Enfermedades Infecciosas y Microbiología Clínica 41 2023 638–639

Enfermedades Infecciosas y Microbiología Clínica

www.elsevier.es/eimc

Diagnosis at first sight

Gluteal tumor of unusual etiology

Tumoración en glúteo de etiología inusual

Cristina Gaona-Álvarez[a,*], Carmen González-Velasco[a], Juan Carlos Luna-Berral[b], Miguel Fajardo-Olivares[a]

[a] Servicio de Microbiología, Hospital Universitario de Badajoz, Badajoz, Spain
[b] Servicio de Cirugía Ortopédica y Traumatología, Hospital Universitario de Badajoz, Badajoz, Spain

Case report

This was the case of a 52-year-old woman of Moroccan origin, who has lived in Spain for 21 years, although she continues to travel occasionally to Morocco. The patient consulted for a lump in her left buttock in July 2015. In this first consultation, an ultrasound revealed an extensive complex hypoechoic collection of 10 cm in the long-axis, multiloculated and located in the deep upper third of the left gluteus maximus muscle. Without ruling out other aetiologies, the possibility of evolving loculated haematoma was raised due to repeated intramuscular injections in the affected area for the treatment of migraine.

In March 2022, the patient consulted again due to pain and an increase in the size of the lesion. The physical examination revealed an indurated tumour with a maximum diameter of 15 cm, with limited movement and fixed to underlying tissues. A contrast-enhanced computed tomography (CT) imaging study of the thorax, abdomen and pelvis was performed with intravenous contrast, revealing a tumour affecting the left gluteus maximus, indenting the piriformis and gluteus medius muscles. The lesion, ostensibly encapsulated, had a multilocular appearance with numerous internal septa (Fig. 1). Hypodense areas with fluid density values suggested possible myxoid liposarcoma.

The study was completed with magnetic resonance imaging of the left hip, whereby an intramuscular multicystic lobulated mass with well-defined internal structures was observed. In view of these findings, surgical exeresis was performed.

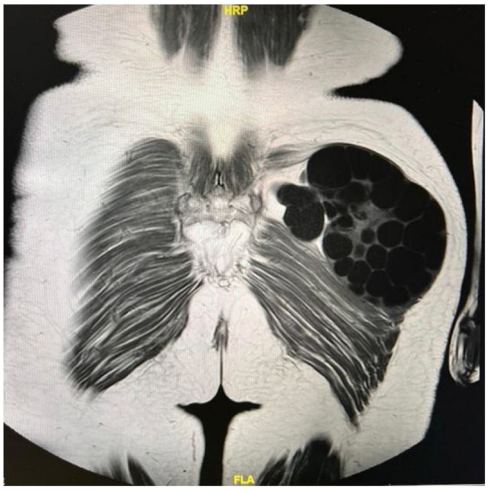

Fig. 1. CT of the thorax, abdomen and pelvis. Multilocular lesion in the buttock.

Clinical course and diagnosis

Once the cyst had been resected by cystopericystectomy, an incision in the adventitia provided access to the multiple vesicles contained inside the cavity (Fig. 2). Several of these vesicles were sent for microbiological study.

At the gross level, the sample received by Microbiology was a vesicle with a gelatinous appearance and a whitish coating. The

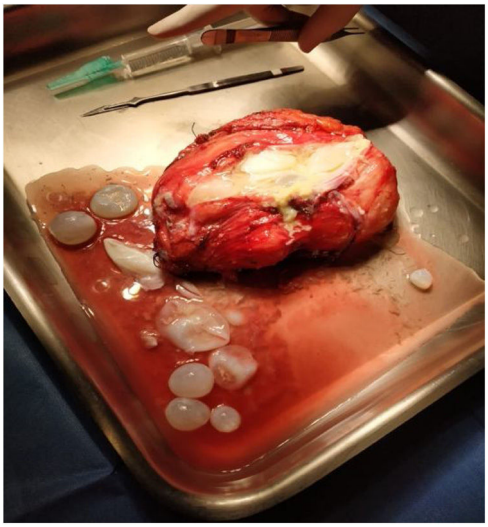

Fig. 2. Cyst with internal vesicles.

