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CASE REPORT

Osseous hydatidosis of the proximal femur: a rare diagnosis in revision total hip arthroplasty

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Abstract

Musculoskeletal hydatidosis is a rare but severe disease in central Europe. This case report presents the incidental finding of an osseous hydatidosis after cementless revision total hip arthroplasty in a patient without a preoperative history of hydatidosis or any clinical symptoms. Revision total hip arthroplasty had been necessary due to a septic osteonecrosis of the femoral head 2 years after osteosynthesis of a traumatic proximal femur fracture with a sliding hip screw. The positive sample was taken out of the greater trochanter in the area of the possible former entry point for the lag screw, which was macroscopic inconspicuous. Sero-analysis could afterwards confirm the suspected diagnosis. Postoperative chemotherapy with albendazole was performed for 6 months. A full-body MRI did not reveal any further cysts. This case demonstrates a possible impact of migration on the expected pathogens in revision arthroplasty. This demonstrates that in revision arthroplasty, an infection with this parasite also has to be taken into account, if the patients come from an area endemic for hydatidosis.

Keywords Osseous hydatidosis · Femur · Prothesis · Total hip arthroplasty · Revision

Introduction

Human echinococcosis is a parasitosis caused by larval forms of the Echinococcus tapeworms. *E. granulosus* and *E. multilocularis* are of medical importance, commonly reported to be responsible for the cystic and alveolar echinococcosis in humans. The endemicity of this zoonosis varies between different countries, with a focus on rural areas. *E. granulosus*, the dog tapeworm, is widespread in countries where large herds of sheep are controlled by sheepdogs. In endemic areas such as regions of South America, the Mediterranean littoral, Eastern Europe, the Near and Middle East, East Africa, Central Asia, China and Russia, it remains a relatively common disease with an annual incidence from < 1 to 200 per 100,000 [1–3]. According to Steinmetz et al., the geographical distribution of echinococcosis has not changed since 1930 [4]. Infection occurs after the ingestion of eggs excreted with the faeces of an infected host (e.g. dogs). The eggs hatches and release larvae (oncospheres) which penetrate the intestinal wall and, having gained access to the portal venous circulation, spread throughout the body, infiltrating various organ systems. An involvement of the liver (approx. 70%) and the lung (15–30%) is common, whereas the musculoskeletal system is only affected in 0.5–2% of all cases. Within the organs the larvae develop into hydatid cysts that can survive for months or even years. Active hydatid cysts usually contain sterile hydatid fluid and a large number of protoscolices, and can reach sizes of 20 cm or more [1, 2, 5]. The most common site of osseous hydatidosis still remains the spine (50% of the cases) followed by the pelvis and the hip (14% of the cases) [2, 4].

In this paper, we report the rare case of an osseous and soft-tissue cystic echinococcosis as a postoperative incidental finding after cementless revision total hip arthroplasty (RTHA).

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Case presentation

An Algerian 18-year-old male patient was referred by a peripheral hospital to our department in August 2017 after a girdlestone resection arthroplasty of his right hip.

The patient had suffered a traumatic proximal femur fracture of the right femur after a motorcycle accident 2 years earlier in Algeria, which was treated with a sliding hip screw in Algeria. Two years later and after migration to Germany over the Eastern Mediterranean route he was complaining of persistent thigh pain in the former fractured hip. Clinical examination, laboratory tests and radiologic analysis in the treating peripheral hospital showed a septic osteonecrosis of the femoral head with cutting-out of the implanted sliding hip screw. The patient was immediately scheduled for open revision. Unfortunately, the X-ray images from Algeria and those taken pre-operatively (septic revision) are not available to us.

According to the foreign surgical report, the remnants of the femoral head were surrounded by pus and had to be resected, followed by a radical debridement that included reaming of the femoral shaft and implantation of antibiotic chains. Several microbiological samples were taken from the surrounding soft-tissue and the femoral head. The microbiological samples revealed *Propionibacterium acnes*. All in all, the intraoperative macroscopic findings in the foreign hospital confirmed the preoperative suspected diagnosis of a septic osteonecrosis of the femoral head with cutting-out of the infected sliding hip screw. Due to a postoperative wound healing disturbance, a secondary wound revision had to be carried out. In this operation the antibiotic chains were removed. Microbiological samples taken during this operation yielded multi-susceptible coagulase-negative Staphylococci (*S. epidermidis*). The antibiotic therapy was changed from cefazolin to penicillin. After 17 days the i.v. antibiotic therapy was switched to oral chemotherapy (clindamycin) and continued for 4 more weeks. Unfortunately, no histological samples were taken from the excised tissues or the femoral head of the first and second operation. After the second operation, a CT scan was carried out to plan a possible RTHA. The CT did not reveal any signs of osteomyelitis or cystic bone lesions in the remaining bone stock.

