Case Report

Cervical Schwannoma Presenting with Acute Intracranial Subarachnoid Hemorrhage

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A subarachnoid hemorrhage (SAH) associated with negative finding on four-vessel angiography is seen in 5 to 30% of patients with intracranial SAH. A previously silent lesion in the spinal canal may be responsible for the angiographically negative finding for cause of intracranial SAH. We report a case of upper cervical (C1-2) intradural schwannoma presenting with acute intracranial SAH. Repeated cerebral angiographic studies were negative, but cervical magnetic resonance imaging study and tissue pathology revealed a intradural-extramedullary schwannoma in C1-2 level. This case illustrates the importance of a high index of clinical suspicion for spinal disease in angiographically negative intracranial SAH patients.

KEY WORDS: Spinal schwannoma · Subarachnoid hemorrhage

INTRODUCTION

Spinal cord tumors, either primary or metastatic, may cause subarachnoid hemorrhage (SAH), with ependymoma of the conus medullaris accounting for most of these cases^{1,4)}. Spinal nerve sheath tumors such as schwannomas rarely cause SAH¹⁻⁹⁾, especially in the absence of spinal cord or root symptoms. Parmar et al.⁹⁾ stated that until 2004 only 20cases of nerve sheath tumors which developed SAH had been reported in the literature.

We report a rare case of cervical schwannoma presenting clinically with intracranial SAH and discuss the mechanisms of SAH.

CASE REPORT

A 44-year-old man presented with sudden onset of severe headache, neck pain after paroxysmal sneezing with intact mental status. Physical examination was normal except for nuchal rigidity. Computed tomography of the head revealed hyperattenuation in the cisterna magna, pre-pontine cistern and 4th ventricle consistent with acute SAH (Fig. 1). Cerebral angiography was performed on two occasions, 1 and 10 days after admission, but both studies were negative for aneurysm and vascular malformations (Fig. 2). Also, there were no vasospasm. On direct systemic review of spinal symptom, he was suffered from intermittent vague occipital



Fig. 1. Initial findings of the brain computed tomography (CT). The Brain CT reveals diffuse high density subarachnoid hemorrhage in the cistern magna, pre-pontine cistern and 4th ventricle.

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and neck pain, without precipitating or aggravating events. 12 days after admission, magnetic resonance image scan of the cervical spine was performed to rule out an intraspinal hemorrhage lesion. An ovoid shaped intradural-extramedullary lesion was identified within canal at the C1-C2 level (Fig. 3A-D). After the intravenous administration of

gadolinium, focal nodule and capsule were enhanced. Demarcation between the lesion and the compressed spinal cord was also demonstrated on the T1- weighted image with gadolinium enhancement (Fig. 3E, F). Two weeks after admission, we performed surgery for removal of tumor. A midline skin incision was made from the torcula area to the

A B

Fig. 2. A: Preoperative 3-dimentional computed tomography angiography demonstrating negative for aneurysm and vascular malformations. B: Ten days after admission, follow-up bilateral verterbral angiography was done without definitive lesion.

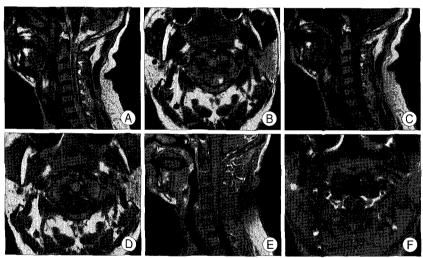


Fig. 3. Preoperative cervical spine magnetic resonance image. A and B: T2-weighted image show intermediate signal intensity with focal necrotic or cystic portion. C and D: T1-weighted image show well defined homogenous low signal intensity mass lesion at Lt lateral epidural space of C2 level. E and F: After the intravenous administration of gadolinium, focal nodule and capsule are faintly enhanced.

C4 in the prone position with head fixation. As soon as partial laminectomy of the atlas and axis was carefully performed, the presence of a bulged dura overlying tumor was identified and a midline vertical incision was made on the dura. And the intradural-extramedullary tumor was disclosed and compressed spinal cord (Fig. 4A). The shape and consistency of the tumor was ovoid, solid encapsulated and $2 \times 1 \times 1$ cm sized. The tumor had no dural attachments, and a small posterior nerve root was identified entering mass. The tumor was completely removed by microsurgical technique without damaging nerve roots. Hemosiderin staining was noted in the posterior portion of cervical cord. No abnormal vessels or dilated venous structures were found. The gross total removal of the mass was accomplished without neurological deficit (Fig. 4B). A pathological examination revealed a benign schwannoma (Fig. 4C).

DISCUSSION

Spinal causes of SAH are rare, representing 1.5% of all cases of SAH⁹. The more common causes include





Fig. 4. A: Operative finding demonstrating a encapsulated ovoid solid well-vascularized mass adherent to the nerve roots of the cervical spinal cord. B: Complete removal of tumor by microsurgical technique without damaging nerve roots. Hemosiderin staining is noted in the posterior portion of cervical cord. C: Histopathologic finding revealing typical schwannoma. The figure shows high cellular area of elongated spindle cells with long cytoplasmic processes arranged in interwoven fascicles (Antoni A area) and loose hypocellular portion with myxoid and microcystic changes (Antoni B area). Nuclei of tumor cells are small round and regular (H & E stain, × 100).

spinal trauma, arteriovenous malformation and saccular aneurysms of spinal arteries. On occasion, spinal cord tumors, either primary or metastatic, may cause SAH, with ependymoma of the conus medullaris accounting for most of these cases. Nerve sheath tumors may also cause SAH, but it is exceedingly rare for these intradural lesions to come to clinical attention with only intracranial SAH without spinal symptoms9. Parmar et al.9 reviewed the 20 reports of spinal nerve sheath tumors that caused SAH, they found only 7 cases including their case presented exclusively with intracranial SAH like our patient. Among the 7 cases, intracranial SAH was developed from the cervical schwannoma in 4 cases and from the cauda equina schwannoma in 3 cases. In our case, patient suffered from suboccipital headache for 1 year without any other spinal symptoms. An intracranial SAH was developed immediately after paroxysmal sneezing. During the operation, we were not able to find abnormal vascular finding in spinal cord and tumor except for the hemosiderin staining on the dorsal portion of cervical cord. Histologically, tumor was schwannoma.

The exact mechanism of initiation of hemorrhage in schwannomas remains obscure⁸⁾, but two theories have been proposed. First theory is a mechanical cause^{1,5)}. Cervoni et al.¹⁾ reported that 79% of the 63 tumors associated with SAH were situated in the region of the filum terminale and medullary conus and even thought tumors of this region account for only 20% of spinal tumors and the acute onset of SAH after physical exercise. Movements of traction along the spine column involving the cervical region may stretch the most superficial portion of the tumor including its vascular connections with the adjacent structures, especially when traction is repeated and prolonged¹⁾. Such tractional force occur most commonly in areas of high mobility, and this probably explains why the onset of symptoms in 50% of cases is related to exertion^{7,9)}. The

other theory proposed that hyalinized ectatic vessels seen in schwannoma undergo spontaneous thrombosis⁴⁾. This is followed by distal tumor necrosis complicated by hemorrhage. In our case, we could not find the abnormal ectatic vessels in the tumor. For our case, the former may be more appropriate to explain the mechanism of SAH from cervical schwannoma.

CONCLUSION

We must always consider that there is a possibility for cause of SAH owing to silent spinal lesion in angiographicnegative intracranial SAH patients like this case.

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