Case Report

Intraosseous Meningioma in a Woman Presenting with Seizure Mimicking Acute Multiple Cerebral Infarction: A Case Report

Shinichi Okonogi* Jun Nomoto Kei Uchino
Sayaka Terazono Yasuhiro Node Shunpei Ando
Daisuke Fukushima Hiroyuki Masuda Kosuke Kondo
Naoyuki Harada Masaaki Nemoto and Nobuo Sugo

Department of Neurosurgery (Omori), School of Medicine, Faculty of Medicine, Toho University

ABSTRACT: Intraosseous meningioma mainly develops in the cranial diploe. Incidence is very low — fewer than 1% of meningioma cases. We evaluated and treated a patient with intraosseous meningioma that was difficult to diagnose on preoperative imaging because of the presence of multiple concomitant lesions. The patient was a 66-year-old woman who was transported to our hospital for treatment of seizure. Emergent diffusion-weighted magnetic resonance imaging (MRI) revealed hyperintense regions in the right mesial temporal lobe and thalamus. On computed tomography (CT), a tumorous lesion with osteosclerotic features was present in the left frontal bone. Contrast-enhanced MR images obtained on the 9th hospital day showed multiple brain parenchymal lesions, in addition to the skull lesion. A new enhanced lesion was present in the right cerebellar hemisphere and required differentiation from malignant lymphoma, metastatic brain tumor, and metastatic skull tumor. The tumor was resected on the 30th day, and the histopathological diagnosis was fibrous meningioma. MRI enhancement of the multiple lesions in brain parenchyma decreased after surgery. Thus, these findings were classified as changes in cerebral infarction over time. The difficulty in diagnosis may have been due to the coexistence of different diseases — intraosseous meningioma and cerebral infarction — temporal change in contrast medium enhancement of cerebral infarct lesions, and the presence of multiple lesions.

Toho J Med 1 (4): 73–79, 2015

KEYWORDS: intraosseous meningioma, multiple lesions, cerebral infarction

Meningioma develops in various regions of the cranium and is frequently encountered in clinical neurosurgery.[1–7] Intraosseous meningioma mainly develops in the diploic layer, in the skull.[4–7] We assessed and treated a patient with intraosseous meningioma that was difficult to diagnose on preoperative imaging because of the concomitant presence of multiple additional lesions.

Case Presentation

Patient: 66-year-old woman

Chief complaint: Seizure

Past medical history: Untreated hypertension; no history of head injury

Familial medical history: Unremarkable

6–11–1 Omorinishi, Ota, Tokyo 143–8541, Japan
*Corresponding Author: tel: +81–(0)3–3762–4151
e-mail: shinichi.okonogi@med.toho-u.ac.jp
DOI: 10.14994/tohojmed.2015.009

Received July 9, 2015: Accepted Sept. 18, 2015
Toho Journal of Medicine 1 (4), Dec. 1, 2015.
ISSN 2189–1990, CODEN: TJMOA2