DOI of original article: https://doi.org/10.1016/j.eimc.2023.03.005
* Corresponding author.
E-mail address: cristina.gaona@salud-juntaex.es (C. Gaona-Álvarez).

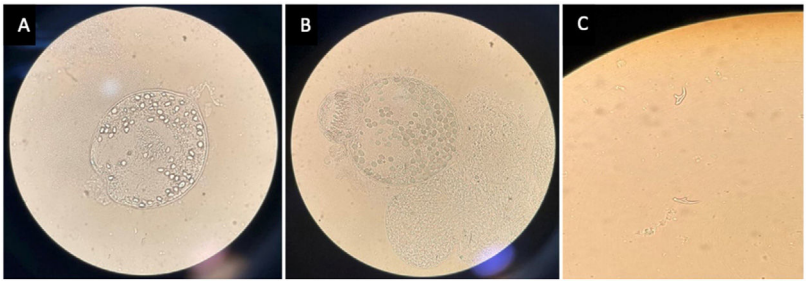

Fig. 3. Microscopic view (40×) of protoscolices A) and B) and free hooks C) in vesicle fluid (magnified image).

contents of the cyst were aspirated with a syringe and hypodermic needle, and a clear and transparent aqueous fluid was obtained. After centrifuging for 10 min at 2000 rpm, the supernatant was discarded. Microscopic observation of the sediment showed protoscolices and free hooks (Fig. 3).

These structures provided microbiological confirmation of *Echinococcus* spp. One day after surgery, determination of serum antibodies against *Echinococcus granulosus (E. granulosus)* by indirect haemagglutination assay (IHA) was negative. Subsequently, PCR for *E. granulosus* and *Echinococcus multilocularis* on hydatid fluid performed at the Centro Nacional de Microbiología [National Centre for Microbiology] confirmed the identification of *E. granulosus*.

According to the World Health Organization's classification of hydatid cysts, the characteristics of the lesion observed by imaging tests allowed it to be classified as a CE2-type active cystic lesion.

The results of the CT of the thorax, abdomen and pelvis ruled out the presence of other cysts in these areas.

After surgery, oral albendazole (400 mg/12 h) was prescribed. The patient was discharged eight days later and scheduled for clinical and therapeutic follow-up by the Infectious Disease Unit. Treatment with albendazole was maintained for three months, with favourable clinical outcome.

Hydatid disease or cystic echinococcosis is a zoonosis caused by *E. granulosus*. Humans can be an intermediate host for the parasite. Most patients (40%–80%) have a single cystic lesion in a single organ,[1] with the liver (69%–75%) and lung (17%–22%) most commonly affected, while location in the muscle is rare (2%).[2]

In this case, the imaging tests ruled out liver and lung cysts, leading to the diagnosis of primary muscle hydatid disease.

Although the pathogenesis of extrahepatic and extrapulmonary primary cysts is unclear, haematogenous or lymphatic spread have been proposed as possible mechanisms.[3]

The increase in size of the cyst in our patient from July 2015 to March 2022 was 5 cm, a prolonged clinical course that makes it impossible to establish the origin and time of acquisition of the infection.

IHA has variable sensitivity depending on the location of the cyst; it is positive in 80% of hepatic hydatid disease and negative in most of the muscular forms, limiting its diagnostic value.[3] Although the cause of this variability is unknown, hydatid disease with negative serology could be associated with encystment of the larval form producing insufficient stimulation of antibody production, resulting in undetectable serum levels.[1] In our case, prior serology was not performed, but the negative IHA the day after surgery demonstrates that it would not have been useful to support the suspected diagnosis of hydatid disease.

References

1. Wen H, Vuitton L, Tuxun T, Li J, Vuitton DA, Zhang W, et al. Echinococcosis: advances in the 21st century. Clin Microbiol Rev. 2019;32(2).
2. Kern P, Menezes da Silva A, Akhan O, Müllhaupt B, Vizcaychipi KA, Budke C, et al. The Echinococcoses: diagnosis, clinical management and burden of disease. In: Thompson RCA, Deplazes P, Lymbery AJ, editors. Advances in parasitology. 2017. p. 259–369.
3. Okus A, Sevinc B, Ay S, Karahan O, Eryilmaz MA, Er C. Relation between serology and grow-up time in atypically localized hydatic cysts. Turk J Parasitol. 2014;37:257–61.