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# Regression of intracranial meningioma following intratumoral hemorrhage

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ABSTRACT

An intratumoral hemorrhage within a meningioma occurs infrequently; in less than 3% of all lesions. When hemorrhage does occur, however, it is associated with a poor prognosis and significant mortality rates. We report a 66-year-old woman with a 10-year history of multiple intracranial meningiomas managed conservatively who underwent surgical resection of a spheno-orbital lesion for decompression of the right optic nerve. Postoperatively, an intratumoral hemorrhage developed in a contralateral lesion, which was managed conservatively. During follow up, the hemorrhaged lesion became significantly smaller. To our knowledge there are no published reports of spontaneous resolution of a meningioma after intratumoral hemorrhage without surgical management. We review the literature on hemorrhage in meningiomas and postulate some pathophysiologic mechanisms for the bleeding and subsequent tumor resolution seen in this patient.



## 1. Introduction

Meningiomas are the most common non-glial intracranial tumor, with an incidence of 2.3 to 5.5 per 100,000 people,[1,2] accounting for 30% of all primary adult brain tumor diagnoses in the United States of America.[3] Hemorrhage secondary to meningioma is rare, occurring in 1.3% to 2.4% of those lesions.[4,5] Tumoral bleeding is associated with worse prognoses and higher mortality rates – the overall mortality for bleeding meningiomas has been reported to be 21.1% and 9.5% in surgically treated patients.[6] Treatment for such hemorrhages has included tumor resection, hematoma evacuation and non-operative medical management.[6,7]

To our knowledge there are no reports in the literature that show regression of an intracranial meningioma following intratumoral hemorrhage. In this report, we describe a patient with multiple intracranial meningiomas who developed an intratumoral hemorrhage in one of the lesions, and experienced subsequent resolution of the tumor. In addition, we review the literature on intratumoral hemorrhage of meningiomas and propose possible pathophysiologic mechanisms by which resolution may have occurred in this patient.

## 2. Case report

### 2.1. History and physical examination

A 66-year-old woman with a history of dizziness presented to our center after being diagnosed with left parietal and right anterior temporal enhancing lesions on MRI consistent with bilateral meningiomas. At that time, the patient had neurological symptoms other than her dizziness, which was attributed to Ménière's Disease, and her extra-axial lesions were followed with serial MRI studies and clinical evaluations. Over the following 10 years, the patient remained both clinically and radiographically stable. She eventually noticed decreased vision in her right eye. At that time, she denied headaches, seizures, motor weakness, and sensory abnormalities. On examination, she was alert and oriented to person, place and time. She was fluent without signs of aphasia. Examination of cranial nerves II through XII was notable for normal vision in the left eye, but detection of hand motion only in the right eye, with a right afferent pupillary defect, and a temporally pale right optic disc on fundoscopy; consistent with a compressive right optic neuropathy. Motor examination was unremarkable except for mild weakness in the distal right lower extremity secondary to sciatic pain. Sensation, reflexes, coordination and gait were also unremarkable.

### 2.2. Imaging

MRI performed after the development of visual impairment showed right temporal (2.4 cm × 2.8 cm × 3.2 cm) (Fig. 1) and left parietal (1.7 cm × 2.85 cm × 2.27 cm) (Fig. 2) extra-axial enhancing lesions with hyperostosis of the right greater wing of the sphenoid bone (Fig. 1B). The presumptive diagnosis was bilateral intracranial meningioma.

### 2.3. Procedure

The patient elected to undergo surgical resection of the right spheno-orbital tumor. A right-sided frontotemporal craniotomy was performed with excision of the right temporal lesion and decompression of the right optic nerve. Pathologic examination of the resected lesion was compatible with meningioma, World Health Organization grade I.

