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Case Report

Osseous hydatid disease: A mimic of other skeletal pathologies ☆,☆☆

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ABSTRACT

Hydatid disease is still endemic in several regions worldwide including Morocco, and is caused in most cases by the larval form of 2 species of the tapeworm Echinococcus: E. granulosus and E. multilocularis. Primary hydatid disease of the bone without systemic involvement is rare. The disease has a silent clinical evolution until it reaches complicated stages. Complications may include pathological fracture, neural deficit, infection, and fistulization of the abscess. Preoperative diagnosis is based on clinical history, imaging findings, and serological tests, which lack high sensitivity and specificity. Although the interpretation of imaging studies can prove to be very confusing because the bone changes evolve with time, and the nonspecificity of these findings often leads to a mistaken diagnosis. The diagnosis requires a high index of suspicion, especially in patients who reside in or travel to sheep-raising areas where hydatid disease is endemic. A high index of suspicion is necessary for the diagnosis, especially in patients that live in or travel to sheep-raising areas where hydatid disease is endemic. The treatment of choice remains surgical, following the principles of a locally malignant lesion. Chemotherapy (albendazole alone or in combination with praziquantel) is indicated when surgery is not possible or as an adjuvant treatment. The prognosis is often poor. We report the case of a 28-year-old woman with long-standing pain in the left hip joint in which the imaging findings were thought of as being either tuberculous or neoplastic. The result of a CT-guided biopsy concurred with an unexpected diagnosis of a hydatid cyst. This case highlights that in the absence of a high index of sus-

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picion for echinococcal infection, the semblance of imaging findings of hydatid disease in the bone to those of other skeletal pathologies can lead to misinterpretation.

Introduction

Hydatid disease, or Echinococcosis, is a zoonotic disease caused by the echinococcus parasite, which belongs to the family of Taeniidaes [1]. Primary hosts are dogs, wolves, and coyotes, while the intermediate hosts are sheep, cattle, and deer. Human transmission occurs through ingestion of infected food or water, or by direct contact with animal hosts. It may develop in almost any part of the body, Although the liver (60%-70%), and lungs (20%-30%) are the most usually affected organs [1,2]. Bone involvement is rare, and accounts for only 0.5%-4% of all locations. The most commonly described sites for hydatid cysts of the bone are the vertebrae, the long bones of the lower limbs, and the pelvis [3]. According to Zlitni et al. [4] echinococcosis of the pelvis accounts for 28% of all cases of osseous hydatidosis [3].

A combination of clinical history, imaging findings, and serological tests are diagnostic of Hydatid disease. However, histopathological examination is the gold standard. Early diagnosis is uncommon and usually present at an advanced stage of the disease. Therefore, management is difficult and recurrence is common [5]. The prognosis is often poor. We report the case of a 28-year-old woman with long-standing pain in the left hip joint in which the imaging findings were thought of as being either tuberculous or neoplastic. The result of a CT-guided biopsy concurred with an unexpected diagnosis of a hydatid cyst. This case highlights that in the absence of a high index of suspicion for echinococcal infection, the semblance of imaging findings of hydatid disease in the bone to those of other skeletal pathologies can lead to misinterpretation.

Case report

We report the case of a 28-year-old otherwise healthy woman patient, without significant history, who presented with 2 years' history of left hip pain while walking. This had increased in severity in the past few months. She had no fever, chills, weight loss, or trauma history. The physical examination revealed restriction of the left hip joint and pain during movement. At the left groin, we observed poorly limited tumefaction, mesearing 4 cm in greatest dimension, renitent and only slightly sensitive on palpation, there were no signs of inflammation of the superficial skin or lymphadenopathy. A laboratory analysis was performed showing marked anemia with a hemoglobin: 13 g/dL (normal value: 14-16 g/dL); eosinophilia at 800 μL (normal value: 100-400 μL) and elevated CRP levels at 56 mg/L (normal value inferior at 5 mg/L).

