Magnetic Resonance Imaging and Histology of a Large Cystic Meningioma

—Case Report—

Akira WATANABE, Ryoji ISHII, Kotaro TAKE, Yasuo SUZUKI,
Kazuhiro HIRANO, Hironari OKAMURA, Masaki KAMADA, and
Teruo SHIRABE*

*Departments of Neurosurgery and *Neuropathology, Kawasaki Medical School,
Kurashiki, Okayama*

Abstract

A 9-year-old boy presented with a meningioma containing a large cyst. Magnetic resonance (MR) imaging demonstrated tumor cells in the solid part of the tumor, part of the cyst wall, and cyst septum. The presence of the tumor cells in the thin cyst wall or the cyst septum is important to recognize preoperatively, and MR imaging is very useful to differentiate intratumoral cysts from peritumoral cysts in meningiomas.

Key words: cystic meningioma, magnetic resonance imaging, tumor cells

Introduction

Cystic meningiomas are somewhat rare, accounting for only 1.7–4.6% of intracranial meningiomas.^{10,26)} Cystic meningiomas are more frequent in childhood and adolescence, with an incidence of about 30%,^{9,16,24,29)} and even higher in infants at 50%.¹⁾ Meningiomas associated with large cysts are rare,^{3,25,30,31)} and the magnetic resonance (MR) imaging appearance of cystic meningiomas is not well described.^{7,22,25,30)}

We describe a 9-year-old boy with a meningioma containing a large cyst, and compared MR imaging findings with the histological study.

Case Report

A 9-year-old boy was admitted with a partial seizure in his right face. He had had a general convulsion at the age of 3, and since then had received anti-epileptic medication. He was the second child of healthy parents, a 37-year-old father and 30-year-old mother. His elder sister had undergone an operation for ovarian cyst.

On examination, multiple pigmentations, depigmentations, and subcutaneous tumors were found on and in his trunk and all four extremities. Histological examination of a biopsy specimen of one of the subcutaneous tumors showed plexiform schwannoma. Chromosome analysis using a total of 20 peripheral blood lymphocytes found a normal karyotype of 46XY chromosomes. Gene analysis was not performed.

Neurological examination found no abnormalities. There was no evidence of optic atrophy, hearing disturbance, or sensorimotor abnormalities. Computed tomography (CT) demonstrated a postcontrast enhanced tumor with a large perifocal cyst in the left frontal lobe (Fig. 1 *upper*), abnormal calcification in both lateral ventricles (Fig. 1 *lower*), and a postcontrast enhanced tumor in the right cavernous sinus. MR imaging showed the tumor with a large cyst containing septum-like structures (Fig. 2). The solid part of the tumor was slightly hypointense on T₁-weighted images and slightly hyperintense on T₂-weighted images. The tumor was heterogeneously enhanced with intravenous administration of gadolinium-diethylenetriaminepenta-acetic acid (Gd-DTPA). Although enhancement of the septum-like structures was unclear, a few tiny parts of the wall at-

Fig. 1 *upper*: Postcontrast CT scans showing a tumor with a perifocal large cyst in the left frontal lobe. Septum-like structures are faintly visible. *lower*: Precontrast CT scans showing abnormal calcification of both lateral ventricles.

tached to the brain surface were enhanced, suggesting that tumor cells were present in these regions. The cyst fluid was slightly hyperintense to cerebrospinal fluid on T₁-weighted images and hyperintense on T₂-weighted images. The tumor in the right cavernous sinus, which invaded the right pterygopalatine fossa, was also demonstrated by MR imaging. Left external carotid angiography disclosed a tumor stain fed by branches of the middle meningeal artery.

Since we considered that the tumor with the large cyst in the left frontal region was the cause of his epilepsy, a left frontal craniotomy was performed. The preoperative diagnosis was meningioma with a large, peripheral intratumoral cyst, because a few tiny parts of the wall were enhanced by Gd-DTPA on the MR images. The tumor involved the overlying bone and was tightly attached to the dura mater. The cyst contained a large amount of xanthochromic fluid (protein 3800 mg/dl). Thin and whitish membranes in the cyst joined the tumor to the surface of the brain. These membranes were considered to be the septum, as shown by MR imaging. Some parts of the septum and a part of the cyst wall overlying the surface of the brain were removed. The arachnoid

membrane was recognized behind most of the cyst wall. Histological examination showed meningotheelial meningioma with microcysts and tumor cell nests in the cyst wall and septum (Fig. 3).

