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Case report

Case report: Hydatid cyst of bone in the distal femur with untypical presentation treated surgically

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ABSTRACT

Introduction: Osseous form is a rare manifestation of hydatid cystic disease, and it is easily misdiagnosed for other musculoskeletal infections and tumor-like lesions as it presents in a solitary form with subtle and unspecific radiological features and clinical symptoms.

Case report: Our study is a case report of a young female patient with hydatid cyst in the distal femur, who was treated successfully with surgical resection, conducted in 2021 and the case was followed up for 8 months.

Discussion: Several cases had been reported of osseous hydatidosis commonly in the axial skeleton and often diagnosed lately, stressing the importance of keeping unfamiliar diagnosis in mind when dealing with untypical lesions of bone.

Conclusion: Hydatid osseous is a rare parasitic disease that has a non-specific clinical and radiological presentation, differentiating it from similar looking tumor-like lesions and early diagnosis is not simple. However it is crucial as it requires precise treatment plan to prevent recurrence, and long term follow up is important.

1. Introduction

musculoskeletal infections are a major cause of morbidity, and they tend to be challenging to treat, osseous infection happens when the bone is invaded with a disease-causing organism which might be pyogenic, non-pyogenic or rarely parasitic [1], of the latter Echinococcus granuloses relatively common in Mediterranean countries is a tapeworm that lay eggs that could invade the humane intense through its eggs released oncosphere larva ultimately penetrating its way into the liver, lungs and other internal organs forming a hydatid cyst [2], Osseous hydatidosis is considered one of the rarest manifestation of hydatid disease due to the rigid structure of bone trabeculae that does not allow for formation of adventitia [3], however we present to you a case of isolated hydatid cyst in the distal femur with unfamiliar presentation. This case report has been reported in accordance with SCARE 2020 standards [4].

2. Case report

A 35 years old female with no comorbidities and an unremarkable medical history came to us complaining of a 3 years old mild progressive mechanical knee pain, it was fairly relieved on analgesics, physical exam revealed mild local tenderness along the distal bony aspect of the distal femur, full range of motion, neurovascular examination was normal, and no other abnormalities in the musculoskeletal system or any signs on knee derangements, x-rays AP and lateral views were obtained showing an ill-defined non-destructive lytic lesion in the distal femur (Fig. 1).

Differential diagnosis was simple cyst, aneurismal bone cyst or chondromyxoid fibroma so MRI was ordered and it showed a well-defined heterogenies non-aggressive lesion in the distal femur with low signal on t1 weighted images and mixed signal on t2 and PD images measuring 7 × 4 × 3 cm (Fig. 2). while a simple bone cyst doesn't correlate with the age of the patient and ABCs usually have a characteristic MRI appearance -multiple fluid lines- and tend to be more expansile and aggressive, chondromyxoid fibroma might be the most probable diagnosis but it is usually more demarcated from the adjacent bone with a scalloped and a sclerotic rim. Malignant bone lesions don't have that subtle hideous presentation, and of course infection the great mimicker of lesions is a possibility, however there is no local nor systemic symptoms of infection and the labs were within normal limits.

Due to the untypical radiological and clinical appearance the decision of biopsy was made to optimize the planning of management which came in as benign bone tissue with intervening fibrous tissue and addition four giant cells suggesting aneurismal bone cyst after correlation with the radiological examination (Fig. 3).

