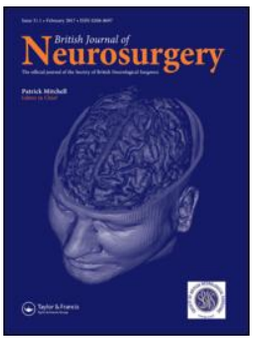

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SHORT REPORT

Very small vestibular schwannoma as the source of fatal subarachnoid hemorrhage: a case report

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ABSTRACT

Background: Subarachnoid hemorrhage (SAH) is commonly caused by an aneurysm, trauma, other vascular diseases, and infrequently by a metastatic tumor or glioma. SAH due to a benign intracranial tumor, such as a vestibular schwannoma (VS), is rare. We report a case in which a very small (1 mm) VS caused fatal SAH.

Case presentation: A 75-year-old woman presented with a sudden severe headache. Computed tomography showed SAH at the right of the cerebellopontine angle. On post-onset day (POD) 27, MRI revealed a 1-mm mass on the cerebellopontine angle's right side. She was discharged with House–Brackmann grade 4 right-side facial weakness and hearing disturbance. She re-presented on POD 45 with headache and loss of consciousness. Computed tomography revealed massive SAH and intracerebellar hemorrhage. She died 4 days later. Histopathological evidence indicated a highly vascular vestibular schwannoma.

Conclusions: Vestibular schwannoma should therefore be considered a source of SAH, particularly in patients with facial weakness and/or hearing disturbance.

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KEYWORDS
Vestibular schwannoma; subarachnoid hemorrhage; case study

Background

Subarachnoid hemorrhage (SAH) is commonly caused by an aneurysm, trauma, and other vascular diseases and infrequently by a metastatic tumor or glioma. SAH due to a benign intracranial tumor, such as a vestibular schwannoma (VS), is rare. Intratumoral hemorrhage in VS occurs in <1% of VSs.[1] Clinically significant SAH or intracerebral hemorrhage occurs in <25% of patients in whom intratumoral hemorrhage is seen.[2–8] The mechanism of hemorrhage in a VS is not completely understood, although some risk factors seem to contribute to it. These factors include (1) a large size (>2 cm), (2) presence of the mixed Antoni type, (3) dilated thin-walled vessels, (4) high vascularity. Rapid growth of the VS is also a possible factor.[9] We report a case in which a very small (1 mm) VS caused fatal SAH.

Case presentation

A 75-year-old woman was admitted to our hospital, presenting with a sudden, severe headache and a history of hypertension. She has never taken any anti-thrombotic medication before. Physical examination revealed neck stiffness but no focal neurological deficits. Blood tests showed no hemostasis disorders. Initial computed tomography (CT) showed SAH in the interpeduncular cistern and on the right side of the cerebellopontine angle (CPA) (Figure 1(a)). CT angiography was negative for an aneurysm and vascular malformations (Figure 1(b)). The day after admission, digital subtraction angiography was performed (Figure 1(c,d)), which revealed no abnormalities. Conservative therapy was maintained.

On day 2, she complained of right-sided facial weakness, which was diagnosed as House–Brackmann grade 2, and hearing disturbance. Her symptoms gradually became more pronounced. Magnetic resonance imaging (MRI) without contrast medium enhancement was performed on post-onset day (POD) 27 and revealed a 1-mm mass in the right side of the CPA. It showed high intensity on fluid-attenuated inversion recovery imaging, high intensity on MR angiography source imaging, and high intensity with a hemosiderin rim on T2-weighted imaging (Figure 2(a–c)). The mass appeared to be what remained of the SAH. The patient was then discharged from the hospital now having House–Brackmann grade 4 right-side facial weakness and hearing disturbance.

On POD 45, she re-presented to the hospital with a sudden, severe headache followed by loss of consciousness. CT showed massive SAH (Figure 2(d)). She died 4 days later.

The histopathological examination showed no aneurysms, dissections, or vascular malformations in the vertebral artery or basilar artery. A hemorrhagic mass attached to the right vertebral artery was seen on the right side of the CPA (Figure 3(a,b)). Histopathologically, the spindle cells were positive for S-100 protein (Figure 3(c)). There were also relatively large areas positive for CD34, indicating hypervascularity (Figure 4(a)). Other areas showed numerous macrophages containing dark-brown hemosiderin pigment, indicating hemoglobin breakdown from older hemorrhagic episodes (Figure 4(b)). Taking these facts into consideration, we suspected that the small vestibular schwannoma was the source of the fatal SAH.