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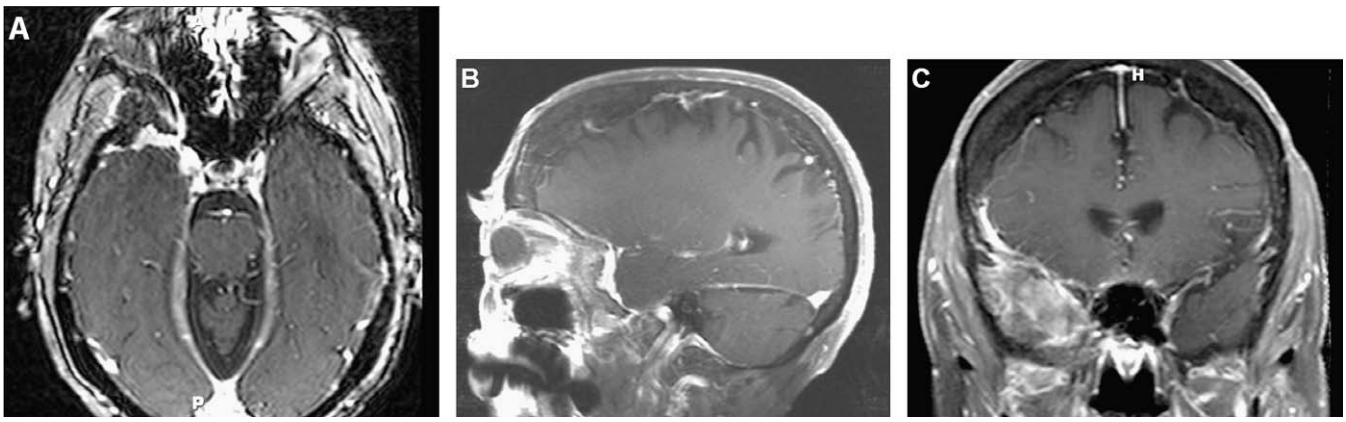


**Fig. 1.** Preoperative T1-weighted MRI with contrast enhancement showing the right spheno-orbital meningioma (A, axial) encroaching on the adjacent optic nerve, (B, sagittal) growing into the petrous temporal bone, and (C, coronal) in the coronal plane growing along the sphenoid wing.

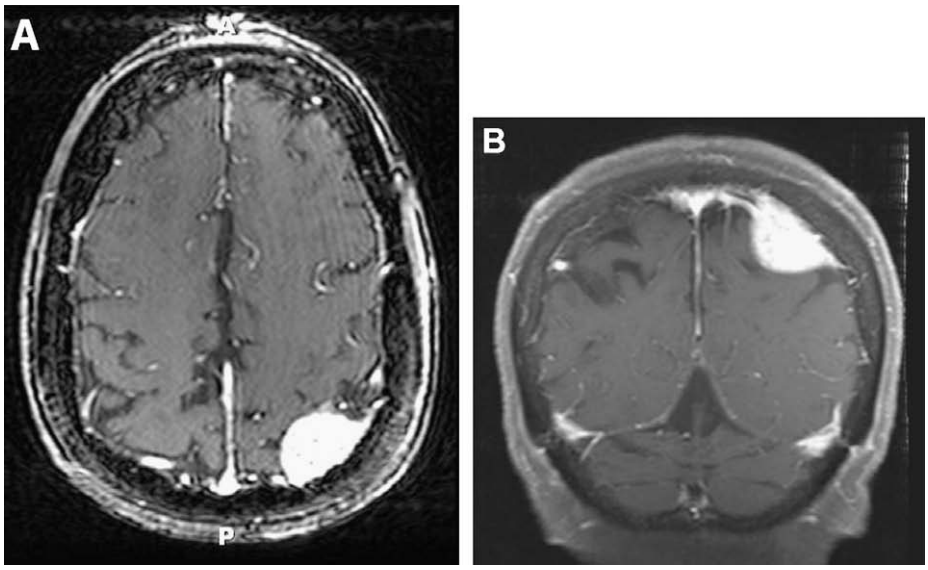


**Fig. 2.** Preoperative (A) axial and (B) coronal T1-weighted MRI with contrast enhancement showing the left parieto-occipital convexity meningioma.

Following surgery, the patient was transferred to the Neurosciences Critical Care Unit (NCCU) for hemodynamic and neurologic monitoring. While in the NCCU, the patient was initially slow to wake and had an expressive aphasia. A head CT scan demonstrated postoperative changes on the right and new hyperdensities in the contralateral parietal tumor, with no new mass effect or midline shift (Fig. 3). These hyperdensities were presumed to be acute hemorrhages rather than tumor calcifications because they were not apparent on previous imaging studies. With the absence of mass effect and the patient's refusal of surgical intervention, aggressive medical therapy was implemented including intravenous mannitol and hypertonic saline for potential elevation of intracranial pressure. Over the next 2 days, her aphasia resolved completely. She was discharged to inpatient rehabilitation on postoperative day 15.

