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Progressive Tentorial Cavernous Malformation

Case Report

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ABSTRACT

Background: Because extra-axial cavernous malformations (CMs) are rare, the common clinical course remains unclear. We report the case of a patient with progressive CM originating from the cerebellar tentorium.

Case Description: A 64-year-old woman was admitted to our hospital with a complaint of diplopia. Magnetic resonance (MR) imaging revealed a lesion attached to the left cerebellar tentorium, close to the cerebral peduncle. This well-demarcated lesion rapidly enlarged for 3 months and eroded into the midbrain. Cerebral angiography showed a branch of the middle meningeal artery supplying the lesion and pooling of the contrast medium in the venous phase. A dark-reddish and mulberry-like mass of the tentorium was observed intraoperatively, allowing diagnosis of a tentorial CM. The feeding artery was identified in the tentorium and was coagulated. Postoperative MR imaging showed remarkable mass reduction and central necrosis of the lesion. However, the lesion recurred in 3 months; consequently, gamma knife radiosurgery was performed. After additional 2 months, the lesion shrank in response to the radiosurgery.

Conclusions: We report an extremely rare case of tentorial CM, which showed rapid growth in a short period. Coagulation of the feeding artery was not sufficient to control the lesion. Gamma knife radiosurgery may prove highly effective for recurrent lesions.

Key Words: cavernous malformation, cerebellar tentorium, gamma knife

INTRODUCTION

Cavernous malformations (CMs), also known as cavernous angiomas or cavernomas, account for 3–13% of all intracranial cerebral vascular malformations^[4, 7, 25] are usually located in the subcortical parenchyma of the supratentorial brain. Extra-axial CMs account for 14% of all CMs.^[21] They usually arise from the dura mater, particularly at the middle cranial fossa, and are related to the cavernous sinus. They rarely occur at the tentorium, convexity, anterior cranial fossa, Meckel's cave, or cerebellopontine angle.^[5] Thus far, only 8 cases of tentorial CM have been reported.^[18] Generally, CM is a stable or slow-growing mass that develops over a long period,^[14, 22] although the natural course of CM is not yet well described. Here, we present the case of a patient with tentorial CM that rapidly enlarged in 3 months.

CASE DESCRIPTION

A 64-year-old woman visited our hospital with a complaint of diplopia. Despite her subjective complaint, an objective neurological examination revealed no obvious cranial nerve disorder. Magnetic resonance (MR) imaging revealed a mass lesion located beside the left cerebral peduncle; this mass appeared isointense on T1-weighted imaging (T1WI), and hyperintense on fluid-attenuated inversion recovery (FLAIR) imaging (Fig. 1A). After 3 months, the lesion had enlarged with obvious perifocal edema in the cerebral peduncle (Fig.1B). Her diplopia had persisted. Contrast-enhanced MR imaging showed a homogeneously enhanced lesion that had eroded into the midbrain (Fig.1C, D). Cerebral angiography revealed that the blood supply originated from a branch of the middle meningeal artery, and revealed contrast pooling in the venous phase (Fig.2A, B). From the findings of radiological examinations, one of the differential diagnoses was tentorial CM. However, malignant meningioma or hemangiopericytoma could not be excluded because of its rapid enlargement.

The patient was placed in the supine position, and a left orbitozygomatic craniotomy was performed. The lesion appeared as a multilobulated, mulberry-like mass located on the tentorium (Fig.3). The intraoperative diagnosis was CM. The feeding artery, detected on the tentorium, was coagulated. However, the lesion was intentionally not removed to avoid hemorrhage in the CM, especially juxta-brainstem hemorrhage, which would be located in a blind area in this approach.

MR imaging 1 week after the operation showed central necrosis of the lesion concomitant with mass reduction and diminution of the perifocal edema in the midbrain (Fig.4A, B). However, 3 months later, the lesion recurred with homogeneous enhancement (Fig.4C). Consequently, gamma knife radiosurgery was performed. The lesion shrank remarkably in size by 2 months after the treatment (Fig.4D). Seven months later, follow-up MRI revealed no recurrence of the lesion.