The tumor in the right cavernous sinus was not removed. The postoperative course was uneventful, and he was discharged without neurological deficit.

Follow-up MR imaging revealed enlargement of the tumor in the cavernous sinus without neurological manifestation (Fig. 4). Approximately 11 months after the first operation, a right frontal craniotomy was performed and the intracranial extension of the tumor was totally removed. The histological diagnosis was meningotheiomatous meningioma with some malignant characteristics including high cellularity, necrosis, and pseudopalisading.

The residual intracavernous tumor invading the right pterygopalatine fossa was treated with the 201-source cobalt-60 gamma knife.

Discussion

Cystic meningiomas can be classified into four types according to the relationships between the tumor, associated cysts, and the surrounding brain: 1) centrally located intratumoral cyst, 2) peripherally located intratumoral cyst, 3) peritumoral cyst in the adjacent parenchyma, and 4) peritumoral cyst between the tumor and the adjacent parenchyma.²³⁾ Our case was considered to be type 2 from the histological findings. Type 2 may be macroscopically difficult to differentiate from type 4, but this differentiation is surgically important to prevent recurrences. Several authors have attributed tumor recurrences to incomplete removal of the wall of cystic meningiomas.^{11,13,17)} Therefore, careful histological examination of the cyst is necessary during surgery and the whole cyst wall should be removed to achieve a permanent cure.^{3,10-13,18,19)} In our patient, tumor cell nests were present in the cyst wall remote from the solid tumor, but radical removal of the cyst wall could not be achieved because of the functional prognosis and the presence of the tumor in the right cavernous sinus. However, every effort should be made to remove the cyst wall, especially the parts of the cyst wall enhanced on MR images.

MR imaging showed the solid part of the tumor in our patient as slightly hypointense on T₁-weighted images and slightly hyperintense on T₂-weighted images. This is the common finding for meningiomas. The cyst fluid was slightly hyperintense to cerebrospinal fluid on T₁-weighted images due to the high protein concentration of the cyst fluid. The MR

Fig. 2 MR images. A: The solid part of the tumor is slightly hypointense on T₁-weighted images (repetition time [TR] 440 msec, echo time [TE] 15 msec). B: The solid part of the tumor is markedly enhanced with Gd-DTPA on T₁-weighted images (TR 440 msec, TE 15 msec). C: The solid part of the tumor is slightly hyperintense on T₂-weighted images (TR 2000 msec, TE 100 msec), and the septum-like structures are visualized. D: Coronal T₁-weighted images with Gd-DTPA (TR 400 msec, TE 15 msec) showing abnormal enhanced lesions of the cyst wall (arrows), the septum-like structures (arrowheads), and a right cavernous sinus tumor encasing the internal carotid artery (thick arrow).

images also demonstrated tumor cell nests in the septum-like structures. The presence of tumor cell nests in the septum has not been reported before.

Several authors have discussed cyst formation in meningiomas. Ruelle *et al.*²⁸⁾ reviewed reported cases of cystic meningioma, and suggested different mechanisms of formation for intratumoral cysts and peritumoral cysts. Intratumoral cysts may be the outcome of microcystic degeneration, ischemic necrosis, intratumoral hemorrhage, or intratumoral metastasis, and peritumoral cysts originate from peritumoral edema, widening of the subarachnoid space, demyelination, or peritumoral hemorrhage.^{6,25,27)} In our patient, there was no communication between the subarachnoid space and the cystic cavity, and examination of the surgical specimen found no hemosiderin deposits.

Kolluri *et al.*¹⁵⁾ suggested that sarcomatous and angioblastic meningiomas are more frequently associated with cyst formation. Bowen *et al.*³⁾ and Maiuri *et al.*¹⁸⁾ found no significant correlation between cyst formation and histological type of meningioma. Several authors have pointed out the presence of vacuolated foam cells, microcysts, and reticulated

pattern in cystic meningiomas.^{3,13,14,19,25)} The tumor in our patient largely contained microcystic areas, so the microcysts probably resulted in the large cyst formation and a small amount of tumor persisted in the collagenous connective tissue forming the septum. Therefore, careful histological evaluation of the membranous structure, both cyst wall and cyst septum, is important.

