Skull Meningioma Associated with Intradural Cyst: A **Case Report**

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ABSTRACT: We present the first report of intraosseous meningioma accompanied by intradural cyst formation. A 76-year-old woman had previously undergone breast cancer treatment, so the preoperative diagnosis was metastatic breast cancer. This case reminds us that the possibility of meningioma should be kept in mind in patients with breast cancer, irrespective of neuroimaging findings.

KEYWORDS: Intraosseous meningioma, intradural cyst formation, skull tumor, breast cancer

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Case Report

A 76-year-old woman who had undergone breast cancer treatment 9 years previously presented with dizziness. Neurological findings were normal. Skull x-rays and computed tomography (CT) showed an osteolytic lesion measuring approximately 3 cm×4 cm in the left frontal bone (Figure 1). Magnetic resonance images demonstrated a mass lesion within the diploe that was isointense on T1-weighted and T2-weighted images and hyperintense on diffusion-weighted and fluid attenuated inversion recovery images (Figure 2). An intradural cyst with similar intensity on T1-weighted, T2-weighted, and diffusion-weighted images, slightly hyperintense compared with cerebrospinal fluid (CSF), was seen adjacent to the mass lesion. T1-weighted images following gadolinium injection demonstrated prominent, homogeneous enhancement of the skull tumor, and continuous enhancement of the dura mater and a small intradural solid mass, whereas the cyst wall remained unenhanced. Systemic CT and bone scintigraphy revealed no other lesions. Tumor markers were all negative. The neuroradiological findings supported a preoperative diagnosis of skull metastasis from breast cancer invading the intradural space, despite the lack of evidence of recurrence or systemic metastasis of breast cancer on physical, hematologic, or radiologic examinations.

Intraoperatively, the skull surface was found to be intact, but the tumor was visible crossing the coronal suture. Craniotomy was performed, allowing a safe margin around the lesion. The dura mater and attached tumor were also resected while ensuring safe margins and, together with the underlying cyst wall, separated easily from the brain surface. After total removal of the lesion, duroplasty was performed using a Gore-Tex membrane (Japan Gore-Tex Institution, Tokyo, Japan), and cranioplasty was performed with a titanium mesh plate.

Histologic evaluation revealed benign transitional meningioma with whorl formation in all excised specimens including the skull, dura mater, and cyst wall (Figure 3). There were

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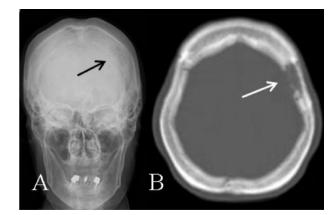


Figure 1. (A) Plain skull x-ray shows an osteolytic lesion in the left frontal bone (arrow). (B) Bone window computed tomography shows a solitary osteolytic lesion.

increasing collagenous fiverb in the skull sections. The postoperative course was uneventful.

Discussion

Intraosseous meningioma is classified as primary or secondary. Primary intraosseous meningioma is defined as a lesion that does not involve the inner layer of the underlying dura mater. However, this type of intraosseous meningioma may involve the dura mater and extend into the intradural space later in the course of tumor growth.¹ In the present case, the tumor was predominantly located in the skull, giving rise to an inwardly bulging dura mater and a minimal intradural solid component. We thus consider that this case probably represented primary intraosseous meningioma, although the possibility of a secondary lesion cannot be excluded. Nonetheless, meningioma predominantly located in the skull and associated with a true intradural cyst is very rare: to the best of our knowledge, this represents the first reported case.

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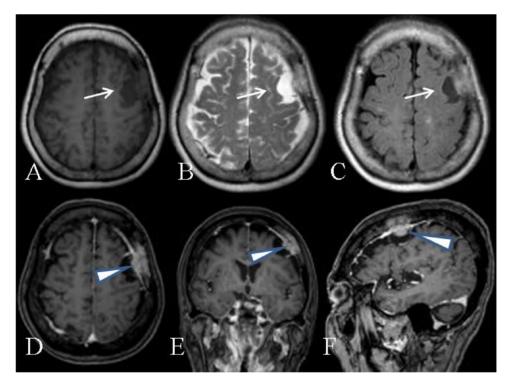


Figure 2. (A) T1-weighted and (B) T2-weighted axial MR images show an isointense intraosseous mass that is hyperintense on (C) FLAIR image. The mass is associated with an intradural cyst showing similar intensity to CSF on T1-weighted and T2-weighted images and slightly hyperintense compared with CSF on FLAIR image. Gadolinium-enhanced (D) axial, (E) coronal, (F) and sagittal MR images demonstrate homogeneous enhancement of the mass and dura mater but no enhancement of the cyst wall. Note that the dura mater is seen along the inner margin of the mass, bulges inward as a hypointense band on the (B) T2-weighted image, and appears as a more enhanced band on gadolinium-enhanced (D, E) T1-weighted images. Arrows indicate tumor with cyst, and arrowheads indicate dura mater between the mass and cyst. CSF indicates cerebrospinal fluid; FLAIR, fluid attenuated inversion recovery; MR, magnetic resonance.