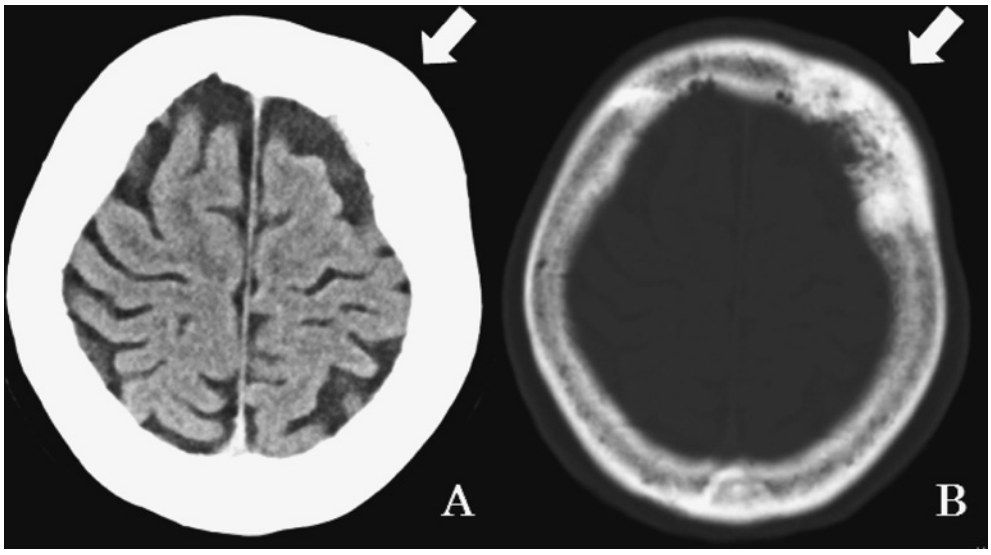

Fig. 1 Computed tomography (CT)
A: Plain CT image of the skull showing a lesion in the left frontal bone (white arrow)
B: Bone window CT image showing hyperostosis of a skull lesion (white arrow)

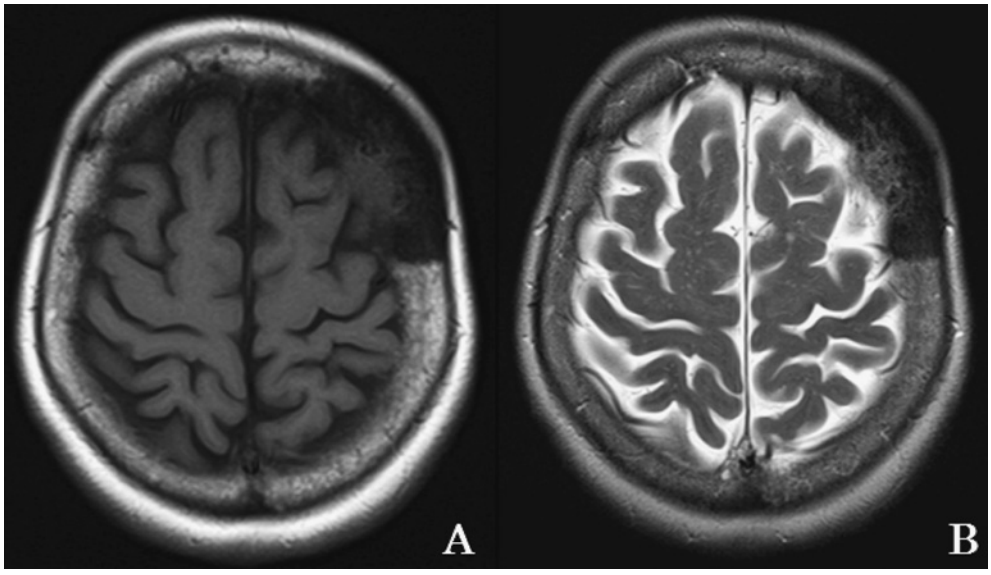

Fig. 2 Magnetic resonance imaging (MRI)
A: T1-weighted MR image showing an isointense lesion.
B: T2-weighted MR image showing a hypointense lesion with partial isointensity.

History of present illness: She suddenly developed a tonic–clonic seizure at about 13: 45 while doing housework in her kitchen; her husband requested an ambulance.

Neurological findings on admission: Consciousness level was Japan Coma Scale (JCS) I-1 and Glasgow Coma Scale (GCS) 15 (E4V5M6). Left homonymous hemianopsia and left unilateral spatial agnosia were observed. The Barre sign was positive in the left upper limb.