A plain radiograph of the pelvis (Fig. 1), showed a mildly expansile lytic lesion involving the periacetabular region of the iliac bone. Scalloped bone erosion appeared more extensive

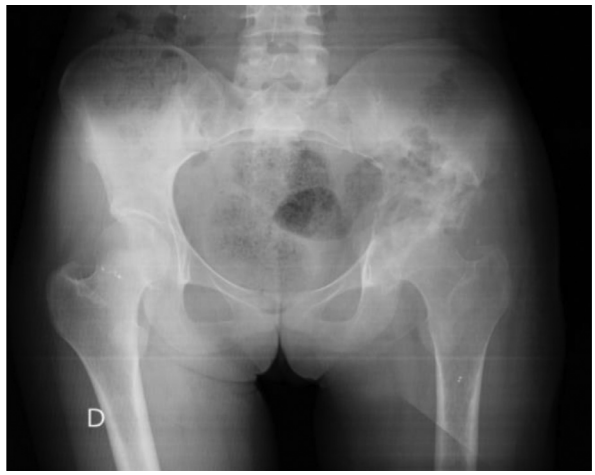

Fig. 1 – A plain radiograph of the pelvis revealed a mildly expansile lytic lesion involving the periacetabular region of the iliac bone. Scalloped bone erosion appeared more extensive along the medial aspect of the iliac wing with scattered hyperdense foci suggesting intrapelvic extension. The femoral head and neck appeared unremarkable. The joint space and acetabular subchondral bone are absent.

along the medial aspect of the iliac wing with scattered hyperdense foci suggesting intrapelvic extension. The femoral head and neck appread unremarkable. The joint space and acetabular subchondral bone are absent. Pelvic CT scan (Figs. 2 and 3) corroborated the above findings and showed an expansile lytic process of the acetabulum to the lytic ilium, multiloculated with a honeycomb appearance, containing calcifications, enhanced at the periphery after injection of the contracted product. CT also showed multiple cortical destruction foci and loss of bony texture in the left iliac bone with a periosteal reaction, and infiltration of the adjacent muscles. The sacroiliac joint, the coxofemoral line and the femoral head were respected. Individualization of a subcutaneous collection communicating with the lesional process. In front of this feature on Imaging, a tubercular or malignant origin was suspected. The workup was completed with an MRI (Fig. 4), which eliminated any sacroiliac or sacral involvement. In the acetabular area, where cystic images were shown on standard X-rays, the MRI demonstrated a microlacunary aspect with a few centimeter-sized formations on hyperintense T2-weighted images. The upper extremity of the femur showed a heterogenous signal in T1- and T2-weighted sequences, but did not seem to be invaded. Individualization of a subcutaneous collection communicating with the lesional process.

At histopathology of a tissue specimen obtained by a CT-guided biopsy, fragments of lamellar membrane were seen that characterized the presence of hydatid cysts (Fig. 5). There

