



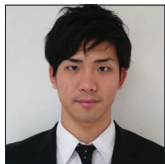
Case Report

Hemichorea induced by a sphenoid ridge meningioma

Hiroataka Inoue¹, Rihito Yamamura², Kazumichi Yamada³, Tadashi Hamasaki¹, Nobuhiro Inoue⁴, Akitake Mukasa¹

¹Department of Neurosurgery, Kumamoto General Hospital, Yatsushiro, ²Department of Neurosurgery, Amakusa Medical Center, Amakusa, ³Division of Speech-Language and Hearing Therapy, Faculty of Health Science, Kumamoto Health Science University, Kita-Ku, ⁴Department of Neurosurgery, Kumamoto Neurosurgical Hospital, Chuo-Ku, Kumamoto, Japan.

E-mail: *Hiroataka Inoue - hiro2866@gmail.com; Rihito Yamamura - r.arcturus@gmail.com; Kazumichi Yamada - yamadakazu1202@gmail.com; Tadashi Hamasaki - thamasaki-nsu@umin.ac.jp; Nobuhiro Inoue - ninoue@knh.co.jp; Akitake Mukasa - mukasa-nsu@umin.ac.jp



*Corresponding author:

Hiroataka Inoue,
Department of Neurosurgery,
Kumamoto General Hospital,
Yatsushiro, Kumamoto, Japan.

hiro2866@gmail.com

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ABSTRACT

Background: Movement disorders are rare in brain tumors. We describe a 45-year-old woman with hemichorea, a concomitant contralateral sphenoid ridge meningioma.

Case Description: The meningioma enlarged as her hemichorea worsened, and after meningioma resection, the hemichorea gradually subsided. N-isopropyl-p-[123I]-iodoamphetamine single-photon emission computed tomography performed preoperatively showed decreased regional cerebral blood flow (CBF) to the basal ganglia circuit ipsilateral to the tumor and, when repeated postoperatively, confirmed improved regional CBF.

Conclusion: We propose that the enlarging sphenoid ridge meningioma had a remote effect on regional CBF and the thalamocortical motor center and that complex changes in the basal ganglia output may have caused the hemichorea.

Keywords: Cerebral blood flow, Chorea, Meningioma, Remote effect

INTRODUCTION

Chorea is defined as a hyperkinetic movement disorder, characterized by involuntary, brief, random, and irregular muscle contractions.^[3] Brain tumors presenting with chorea are rare,^[2] and to the best of our knowledge, there has only been one case of chorea with a meningioma reported to date.^[10] Herein, we report the clinical course and discuss the mechanism underlying this phenomenon.

CASE PRESENTATION

Our patient was a 45-year-old woman who experienced twitching of the left lip, anarthria, and abnormal movements involving the distal part of her left limb 2 years previously. A diagnosis of chorea was made after consultation with the department of neurology, and the symptoms failed to improve despite administration of clonazepam. A small meningioma arising from the right sphenoid ridge was discovered. The meningioma enlarged over time [Figures 1a-c], alongside worsening symptoms [Video 1]. She was referred to our department for surgical treatment of the tumor. However, she tested positive for anticardiolipin antibody and lupus anticoagulant preoperatively. She was diagnosed with anti-phospholipid syndrome (APS); however, the D-dimer level (0.8 µg/mL) and platelet count (169,000 /µL) were normal. The coagulation disorder did