Test findings on admission

Computed tomography (CT) revealed no abnormal findings in the cranium (Fig. 1A), but a tumorous lesion measuring 6 cm in the left frontal bone exhibited hyperostosis (Fig. 1B). The lesion was isointense on T1-weighted magnetic resonance (MR) images obtained on the same day (Fig. 2A) and hypointense on T2-weighted images, with partial isointensity (Fig. 2B), as compared with brain parenchyma. On diffusion-weighted imaging (DWI), hyperin-

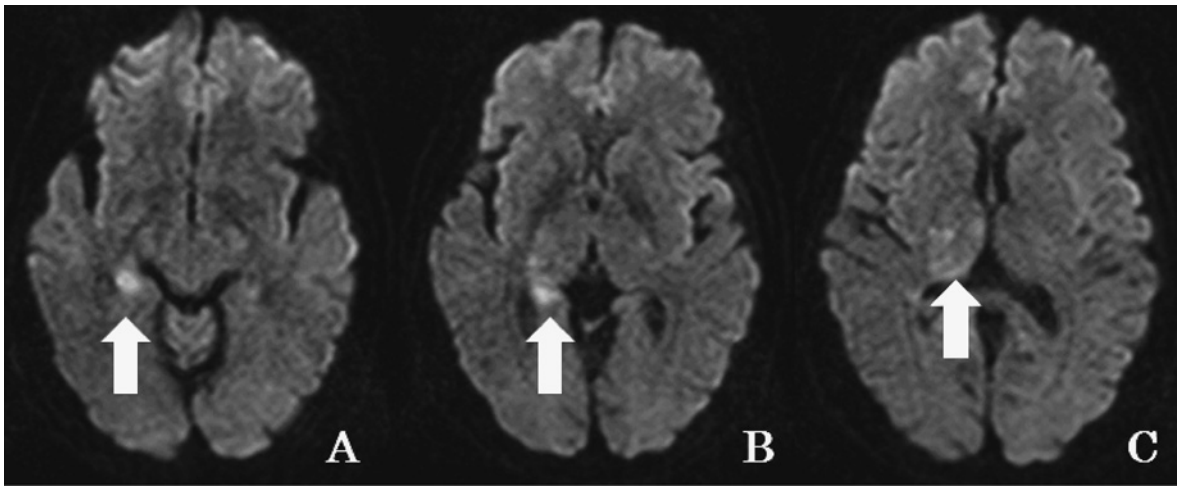

Fig. 3 Diffusion-weighted images (DWI) with strong signals
A: Medial site of the right temporal lobe (white arrow)
B: Corpus callosum (white arrow)
C: Right thalamus (white arrow)

tense regions were noted in the right mesial temporal lobe, over the thalamus and corpus callosum (Fig. 3A-C). These regions were hypointense on apparent diffusion coefficient (ADC) mapping. The lesion was isointense, with partial hyperintensity, on DWI. The results of general blood testing and plain chest radiography findings were within normal ranges, and atrial fibrillation was detected on electrocardiography.

Course after admission

No seizures occurred after admission. On electroencephalography, 6- to 7-Hz slow waves were dominant in the right temporal region, and no clear spike wave was seen. Cerebral infarction was diagnosed because the intracranial lesions were hyperintense on DWI, and treatment was started. Cardiogenic embolism was considered the cause of cerebral infarction because atrial fibrillation was detected on electrocardiography. Findings for the intracranial lesions suggested that left homonymous hemianopsia was caused by impairment of the right thalamus lateral geniculate body.

