brief report

Unusually located thoracic hydatid cysts

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Human hydatid disease caused by the larval form of *Echinococcus granulosus* has a worldwide distribution and is endemic in many countries. The disease, known since the time of Hippocrates, is primarily an illness of residents in rural areas who frequently come into contact with sheep, cows and carnivores.[1-4]

The estimated surgical case rate of cystic echinococcus is 0.87-6.6 per 100 000 in Turkey.[2] The prevalence of *Echinococcus granulosus* infestation in dogs in Turkey is between 0.32% and 40% and the reported prevalence of cystic echinococcus in domestic animals has ranged from 11.3 to 50.7% and varies widely by geography.[5,6]

Echinococcal cysts may develop in almost any part of the body. The liver and the lungs are the most commonly affected areas in adults. Within the thorax most of the cysts settle in the lung parenchyma. In this situation surgical treatment is usually easy and quick. Sometimes the cyst grows as an extrapulmonary lesion or passes over the lung parenchyma to other structures. Location of the disease outside the lung parenchyma in the thorax is rare and surgical procedures can be considered that may differ from those used for pulmonary cysts.[7] The aim of this study was to review surgical techniques and possible perioperative complications. We present our experience, documenting the clinical features and the treatment employed for unusually located thoracic hydatid cysts.

METHODS

Of 552 patients with thoracic hydatidosis who underwent surgery between 1980 and 2005, 29 (5.25%) had cysts located in extrapulmonary sites. Medical records of the patients were examined for age, sex, history of living in a rural area, symptoms, localization, dimensions and number of cysts, surgical treatment methods, complications, morbidity, mortality and hospital stay after operations and recurrence.

RESULTS

Extrapulmonary thoracic hydatid cysts were detected In 29 patients, 25 (86.2%) of whom were male and 4 (13.8%) were female. Their ages ranged from 22 to 62 years with an average age of 26.8 years. Fourteen men and 3 women lived in a rural area and had a history of contact with sheep and sheep dogs. Three patients had a history of previous surgery for hydatidosis (two lungs and one liver). Chest pain, back pain, cough, fever and dyspnea were the most common symptoms. One patient with acute onset pain in the lower extremities was included due to embolus of the cyst material to the bilateral iliac arteries. Two patients (6.89%) were asymptomatic.

Preoperative radiologic diagnostic tests were based primarily on plain chest roentgenogram and CT. Chest roentgenograms led to a correct diagnosis in 6 (20.7%) cases. An additional CT scan in 21 (72.4%) cases, an MRI in 6 cases (20.7%), and echocardiography in 8 patients (27.6%) was required for diagnosis and surgical preparation. The diagnosis was established intraoperatively in 5 (17.2%) cases. In 7 (24.1%) patients there were multiple cysts. The cysts were greater then 10 cm in 5 (17.2%) patients and in 1 (3.4%) was greater then 15 cm. In one patient bilateral multiple hydatid disease was present. The cysts were located in the mediastinum in 10 patients (34.5%) (Figure 1), in the heart in 4 patients (13.8%), on the chest wall in 9 patients (31.0%) and on the diaphragm in 6 patients (20.7%). In three patients the cysts were concomitant with the liver. In one patient settlement was in the paravertebral region (Figure 2). Mediastinal cysts were the most frequent types among extrapulmonary intrathoracic hydatid cysts (Table 1).

Patients with cardiac cyst were operated on via median sternotomy and under extracorporeal circulation. To reduce the risk of pulmonary embolus, especially for the cysts in the right cardiac chambers, a cross-clamp was placed on the pulmonary artery.

All the cysts were totally excised. Twenty-three posterolateral thoracotomies (9 left and 14 right) and four median sternotomies were performed for cyst excision. The hydatid cysts were in the supraclavicular region in one patient and in the serratus muscle on the thoracic wall in another patient and therefore no thoracotomy was performed. An enucleation was performed in 6 (20.1%) cases. We were unable to perform an enucleation in 23 (79.3%) cases and the patients underwent a cystotomy operation instead. Primary diaphragmatic repair was required in four patients because of diaphragmatic defects. In addition, pleural decortication was performed in one patient for the dense pleural thickening.

There was no mortality. Intraoperative vena cava superior rupture occurred in one patient and was repaired successfully. One patient underwent rethoracotomy for postoperative bleeding. Prolonged air leak was seen in two patients. A postoperative emphyema developed in one of these patients, but none needed additional intervention. There was no other major complication.

Because of intraoperative suspicion that uncontrolled spilling of vivid cyst fluid into the pleural cavity might occur postoperatively, albendazole treatment 10 mg/kg/day for 28 days was started in six patients after surgery. However, no recurrence was observed in these or in other patients after surgery. The median duration in hospital for patients after surgery was 7.6 days (3-17 days).

