

Spinal Involvement in Alveolar Echinococcosis: Assessment of Two Cases¹

Two cases of spinal involvement in alveolar echinococcosis are reported. Conventional radiographs showed bone lysis and spondylitis; computed tomographic scans showed detail of local spread to the ribs, other vertebrae, and soft tissues. Differential diagnosis, which includes other infections and hydatidosis, is difficult, and the diagnosis may be suggested by evidence of a primary hepatic focus, geographic propensity to the infection, and laboratory findings.

Index terms: Echinococcosis, 761.2083 • Liver, cysts, 761.3121 • Parasites • Spondylitis

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ALVEOLAR echinococcosis (*Echinococcus multilocularis*, causitive agent) can be distinguished from unicellular echinococcosis (*E. granulosus*). The radiologic patterns of alveolar echinococcosis and its primary hepatic focus have been described (1, 2). Secondary involvement of other sites is rare, and involvement of bone is exceptional (3-6). We describe two patients with spinal involvement secondary to a hepatic focus.

CASE REPORTS

Case 1.—A 73-year-old man was seen with a 2-month history of symptoms suggesting progressive spinal compression at T-8. Standard radiographs and computed tomographic (CT) scans showed destruction of the T-8 vertebral body and a lysis of the right pedicle of T-8, with a paraspinal abscess. Spinal compression was confirmed at myelographic examination (Fig. 1). During decompressive laminectomy, the surgeon found evidence of pseudo-Pott disease. Histologic study of the lesion showed multiple vesicles surrounded by a wrinkled, acellular cuticle floating in a granular, mucoid substance with a dense, fi-

brous stroma (no germinal membrane or scolex). Immunologic tests were positive for *E. multilocularis*. Further evaluation demonstrated a 1-cm-diameter lesion in the right hepatic lobe. This was hyperechogenic and homogeneous on ultrasound (US) study and had low attenuation on CT scans. Despite fluoromebendazole (Flubendazole, Janssen) therapy, the patient's paraplegia from unrelated cardiorespiratory causes worsened 5 months after laminectomy. The patient died 13 months after diagnosis.

Case 2.—A 54-year-old woman was hospitalized because of rapidly progressing spinal compression at the T-7 level. CT scans revealed compression of the bodies of T-7 and T-8 and spondylitis and kyphosis at T-7 and T-8 (Fig. 2a); myelographic study showed a complete block, with signs of contiguous epiduritis. The findings at surgery gave the appearance of a neoplastic epiduritis. Biopsy specimens showed lesions compatible with alveolar echinococcosis, and this was confirmed by immunologic study. US examination of the liver revealed two small hyperechogenic nodules in the right lobe. At CT scanning these were seen as areas of low density, with slight peripheral calcifications, that did not enhance on administration of intravenous contrast material. Despite high doses of Flubendazole and attempted bone transplant, standard radiographs showed a slight increase in bone lysis. Epiduritis was more pronounced on myelographic studies. CT scans demonstrated the extent of bone destruction and spread to adjacent soft tissues and bone (Fig. 2b). These lesions did not enhance with administration of intravenous contrast material.

The patient's condition then appeared to stabilize. At follow-up study 1 year later, the hepatic lesions showed no change on US scans; CT scans revealed greater calcification. The extent of lysis in vertebral bone appeared identical to previous examinations, while soft-tissue infiltration decreased (Fig. 2c). When metal rods were implanted in the paravertebral spine, biopsies of bone revealed the continued presence of parasitic structures.

