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Recurrent hydatid cyst of the Pelvis: A rare case

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Abstract
Hydatic echinococcosis is preferentially localized to the liver. Primary bone localization is rare, characterized by high clinical latency, lack of radiological specificity and difficulties of its surgical treatment. We report the case of a 20-year-old young man with a hydatid cyst in the pelvis, diagnosed by a pain in his left hip and a lytic image of the ilio-pubic branch on X-ray. Treatment consisted on a resection of the cyst by both posterior and abdominal approaches, combined with antiparasitic treatment. The functional result was satisfactory, but the hydatid cyst recurred after 3 years, a medical treatment was considered using albendazole for one year. Currently at 12 years of follow up, the patient was asymptomatic and radiologically the hydatic cyst was stationary. The quality of the surgical resection is very important for the risk of recurrence. Pelvic localization of hydatid disease is particularly difficult to treat. Medical treatment using albendazol could be a reel alternative for the difficult cases not treated surgically.

Keywords: hydatid cyst, pelvis, albendazole

1. Introduction
Hydatid disease is a parasitosis caused by human development of the larval form of Echinococcus granulosus [1]. This disease is relatively widespread in the Mediterranean, the Middle East, Central Asia, East and parts of South and North America [2]. Bone echinococcosis desease is commonly observed in the liver and lungs, and presents only 0.9 to 2.5% of all sites [3]. The most frequently described osseous sites are the vertebrates, the long bones of the lower limbs, the pelvis, the hip joint, the ribs and the scapulas [2]. Its high clinical latency and lack of radiological specificity often lead to a delayed diagnosis contributing to the extension of the parasitic osteitis. Despite the progress of the reconstructive bone surgery and antiparasitic drugs, the treatment remains difficult. We report a rare case of a hydatid cyst of the pelvis illustrating the contribution of imaging in the diagnosis and the different therapeutic difficulties.

2. Patient and Observation
A 20-year-old patient living in the rural area, with a history of liver hydatid cyst in his brother, who is presented at the orthopedic consultation for a mechanical pain in his left hip with a slight lameness at walking, without fever or alteration of the general state. The general examination found a patient in good general conditions, apyretic, with a normal range of motion of his left hip wich is painful at walking with no palpable mass or an inflammatory sign, the rest of the somatic examination was normal. A standard x-ray of this hip was performed and showed a lytic image of the ilio-pubic branch and the bottom of the acetabulum on the same side of the pain (Figure 1), the bone lysis also involved a part of the femoral head. The standard biological tests had not shown any abnormality with white blood cells at 6000 / mm3 and a c-reactive protein at 6 mg / l. In front of this table the diagnosis of hydatic desease was mentioned, among others, and a complementary tests were obtained, in particular a hydatic serology which was positive, a pelvic ultrasound which has shown an image of compartmentalized liquid echo-structure and a computed tomography of the pelvis (Figure 2), which described multiple lytic and cystic lesions involving the left ischium, the roof of the acetabulum and the femoral head, with an extension of cystic lesions into the laterovesical side. The diagnosis of pelvis hydatid cyst was then retained without other localizations.

The therapeutic decision was to operate this hydatid cyst, the surgical procedure involved two surgical times with two first seeing: a posterior approach of Kocher Langenbeck and an abdominal approach and two surgical teams: orthopedic and visceral surgery teams. The surgical procedure consisted on a resection of the tumor and sterilization of the site by hypertonic serum. Medical treatment using albendazole at a dose of 10mg / kg / day, 4 weeks / 6 weeks, for a period of 6 months.

Controls performed up to one year follow up were satisfactory; the one-year radiography (FIG. 3) had shown a nearly complete filling of the curetted cavity. At 3 years of follow-up the patient was admitted in consultations for a symptomatology close to the initial symptomatology, complementary examinations have been carried out in particular a standard radiography (FIG. 4) and an MRI (FIG. 5) which have detected a recurrence of the hydatid cyst with a much larger bone lysis. The patient's case was discussed between both surgeons and infectiologists and the decision was to treat medically without surgical resumption, the patient was then put on albendazole at a dose of 15mg / kg / day 4 Weeks / 6 weeks for a period of one year with good clinical evolution including a disappearance of pain. Currently at 12 years of follow-up, the patient is asymptomatic, and the CT scan control (FIG. 6) shows a stationary state of the lesions.

