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Alveolar echinococcosis of the femur

A case of alveolar echinococcosis with involvement of the femur is described. Bone involvement is extremely rare in alveolar echinococcosis. Attention is drawn to the difficulty of diagnosis, and the therapeutic possibilities are discussed.

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Echinococcosis in humans is a parasitical disease which can be traced to two different kinds of tape worm: *Echinococcus multilocularis* (alveolaris) and *Echinococcus granulosus* (hydatidosus). Animals of prey may be considered as being final hosts for both types of worm. The tape worm eggs, excreted in the faeces, are absorbed by an intermediate host (sheep, cow, horse, human) (Boch & Supperer 1983). This can occur by direct contact with the final host, or by eating contaminated vegetables or forest fruits. The fins, which are released in the duodenum, penetrate the wall of the intestine and become lodged mainly in the liver and the lungs. Almost 70% of primary *Echinococcus* cysts are found in the liver; the right lobe of the liver is more affected than the left (Marcial-Rojas 1966). Extrahepatic and extrapulmonary infections are rare.

The two types of *Echinococcus* exhibit differing forms of growth: the *Echinococcus granulosus* grows into the affected host tissue and forms large single cysts, while the *Echinococcus alveolaris* forms multilocular small cysts.

In intraosseous infection caused by *Echinococcus granulosus*, mainly cancellous bone, infiltrated by many small, separate, thin-walled cysts is affected. On the other hand, marginal *Echinococcus* infections, which originate in the bone, may exhibit the typical picture of large cysts.

While the diagnosis of bone infection due to *Echinococcus granulosus* is easy to make and can be confirmed by biopsy, that of *Echinococcus alveolaris* is much more difficult. The ac-

companying necrosis of bone and soft tissues, and the formation of fluid, often lead to the wrong diagnosis of tuberculosis. The difficulty of diagnosis and the rarity of the disease explain why alveolar echinococcosis has seldom been described up to now (Jaffé 1971).

Case report

A 45-year-old female, Greek guest worker had noticed recurring, increasing pain in the right thigh for about 2 years. There had been no preceding trauma. About 4 weeks before admission to hospital, she had consulted her doctor, and radiographs of the right thigh then showed extensive patchy osteolytic foci; she was admitted to hospital because of suspected osteomyelitis (Figure 1).

The bone-scan showed a marked accumulation in the central and distal areas of the thigh, but no further bone involvement. In order to find out whether there was a malignant, neoplastic process, a percutaneous biopsy was taken from some foci under image intensifier control.

The histological examination revealed an extensive, partly chronic, partly still very acute osteomyelitis. The inflammatory reaction was distinctly varied. Beside granulomatous, in part epithelioid cell changes, an infiltrate with a high density of inflamed cells was established, in which mainly plasma cells, lymphocytes, macrophages and histiocytic giant cells dominated the picture. The most marked and impressive finding – lying between the destroyed and necrotic trabeculae of bone – was a system of communicating cavities, consisting of a PAS-positive layered cuticula free of cells, and containing necrotic, slightly granular, gelatinous material (Figure 2).

In the course of further diagnostic work-up, i.e. with the aid of a sonograph, we were able to verify the presence of alveolar *Echinococcus* cysts in the right lung. It was therapeutically necessary to remove the *Echinococcus* cysts in the lung and in the liver as completely as possible. According to the patient's wish, this operation was carried out in her native country. After complete healing, the patient was admitted to our department, where we removed the lesion in the upper thigh by curettage of the foci and irrigation with a hypertonic saline solution.

Post-operatively, the patient was given a Thomas splint. The Nebendenzol-Vermox therapy, which had been started pre-operatively, was now continued post-operatively. This compound effects only a stoppage of growth in the *Echinococcus alveolaris*, not its destruction. The aim of this post-operative treatment

was to prevent the spreading of any material which might still have been present.

At the last examination, 11 months after the operation, the patient was subjectively without symptoms, and radiography showed no increase in osteolytic foci (Figure 1B). The corresponding blood parameters are at present moving towards normal.

B

Figure 1. Echinococcus infection in the left thigh.
A. Extensive osteolytic foci in the distal femur.
B. Condition 11 months post-operatively.

