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Case Report

# De novo meningiomas and cavernous malformations developing after coil embolization for dural arteriovenous fistula ☆

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ABSTRACT

A 25-year-old woman presented with exophthalmos. With the diagnosis of dural arteriovenous fistula of the transverse sinus, the patient underwent coil embolization. Fifteen years later, she sustained visual disturbance due to de novo tuberculum sellae meningioma that was resected. Surveillance magnetic resonance imaging (MRI) performed at the age of 42 years detected tumor recurrence and small, apparent meningioma in the cerebral convexity. The patient underwent the second tumor resection at the age of 46 years. The histological appearance was consistent with meningothelial meningioma. In addition, surveillance MRI at the age of 43 years detected de novo cerebral and cerebellar cavernous malformations (CMs). These CMs showed repeat hemorrhages on MRI but remained asymptomatic during the next 38 months. De novo meningiomas and CMs may develop in association with radiation exposure during endovascular therapy. Sufficiently long-term follow-up is recommended after endovascular therapy for monitoring secondary pathologies.



## Introduction

Radiotherapy can result in the development of secondary pathology in the radiation port more than 10 years after administration [1,2]. Radiation-induced meningiomas have been documented as distinct entities represented by lower patient age at diagnosis, higher proportion of multiple tumors, and high recurrence rate with the latency period between radiotherapy and the onset of meningiomas 22.9±11.4 years [1,3,4]. Cranial irradiation at older ages, higher doses, and concomitant chemotherapy are thought to be associated with a shorter latency period for developing these tumors [5,6]. Also, radiation-induced cavernous malformations (CMs) have been documented as late complications associated with radiotherapy [2,7–12]. Infrequently, simultaneous or consecutive development of radiation-induced meningioma and CM has been reported [13,14]. However, to the best of

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our knowledge, there has not been a report documenting the development of radiation-induced lesions after endovascular therapy.

Here, we present a unique case of radiation-induced meningiomas and CMs that probably developed in association with coil embolization for the treatment of dural arteriovenous fistula.

## Case report

A 25-year-old woman presented with nontraumatic exophthalmos and was diagnosed with dural arteriovenous fistula of the right transverse sinus. The patient had not received any previous radiation therapy. She underwent coil embolization combined with an occipital craniotomy that exposed the sinus and directly punctured the sinus (Fig. 1). Fifteen years later, the patient sustained visual disturbance due to a dural-based tumor developed in the tuberculum sellae that was totally resected (Figs. 2A and B). Histological diagnosis was meningothelial meningioma. Surveillance magnetic resonance imaging (MRI) performed at the age of 42 years detected tumor recurrence and development of small, dural-based tumors in the cerebral convexity (Figs. 2C and D). The patient underwent the second surgery at the age of 46 years. Through a left frontotemporal craniotomy, a gross total tumor resection was achieved. Histologically, the tumor comprised of arachnoid cell-like neoplastic cells with frequent whorl formations. Neither cell atypia nor pleomorphism was found. There were scant mitotic figures. Immunohistochemical examination showed positive staining for epithelial membrane antigen and progesterone receptor, while negative for glial fibrillary acidic protein (Fig. 3). These findings were consistent with meningothelial meningioma. In addition, surveillance MRI at the age of 43 years detected de novo lesions in the cerebral and cerebellar hemispheres, suggesting CMs. They showed findings of repeat hemorrhages but remained asymptomatic during the next 38 months (Figs. 4 and 5). Both the cerebellar lesion and pre-existing cavernous malformation were located adjacent to a venous malformation (Fig. 6). The patient has now been placed under close observation with a periodical MRI performed every 6 months.

## Discussion

Endovascular therapy is currently adopted as the first-line modality in the treatment of dural arteriovenous fistulas. On the other hand, as the therapy is performed under fluoroscopy, risks of radiation injury can increase when they are prolonged or complicated. In this case, multiple dural-based tumors and CMs developed after coil embolization of dural arteriovenous fistula, with a latent period more than 17 and 18 years, respectively. These lesions showed clinical progressions with consecutive tumor developments and repeat hemorrhages. Furthermore, given that the coils deployed in the transverse sinus formed huge mass, the present patient could have undergone a prolonged embolization with exposure to considerably high dose X-ray during the procedures. Therefore, we assumed that these de novo tumors and CMs may be radiation-induced lesions. Also, given that the tuberculum meningioma became symptomatic 15 years after coil em-