DISCUSSION

Intracranial extra-axial CMs located in the middle cranial fossa account for 3% of all benign cavernous sinus masses and usually attach to the cavernous sinus. Our case, however, was independent of the cavernous sinus.^[15] To our best knowledge, only 8 cases of extra-axial CM originating from the tentorium have been reported.^[18]

Most parenchymal CMs are stable in size for long periods, but occasionally grow by recurrent hemorrhages.^[19] Some reports showed aggressive clinical behavior of parenchymal CMs during pregnancy, indicating an involvement of hormonal factors such as estrogen and progesterone.^[1, 2, 6, 27] As other possible causes, capillary budding, ectasia, and thrombosis of vascular spaces might occur in during the enlargement of CMs.^[14, 17, 24]

Extra-axial CMs are also stable, except for 2 cases that showed slow growth over a period of 2 years.^[14, 22] The diameter of the lesion in our case increased by 3 times within 3 months, suggesting that our case was the most aggressive extra-axial CM reported thus far.

Our case, and the 2 cases described above, showed no evidence of hemorrhage on the radiological examinations. Hormonal levels in our patient might be low owing to

menopause. Intriguingly, the lesion in our case shrank once by feeding artery occlusion, but recurred in just 3 months. It is likely that recanalization of the feeding artery occurred, or other vascular channels feeding the CM opened promptly after occlusion of the main feeding artery. Presumably, the CM in our case is apt to promote neovascularization.

On MR imaging, the lesion in our case appeared isointense to gray matter on T1WI and intensive enhancement, which are consistent with the typical findings in CM. At the border of the lesion, extra-axial CM sometimes exhibits a dural tail sign, as observed in our case,^[8, 13, 18] which renders distinguishing of this lesion from meningioma difficult. Angiography is useful to distinguish extra-axial CMs from meningiomas.^[3, 4, 13, 18] Meningiomas show a clear tumor blush, commonly called the “sun-burst appearance”. By contrast, CMs show a stain usually described as “flecked” in appearance, with pooling of contrast in small lakes visible in the late venous phase, as was observed in our case.^[3, 18] The progressive clinical course in our case obfuscated the correct preoperative diagnosis, although the overall findings of radiological examinations were consistent with those of extra-axial CM.

Surgical resection is a common treatment for extra-axial CM, although a lesion with a dural origin close to the cavernous sinus tends to bleed massively during removal.^[11, 13, 18]

Preoperative embolization is useful to avoid bleeding, if the feeding artery is accessible with the catheter.^[10] In our case, the feeding artery was so narrow that it could not be selectively catheterized; however, we could detect and coagulate the artery during the operation. The lesion recurred in 3 months, even after occlusion of the main feeding artery, suggesting that feeder obstruction was not sufficient to cure the lesion.

In contrast to intraparenchymal CM, both fractionated radiotherapy and stereotactic radiosurgery have been reported as effective tools for the adjunct treatment of extra-axial CM, particularly located at the cavernous sinus.^[9, 16, 20, 26] Conventional radiation can be recommended for large lesions with minimal risk.^[26] Stereotactic radiosurgery is indicated for lesions at deep locations, small lesions, residual lesions, or lesions that recur after operation.^[9, 12, 23] Stereotactic radiosurgery was useful for our patient, in whom the lesion was close to the midbrain and recurrent. Treatment should be tailored to the location and

size of the lesion and to the patient's age and clinical symptoms.

CONCLUSIONS

We treated a rare case of progressive tentorial CM. Extra-axial CMs often mimic meningiomas. Their clinical behavior and appearance on imaging are quite different from those of intra-axial CMs. Fractionated or stereotactic radiotherapy is an effective treatment tool for the residual or recurrent extra-axial CM. Further research and longer follow-up periods are required for a better understanding of the natural history of CMs of the cerebellar tentorium.