The patient presented in our department 2 months after completion of his antibiotic therapy. At that time, the patient showed no clinical or laboratory (CRP and WBC) signs of a persisting infection. A radiograph of the pelvis showed a distinct femoral and acetabular osseous bone stock loss, which could be classified as a type 2c (acetabular defect) and type 2 (femoral defect) defect according to the Paprosky classification [6]. Furthermore, an old subtrochanteric fracture line with callus formation and dislocation ad axim and ad latus of the proximal femur was visible. Cystic bone lesions were not described. The preoperative X-ray is shown in Fig. 1. We decided on an operative revision of the right hip with radical debridement. Without macroscopic signs for a persisting infection, we further planned to implant a modular RTHA while preserving the greater trochanter and the gluteal muscles in order to achieve the best possible, postoperative functional results. The patient did not have any other illnesses, nor did he take any permanent medication, or report any allergies. His family history was unremarkable for infectious diseases or musculoskeletal disorders.

Intraoperatively, an enlarged lateral approach to hip, pelvis and proximal femur was used. The debridement showed no macroscopic sign of a persisting infection. Representative samples for the microbiological and pathohistological examination were taken of the excised tissue as described previously [7]. After debridement, we implanted an uncemented modular RTHA using modular implants (Peter Brehm, Weisendorf, Germany) and used cerclages to refix the greater trochanter with the gluteal muscles at the stem neck (used implants: MRP hip stem and MRS-Titan Comfort Cup with a 14-mm dorsocranial modular augment). A strut graft was used to cover a remaining lateral corticalis defect, which we held for the possible former entry point of the lag screw, and was afterwards secured with cerclages. Homolog spongiosa was used to augment a medial acetabular osseous bone stock loss in impaction bone grafting technique. Postoperatively the patient did well, without complicated wound healing. Postoperative X-rays are shown in Figs. 2 and 3.

Microbiological samples did not detect any (bacterial) signs of a persisting infection. Pathohistological examination of the intraoperative samples, however, revealed small

