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CO-OCCURRENCE OF DIAPHRAGMATIC AND SERRATUS ANTERIOR MUSCLE HYDATIDOSIS: AN UNUSUAL LOCALIZATION

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□ **Abstract—Background: With hydatid cyst, the skeletal muscles and diaphragm are rarely affected, and hepatic and pulmonary hydatid cysts are far more common. We report a case with an unusual localization of diaphragmatic and serratus muscle anterior hydatidosis that occurred simultaneously. Case Report: A 37-year-old developmentally disabled woman presented to the Emergency Department (ED) of Harran University with tachycardia, tachypnea, and dyspnea. On pulmonary auscultation, breath sounds were decreased on the right side. A chest X-ray study revealed a radiopaque right hemithorax with a mediastinal shift and tracheal displacement. Thoracic computed tomography scan revealed a hydatid cyst in the serratus muscle anterior and cystic vesicles in the pleural cavity. The patient underwent chest drainage. During drainage, daughter vesicles within the pus were detected macroscopically. An elective thoracotomy was performed after hemodynamic stabilization of the patient. Postoperative chest X-ray study demonstrated that the lungs had re-expanded. The patient had no postoperative complications and was discharged with relief of all symptoms. Conclusion: Hydatid cyst should be considered, especially in endemic regions, in the differential diagnosis in the presence of a rare localization or unexpected clinical presentation. Surgical intervention is the appropriate approach for the treatment of hydatid cyst when there is concomitant intrathoracic involvement. © 2012 Elsevier Inc.**

□ **Keywords—serratus muscle anterior; hydatid cyst; diaphragm; unusual localization**

INTRODUCTION

Hydatid cyst is an infection caused by the cestode *Echinococcus* (1). *Echinococcus* tapeworms are parasitic organisms with a two-stage life cycle. In the adult phase, the tapeworm lives in the intestines of the dog; the so-called "definitive" host. A great number of other animals, especially sheep and cattle, and occasionally humans, can become infected with the larval stage of the worm; they are called "intermediate" hosts. Intermediate hosts become infected when they ingest food or water contaminated by dog feces. Most human cases occur in areas where dogs and livestock are raised together (2). It affects whole organs or tissues in the human body, especially the liver (65–75% of cases) and lungs (25–30% of cases) (1–4). Primary hydatid cyst of the diaphragm is estimated to comprise $< 1\%$ of cases (5). Muscular or soft-tissue hydatidosis accounts for about 1–5% of all echinococcal infections in endemic regions (6).

We report a rare case of hydatid cyst that occurred simultaneously in the diaphragm and in the serratus muscle anterior. To our knowledge, this has never before been reported in the literature.

CASE REPORT

A 37-year-old, developmentally disabled woman presented to the Emergency Department (ED) with new-

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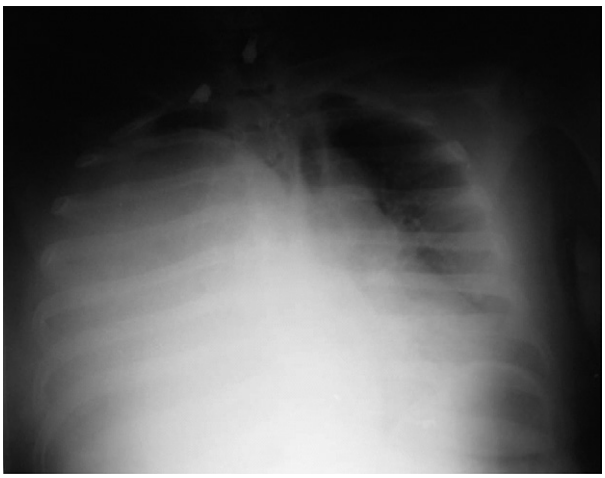

Figure 1. Chest X-ray study reveals opaque right hemithorax and mediastinal shift with tracheal displacement.

onset tachycardia, tachypnea, and dyspnea. On arrival in the ED, her skin was tense and hyperemic on the right hemithorax. Pulmonary auscultation determined that breath sounds were decreased on the right side. Controlled intravenous fluid support and supplemental oxygen by face mask were started in the ED. A chest X-ray study revealed a totally radiopaque right hemithorax with mediastinal shift and tracheal displacement (Figure 1). Thoracic computed tomography (CT) scan was performed, and it revealed hydatid cyst in the serratus muscle anterior and cystic vesicles in the pleural cavity (Figure 2). After the pus was aspirated by thoracentesis, the patient underwent chest drainage, at which time daughter vesicles within the pus were detected macroscopically. An elective thoracotomy was performed after hemodynamic stabilization of the patient.

