



Case Report

Hip joint hydatidosis after prosthesis replacement

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SUMMARY

Hydatidosis is a rare parasitic disease caused by the *Echinococcus* tapeworm, which only occasionally affects the musculoskeletal tissues. In this article we describe the case of a patient who underwent a total hip replacement procedure for a pathological fracture of the femur neck. At the next histological examination it was shown to be a consequence of secondary bone hydatidosis. This clinical case is exceptional in that the infection spread to the cotyloid and femoral prosthesis components and, in the following years, caused repeated episodes of joint dislocation.

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Introduction

Hydatidosis is a parasitic disease that is endemic in the Mediterranean area. Although the definitive host is the dog, it occasionally affects man as an intermediate host when the larval form of the *Echinococcus granulosus* tapeworm implants in the tissues.¹ The larvae develop in cystic form, usually in the liver and lung, and over a period of time provoke symptoms due to local compression and generalized allergic-anaphylactic phenomena.² Localization in the bone tissue is extremely rare, having an incidence of 0.5–2% of all cases of hydatidosis.³ In such cases the sites most commonly involved are the more vascularized areas; the vertebral bodies are involved in 50% of cases, followed, in descending order of frequency by the epiphysis of the long bones, the ileum, the cranium and the ribs.⁴ Radiography, echotomography and magnetic resonance imaging raise the diagnostic suspicion of the disease, but confirmation is based on immunological tests, such as indirect hemagglutination, ELISA and immunoelectrophoresis, and on the subsequent histological examination.⁵ Treatment consists of surgical removal of the mass and subsequent chemotherapy.

The clinical case

In October 2004, a 53-year-old countrywoman from southern Italy presented to our casualty department with a dislocated

arthroprosthesis of the left hip. While taking her medical history it was discovered that since 1985 she had started to feel pain in the left pelvic region. In 1991, after a fall, she suffered a contusion trauma of the pelvic region and was admitted to an orthopedics and traumatology ward to undergo clinical and instrumental tests. In particular, pelvic computed tomography (CT) scans showed thinning of the cortical bone with some sclerotic areas affecting the left iliac crest up to the superior two-thirds of the sacral ala. These alterations were considered to be idiopathic with no clinical relevance, and no further investigations were made. The patient was prescribed only a few days of rest.

Owing to persistence of the pain in the abdomino-pelvic region, she later consulted a specialist surgeon, who performed abdominal CT scans that revealed a multilobed retroperitoneal mass with a cystic appearance, of an unknown nature. This was surgically removed. Postoperative immunohistological tests led to a diagnosis of hydatid cysts. The patient received no indications for further treatment and no instructions to undergo periodical monitoring for recurrence.

In the following years the patient continued to suffer left coxalgia until, in 1996, without any evident traumatic event having occurred, an X-ray showed a left pertrochanteric fracture (Figure 1). The etiopathogenesis was not further studied and the patient was submitted to a total left hip replacement procedure (Figure 2). In 1998, 2 years after the operation, she suffered a dislocation of the prosthesis (Figure 3). This was non-surgically reduced and stabilized with a pelvic-podalic plaster. In 1999 the prosthesis disassembled and a new operation was necessary, when a new self-retaining cotyle with a metal–polyethylene cup was

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Figure 1. Femoral X-ray showing cystic bone lesions at the level of the pertrochanteric fracture.

positioned (Figure 4). In 2001 a second dislocation occurred, that was again reduced and stabilized.

In 2004 the patient came to our attention for yet another disassembly of the prosthesis (Figure 5). After thorough investigation, repeat surgery was considered necessary to replace the cup with a Wagner type (Figure 6). Perioperatively, together with signs of metallosis, a cystic sac filled with a translucent fluid and a few centimeters long was observed (Figure 7), adhering to the muscle planes of the quadriceps and the abductors and extending to the articular space. This was removed and histologically evaluated. The prosthesis revision procedure proceeded as planned, with no subsequent complications. Examination of a biopsy sample led to the diagnosis of hydatid cysts. We reviewed the patient's clinical history in the light of this finding. Radiography of the pertrochanteric fracture revealed oval osteolytic areas that had not previously



Figure 2. Post-operative femoral X-ray after the first total hip replacement procedure.