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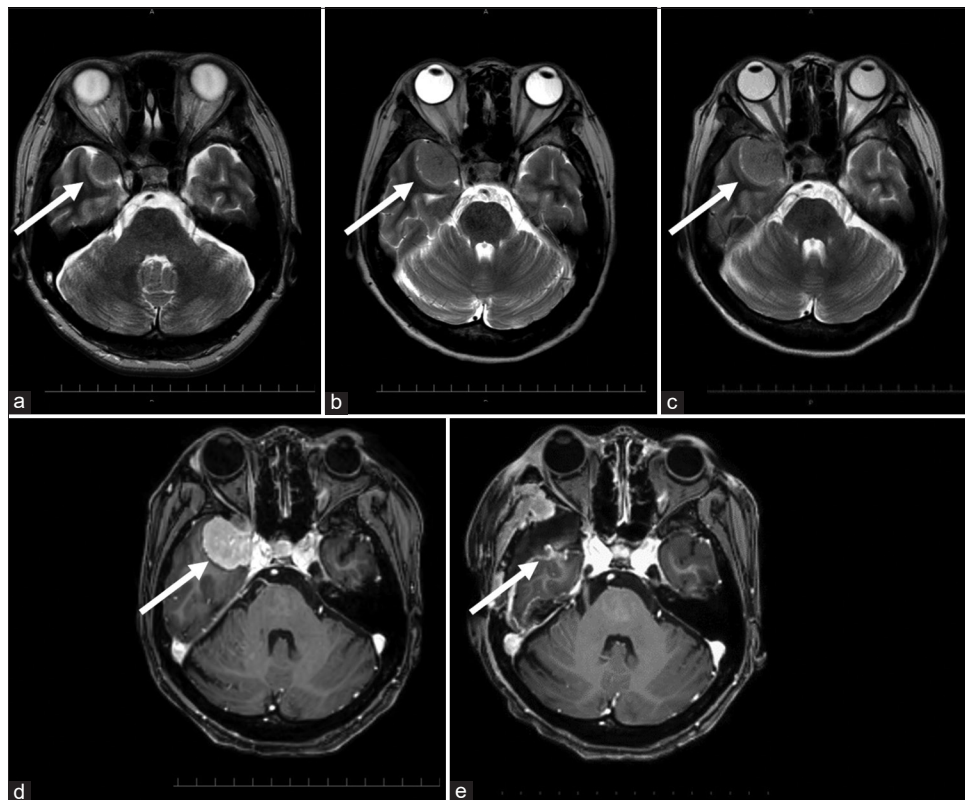


Figure 1: Preoperative T2-weighted magnetic resonance imaging (MRI) performed at the age of 43 (a), 44 (b), and 45 years (c) shows an enlarging mass lesion in the right middle cranial fossa. (d) Preoperative postcontrast T1-weighted MRI. (e) Postoperative postcontrast T1-weighted MRI.

not require treatment, and it was considered safe to proceed with neurosurgery. The tumor was resected without any complications; a small amount of tumor was left on the dura around the superior orbital fissure. The postoperative magnetic resonance imaging (MRI) demonstrated complete resolution of the mass effect on the brain [Figures 1d and e]. Her symptoms gradually improved, and she was able to return to work 7 months after the surgery [Video 2]. Her Unified Dyskinesia Rating Scale score also improved from 56/104 to 18/104. N-isopropyl-p-[^{123}I]-iodoamphetamine single-photon emission computed tomography (^{123}I -IMP SPECT) performed before the surgery demonstrated a decrease in the regional cerebral blood flow (CBF) of the right ventral midbrain, right thalamus, and frontotemporal lobe cortex. ^{123}I -IMP SPECT performed 5 months after the surgery revealed a slight improvement in the regional CBF [Figure 2].

DISCUSSION

Hemichorea generally occurs as a complication of a vascular malformation, basal ganglia tumor, stroke, metastasis, or metabolic disorders.^[2] None of these were present in our patient, who only had a sphenoid ridge meningioma and

APS. Only one case of chorea with a meningioma has been reported to date; however, the postoperative clinical course was not described.^[10] Interestingly, 1.3% of patients with APS have choreiform symptoms.^[4]

We believe that the meningioma caused hemichorea in our patient for the following reasons. First, the laterality of hemichorea corresponded to the tumor location. Second, the hemichorea worsened when the meningioma increased in size, and subsided after surgery, while the APS worsened throughout this period. Third, after tumor resection, the reduced CBF in the midbrain and thalamus normalized.