DISCUSSION

In our series, 17 of the patients (58.5%) had a history of living in rural areas. Hydatid cysts cause an inflammatory reaction and destruction of nearby tissues by expanding. Especially when complicated, they tend to be severe and cause pleural thickening.[8,9] Therefore, additional surgical procedures may be required, such as decortications, wide parenchymal resections, diaphragmatic repair, and rib debridement or resections. Pleural thickening due to cyst rupture into the pleural cavity is the complication most often encountered. In the studies of Ozvaran and Kuzucu, approximately 70% of patients needed decortication for pleural complication.[8,9] In our series, dense pleural thickening, which required decortication, was seen only in one (3.4%) patient because of the low pleural involvement rate in our series.

Hydatid disease is initially diagnosed by its familiar radiographic findings in endemic areas. Currently, the chest radiograph and thoracic CT are sufficient to reach a diagnosis.[10,11] MRI and serologic tests may be helpful in the diagnosis.[12] For serological diagnosis, the most sensitive test, the IgG ELISA, should be used.

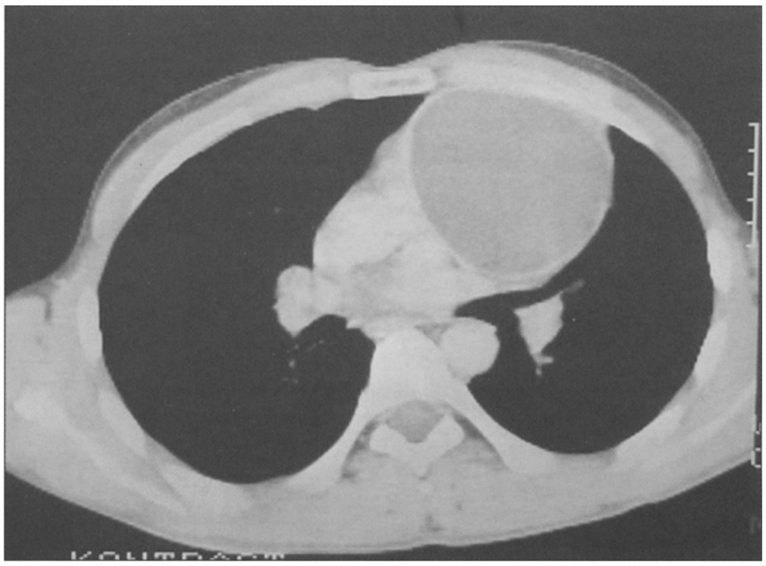

Figure 1. CT scan of a mediastinal hydatid cyst.

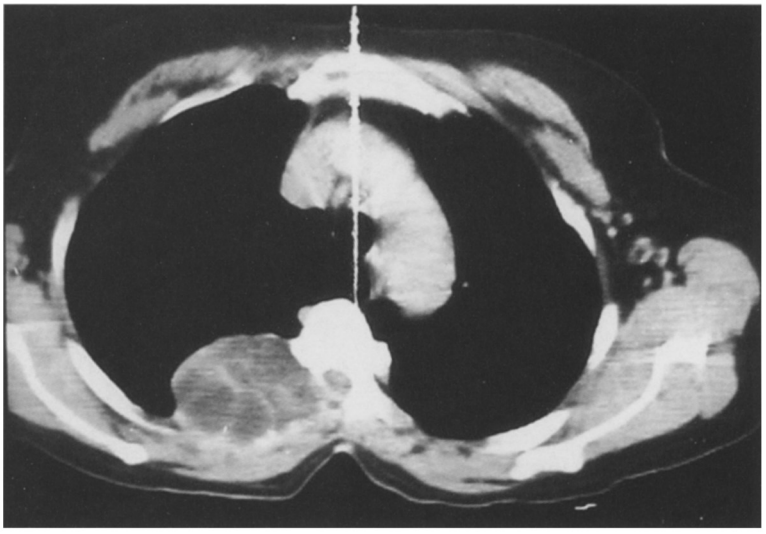

Figure 2. MRI scan of hydatid cysts in the right paravertebral region.

However, in cases with unusual localization or in complicated hydatidosis, with atypical radiological appearance, diagnosis is more difficult.

Location of the hydatid cyst in the chest wall is a very rare occurrence. Bony structures as well as soft tissues may be involved. The rate overall bone involvement in hydatid disease has been reported as 0.5% to 2.5%.[4,13] In our series chest wall involvement was seen in 9 patients. Paravertebral structures were involved in 1 patient, subcutaneous soft tissues in 2 supraclavicular region in 1 patient and the lateral chest wall subcutaneous tissue in one other patient. The other six may have

Table 1. Sites of hydatid cysts.

Affected site	N = 29	%
Mediastinum	10	34.5
Pericardial involvement	7	
Outer myocardial wall involvement	2	
Phrenic nerve involvement	1	
Chest wall	9	31.0
Rib involvement/destruction	6	
Subcutaneous or muscular chest wall	2	
Paravertebral region	1	
Diaphragmatic location	6	20.6
Diaphragmatic pleura	3	
Diaphragmatic with liver involvement	3	
Heart	4	13.8
Left ventricle	1	
Interventricular septum	1	
Right atrium	2	

been of pleural or parenchymal origin and caused destruction of the wall structures. Rib debridement and curettage were required for two patients because of bone destruction.

