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Case Report

Hydatid cyst of the humerus presenting as a suspicious lesion: A rare case report and review of literature ☆

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ABSTRACT

Hydatidosis is a parasitic disease caused by the tapeworm Echinococcus. Echinococcus Granulosus is the most common cause of hydatid disease in humans. Bone involvement is rare, accounting for only 0.9% to 2.5% of all cases. We report the case of an 8-year-old child admitted with right arm pain, revealing a hydatid cyst on the humerus. Lesion assessment revealed a hydatid cyst of the humerus with extension to the adjacent soft tissues. The surgical procedure involved the excision of the cyst along with drainage. In this case report, we review the epidemiological, clinical, and paraclinical aspects of the disease, as well as the treatment modalities. Bone hydatid disease is infiltrative, diffuse, slow, and progressive, making diagnosis late, and compromising the quality of treatment.

Introduction

Hydatid cysts are a parasitic disease endemic in certain countries, particularly in South America and Australia, but also around the Mediterranean basin and in Central Europe [1]. It frequently affects the liver and lungs. Skeletal involvement is rare; its frequency varies from 1% to 2%. This localization is characterized by a long clinical latency [1].

Imaging plays a significant role in making the diagnosis. Computed tomography (CT) and magnetic resonance imaging (MRI) clarify the location, size, extent, and severity of the lesion [2].

We report a rare case of hydatid cyst of the humerus, illustrating the contribution of imaging to the diagnosis and therapeutic difficulty of this condition.

Case report

An 8-year-old patient, right-handed, living in an endemic area, has been under follow up for pleural hydatid cysts since 2022,

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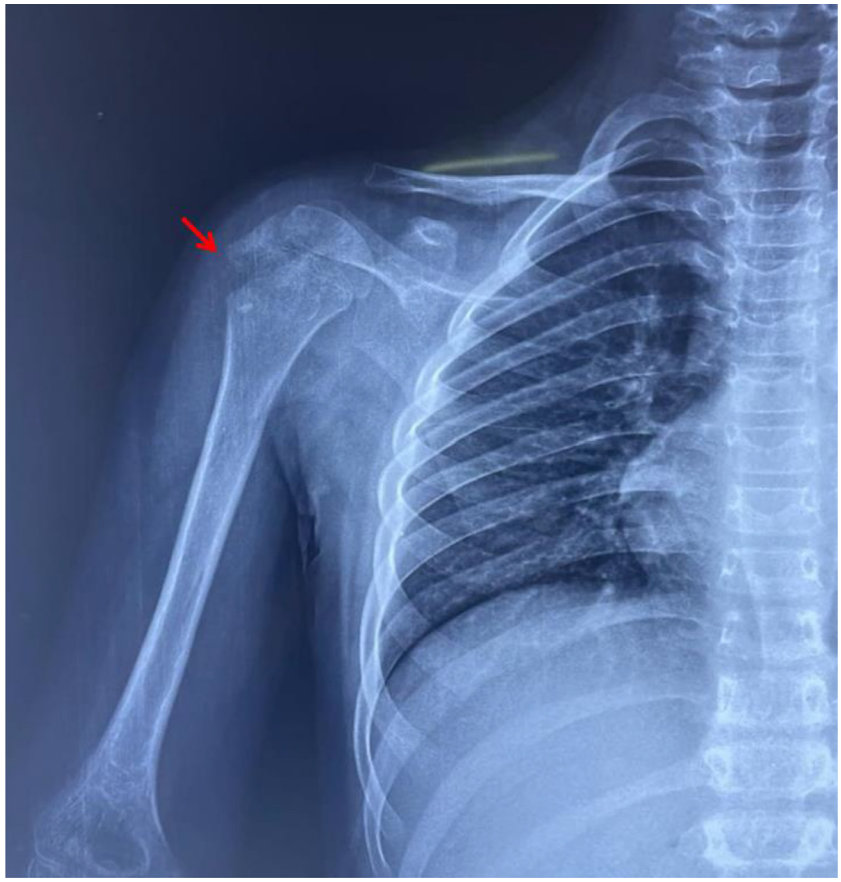

Fig. 1 – X-rays of the right arm showing a lytic metaphyseal lesion of the upper end of the right humerus without calcification or marginal sclerosis, breaking and blowing the cortex Lodwick 1c (red arrow).

and was put on albendazole 15 mg/kg during 15 days, followed by a 15-day therapeutic window, for a total duration of 6 months. The patient attended an orthopedic consultation for pain in his right arm for 6 months. The pain located in the upper third of the right arm had moderate intensity and was nonradiating. The patient had no fever and no weight loss.

