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Primary hydatid cyst of the posterior cervical triangle

Murat Akal, Murat Kara

Abstract
Hydatid cysts in the cervical region are extremely rare. We report herein a case with a hydatid cyst that was primarily located in the posterior cervical triangle without any pulmonary or hepatic involvement. A hydatid cyst of the neck should be considered in the differential diagnosis of lesions in the cervical region, in endemic areas, so as to avoid any dangerous complications such as contamination and a fatal anaphylactic reaction.

Key words: Echinococcosis; Neck

Introduction

Hydatidosis is a cyclozoonotic infestation caused by the cestode genus *Echinococcus*. The dog and other canine species are primary hosts, while sheep, cattle, horses and occasionally humans remain as intermediate hosts. The disease is most commonly seen in the sheep-raising areas of the world such as Australia, New Zealand, the Mediterranean countries, the Middle East and South America.[1,2]

The most common form is *Echinococcus granulosus* which gives rise to cysts primarily in the liver and lungs (70–85 per cent).[2–4] Unusual locations including peritoneum, heart, spleen, kidney, spine, bones, chest wall, and other organs also have been reported.[3–5] Although, we have not detected any primary hydatid cyst of the cervical area in a review of 7532 patients who had been operated on for hydatidosis during a 40–year period in our department,[5] some cases have been presented in the literature.[1,6–8] We are aware of only two cases of a primary hydatid cyst located in the posterior cervical triangle in the literature.[7,8]

We report herein a case of a primary hydatid cyst that was located in the posterior cervical triangle and report on the potential pitfalls in the management of hydatid disease.

Case report

A 30-year-old man presented with a slowly growing swelling on the right side of his neck of one-year duration. He had an unremarkable medical history. On physical examination, a painless, semi-solid mass 8 cm in greatest diameter was detected in the supraclavicular area of his neck. The mass was in close association with the sternocleidomastoid muscle and extended behind it. Laboratory data were within the normal limits. A chest X-ray was normal. Cervical ultrasonography (US) revealed a calcified, multilocular cystic lesion with a double-layered membrane, highly suggestive of a hydatid cyst (Figure 1). Computed tomography (CT) showed a well-circumscribed, lobulated, 8 cm cystic mass in the right supraclavicular area, with a density of 20 HU (Housfield Unit) and no evidence of invasion to the contiguous structures (Figure

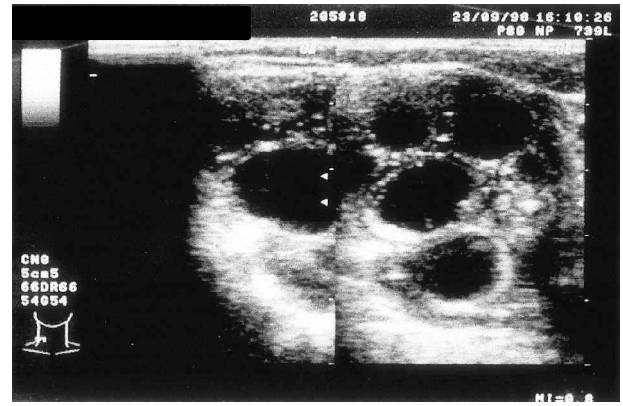

Fig. 1
Cervical ultrasonography showing a calcified, multilocular cystic lesion with a double-layered membrane.

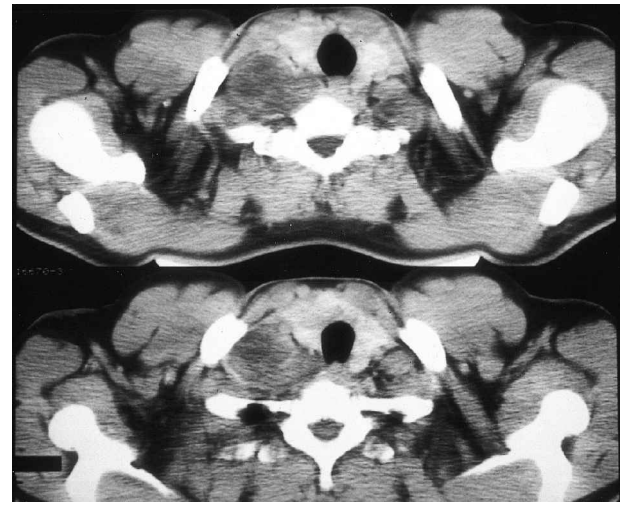

Fig. 2
CT scans show a cystic mass in close association with the right sternocleidomastoid muscle.

From the Departments of Thoracic Surgery, Ankara University School of Medicine, İbn-i Sina Hospital, Ankara, Turkey and University of Kırıkkale, School of Medicine*, Kırıkkale, Turkey.
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FIG. 3
Germinative membrane and cystic vesicles removed at surgery.

2). Further investigation including abdominal ultrasonography, bone scan and cranial CT revealed no evidence of a possible extracervical site for a hydatid cyst.