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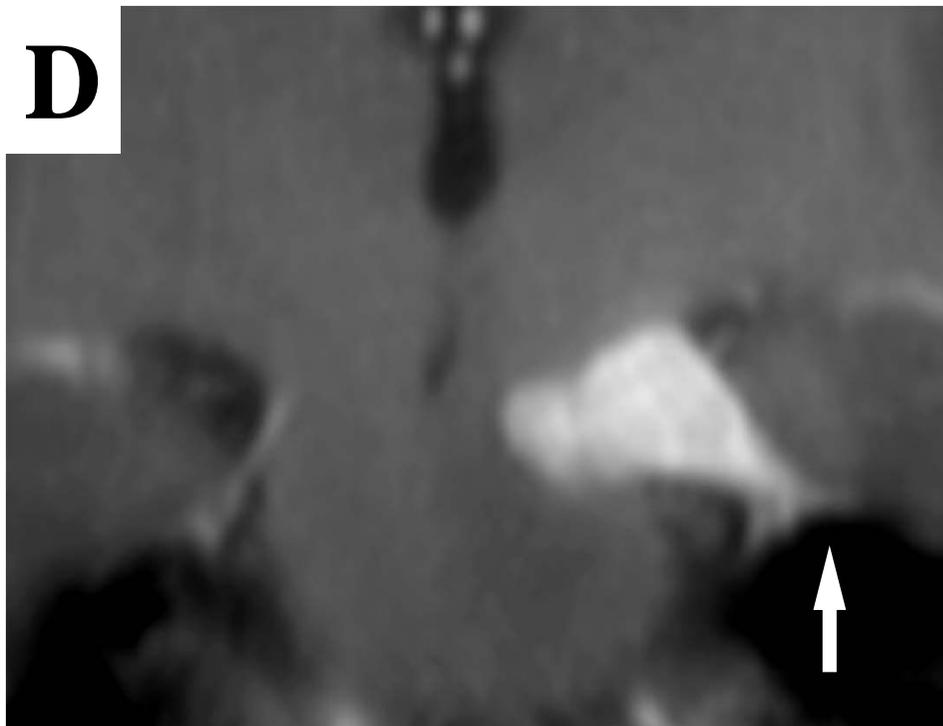
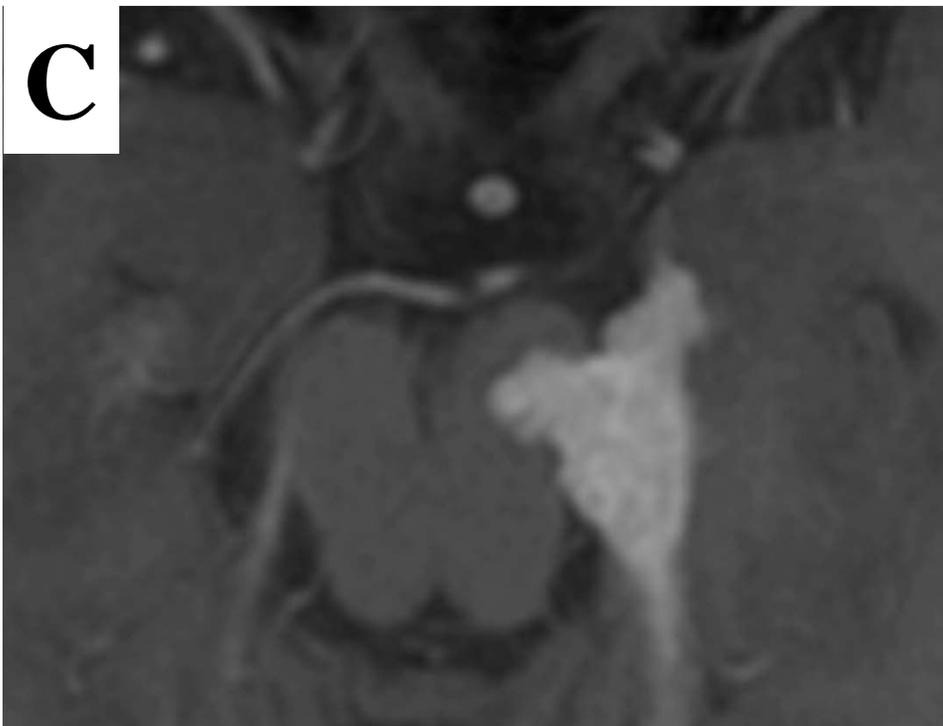
