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Widespread Inflammatory Response to Osteoblastoma: The Flare Phenomenon¹

A case of vertebral osteoblastoma caused a diffuse, reactive inflammatory infiltrate in two vertebrae, adjacent ribs, and the paraspinous soft tissues. The authors call this the flare phenomenon. On magnetic resonance images the diffuse inflammatory response caused a misleading appearance that simulated a malignant process (lymphoma or Ewing sarcoma). A computed tomographic myelogram was diagnostic.

Index terms: Osteoblastoma, 322.3123 • Spine, CT, 322.1211 • Spine, primary neoplasms, 322.3123 • Spine, MR studies, 322.1214

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THE main purpose of this report is to emphasize the nonspecificity of magnetic resonance (MR) imaging in the workup of bone lesions and dangers in relation to biopsy procedures in the absence of adequate correlation with standard radiographs and especially with computed tomographic (CT) scans.

CASE REPORT

A 19-year-old man complained of pain in the upper back that had been present for 2 years. The pain had gradually increased in severity until it was no longer relieved by narcotics, and often it awakened him from sleep. Radiographs were interpreted as negative, although in retrospect the right pedicle of the third thoracic vertebra was obscured. A bone scan obtained after administration of methyl-

ene diphosphonate showed diffusely increased activity of the upper thoracic vertebrae; this activity was most marked at T-3. MR imaging showed regions of low signal intensity in the second and third thoracic vertebral bodies and posterior elements and the adjacent ribs on T1weighted images (this low signal intensity became high signal intensity on T2weighted images), and a right-sided paraspinous mass in which signal intensity went from intermediate to high. Examination of biopsy samples of the soft-tissue mass, obtained with both needle aspiration and open right thoracotomy, showed only nonspecific inflammatory tissue. A Horner syndrome was noted after thoracotomy.

The patient was referred to our institution. Repeat MR imaging again showed a paraspinous soft-tissue mass and abnormal signal in both the T-2 and T-3 vertebral bodies (Fig 1), their posterior elements, and the adjacent portions of the second and third ribs bilaterally. Additionally, compression of the thecal sac by an expanded right pedicle at T-3 was seen. All of these regions showed signal enhancement after administration of gadopentetate dimeglumine.

A CT myelogram was performed to better assess the cord compression. At CT, the trabeculae of the vertebral bodies appeared entirely normal, no evidence of an infiltrative process was seen, and the bony cortices were intact. However, the lesion in the right pedicle of T-3 could be better seen (Fig 2) than at MR imaging. The pedicle lesion was expansile, without cortical breakthrough, and blastic regions were visible within it. The thecal sac was compressed, but there was no evidence of intrathecal tumor. A paraspinous soft-tissue mass was again seen and was homogeneous in appearance. A complete blood count and results of routine biochemical tests were normal; the erythrocyte sedimentation rate was not obtained.

It was believed that the T-3 pedicle lesion was causing the patient's symptoms, and therefore the pedicle was excised through a direct posterior approach. At surgery, biopsy samples of the T-2 and T-3 vertebral bodies and spinous processes and bacterial, tuberculosis, and fungal cultures were also obtained in order to assess the infiltrative process suggested by MR imaging. The patient reported complete relief of his preoperative symptoms by 1 week after surgery. One month after surgery he returned to work as a laborer. Repeat MR imaging 3 months after sur-



Figure 1. Sagittal spin-echo (480/30 [repetition time msec/echo time msec]) image. Low signal intensity is seen throughout the T-3 vertebral body, T-2 and T-3 spinous processes, and the inferior portion of the T-2 vertebral body. Disk space is not involved. Cortices appear intact.

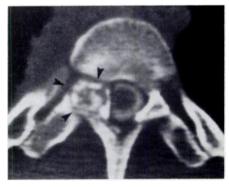


Figure 2. CT myelogram. There is marked "expansion" of the right pedicle of T-3 (arrowheads) by a mass that contains both lytic and blastic regions. The lesion is surrounded by a rim of mature periosteal new bone and contains foci of amorphous, dense bone. Soft-tissue mass is seen in the right paraspinous region. The thecal sac is compressed but not invaded.

gery showed that the marrow signal in T-2 and T-3 had returned to normal.