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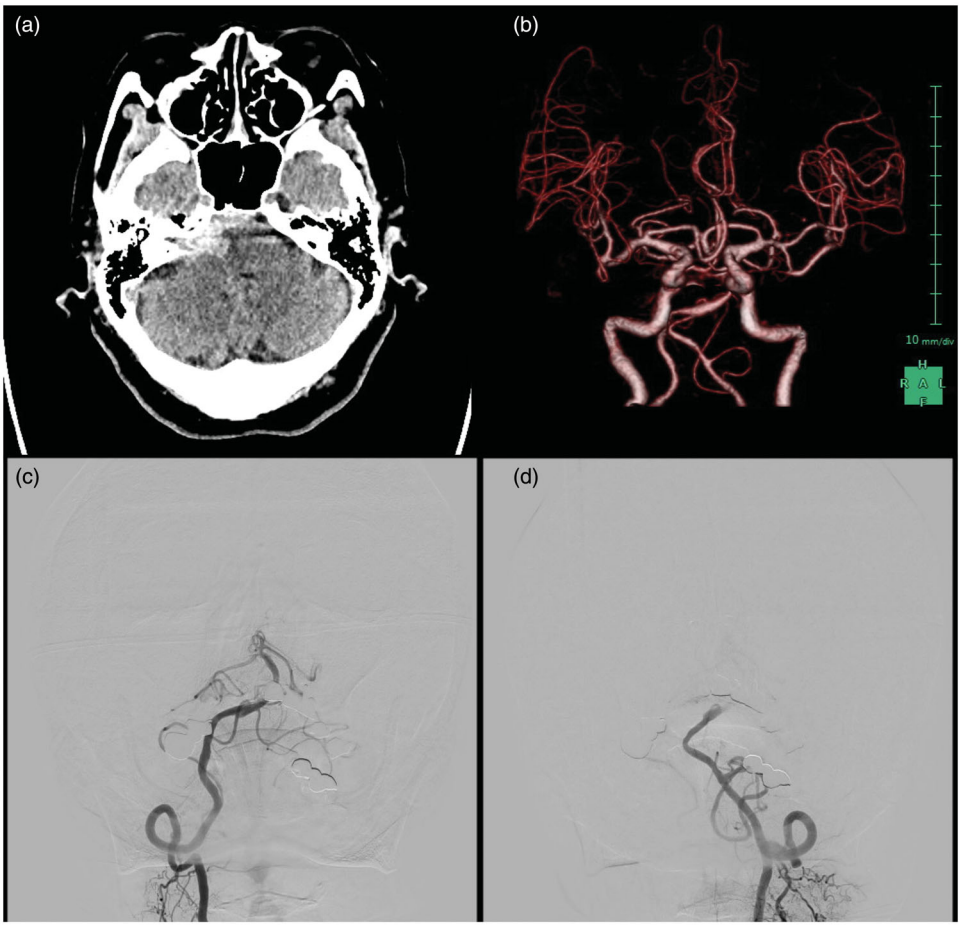

Figure 1. (a) Computed tomography (CT) on the day of admission shows subarachnoid hemorrhage (SAH) in the interpeduncular cistern and on the right side of the cerebellopontine angle (CPA). (b) CT angiography shows no aneurysms, dissections, or vascular malformation. Right (c) and left (d) vertebral angiography shows no abnormalities.

Discussion

SAH secondary to an intracranial tumor is rare. Hemorrhage from intracranial tumors accounts for 1%–11% of all intracranial hemorrhage and is mostly accounted for by aggressive tumors.[10,11] There have been only a few reports of SAH associated with a VS.[2,6,9–16] Carlson *et al.* reported that tumor (VS)-associated clinically significant hemorrhage occurred in 0.4% of cases at their institution.[1] The risk of clinically significant hemorrhage is greater in VS patients who are under anticoagulant treatment compared with the general VS population. Other factors also seem to contribute to the occurrence of hemorrhage such as a large size (>2 cm) of the VS, mixed Antoni type, dilated thin-walled vessels, high vascularity, and rapid growth of the tumor.[9]

In our case, an especially small (1 mm) VS caused massive SAH. As far as we can determine, this report describing fatal SAH due to a 1-mm VS is unique. The patient was not on any anticoagulants, and CT angiography and digital subtraction angiography showed no vascular malformations or tumor staining. The histopathological findings revealed the presence of a hypervascular vestibular schwannoma.

SAH of unknown etiology is relatively common, representing approximately 10%–15% of all non-traumatic SAH. Patients with perimesencephalic, sulcal, and CT-negative hemorrhage of unknown etiology generally have good prognoses, whereas those with intraventricular or diffuse SAH with a poor Fisher grade have worse short- and long-term clinical outcomes.[17] The literature contains several reports of VS patients with SAH who presented with symptoms associated with cranial nerves VII and VIII.[9,10,12,13,15,18,19] Some of the patients also presented with right-side facial weakness and hearing disturbance starting several days after the onset of the initial symptoms. Hence, it is necessary to investigate the causes of SAH of unknown etiology with every modality available if the patient's symptoms become increasingly pronounced and CT reveals diffuse SAH.

