Benign Sphenoid Wing Meningioma Presenting with an Acute Intracerebral Hemorrhage – A Case Report

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ABSTRACT

BACKGROUND AND STUDY OBJECT: We report an unusual case of a benign lateral sphenoid wing meningioma that presented with, and was masked by, an acute intracerebral hemorrhage.

CASE REPORT: A 68-year-old woman was admitted after sudden onset of coma. Computed tomography (CT) revealed an intracerebral hemorrhage, without any underlying vascular pathology on CT angiography. During the surgery, we found a lateral sphenoid wing meningioma with intratumoral bleeding that extended into the surrounding brain parenchyma.

RESULTS: We removed the hematoma and resected the tumor completely in the same session. The histopathological classification of the tumor was a WHO grade I meningothelial meningioma. The patient recovered very well after surgery, without significant neurological sequelae.

CONCLUSIONS: Having reviewed the relevant references from the medical literature, we consider this event as an extremely rare presentation of a benign sphenoid wing meningioma in a patient without any predisposing medical factors. The possible mechanisms of bleeding from this tumor type are discussed.

KEYWORDS: intracranial meningioma, intracerebral hemorrhage, brain tumors, sphenoid wing

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Introduction

Intracranial meningiomas are commonly encountered in neurosurgical practice. Most of these tumors are benign lesions with a slow growth. They usually present with clinical symptoms evolving over time, though the severity of the symptoms is dependent on the tumor's location and size. In contrast to intracranial malignancies such as high-grade gliomas and metastases, an acute intracranial hemorrhage – intracerebral, subdural, or subarachnoid – from a benign intracranial meningioma is considered a rare event. Its main occurrences in the literature are case reports or very small series,[1–19] and its reported relative incidence is 1.3%.[20] Other reported cases presented with either subacute or chronic subdural bleeding.[21–23]

In this report, we present a case of a patient with a benign lateral sphenoid wing meningioma presenting with a large intracerebral hemorrhage, causing acute onset of symptoms and requiring urgent surgery. The patient has given consent for publication of this report.

Case Report

The patient was a 68-year-old woman without any significant previous medical comorbidity except for hypertension that was well controlled by monotherapy medication. She was not taking any antiplatelet medication or anticoagulants.

She was found comatose in her own home, after not having talked to anyone since the previous evening. The Glasgow Coma Scale score was seven on admission to the local hospital, with small equal pupils and spontaneous movement in all four extremities, although possibly less on the right side. She was intubated and transported to our institution. On admission, the right pupil showed reaction to light, but almost no reaction was observed in the left pupil; in addition, the corneal reflex was present on the right, but not on the left side. The coagulation profile from blood tests was normal.

Her computed tomography (CT) scan showed an intracerebral hemorrhage $45 \times 67 \times 54$ mm, localized mainly in the left temporal lobe, but extending also to the basal part of the left frontal lobe, and giving approximately 1 cm midline shift toward the right side (Figs. 1A and B). Although the hemorrhage was centered around the left Sylvian fissure, CT angiography revealed neither intracranial aneurysm nor arteriovenous malformation, only a dislocation of the middle cerebral artery and marked vasculature in the lateral part of the hematoma (Fig. 1C). Due to the patient's

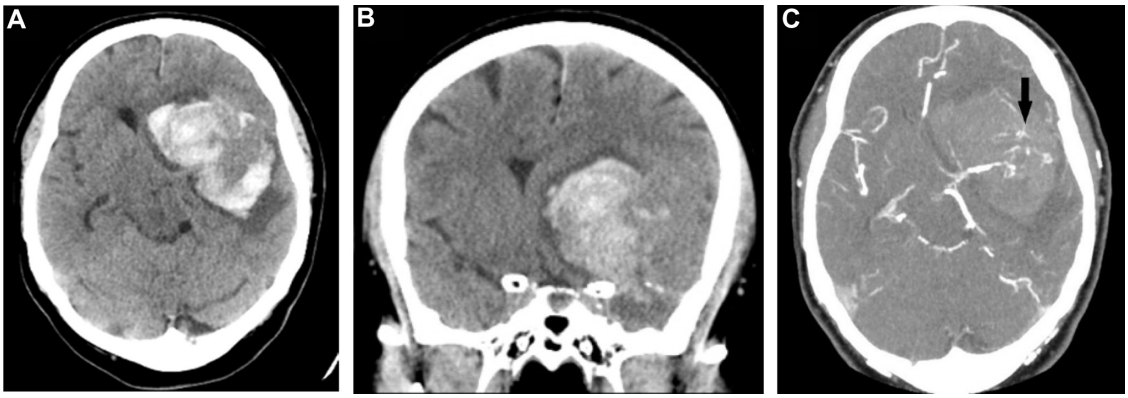

Figure 1. Preoperative CT scan showing the extent of the hemorrhage in the (**A**) axial and (**B**) coronal plane as well as (**C**) marked vasculature in the lateral part of the hematoma (black arrow), masking the underlying tumor.

poor condition, there was no time for emergency magnetic resonance imaging.