Examination of the musculoskeletal system revealed a right shoulder with no swelling or skin changes compared with the left side. There was no pain on palpation, and active mobility was preserved. The right elbow was mobile and painless. Vascular and nerve examinations of the right upper limb and the rest of the musculoskeletal system were without abnormalities.

During the pulmonary examination, no significant cough has been observed in the patient. However, the patient reported having some occasional nonproductive coughing. On the pulmonary auscultation, the following observations were made: a decreased vesicular breath sounds in the left side and the respiratory sounds are less audible compared to the right lung. Thoracic percussion found a dullness on percussion in the left lung.

The rest of the clinical examination was normal.

X-rays of the right arm revealed a lytic metaphyseal lesion of the upper end of the right humerus without calcification or marginal sclerosis, breaking and blowing the cortex classified as Lodwick 1c. No periosteal reaction was observed (Fig. 1). Based on clinical and X-rays data, the lesion was believed most likely to represent an infectious process or a neoplastic lesion.

CT scan of the right arm revealed a liquid-dense lesion of the upper end of the humerus, containing thin partitions, breaking the cortex and extending into the soft tissues. The lesion was also coming into contact with the deltoid muscle, from which it remains separated by a thin wall with no signs of invasion (Fig. 2). A chest CT scan was also performed showing multiple uniloculated left subpleural cysts, some of which are contiguous (Fig. 3) .

An MRI was not performed, as the X-ray and CT scan were sufficient to characterize the lesion given the patient's clinical context.

Biology reported a CRP at 150 mg/L (<6 mg/L), white blood cells at 20,000 cells/m^3 (5000-11000 cells/m^3), ASAT at 33 IU/L, ALAT at 29 IU/L (<35 IU/L) and Alkaline phosphatase at 233 IU/L (86-315 IU/L). We completed the assessment with a hydatid serology, which was positive.

The patient's case was presented to a multidisciplinary meeting. Since the patient had a single bony hydatid cyst, of small size, with no complications such as a pathological fracture or extensive destruction of the adjacent bone, the surgical team opted for a hydatid cyst curettage associated with 1-

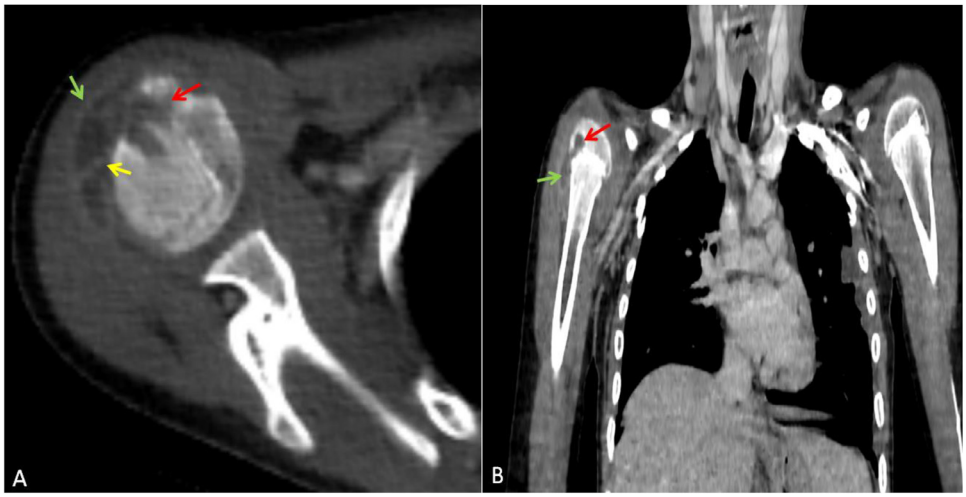

Fig. 2 – CT scan in axial (A) and coronal (B) section showing a liquid-dense lesion of the upper end of the humerus (red arrows), containing thin partitions (yellow arrow), breaking the cortical bone and extending to the adjacent soft tissues (green arrow).

