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Clinical experience of surgical therapy in 207 patients with thoracic hydatidosis over a 12-year period

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Summary

Principles: Hydatid disease is the most severe helminthic zoonosis, with a major medical, social, and economic impact in Turkey. The aim of this study was to evaluate retrospectively 207 patients diagnosed with hydatid cyst and treated surgically in our department between January 1990 and December 2001.

Methods: Hundred and ninety three patients were male and 14 female. They ranged in age from 19 to 72 years (mean 25.3 years). The most common presenting symptoms were cough, expectoration and chest pain. The surgical approach was thoracotomy in 198 patients, bilateral staged thoracotomies in 5 patients, median sternotomy in one patient and video-assisted thoracic surgery in 3 patients.

Results: Hundred and thirty eight of the 265 intrapulmonary cystic lesions were found in the right lung and 127 in the left lung. Intrathoracic extrapulmonary cystic lesions were detected in 13 patients. 38 patients also had cystic lesions in the liver. Conservative sur-gical procedures were adopted except for small wedge resections in 8 patients, segmentectomy in 4 patients and lobectomy in one. Operative and postoperative mortality was nil. Albendazole treatment was given to patients who had multiple intrathoracic cysts or additional hepatic cysts after 1994.

Conclusions: Our preferred surgical techniques for removal of cysts were conservative surgical procedures such as enucleation of cysts or removal by cystotomy. Radical procedures such as pneumonectomy, lobectomy and segmentectomy should be avoided as far as possible.

Key words: hydatid disease; hydatid cyst; thoracic hydatidosis

Introduction

Hydatid disease, which is caused by the tapeworm *Echinococcus granulosus* or *Echinococcus multilocularis*, is known as echinococcosis or hydatidosis [1]. Hydatid disease is the most severe helminthic zoonosis, with a major medical, social, and economic impact in Turkey [2].

Hydatid disease had been known since the time of Galen and Hippocrates, and was described by Thebesius in the 17th century [1, 3]. Rudolphi (1808) first used the term hydatid cyst to describe echinococcosis in humans [1]. Infestation is confined to geographic areas in which there is continuous contact between humans and certain domestic carnivores such as dogs and some ungulates including cats and sheep [3]. Echinococcosis is endemic to the Mediterranean region, South America, Australia, New Zealand, the Middle East, Alaska and Canada, where it is widespread among Indian tribes [3]. The disease is encountered endemically in the eastern and southeastern region of Turkey [4]. Its incidence is said to be between 1/50,000 and 1/20,000 in Turkey [5], where it is particularly common in the rural population [5]. The lung is the second most commonly affected organ (10–40%) after the liver [2].

This study is a retrospective evaluation of patients diagnosed with hydatid cyst and treated surgically in our department between January 1990 and December 2001.

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Material and methods

Two hundred and seven patients with thoracic hydatid disease have undergone surgery in our department over the last 12 years. Hundred and ninety three were male and 14 female. The age range was 19–72 years (mean 25.3 years). 28 patients (13.5%) had a long history of close contact with animals, such as tending of domestic animals or sheep-farming.

Common presenting symptoms were cough, expectoration and chest pain in symptomatic patients (table 1). However, 41 patients (19.8%) were symptom-free and were diagnosed by chest radiography during medical checkups in a military dispensary. When oval or spherical opacities were seen in the lung we performed thoracic and upper abdominal computed tomography and abdominal ultrasonography to detect liver cysts. In 38 patients cystic lesions were detected in the liver. In two cases liver hydatid cysts were localised thoracoabdominally as dumbbell images around the diaphragm in computed tomography. The Casoni intradermal test was carried out in 83 patients and was positive in 37. The Weinberg complement fixation test was positive in 30 out of 68 patients and the indirect haemagglutination test was positive in 82 out of 124.

Table 1.

Major clinical manifestations in 166 symptomatic patients.

