Cystic Lymphoplasmacyte-Rich Meningioma

—Case Report—

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Abstract

A 47-year-old female presented with an unusual cystic meningioma. Computed tomography and magnetic resonance imaging showed a cystic tumor with marked edema in the left parietal region of the brain, resembling a metastatic tumor. The tumor was totally removed. Histological examination revealed meningioma with dense lymphoplasmacytic infiltrates.

Key words: cystic meningioma, lymphocyte, magnetic resonance imaging, plasma cell

Introduction

Meningothelial, transitional, and fibrous meningiomas with dense lymphoplasmacytic infiltrates are listed in the new World Health Organization classification of tumors of the central nervous system.⁷⁾ Lymphoplasmacyte-rich meningioma is an extremely rare variant of this tumor. Cystic meningiomas are also uncommon with an incidence of 1.7% in a large series of 1313 intracranial meningiomas.^{4,11,14,15)} Meningioma occurring as a tumor with the features of a plasmacytoma and meningioma is very unusual.^{1,3,5,8,9,16,17)} We describe an unusual case of meningioma with both lymphoplasmacytic and cystic characteristics.

Case Material

A 47-year-old female was referred to our service with a 6-month history of dizziness, dyscalculia, and dysgraphia. She had no complaints of headache, nausea, or vomiting. She was alert and well oriented. On admission, no further neurological signs were found. Magnetic resonance (MR) imaging showed a cystic tumor with marked brain edema in the left parietal region (Fig. 1). After the administration of gadolinium-diethylenetriaminepenta-acetic acid, the tumor showed ring enhancement and a mural nodule (Fig. 1). Computed tomography (CT)

also showed ring enhancement (Fig. 2). Cerebral angiography showed a slight tumor stain in the venous phase. Therefore, we suspected that this tumor was a metastasis.

Whole-body CT, technetium-99m-hydroxymethylene diphosphonate bone scintigraphy, gallium-67 scintigraphy, and blood chemistry tests were performed. The blood chemistry tests included tumor markers, such as alpha-fetoprotein, carcinoembryonic antigen, CA19-9, CA125, squamous-cell carcinoma antigen, and neurospecific enolase. These examinations identified no primary malignancy, but showed increased serum C-reactive proteins (6.6 mg/ml) and white blood cell count (9900/ μ l).

A left parieto-occipital craniotomy performed under general anesthesia revealed the well-encapsulated tumor which arose from the dura mater. The tumor had not invaded the superior sagittal sinus or the cortical veins. Xanthochromic fluid was observed and a mural nodule was resected. The mass including the cavity wall was totally removed (Simpson's grade II).

Histological examination of the tumor specimen showed that the cavity wall consisted of fibrous connective tissue containing nests of tumor cells. The tumor cells were meningotheliomatous meningioma, with eosinophilic cytoplasm and ill-defined cell borders. The connective tissue was infiltrated by numerous plasma cells and lymphocytes, with a few scattered lymph follicles. Some of the tumor cell nests were also infiltrated by lymphoplasmacytic cells (Fig. 3). The small mural nodule was attached to

Fig. 1 Preoperative sagittal (A-C) and coronal (D-F) magnetic resonance (MR) images showing a cystic tumor in the parietal region. The T_2 -weighted MR images (B, E) show the signal intensity in the cyst is similar to that of cerebrospinal fluid, and the large area of edema surrounding the cyst. The T_1 -weighted MR images before (A, D) and after (C, F) gadolinium-diethylenetriaminepenta-acetic acid administration show ring enhancement and a mural nodule (arrow).

Fig. 2 Computed tomography scans with contrast medium showing ring enhancement.

Fig. 3 Photomicrograph of the tumor specimen showing meningotheiomatous cells in the tumor infiltrated by numerous lymphoplasmacytic cells. HE stain, $\times 350$.

Fig. 4 Immunohistochemical staining for the λ light chain showing approximately half of the infiltrating plasma cells are positively stained, and about an equal number of plasma cells are positive for the κ chain, implying that the lymphoplasmacytic cells are not neoplastic in nature. $\times 270$.