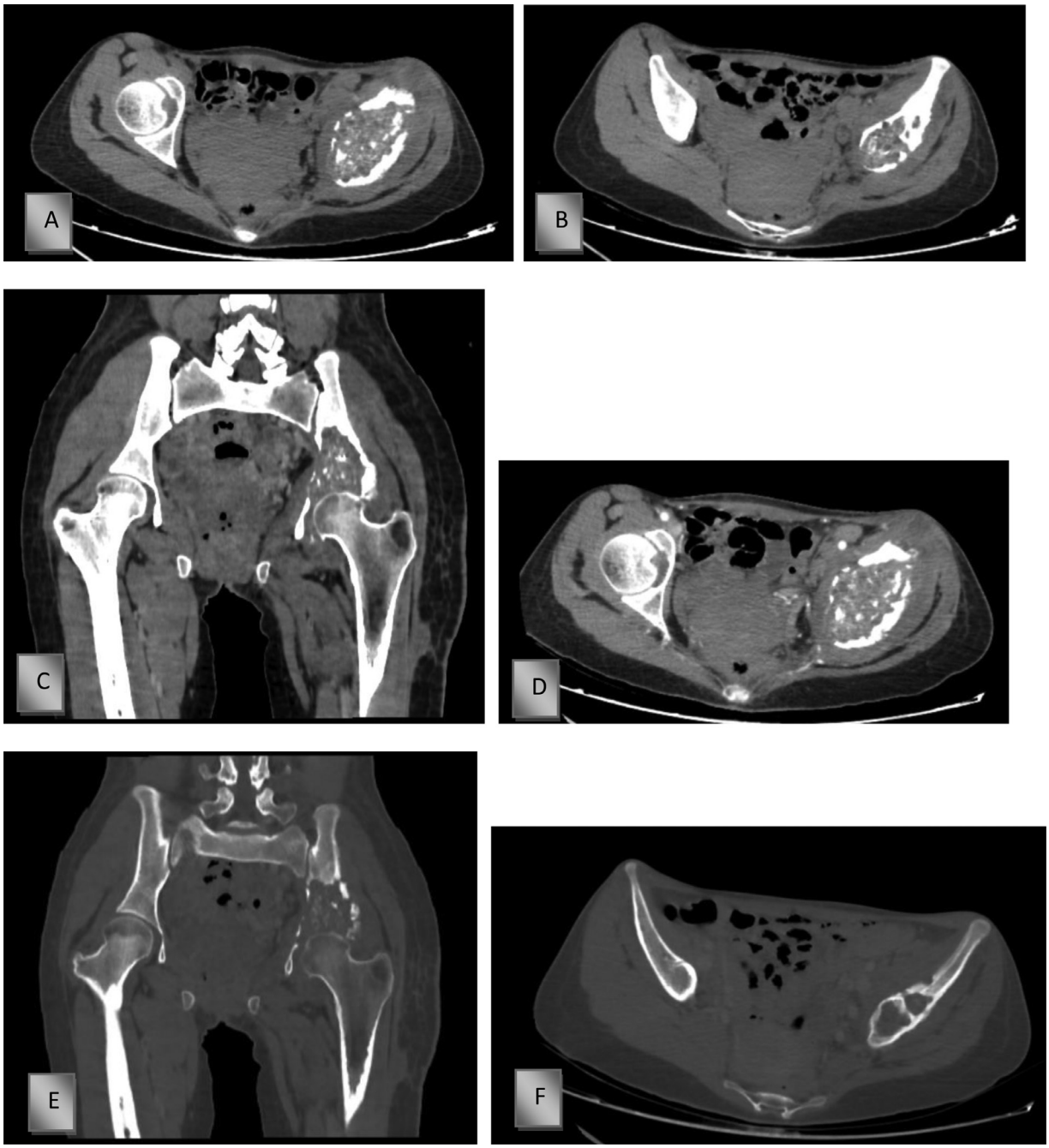

Fig. 2 – Pelvic CT scan. (A, B) Axial section without contrast-enhanced. (C, D) Axial section and coronal reformatted scan with contrast-enhanced. (E, F) Bone window in coronal reformatted scan and axial section, revealing an expansile lytic process of the acetabulum to the lytic ilium, multiloculated with a honeycomb appearance, containing calcifications, enhanced at the periphery after injection of the contracted product. CT also showed multiple cortical destruction foci and loss of bony texture in the left iliac bone with a periosteal reaction and infiltration of the adjacent muscles. The sacroiliac joint, the coxofemoral line, and the femoral head were respected. Individualization of a subcutaneous collection communicating with the lesional process.

was no evidence of atypical cells that could have suggested malignancy. The hydatid serology performed after the biopsy diagnosis was positive. Abdominal ultrasonography and radiographs of the chest showed no evidence of hepatic or pulmonary involvement. In light of those findings the the diagnosis of a primary osseous hydatid disease was made.

Given the high rates of recurrence and the extension of lesions, the patient was treated with medical treatment based

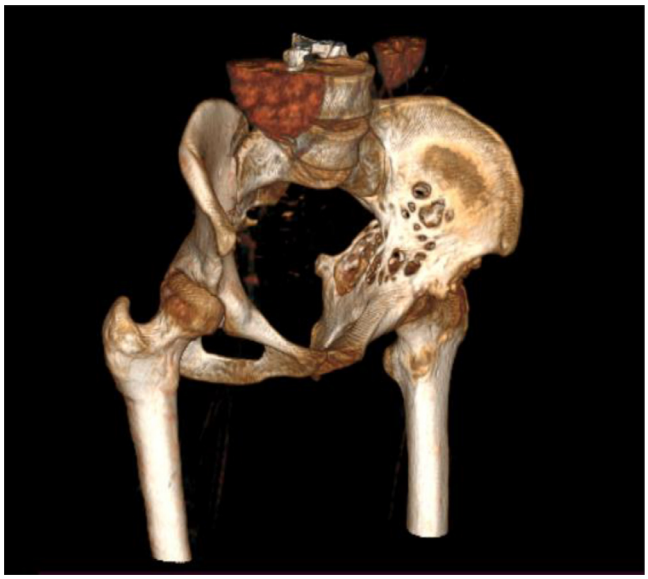

Fig. 3 – Ilium bone destruction on 3 dimensional CT scan reconstruction.

on albendazole, following the WHO protocol: 10-15 mg/kg/d, 4 weeks out of 6, for a duration of 6 months, and a CT scan re-evaluation is scheduled at the end of the treatment to discuss surgery.

