

Rare case of tentorial cavernous malformation mimicking a meningioma: illustrative case

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BACKGROUND Cavernous malformations of the dura, especially of the tentorium, are exceedingly rare. In the available literature, only 10 cases have been described to date.

OBSERVATIONS The authors present the case of a 46-year-old male patient with a 1-cm infratentorial lesion suspicious for meningioma that was found on routine magnetic resonance imaging (MRI) performed for vertigo. The lesion was followed for 1.5 years with no change in signal and size. Nevertheless, the patient was concerned about the lesion and requested removal. The removal was successful and without any neurological sequelae. However, histological evaluation demonstrated a cavernous malformation. Postoperative computed tomography and MRI showed complete removal. Preoperative MRI characteristics, intraoperative images, and a video, as well as histological evaluation, are shown. The case is discussed with respect to the literature.

LESSONS Cavernous malformations of the tentorium are extremely rare and mimic meningiomas; thus, they need to be taken into account. DOTATOC positron emission tomography may help to differentiate in these cases. Considering the cases reported in the literature, in cases of large tumors, preoperative angiography and possibly embolization may be helpful.

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Cavernous malformations are rare, nonneoplastic intracranial lesions occurring in 0.17%–0.9% of the population.¹ They account for 3%–13% of intracranial vascular malformations.² Approximately one-half are the familial form, with at least 3 known gene loci. Most cavernous malformations are supratentorial, with the remaining 23% in the posterior fossa and 5% in the spine. Extra-axial cavernous malformations are thus rare.³ Tentorial cavernous malformations have only been reported in 10 cases in the available literature.^{3–12} In 98% of cases, cavernous malformations occur in the brain parenchyma or spinal cord, often associated with a developmental venous anomaly. On computed tomography (CT) or magnetic resonance imaging (MRI) without and with contrast, differentiating these lesions from meningiomas is challenging. The dural tail sign can be absent,^{7,10} which can provide a possible clue. In the pre-MRI and -CT era, diagnostic cerebral angiography showed dural feeding arteries, which may also be present in meningiomas. Cavernous malformation can become large and can

be highly vascularized, leading to profuse bleeding, as reported in the case of Mori et al.¹⁰ According to the definition, cavernous malformations are slow-perfused vascular malformations with long pooling of contrast medium until the late venous phase.

Histologically, cavernous malformations are vascular malformations that emerge from sinusoidal vessels and are lined with thin endothelial walls without intervening tissue.^{13,15} Larger vessels can demonstrate luminal fibrosis and thrombosis. Endothelial walls do not contain elastic lamina or smooth muscle fibers.

Illustrative Case

An otherwise healthy 46-year-old male had cervical degenerative disease as the only significant history. During the evaluation for headache and vertigo symptoms, a small, 1-cm, left infratentorial lesion was discovered during a routine MRI evaluation (Fig. 1). The lesion

ABBREVIATIONS ADC = apparent diffusion coefficient; CCM = cerebral cavernous malformation; CT = computed tomography; DWI = diffusion-weighted imaging; MRI = magnetic resonance imaging; PET = positron emission tomography; SPECT = single-photon emission computed tomography; SSTR2A = somatostatin receptor type 2A.

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was followed for 1.5 years, after which the patient requested surgical removal in lieu of long-term follow-up.

Surgical Removal

The patient was placed in the right lateral position with the head in 3-point Mayfield fixation turned 45° down and the shoulder retraced caudally. Neuronavigation was performed using a Medtronic StealthStation. A left-sided craniotomy was performed suboccipital to the transverse sinus. The dura was opened, cerebrospinal fluid was drained, and the tumor was accessed superior to the cerebellum and infratentorially with attention to the draining veins. No spatula was used. The tumor was approached, and no parenchymal attachment was found. It was then resected in total, and the attachment was curetted and coagulated (Fig. 2, Video 1). The dura was closed with sutures and fibrin glue in a standard fashion, and the cranial bones were replaced with a small Palacos plate. The fascia was sutured, and the skin was closed with a continuous 4-0 Prolene suture. Postoperatively, the patient awakened without any deficits. Postoperative CT (Fig. 3A and B) showed no evidence of rebleeding, and the follow-up MRI (Fig. 3C) confirmed complete removal.