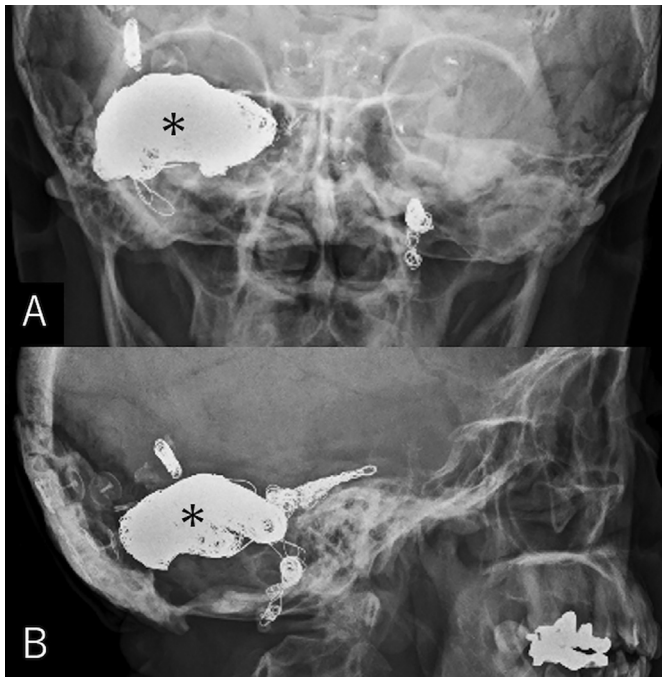


**Fig. 1 – Cranial X-rays, anteroposterior (A) and lateral (B) views, showing a huge coil mass deployed in the right transverse sinus (*asterisk*).**

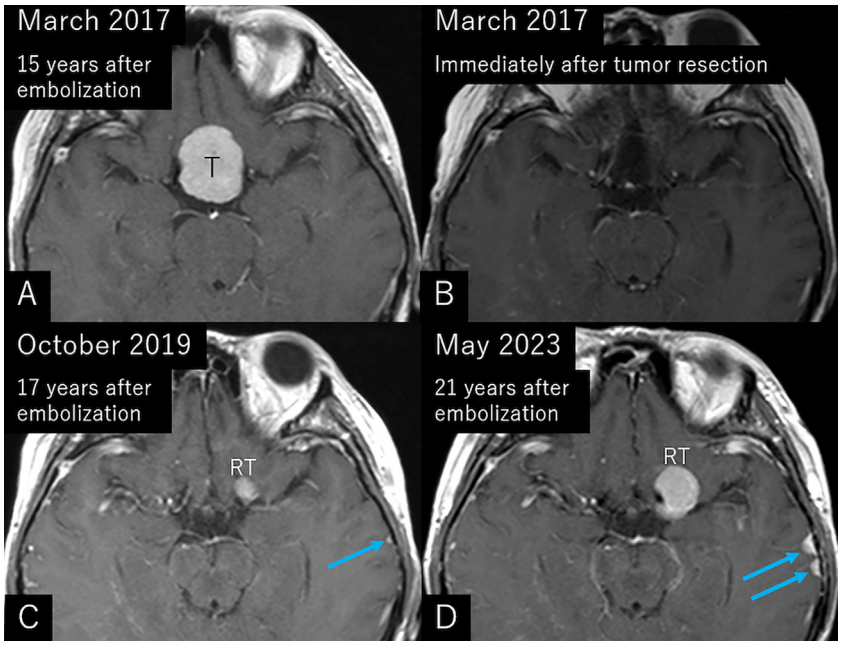


**Fig. 2 – Serial postcontrast axial T1-weighted magnetic resonance images at the same level that were performed at the initial presentation with a tumor in the tuberculum sellae (A), after tumor resection (B), at the tumor recurrence in the lateral aspect of the left anterior clinoid process with a presence of small, dural-based tumor in the left temporal convexity (C), and enlargement of the recurrent tumor with increases in size and number of the temporal convexity tumor (D). RT: recurrent tumor; T: tumor; Arrow: de novo tumor.**