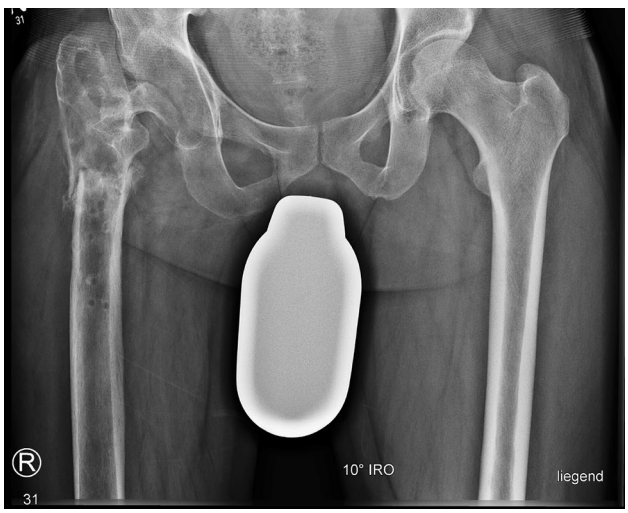

Fig. 1 Preoperative AP radiograph showing the situation before arthroplasty

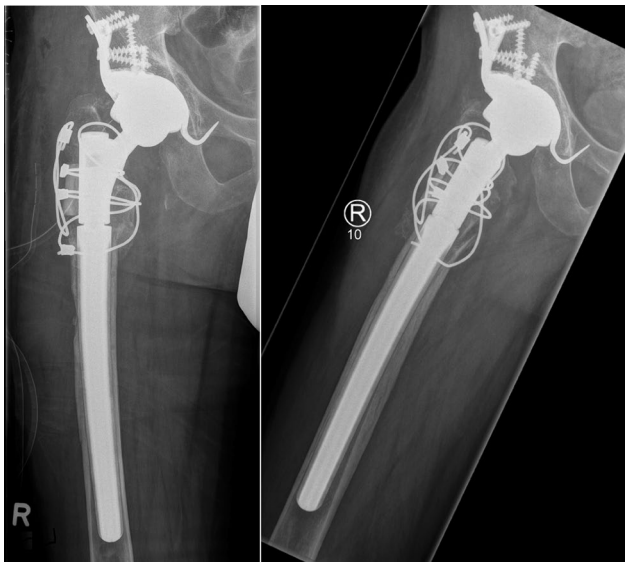

Fig. 2 Direct postoperative AP and axial radiograph after RTHA

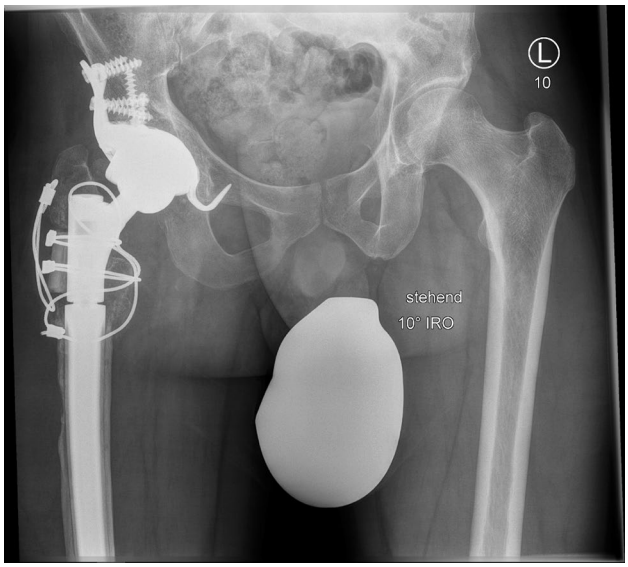

Fig. 3 6 weeks postoperatively, the AP radiograph shows the RTHR with beginning osseous integration especially of the modular acetabular cup

Echinococcus cysts (daughter cysts) in one out of seven samples (see Fig. 4). This sample was taken out of the greater trochanter in the area of the possible former entry point for the lag screw of the sliding hip screw. Intraoperatively this region was lined with granulation membranes, which did not show any macroscopic sign of a persisting infection, e.g. pus or cystic structures. We regarded this membrane as a remnant of a former insufficient debridement. The serological examination confirmed an infection with *E. granulosus* (ELISA and Western blot).

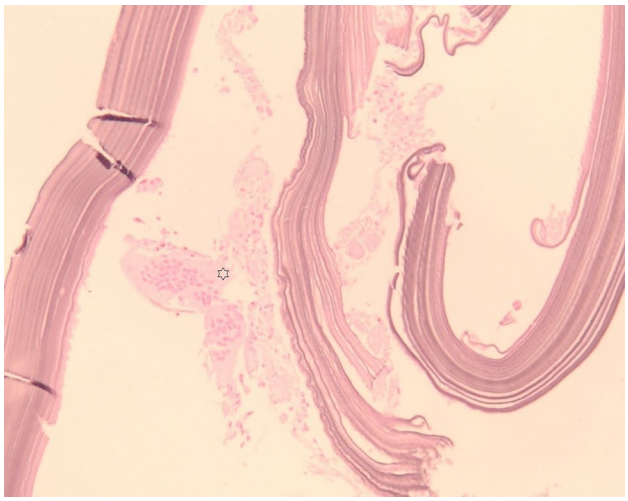

Fig. 4 Cystic echinococcosis with multilaminated membrane and protoscolices budding (star) from the germinal membrane

A full-body MRI was performed in search of further infection sites. Although the scan was inconclusive for further Echinococcus cysts, we set the patient on a course of albendazole, which is the recommended antihelminthic therapy in treatment of echinococcosis. The rest of the postoperative period was uneventful. He remains under close gastroenterological and orthopaedic (3-monthly) follow-up. At the time of our latest follow-up the patient was asymptomatic, with no signs of reinfection.

Discussion

Osseous echinococcosis is a severe and in central Europe relatively rare parasitic disease [1–4]. An early diagnosis is essential to find the right therapy. Typical symptoms of musculoskeletal echinococcosis are pain, swelling and pathological fractures, which usually lead to the diagnosis of bone hydatidosis [4, 5]. Three mechanisms are held responsible for bone destruction: local ischemia, compression and osteoclast proliferation [2].

Due to the unspecific radiographic and CT-morphologic findings of osseous echinococcosis, bone lesions are often misdiagnosed. In the absence of elements pointing towards echinococcosis in the patient's history, bone hydatids can be mistaken for benign and malignant bone tumours, which are important differential diagnoses [1, 3].

