Coexistence in unusual locations of hydatid cysts: thigh, breast and lung involvement

Hydatid disease is a parasitosis caused by *Echinococcus granulosus*. It has been known since the time of Hippocrates and is still an endemic disease in some regions of the world, particularly in South America, North Africa, Asia and Australia. The incidence of hydatid cysts is 18—20/100 000 in Turkey, one of the endemic countries.²

A hydatid cyst may develop in almost any part of the body, and in adults the lung is the second most common target for hydatid disease after the liver.² A subcutaneous cyst in the proximal thigh and breast involvement are very rare. We describe herein a case presenting with subcutaneous hydatid disease of the thigh associated with breast and lung involvement.

A 32-year-old woman was admitted to our clinic with complaints of thigh and left-sided chest pain. Laboratory tests were normal. Physical examination revealed a painful mass, palpated subcutaneously on the lateral thigh, over the fascia lata (Figure 1). Ultrasonography of the left breast revealed a cystic lesion measuring 1 cm in diameter.

The patient underwent single-stage surgery. The first step of the surgical procedure was excision of the cyst in the breast. The second step involved cystectomy plus pericystectomy performed on the lung lesion via a left posterior-lateral thoracotomy, and pericystectomy performed on the thigh lesion via a left lateral longitudinal incision. The last two surgical interventions were carried out with the patient in a lateral decubitus position. The diagnosis of hydatid cyst was confirmed by histopathological examination (Figure 2); scolecites were seen in histopathologic section of the hydatic cyst. The patient was given a course of medical treatment (albendazole 10 mg/kg/day) and after 6 months of follow-up, there was no evidence of recurrent disease.

Hydatid disease is still a major health problem that affects both humans and animals in our country. Human hydatid cyst disease caused by *E. granulosus* can develop in almost any organ or tissue by hematogenous dissemination.² Three common locations of involvement are the liver, lung and the peripheral nervous system.¹ The liver is the most frequently involved organ (50—60%), followed by the lung (10—30%). Soft-tissue echinococcosis occurs in 2.3% of cases reported from endemic areas and is usually associated with other solid organ involvement.²

Musculoskeletal echinococcosis is observed in 1—5.4% of all cases of hydatid disease.²—⁴ Cases of musculoskeletal hydatid cysts have been reported involving the supraspinatus, pectoralis major, brachial biceps, thigh and chest wall.
However, there have been very few reports of thigh involvement. In the series of Safioleas et al., thigh involvement was seen in only 0.37% of 272 cases of hydatid cysts. A hydatid cyst of the breast is rare even in endemic regions, accounting for 0.27% of hydatid disease cases.

Mammography and ultrasonography are not entirely helpful in the differential diagnosis of a hydatid cyst of the breast. Fine-needle aspiration has diagnostic value but contamination risk should be kept in mind when this is performed in cases of hydatid disease. Serological tests may not always be positive, as in this case. MRI can adequately demonstrate most features of the hydatid cyst, and ultrasound and CT are also helpful in making a preoperative diagnosis. Total cyst excision is the treatment of choice, and combination adjunctive chemotherapy with anthelmintics is recommended to reduce the risk of dissemination during surgery and to prevent recurrence.

If the pulmonary hydatid disease is complicated, it may resemble other malignant and benign lesions of the lung. In the current case, however, there was suspicion that the lung lesion was a ruptured hydatid cyst because of the multiple other organ involvement and the fact that this is an endemic region. The diagnosis of complicated hydatid cyst was confirmed by histopathological examination.

Our case concerns multiple atypical involvement including thigh, breast and lung. Coexistence of hydatid cysts in these localizations has not been previously reported. This case supports the overall opinion that hydatid cysts can be located simultaneously in various tissues. Treatment consists of the excision of the cyst and postoperative medical treatment.

In conclusion, hydatid disease should be considered in the differential diagnosis of all cystic lesions even if they are in atypical locations, especially if they occur in Echinococcus endemic regions.

References

Imipenem-resistance in *Klebsiella pneumoniae* in Malaysia due to loss of OmpK36 outer membrane protein coupled with AmpC hyperproduction

Carbapenems are frequently used to treat infections caused by extended-spectrum β-lactamase (ESBL)-producing *Klebsiella pneumoniae*. Resistance to carbapenems in *K. pneumoniae* is infrequent and may be due to production of AmpC cephalosporinase combined with decreased outer membrane permeability due to loss or alteration of porins.\(^1\)

A second possible mechanism of resistance is production of a β-lactamase that is capable of hydrolyzing carbapenems. This includes the metallo-β-lactamas (MBLs) of class B and the class A imipenem-hydrolyzing non-MBLs such as KPC-1.\(^2\)

In June 2004, an imipenem-resistant strain of *K. pneumoniae* (1/B37) was isolated from the blood culture of an inpatient at the University of Malaya Medical Centre (UMMC), Kuala Lumpur, Malaysia. We present a brief clinical history of the patient and report our findings on the characterization of the mechanism of resistance to imipenem in the strain.

The patient, a 42-year-old woman who had undergone a myeloablative allogeneic peripheral blood stem cell transplant for acute myeloid leukemia, presented on day 22 post-transplant with fever and abdominal pain. Her blood counts were normal, with hemoglobin of 120 g/dl, white cell count of 13.2 \(\times\) \(10^9\)/L with 83% neutrophils, and a platelet count of 146 \(\times\) \(10^9\)/L. A working diagnosis of septicemia with likely acute graft-versus-host disease (GVHD) was made, and routine blood cultures and blood fungal cultures were carried out. It was noted at this time that the site of the peripherally inserted central catheter (PICC), which had been inserted during the transplant, was inflamed; the catheter was removed. Empirical treatment with intravenous imipenem, vancomycin and methylprednisolone was started, and two days later a triple lumen catheter was inserted. The blood cultures for bacterial and fungal pathogens were negative. She developed multiple macular rashes that became progressively worse and the steroid dose was increased. Cyclosporin, which had been instituted immediately after her transplant, was then changed to intravenous tacrolimus. Intravenous immunoglobulin was also given at this time and repeated fortnightly. On day 28 post-transplant, in view of the progressive and severe nature of the acute skin GVHD (stage 4), intravenous infliximab 600 mg weekly was instituted. Earlier skin biopsy findings confirmed the diagnosis of acute GVHD. Her fever settled and repeated blood cultures showed no growth; vancomycin was stopped but imipenem was continued. However, the skin GVHD progressively worsened and antifungal and antiviral prophylaxis (flucnazole and acyclovir) were started in view of her severe immunosuppressed state. A skin swab taken at this time grew *Proteus spp* and *Acinetobacter baumannii*, and intravenous Unasyn\(^6\) was added. A day later, small skin nodules suggestive of systemic fungal infection were noted on her back and neck folds. Fluconazole was stopped and intravenous amphotericin B was commenced. Following this the patient developed high fever with new findings in her lungs. Intravenous vancomycin was added to the antimicrobial regimen. A septic workout...