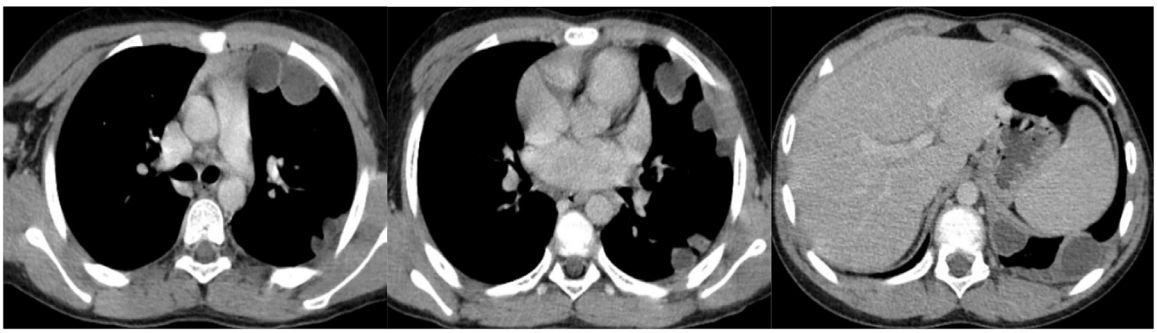

Fig. 3 – Axial chest CT showing uniloculated left subpleural cystic formations.

month treatment of albendazole (15 mg/kg/day orally) before the operation and after the operation for 6 months as adjuvant therapy.

The curettage was performed by a pediatric orthopedic surgeon using a direct approach under general anesthesia. Cortical destruction was evident. The orthopedic surgeon performed a curettage aspiration, evacuation of the collection, and extensive cleansing with hypertonic saline solution. During the curettage procedure, a proligerous membrane was discovered within the cystic cavity (Fig. 4).

After curettage, the wound was cleaned with H2O2 solution and hypertonic saline to prevent cyst recurrence. The intervention was performed with no incidents and the patient was discharged after 1 week with oral albendazole treatment. There was a complete resolution of clinical symptoms and his arm mobility is improving gradually.

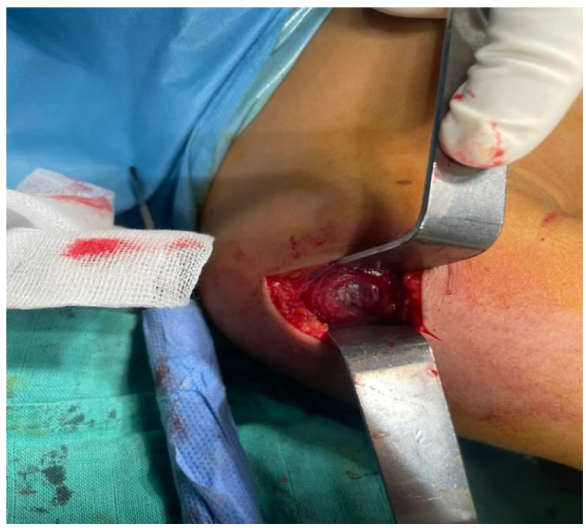

Fig. 4 – Intraoperative image showing the proligeral membrane of the hydatid cyst.

Discussion

Hydatidosis is a parasitic disease caused by the larval form of Echinoccocus granulosus, a zoonosis that usually affects dogs

Table 1 – Clinical characteristics and imaging findings relating humeral hydatid cysts: published cases vs our case.

Year of publication	Authors	Number of cases	Clinical findings	Imaging findings		
				X-rays	CT	MRI
2011	Nourbakhsh et al	01	- Pain - Limited motion	- Cystic lesion in the proximal part of the humerus in the presence of nonunion		
2000	Markonis et al	01	- Tenderness - Moderate powerloss - Limited abduction	- Distortion of the axis, - Minimal cortical thinning areas, - Radiolucent areas of the humeral shaft, - No calcification of soft tissues.	- Destruction of the trabecular bone of the humeral head, - Polycystic appearance of the bone marrow - Large collection between medial and lateral head of the triceps muscle.	- Multiloculated lesion into the humeral lumen, - Cystic lesion of the soft tissues of the upper arm.
2019	Patino et al	01	- Pain - Limited motion of shoulder and elbow.	- Oblique fracture in the distal third of the diaphysis, - Osteolytic and multiloculated images.	Not performed	- Expansion to soft tissues, - Cortical thinning.
Our case			- Pain - Normal range of motion	- Lytic metaphyseal lesion of the upper end of the humerus Lodwick 1c, - Cortical destruction.	- Liquid-dense lesion, - thin partitions, - Cortical destruction, - Extension to adjacent soft tissues.	Not perfomed

and sheep, but can also affect humans. It occurs on all continents except Antarctica but is more common in the Mediterranean basin, the Middle East, Central Asia, Western China, the Russian Federation, and North and West Africa [3]. In Morocco, it is a major public health problem, with an incidence of 4.5%.