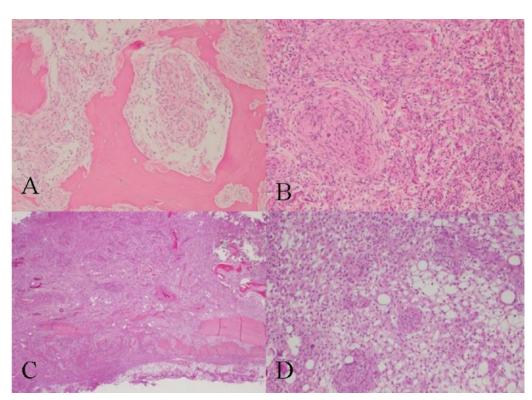


Figure 3. Hematoxylin-eosin–stained sections of intraosseous tumor—(A) ×10 and (B) ×40; dura mater—(C) ×10 and cyst wall—(D) ×40. (A, B) Transitional meningioma is shown with whorl formation and an increased proportion of collagenous fibers in the skull. Tumor cells have infiltrated through (C) the dura mater and are present in (D) the cyst wall.

Cystic meningiomas account for 2% to 4% of all intracranial meningiomas.^{2,3} Nauta et al³ classified cystic meningioma into 4 groups. In type 1, the cyst is surrounded by macroscopic tumor throughout; in type 2, the cyst is on the periphery of the tumor, but still wholly within the margins of microscopically visible tumor; in type 3, the cyst lies within the adjacent brain rather than within the tumor itself; and in type 4, the cyst appears at the interface between the tumor and brain as CSF sequestered within the subarachnoid space. Several mechanisms of cyst formation have been postulated.4,5 According to Fortuna et al,⁵ intratumoral cysts may be caused by microcystic degeneration, ischemic necrosis, or hemorrhage. Microcystic degeneration is considered to represent the confluence of cellular degenerative phenomena such as vacuolar, myxomatous, mucoid, and fatty degeneration, leading to the formation first of microcavitations and then of macrocavitations within the tumor.^{4,5} In the present case, which was categorized as Nauta type 2, the inwardly bulging dura mater infiltrated with tumor cells might have trapped CSF within the subarachnoid space and could then have enlarged by a check valve mechanism. Tumor cells might have extended through the arachnoid membrane and along the brain surface, forming the true cyst, because the cyst contained protein-rich fluid resembling CSF on magnetic resonance (MR) images.

The presence of cyst wall enhancement after intravenous injection of gadolinium may facilitate diagnosis of cyst type and thus determination of whether the cyst wall warrants removal.^{2,6} Zee et al⁷ described enhancement of the cyst wall in 5 of 5 type 2 cases but no enhancement in 4 of 4 type 3 cysts and concluded that this parameter is very important for differentiating between these 2 types of cysts. In contrast, Ferrante et al² described no MR enhancement of the cyst wall in 7 surgically verified cases of Nauta type 2 and 3 cysts. Differentiating between type 2 and 3 cysts preoperatively is thus often difficult. In the present case, the cyst wall was not enhanced on MR images, but histologic findings indicated Nauta type 2 cyst. Regardless of a lack of enhancement, and even if the cyst had been difficult to separate from the brain surface, removal of the cyst wall as completely as possible would have been necessary to minimize the risk of tumor recurrence.

Both cystic and intraosseous meningiomas are diagnostically confused with other tumors. Gastavo et al⁶ noted that cystic meningioma sometimes resembles glioma or metastatic tumor. Moreover, intraosseous meningioma has many differential diagnoses, including metastasis, plasmacytoma, giant cell tumor, hemangioma, epidermoid cyst, osteogenic sarcoma, and eosinophilic granuloma. However, the most plausible diagnosis is metastasis when a patient with a history of cancer presents with skull or dural lesions. Dural and skull metastases from breast cancer have often been described, even when the primary tumor was in long-term remission.⁸⁻¹⁰ Breast cancer is one of the most frequent malignancies associated with intracranial dural metastasis, which is often a single lesion.8 Direct extension from osteolytic skull metastasis is the most common mode of spread.^{8,10} However, the unique association between carcinoma of the breast and meningioma is also well known.9 As neuroimaging findings in the present case with a past history of breast cancer were atypical for meningioma and compatible with metastasis, the presumptive diagnosis of metastatic breast cancer seemed reasonable. However, we should also have suspected meningioma.

In conclusion, this is the first case report of meningioma predominantly located in the skull and associated with true intradural cyst. We should not exclude the possibility of meningioma without histologic confirmation because meningioma is sometimes difficult to distinguish from metastasis in patients with breast cancer.

Author Contributions

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