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Primary Intraosseous Hydatid Cyst: A Case Report

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ABSTRACT

Introduction: Hydatid cyst is primarily localized in the liver. Primary involvement of long bones is rare.
Case Presentation: We report a case of primary hydatidosis of the thigh, collected in the Department of Traumatology and Orthopedic Surgery at Ibn Sina University Hospital in Rabat. The patient was a 22-year-old woman. The clinical symptoms were nonspecific. Standard radiography and bone CT scans were suggestive. Chest radiography and abdominal ultrasound were normal. The hydatid serology was positive. She underwent surgical excision and was treated with Albendazole for six months. Histology confirmed the diagnosis. The outcome was favorable, with a median follow-up of two years.
Conclusion: Through this case, a review of the literature helps to highlight the epidemiological, clinical, and paraclinical features, as well as the therapeutic modalities for primary bone hydatidosis.

KEYWORDS

Hydatid disease, hydatid cyst of the bone, bone tumor.

MAIN ARTICLE

Introduction:

Bone echinococcosis, or bone hydatid cyst, is a parasitic condition caused by the larval stage of *Echinococcus granulosus* (or more rarely *Echinococcus multilocularis*) in humans, who serve as an accidental intermediate host. The adult form of the parasite lives in the intestines of dogs or other carnivores [1]. Bone echinococcosis is rare [2,6], accounting for only 0.9 to 2.5% of localizations [2,4,7]; its frequency is much lower than that of liver (60 to 70%) and lung (20 to 30%) localizations [2,4,8]. This condition remains asymptomatic for a long period (15–40 years), with the first manifestations appearing late. Treatment is both medical and surgical. We report a case of primary hydatidosis of the femur, collected from the Department of Traumatology and Orthopedic Surgery at Ibn Sina University Hospital in Rabat.

Case report:

A 22-year-old woman from a rural background, with no significant medical history, presented to the general medical consultation for an inflammatory pain and swelling of the right thigh persisting for several weeks. Laboratory tests showed leukocytosis (11,200 WBC/mm^3) without eosinophilia, an elevated erythrocyte sedimentation rate (70 mm in the first hour), a C-reactive protein (CRP) of 92 mg/l, and normal liver function tests. The patient was treated with analgesics (paracetamol) and antibiotics (ciprofloxacin 500 mg a day for 14 days), but after several weeks, the pain persisted. She was referred to the Traumatology and Orthopedic Surgery clinic. X-rays revealed bone tumor in the femur, and MRI showed a cystic tumor mass, non-fistulated, compatible with a hydatid cyst. Abdominal ultrasound and chest radiography showed no abnormalities. Hydatid serology was positive. Surgery confirmed the presence of a hydatid cyst at the junction of the middle and lower thirds of the femur, with vesicles present. Complete excision of the lesion was performed, and histological analysis confirmed the diagnosis of a hydatid cyst of the bone with non-hooked membranous elements within bone fragments. Postoperatively, the patient was treated with Albendazole at a dose of 10 mg/kg/day for six months, with six one-month cycles spaced 15 days apart. The outcome was favorable, with a three-year follow-up and no local recurrence.