VIDEO 1. Clip showing the intraoperative removal of cavernous malformation. Click here to view.

Histology

Microscopically, the lesion was composed of a cluster of sinusoidal vascular spaces of varying sizes lined by endothelial cells and embedded in a fibrous stroma. The endothelial cells were immunolabeled with CD31 (Fig. 4A). Somatostatin staining with somatostatin receptor type 2A (SSTR2A) was negative (Fig. 4B)

Patient Informed Consent

The necessary patient informed consent was obtained in this study.

Discussion

Observations

Extra-axial cavernous malformations, especially of the tentorium, are rare. In a recent review by Hassanzadeh et al., 91 extra-axial cavernous malformations were identified in the literature dating from 1947 to the present.³ Of these, 11 were located on the tentorium. This figure corresponds to 12% of the reported cases (Table 1). However, there might be a bias of under-sampling, as smaller cavernous malformations like in our case can mimic a meningioma and, due to the lack of histological confirmation, may never be correctly identified. The clinical presentation varies depending on the location and size of the lesion. Focal neurological deficits and seizures have been reported in the literature.³ The exact rate of hemorrhage is unknown, with the reported 9% risk for hemorrhage possibly overestimated, as the natural history is based largely on intra-axial cavernous malformations.¹⁵

The exact nature of the origin of cavernous hemangiomas is unclear. In intra-axial cavernous malformations, several genes have been found in correlation with their presence, such as cerebral cavernous malformation (CCM) genes *CCM-1*, *-2*, and *-3*.^{16,17} In dura-based cavernous malformations, no correlation to specific genes has been attempted. The factors leading to the growth of dura-based cavernous malformations are not known. However, factors such as endocrine influences, thrombosis, and capillary budding have been discussed.¹⁸

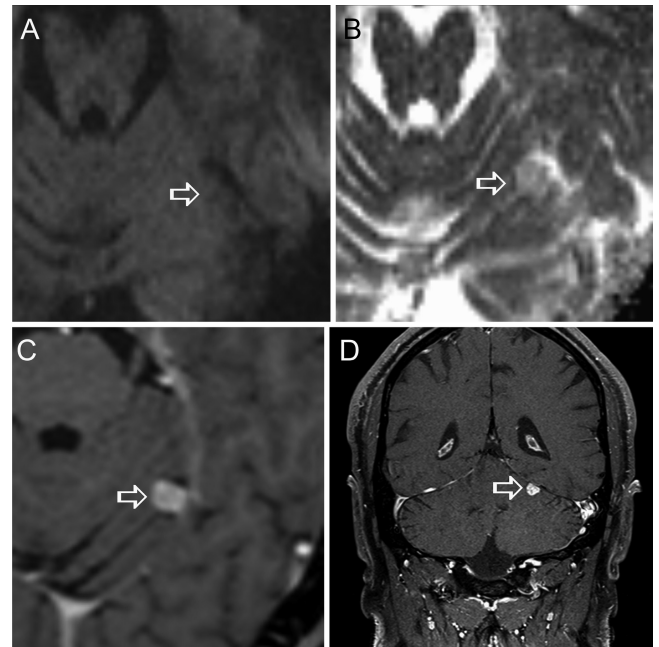


FIG. 1. MRI of the suspected tentorial lesion. Axial DWI at B1000 (A) and corresponding ADC map (B), as well as an axial T1-weighted image with contrast on the same slice (C), depicting the supracerebellar lesion (arrows) with strong contrast enhancement but without any restricted diffusion. Coronal T1-weighted contrast-enhanced MRI (D) showing the right-sided tentorial location of the lesion. No obvious dural tail is visible, which, in retrospect, is evidence against a dural meningioma.