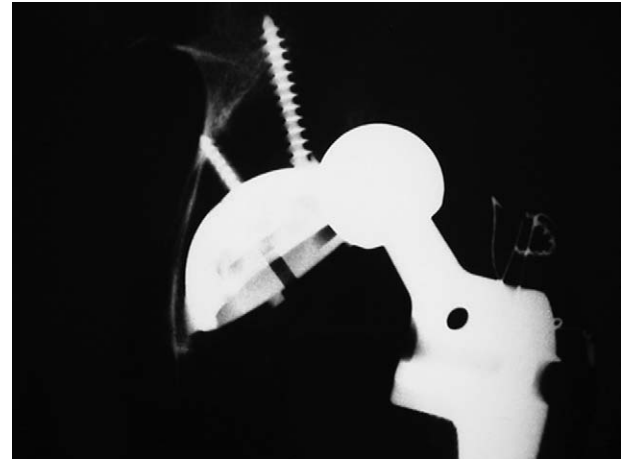


Figure 3. Femoral X-ray showing the first episode of prosthetic dislocation.

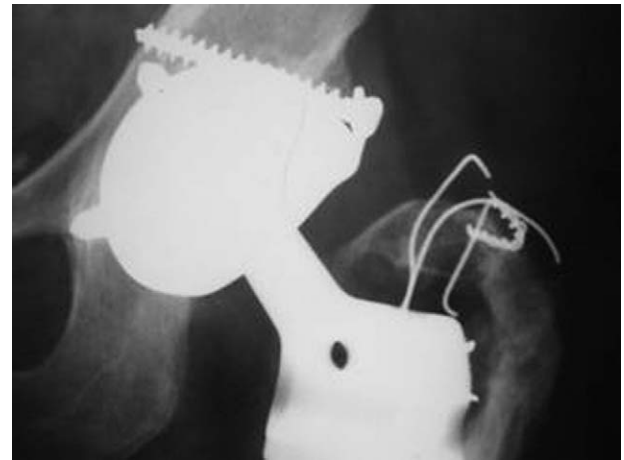


Figure 4. Post-operative femoral X-ray after the second procedure to position a new cotyle.

been recognized as hydatid cysts (Figure 1). On this basis, we reconstructed the dynamics, concluding that the primary localization of the hydatid infection in the retroperitoneum had been followed by a secondary spread to the femur, and that it was this that had caused the pathological fracture. In turn, this had led to further joint invasion, resulting in mobilization and recurrent



Figure 5. Femoral X-ray revealing disassembly of the prosthesis.



Figure 6. Post-operative femoral X-ray after the third procedure to repair the head.

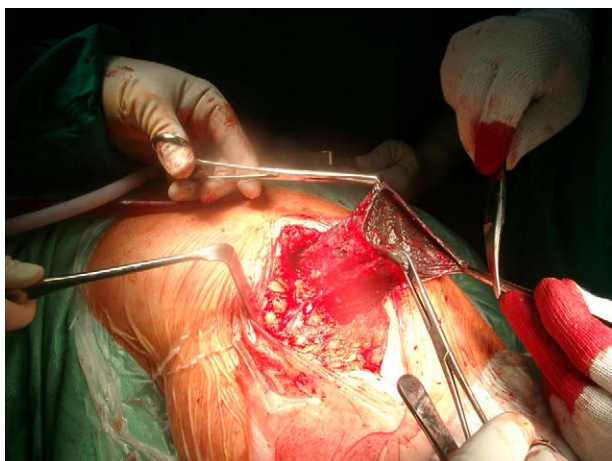


Figure 7. Image of the operative field during the third surgical procedure, showing the presence of a voluminous, fluid-filled cystic sac extending to the muscle planes and originating in the prosthetic joint space.

dislocation of the prosthesis. We decided to institute chemotherapy with albendazole (10 mg/kg) for 3 months, with monitoring of liver function. At the last follow-up, 5 years later, the patient has had no further secondary localization of the hydatidosis and the prosthesis is correctly positioned and stable.