The overall incidence of multiple meningiomas is 2.5%,⁸⁾ but multiple meningiomas are more common in children.²⁰⁾ The association of multiple meningiomas with neurofibromatosis is well recognized,^{8,21)} but the plexiform schwannoma removed from our patient is not consistent with neurofibromatosis. The abnormal calcification in both lateral ventricles was thought to arise from the choroid plexus, and may indicate intraventricular meningioma, which occurs very frequently in pediatric cases.⁴⁾ Arinami *et al.*²⁾ reported a patient with multiple meningiomas associated with a ring chromosome 22. Chromosome analysis was normal in our patient, although gene analysis was not performed. There was no evidence for the presence or absence of neurofibromatosis in this patient.

Fig. 3 A: Photomicrograph of the solid part of the tumor showing meningothelial meningioma largely composed of microcystic areas. B: Photomicrograph of the septum-like structure showing a tumor cell nest in the collagenous connective tissue. C: Photomicrograph of a part of the cyst wall attached to the brain surface showing meningothelial meningioma with microcysts. Hematoxylin and eosin stain, $\times 100$.

Fig. 4 Coronal T_1 -weighted MR image with Gd-DTPA (TR 400 msec, TE 15 msec) 11 months after the previous MR imaging (Fig. 2D) showing intracranial enlargement of the right cavernous sinus tumor.

Drape *et al.*⁷⁾ found that aggressive meningioma frequently appeared as a heterogeneous signal on both T_1 - and T_2 -weighted images. The meningioma in the cavernous sinus of our patient had some histological characteristics of malignancy, but it was difficult to estimate the histological type of the meningioma in the cavernous sinus and the large cystic meningioma in the left frontal lobe by pre-operative MR imaging. Children with meningeal tumors have a significantly shorter survival time because of the increased incidence of sarcomatous change, the multiple sites of origin, and the larger size of the tumor.⁹⁾ Such patients require treatment to ensure a high quality of daily life and very close follow-up to detect any tumor progression as early as possible.

MR imaging can demonstrate the presence of tiny tumor cell nests in the thin cyst wall and the cyst septum of meningiomas, and can help to differentiate intratumoral from peritumoral cysts.