On gadolinium (Gd)-enhanced MR images obtained on the 9th hospital day (Fig. 4A-F), the skull lesion was heterogeneously enhanced and a dural tail sign was observed. Enhancement was noted in the right mesial temporal lobe, over the thalamus, corpus callosum, and skull lesion, and a new enhanced lesion was present in the right cerebellar hemisphere. No abnormality was observed in the right cerebellar hemisphere on MR images obtained on admission, but a new lesion was seen in Gd-enhanced MR images and was hyperintense on T2-weighted imaging. Additional examinations were performed to differentiate these multiple lesions from malignant lymphoma, metastatic brain tumor, and metastatic skull tumor. Blood tumor marker levels were carbohydrate antigen (CA) 125, 15.7 U/ml (normal range, 0.0-35.0 U/ml); CA19-9, 3.4 U/ml (0.0-37.0 U/ml); CA 72-4, 1.9 U/ml (<8.0 U/ml); carcinoembryonic antigen (CEA), 2.0 ng/ml (0.0-5.0 ng/ml); neuron-specific enolase (NSE), 12.3 ng/ml (<16.3 ng/ml); anti-p53 antibody, <0.40 U/ml (<1.3 U/ml); sequamous cell carcinoma (SCC), 1.0 ng/ml (<1.5 ng/ml); and serum interleukin 2 receptors (S-IL2R), 279 U/ml (220-530 U/ml); no clinically significant elevation was observed. Cerebrospinal fluid testing showed a cell count of 4 per microliter (monocytes, 100%); protein, 32 mg/dl (normal range, 10-40 mg/dl); and glucose, 57 mg/dl (50-75 mg/dl), which were all within normal ranges. With regard to malignant lymphoma markers, cerebrospinal fluid β2-microglobulin level was 2.1 mg/l (1.0-1.9 mg/l) and S-IL2R was <54.5 U/ml (220-530 U/ml) — both within normal ranges. On nuclear medical examination, a mild increase in tracer uptake was observed in the left frontal region on bone scintigraphy, but no morbid high-level accumulation was noted in any other region. On single-photon emission CT using ^{123}I-N-isopropyl-p-iodoamphetamine (^{123}I-IMP), ^{123}I-IMP was not incorporated into the lesion during the early or delayed phases. Gallium scintigraphy, upper gastrointestinal endoscopy, and chest and abdominal CT were performed to identify the primary lesion, but all findings were normal. To make a definite diagnosis, the skull