Conclusions

We reported a rare case of a very small VS being the source of fatal SAH. Tumor-derived SAH tends to have a more benign course than aneurysmal SAH. Although it is unlikely that a benign tumor such as VS causes fatal SAH, VS should be added to the list of causes of SAH of unknown origin, particularly in patients presenting with facial weakness or hearing disturbance.

Ethics approval and consent to participate

The authors certify that this study was performed in accordance with the ethical standards as laid down in the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards.

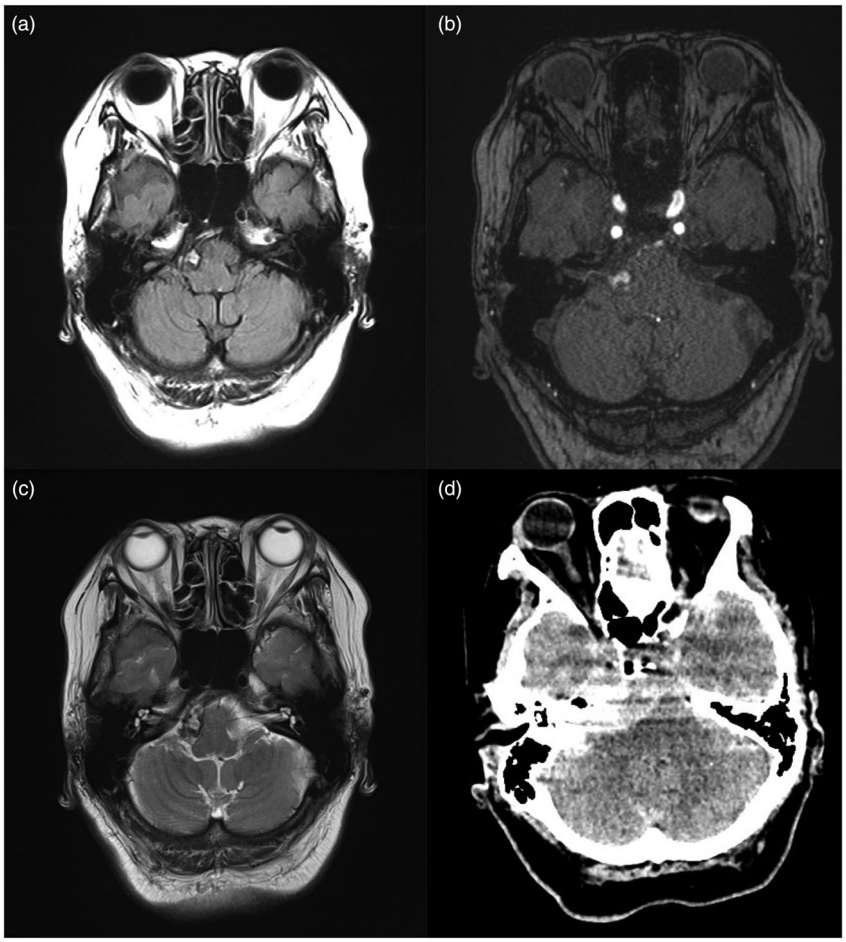

Figure 2. Axial fluid attenuated inversion recovery imaging (a), magnetic resonance angiography source imaging (b), and T2-weighted imaging (c) show a small, 1-mm mass near the CPA attached to the pons. d CT shows massive SAH.