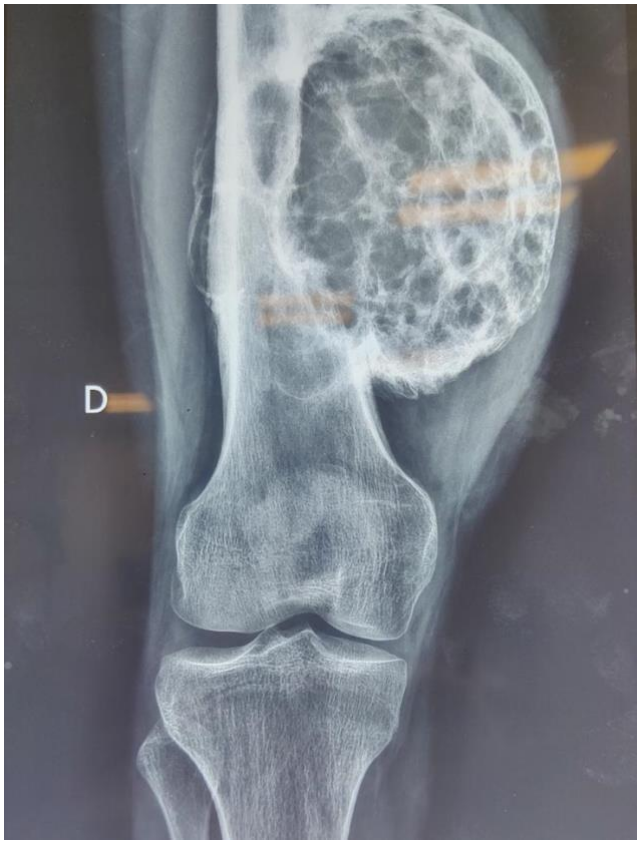
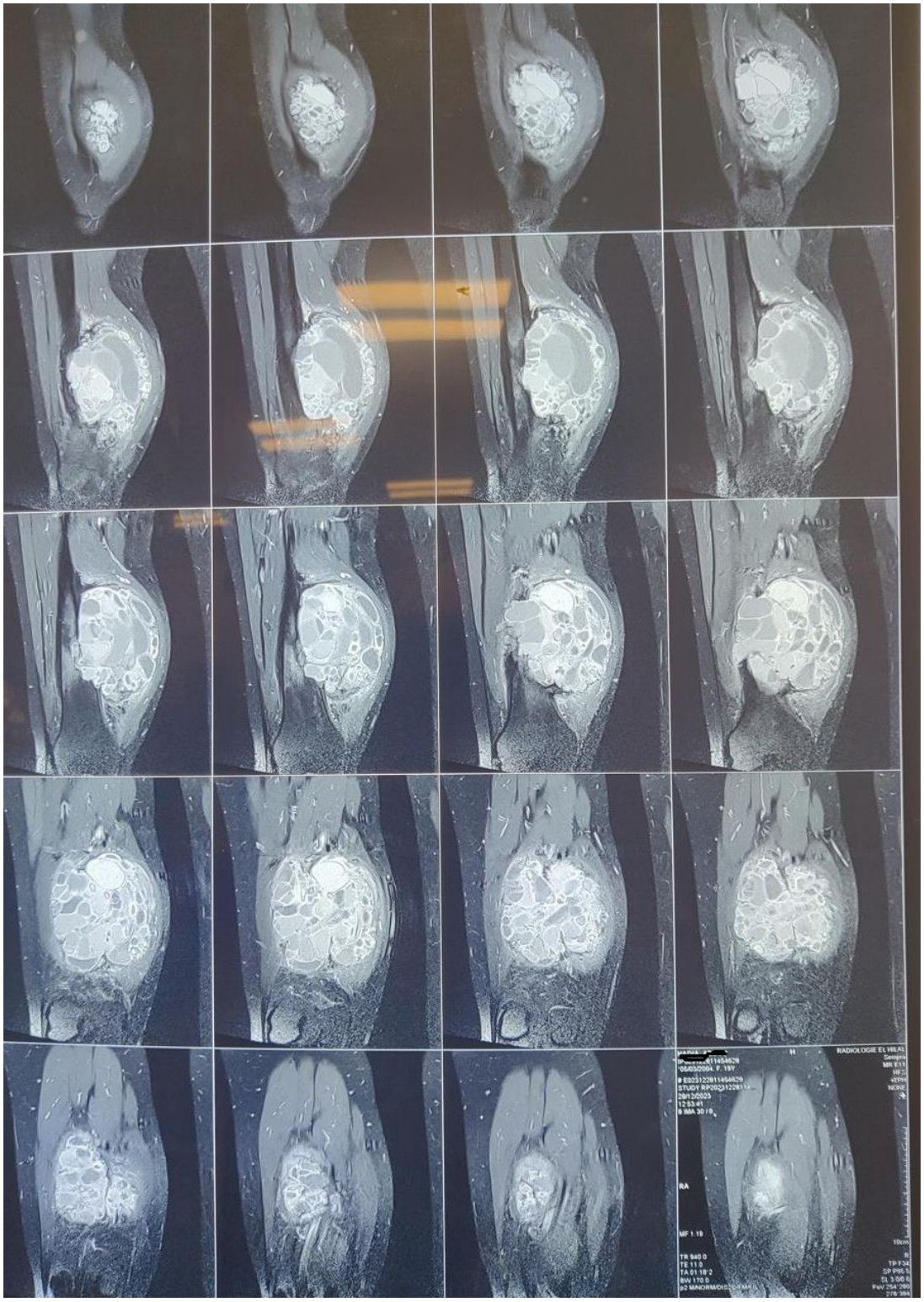