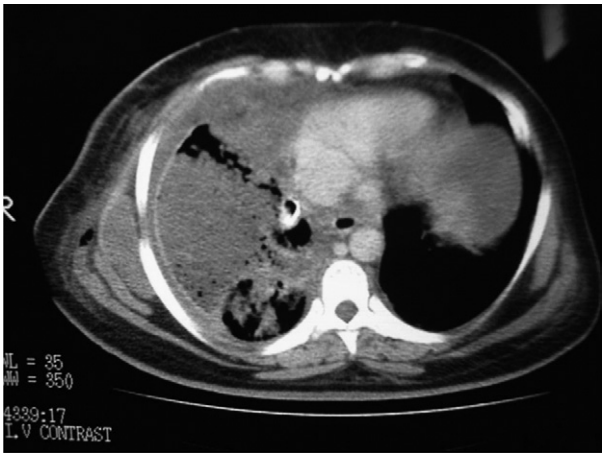

Figure 2. Thorax computed tomography scan showing cystic vesicles in the pleural cavity as well as a hydatid cyst in the serratus muscle anterior.

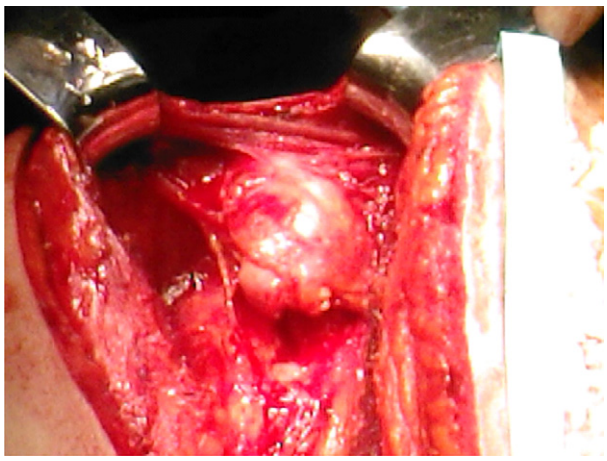

Figure 3. Intraoperative appearance of hydatid cyst in the fibers of serratus muscle.

At thoracotomy, a hydatid cyst was found in the fibers of the serratus muscle anterior and its extirpation was performed (Figure 3). Upon opening the thorax, many vesicles were found in the empyema cavity, and these were removed. In addition, it was found that the empyema cavity related to the diaphragm from the inferior (Figure 4). Due to the possibility of the presence of a hydatid cyst in the liver, the patient underwent a preoperative consultation with General Surgery. Liver exploration was performed by opening the diaphragm circularly, so that the transitional zone of the lesion was left in the center. The liver tissue was healthy and the lesion was determined to be due to multiple hydatid cysts originating from the right diaphragm with transition to the pleura. The operation was completed after resection of the empyema pouch. Postoperatively, the patient was

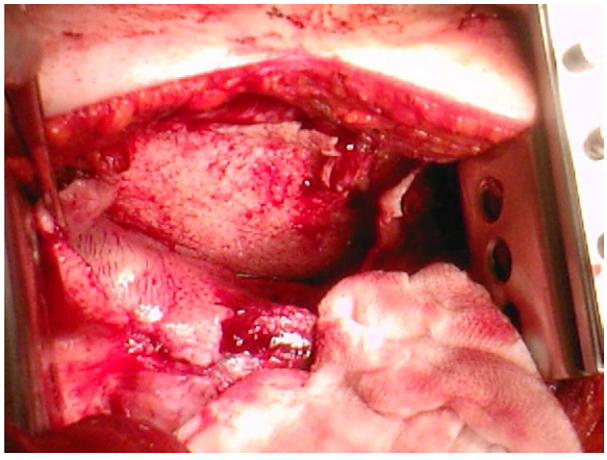

Figure 4. Intraoperative appearance of hydatid cyst located in the chest wall that related to the diaphragm.

given albendazole therapy, 10 mg/kg per day, for 3 months.

