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CASE REPORT

Multifocal hydatidosis with extensive involvement of the sacrum and the femoral head

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Key Clinical Message:

Hydatidosis is a parasitic infection caused by the larval form of "Echinococcus granulosis." Bone localization is rare even in endemic areas. We report an unusual case of an extensive hydatidosis of the right sacrum and femur with muscle involvement. **Abstract:**

We report the case of a man, with a history of visceral hydatidosis, who developed after 14 years an extensive hydatidosis of the sacrum and femur with muscle involvement. The treatment of visceral hydatidosis was chemotherapy with albendazole for a long term. Conservative surgical treatment is considered for bone locations.

KEYWORDS

bone, echinococcosis, femur, hydatid disease, magnetic resonance imaging, sacrum

JEL CLASSIFICATION

Haematology, Oncology, Infectious Diseases, Orthopaedics

1 | INTRODUCTION

Hydatidosis is a parasitic infection caused by the larval form of "Echinococcus granulosis." The most frequent localization is hepatic and pulmonary. Bone localization is rare even in endemic areas. Bone hydatidosis remains latent for a long time as the clinic is poor and the evolution is progressive and slow, and its treatment combines chemotherapy and surgery.^{1,2}

2 | CASE PRESENTATION

A 29-year-old patient, with a history of operation of hydatid cysts of the liver and the kidney in 2008, oral

anthelmintic albendazole was prescribed for a long term. For the past 20 days, he complained from a right lumbosacral pain mimicking a truncated sciatica.

The clinical examination showed a swelling in the right buttock. The right hip was painful on mobilization but not limited. There was no sign of spine disorder. Neurological examination was normal.

Laboratory results revealed normal blood cell counts, normal sedimentation rate, and normal serum C-reactive protein.

The plain radiograph of the pelvis showed lytic areas on the right sacral wing, with invasion of the right sacral foramina and a lacunar lesion on the right femoral head (Figure 1).

Magnetic resonance imaging (MRI) of the pelvis revealed a multivesicular cystic lesion on the right sacral

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial-NoDerivs License, which permits use and distribution in any medium, provided the original work is properly cited, the use is non-commercial and no modifications or adaptations are made. © 2023 The Authors. *Clinical Case Reports* published by John Wiley & Sons Ltd. wing (the largest being of $8 \text{ cm} \times 7 \text{ cm} \times 1 \text{ cm}$ size) with invasion of the right sacroiliac joint (Figure 2), the lesion passed the large ischial notch and invades the piriformis and small gluteal muscles. There was a second cystic lesion on the femoral head with the same characteristics (Figure 3).

A computed tomography (CT) scan of the chest and abdomen was performed to detect other localizations, it showed in addition to the bone location (Figure 4); hepatic hydatid localization with partially calcified hypodense formations, left mid-renal, and adrenal hydatid localizations.

The treatment of hydatdosis of the liver and the kidney was chemotherapy for a long term.

Conservative surgical treatment of the sacrum cystic lesion was performed with puncture, aspiration, and abundant washing with hypertonic saline serum. The postoperative course was simple.

The treatment of the hydatid cyst of the hip will be performed, in a second step, a drilling, excision of the lesion and then filling with an autograft.

3 | DISCUSSION

The hydatid cyst is caused by a parasite "echinococcus granulosis" which lives in its adult state in the bowel of the dog. Human infestation is accidental and occurs only through the digestive tract by ingestion of eggs.^{1,3} The two filters that the egg must pass after its intestinal passage are the liver and the lung, which explains the frequency of these two localizations. Bone lesions contribute only 0.5%–2.5% of hydatid cysts; vertebral localization is the most frequent (44%), followed by the pelvis (16%), the femur, and the tibia.² Bone contamination is essentially hematogenous. The characteristic of the bone hydatid cyst is the absence of adventitia, which allows the cyst to enlarge and become polycystic.¹ It can spread through the large ischial notch to the buttocks⁴ and can compress the sciatic nerve, as in our case.

The clinic remains latent for a long time and when the signs appear, it is dominated by pain and swelling,⁵ and we can find also radicular irritation signs.^{2,6}

Plain radiography is essential for the diagnosis. It shows osteolysis images without periosteal reaction, realizing a honeycomb appearance.⁷

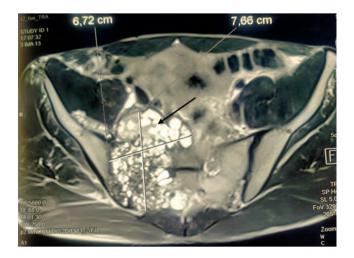


FIGURE 2 Axial section of MRI T2 sequence showing a mass occupying the right sacrum measuring 80×70 mm in axial and 100 mm in height, this mass is in hypersignal T2 with a multivesicular aspect (Black arrow).



FIGURE 1 Standard radiograph of the pelvis showing lysis of the cortical bone of the right sacral wing with invasion of the right sacral foramina (black arrow). There is also a lacunar lesion in the right hip (Asterix).



FIGURE 3 Axial section of MRI T2 sequence showing the cyst in hypersignal with a multivesicular aspect in the right femoral head measuring 17×12 mm.

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FIGURE 4 CT scan transverse section of the pelvis showing a hypodense formation in the right sacrum (black arrow) with bone destruction as well as sequestrated bone pieces (Asterix).

Ultrasonography is useful for detecting abscesses in soft tissues and presents with the chest X-ray, the first extension assessment to be requested.⁷

The interest of the CT scan is to make an overall assessment of the extent of the lesions. It shows hypodense images that are not enhanced after injection of contrast product. MRI typically shows a multivesicular cystic lesion, and its interest is in the early detection of recurrences after surgery.²

The treatment often combines chemotherapy and surgery.

Medical treatment: as oral anthelmintic albendazole is used mainly in patients with inoperative lesions, it reduces the size of the cysts and cure the unnoticed cysts before and after surgery, but it is effectiveness is controversial.⁸

Surgical treatment must be radical with carcinologic excision; removal of the hydatid lesions with a wide margin of resection of the surrounding healthy tissue which is microscopically infiltrated. The defect generated is filled by a reconstruction. When the lesion is too extensive invading the muscles and the nerves, puncture aspiration and cleaning with a hypertonic saline solution is performed,⁷ but with a high local recurrence rate of 70%–80%, although cases of success have been published.¹ Concerning the surgical treatment of the hip hydatid cyst, resection of the affected segment is performed when the lesion is limited, and if it is diffuse, amputation or disarticulation is the only solution.¹

4 | CONCLUSIONS

Bone hydatidosis is rare even in endemic areas, and its slow evolution is responsible of a delay in the diagnosis. Medical imagery allows an assessment of the extent of the lesions. The principle of surgical treatment is a carcinologic excision, but if the excision is partial, the relapse rate will be very important. Prevention is the best solution regarding difficulties in the diagnosis and the treatment.

AUTHOR CONTRIBUTIONS

Afef Feki: Conceptualization; writing – original draft. Cyrine Abid: Conceptualization; data curation. Samar Ben Djmeaa: Formal analysis. Zouhour Gassara: Formal analysis. Ezzeddine Mariam: Data curation. Mohamed Hedi Kallel: Visualization. Akrout Rim: Validation. Hela Fourati: Data curation; resources. Sofien Baklouti: Supervision; validation; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declared no conflict of interest with respect to the authorship and/or publication of this article.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no datasets were generated or analyzed during this study.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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