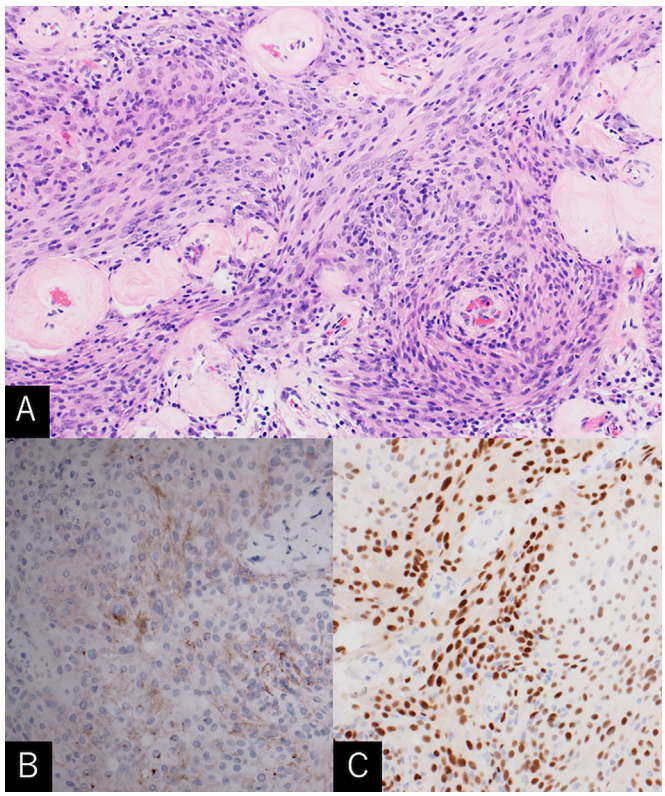


**Fig. 3 – Photomicrographs of the resected specimen showing arachnoid cell-like neoplastic cells and frequent whorl formations. Neither cell atypia nor pleomorphism is found. Immunohistochemical examination showed positive staining for epithelial membrane antigen (B) and progesterone receptor (C). (A) Hematoxylin and eosin stain, ×200; (B, C) ×200.**

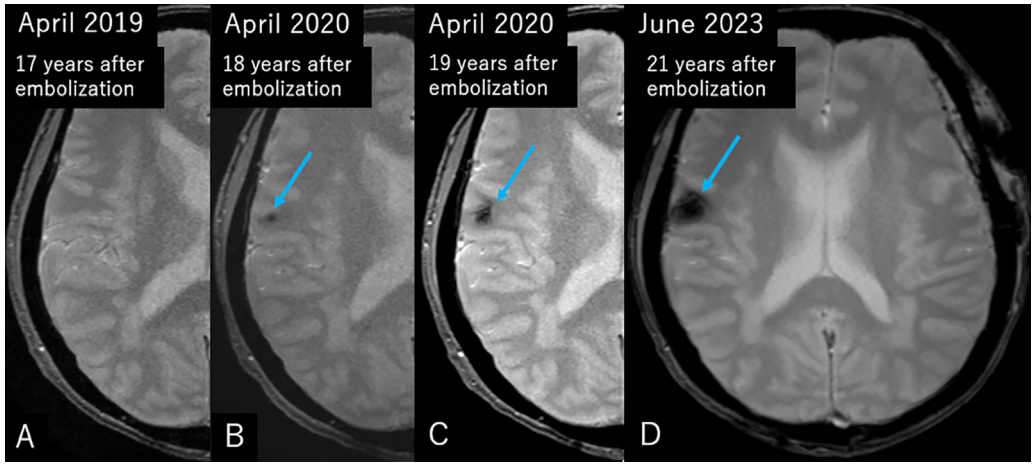


**Fig. 4 – Serial images of axial T2*-weighted magnetic resonance images performed at the same level showing development of hemosiderin deposition in the right frontal lobe 18 years after coil embolization (A, B) and its temporal enlargement (C, D).**