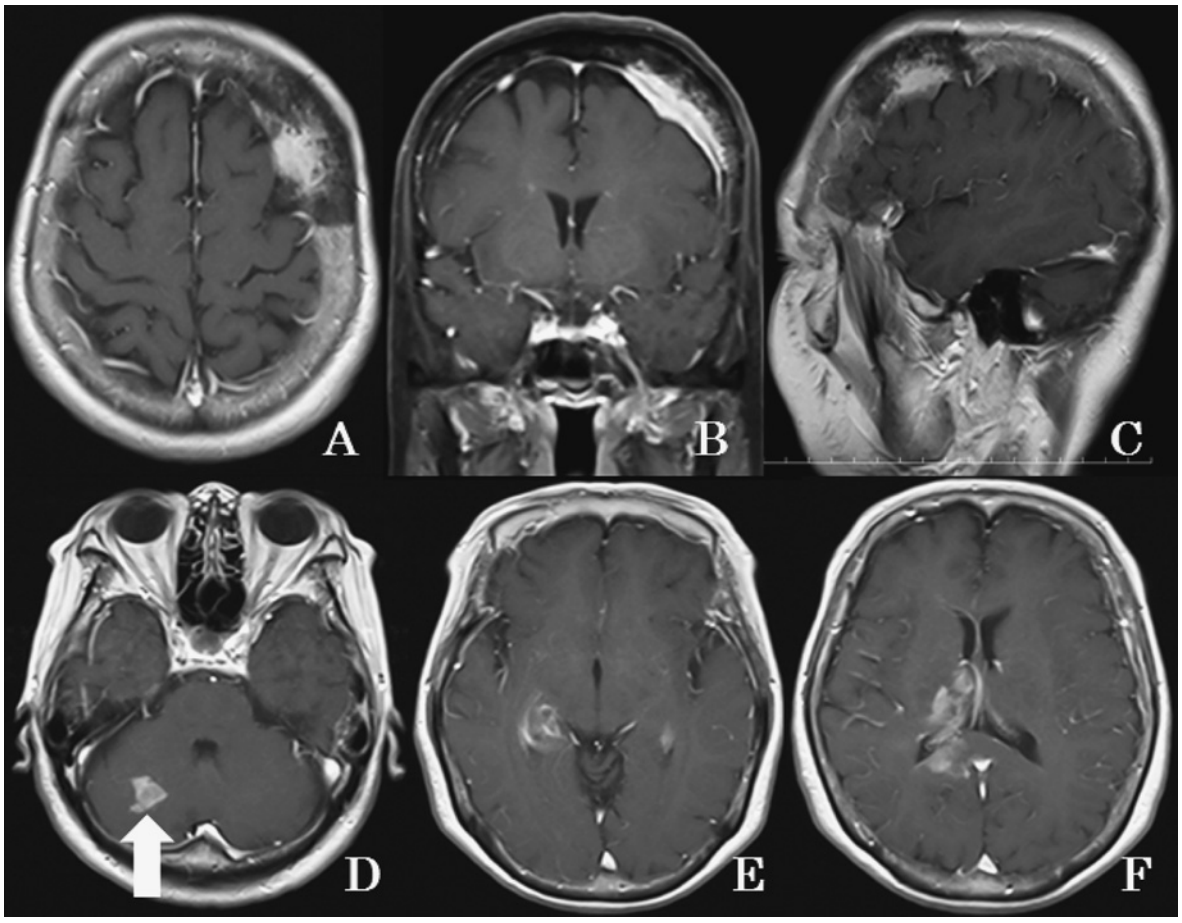

Fig. 4 Gadolinium (Gd)-enhanced magnetic resonance (MR) images obtained on the 9th day after onset
MR image showing multiple enhanced brain lesions, including the skull tumor (A–F). B shows only fat suppression.

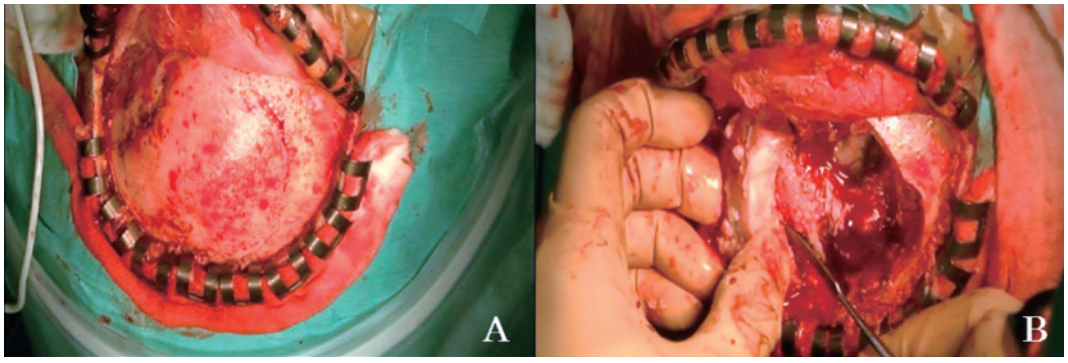

Fig. 5 Operative photographs (A, B)
A: The cranial tumor was distended and white.
B: The cranial tumor and dura mater strongly adhered.

tumor was excised on the 30th day. The bone tumor was pale white, and the bone and dura mater strongly adhered to each other. The tumor was resected in a range wider than the dural region enhanced on preoperative MR imaging (MRI) (Fig. 5A, B). Pathologically, whorl formation and psammoma bodies were noted on hematoxylin-eosin staining, and the tumor was diagnosed as fibrous meningioma (Fig. 6A). The MIB-1 index was lower than 1% (Fig. 6B). The postoperative course was favorable, and the patient was able to walk without assistance at discharge. Gd-