Fig 1 : Standard radiograph showing an oval-shaped formation displacing adjacent soft tissues, containing multilocular formations that erode the cortical bone on the internal side.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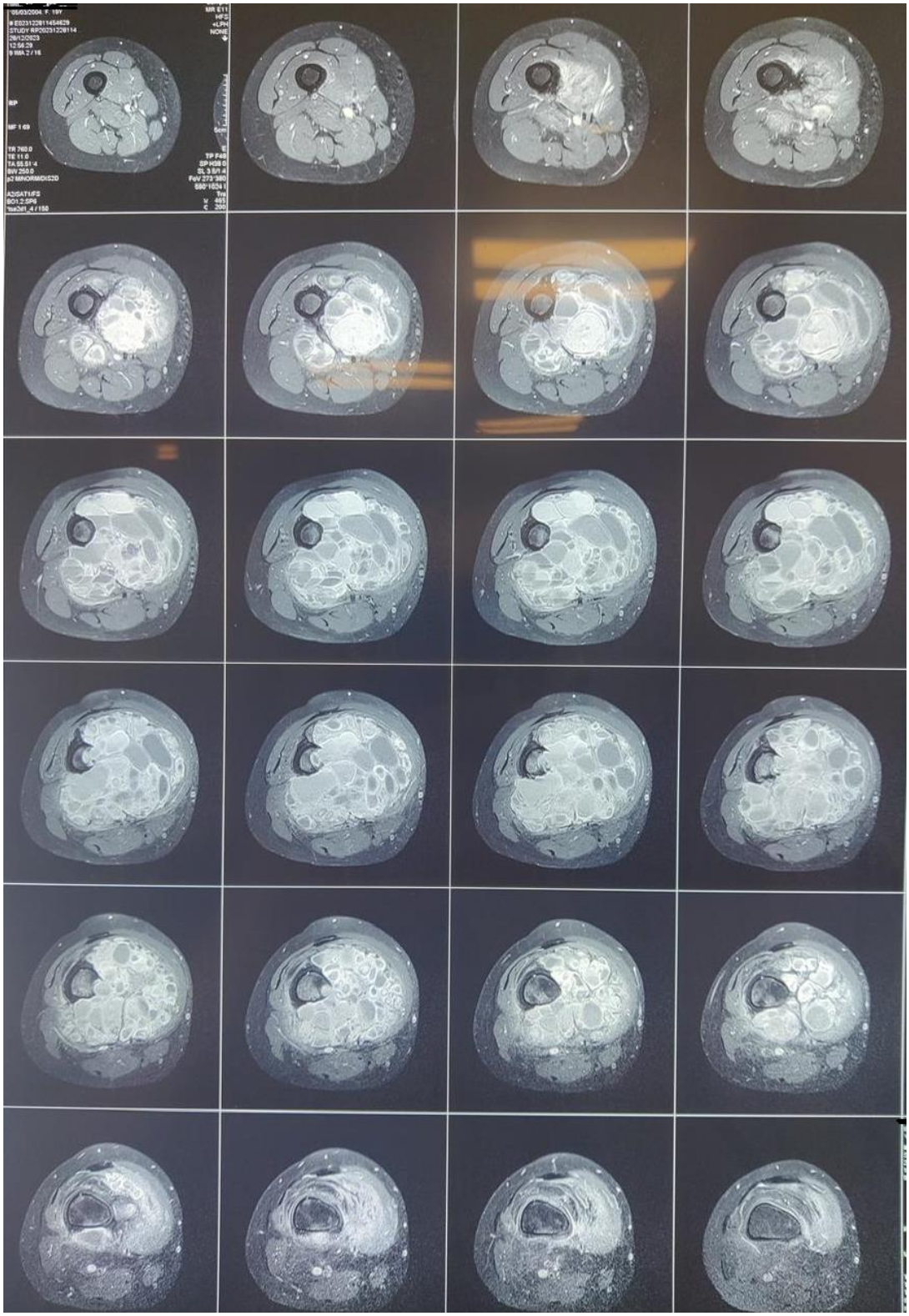