Movement disorders are rare clinical features of brain tumors.^[2] Movement disorders in astrocytomas^[9] and metastatic brain tumors^[6] of the basal ganglia and thalamus were reported. It seems that such areas, when damaged, may be more likely to produce movement disorders than others. However, Chorobski reported some cases that movement disorders occurred with the lesions involved only the cortical area.^[5] Two possible underlying mechanisms are known. First, movement disorders could be a direct result of basal ganglia pathway compression by the tumor and/or the resulting edema. Second, the symptoms could arise due to impaired vascular flow to the basal nuclei.^[11] The

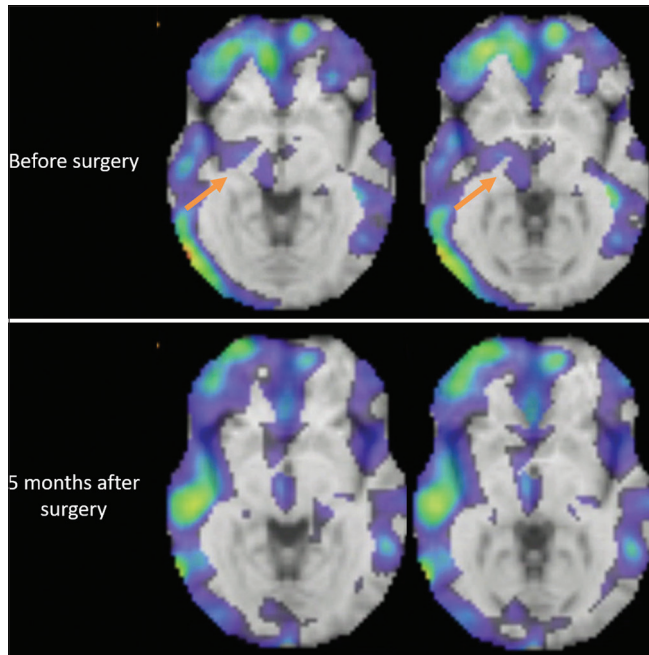


Figure 2: N-isopropyl-p-[123I]-iodoamphetamine single-photon emission computed tomography (123I-IMP SPECT) performed before surgery (upper panels) demonstrates decreased regional cerebral blood flow to the right ventral midbrain, thalamus (arrows), and frontotemporal lobe cortex. SPECT performed 5 months after surgery (lower panels) reveals a slight improvement.

meningioma in our patient occurred in the right sphenoid ridge. Therefore, it was difficult for the tumor or resulting edema to have directly compressed the basal ganglia pathway. Furthermore, the tumor was supplied by the artery of foramen rotundum, middle meningeal artery, and inferolateral trunk of the internal carotid artery. It would be difficult for such a plethoric tumor to impair vascular flow to the basal nuclei. However, the regional CBF to the right ventral midbrain and right thalamus decreased in our patient and improved after the resection of the tumor. Urasaki *et al.* described a case of paroxysmal kinesigenic choreoathetosis associated with a frontotemporal arachnoid cyst.^[12] The authors of that report suggested that the arachnoid cyst might affect the ipsilateral cortico-subcortical connections. Any disturbance in the cortical control of the striatum and its thalamic connection could induce paroxysmal kinesigenic choreoathetosis. Furthermore, Hwang *et al.* investigated the pathological mechanisms of hemichorea or hemiballism secondary to cortical lesions.^[7] They also suggested that cortical dysfunction might have an important role in the pathogenesis of cortical hemichorea or hemiballism, even without significant involvement of the basal ganglia and thalamus. Similar to these cases, localized brain lesions are known to cause neurological impairments in remote sites (a “remote effect”).^[8] Iwasa *et al.* confirmed that reduced 2-deoxy-2-[fluorine-18]-fluorodeoxyglucose uptake in the

ipsilateral thalamus was observed in 21% of the patients with brain tumors, and a simultaneous reduction in cortical metabolism around the tumor was observed in 70% of the patients.^[8] The meningioma in our patient may have had a similar remote effect, which would explain our aforementioned ¹²³I-IMP SPECT findings.