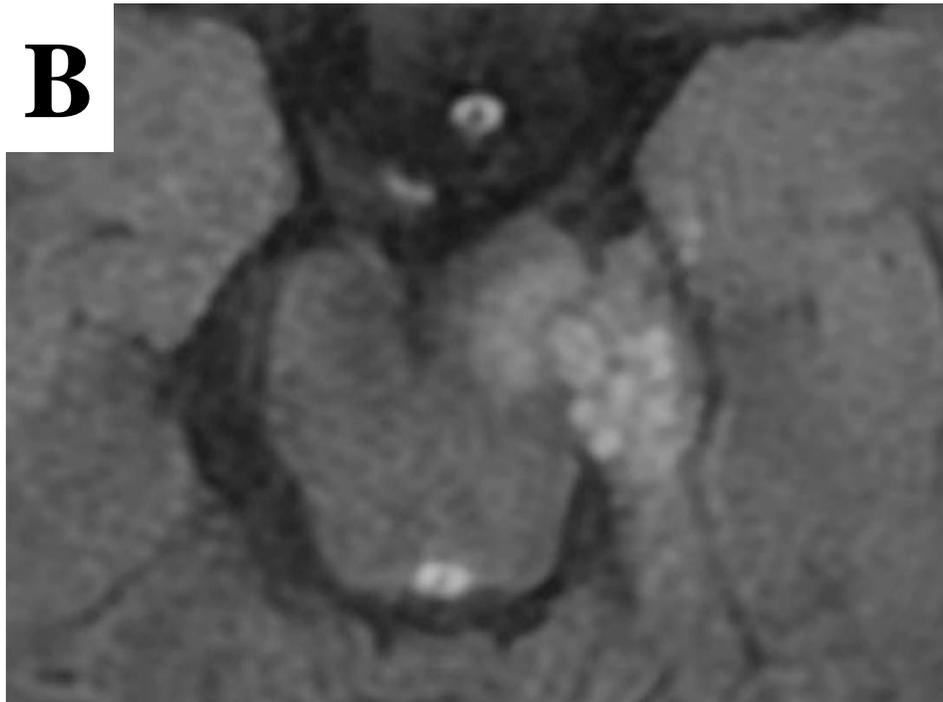
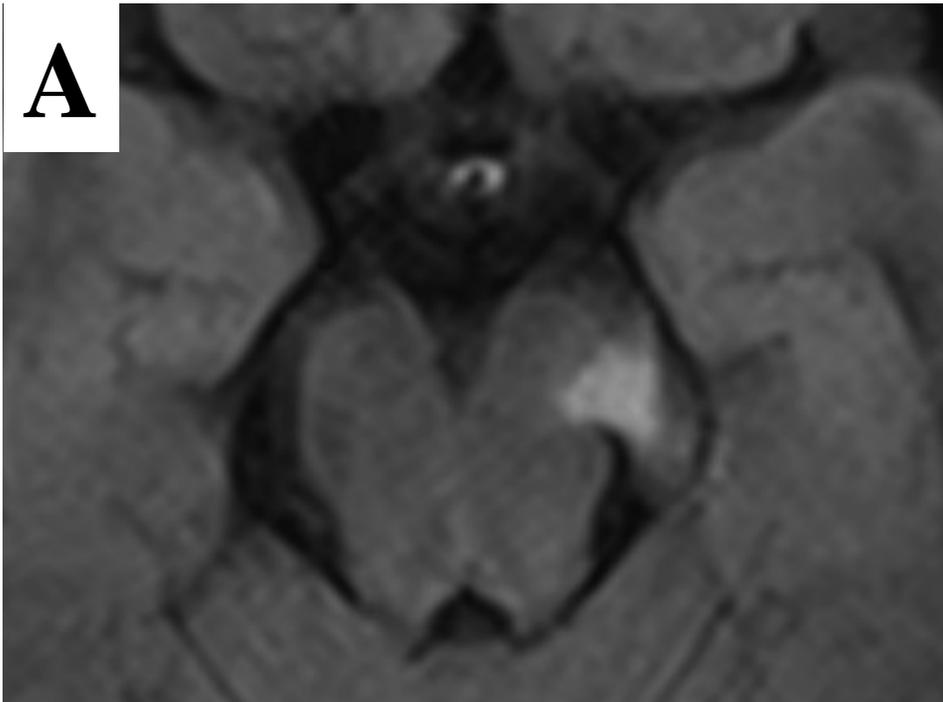
FIGURE LEGENDS