Discussion

Osseous hydatid disease is an involvement of the bone by an anthropozoonosis caused by the larval form of the tapeworm Echinococcus granulosus [6], and first described by Didlou in 1706 [6,7]. Osseous hydatid disease is rare, even in endemic areas, such as Morocco, contributing only 0.5%-2.5% of all hydatid cysts [8], and among them, 30%-50% involves the vertebrae, 15% involves the pelvis, and less frequently involves long bones [9,10]. Men and women seem equally affected [8]. Bone contamination is mainly hematogenous [11], but a secondary bone invasion from primary soft tissue involvement is possible. Hydatid osteopathy is infiltrative, diffuse, slow, and progressive with numerous microvesicles without encystation of the parasite [2,12]. The importance of spongy tissues in the pelvis constitutes favorable ground for the spread of hydatid lesions [4]. The slow growth of numerous vesicles results in the replacement by the parasites of osseous tissue between

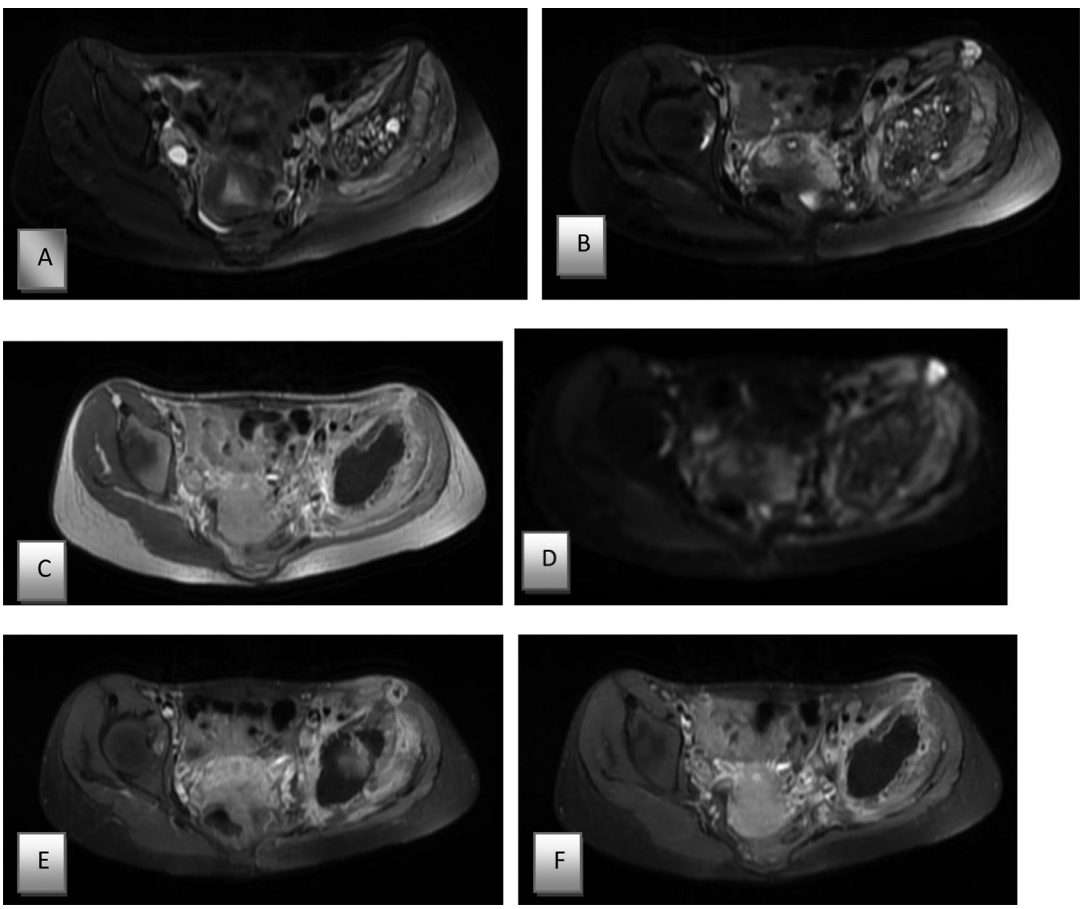

Fig. 4 – MRI images (A, B) Axial T2 weighted images. (C) Axial T1 weighted Image. (D) Axial diffusion B1000. (E, F) Postcontrast T1 FAT SAT images), revealed multiple small vesicles in the left iliac wing and cystic lesions in the supraacetabular soft tissues.

