Current problem cases

Osteosarcoma with multiple skeletal metastases

A case of “nonstochastic” metastasis

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Summary. Osteosarcoma of the thoracic spine developed in a 15-year-old Japanese boy. After his first admission with paralysis, multiple skeletal metastases were demonstrated in the absence of pulmonary metastasis. This rare condition may possibly be considered as a unicentric osteosarcoma with bone metastases, since there were no precursor lesions or history of exposure to radioactive materials or chemical agents. These multiple lesions may be an example of so-called “organ-specific metastasis,” although this “nonstochastic” process is rare.

Multiple osteosarcomas have been interpreted as a distinct clinical entity [18, 21]. However, it is not always easy to determine whether multiple skeletal tumors represent multicentric primary tumors or metastatic foci [3]. Several features, such as the absence of previous systemic bone pathology, absence of a history of exposure to radiation, simultaneous appearance of lesions in affected bones, and absence of pulmonary metastases, are considered to establish a multicentric origin of an osteosarcoma [4]. Nevertheless, multicentric origin is still not sufficiently established in this way, since synchronous occurrence is not likely to be accepted and the biology of bone metastasis is poorly understood [5, 7]. In this paper, we report the case of a patient with thoracic spinal osteosarcoma [11, 16, 20, 26] who has atypical multiple skeletal metastases [25].

Case report

On November 2, 1990, a 15-year-old boy with complete paraplegia was admitted to Gunma University Hospital. He had complained of severe back pain for a month. He denied any family history of Paget’s disease or bone disorders, or a history of previous radiation therapy. Physical examination demonstrated complete motor deficits below the level of the seventh thoracic vertebra. He had noticed disturbances in bowel and bladder functions. A mild sensory disturbance was evident below the level of the epigastric region and complete deficit below the level of the inguinal region.

Laboratory investigations revealed extreme increases alkaline phosphatase level (8172 IU/L; norm 75–220 IU/L) and the lactate dehydrogenase level (1175 IU/L; norm 230–420 IU/L). The blood sedimentation rate was 38 mm/h. Other laboratory findings were uniformly within normal limits.

Roentgenograms of the thoracic spine showed indistinctness of the spinous process of the seventh thoracic vertebra (Fig. 1), although plain roentgenograms of the chest showed no abnormalities. A myelogram and axial plain computed tomographic (CT) scan demonstrated that a mass was producing extradural spinal cord compression at the level of the seventh thoracic vertebra (Fig. 2). Cerebrospinal fluid analysis showed an increased protein level. Sagittal magnetic resonance imaging (MRI) also showed the extradural posterior compressive mass at the same level (Fig. 3). The sagittal plain CT scan demonstrated a posterior bony mass extending from the lamina of the seventh thoracic vertebra (Fig. 4). Technetium scintigraphy showed increased radioisotope uptake at the following sites: upper cervical spine, mid-thoracic spine, lumbar spine, left sacroiliac joint, left femoral neck, and right femoral shaft (Fig. 5). Osteoblastic lesions appeared at these sites in skeletal roentgenograms (Fig. 6). On November 7, open biopsy was performed. The histological diagnosis was osteoblastic osteosarcoma. After surgery, the patient underwent chemotherapy and palliative radiotherapy. CT tomography and plain radiograms showed no abnormalities in the pulmonary fields up to June 10, 1991.

Discussion

It is logical to suppose that the initial lesion of multiple osteosarcomas will usually be larger than subsequently noted lesions [3]. Furthermore, there has always been an apparent interval, however short, between the appearance of the first two symptomatic lesions in all reported patients with multiple osteosarcomas [3]. In general, osteosarcomas have been considered to be of multiple origin if pulmonary metastasis is not demonstrated [4], since osteosarcomas most often metastasize to the lung and less often to other organs such as the skeleton [30]. Based on these concepts, synchronous multiple osteogenic sarcomas are occasionally reported in the literature [4, 10, 14, 18, 21, 27, 29]. On the other hand, micrometastases may be present at the time of the initial diagnosis of osteosarcoma [17, 23, 28]. However, whether multiple bone lesions are multifocal in origin or result from a single primary tumor is not clear, especially without a
Fig. 1a, b. Roentgenograms of the thoracic spine reveal indistinctness of the seventh spinous process. No other abnormal findings are apparent. a Anteroposterior view, b lateral view

Fig. 2. a Myelogram and b axial plain computed tomogram show a mass producing extradural spinal cord compression at the level of the seventh thoracic vertebra

Fig. 3. Sagittal magnetic resonance image shows an extradural posterior compressive mass at the level of the seventh thoracic vertebra. (TR 300 ms, TE 14 ms)

Fig. 4. Sagittal plain computed tomogram shows a posterior bony mass extending from the lamina of the seventh thoracic vertebra

Fig. 5. A total body bone scan shows increased uptake at the following sites: upper cervical spine, midthoracic spine, lumbar spine, left sacroiliac joint, left femoral neck, and right femoral shaft. Artificial uptake at the right leg shows urine collection