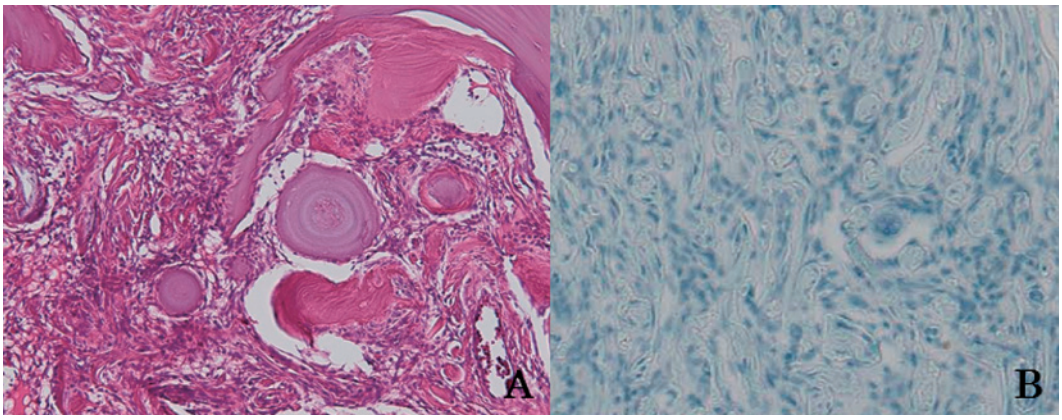

Fig. 6 Pathological findings
A: Hematoxylin-eosin (HE)-stained section showing fibrous meningioma with psammoma bodies and whorl formation. (×40)
B: The MIB-1 index was less than 1% on MIB-1 staining. (×10)

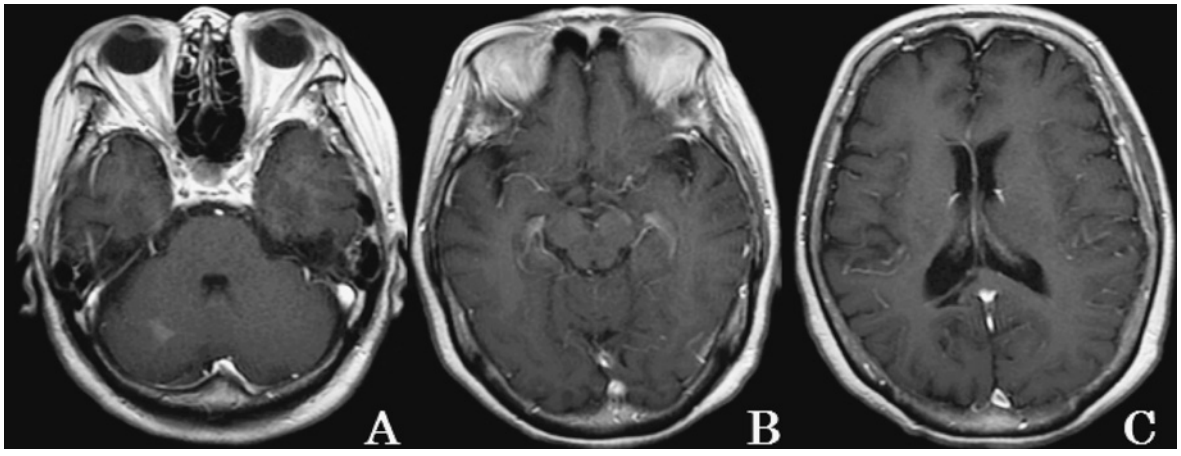

Fig. 7 Contrast-enhanced magnetic resonance (MR) images obtained 4 months after onset
The multiple lesions are no longer contrast-enhanced (A–C).

enhanced MR images obtained 4 months after onset confirmed the disappearance of the enhanced lesions in the right mesial temporal lobe, over the thalamus, corpus callosum, and right cerebellar hemisphere. All lesions were hyperintense on T2-weighted images, which indicated that these may have been temporal changes in cerebral infarction (Fig. 7A-C).