DISCUSSION

E. multilocularis is a parasite once believed to be limited to Europe, Japan, the Middle East, and Alaska (7) but is

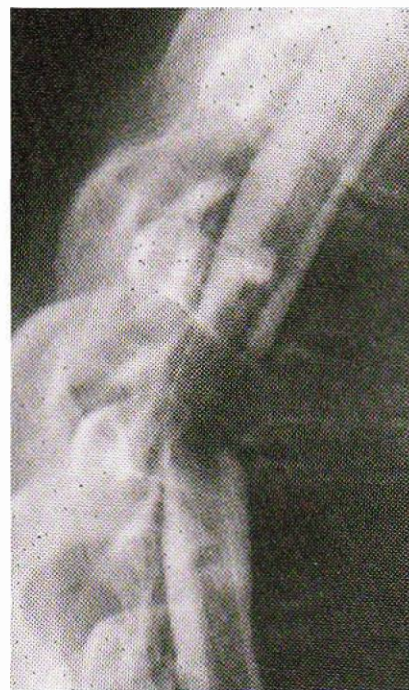


Figure 1. Lateral view obtained at myelography shows compression of the body of T-8 and of the anterior extra-dural spine with no evidence of epidural inflammation.

now found in the United States, China, India, and Tunisia (8, 9). The primary site of infestation is the liver, and its radiologic patterns have been reported (2, 9).

The pathogenesis of the secondary infestation remains poorly understood. There may be vascular or lymphatic migration of a fertile cell from the hepatic focus (10). The rarity with which fertile cells (germinal membrane or scolex) are found in the liver (11) may explain why metastases are rare, even though lesion development is slow, sometimes requiring several decades. The lungs and brain are involved only occasionally (1.5%-10% of cases) (3, 4, 6). Bone involvement is exceptional, but several early observations reported involvement of the skull, ribs, and spine, the latter of which were believed to be extensions of liver involvement (5).

The essential radiologic sign is bone lysis, which appears to begin in

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the vertebral body and spread to the posterior arch, disks, adjacent vertebrae, and the epidural space, creating a picture of spondylitis, with a paravertebral "spindle." These signs are not specific for alveolar echinococcosis; costal involvement and lack of uptake of contrast medium at the lesion periphery at CT have also been described in vertebral unilocular echinococcosis (12). A history of possible exposure to the organism (agricultural work, contact with foxes) and the finding of a slowly progressive, well-tolerated nodular hepatomegaly may suggest the diagnosis and the need for a serologic examination. The appearance of neurologic signs may lead to decompressive laminectomy; surgery may then allow a histologic diagnosis.

Macroscopically, the lesions take on the appearance of pseudo-Pott disease or a pseudoneoplasm, depending on the extent of the inflammatory reaction relative to the bone necrosis. Microscopically, a cuticle is the sign of an echinococcosis (Fig. 2d), as there are no fertile parasite cells. It is difficult to distinguish from a hydatid cyst at histologic study, as the latter takes on a multilocular appearance in the bone (13). A middorsal site of involvement has also been noted in vertebral hydated disease (12). Immunology now provides reliable means for differentiating the two *Echinococcus* species: the enzyme-like immunoabsorbant assay (ELISA) and immunoelectrophoresis (14).

Radiologic examinations are required to follow the extent of bone lysis and degree of spinal compression. As shown by case 2, however, an apparent stability of bone lesions, even combined with partial regression of the infiltration of soft tissues, does not signify eradication of infection. The role of Flubendazole treatment is difficult to evaluate in case 2, given the spontaneous and capricious course of alveolar echinococcosis. ■