**Fig. 3.** Postoperative day 1 axial CT scan without contrast enhancement showing blood in the left parietal meningioma.

### *2.4. Follow-up*

The patient returned for follow-up 2 months after surgery and reported improved vision in her right eye. She was able to count fingers at two feet and single field-testing of the right eye was moderately

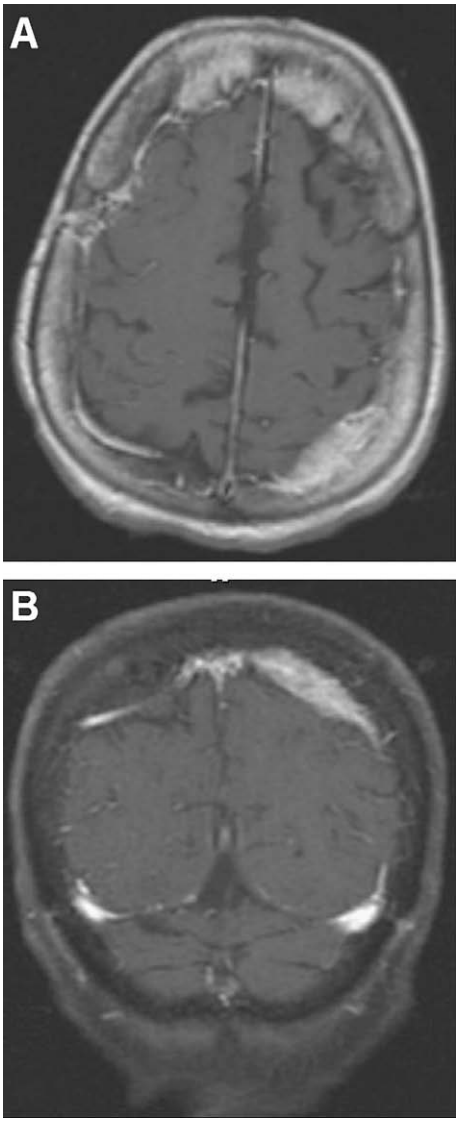


**Fig. 4.** Two-month postoperative (A) axial and (B) coronal T1-weighted MRI with contrast enhancement showing significant reduction in size of the left parieto-occipital lesion.

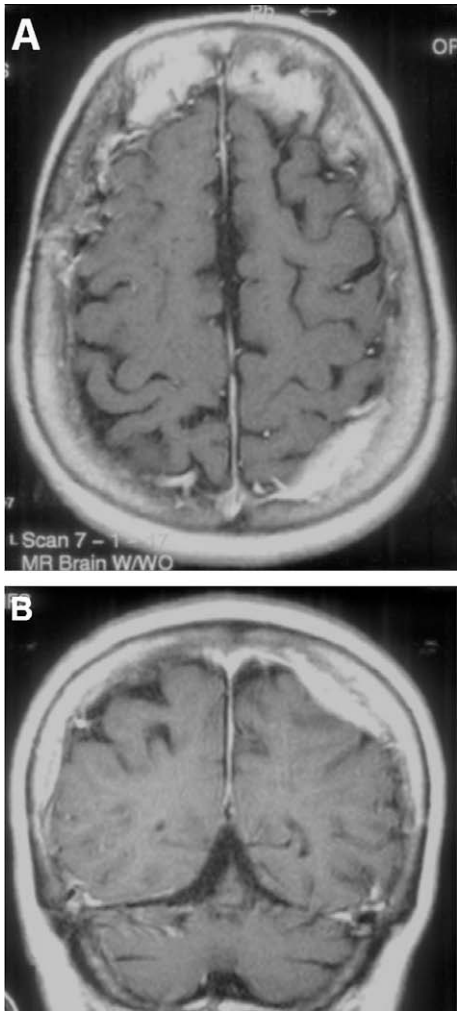


**Fig. 5.** One-year postoperative (A) axial and (B) coronal T1-weighted MRI with contrast enhancement showing stable reduction in size of the left parietal lesion.

improved. She also denied headaches, seizures, motor weakness, and sensory abnormalities. Surgical treatment for the left parietal meningioma was once again offered, but the patient refused and decided to opt for serial imaging studies. Interestingly, MRI studies at 2 months (Fig. 4) and 12 months (Fig. 5) postoperatively showed substantial reduction in size (about 90%) of the contralateral left parietal lesion in the absence of any additional treatment.