3. Discussion
The hydatic echinococcosis is preferentially localized in the pulmonary (20 to 30%), and hepatic sites (60 to 70%). Unusual localizations are numerous: splenic, renal, cardiac, muscular, cerebral.The osseous lesions represent from 0.9 to 2.5% of all localizations, bones in order of frequency are: spine (44%), ilium bone (16%), femur (15%), humerus (7%), tibia (6%) and

skull bones (3%) [1]. This rarity is explained by the path taken by the hexacanth embryo, which after its digestive absorption must pass through the capillaries of the liver and then the lungs, which are smaller than the bone capillaries, and produce a more selective filter [4].

After several years of latent evolution, clinical signs are uncharacteristic and rarely refer to the pathology in question as pain, swelling, fractures or signs of compression. The pelvic lesion involving iliac bone in 16% for wheat, 14.4% for Deve. This localization is of poor functional prognosis byecause of the extension to the coxofemoral joint and more rarely to the sacrum. Abscesses can be spread in the sacrococcygeal region be responsible for nerves compression causing a sypmtomatology of crural and sciatic neuralgia, or sphincter disorders [5].

The clinical examination is poor; it is marked by the preservation of the general state and the apyrexia of the patient. A discreet lameness at walking is observed when the parasitosis is localized in the lower limbs or in the pelvis, the pressure of the concerned region can trigger pain, the neurological examination may reveal signs of root or pyramidal irritation [5].

Conventional radiology is the essential element in the exploration of bone echinococcosis, it can be revealing when the lesion is still asymptomatic, the lesions are characteristic without being pathognomonic, the affected bone is not morphologically modified, this examination Allows to locate the lesion, to appreciate its extent and its regional diffusion. The multigeodic, honeycombing area aspect, infiltrating the bone piece without periosteal reaction is the most characteristic of the hydatic origin of a bone lesion. Perilesional condensation is only seen in the case of surinfection, there is no clear limit between the lesion and the healthy region [5]. Ultrasound allows the exploration of soft tissues in search of the abscess Ossifluent, it contributes as well as chest x-ray, to the assessment of the hydatic disease, in the search for associated visceral localizations that can guide the diagnosis [4]. CT and MRI indicate bone involvement, appreciate locoregional extent, and are an excellent means of monitoring the course of the disease [4].

The curative treatment of bone hydatidosis can only be surgical, its indication and its results depend on the evolutionary stage and especially on the localization of the disease [1]. It consists of a "carcinological" excision of the lesions that are assimilated to a real malignant tumor [4]. At the level of the pelvis, the large excision leading to a major mutilation causes push surgeons to perform successive curettages, the prognosis is often very reserved when the injury is extensive [5]. Disappointing results have been reported following total hip replacement surgery at the hip. Large bone grafts and customized prostheses are required, and the rate of complications is high when there is a large destruction of the hip joint, a hip replacement probably should not be attempted [2].

Until 1977, the date of the first successful use of medical treatment in humans, surgery was the only option to treat hydatid disease, since then several studies using benzimidazole carbamates have been repeatedly reported from the size reduction of the cysts before the surgery, to the complete cure of certain cysts under the medical treatment only, this treatment was initially used for the patients considered inoperable, later other indications were proposed in particular to reduce the size of the cysts, to sterilize their contents before surgery, to avoid the risk of secondary dissemination, and in postoperative treatment to act on small cysts that have gone unnoticed, as well as on any scolex scattered during surgery in order to prevent their cystic evolution [6]. Of all the benzimidazoles, albendazole appears to have better digestive absorption and its clinical and biological tolerance is good even at high doses for prolonged periods. According to WHO recommendations, albendazole is prescribed at a daily dose of 10-15 mg / kg / day; the administration is divided into two postprandial intakes, four to six cures per week framing the surgery, and spaced by two weeks [3].

The optimal time limit for the definitive evaluation of the treatment effectiveness remains unclear. WHO recommended a minimum of 12 months for an objective evaluation, however, a longer follow-up, or even for life, seems necessary both to detect late morphological changes and also possible recurrences, usually occurring in the 2nd or 3rd year after treatment, and which are most often susceptible to a new cure of albendazole [6].