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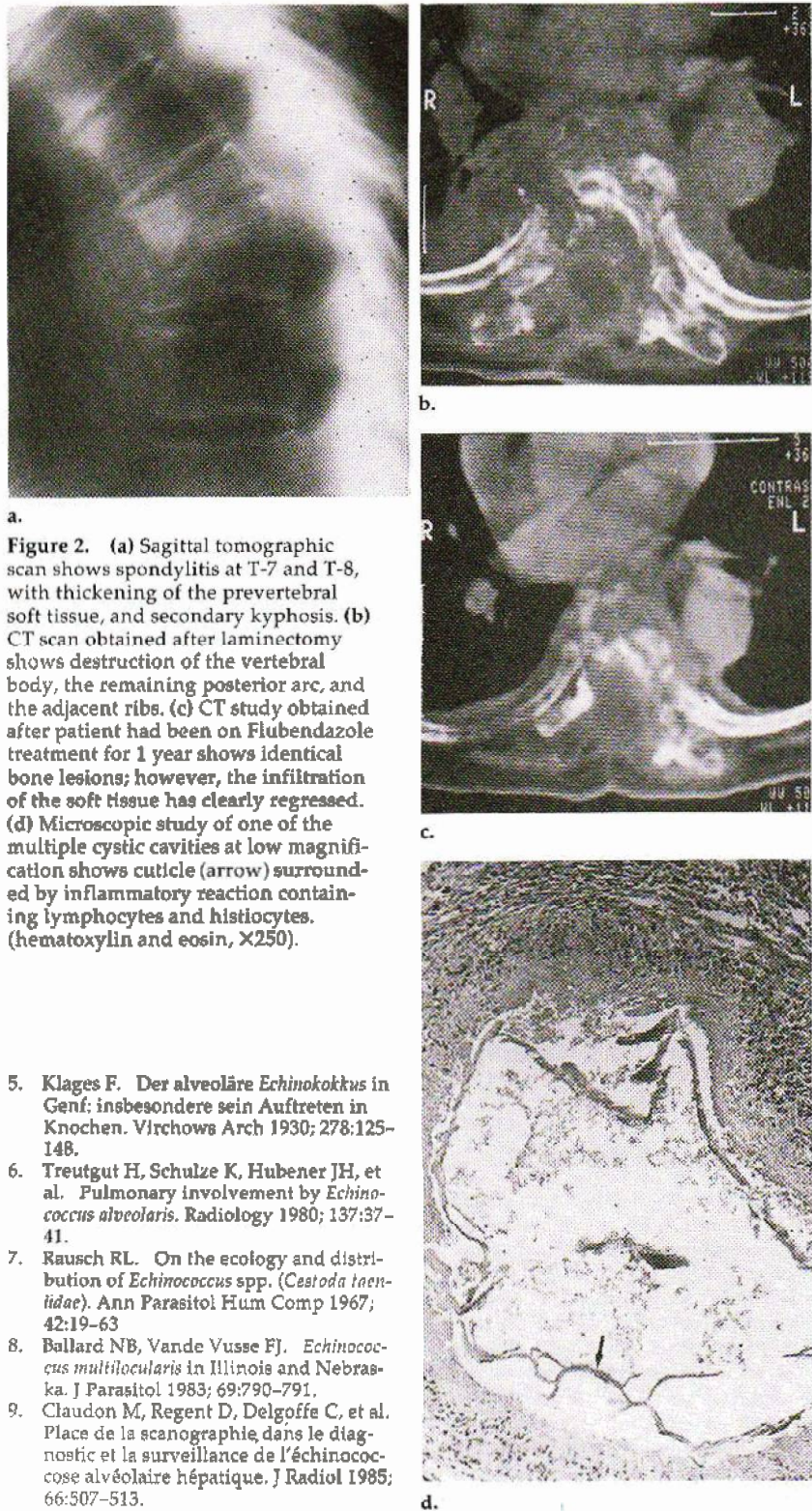


Figure 2. (a) Sagittal tomographic scan shows spondylitis at T-7 and T-8, with thickening of the prevertebral soft tissue, and secondary kyphosis. (b) CT scan obtained after laminectomy shows destruction of the vertebral body, the remaining posterior arc, and the adjacent ribs. (c) CT study obtained after patient had been on Flubendazole treatment for 1 year shows identical bone lesions; however, the infiltration of the soft tissue has clearly regressed. (d) Microscopic study of one of the multiple cystic cavities at low magnification shows cuticle (arrow) surrounded by inflammatory reaction containing lymphocytes and histiocytes. (hematoxylin and eosin, X250).