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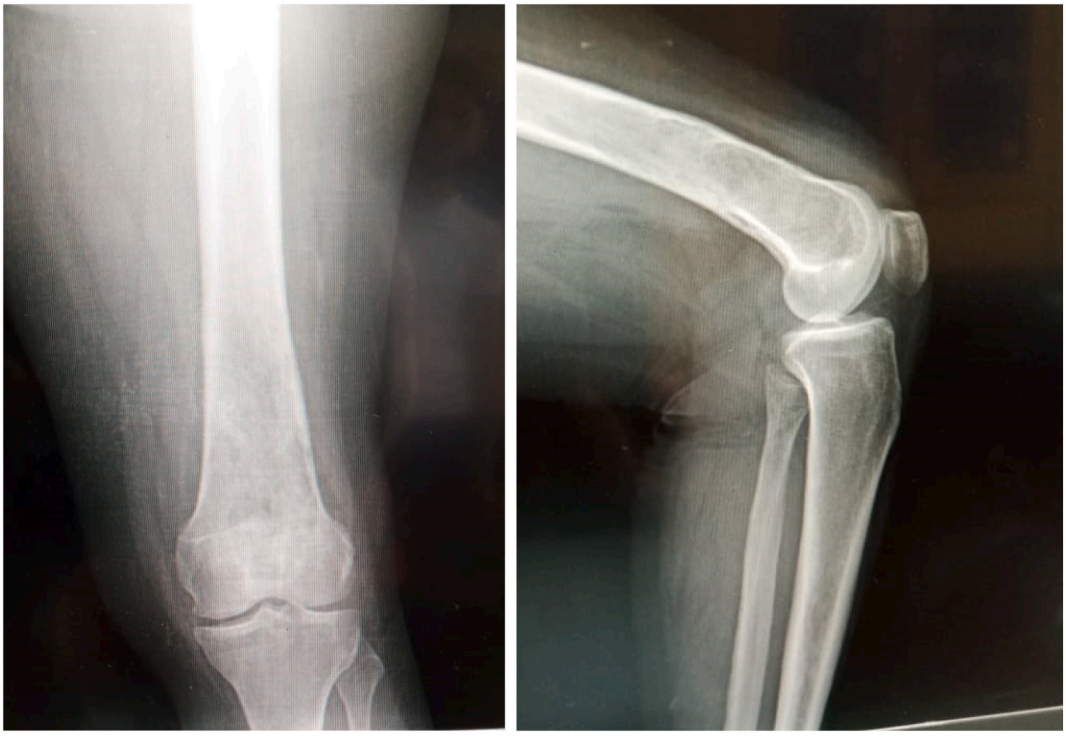

Fig. 1. Plain x-ray of the distal femur showing non-destructive lytic lesion.

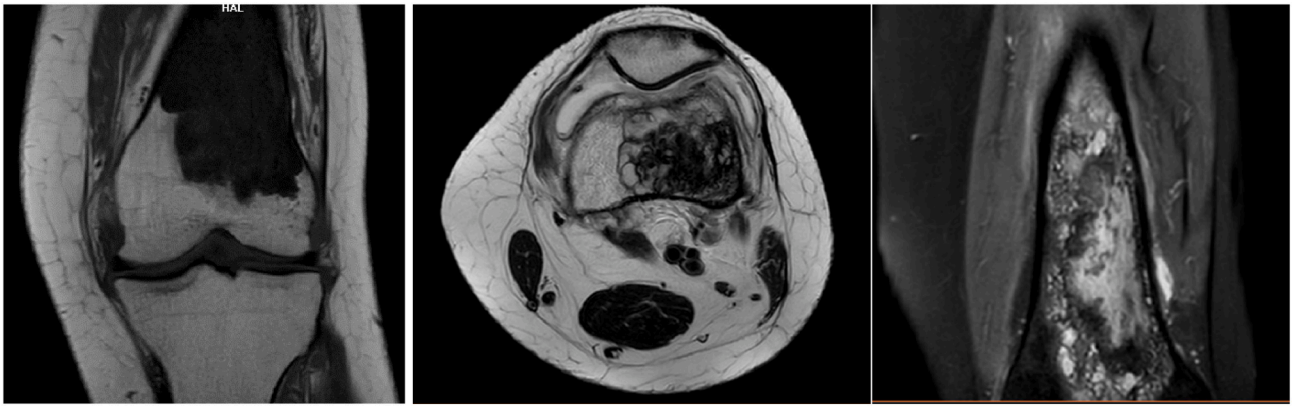

Fig. 2. MRI images showing the lesion from left to right: T1 weighted image, T2 weighted image, PD weighted image.