Rupture of a pulmonary hydatid cyst into the pleural space either spontaneously or during surgery is the most common cause of pleural or chest wall hydatidosis. Therefore, in patients who have undergone surgery for hydatidosis, the surgeon must be prepared for an unexpected hydatidosis.[14] Three patients in our series had a history of previous surgery for hydatidosis. The diaphragm or diaphragmatic pleura were less commonly involved sites for hydatid cysts in our series. Diaphragmatic involvement was seen in six patients and four required reconstruction by primary closure.

Although many uncommon locations for hydatid cysts have been reported, the disease is rarely present in the mediastinum. A large mediastinal hydatid cyst may compress the vital organs and produces pressure symptoms. Similar symptoms occur with other mediastinal cystic lesions. Differentiation may be impossible even with sophisticated radiological imaging techniques.[15-17]

In 10 of our patients mediastinal involvement was seen. In 7 patients, pericardial sacs were affected, and in two of those patients, outer myocardial wall involvement was detected. One patient had dense adhesion of the epicardium with surrounding tissues and in the other patient there was erosion of the outer myocardial wall by complicated hydatid cysts. Each patient was treated without complication.

When hydatid cysts are bounded by lung parenchyma, the usual procedures from diagnosis to treatment are simple and have very low mortality and morbidity. However, when the cyst settles in the thorax outside of lung parenchyma or extends to the chest wall, mediastinum, diaphragm or liver, especially in complicated hydatid cysts, diagnosis and treatment can be complicated. In conclusion, extra-pulmonary located thoracic hydatidosis is an uncommon condition that requires careful preparation and surgical planning to reduce peri-operative mortality and morbidity.

REFERENCES

1. Eroglu A, Kurkcuoglu C, Karaoglanoglu N, Tekinbas C, Kaynar H, Onbas O. Primary hydatid cysts of the mediastinum. Eur J Cardiothorac Surg 2002 Oct;22(4):599-601.
2. Burgos R, Varela A, Castedo E, Roda J, Montero CG, Serrano S, et al.. Pulmonary hydatidosis: surgical treatment and follow-up of 240 cases. Eur J Cardiothorac Surg. 1999Dec;16(6):628-34.
3. Dhaliwal RS, Kalkat MS. One-stage surgical procedure for bilateral lung and liver hydatid cysts. Ann Thorac Surg. 1997Aug;64(2):338-41.
4. Dakak M, Genc O, Gurkok S, Gozubuyuk A, Balkanli K. Surgical treatment for pulmonary hydatidosis (a review of 422 cases). J R Coll Surg Edinb. 2002Oct;47(5):689-92.
5. Altintas N. Past to present: echinococcosis in Turkey. Acta Trop. 2003 Feb;85(2):105-12.
6. Yildiz K, Gurcan S. Prevalence of hydatidosis and fertility of hydatid cysts in sheep in Kirikkale, Turkey. Acta Vet Hung. 2003;51(2):181-7.
7. Oguzkaya F, Akcali Y, Kahraman C, Emirogullari N, Bilgin M, Sahin A. Unusually located hydatid cysts: intrathoracic but extrapulmonary. Ann Thorac Surg. 1997Aug;64(2):334-7.
8. Kuzucu A, Soysal O, Ozgel M, Yologlu S. Complicated hydatid cysts of the lung: clinical and therapeutic issues. Ann Thorac Surg. 2004Apr;77(4):1200-4.
9. Ozvaran MK, Ersoy Y, Uskul B, Unver E, Yalcin E, Baran R, et al. Pleural complications of pulmonary hydatid disease. Respirology. 2004Mar;9(1):115-9.
10. Von Sinner WN. New diagnostic signs in hydatid disease; radiography, ultrasound, CT and MRI correlated to pathology. Eur J Radiol. 1991Mar;12(2):150-9.
11. Solak H, Ozgen G, Yuksek T, Eren N, Solak N, Kirca NK, et al. Surgery in hydatid cyst of the lung. A report of 460 cases. Scand J Thorac Cardiovasc Surg. 1988;22(2):101-4.
12. Col C, Col M, Lafci H. Unusual localizations of hydatid disease. Acta Med Austria. 2003 May;30(2):61-4.
13. Aarons BJ. Thoracic surgery for hydatid disease. World J Surg. 1999 Nov;23(11):1105-9.
14. Loudiye H, Aktaou S, Hassikou H, El-Bardouni A, El-Manouar M, Fizazi M, et al. Hydatid disease of bone. Review of 11 cases. Joint Bone Spine. 2003Sep;70(5):352-5.
15. Ozdemir N, Akal M, Kutlay H, Yavuzer S. Chest wall echinococcosis. Chest. 1994Apr;105(4):1277-9.
16. Thameur H, Chenik S, Abdelmoulah S, Bey M, Hachicha S, Chemingui M, et al. Thoracic hydatidosis. A review of 1619 cases. Rev Pneumol Clin. 2000 Feb;56(1):7-15.
17. Ozpolat B, Ozeren M, Soyal T, Yucel E. Unusually located intrathoracic extrapulmonary mediastinal hydatid cyst manifesting as Pancoast syndrome. Journal of Thoracic and Cardiovascular Surgery. 2005Mar;129(3):688-9.