Figure 1: FLAIR image showing a hyperintense lesion beside the brain stem (A). Three months later, the lesion enlarged and eroded into the midbrain with peritumoral edema (B). Contrast-enhanced MR imaging showing a homogeneously enhanced mass (C, D) with a dural tail (arrow).

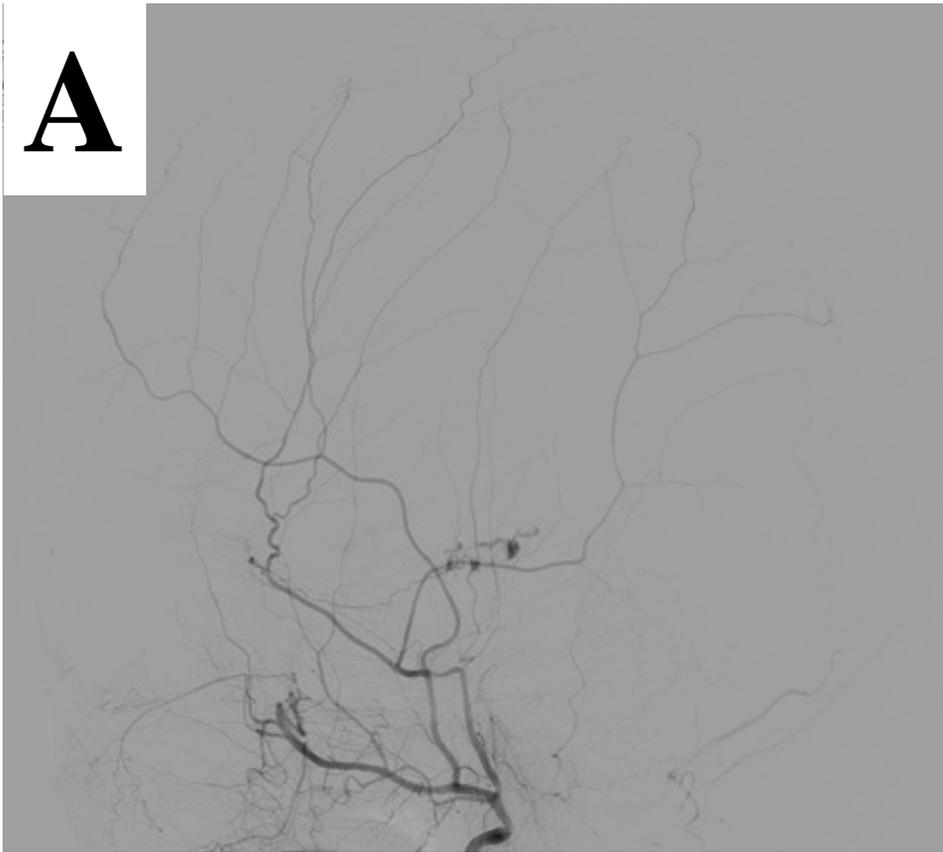
Figure 2: Lateral views of the left external carotid artery angiogram, in late arterial (A) and venous (B) phases, showing filling of the lesion from the periphery to the center with a sustained and persistent blush from the middle meningeal artery.