Hydatidosis can affect any organ, but the most common sites are the liver and lung. These 2 organs are the most affected because of their role as capillary filters [4], but other organs can also be affected, such as lymph nodes, the greater omentum, the central nervous system, the skin, and the retroperitoneum [5]. Involvement of the musculoskeletal system is rare compared with other localizations, accounting for 0.9%-2.5% [1]. The spine is affected in 50-60% of cases, followed by the femur (10%), ribs (8%), tibia and skull [6].

Involvement of the humerus is rare. In the literature, we found only 3 cases of humeral localization of hydatid disease. In 2011, Nourbakhsh et al described a case presenting with pseudarthrosis of the left humerus on a hydatid cyst [7]. In 2000, Markonis et al. described a case of a 17-year-old boy with a hydatid cyst of the left humeral shaft [8]. In 2019, Patino et al. described a case of a 24-year-old patient who presented with a pathological fracture of the humeral shaft [9] (Table 1).

Most cases arise from hematogenous dissemination of hepatic cysts, and their metastasis rate is around 1.3% [10,11]. Systemic circulation of metacestodes and invasion of bone may depend on local blood supply and slow metacestode growth. Larval expansion and growth are limited by dense bone tissue and narrow trabecular spaces, so the onset of disease is often sub-clinical, and difficult to detect at an early stage [12].

Clinical manifestations are variable and nonspecific. Involvement of the long bones is often discovered by the appearance of pain, swelling, pathological fracture, or functional impotence. Bone echinococcosis has no specific clinical signs. After several years of latent evolution, pain is often the first revealing sign. The pain is moderate and intermittent, synonymous with a long-standing condition. Local swelling may or may not be associated with pain, and generally reflects extension into the soft tissues. Spontaneous fractures or fractures resulting from minor trauma are a frequent mode of revelation. The rest of the clinical examination is unremarkable, and the patient's general condition is often preserved [13].

There are no specific radiographic signs. In the early stages, marginal sclerosis and periosteal reaction are not evident [4]. In the advanced stage, a lytic lesion with or without marginal sclerosis may be found [14]. Cortical rupture associated with calcifications may also be found, but periosteal involvement is not evident except in the case of pathological fracture [13].

CT is more accurate than standard radiography, particularly in the more frequent localizations (spine and pelvis) [14]. In other patients, bone involvement may have a typical appearance: round or oval lesions containing fluid, with sharp, thin margins and no contrast within the lesion. The lesion may resemble an abscess or take on a pseudotumoral form, mimicking malignant bone tumors. CT scans also allow better assessment of extension to neighboring tissues [14].

MRI remains the best imaging method. It allows us to determine the exact location of the disease and its extension in the soft tissues. Cysts appear in homogeneous T1 hypointense and homogeneous T2 hypeintense. However, MRI must be coupled with CT scanning, as the latter enables a better study of the bone and cyst calcifications [4].

Imaging does not provide any criteria of diagnostic certainty. If the chest X-ray or liver ultrasound reveals other sites of involvement, the diagnosis becomes easy. On the other

hand, in the absence of other localizations, clinical manifestations, or positive hydatid serology, it becomes impossible to make a differential diagnosis and even exclude a malignant lesion, as was the case in our observation [14]. In these patients, only histological studies can provide a definitive diagnosis.

In terms of treatment, the behavior of bone lesions is similar to that of a cancerous tumor. The disease has a poor prognosis since it can be hard to treat. Complete excision of the afflicted region is the sole treatment option for bone involvement. To prevent recurrence, the resection must be complete with a large healthy margin. However, surgical intervention is also linked to significant mortality, morbidity, and recurrence rates of between 70% and 80% [7].

When surgery is not an option, mebendazole or albendazole may be administered as an isolated medicinal treatment. Medical treatment can be added to surgery, to prevent or at least reduce the risk of dissemination. This adjuvant therapy controls the disease locally, stops its systemic spread, and prevents recurrence. It is taken both preoperatively if the diagnosis is known and postoperatively. Chemotherapy results are debatable, and there is currently little information on how well these medications treat bone infections [7].

Conclusion

In conclusion, hydatid disease is common, while bone echinococcosis is not as common. Misdiagnosis of bone echinococcosis is frequent. MRI is the method of choice for evaluation, preoperative planning of the surgical approach, and correct diagnosis of such lesions.

Patient consent

Written informed consent for the publication of this case report was obtained from the parent of the patient.

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