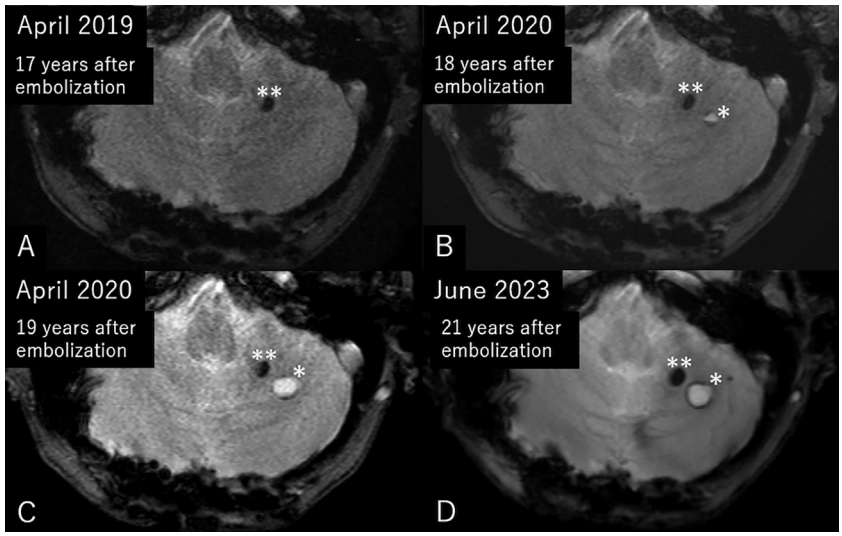


**Fig. 5 – Serial images of axial T2*-weighted magnetic resonance images performed at the same level showing a cystic lesion lined by low-intensity rim in the left cerebellar hemisphere (*asterisk*), suggesting cavernous malformation developed 18 years after coil embolization (A, B) and its temporal enlargement (C, D). Double asterisk: pre-existing cavernous malformation.**

bolization, it might be radiation-induced tumor with a typical histological appearance of meningothelial meningioma. For a better understanding of radiation-induced meningiomas, exploration of detailed genomic profiles is desirable, although it was not performed in the present tumor.

In our case, both pre-existing and de novo cerebellar CMs were located adjacent to a venous malformation, consistent with previous reports suggesting a causal relationship between developmental venous malformation and some CMs [7,12]. A fraction of CMs may acquired develop in association with venous anomalies.

De novo meningiomas and CMs may develop in association with radiation exposure during endovascular therapy especially when the procedure is prolonged. Sufficiently long-term follow-up is recommended after endovascular therapy for monitoring secondary pathologies.

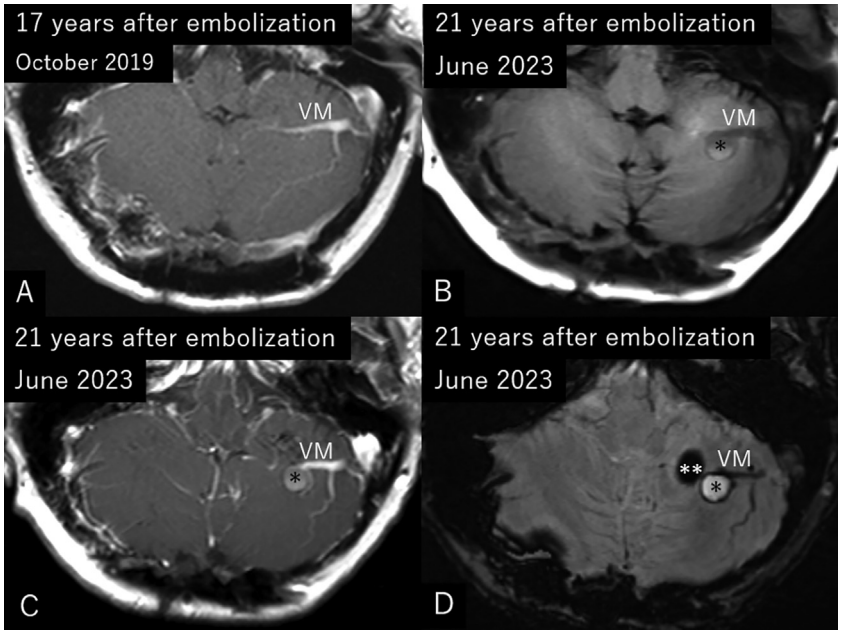


**Fig. 6 – Postcontrast axial T1-weighted magnetic resonance images performed at 17 years after coil embolization (A), pre- (B) and postcontrast (C) T1-weighted images at 21 years, and susceptibility-weighted images at 21 years (D), at the same level of Fig. 5, showing that both the cerebellar lesion and pre-existing cavernous malformation (*double asterisk*) are located adjacent to a venous malformation (VM).**

## Author contributions

All the authors contributed equally to the study.

## Ethical standards and patient consent

We declare that all procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its later amendments. Written informed consent was obtained from the patient for publication of anonymized data.

## Patient consent

We have obtained written informed consent from the patient documented in the manuscript. The patient fully understood and agreed that the authors use information materials of the patient in an anonymized manner for possible publication in Radiology Case Reports.

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