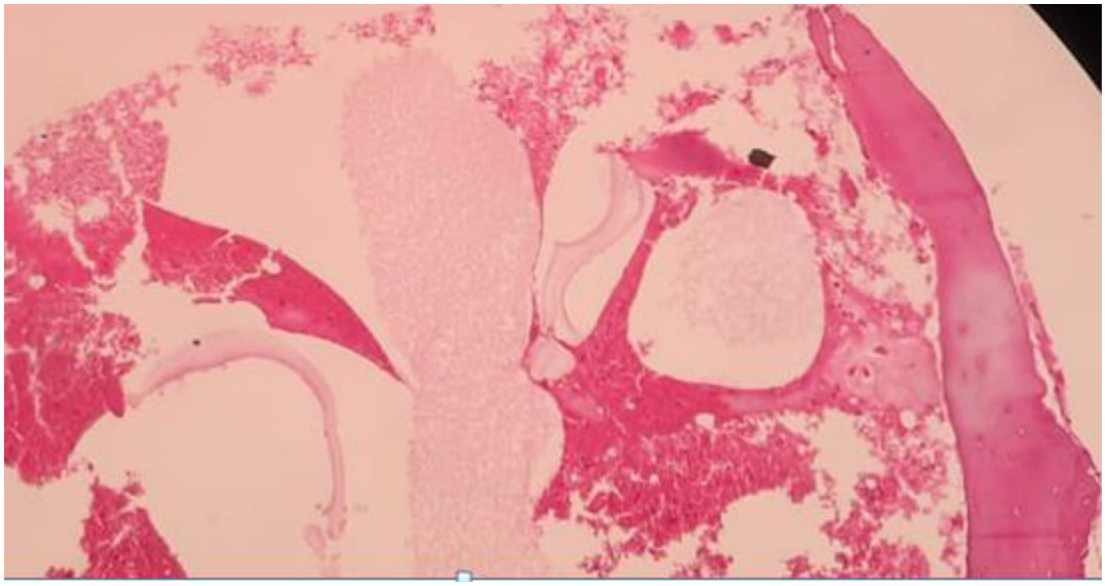

Fig. 5 – Lamellar anhistic membrane of hydatid cyst (immunohistochemical stain H&E, x100).

the trabeculae. The parasites ultimately reach and destroy the cortex, with the subsequent spread of the disease to surrounding tissues [13–16].

Clinically, hydatid disease of bone remains asymptomatic over a long period, and its diagnosis is usually made when lesions have become extensive [5]. The pain is often well-tolerated by the patient, evolving intermittently and becoming strong only at a late stage [4]. Tumefaction about the abscess may have been seen, such as in our case. It is a cold migratory abscess without a fistula that may, however, infect, simulating a hot abscess [4]. These 2 symptoms are rarely alarming, and hydatidosis of bone is usually detected in the stage of complications [4], which may include pathological fracture, mostly seen with long bone hydatidosis, neural deficit secondary to vertebral involvement, infection and fistulization of the abscess [5,4]. The notion of contagion and a history of hydatid disease are the signs that orient the diagnosis. The biological tests are also contributive. Hypereosinophilia is inconstant since it is only present in 25% of cases and it is not specific [17] Serology has an important role in the diagnosis. The results should be interpreted with caution: a positive result with a significant titer suggests hydatidosis. On the other hand, a negative result does not exclude it. Negative serology was reported in 10%-15% of cases of hydatid cysts [18]. Serology is also used to monitor the efficiency of treatment: the serum antibody level rises then decreases to disappear in 12-24 months. The persistence of a high level of antibodies beyond this period or the re-ascension of this level after negativation suggests a recurrence of the disease [19].