Preoperative evaluation is usually assessed by MRI and CT, and possibly angiography in larger lesions. It is worth noting that the lesion may not be distinguished by conventional MRI or CT sequences. However, Wang et al. recently reported that the application of a radiomics-based algorithm using diffusion-weighted imaging (DWI) and apparent diffusion coefficient (ADC) facilitates the distinction of meningiomas from cavernous malformations.¹⁹ Future automated radiomics may assist in the preoperative distinction, which may have

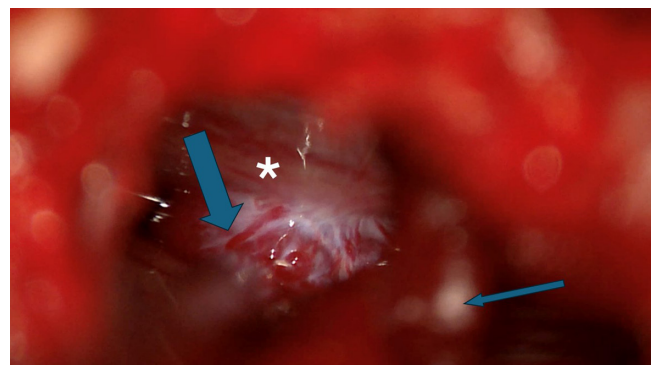


FIG. 2. Intraoperative view of the cavernous tentorial malformation. The patient was in the right Fukushima position. *Large arrow* indicates the cavernous malformation; *asterisk*, the tentorium; and *small arrow*, draining cerebellar vein en passant. No spatula was used for removal. See Video 1 for the intraoperative procedure.

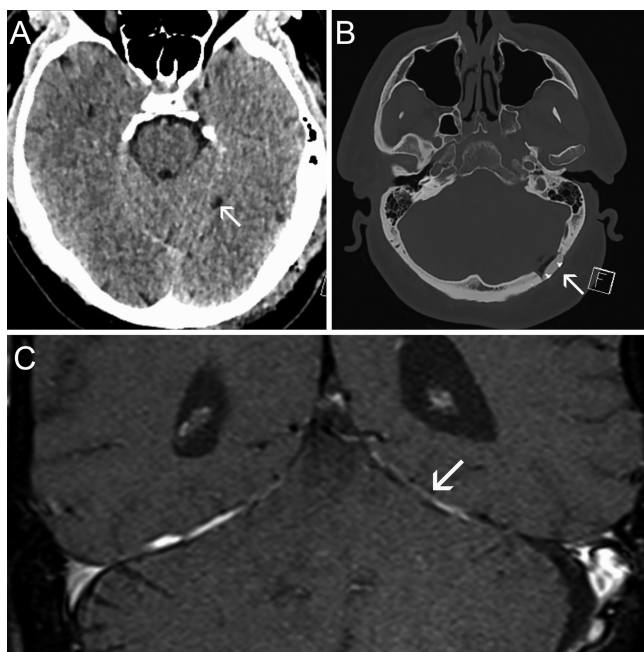


FIG. 3. Postoperative CT in brain parenchyma windowing (A) shows a small defect without any postoperative hematoma or residual mass (arrow). The suboccipital access was closed with a Palacos plate (arrow), clearly visible on the bone window (B). Four weeks after resection, coronal T1-weighted contrast-enhanced MRI (C) showed complete resection of the right-sided tentorial lesion, without residual neoplastic or granulomatous tissue, with only minimal postoperative residual enhancement of the tentorium (arrow).

implications for treatment evaluation, such as additional angiographic imaging for larger lesions. In single-photon emission computed tomography (SPECT), cavernous malformations are usually low in uptake, whereas in other tumors such as meningiomas, the metabolic uptake may be higher. Despite these observations, SPECT does not sufficiently contribute to securing the preoperative diagnosis.²⁰

Surgical removal can be challenging and profuse bleeding can occur, especially in cases that are large and located near the cavernous sinus.²¹ In these cases, cerebral angiography and endovascular embolization of feeding arteries may be beneficial prior to surgical