Figure 3: Intraoperative view: A multilobulated, mulberry-like lesion attached to the cerebellar tentorium, which is a typical macroscopic feature of CMs.

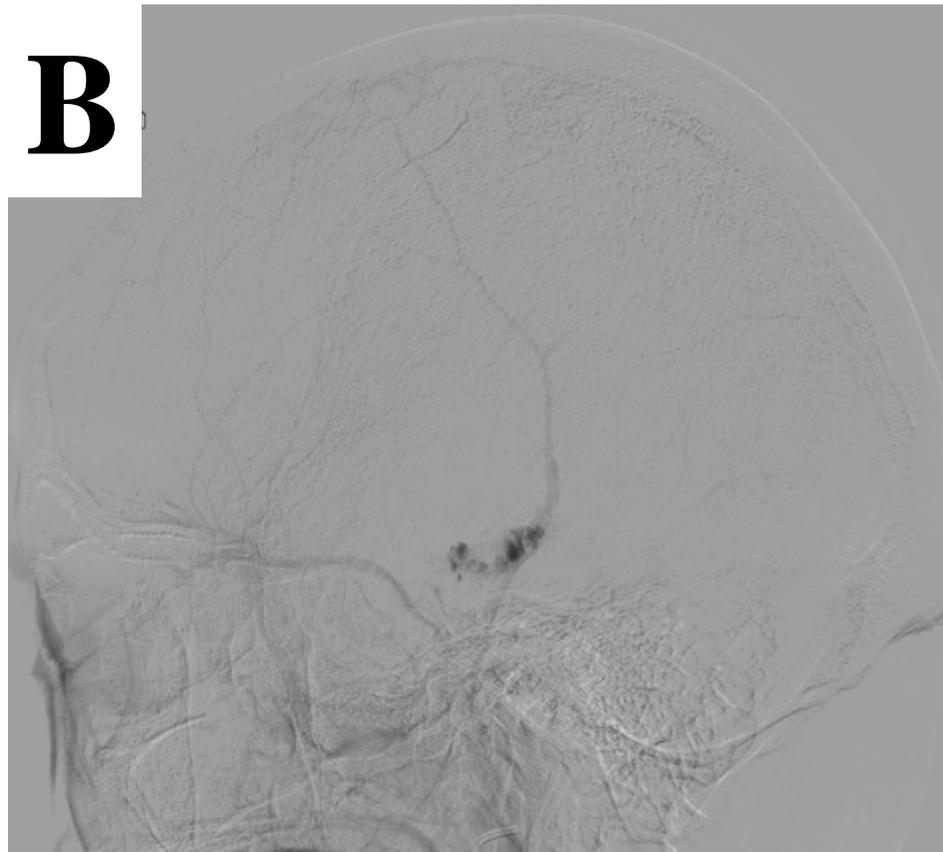
Figure 4: Postoperative axial MRI showing size reduction and central necrosis in the lesion (A) with decreased perifocal edema (B). Three months later, the lesion appeared with homogeneous enhancement (C). Two months after gamma knife radiosurgery, the lesion shrank remarkably (D).