A

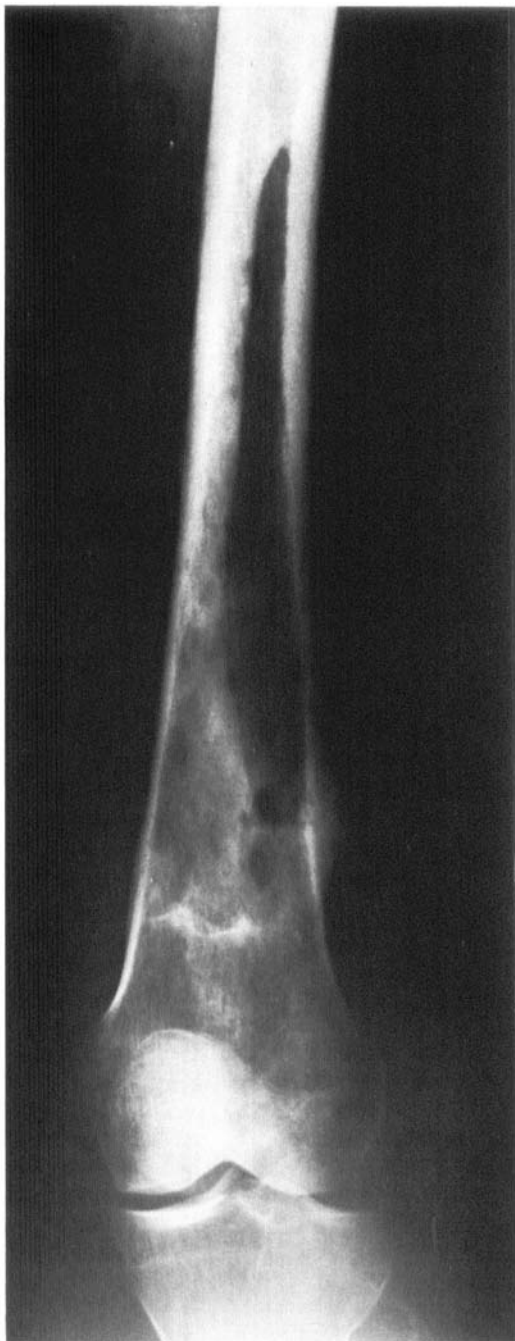
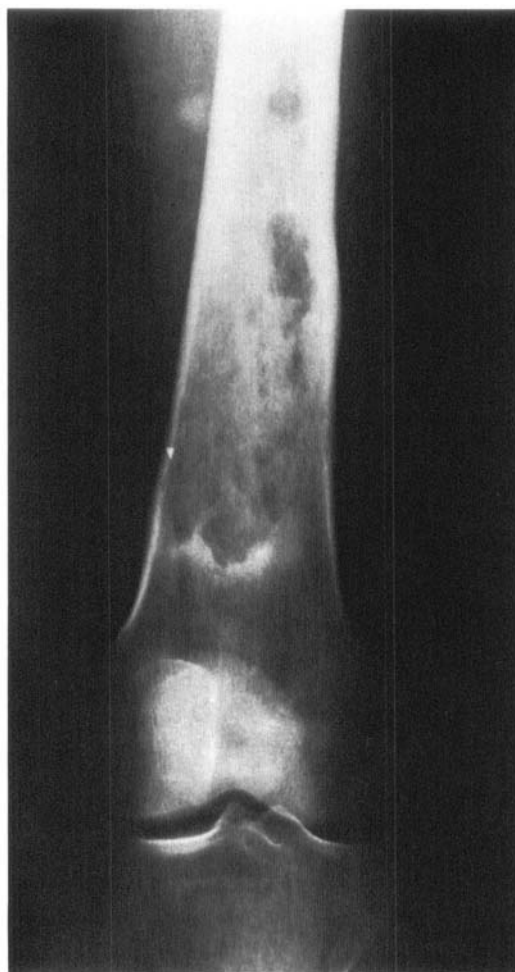




Figure 2. Histological sample of the osteomyelitis material removed at biopsy.

Discussion

Although echinococcosis is the most frequent bone disease in the world which is caused by a worm infection (Jaffé 1972), this condition is very seldom observed. Hooper & McLean (1972) estimated that the skeleton is involved in only 1 percent of manifest *Echinococcus* infections. Little (1976) did not find one single case where bone was involved among 111 cases of *Echinococcus hydatidosus* in Australia.

In the industrialized countries of the Western world, a high level of hygiene is found in the keeping of pets and domestic animals, thanks to a well-developed veterinary control system. The observations in the more recent literature about bone involvement in echinococcosis have come mainly from the sheep-breeding countries of the Mediterranean, the

Near and Far East and Australasia (Hogg et al. 1970, Bouz 1975, Hooper & McLean 1977, Mnaymneh et al. 1977, Duran et al. 1978).

Whilst the positive Casoni skin-test and the classical X-ray findings make diagnosis relatively easy, the extremely rare bone involvement in *Echinococcus alveolaris* infection is much more difficult to diagnose. As in our case, there is the danger of a wrong interpretation (tubercular osteomyelitis) clinically, and by X-rays. Only microscopic examination can reveal the correct diagnosis.

Apart from the macroscopic appearances of the different cystic structures of *Echinococcus granulosus* and *Echinococcus alveolaris*, it might be difficult to distinguish these two types of *Echinococcus* infection by microscopic examination. However, the larval stage in tissues differs in several respects (Bähr 1981, Rausch 1956, Vogel 1957). Whereas in granulous infection the vesicle is surrounded by a connective-tissue capsula which has no budding, the vesicle in alveolar echinococcosis has extensive budding, lacks a retaining capsula, and invades and destroys adjacent tissue. In *Echinococcus granulosus*, numerous scolices are found attached to the cyst membrane or floating free in the hydatid fluid, but in *Echinococcus alveolaris* there is little fluid in the cavities and scolices are rare. Taking into consideration these morphological aspects, we diagnosed alveolar echinococcosis in our case.

Hooper & McLean (1977) and Mnaymneh et al. (1977) agree that the only definite treatment of bone infection due to echinococcosis is en-bloc resection of the bone or amputation of the limb. In those cases in which such radical treatment cannot be considered, Hooper & McLean (1977) recommend curettage of the infected bone area, followed by irrigation with hypertonic saline solution, as this inhibits the growth ability of the parasites. This procedure does not cure the illness, but halts it, so that an asymptomatic course can be achieved, as Duran and colleagues (1978) noted in more than half of their patients.

Our case underlines the necessity of including *Echinococcus* infections in the differential diagnosis in lytic bone processes, and of backing up the diagnosis with biopsy and histology.

This is especially important for patients from the countries of Eastern and Southern Europe, and from the Mediterranean.

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