The patient was immediately operated on. After performing a left pterional craniotomy and opening of the dura, we identified a typical-looking meningioma attached to the lateral sphenoid wing, with the hematoma hidden medially behind the tumor. The Sylvian fissure was opened, and the tumor was dissected free and removed by standard microsurgical technique. The blood clot was then evacuated. We noticed that the tumor was intact and typically stromatous in its lateral part but was obviously destroyed by bleeding more medially, giving a clear impression of a true intratumoral hemorrhage with extension into the adjacent brain tissue. Due to the patient's acute condition, her age, and the risk of postoperative cerebrospinal fluid leakage, we decided not to excise the dura underneath the tumor on the sphenoid wing, thus classifying this tumor removal as Simpson grade II, although no macroscopically visible tumor remains were left in place.

The CT taken on the following day (Fig. 2) showed no significant remaining hematoma; there were signs of ischemic damage in the surrounding brain tissue, but the cerebral vasculature was open without vasospasms. After withdrawal of the patient's sedation, she woke up gradually during the next two days and was discharged from our hospital on the fifth postoperative day; thereafter, she was fully awake and oriented, with only mild dysphasia and very mild right hemiparesis. At the time of latest clinical follow-up, eight months after surgery, she had no hemiparesis and was fully active, although she suffered from minor cognitive problems and very mild sensory dysphasia.

The histopathological classification of the tumor was a meningioma. Microscopy revealed an epithelial membrane antigen (EMA)- and progesterone receptor (PGR)-positive tumor build up of meningothelial cells exhibiting whorled growth, no mitoses, and Mib labeling index of far less than 1%, morphologically corresponding to a meningothelial meningioma, WHO grade I. Tumor tissue was seen between meningeal vessels and growing into fibrous connective tissue interpreted as dura. Focally, degenerative features such as nuclear atypia and myxoid degeneration demonstrated by Alcian Blue stain were present, whereas tumor and vessel wall necrosis and vessel wall and brain tissue invasion were absent (Figs. 3A–C).

Discussion

In some cases, the coincidence of an asymptomatic meningioma and spontaneous intracerebral hemorrhage might have an alternative explanation, such as in elderly patients with significant medical comorbidities. However, in the present case, we found clear evidence intraoperatively of a true intratumoral hemorrhage with extension into the cerebral parenchyma.

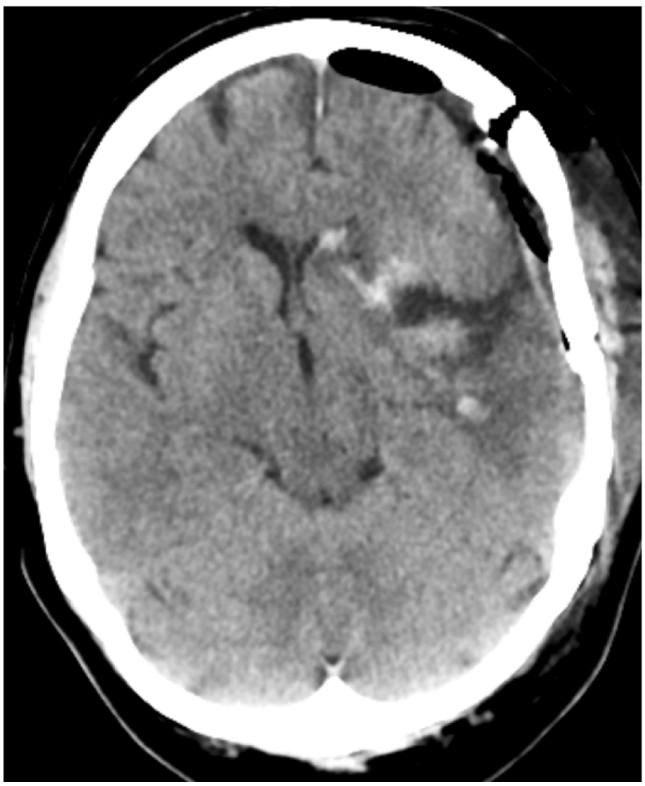

Figure 2. CT control one day after surgery.