Fig 2-3 MRI of the right lower limb in T1, FATSAT sequence after gadolinium injection. Axial and coronal cuts show a roughly oval formation containing rounded, septated structures with enhanced walls, consistent with daughter vesicles, suggesting a probable hydatid cyst that does not fistulize to the skin and shows no signs of soft tissue invasion. The cortical bone is eroded without periosteal reaction of a suspicious nature, and there are no signs of aggressiveness.

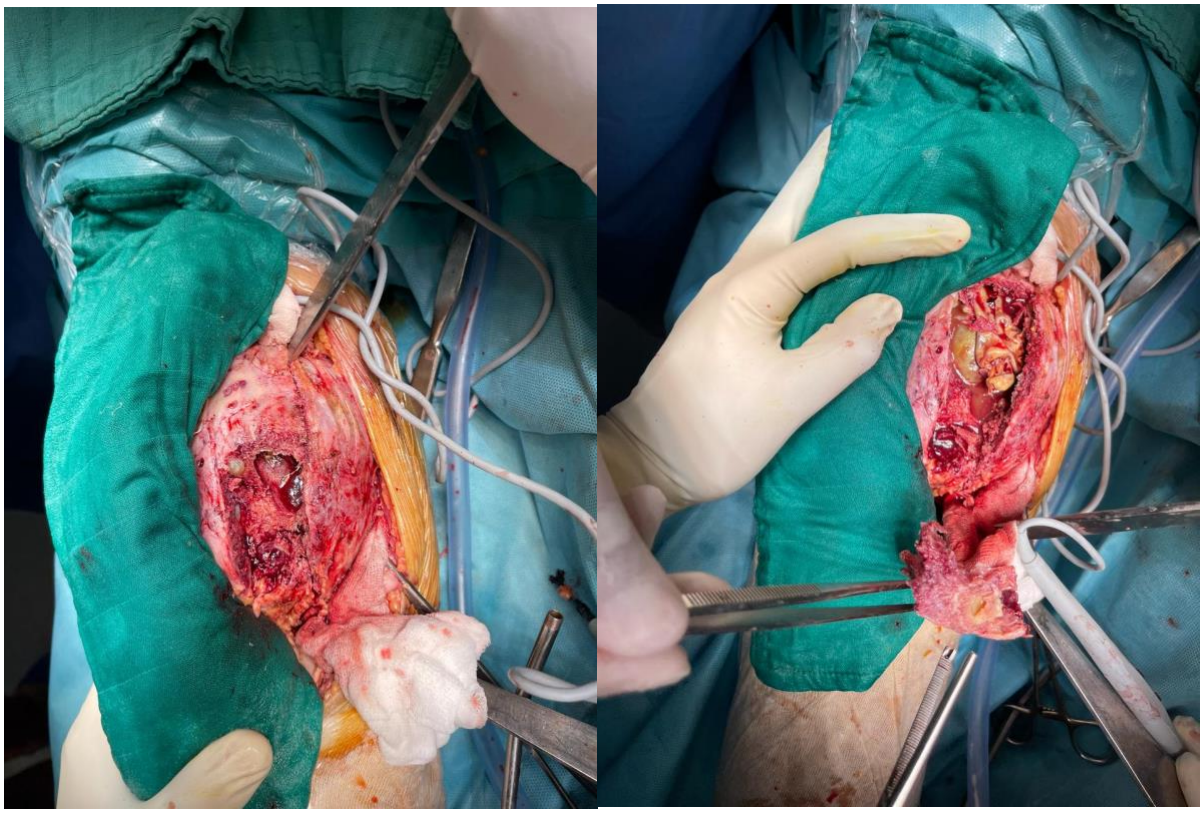

Fig 4 : Peroperative images.