Radiollocaly, plain radiography usually reveals single or multiple expansile osteolytic lesions containing trabaculae in the classic honeycomb pattern with cortical thinning [16,17,20]. Osteosclerosis is typically seen in the advanced stages of the disease, whereas periosteal reaction is typically a sign of a pathological fracture in most cases. If the cyst has breached the cortex, an adjacent soft tissue mass with calcification may be seen [16,21]. The initial lesion in the pelvis is classically situated on the ilium, but primary affection of the ischium or the superior ramus of the pubis may be observed. The CT scan is still the best method for diagnosis, appraisal of the extension, and post-therapy follow-up of osseous hydatidosis [4,22]. It shows well-defined single or multiple cystic lesions that may cause cortical thinning without contrast enhancement or calcification [16]. It may also show pathologic fracture, cortical destruction, and soft tissue extension [16]. CT helps when studying the bone and extraosseous extension, especially the ossifluent abscess, by evaluating its size and expansion [4]. Magnetic resonance is particularly useful in depicting the extent of the disease, especially in soft tissue and the spine [16,17], as it gives a full image of the vertebral axis of the spinal canal [23]. It is also helpful in evaluating recurrences [16]. Ultrasonography helps appraise the extension of the ossifluent abscess by homogeneous cystic views corresponding to it [4]. Abdominal ultrasonography along with chest radiography is the basic investigation for detecting hydatid cysts in the viscera, especially the liver and lung, which may co-exist with osseous cysts. Herrera et al. [23] reported that 45% of their patients had liver or lung cysts [16].

The differential diagnosis includes infectious pathologies, especially tuberculosis, and tumors [4,16], depending on the location, other differential diagnoses are discussed, at the pelvic localization, chronic osteomyelitis of the ilium, hip tuberculosis, sacroiliac tuberculosis, a tumor with giant cells, osteosarcoma, or a brown tumor must be eliminated. Spinal involvement is a differential diagnosis problem with Potts' disease, Aneurysmal cysts, vertebral plasmocytoma, Kahler's disease, chordoma, and spinal metastases must also be eliminated. In the case of long bone involvement, the differential diagnosis includes chronic osteomyelitis, fibrous dysplasia of bone, osteosarcoma, and cystic benign lesions, but the pres-

ence of a periosteal reaction, osteocondensation, calcification, and distinct delimitation of the lesions enables the diagnosis of osseous hydatidosis to be excluded [4]. As a result, it is challenging to make a preoperative diagnosis of skeletal hydatid disease and an accurate diagnosis is generally made during surgery and confirmed histologically [24]. The macroscopic view shows diffuse infiltration by small vesicles of different sizes without cysts. There is no clear delimitation between healthy and pathologic tissues, which explains the difficulty of surgical resection. We sometimes find sequestered tissues or infection of the hydatic cyst [4]. Histologic lesions are characterized by the presence of hydatid fragments in bone trabeculae (germinal layer, scolex) but without adventitia. Bone lesions are nonspecific, indicating destructive osteitis. Bacterial infection of hydatid lesions produces the aspect of condensing osteitis. Often there is a discrepancy between the macroscopic and microscopic boundaries of the parasitic infestation, which explains the frequent recurrences after apparently wide excisions [4].

The management of osseous hydatidosis is highly challenging from diagnosis to treatment, due to its long clinical latency, delay in diagnosis, the absence of radiological specificity, lack of standardized treatment regimens, and high recurrence [4,13,25]. The treatment is medical and surgical. Medical treatment aims to reduce cyst size and sterilize the cysts' contents before surgery, and to treat the small cysts that had not been detected after surgery. Albendazole appears to have higher digestive absorption than the other benzimidazoles, and it has strong clinical and biological tolerance, even at high doses for a long period. According to WHO recommendations, albendazole is administered in four to six 4-week courses before and after surgery at a daily dose of 10-15 mg/kg divided into 2 postprandial doses, spaced 2 weeks apart. Regular follow-up of hepatic function is recommended [17] surgical treatment aims for complete excision of the hydatid lesions [17]. About the pelvis, the radical resection of the affected bone is often met with technical difficulties, and total eradication of the parasitic infestation is usually impossible. Implanting a prosthesis carries a risk due to the septic potential of hydatid disease [16]. Osseous hydatidosis has significant morbidity and mortality rates [4]. The severity of its prognosis has given it the label of "white cancer" [17].

Conclusions

Osseous hydatid disease is a rare entity, even in endemic areas, such as Morocco. Because of the long clinical latency and the lack of specific clinical symptoms, as well as the absence of radiological specificity, the diagnosis is often established late. Medical imaging allows establishing of a precise lesion assessment to plan a large surgical resection. The hydatid disease of bone should be considered a differential diagnosis in osteolytic lesions, though it is rare. The most significant measures to minimize the severe damage that this parasitosis causes continue to be early diagnosis and hygiene education in endemic regions. Medical treatment appears to be an additional contribution whose effectiveness has not yet been proven. Pelvic locations are particularly challenging to treat surgically.

Author's contributions

All authors contributed to this work. All authors have read and approved the final version of the manuscript.

Patient consent

Written informed consent for publication was obtained from patient.

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