References

- 1) Amano K, Miura N, Tajika Y, Matsumori K, Kubo

- O, Kobayashi N, Kitamura K: Cystic meningioma in a 10-month-old infant. Case report. *J Neurosurg* 52: 829-833, 1980
- 2) Arinami T, Kondo I, Hamaguchi H, Nakajima S: Multifocal meningiomas in a patient with a constitutional ring chromosome 22. *J Med Genet* 23: 178-180, 1986
 - 3) Bowen JH, Burger PC, Odom GL, Dubois PJ, Blue JM: Meningiomas associated with large cysts with neoplastic cells in the cyst walls. *J Neurosurg* 55: 473-478, 1981
 - 4) Byard RW, Bourne AJ, Clark B, Hanieh A: Clinicopathological and radiological features of two cases of intraventricular meningioma in childhood. *Pediatr Neurosci* 15: 260-264, 1989
 - 5) Crouse SK, Berg BO: Intracranial meningiomas in childhood and adolescence. *Neurology* 22: 135-141, 1972
 - 6) Dell S, Ganti SR, Steinberger A, McMurtry J III: Cystic meningiomas: A clinicoradiological study. *J Neurosurg* 57: 8-13, 1982
 - 7) Drape JL, Krause D, Tongio J: MRI of aggressive meningiomas. *J Neuroradiol* 19: 49-62, 1992
 - 8) Eljamel MSM, Foy PM: Multiple meningiomas and their relation to neurofibromatosis. *Surg Neurol* 32: 131-136, 1989
 - 9) Ferrante L, Acqui M, Artico M, Mastronardi L, Rocchi G, Fortuna A: Cerebral meningiomas in children. *Childs Nerv Syst* 5: 83-86, 1989
 - 10) Fortuna A, Ferrante L, Acqui M, Guglielmi G, Mastronardi L: Cystic meningiomas. *Acta Neurochir (Wien)* 90: 23-30, 1988
 - 11) Hadani MA, Rappaport ZH, Findler G, Tadmor R, Sahar A: Recurrent cystic meningioma. *Neurochirurgia (Stuttg)* 28: 202-204, 1985
 - 12) Imagawa K, Nomura T, Asai A, Hayashi M, Toda I, Kawasaki M, Yokoi K: Cystic meningioma. Report of two cases. *No Shinkei Geka* 11: 513-518, 1983 (in Japanese)
 - 13) Inoue T, Kuromatsu C, Sawada K, Matsushima T: Recurrent cystic meningioma. *Surg Neurol* 26: 399-404, 1986
 - 14) Kleinman GM, Liszczak T, Tarlov E, Richardson EP Jr: Macrocystic variant of meningioma. *Am J Surg Pathol* 4: 383-389, 1980
 - 15) Kolluri VRS, Reddy DR, Reddy PK, Naidu MRC, Devi S: CT-findings in cystic meningiomas. *Acta Neurochir (Wien)* 87: 31-33, 1987
 - 16) Kolluri VRS, Reddy DR, Reddy PK, Naidu MRC, Rao SBP, Sumathi C: Meningiomas in childhood. *Childs Nerv Syst* 3: 271-273, 1987
 - 17) Lake P, Heiden JS, Minckler J: Cystic meningioma. Case report. *J Neurosurg* 38: 638-641, 1973
 - 18) Maiuri F, Benvenuti D, De Simone MR, Cirillo S, Corriero G, Giamundo A: Cystic lesions associated with meningiomas. *Surg Neurol* 26: 591-597, 1986
 - 19) Matsushima T, Kinoshita K, Numaguchi Y, Oda K: Cystic meningioma. A case report. *No Shinkei Geka* 6: 167-171, 1978 (in Japanese)
 - 20) Merten DF, Gooding CA, Newton TH: The radiographic features of meningiomas in childhood and adolescence. *Pediatr Radiol* 2: 89-96, 1974
 - 21) Merten DF, Gooding CA, Newton TH, Malamud N: Meningiomas of childhood and adolescence. *J Pediatr* 84: 696-700, 1974
 - 22) Namba H, Sueyoshi K: Magnetic resonance imaging of a cystic meningioma. *Neuroradiology* 32: 536, 1990
 - 23) Nauta HJW, Tucker WS, Horsey WJ, Bilbao JM, Gonsalves C: Xanthochromic cysts associated with meningioma. *J Neurol Neurosurg Psychiatry* 42: 529-535, 1979
 - 24) Numaguchi Y, Hoffman JC, O'Brien MS, Fukui M, Matsuura K, Kitamura K: Meningiomas in childhood and adolescence. *Neurol Med Chir (Tokyo)* 18 [Part I]: 119-127, 1978
 - 25) Odake G: Cystic meningioma: Report of three patients. *Neurosurgery* 30: 935-940, 1992
 - 26) Parisi G, Tropea R, Giuffrida S, Lombardo M, Giuffrè F: Cystic meningiomas. Report of seven cases. *J Neurosurg* 64: 35-38, 1986
 - 27) Pinna G, Beltramelli A, Buffatti P, Signorini G, Colombari R, Bricolo A, Ore GD: Cystic meningiomas. An update. *Surg Neurol* 26: 441-452, 1986
 - 28) Ruelle A, Mariotti E, Boccardo M: "True" cystic meningioma. *J Neurol Neurosurg Psychiatry* 48: 716-718, 1985 (Letter)
 - 29) Suematsu K, Tokuda S, Miyazaki Y: Intracranial meningioma in a five-month-old infant. A case report and review of the literature. *No To Shinkei* 26: 453-464, 1974 (in Japanese)
 - 30) Tomita H, Kuwamura K, Sugimoto M, Saito M: Cystic meningioma: Report of a case and review of literatures. *Neurosurg Lett* 3: 209-214, 1993
 - 31) Umansky F, Pappo I, Pizov G, Shalit M: Cystic changes in intracranial meningiomas. A review. *Acta Neurochir (Wien)* 95: 13-18, 1988

Address reprint requests to: A. Watanabe, M.D., Department of Neurosurgery, Kawasaki Medical School, 577 Matsushima, Kurashiki, Okayama 701-01, Japan.