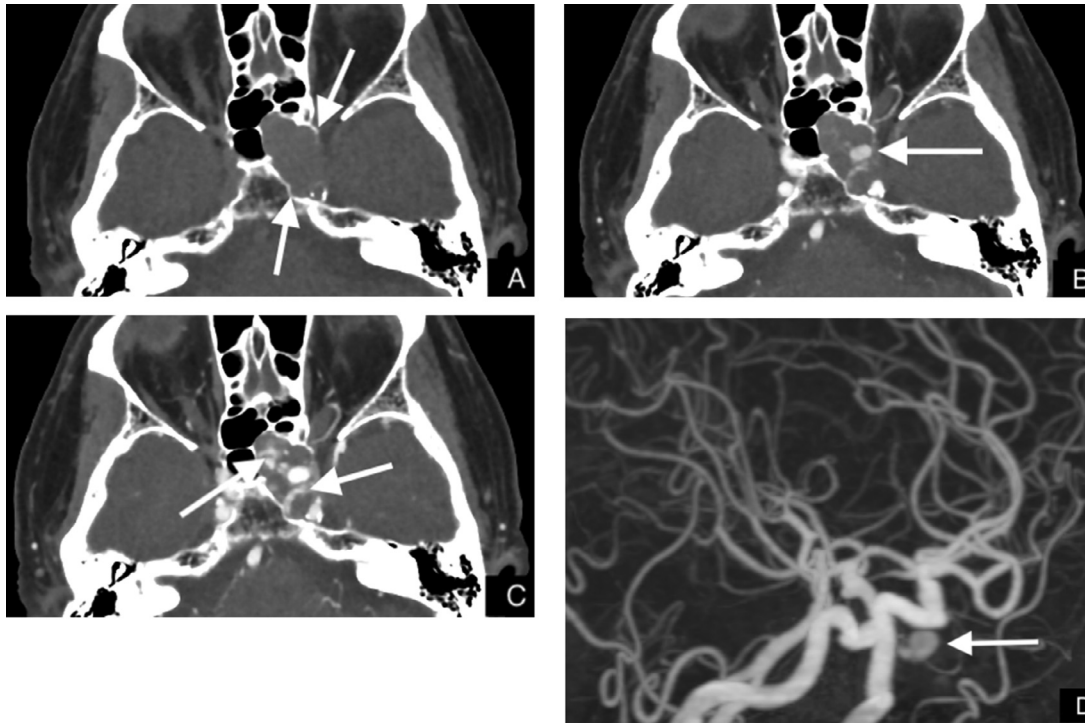


Fig. 2 – Computed tomography angiography (CTA) scan revealed osteolysis of the walls of sphenoid sinus and clivus with osteosclerotic, scalloping edges (arrows, 2A). An enhancing vessel-like structure was observed within the lesion during the arterial phase (arrow, 2B), which was observed to expand further (arrows, 2C) in the later phase of the investigation. 3D CTA image shows pooling of contrast agent (arrow, 2D), and vessel continuity could not be visualized.

diplopia. Laboratory tests showed elevated prolactin levels (192.4 ng/ml).

Magnetic resonance imaging (MRI) revealed a mass-like lesion involving the left posterior ethmoid and sphenoid sinuses and left cavernous sinus, extending into the intrasellar and parasellar regions (Figs. 1A-E). The lesion was observed to be hypointense on T1-weighted images and presented a marked hyperintensity with hypointense, thin internal septal structures on T2-weighted images. Diffusion-weighted imaging did not reveal any diffusion restriction. The apparent diffusion coefficient values ranged between 1.1 and 1.4×10^{-3} mm²/s. Almost the entire lesion was homogeneously enhanced on T1-weighted images after gadolinium administration.

Unenhanced CT images revealed osteolysis of the walls of the sphenoid sinus and clivus with osteosclerotic, scalloping edges, without internal calcifications. The subsequent dynamic contrast-enhanced CT scan revealed enhancing vessel-like structures within the lesion during the arterial phase, which were observed to expand further in the later phase of the investigation (Figs. 2A-D).

The patient underwent endoscopic resection using the endonasal transpterygoid approach. Histopathological examination revealed capillary-like structures (Fig. 3), with an immunostaining profile of a hemangioma. Based on the clinical, radiographic, and histopathological findings, the lesion was diagnosed as a capillary hemangioma. In retrospect, the high prolactin levels were attributable to the adverse effects of antipsychotic therapy with sulpiride (SULPIRIDE CAPSULES

50mg, Towa Pharmaceutical Co, Ltd, Japan; administration: 50 mg twice a day).

Following resection, the patient underwent neurosurgical observation. Three months after the surgery, a follow-up contrast-enhanced MRI scan revealed an enhancing region of postoperative changes in the left cavernous sinus, which did not increase in size on subsequent imaging studies. The patient continues to be under observation.

