Tentorial meningioma and painful tic convulsif

Case report

HIDENORI OGASAWARA, M.D., SHUICHI OKI, M.D., HIROAKI KOHNO, M.D., SEIICHIRO HIBINO, M.D., AND YOKO ITO, M.D.

Department of Neurosurgery, Hiroshima City Asa Hospital, Hiroshima, Japan

A case is presented of painful tic convulsif caused by a posterior fossa meningioma, with right trigeminal neuralgia and ipsilateral hemifacial spasm. Magnetic resonance images showed an ectatic right vertebral artery as a signal-void area in the right cerebellopontine angle. At operation the tentorial meningioma, which did not compress either the fifth or the seventh cranial nerves directly, was totally removed via a suboccipital craniectomy. The patient had complete postoperative relief from the trigeminal neuralgia and her hemifacial spasm improved markedly with decreased frequency. From a pathophysiological standpoint, the painful tic convulsif in this case was probably produced by the tumor compressing and displacing the brainstem directly, with secondary neurovascular compression of the fifth and seventh nerves (the so-called “remote effect”).

KEY WORDS • painful tic convulsif • trigeminal neuralgia • hemifacial spasm • meningioma

Posterior fossa tumors (neurinomas, meningiomas, and epidermoids) are likely to cause trigeminal neuralgia and facial spasm.7,11,16 Painful tic convulsif was first described in 1920 by Cushing3 in those patients displaying a combination of facial pain and ipsilateral hemifacial spasm. Since that time, only a few cases of painful tic convulsif have been reported. To our knowledge, there have been only eight cases of these tumors causing this condition. In these eight patients described in the literature, the tumors directly compressed the fifth and/or seventh cranial nerves. We present a ninth case, a patient who suffered from painful tic convulsif caused by a tentorial meningioma that did not involve the cerebellopontine angle, and a review of the literature on the unusual pathophysiology in this case.

Case Report

This 79-year-old woman was referred to our department for neurological assessment in June 1993. She had a 3-year history of right-sided hemifacial spasm and severe episodic pain on the right side of the maxillary region brought on by brushing her teeth and eating. Her facial pain increased despite treatment with carbamazepine (400 mg/day) given over a 2-year period.

Examination. Her physical condition was unremarkable and hematological and biochemical studies were normal. Neurological examination revealed right-sided hemifacial spasm and severe neuralgia in the region of the second branch of the right trigeminal nerve. A trigger point for touch was found in the right maxillary area. No cerebellar signs were noted. Neurootological assessment indicated bilateral high-frequency sensorineural hearing loss. Magnetic resonance (MR) images revealed a 4-cm round mass displacing the fourth ventricle and distorting the brainstem in the right cerebellar hemisphere. The tumor enhanced homogeneously with gadolinium-diethylenetriamine pentaaetic acid administration and was attached to the inferior surface of the tentorium cerebelli (Fig. 1). An ectatic right vertebral artery and its branch could be detected as a signal-void area in the right cerebellopontine angle (Fig. 2). A right brachial angiogram was remarkable only for tumor stain in the right posterior fossa (Fig. 3).

Operation. The patient underwent a right suboccipital craniectomy in July 1993. A large tumor was attached to the inferior surface of the tentorium cerebelli, but did not directly compress the cranial nerves. The right cerebellopontine angle was not inspected during the operation. The tumor was gross totally removed, and a histopathological
examination revealed that the tumor was a transitional meningioma.

Postoperative Course. Both the right-sided hemifacial spasm and trigeminal neuralgia gradually improved after operation. Six months later, the patient’s right-sided trigeminal neuralgia had resolved completely and her hemifacial spasm was markedly improved, with decreased frequency.

Discussion

Taken alone, either trigeminal neuralgia or hemifacial spasm is well recognized. This neuralgia is commonly caused by compression of the trigeminal nerve at its entry zone by ectatic arteries. Neurovascular compression of the facial nerve at its entry zone is similarly thought to be responsible for hemifacial spasm.

The coexistence of trigeminal neuralgia and ipsilateral hemifacial spasm is uncommon. Since Cushing’s description of this condition as “painful tic convulsif,” few cases have been reported. Iwasaki, et al., reported that the incidence of painful tic convulsif has been eight of 800 cases of hemifacial spasm and 400 cases of trigeminal neuralgia. The majority of reported cases of painful tic convulsif have had vascular abnormalities around the fifth and seventh cranial nerves, such as ectatic vertebral or basilar arteries, arteriovenous malformations, and aneurysms.

Tumors causing painful tic convulsif are extremely rare, with only eight cases reported until now (Table 1). In these cases, seven patients had epidermoid cysts and only two, including our case, had meningiomas. Moreover, all tumors reported to cause painful tic convulsif have involved the cerebellopontine angle tumors. In the cases of epidermoid cysts, the fifth and seventh cranial nerves were surrounded or compressed by the tumor. The capsule and contents of the tumor, which are irritants to the fifth and seventh nerves, might have caused trigeminal neuralgia and hemifacial spasm. Cook and Jannetta have reported a meningioma that was pushing the anterior inferior cerebellar artery into the root entry zone of the seventh nerve. The tumor was removed and the artery mobilized and padded away by muscle, and the patient’s facial pain and spasm improved.

In our case, the tentorial meningioma did not directly compress either the fifth or seventh nerve. The tumor caused compression, distortion, and rotation of the brainstem. In addition, MR images showed distortion and rotation of the brainstem, an ectatic right vertebral artery, and an elongated loop of the branches of the vertebral or basilar artery. This distortion and rotation of the brainstem was corrected, and the painful tic convulsif was relieved by removing the tumor without vascular decompression of the root entry zone of either nerve. Therefore, the distortion and rotation of the brainstem may have caused vascular structures (possibly the branches of the right verte-
bral or basilar artery) to compress the fifth and seventh nerves at the root entry zone.

When painful tic convulsif is caused by tumor, two different mechanisms might cause symptoms. In a cerebellopontine angle tumor, the tumor directly compresses the fifth and/or seventh nerve. In a posterior fossa tumor not involving the cerebellopontine angle, the tumor compresses and displaces the brainstem directly, causing secondary neurovascular compression of the fifth and seventh nerves (the so-called "remote effect"). Therefore, at operation for painful tic convulsif caused by a posterior fossa tumor not involving the cerebellopontine angle, decompression of the fifth and seventh nerves by removing the tumor might be the first choice. Additional microvascular decompression of the root entry zone of one of these nerves might be necessary if facial pain and/or hemifacial spasm remain despite tumor removal.

References


Manuscript received June 3, 1994.
Address reprint requests to: Hidenori Ogasawara, M.D., Department of Neurosurgery, Hiroshima City Asa Hospital, 2–1–1, Kabeiminami, Asakita-ku, Hiroshima 731–02 Japan.