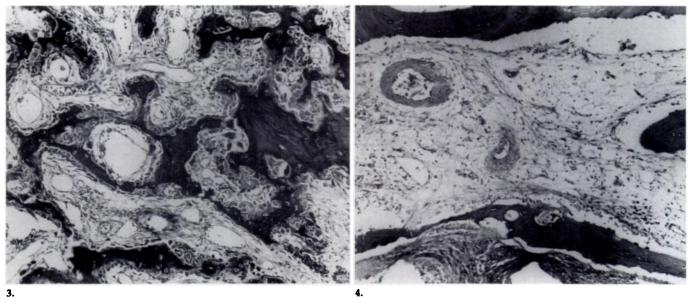
DISCUSSION

This patient had an abnormality of the spine that appeared very different on MR images than on CT scans: MR images showed a diffuse process involving several bones, and CT scans showed a highly localized tumor. On

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Figures 3, 4. (3) Lesion from T-3 pedicle showed irregular trabeculae of woven bone with osteoblastic rimming, osteoclastic hyperplasia, and telangiectasia, features that typify osteoblastoma. (Hematoxylin-eosin stain; original magnification, ×125.) (4) Biopsy sample of T-2 vertebral body showed osteoporosis and replacement of marrow fat by edema, loose fibrous tissue, hypervascularity, and an infiltrate of round cells, which at high-power examination were seen to consist of an equal mixture of mature lymphocytes and plasma cells. (Hematoxylin-eosin stain; original magnification, ×125.)

the basis of the MR images alone, lymphoma or Ewing sarcoma was the most likely diagnosis to explain the involvement of several bones and the soft-tissue mass. Infection was unlikely, given the preservation of cortical bone and intervertebral disk spaces and the normal white blood cell count.

In contrast, the appearance at CT was most consistent with osteoblastoma. Low-grade osteosarcoma could have a similar appearance but is usually centered in the vertebral body rather than in the posterior elements. Clinically, the long history of severe and increasing pain in the upper back in a young man should raise the suspicion of osteoid osteoma or osteoblastoma.

Review of the microscopic anatomy (Fig 3) showed a tumor forming thick, irregular bone trabeculae rimmed by osteoblasts. The stromal cells showed no anaplasia, and mitotic figures were absent. Because osteosarcomas, which are heavily ossified, may show "normalization" of nuclei (1), the differential diagnosis was osteosarcoma versus osteoblastoma. The lesion had a loose stroma, and prominent osteoclasts and capillaries characteristic of osteoblastoma. The argument against osteosar-

coma was based on the absence of cartilage production or trapped spicules of lamellar bone indicative of a malignant infiltrative process.

What caused the diffuse marrow abnormality at MR? Biopsy samples taken from two of these areas (Fig 4) showed replacement of the normal marrow fat by considerable edema fluid, fibroplasia, and chronic inflammatory cells, predominantly plasma cells. The softtissue mass showed a similar inflammatory infiltrate. Cultures obtained at surgery were negative for aerobic and anaerobic bacteria, tuberculosis, and fungi. The patient's pain and the diffuse MR marrow signal abnormalities resolved after removal of the right T-3 pedicle.

The severe inflammatory response involved several adjacent bones and adjacent soft tissue. Although a soft-tissue mass in osteoblastoma has been described (2), the soft-tissue mass seen in our patient was merely part of an inflammatory reaction. Diffuse gadolinium enhancement reflected the inflammatory nature of the process and caused marked overestimation of the size of the lesion. Because of the diffuse abnormalities, the actual tumor site was not appreciated on MR images, and this led to inappropriate needle biopsy and

pen thoracotomy. These procedures not only failed to achieve a positive diagnosis but induced a Horner syndrome because of injury to the sympathetic chain.

This case, in which MR imaging suggested an entirely different diagnosis than did CT, indicates the need for caution in basing diagnosis and choice of biopsy site on MR imaging findings alone. CT is the modality of choice in diagnosis of bone tumors that are poorly seen on conventional radiographs, because CT is superior to MR in characterizing the morphology and matrix of a tumor (3).

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