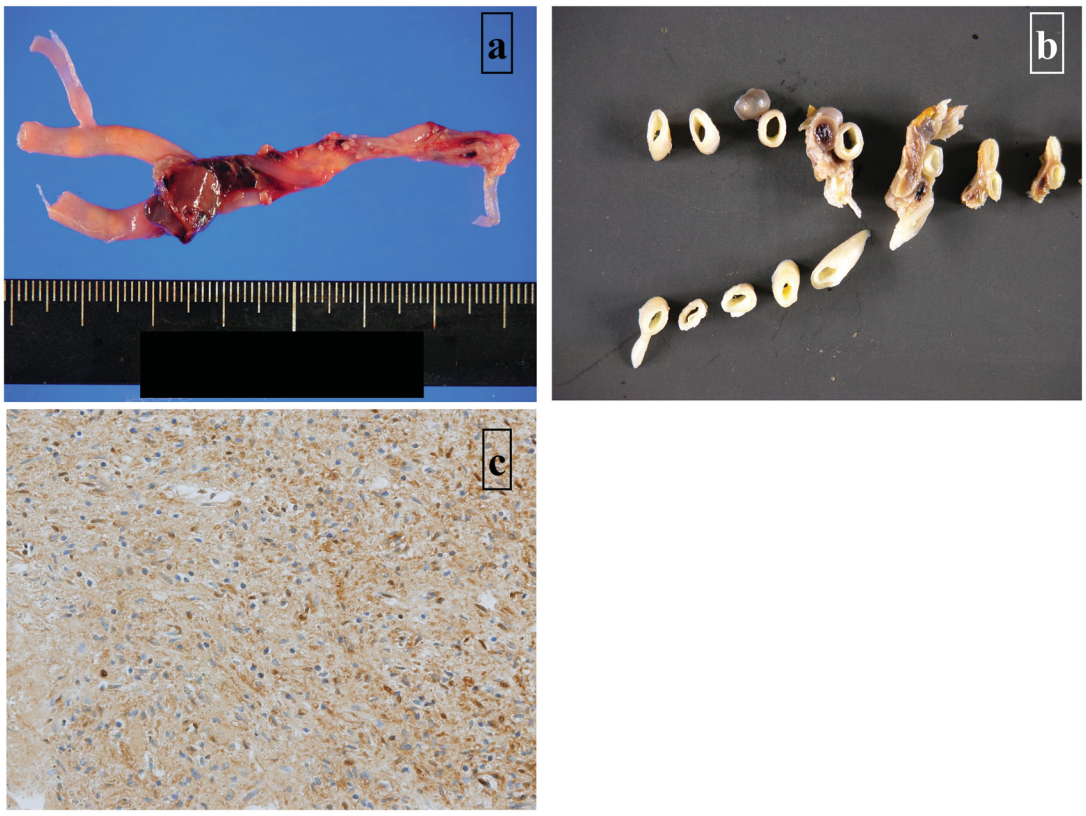

Figure 3. Histopathological findings. (a) Hemorrhagic mass attached to the right vertebral artery was located at the right side of the CPA. (b) There were no aneurysms, dissections, or vascular malformations in the vertebral artery or basilar artery. c Spindle cells were positive for S-100 protein.

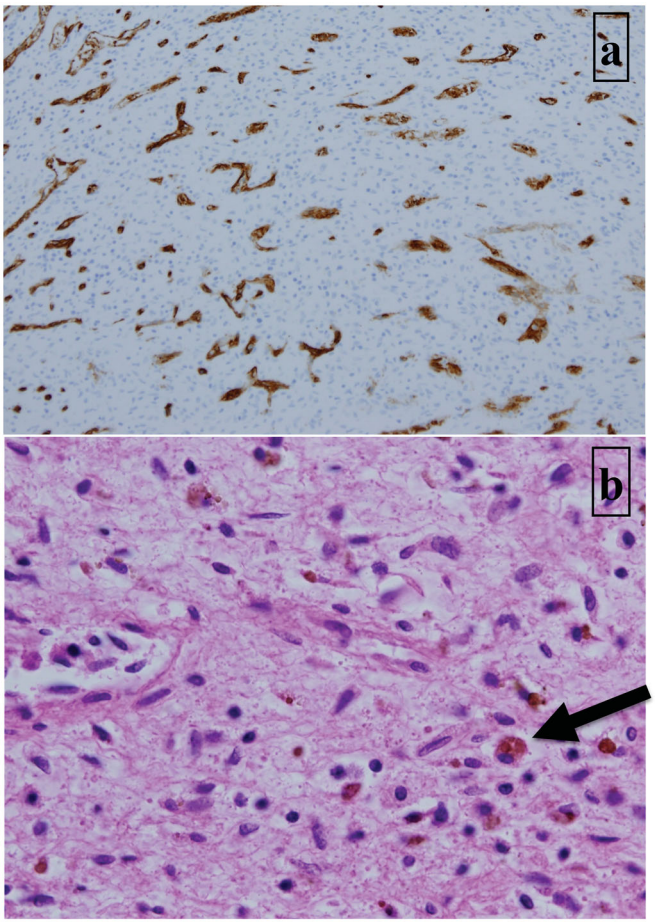

Figure 4. (a) Histopathological analysis shows relatively large areas that are positive for CD34, indicating hypervascularity. (b) There were numerous macrophages (*arrow*) containing dark-brown hemosiderin pigment, indicating hemoglobin breakdown during previous hemorrhagic episodes.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Acknowledgment

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Author contributions

Masafumi Kuroiwa: Writing, original draft preparation and editing.
Takahiro Murata: Supervision, validation.
Shuichi Hirayama: Supervision.
Masanobu Hokama: Supervision.
Toshihiko Miyashita: Supervision.

Disclosure statement

No potential conflict of interest was reported by the author(s).

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

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