Fig. 5 Postoperative T₂-weighted sagittal magnetic resonance image 1 month after surgery showing the area of edema in the parietal lobe is smaller.

the dural side of the cavity wall. This nodule was composed of tumor cells and connective tissue, both infiltrated by lymphoplasmacytic cells. The dura was infiltrated by small nests of meningioma cells.

Immunohistochemical staining showed the tumor cells were positive for vimentin (DAKO Japan Co., Ltd., Kyoto) and negative for cytokeratin (AE1/AE3; Boehringer Mannheim, Mannheim, Germany). Immunohistochemical analysis using antibodies for B

(CD20; DAKO) and T lymphocytes (CD3 and CD45RO; DAKO) showed that B cells were distributed near lymph follicles and T cells were scattered diffusely in interfollicular areas. Plasma cells, the main component of the infiltrating lymphoplasmacytic cells, were positive for κ or λ chains in about equal numbers, implying that they were reactive cells and not neoplastic (Fig. 4).

One month after the operation, the patient's clinical symptoms, such as dizziness, dyscalculia, and dysgraphia, were improved and MR imaging showed a decrease in brain edema (Fig. 5). Hematological examination was done to assess the possible association of hypergammaglobulinemia,⁵⁾ but the serum levels of immunoglobulin G (1350 mg/dl) and immunoglobulin A (233 mg/dl) were within normal limits.

Discussion

Various lesions, such as gliomas, metastatic tumors, abscesses, inflammation, infarction, and hematomas, show ring enhancement on CT or MR imaging after contrast material is injected.^{2,8,10,12,13)} In our patient, the tumor showed ring enhancement with marked surrounding edema, but angiography did not demonstrate any remarkable stain. Therefore, we considered the tumor to be metastatic preoperatively. The postoperative results revealed the tumor to be a cystic lymphoplasmacyte-rich meningioma. Therefore, ring enhancement is not a specific finding.

Cystic meningioma is difficult to distinguish from other tumors, such as astrocytomas, hemangioblastomas, neuroblastomas, or metastatic tumors. The chance of diagnosing a cystic meningioma based on CT findings is less than 38%, and the chance of diagnosis based on angiography is less than 18%.⁴⁾ Differentiating this tumor from a partially enhanced glioma or metastasis was difficult in three previous patients even with MR imaging because of the nonenhanced cysts and focal edema.¹⁴⁾

The definition of lymphoplasmacyte-rich meningioma was established by the World Health Organization in their new histological classification.⁷⁾ Various hypotheses to account for lymphoplasmacytic infiltrates have been presented. These lesions may develop from the collision of a plasmacytoma and a meningioma, or a plasmacytoma of the meninges with a leptomeningeal reaction.¹⁾ The inflammatory cell reaction to the meningioma may be a mechanism of host resistance to the tumor.⁶⁾ However, the detailed relationship of inflammatory lesions and meningiomas is still uncertain.

Our patient did not have hypergammaglobuline-

mia and the preoperative general examinations revealed no primary malignancy. Immunohistochemical examination suggested that the lymphoplasmacytic infiltrate was not neoplastic but an inflammatory reaction to the cystic meningioma. We suggest that this biological reaction caused the marked brain edema. The tumor had not disturbed the venous circulation and the brain edema disappeared after resection of the tumor. In addition, 1 month after the operation, the serum C-reactive proteins (<0.5 mg/ml) and the white blood cell count ($4200/\mu\text{l}$) had returned to within normal limits.

Preoperative diagnosis is difficult for both cystic meningiomas and lymphoplasmacyte-rich meningiomas. Therefore, a tumor combining these characteristics will confuse the correct diagnosis. Nonspecific radiological findings, such as ring enhancement, marked edema on CT or MR imaging, and the absence of remarkable tumor stain on angiography can easily mislead the differential diagnosis. Therefore, careful general physical and radiological examinations, blood chemistry tests, and neuropathological examinations, including immunohistochemistry, are necessary for the correct diagnosis.

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