Case report

Sciatica and femoral head osteonecrosis complicating a gluteal hydatid cyst: A case report

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ABSTRACT

Introduction and importance: Hydatid disease is an endemic echinococcal infestation in Tunisia. Only 4% of hydatid cysts are located in the muscle. The gluteal localization is a very rare entity. The present case illustrates a gluteal hydatid cyst resulting in sciatica and osteonecrosis of the femoral head, an exceptional entity.

Case presentation: A 49-year-old man of urban origin, with free medical history, presented to our outpatient clinic with left sciatica and mechanical hip pain, that have been ongoing for 2 years. Pelvic radiograph showed femoral head osteonecrosis and lytic lesions of the ischium. MRI scan identified an extra-articular vesicular cyst in the left gluteal region involving the gluteus maximus. It suggested the diagnosis of musculo-skeletal echinococcosis. The patient underwent surgery and intraoperative findings showed a gluteal vesicular cyst with direct compression of the sciatic nerve, it also revealed femoral head necrosis. The cyst was drained and the pericyst excised. Total hip arthroplasty was postponed and the patient received a course of anthelmintic chemotherapy.

Clinical discussion: Muscular involvement of hydatid cysts is rare and seen only in 4% of cases. Neurological complications from sciatic nerve compression can present as sciatica, with a principal differential diagnosis of nerve root compression. Aseptic femoral head osteonecrosis, an exceptional complication of an extra-articular gluteal hydatid cyst, can result from three mechanisms: a direct mechanical compression, an ischemic process due to vessel obstruction or a cellular process mediated by osteoclasts. Treatment is based on surgical excision combined with anthelmintic chemotherapy, the latter reduces the number of live cysts and the risk of recurrence.

Conclusion: Although gluteal cysts are extremely rare with different clinical symptoms, surgery associated with anthelmintic treatment has a good functional outcome.

1. Introduction

Hydatid disease is an endemic echinococcal infestation in Tunisia. Only 4% of hydatid cysts are located in the muscle [1]. The gluteal localization is a very rare entity. The present case illustrates a gluteal hydatid cyst resulting in sciatica and osteonecrosis of the femoral head, an exceptional entity that was managed in an academic institution.

2. Case presentation

A 49-year-old man of urban origin, with free medical history, presented to our outpatient clinic with left sciatica and mechanical hip pain that have been ongoing for 2 years. Physical examination revealed an antalgic gait, a swollen and painful left hip, especially when attempting rotation, and left sciatic palsy with a neurological score of 3/5. Pelvic radiographs showed osteolytic lesions of the ischium, and femoral head osteonecrosis aspect (Fig. 1). Magnetic resonance imaging identified a gluteal cyst of vesicular content, lodged in the gluteus maximus (Fig. 2). It suggested the diagnosis of Musculo-skeletal echinococcosis with sciatic nerve compression. The extension assessment did not show any other hydatid localization. Surgical exploration, using a posterior approach (Fig. 3) revealed a...
25-cm extra-articular cyst, lying deep to gluteus maximus with direct compression of the sciatic nerve (Figs. 4, 5, 6, 7).

The cyst was drained, the pericyst excised (Fig. 8) and the surgical field was washed with hypertonic saline to reduce recurrence risk. We performed sciatic nerve neurolysis and the femoral head and neck were removed (Fig. 9). Total hip arthroplasty was postponed. The procedure was performed by a senior orthopaedic surgeon. The patient received a course of anthelmintic chemotherapy. The vesicles and pericyst were sent for histopathologic examination. A rehabilitation protocol has been established in a rehabilitation center. At the last follow-up of 02 years, the patient walks without limping. The neurological evaluation of the left sciatic nerve is 4/5. No hydatid recurrence at the last MRI. The patient returned to work and was satisfied with the clinical outcomes.

3. Discussion

Hydatid muscle involvement is rare and seen in 0.5–4% of cases [1]. This is due to the high lactic acid level in muscle tissue, unfavorable for parasite survival, and the muscular contractions that prevent fixation of larvae to the tissue [2].

The affected muscular sites are most commonly the diaphragm, pectoralis major, biceps brachii, sartorius and psoas [3]. The gluteal localization is extremely rare and our brief review of literature revealed less than 20 reported cases.

The gluteal hydatid cyst usually presents as a slow-growing painful swelling without systemic symptoms. Our case, is exceptional, describing a hydatid cyst in the gluteal region responsible not only for sciatic compression, but also for an osteonecrosis of femoral head.

Sciatic injury has been rarely described, we cite Ajay and al. who reported a case of foot drop by cystic erosion of the roof of the S2 foramen [4]. We reviewed three more report cases describing sciatica and

Fig. 1. Pelvic radiograph showing osteolytic lesions of the ischium and osteonecrosis of the femoral head.

Fig. 2. MRI study showing a cyst lodged in the gluteus maximus containing multiple vesicles.

Fig. 3. Surgical posterior approach and the clinical aspect of the gluteal mass.
foot drop caused by a gluteal hydatid cyst, by Shannak et al. [5], Ergin et al. [6] and Çetintas et al. [7].

But no review was found relating osteonecrosis of the femoral head complicating a gluteal hydatid cyst which makes the unicity of our case.

Zlitni et al. proposed three mechanisms to account for the bony destruction: a mechanical process whereby the expanding cyst compresses the surrounding soft tissues; an ischemic process due to obstruction of the nutrient blood vessels; and a cellular process which is mediated by the proliferating osteoclasts around the compressed bony tissue [8]. In our case, we believe that these 3 mechanisms have played a role in the femoral head osteonecrosis.

Treatment of the musculoskeletal hydatid disease, is based on surgical excision combined with anthelmintic chemotherapy, the latter reduces the number of live cysts and the risk of recurrence [9].

4. Conclusion

Muscle involvement in hydatid disease accounts for 4% of cases and the gluteal localization is extremely rare. The hydatid cyst can cause symptoms from pressure effect, obstruction or allergic reactions.

Adequate treatment consists of surgical resection and anthelmintic chemotherapy, in order to prevent recurrences.

Statement

This work has been reported in line with the SCARE criteria [10].

This work has been reported in line with the SCARE 2020 criteria [11].

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Author contribution
Mehdi Bellil: Writing the paper
Meriem souissi: Study concept
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Mohamed Zaibi: Study concept
Walid Balti: Supervision
Mohamed Ben Salah: Supervision

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Declaration of competing interest
The authors have no financial, consultative, institutional, and other relationships that might lead to bias or conflict of interest. The authors declare that no funding was received for this study.

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Fig. 8. The pericyst was completely excised.

Fig. 9. The femoral head aspect of an aseptic osteonecrosis.