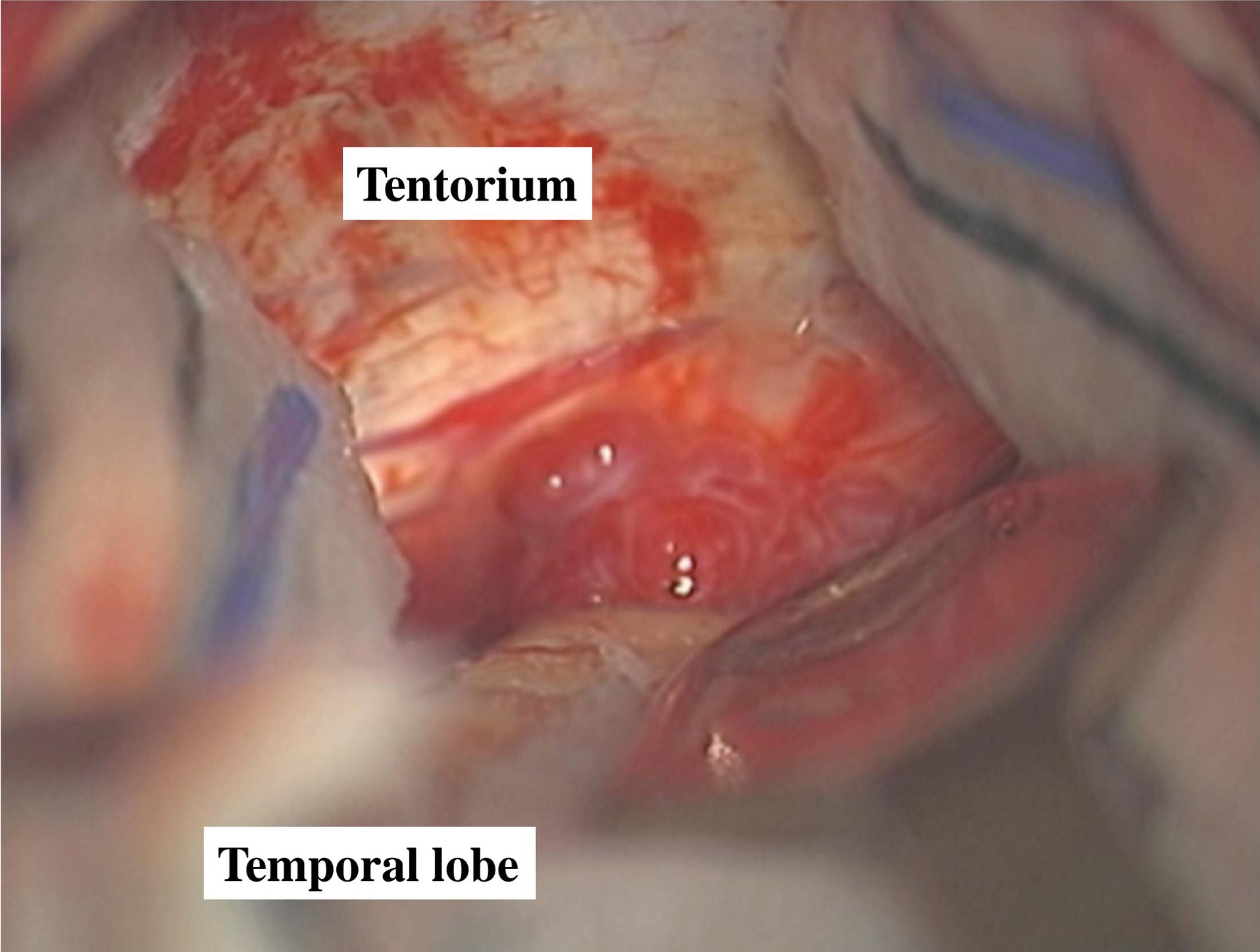


A



B

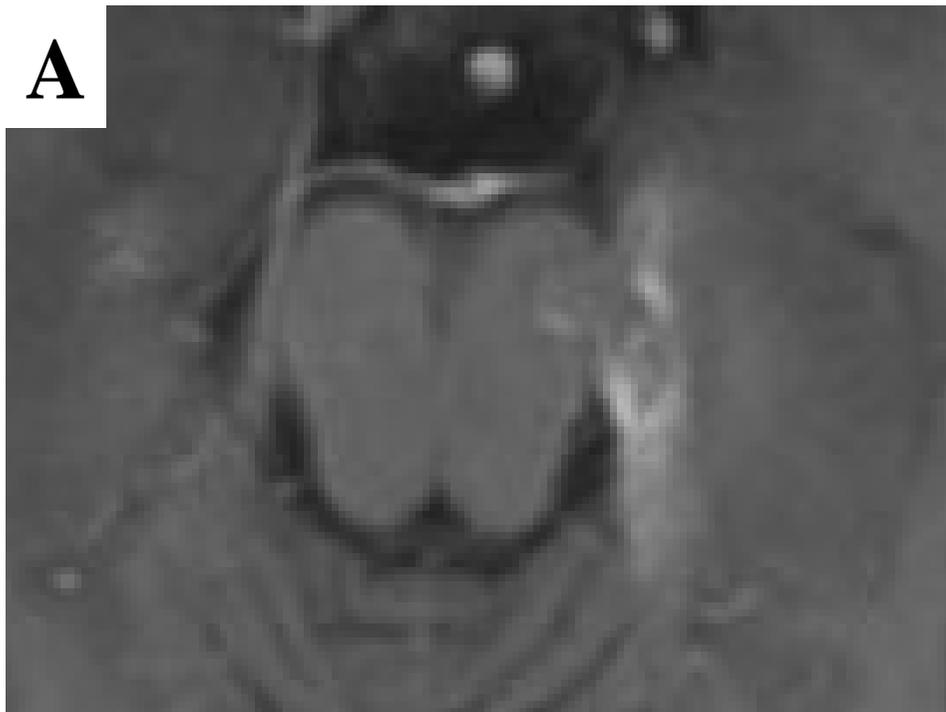
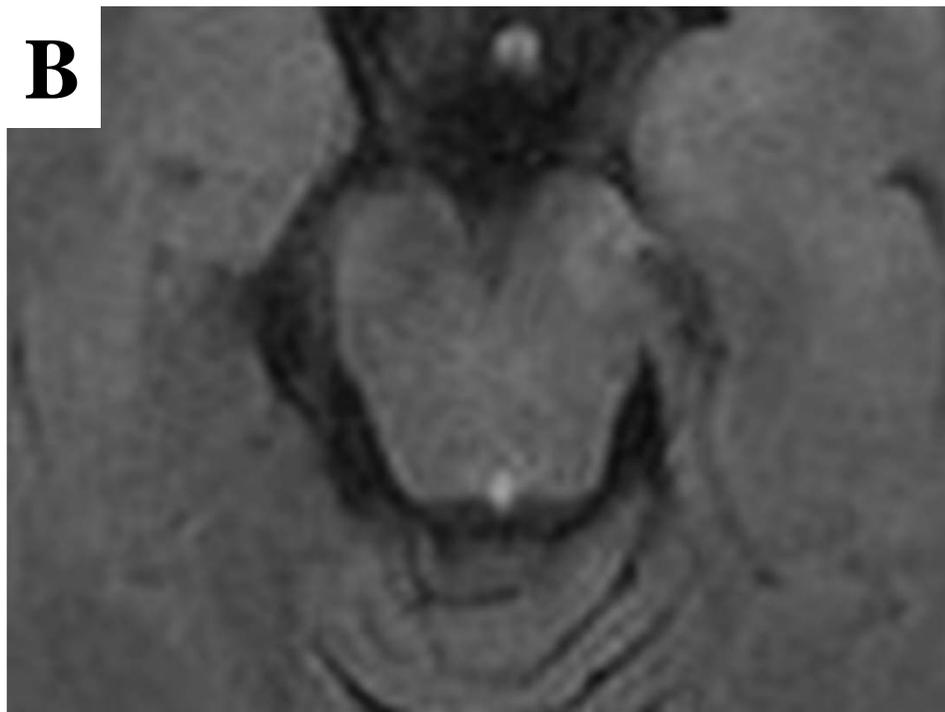




Tentorium

This is an intraoperative photograph showing a surgical dissection. The central focus is a reddish, fibrous structure, which is the tentorium. It is partially retracted, revealing underlying brain tissue. The surrounding area is filled with various anatomical structures, including muscles and connective tissue, some of which are stained with a blue dye. The overall scene is illuminated by surgical lights, creating a clear view of the surgical field.

Temporal lobe

A**B****C****D**