We used the Alexander and Crutcher model to determine the causal association of CBF change and hemichorea.^[1] Hemichorea has been speculated to result from a disinhibition of the motor thalamus. This disinhibition is caused by a reduced excitatory output from the subthalamic nucleus to the pallido-thalamo-cortical motor loop.^[3] In earlier reports, patients with hemichorea showed a significant CBF increase in the contralateral thalamus.^[14] We did not observe this phenomenon in our patient. Vitek *et al.* suggested that alterations in the firing rate and the pattern of neuronal discharges in the basal ganglia provoke anomalous activities throughout the pallido-thalamo-cortical circuit.^[13] Thus, an increase in CBF to the contralateral thalamus may not be necessary to induce hemichorea.

CONCLUSION

Herein, we report a rare case of hemichorea induced by a sphenoid ridge meningioma. The symptoms underwent remission after resection of the tumor. We believe that the growing meningioma had a remote effect, altering the firing pattern of the thalamocortical motor pathways and leading to the manifestation of hemichorea. APS can also cause hemichorea; however, the status of the APS in our patient did not correlate with her clinical course.

Ethical approval

The authors confirm that the approval of an Institutional Review Board was not required for this work. Written informed consent for the publication of this report was obtained from the patient, and this report does not identify the individual. We confirm that we have read the journal’s position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Alexander GE, Crutcher MD. Functional architecture of basal ganglia circuits: Neural substrates of parallel processing. *Trends Neurosci* 1990;13:266-71.
2. Bhatoe HS. Movement disorders caused by brain tumours. *Neurol India* 1999;47:40-2.
3. Cardoso F, Seppi K, Mair KJ, Wenning GK, Poewe W. Seminar on choreas. *Lancet Neurol* 2006;5:589-602.
4. Cervera R, Piette JC, Font J, Khamashta MA, Shoenfeld Y, Camps MT, *et al.* Antiphospholipid syndrome: Clinical and immunologic manifestations and patterns of disease expression in a cohort of 1,000 patients. *Arthritis Rheum* 2002;46:1019-27.
5. Chorobski J. Involuntary movements in patients with intracranial tumors. Their occurrence and possible pathogenesis. *Arch Neurol* 1962;6:27-42.
6. Glass JP, Jankovic J, Borit A. Hemiballism and metastatic brain tumor. *Neurology* 1984;34:204-7.
7. Hwang KJ, Hong IK, Ahn TB, Yi SH, Lee D, Kim DY. Cortical hemichorea-hemiballism. *J Neurol* 2013;260:2986-92.
8. Iwasa H, Murata Y, Nishimori M, Miyatake K, Tadokoro M, Kohsaki S, *et al.* Remote effects in the ipsilateral thalamus and/or contralateral cerebellar hemisphere using FDG PET in patients with brain tumors. *Jpn J Radiol* 2018;36:303-11.
9. Krauss JK, Nobbe F, Wakhloo AK, Mohadjer M, Vach W, Mundinger F. Movement disorders in astrocytomas of the basal ganglia and the thalamus. *J Neurol Neurosurg Psychiatry* 1992;55:1162-7.
10. Rana AQ, Yousuf MS, Hashmi MZ, Kachhvi ZM. Hemichorea and dystonia due to frontal lobe meningioma. *J Neurosci Rural Pract* 2014;5:290-2.
11. Salvati M, Frati A, Ferrari P, Verrelli C, Artizzu S, Letizia C. Parkinsonian syndrome in a patient with a pterional meningioma: Case report and review of the literature. *Clin Neurol Neurosurg* 2000;102:243-5.
12. Urasaki E, Tokimura T, Genmoto T, Yokota A. Paroxysmal kinesigenic choreoathetosis associated with frontotemporal arachnoid cyst-case report. *Neurol Med Chir (Tokyo)* 1999;39:169-73.
13. Vitek JL, Chockkan V, Zhang JY, Kaneoke Y, Evatt M, DeLong MR, *et al.* Neuronal activity in the basal ganglia in patients with generalized dystonia and hemiballismus. *Ann Neurol* 1999;46:22-35.
14. Yamada K, Harada M, Inoue N, Yoshida S, Morioka M, Kuratsu J. Concurrent hemichorea and migrainous aura-a perfusion study on the basal ganglia using xenon-computed tomography. *Mov Disord* 2008;23:425-9.

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