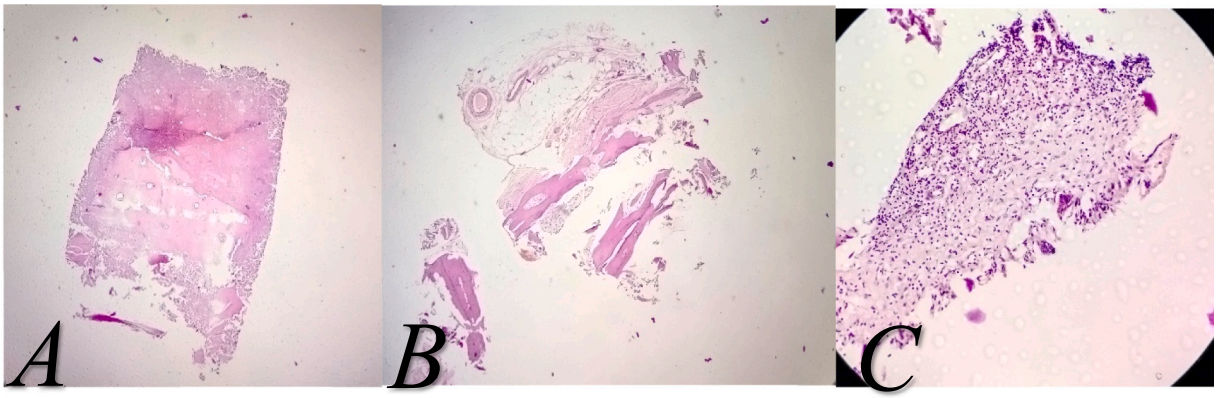

Fig. 3. A, B: benign osseous and fibroadipose tissues. C: edematous fibrous tissue with mild inflammatory infiltrate and multinucleate giant cells. From the CT guided biopsy.

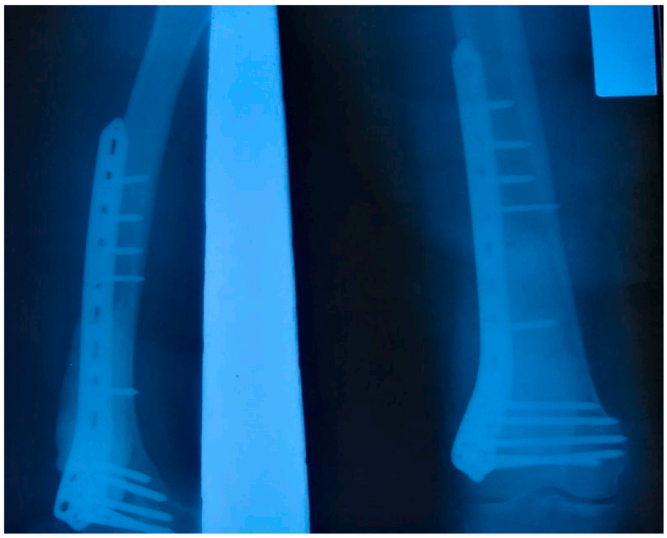

Fig. 4. A follow-up x-ray 6 months post-surgery.

So we went for surgery with extensive curettage and reconstruction of the gap left behind, however the visual look of the lesion intra-operatively was not correlating with ABC so the decision of marginal resection was made and the gap was filled with bone cement and the bone was prophylactically fixed with an anatomical distal femur plate (Fig. 4). The resected tissue was send again for histological study and came in as fragments of bony and inflamed fibrotic tissue showing granulation tissue formation and fragments of pink acellular laminated material suggestive of germinal layer of the hydatid cyst (Fig. 5).

The drain was withdrawn the next day of surgery and the patient was discharged into physiotherapy program with partial weightbearing and prophylactic anticoagulant medicine based on her weight and it was referred to a general surgeon consultant where full body CT scan was made and it came negative with no other lesions identified. Infections control consultant was made and the patient was given a course of Albendazole for 3 months.

3. Discussion

When it comes to pathological lesions in orthopedics it is very important that proper and precise methodology of management is done, as there are malignant, benign and tumor-like lesions that might have similar presentations but require a totally different plan of management and if done wrong it could lead to catastrophic results. While it is obvious that a comprehensive approach should be taken, one must also have high suspicion of unfamiliar diagnoses, hydatid osseous is a rare manifestation of Echinococcosis that has an unspecific clinical and radiological picture, cystic echinococcosis mainly prevail in the third world countries in the Middle East, central Asia, western China and around the Mediterranean [5]. The visceral form of the disease is the most common form, and the liver is the most common site for cyst formation where it might occur in any soft tissue organ [2].whereas osseous tissue infestation is not characteristic due to the special structure of bone and the periosteum, many theories is thought to be explaining osseous invasion starting by mechanical expensive process leading to ischemic osteitis and eventually destruction of bony tissue without an inflammatory process [6,7], while the osseous form incidence is reported to be only 0.5–4 % and rarely in the appendicular skeleton [8,9]. As the blood-borne scolex invade bone pericyst formation does not occur, rather aggressive proliferation in the line of least resistance bone canals, thus the normal osseous tissue between trabeculae is slowly destructed and replaced due to the growth of multiple vesicles [10,11]. This slow pathological process along with the common subtle symptoms like: unspecific pain, swelling and at worse a pathological fracture makes early diagnosis a challenge, or even commonly misdiagnosed and mistreated. The radiological appearance is also nonspecific as on plain x-rays the disease present as a cystic or irregular lytic lesion, on CT scan local ovoid destruction of bone might be found with rarely a specific mark as “double layered arcuate calcification” which happens due to calcification of the capsule wall [11,12], MRI is definitely more reliable as it might shows a multilocular appearance and the pericystic wall and the relation of the lesion with the surrounding tissues but still rather not highly specific [10,13]. Serological tests are very important but they might be negative as the cyst is aging, calcified or dead [14]. There is no effective conservative treatment for hydatid osseous, high recurrence rate is expected without aggressive surgical resection -with healthy normal margins-, and extended adjuvant therapy with Albendazole is

