Clinical Image

Metastatic Cholangiocarcinoma Presenting As Punched-out Lesions of the Skull

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A 60-year-old female with diabetes presented to the department of internal medicine with a 1-month history of headache. The patient's Eastern Cooperative Oncology Group performance status was 1. A moderate tenderness was elicited by soft palpation of the skull. Laboratory investigations showed elevated alkaline phosphatase level of 604 (normal range 100-350) U/L. The level of hemoglobin, creatinine and calcium were within normal range. Plain radiography and computed tomography (CT) demonstrated many punched-out osteolytic lesions in the skull (Figure 1). Cranial magnetic resonance imaging depicted multiple skull lesions, with T1 hypointense signal, T2 slightly hyperintense signal, and restricted diffusion (Figure 2). We suspected multiple myeloma, but monoclonal protein was not detected in serum. Contrast-enhanced chest-abdomen-pelvis CT revealed multiple hepatic masses with lymph node metastases and distant metastases including vertebral bodies (Figure 23). Other malignant tumor workup was negative. Ultrasound-guided biopsy of the hepatic tumor indicated cholangiocarcinoma. The patient underwent chemotherapy with gemcitabine, in combination with radiotherapy, but died one month after admission. Punched-out lesions are osteolytic lesions without sclerotic rim on X-ray examination, which are considered as

hallmarks of multiple myeloma. Punched-out lesions, however, are created by the absence of reactive bone formation because of tumor factors that combine to activate osteoclasts and inhibit osteoblasts (1), so any malignancy can cause punched-out lesions. Majority of lytic bone metastases originate from breast, lung, kidney, colon, melanoma and thyroid. Punched-out lesions in the skull arising from cholangiocarcinoma has not been previously reported.

Cholangiocarcinoma is a heterogeneous group of tumors derived from bile ducts cells, which represents the second most frequent malignant liver cancer. Incidence is globally increasing with 2.1/100,000 person-years in Western countries. Patients with cholangiocarcinoma commonly have locally advanced diseases (2). It usually metastasizes to the regional lymph nodes through lymphatic vessels, followed by hematogenous metastasis to the liver, peritoneum and lungs. Distant metastasis is uncommon. But, according to a few case reports, lytic bone metastasis can be a first clinical presentation of cholangiocarcinoma (3).

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FIG. 1. (A) Lateral skull radiograph showing punched-out lesions. (B) Computed tomography of the head showing multiple osteolytic lesions.

FIG. 2. Cranial magnetic resonance imaging depicted multiple skull lesions, with T1 hypointense signal (A), T2 slightly hyperintense signal (B), and diffusion-weighted imaging hyperintense signal (C).

FIG. 3. (A) Axial contrast-enhanced computed tomography of the abdomen showing multiple liver masses. (B) Sagittal contrast-enhanced computed tomography of the abdomen showing vertebral bodies metastases.