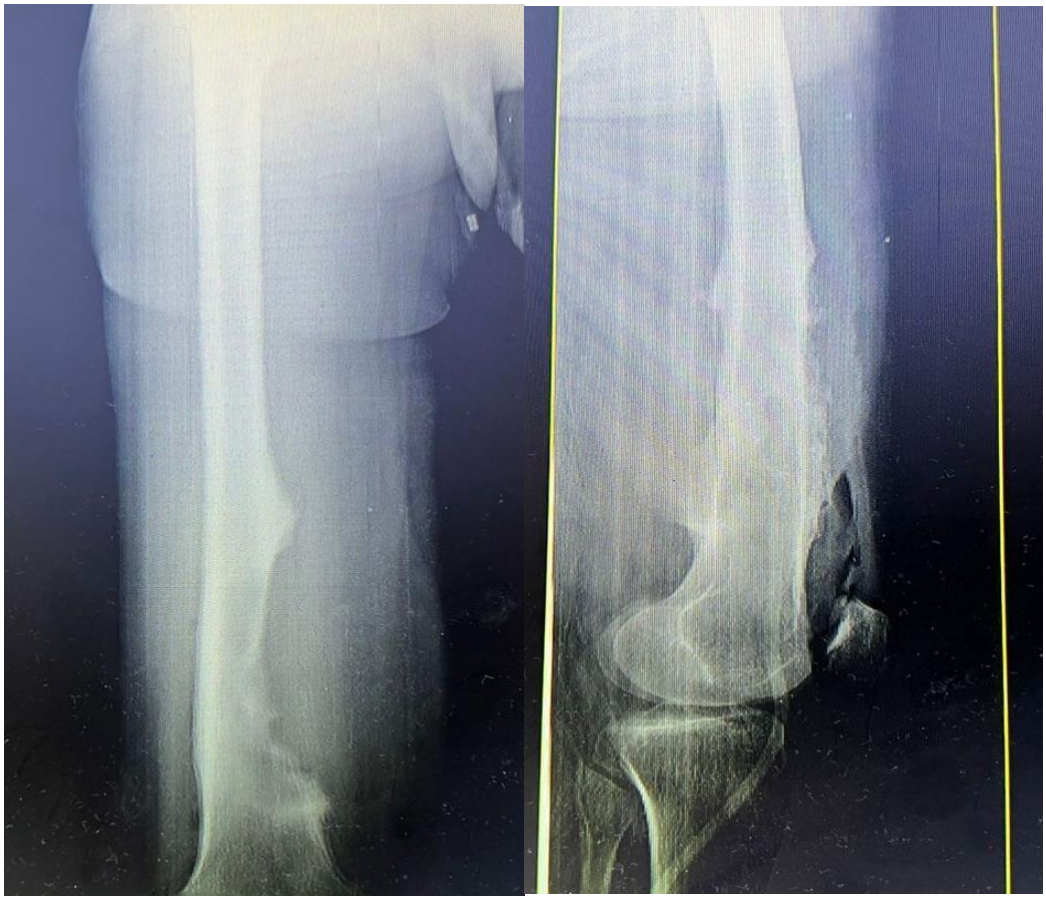

Fig 5 : Postoperative standard radiographs, front and lateral views.