A postoperative chest X-ray study demonstrated that the lungs had re-expanded. The patient had no postoperative complications and was discharged with relief of all symptoms. In addition, no recurrence was observed during an 18-month follow-up.

DISCUSSION

Hydatid cyst is one of the most common infectious diseases in our region (southeast Turkey) (1). Although the echinococcus cyst may localize in any organ, the most common sites are the liver and the lungs (1,3). Localization of the cysts in other tissues or organs is usually secondary to the primary involvement of these organs. Prevalence of primary involvement of the skeletal muscle is 1–5% (4,6). Chest wall hydatid disease generally arises from the soft tissues of the chest wall or costae. Hydatid cysts may occur primarily in the chest wall, or chest wall involvement can be seen secondary to contamination during a surgical procedure. In a study of 6500 hydatid cyst cases, chest wall involvement was seen in only six cases (0.09%) (7). In muscular hydatid cyst, incisional biopsy or partial excision are contraindicated. In this case, due to the preoperative diagnosis of hydatid cyst, we did not incise the cystic mass of serratus muscle. After the cyst was totally resected, a thoracotomy was performed and the pleural cavity was accessed.

Diaphragmatic localization of a hydatid cyst is very rare, with an incidence of approximately 1%, and most are generally associated with the liver (5). A possible explanation for primary diaphragmatic involvement is that the parasitic embryo may have reached the diaphragm by way of the lymphatic or arterial route, without the presence of a hepatic or pulmonary hydatid cyst (8).

The symptoms and signs of hydatid cyst disease depend on the involved organ, site of localization, effect on the adjacent tissue, complications after rupture, secondary infection, and immunological reactions (9). In this case, the diaphragmatic cyst ruptured into the pleura; however, the surface of the visceral pleura was too thick to allow invasion into the lung. During the preoperative radiological investigation, we could not diagnose the diaphragmatic cyst rupture because both the clinical and the radiological presentations were the same as those for liver or lung cysts rupturing into the pleura. In this case, intrapleural rupture caused respiratory distress. Chest drainage was performed for the treatment of hydrothorax and the diagnosis was confirmed after recognition of the so-called "daughter" vesicles (see below) and perforated membrane pieces in the infected fluid.

In hydatid cyst, ultrasonography or CT scan is performed for diagnostic localization and exploration (6). In this case, however, the fact that the liver was not involved was not determined until the operation.

Surgery is the best method, both for confirming noninvolvement of the lung or the liver and for definitive treatment. However, removal of the main cyst mass may not be curative because small "daughter" cysts may be left behind (8,10). Chemotherapy with albendazole or mebendazole is effective against tapeworm disease. They may be used for the treatment of patients with inoperable disease, or as intraoperative and postoperative treatment to reduce the risk of operative spillage and recurrence (10,11). Medical treatment with these drugs as an alternative to surgical treatment has a low rate of success because the drugs are not curative by themselves (10,12).

Ultrasonographic or CT-guided fine-needle aspiration of hydatid cyst contents followed by infusion of a killing agent (e.g., 95% ethanol) and reaspiration, known as PAIR (Puncture, Aspiration, Injection, Reaspiration) therapy, has been used successfully at certain institutions; however, this therapy carries a risk of dissemination of infection or anaphylactic reaction caused by cyst puncture and leakage. Surgical removal of cysts with perioperative antihelmintic medications is the curative treatment for hydatid disease (13).

In the case presented, total removal of the cysts was performed from the diaphragm and serratus muscle anterior followed post-surgery by chemotherapy with albendazole.

CONCLUSION

Hydatid cyst should be considered, especially in endemic regions, when attempting to determine the underlying cause of a rare localization or an unexpected clinical presentation. Chest wall or diaphragmatic cysts are very rare localizations. Surgical intervention is the appropriate approach for their diagnosis and treatment and should be performed in association with diagnosis and treatment of concomitant intrathoracic involvement. To the best of our knowledge, in the literature there is no reported simultaneously localized hydatid cysts within the diaphragm and the serratus muscle anterior.

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