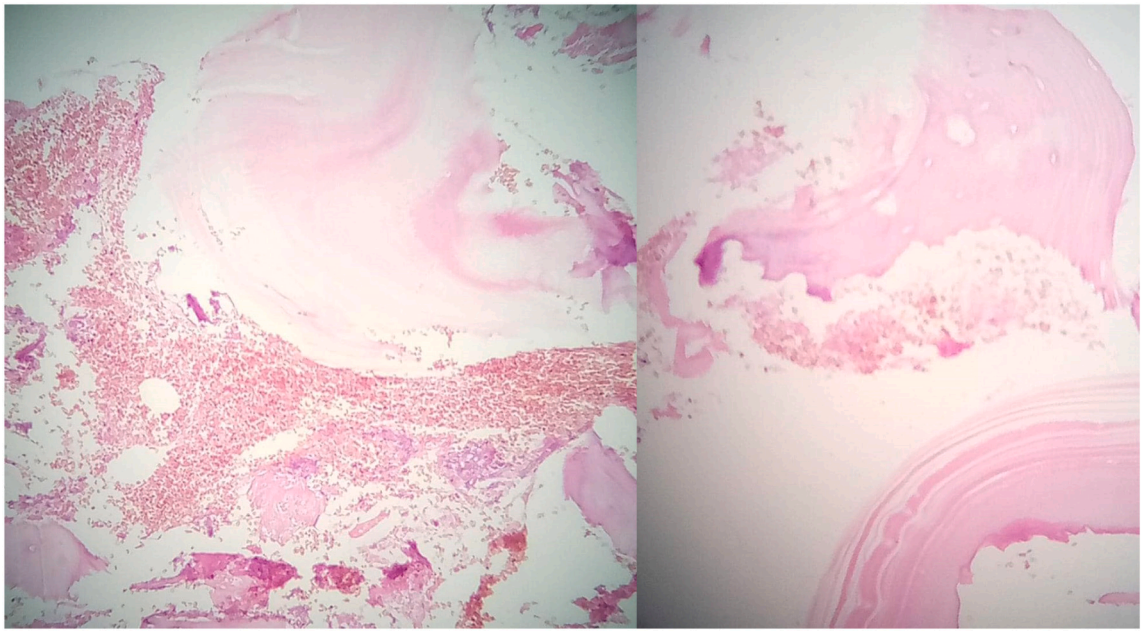

Fig. 5. Histological study showing pink laminated gelatinous membrane suggesting hydated cyst within bone.

recommended [9,15]. The high recurrence rates stress the importance of accurate diagnosis and the following plan of management, clinical suspicion when the patient is living or have visited one of the endemic countries is mandatory. In our case the diagnosis was established intra-operatively and was confirmed with the histopathological study post-operatively. Extended resection of the lesion was done as the surgeon had operated a similar case before, and the decision to reconstruct the bone by filling the gap with PMMA as the literature supports its usage to decrease the risk of recurrence [16]. Our patient is well-collaborated and we have been following her for 8 months with no clinical nor radiographical signs of recurrence. Several cases have been reported in the literature -mainly in the axial skeleton- about osseous hydatidosis [9,17,18], and they all share as in our case the difficulty of early diagnosis and the unfamiliar presentation, which emphasize on keeping rare diseases such as hydatid cyst of bone in differential diagnosis when dealing with uncharacteristic bony lesions and specially a cystic lesion in a patient living in an endemic region.

4. Conclusion

In conclusion, hydatid osseous is a rare parasitic disease that has a non-specific clinical and radiological presentation, differentiating it from similar looking tumor-like lesions and early diagnosis is not simple, however it is crucial as it requires precise treatment plan to prevent recurrence, and long term follow up is important.

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Ethical approval

This study is exempt from ethical approval in our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

CRediT authorship contribution statement

Hakam Hekmat Alasaad M.D.: conceptualization, investigation, data curation, writing, editing and reviewing.

Doried Diri M.D.: investigation, data curation, writing, editing and reviewing.

Firas Hwaij M.D.: investigation, data curation, editing and reviewing.

Guarantor.

Hakam Hekmat Alasaad M.D.

Declaration of competing interest

The author has no conflicts to disclose.

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