## 3. Discussion

Hemorrhages secondary to meningiomas may present as subarachnoid hemorrhages, subdural hematomas, and intracerebral or intratumoral bleeds,[4] but occur only rarely in such lesions.[5,8,9] Some factors have been associated with an increased propensity for meningiomas to bleed, including age 30 years to 70 years, convexity and intraventricular tumors, and malignant, fibrous and angioblastic type meningiomas.[6] The presence of microcysts, intratumoral necrosis,[7] and a higher proliferation rate of the lesion[9] may also predispose lesions to hemorrhage.

Spontaneous regression of meningiomas (without surgery, radiosurgery, embolization, or chemotherapy) has been reported rarely. Previous studies described "spontaneous control" of meningiomas after discontinuation of hormonal therapy, specifically progesterone agonists.[10,11] Pozzati et al. reported a postmenopausal woman with lymphangioleiomyomatosis who presented with rapid growth of multiple meningiomas after initiation of progesterone therapy and subsequent regression of the lesions after suspension of the drug.[10] Shimizu et al described a similar situation in an 80-year-old male who experienced spontaneous regression of an asymptomatic meningioma 15 months after suspension of progesterone agonist therapy.[11] However, there have been no reports of spontaneous regression of such tumors following intratumoral hemorrhage, as occurred in our patient.

It is unclear what mechanism led to such resolution, but there are several reasonable possibilities. First, an intratumoral hemorrhage might have led to necrosis of the tumor. Spontaneous regression of intracranial tumors following intratumoral hemorrhage has been reported for other lesions, specifically pituitary adenomas. Hemorrhagic events within pituitary adenomas (that is, pituitary apoplexy) are more common than in meningiomas. In Wilson's series of pituitary macroadenomas, 3% of patients had an episode of apoplexy,[12] while in another series of 560 patients, the incidence was 17%.[13] The spontaneous resolution of pituitary lesions following apoplectic events has been reported.[14–17] Bjerre et al. suggested that some growth hormone-releasing adenomas could show complete or partial disappearance of the adenoma following hemor-

rhage, probably as a result of infarction.[18] These cases raise the possibility of meningioma necrosis and resolution following intratumoral hemorrhage.

Second, resection of the original tumor possibly modified the dural blood supply and the venous drainage to the contralateral tumor. Meningiomas are known to cause local vascular disturbances, mainly venous congestion and enhanced circulation that give rise to the characteristic radiographic finding of a dural tail.[19] Operative interruption of the already abnormal dural vasculature may have predisposed the distant lesion to further vascular irregularities and precipitated intratumoral hemorrhage. Such findings were described by Ahn et al. who reported the spontaneous resolution of a transverse sinus intradural arteriovenous fistula (AVF) after excision of an ipsilateral non-occlusive convexity meningioma without any surgical manipulation of the AVF.[20] Resolution of this lesion was attributed to subtle hemodynamic changes induced by surgical excision of the meningioma.[20] Similar hemodynamic alterations in our patient may have contributed to vessel and tissue ischemia and, consequently, led to tumor hemorrhage and eventual necrosis.

Third, craniotomy and contralateral tumor resection might have led to resolution of the untouched lesion. Distant disease control following resection of the primary lesion has been shown for other tumors, mainly for renal cell carcinoma.[21,22] For such lesions, Freed et al. postulated that surgical resection of the primary tumor leads to activation of macrophages, lymphocytes and immunoglobulin secretion causing an immunomediated response that promotes regression of disseminated disease.[22] However, such control has never been demonstrated after resection of meningiomas.

## 4. Conclusion

Hemorrhage within a meningioma is rare, but is associated with increased morbidity and mortality. Spontaneous resolution of a meningioma after hemorrhage is very rare, and to our knowledge, this is the first report. Although the definitive etiology of tumor resolution remains unclear, it is possible that hemorrhage led to necrosis, postsurgical perturbations of the dural vasculature caused infarction and hemorrhage, or an immune-mediated process occurred. Indeed, any traumatic event within a tumor, hemorrhage or otherwise, may lead to the activation of multiple processes that result in tumor resolution. Regardless of the mechanism, we feel that a stable intratumoral hemorrhage of a meningioma does not necessarily require surgical intervention, if signs of mass effect or elevation of intracranial pressure are absent. These lesions can be managed conservatively and may, in rare circumstances, resolve.

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