Discussion

Meningiomas that develop outside the dura mater, *e.g.*, in the skin, paranasal and nasal cavities, parotid gland, oral cavity, or skull, are collectively termed extracalvarial meningiomas.[1–8] Those that mainly present in the skull are referred to as intraosseous meningiomas.[5] Intraosseous meningiomas mainly develop in the diploic layer of the skull and are rare, accounting for fewer than 1% of all meningioma cases.[2–5] The mean age of patients is 45 years, and incidence in females is 2 times that of males. A frequent site of development is near the suture of the skull.[4–7] Three pathogenic mechanisms are believed to be responsible: (1) part of the dura mater is incorporated into the bone suture during embryonic development, (2) the dura mater is incorporated into a fracture caused by trauma and becomes tumorous, and (3) a tumor develops in the outer layer of the dura mater and grows in the skull, along the bone suture or a fracture region.[4–7] The mechanism in our patient may have been (1) or (3) because she had no past history of trauma.

The initial symptom was seizure, and two different diseases were present: cerebral infarction and intraosseous meningioma. Intraosseous meningioma is asymptomatic in most cases. Seizures are rare and occur when the tumor

grows in the skull and exerts pressure on the brain.[7, 9] In contrast, the frequency of seizure is relatively high (2-33%) during the early phase of cerebral infarction. In addition, it has been reported that seizures can be induced by cerebral infarctions in the cortex and subcortical region, *e.g.*, the temporal, parietal, and frontal lobes, and also in the thalamus, lenticular nucleus, and caudate nucleus.[10] Because brain displacement due to intraosseous meningioma was not observed in our patient, electroencephalographic findings and the patient's course suggest that the seizure was caused by cerebral infarction.

The presence of multiple lesions observed on contrast MR images obtained on the 9th day, and the identification of a new lesion in the right cerebellar hemisphere, complicated preoperative imaging-based diagnosis. In general, enhancement of brain parenchyma is mainly observed in gray matter after cerebral infarction.[11] Contrast enhancement begins to appear during the subacute phase, 7 days after onset, but tends to appear earlier in infarctions in the thalamus and basal ganglia and may be observed earlier than 7 days after onset.[11] The mechanism responsible for enhancement of brain parenchyma during the subacute phase has been reported to be leakage of contrast medium from the immature blood-brain barrier of new blood vessels growing in necrotized tissue during the cleaning process.[11] The contrast effect disappears during the chronic phase, 30 days after onset and later.[11, 12] In our patient, there was no MRI enhancement of the multiple brain lesions at 4 months after onset, which suggests that these findings were changes in cerebral infarction over time. Although it is unclear why the lesion in the right cerebellar hemisphere was not visualized on DW images obtained at the initial examination, we assume that an embolus later formed in this region after the first seizure.

Intraosseous meningiomas exhibit hyperostosis on CT, which requires differentiation from metastatic skull tumor.[7] The incidence of metastatic skull tumor is the highest of all skull tumors, and the most frequent primary lesion is breast cancer (48% of cases), followed by lung cancer (14%), and head and neck cancer (12%).[7] Cranial metastases of prostate cancer, renal cancer, hepatocellular carcinoma, and malignant lymphoma have also been reported.[7] Osteolytic lesions with unclear boundaries are a characteristic CT finding in bone metastases of malignant tumors, but prostate cancer is visualized as hyperostosis.[7] Because 65% and 35% of intraosseous meningioma cases exhibit hyperostosis and osteolytic findings, respectively, such cases are often difficult to differentiate from metastatic skull tumor.[13] In many cases, intraosseous meningioma shows low isointensity on T1-weighted MR images and iso-and hyperintensity on T2-weighted MR images, and is heterogeneously enhanced.[14, 15] However, a hypointensity was noted on T2-weighted images of this patient. Intraosseous meningioma exhibits varied signal intensity, from low to high, on T2-weighted MR images, depending on the grades of the collagen component, psammoma body, and cell density.[14, 15] Because osteogenic tumors, such as metastatic skull tumors, also exhibit hypo- to isointensity on T1-weighted images and low-hyperintensity on T2-weighted images, differentiation between intraosseous meningiomas and osteogenic tumors may be challenging.[4, 14, 15] When differentiation from metastatic skull tumor is difficult, as in our patient, pathological diagnosis by active surgical resection should be considered.

