

Primary Hydatid Cyst of the Axillary Region: Report of a Case

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Abstract A 53-year-old woman presented to our hospital with a painful mass in her axilla. Magnetic resonance imaging revealed a lobular cystic mass localized in the right pectoralis major muscle, with no significant invasion of the peripheral soft tissue or bone structures and no pulmonary or hepatic involvement. An echinococcal hemagglutination test was positive. Excision of the cystic mass as definitive therapy and histopathologic examination of the resected specimen confirmed that it was a hydatid cyst. For treating hydatidosis in uncommon sites, the cyst must be totally removed whenever possible. The combination of albendazole and praziquantel seems to be the most effective medical treatment, but not an alternative to surgery. This case report serves to demonstrate that hydatid disease should be borne in mind as the possible cause of a palpable lesion in the breast, axillary region, or chest wall, especially in endemic locations.

Key words Axillary mass · Echinococcosis · Hydatid cyst

Introduction

Hydatid disease is caused by the larval stage of a small zoonotic tapeworm primarily found in dogs.¹ Hydatid cysts develop most frequently in the liver and lungs, but they are occasionally found in other organs, although their occurrence in the axillary region is extremely rare, even in countries where *Echinococcus* infestation is endemic.^{2,3} According to Barret and Thomas,⁴ 60% of hydatid cysts are found in the liver, 30% in the lung, 2.5% in the kidney, 2.5% in the heart, 2% in the bone,

Reprint requests to: A.E. Unal, Sedat Simavi sk Mesa Sitesi 5, Blok 39/29, Çankaya, Ankara 06550, Turkey Received: June 27, 2000 / Accepted: March 6, 2001 1.5% in the spleen, 1% in the muscle, and 0.5% in the brain. In a series of 532 cases reported by Bickers,⁵ other rare sites included the orbit, urinary bladder, heart, chest wall, subcutaneous tissue, tibia, parotid, breast, cervicofascial region, and thyroid. We report this case of a hydatid cyst in the axilla because of its outstanding rarity and clinical confusion with other causes of axillary masses.

Case Report

A 53-year-old woman was admitted to Ankara University Hospital in August 1999 for investigation of right shoulder and cervical pain that she had experienced since May 1999. On admission she was examined by a physical therapist who advised her to do physical exercises and prescribed anti-inflammatory drugs, which in fact alleviated some of her pain. In January 2000, the patient presented again with a palpable mass in the right axillary region. Magnetic resonance imaging (MRI) localization of the right pectoralis major muscle group, in T1-weighted image hypointensity and the T2-weighted image hyperintensity, showed a lobular cystic mass measuring $75 \times 45 \times 35$ mm with no significant invasion to the peripheral soft tissue or bone structures (Fig. 1). She was referred to our department for a surgical consultation. Physical examination revealed a dense semimobile and painful mass. Laboratory findings were normal, except that an echinococcal hemagglutination test was positive, suggesting hydatid disease. The patient underwent surgery on February 2, 2000. The mass was excised, and both macroscopic and microscopic examination confirmed a primary hydatid cyst in the axillary region with germinative membranes, scoleces, and hooklets (Figs. 2 and 3). The patient had an uneventful postoperative course, and radiological examination showed no evidence of hepatic or pulmonary involvement. She was discharged on postoperative day 6 with

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Fig. 1. Magnetic resonance image of the axillary mass (arrow)



Fig. 2. Macroscopic findings of the excised cyst and its germinative membrane

no further treatment, and has remained well in the 9 months since her operation.

Discussion

Hydatid disease is a cyclozoonotic infection caused by the cestode genus *Echinococcus*. The dog is the primary host in echinococcal infestation while the intermediate hosts are sheep, cattle, horses, and occasionally, man. For this reason, the highest incidence of hydatid disease is found in sheep- and cattle-rearing regions such as Australia, New Zealand, Iceland, South America (especially Uruguay, Chile, and Argentina), East Africa, Central Europe, the Middle East, some Mediterranean countries, and parts of the former USSR. The hydatid



Fig. 3. Microscopic examination revealed a thick collagenous fibrotic cyst wall surrounded by a striated muscle fibril with lymphocyte infiltration (*arrow*), and a free scolex in the cyst lumen (*arrow*). (Original magnification $\times 100$)

disease that develops in humans is caused by the larval stage of the parasite and follows accidental ingestion of the eggs, often several years earlier. Cysts can develop almost anywhere in the body, but are rarely found in the breast, axillary region, and chest wall.^{6,7} In our patient, the disease was relatively easy to diagnose because of the clinical signs and imaging findings. MRI showed a hypointense lobular cystic mass with a low-intensity rim (pericyst-rim sign) and high-intensity peripheral soft tissue infiltration, suggesting a hydatid cyst.^{8,9} Unilocular cysts are considered to be an initial stage in the development of the parasite. In our patient, the mother cyst showed high signal intensity on T2-weighted images. The MRI findings of the soft tissue hydatidosis include a cyst or a cyst inside a cyst, a hypointense peripheral ring around the cyst (rim sign),^{9,10} collapse of the germinative membrane due to death of the scoleces and hexacant embryo (serpent or snake sign), and no production of cystic fluid or damage by trauma or aspiration.8.9 However, a unilocular intermediate or hyperintense mother cyst can be misdiagnosed as a soft tissue sarcoma or conglomerated lymphadenopathy in the axillary region. Therefore, physicians should consider the possibility of hydatid cysts in the differential diagnosis of soft tissue masses, especially in endemic regions. Today there is controversy about the pharmacological treatment alternatives for hydatidosis. High-dose mebendazole was first used in the 1970s as medical treatment, but this was toxic and not very effective. In the 1990s, several studies on albendazole showed that it was effective when given as repeated courses.¹¹ On the other hand, Yao Ping Li suggested that praziquantel, which had been as a scolicidal agent in animal models, might be benefical against human hydatidosis.12 Mohammed et al. reported that combination treatment with albendazole and praziquantel, compared with albendazole alone, reduced the duration of therapy and was effective over a short period. Currently, the combination of albendazole 400 mg/kg and praziquantel 50 mg/kg seems to be the most effective choice for medical treatment but the protocols have not yet been standardized.¹³

For treating uncommon sites of hydatidosis the general rules of hydatid surgery must be followed. If possible, the cysts must be totally removed without sacrificing the containing organs. Partial pericystectomy and evacuation of the cystic content can be performed whenever a cyst seems to be incorporated into an organ, such as renal, pelvic, or peritoneal cysts. Nephrectomy can be performed for large and organ-damaging cysts and splenectomy is mandatory for splenic cysts.¹⁴ Soft tissue cysts can be easily ruptured, and the thickness or the occurrence of a pericyst depends on the response of the host to the parasite and localization; the walls being thick in the liver, less developed in muscle, absent in bone, and sometimes visible in the brain.¹⁰ Total cystectomy is the only effective treatment for soft tissue hydatidosis, but rupture of the cyst must be avoided to prevent recurrence.14

In conclusion, although very rare, the possibility of a hydatid cyst should be borne in mind in the differential diagnosis of a palpable lesion in the breast, axillary region, or chest wall, especially in endemic regions.

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