Discussion

Intracranial capillary hemangiomas can present as both intra-axial and extra-axial lesions. Extra-axial lesions are more common, with convexities, the middle cranial fossa, and cavernous sinuses as their most prominent locations [10,11]. Contrary to typical capillary hemangiomas involving the skin and/or soft tissues, intracranial lesions are diagnosed in both infants and adults. The age distribution of intracranial capillary hemangiomas displays a male predilection in the age group of 0-10 years and a female predilection in the age group of 20-80 years (our patient was 71 years of age). The statistically significant median age with reference to the male and female patients is 9 and 30 years, respectively [10]. The most frequent clinical symptoms associated with these intracranial lesions include headache and visual disturbances. However, previous studies have also described asymptomatic cases and patients with a wide spectrum of neuro-

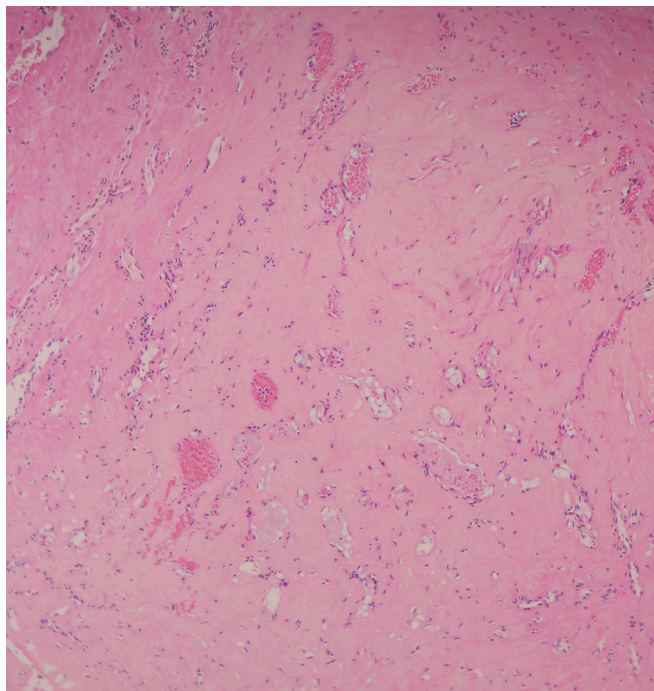


Fig. 3 – Hematoxylin and eosin staining (x 100) of the tissue specimen of capillary hemangioma. Histopathological examination revealed the presence of vessels of different sizes within the lesion, some of them partially filled with erythrocytes.

logical disorders [9]. Capillary hemangiomas are considered benign, but the confines of the skull and the importance of surrounding structures often favor surgical resection [10]. Other options for the management of the condition include fractionated stereotactic radiotherapy, endoscopic biopsy, and embolization [9].

The characteristic MRI findings pertaining to capillary hemangiomas are mass-like lesions that appear hypointense on T1-weighted images, hyperintense on T2-weighted images and show no diffusion restriction. They are comparable to the radiographic features of cavernous hemangiomas [12]. Occasionally, the radiographic appearance mimics meningiomas [7]. Capillary hemangiomas display enhancement after contrast administration, and some cases may present with a peripheral enhancement pattern [13]. Avid enhancement on post-contrast T1-weighted images and hyperintensity with flow voids on T2-weighted images indicate high vascularity and strongly suggest capillary hemangioma [14], although flow voids have not been reported in all cases. In addition to bright T2 signal, T2 dark septa between lobules were observed in our patient. This appearance has been reported previously [15] and is considered to be a characteristic feature of capillary hemangiomas. However, similar appearance is also observed in chordoma and chondroma/chondrosarcoma [16], and diagnosis based on MRI images alone can be challenging.

The CT findings related to capillary hemangiomas include hyperdense masses with or without surrounding vasogenic edema, scalloping pattern of bone erosion, which was observed in our case, and, rarely, internal cysts [14]. Suss et al. described an irregularly mineralized capillary hemangioma of the sphenoid bone [17], but reviewing previous literature

we found no confirmation that internal calcifications could be suggestive of intracranial capillary hemangiomas.

Capillary hemangiomas are reported to exhibit centripetal enhancement on conventional angiography [14]. Conversely, in the present case, dynamic contrast-enhanced CTA revealed enhancing vessel-like structures within the lesion during the arterial phase that were observed to expand further in the later phase of the examination. Although this observation has not been reported as a characteristic radiographic feature of the condition in previous literature, it seems to indicate the highly vascular nature of capillary hemangiomas. Moreover, this feature has not been observed in relation to cavernous hemangiomas, which display similar MRI findings [12].

Conclusion

Intracranial capillary hemangiomas are mostly extra-axial, rare, benign tumors that are usually located near the convexities, middle cranial fossa, and cavernous sinuses. The lesions can be observed in patients of any age, with a predilection for male children and female adults. Preoperative diagnosis of the condition can be challenging, since imaging features are similar to those of cavernous hemangioma, chordoma, and chondroma/chondrosarcoma. In our case, the capillary hemangioma mainly occupied the left sphenoid sinus and extended into the intrasellar and parasellar regions. Dynamic contrast-enhanced CT revealed enhancing vessel-like structures within the lesion during the arterial phase, which were observed to expand further in the later phase of the investi-

gation. This characteristic feature indicated increased vascularity and suggested the presence of a capillary hemangioma.

Patient consent

Written informed consent was obtained from the patient.

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