Discussion:

Hydatid cyst preferentially affects the lungs (20-30%) and liver (60-70%)【2,4,8】. Uncommon localizations are numerous, including the spleen, kidneys, heart, muscles, and brain【4,8】. Bone involvement is rare, accounting for 0.9 to 2.5% of all localizations【2–6】. The most commonly affected bones are the spine (44%), ilium (16%), femur (15%), humerus (7%), tibia (6%), and skull bones (3%).

Human infestation is accidental, occurring solely through ingestion of eggs, which suggests proximity to livestock areas【1,4】. Bone involvement typically occurs through hematogenous spread【9】, though it can also result from primary soft tissue involvement. Initially, medical treatment with benzimidazoles is recommended for inoperable cysts. These drugs have since been used to reduce cyst size, sterilize their contents before surgery, and in the postoperative phase for small cysts that were overlooked【9–12】. The mechanism of action of albendazole is to inhibit glucose absorption by the parasite. The results of medical treatment for hydatid cysts are variable across studies, with success rates ranging from 43.5% to 80%. This variability is due to the different compositions of the study groups, variations in therapeutic regimens, and the localization of cysts. The current treatment for bone echinococcosis is medical-surgical. Albendazole is preferred due to its better gastrointestinal absorption【2,3,14】. The distinctive feature of this molecule compared to other benzimidazoles is its good absorption in the intestinal mucosa, which enables an effective action against extra-intestinal visceral helminthiases. It is rapidly metabolized to albendazole sulfoxide. In humans, clinical and biological tolerance is good, even at high doses over extended periods, as seen in the treatment of hydatid disease. Minor digestive side effects have been reported. In patients operated on after albendazole treatment, samples from the wall and hydatid fluid revealed variable levels (50 to 4000 ng/ml) of the drug (921 ± 314 ng/ml in hydatid fluid). According to World Health Organization (WHO) recommendations, albendazole is prescribed at a daily dose of 10-15 mg/kg, divided into two postprandial doses, in four to six 4-week courses surrounding the surgical procedure, with intervals of two weeks between courses【1,4,13】. Regular monitoring of liver function is advised. We adopted this therapeutic protocol for our two patients, with good results. These two favorable outcomes also raise the issue of defining cure criteria. Some authors have emphasized the importance of bone CT or, preferably, MRI imaging, which allows for a more precise analysis of lesions, confirmation of the cystic nature of geodes, and evaluation of the extent of the disease, regional extension to soft tissues, lack of enhancement after contrast injection, and detection

of extraosseous abscesses【3,6,9】. These exams are crucial for assessing surgical possibilities and are useful in postoperative follow-up to detect residual lesions and recurrences【5,15】.

Bone malignancies and hydatic cyst disease are treated surgically however, more aggressive surgery is needed in malignant diseases. In addition, patients with hydatid disease should be treated with antiparasitic drugs before surgery. In the past years these cases were considered as malignant diseases and hemipelvectomy surgery was applied5-8.

Current surgical treatment is wide excision of hydatid cyst cavity after the application of hypertonic solution. And also albendazole treatment are suggested after surgery. Recently, systemic chemotherapy is recommended as a benzimadozole, particulate mebandozol, albendazole, and combination treatment in clinical and experimental studies [16]

Bone hydatid cyst disease is a benign disease and may be confused with malignant diseases of the bone because of the behavior. Therefore, the differential diagnosis should be kept in mind. Treatment is extended surgical excision and albendazol.

Cardona et al. highlight the importance of serology for monitoring disease progression when positive before treatment【12】. However, there is a significant proportion of false negatives related to the location of the cyst (brain, bone, eye)【1】. For example, Lapierre et al. [13] reported 11% false negatives for liver hydatid cysts and 35% for bone localizations.

Antibiotic therapy can prevent severe and sometimes fatal infectious complications. As for surgery, it should be "oncological" in nature, as the recurrence rate after partial excision is very high【2,3】.

Conclusion :

Bone hydatidosis remains a rare localization, even in endemic areas such as Tunisia. Its diagnosis is often delayed due to its insidious progression. The best treatment appears to be the combination of Albendazole and surgery. Currently, it is difficult to consider a definitive cure for echinococcosis.

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