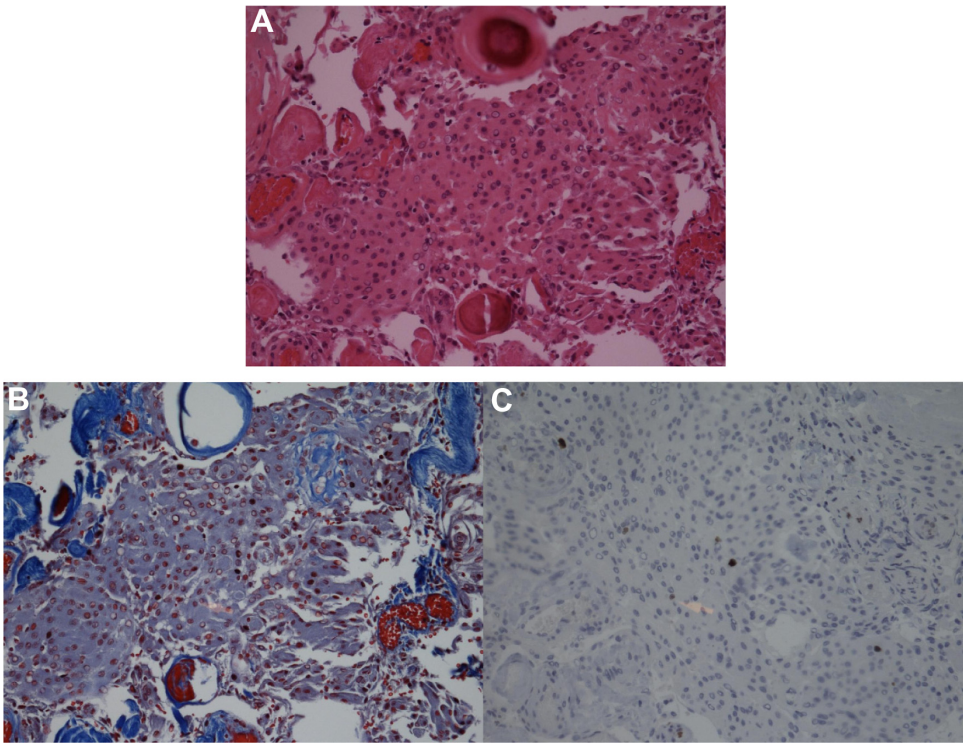

Figure 3. Histopathological findings supporting the diagnosis of meningothelial meningioma (WHO grade I) in the (**A**) hematoxylin/eosin, (**B**) trichrome (connective tissue), and (**C**) Ki67 (proliferation marker) staining.

Furthermore, the localization of the hematoma was not typical for hypertensive bleeding.

As mentioned above, an acute intracranial hemorrhage is an extremely rare presentation of a benign meningioma. In an autopsy series of 430 spontaneous intracerebral hematomas, 44 cases (10.2%) were caused by a proved neoplasm, and, of those proved cases, only one meningioma was present (2.3%).[24] Several possible mechanisms of bleeding related to an underlying meningioma have been suggested as follows: tumor infarction with secondary bleeding,[6] direct tumor invasion into one of the cerebral arteries,[25] mechanical stretching and distortion of cortical bridging veins,[7] as well as histamine-related vasodilation[23] or venous hypertension due to occlusion of the venous sinus.[12] We assume that one of the first two mechanisms mentioned here was probably the case in our patient, although not being exactly proved by histopathological findings.

It is also unclear whether some specific subtype of a benign meningioma should be more prone to intratumoral bleeding.[6,8,26,27]

In the analysis of 143 published cases and two cases of their own study, Bosnjak et al.[28] found that, interestingly, increased bleeding tendency in intracranial meningiomas was associated with two age groups (<30 years and >70 years, respectively), convexity and intraventricular locations, and fibrous subtype. Only 3 cases of intracerebral hemorrhage out of 10 sphenoid wing meningiomas with bleeding were mentioned in this review, making our own case one of the very few published to this date.[2,17]

The documented historical overall mortality rate due to meningioma-associated hemorrhages was reduced from 21.1% to 13.9% after the advent of the widely available CT scanning. Expected mortality in the post-CT era is now lower (7.5%) in surgically treated cases, and, in patients conscious at presentation, the mortality appears to be similar (<3%) to elective cases of meningiomas without documented bleeding.[28]

Conclusion

The presented case of an acute intracranial hemorrhage from an underlying benign sphenoid wing meningioma is one of the very few ever published. Urgent evacuation of the intracerebral hemorrhage and optimally total removal of the underlying tumor in the same session, whenever feasible, seems to be an optimal treatment strategy in cases of meningiomas presenting with acute intracerebral bleeding. Given the rarity of this event, however, the impact on decision-making with regard to surgical indication in patients with incidental, asymptomatic intracranial meningiomas would be doubtful.

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Author Contribution

Conceived and designed: RF. Analyzed the data: RF, JKH, EAA. Wrote the first draft of the manuscript: RF. Contributed to the writing of the manuscript: RF, JKH, EAA.

Agree with manuscript results and conclusions: RF, JKH, EAA. Jointly developed the structure and arguments for the paper: RF, JKH, EAA. Made critical revisions and approved final version: RF. All authors reviewed and approved the final manuscript.

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