4. Figures

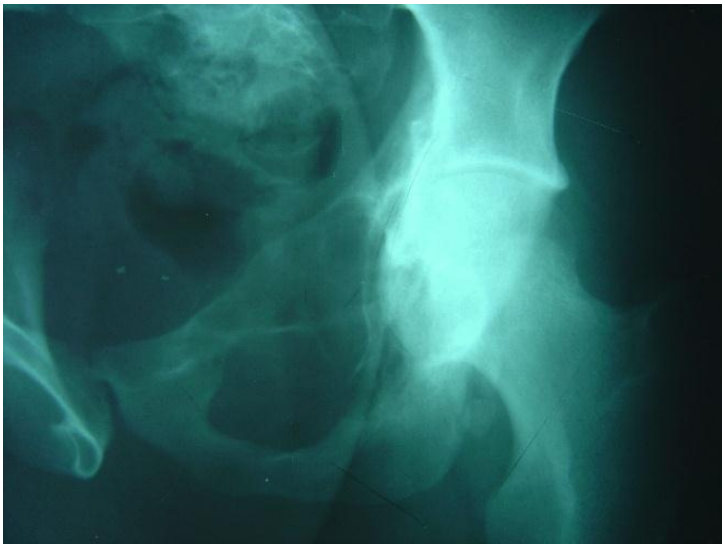

Fig 1: Lytic image of the ilio-pubic branch, the base of the acetabulum and the femoral head.

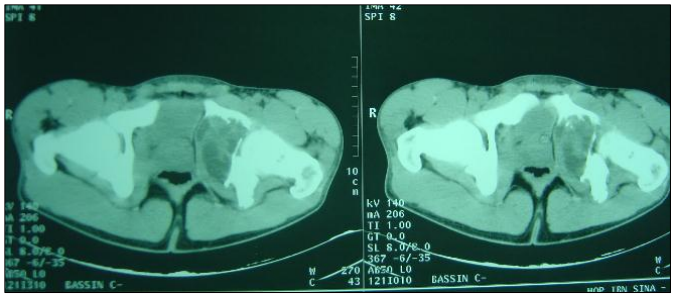

Fig 2: CT imaging showing in addition to the bone lysis intra-osseous, multi-vesicular and latero-vesicular cystic images.

Fig 3: Radiological monitoring at one year after surgery and medical treatment showing good progression with improved bone lesions.

Fig 4: Radiological control at 3 years follow up showing a recurrence of the hydatid cyst with very large bone lysis.

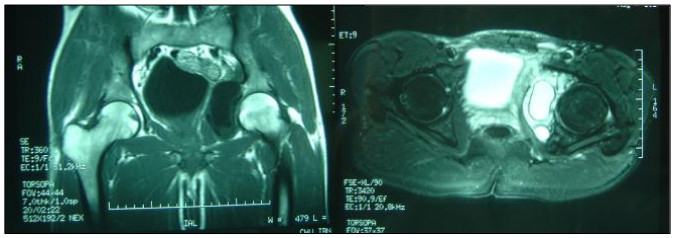

Fig 5: MRI at 3 years follow up to assess the importance of the lesions due to hydatid cyst recurrence.

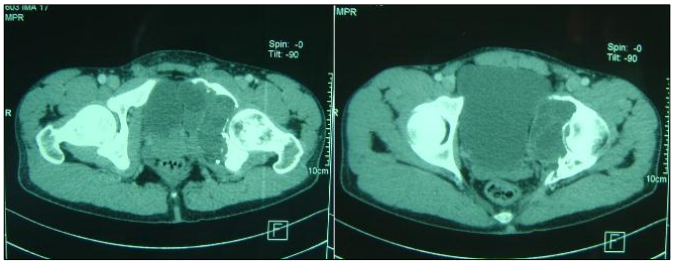

Fig 6: CT scan control at 12 years follow up showing a steady state of the lesions.

5. Conclusions

Bone hydatidosis is a rare, even in endemic areas such as Morocco. Medical imaging allows a precise lesional assessment to plan a surgical procedure and also the follow-up of lesions. The very dilapidating surgical treatment never leads to recovery but only remissions more or less long. Medical treatment seems to be a supplementary option whose effectiveness needs to be proved. Evaluation criteria and timeframes require better definition on larger series.

6. Declaration of Interest

The authors declare that they have no conflicts of interest in relation to this article.

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