Positive serology can confirm a present or past infection with *Echinococcus* spec. and help to identify the species (ELISA and Western blot). It has to be taken into account, though, that a positive serology depends on the presence of parasitic antigens. Antigens are released especially when a cysts ruptures, a complication of the echinococcal infection

[2, 8]. Thus, the combination of sero-analysis and pathohistological examination is essential to confirm a suspected diagnosis.

Currently, there are several therapy options available for the treatment of echinococcosis, from wait-and-watch to percutaneous treatments such as puncture, aspiration, injection, re-aspiration (PAIR) and oral antiparasitic drug treatment, to radical surgical excision [1].

The recommended therapy for osseous and extraosseous cysts in hip and pelvis is a radical surgical excision combined with pre- and postoperative chemotherapy to prevent recurrence, followed by a one- or two-stage THR [2, 3]. At the moment, the role of preoperative (neoadjuvant) therapy is difficult to evaluate. In some studies it did not show any benefits in terms of reduction of the recurrence rate [4, 9]. The same applies to a "second" surgical look, even if the initial approach was similar to oncologic surgery [10].

The duration of postoperative adjuvant chemotherapy for musculoskeletal cystic echinococcosis is unclear. According to Steinmetz et al., antibiotic therapies of <3 months yield outcomes similar to therapies >6 months [4]. Albendazole is regarded by many specialists as the first-line chemotherapeutic in the treatment of bone lesions, because of its higher blood plasma levels compared to mebendazole [11]. However, comparative studies between these two agents in alveolar echinococcosis suggest that they have the same clinical activity for bone [12]. The same applies to possible advantages of a combination of albendazole and praziquantel [1, 2].

The high recurrence rate of cystic echinococcosis is important especially for young patients. Steinmetz et al. describe a local recurrence rate of 17% for cystic echinococcosis, whereas others report a local recurrence rate of up to 50% [3–5]. Local recurrences after 10 years have been reported [4]. Thus, it remains unclear in some cases, whether the patient is cured or merely in a long-lasting remission of their infection [4, 13].

In our case there were no preoperative, especially radiological, signs of a persisting low-grade infection or even a parasitic disease. Preoperative knowledge of the disease would have necessitated a different surgical procedure, with a wide radical surgical excision with removal of the affected bone stock of the femoral shaft, followed by the implantation of a megaprosthesis. Such procedures are associated with increased postoperative complications such as wound infection or periprosthetic infections [14]. Furthermore, the resection of the potentially infected soft-tissue (daughter cysts) and muscle including the gluteal muscles surrounding the cysts would have had a decisive impact on the postoperative functional result. It has to be critically discussed if the patient's echinococcosis could have been diagnosed earlier if representative samples for histopathological examination had been taken during the septic revision surgeries or if the proximal femur was already weakened through small osseous cysts even before the initial trauma that occurred 2 years previously.

Furthermore, it remains unclear if the reaming of the femoral shaft as preparation before implantation of the femoral stem could have caused a spreading of the echinococcosis. In this context, it has to be critically discussed if a second operation with explantation of the modular hip stem, resection of the possible infected femur and reimplantation of a megaprosthesis would have been obligatory or if the sole postoperative oral antihelminthic therapy, due to the performed debridement, could be in this special case sufficient.

In view of the final histopathological results as well as the patient's age and his active life-style, we decided together with the patient and his family to perform a wait-and-watch strategy under close follow-up. The patient was informed in detail regarding potential complications and the possible need for further surgeries.

To our knowledge, this is the first case report describing a primary osseous echinococcosis in central Europe/Germany as incidental finding after performing a RTHA. It highlights the importance of a rapid tissue diagnosis followed by sero-analysis to confirm a suspected diagnosis. Moreover, it outlines the need for awareness of this rare and unusual diagnosis is RTHA.

Learning points:

- Osseous hydatidosis is a very serious parasitosis requiring a combined approach of surgical excision and chemotherapy.
- Cystic echinococcosis is or can be an important differential diagnosis in revision arthroplasty for patients from endemic areas.
- Sero-analysis and histopathological examination are essential in diagnosing an echinococcosis. Due to a high recurrence rate, a long-term chemotherapy with albendazole is necessary.

Compliance with ethical standards

Conflict of interest On behalf of all authors, the corresponding author states that there is no conflict of interest.

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