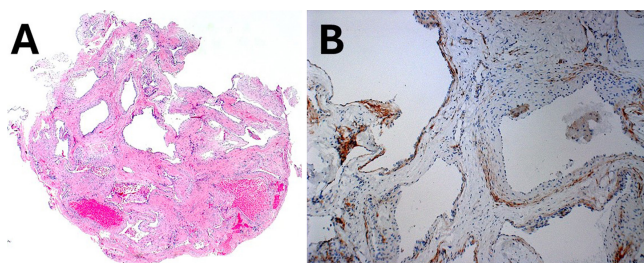


FIG. 4. A: Microscopically, the lesion is composed of a cluster of sinusoidal vascular spaces of varying sizes lined by endothelial cells and embedded in a fibrous stroma (hematoxylin and eosin stain). The endothelial cells were immunolabeled with CD31 (not shown). Original magnification $\times 25$. B: Lining endothelial cells express no immunoreactivity for SSTR2A. Original magnification $\times 100$.

TABLE 1. Historical cases of tentorial cavernous malformation

Authors & Year	Location	No. of Cases
McCormick & Boulter, 1966 ⁴	Tentorium	2
Moritake et al., 1985 ⁵	Tentorium	1
Matsumoto et al., 1988 ⁶	Tentorium	1
Quattrocchi et al., 1989 ⁷	Tentorium	1
Lee et al., 1998 ⁸	Tentorium	1
van Lindert et al., 2007 ⁹	Tentorium	1
Mori et al., 2009 ¹⁰	Tentorium	1
Bhatia et al., 2011 ¹¹	Tentorium	1
Yoshimura et al., 2014 ¹²	Tentorium	1
Hassanzadeh et al., 2024 ³	Tentorium	1

A total of 12 cases including our case were reported. Other locations were more often reported as the cavernous, falx, and convexity. Tentorial cavernous malformations account for around 12% of reported cases in the literature.

removal. Furthermore, radiotherapy and radiosurgery can be considered in deeper-seated lesions. However, the radiosensitivity of the lesions is not clearly predictable.^{22,23}

Lessons

Dural cavernous malformations are rare locations reported in the literature. Tentorial cavernous malformations account for around only 12% of reported cases. In total, 12 cases, including ours, with a tentorial location have been reported. Surgical removal is the treatment of choice and can be complicated, as these tumors can cause profuse bleeding, especially in cases with large tumors. Although intraparenchymal cavernous malformations are angiographically occult, the few cases with intradural large tumors reported in the literature were angiographically positive.¹⁰ In these cases, cerebral angiography and embolization may be advisable prior to surgery. Differentiating cavernous malformation from meningioma prior to surgery can often be difficult; however, the dural tail sign may be missing. Radiomics may possibly help in these cases in the future. Furthermore, as meningiomas express somatostatin receptors, which can be delineated by positron emission tomography (PET) after the injection of somatostatin analogs such as ⁶⁸Ga-DOTATATE (DOTA-D-Phe1-Tyr3-octreotate) or ⁹⁰Y-DOTATOC (DOTA D-Phe1-Tyr3-octreotide),²⁴ PET may help to differentiate cavernous malformations from meningiomas prior to surgery. In our case, somatostatin receptor staining was negative.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: M Reinert, Rushing. Acquisition of data: M Reinert, Rushing, Fischmann, V Reinert. Analysis and interpretation of data: M Reinert, Rushing, V Reinert. Drafting the article: M Reinert, Kolenc, Fischmann, V Reinert. Critically revising the article: M Reinert, Rushing, Fischmann. Reviewed submitted version of manuscript: M Reinert, Rushing, V Reinert. Approved the final version of the manuscript on behalf of all authors: M Reinert. Statistical analysis: M Reinert. Administrative/technical/material support: M Reinert. Study supervision: M Reinert.

Supplemental Information

Videos

Video 1. <https://vimeo.com/949558488>.

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