Conclusion

We evaluated and treated a woman with intraosseous meningioma who presented with seizure mimicking acute multiple cerebral infarction. Diagnosis was complicated by the presence of different diseases — intraosseous meningioma and cerebral infarction — temporal change in contrast medium enhancement of cerebral infarction, and the presence of multiple lesions.

References

1) Tsutsumi S, Izumi H, Yasutomo Y, Ito M. Convexity en plaque meningioma manifesting as subcutaneous mass: cace report. Neurol Med Chir (Tokyo). 2013; 53: 727-9.
2) Yun JH, Lee SK. Primary osteolytic intraosseous atypical meningioma with soft tissue and dural invasion: report of a case and review of literatures. J Korean Neurosurg Soc. 2014; 56: 509-12.
3) Ilica AT, Mossa-Basha M, Zan E, Vikani A, Pillai J, Gujar S, et al. Cranial intraosseous meningioma: spectrum of neuroimaging findings with respect to histopathological grades in 65 patients. Clin Imaging. 2014; 38: 599-604.
4) Nanto M, Tsuji N, Miki J, Tanaka S, Uematsu Y, Itakura T. [A case of intraosseous meningioma with extracranial progression having difficulty in making a preoperative diagnosis]. No Shinkei Geka. 2005; 33: 51-6. Japanese.
5) Azar-Kia B, Sarwar M, Marc JA, Schechter MM. Intraosseous meningioma. Neuroradiology. 1974; 6: 246-53.
6) Lell M, Tudor C, Aigner T, Kessler P. Primary intraosseous meningioma of the mandible: CT and MR imaging features. AJNR Am J Neuroradiol. 2007; 28: 129-31.
7) Kawamoto K, Kawakami K, Iwase M, Suyama T, Li Q, Nakashima Y. [Metastatic skull tumor, epidermoid cyst, dermoid cyst, and intraosseous meningioma]. No Shinkei Geka. 2004; 32: 895-903. Japanese.
8) Siegel GJ, Anderson PJ. Extracalvarial meningioma. case report.

J Neurosurg. 1966; 25: 83-6.
9) Turner OA, Laird AT. Meningioma with traumatic etiology. report of a case. J Neurosurg. 1966; 24: 96-8.
10) Camilo O, Goldstein LB. Seizures and epilepsy after ischemic stroke. Stroke. 2004; 35: 1769-75.
11) Crain MR, Yuh WT, Greene GM, Lose DJ, Ryals TJ, Sato S, et al. Cerebral ischemia: evaluation with contrast-enhanced MR imaging. AJNR Am J Neuroradiol. 1991; 12: 631-9.
12) Uchino A, Miyoshi T, Ohno M. Fogging effect and MR imaging: a case report of pontine infraction. Radiat Med. 1990; 8: 99-102.
13) Nishijima Y, Utsunomiya A, Suzuki S, Endo T, Suzuki I, Nishimura S, et al. [A case of intraosseous and subcutaneous meningioma transforming head like a rugby ball]. No Shinkei Geka Janaru. 2012; 21: 32-8. Japanese.
14) Paek SH, Kim SH, Chang KH, Park CK, Kim JE, Kim DG, et al. Microcystic meningiomas: radiological characteristics of 16 cases. Acta Neurochir (Wien). 2005; 147: 965-72.
15) Lee HY, Prager J, Hahn Y, Ramsey RG. Intraosseous meningioma: CT and MR appearance. J Comput Assist Tomogr. 1992; 16: 1000-1.