Under general anaesthesia, the overlying skin was incised and the neck was explored. The mass was found to be located in the right posterior cervical triangle. Dissection of the dense adhesions between the cyst and adjacent structures, particularly those of the right internal jugular vein was performed. Following the protection of surrounding structures with a scolicidal agent, Savlon (15 per cent cetrimide, 1.5 per cent chlorhexidine gluconate) solution-soaked pads, from a possible contamination of the cystic contents, a fine-needle aspiration biopsy confirmed the lesion to be a hydatid cyst. Two hundred and fifty ml of crystal clear fluid was aspirated until the cyst walls collapsed. Numerous daughter cysts with the germinative membrane were evacuated from the cyst cavity following cystotomy (Figure 3). Subsequent excision of the pericystic tissue also was performed. The patient was given post-operative 15 mg/kg albendazole chemotherapy for two months. He showed an uneventful recovery and remains disease-free two years after surgery.

Discussion

Echinococcus lives in the small intestine of the primary hosts. The terminal segment of *Echinococcus granulosus* contains hundreds of eggs, which are expelled with the faeces. Embryos are liberated in the duodenum following the ingestion of the parasite's eggs by the intermediate hosts such as humans, sheep, and cows. Embryos pass through the intestinal wall to enter the portal circulation and most of them are caught in the hepatic sinusoids and hydatid cysts develop in the liver. Although the liver remains the most frequent location for hydatidosis, a few embryos may pass through the liver via the hepatic veins, inferior vena cava, heart, and pulmonary arteries. Those, entrapped within the lung parenchyma develop hydatid cysts of the lung, while some may even pass through the pulmonary capillaries and reach the systemic circulation.[2] The most common sites for hydatid cysts are the liver and the lungs, the embryos may lodge in any part of the body and result in different clinical features. Hydatid cysts located in the neck are extremely rare even in endemic areas and to date, very few cases have been reported. In a review of 49 patients with hydatid cysts, especially those located in various organs other than the liver and lungs, none was located in the cervical region.[3] Moreover, chart analysis of patients who had undergone operations for hydatid cysts in our department during a 40-years' period revealed no patient with a hydatid cyst primarily located in the cervical region. In this case the cyst was located in the posterior cervical triangle with no extra-cervical manifestations of the disease, whereas in a previously reported case of cervical hydatid disease there was a history of hepatic involvement.[8]

The diagnosis of hydatid disease may be established with clinical presentation, plain radiographs and current imaging techniques such as US and CT. Unless suspected or demonstrative radiological findings are available, pre-operative diagnosis may be missed. Hence, the cystic fluid may spurt out during the operation,[7] which may present as an early or a late complication. Our experience is that if spillage of the cyst contents occurs in the pre-operative or per-operative period, the lesions is very likely to transform into an untreatable multiple hydatidosis. Thus, pre-operative diagnosis is of clinical significance in hydatid disease. Ultrasonography and CT examination is useful in visualizing cystic masses by demonstrating internal septae and daughter cysts.[1,7,8] Laboratory tests, those of the intradermal Casoni's test and serological examinations no longer have a place in the diagnosis as these have low diagnostic sensitivity and specificity.[3] Macroscopic view of the whitish, clear cystic fluid and the germinative layer is often adequate for an intra-operative diagnosis of hydatid disease.

The diagnostic use of a fine-needle aspiration biopsy in hydatid disease is controversial at present because it has dangerous potential risks and pitfalls such as spillage of the embryos with resultant contamination and fatal anaphylactic reaction following the rupture of the cyst.[2,9] Our policy in dealing with hydatid cysts is to avoid any attempt at pre-operative fine-needle aspiration biopsy if any suspicion appears about this infestation. Clinicians often lack sufficient experience of the management of hydatid cysts in the areas of low endemicity, thus a clinician should be aware of a possible diagnosis of a hydatid cyst, which may even be located in the cervical area and should preferably avoid any biopsy before surgery.

Hydatid cysts consist of three main layers. The outer most layer is the pericyst, occurring as a result of host reaction. The middle layer, the so-called cuticular membrane, is acellular and permits the passage of nutrients to the cyst cavity. The most inner, germinative layer produces the scolices that represent the larval stage of this infestation.[2] To our knowledge, the management of a hydatid cyst in any organ is inadequate, and in addition to recurrence, suppuration of the cyst cavity is very likely unless the germinative layer is removed. Thus, the optimal treatment of choice is surgical removal of the cystic material including the germinative layer with subsequent obliteration of the potential space occupied by the cyst, which is referred to as cystotomy and capitonnage. Cystectomy or so-called enucleation is an alternative surgical procedure. Similarly with the previously reported cases of hydatid cyst of the posterior cervical triangle, we performed a cystotomy and subsequent excision of the pericystic tissue in the presented case. The pericystic tissue should preferably be left, particularly in patients with hydatid cyst of the lungs as the main goal is to preserve as much lung tissue as possible and perform a parenchyma-sparing procedure in these patients.

Surgery still remains the optimal treatment of choice. Alternative therapy with non-toxic scolocidal agents or combination chemotherapy by using imidazole derivatives, particularly albendazole has been advocated to be of therapeutic value in the management of patients with recurrence and high risk of contamination. Although we detected no signs of any other organ involvement,

considering the possible presence of embryos in the circulation, we initiated a supplementary albendazole medication in the presented case.

A hydatid cyst of the neck should be considered in the differential diagnosis of lesions in the cervical region so as to avoid any dangerous complications such as contamination and a fatal anaphylactic reaction.

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Address for correspondence:
Dr Murat Akal,
Tezel sokak 9/12,
06550, Aşağiayranci,
Ankara, Turkey.

Fax: +90 312 3106371
E-mail: akal@medicine.ankara.edu.tr; muratakal@turk.net

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