Discussion

Bone hydatidosis, caused by *E. granulosus*, occurs when the protoscolice colonizes the bone tissue, generally by secondary spread from a parenchymal organ.⁶ One of the main characteristics of this localization is a much slower and less uniform growth than in other tissues. The resistance of bone tissue opposes the growth of the parasite, which attempts to extend at a distance through the

bone lacunae delimited by the osteones. Firstly, degradation of the trabecular component occurs, with no cortical expansion, then the increased pressure of the cysts breaks through the cortical layer and involves the neighboring tissues. Finally, the cysts break up and agglomerate in daughter cysts rather than persisting as a single hydatid cyst.⁷ This leads to localizations in the surrounding soft tissues, which offer less resistance and so facilitate loco-regional growth.⁸ Fewer than 20 cases of a joint hydatidosis localization have been reported in the literature. In these cases, the spread of the disease was largely to adjacent areas, whereas a spread through the bloodstream was less frequent. In the patients described, cystic growth was slow and the capsular organization, which acted as a barrier, did not cause any eosinophilic or antibody reaction in the blood, so serum echinococcal antigen or antibody tests were usually negative. It is also true that cysts localized in the muscular-skeletal tissue do not generally induce an antigen reaction, unlike those localized in the liver.⁶ The clinical picture is often multiform, and variable symptoms such as lumbosacral pain⁹ or a cauda equina syndrome have been described for intervertebral lumbosacral localizations,² or a reactive synovitis in the knee joint¹⁰ or cervicobrachialgia when the atlanto-occipital or cervical joints are involved.¹¹ The diagnosis of joint hydatidosis is made on the basis of histological findings in the surgical specimen, because the objective examination and laboratory or instrumental tests yield only aspecific findings.¹² A joint localization can produce various complications, ranging from a simple block or mechanical instability, as occurred in our patient, to more severe events such as rupture of the cysts and dissemination of the hydatid fluid, triggering anaphylactic shock.

Our case report is peculiar not only in view of the joint localization, but above all because of the subsequent spread to the prosthesis components. In fact, only one other case of a similar localization has been described in the literature, caused by dissemination after a hip replacement procedure for primary bone hydatidosis. The latter case had a poor outcome, since the patient gradually developed sepsis and died, despite having complained for years of a severe functional limitation and pain, which were badly underestimated.¹⁰

The fact that the prosthesis components lack the immune system recognizing non-self antigens resulted in a failure to prevent implantation and growth of the cysts,¹³ which gradually extended from the joint spaces towards the muscle planes, where we found them at surgery. This led us to review the clinical history and arrive at the correct diagnosis of Echinococcus cysts.

This case underlines the need to bear in mind the possibility of hydatid infections when making a differential diagnosis of undefined masses in patients coming from endemic areas. This is particularly important when the patient's history is suggestive of hydatidosis, because the possibility of local or distant recurrence should be considered. In fact, echinococcosis is difficult to eradicate and unless adequate surgical and/or chemotherapeutic treatment is administered, the infection can reimplant at a new site as a secondary localization.² In the case of our patient, failure to recognize the bone localization led to a subsequent spread of the disease, causing instability of the hip prosthesis and hence the need for repeated corrective surgical procedures. A closer consideration and better interpretation of the patient's history, together with proper diagnostic and immunological tests, would have enabled a more timely diagnosis and avoided the subsequent complications.

In 2001, Santavirta et al.¹⁴ advised against surgery to treat bone hydatidosis; on the basis of their own experience and a review of the literature they concluded that surgical removal of the cysts and subsequent joint replacement increases the risk of spread and can pose a life-threatening risk. They believe that chemotherapy alone to eradicate the infection is the best treatment option. Our case report confirms that surgery can allow hydatid cysts to spread to

the joint spaces. The successful outcome in our case was due to the combination of surgery with chemotherapy.

Our patient's history was remarkable for a previous diagnosis of abdominal hydatidosis, and then of a pertrochanteric femoral fracture treated with a hip replacement procedure. The eventuality of a secondary bone localization of the echinococcal infection had not been envisaged, and failure to do so caused spread of the disease to the adjacent prosthetic components and hence repeated episodes of mechanical instability of the implant. Despite the late recognition of the secondary localization, no severe complications arose, since the subsequent prosthesis replacement and specific chemotherapy prevented any further recurrence.

Conclusions

The case we have described raises a number of points for consideration. First of all, the optimal treatment for musculoskeletal hydatidosis is a combination of surgery and chemotherapy, since the former can eradicate the infection and the latter can strongly limit the risk of spread.

Another important point is that patients with a history of hydatid cysts should undergo periodic controls and long-term follow-up to prevent or limit recurrence.

Finally, when the patient has already suffered a previous episode of *Echinococcus* cysts, the possibility of a secondary localization should always be taken into account in the differential diagnosis when the clinical picture is compatible with a mass of an undefined nature, even after the performance of serological and instrumental tests.

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