Clinical findings	patient no. (%)
Cough	86 (51.8%)
Expectoration	73 (43.9%)
Chest pain	46 (27.7%)
Weakness	37 (22.3%)
Dyspnoea	26 (15.6%)
Fever	11 (6.6%)
Haemoptysis	3 (1.8%)
Allergic reactions	2 (1.2%)

The surgical approach was lateral or posterolateral thoracotomy in 198 patients, bilateral staged thoracotomies in 5 patients, median sternotomy in one patient with bilateral hydatid cysts, and video-assisted thoracic surgery (VATS) in 3 patients with solitary small peripheral hydatid cysts. VATS was performed with three thoracoports and a mini-thoracotomy (3–4 cm). The cyst was removed after needle aspiration via mini-thoracotomy and bronchial fistulas were then sutured. Phrenotomy was performed in addition to thoracotomy in two patients. Thus, 212 operations were performed in 207 patients. The tissue surrounding the hydatid cyst in the operative area was covered with towels moistened with hypertonic saline to prevent inadvertent implantation of scolices or daughter vesicles. Depending on the localisation, size and intact state of the cyst we employed various surgical techniques for cyst removal such as cystotomy (incision in the lung parenchyma and removal of cyst), enucleation of the intact cyst and removal of the cyst after needle aspiration. Enucleation was performed for superficial and small cysts, cystotomy for those deep inside the parenchyma and needle aspiration for large cysts within the parenchyma to prevent implantation of cyst fluid for removal of cysts. The residual cavity and lung damage were treated by conservative methods (such as pericystectomy, which is always associated with suturing of bronchial fistulas and capitonnage of the cavity) or by adapted lung resections (such as wedge resection, segmentectomy and lobectomy).

Albendazole treatment was given to patients with multiple intrathoracic cysts or additional hepatic cysts after 1994. The albendazole regimen was 800 mg daily for three 21-day courses with 10-day rest periods between courses.

Results

The surgical procedures employed for intrapulmonary cystic lesions are given in table 2. Our preferred surgical techniques for removal of cysts were enucleation of the cyst or cystotomy. In most patients the cyst cavity was closed by capitonnage.

Of our 207 patients 194 had only intrapulmonary, 4 only extrapulmonary and 9 both intrapulmonary and extrapulmonary hydatid cysts. In 203 patients with intrapulmonary cysts a total of 265 intrathoracic hydatid cyst lesions were detected, and 32 of these patients had multiple hydatid cysts. The hydatid cysts were located in the right lung in 102 patients, in the left lung in 96, and in both lungs in 5. Thirty eight patients also had cystic lesions in the liver. Intrathoracic extrapulmonary cystic lesions were detected in 13 patients. The patient with the most abundant intrapulmonary cysts had 8 in the right lung and 3 in the left lung. In this patient no cysts were present in other organs. Hundred and thirty eight of the intrapulmonary cystic lesions were localised in the right lung and 127 in the left lung (table 3). The most common cyst localisations were the lower lobes of both lungs (173 out of 265 cysts). Of 265 intrapulmonary lesions 51 were ruptured towards the bronchus in 42 cysts, the pleural cavity in 7 and the diaphragm via the pleural cavity in 2. Twenty of 51 ruptured cysts had been infected. It was observed that among 214 intact and uncomplicated intrapulmonary cystic lesions 101 were smaller than 6 cm, 52 were between 6–10 cm and 61 larger than 10 cm. The size of the largest cyst was approximately 18×14×10 cm (figure 1).

Thirteen patients (6.2%) had intrathoracic extrapulmonary hydatid cysts. Four of these patients had no pulmonary cysts. Multiple intact and ruptured cysts in the pleural space of a patient without an intrathoracic cyst are shown as figure 2. We suggest that this patient had secondary hydatidosis having undergone surgery for hydatid cyst in the liver 13 years before. The second patient with no pulmonary cyst had 13 multilocular chest wall cysts involving muscles and destroying the left 2nd, 3rd and 4th ribs (figure 3). This patient had a history of surgery for hydatid cyst in the cerebral frontal lobe 8 years previously, which suggested the present case was secondary hydatidosis. The other patient had 12 cysts between the parietal pleura and

Table 2

Operative techniques employed for 265 intrapulmonary cystic lesions in 203 patients.

surgical procedures for removal of hydatid cyst (%)		additional palliative procedures		additional radical procedures		
		pericystectomy (%)	capitonnage (%)	wedge resection (%)	segmentectomy (%)	lobectomy (%)
Cystotomy	143 (54%)	9 (3.4%)	98 (37%)	6 (2.3%)	2 (0.7%)	1 (0.3%)
Removal of cyst after needle aspiration	47 (17.7%)	6 (2.3%)	28 (10.6%)	4 (1.5%)	2 (0.7%)	–
Enucleation of intact cyst	75 (28.3%)	4 (1.5%)	39 (14.7)	1 (0.3%)	–	–
Total	265 (100%)	19 (7.2%)	165 (62.3%)	11 (4.1%)	4 (1.5%)	1 (0.3%)

Table 3

Localisations of 265 intrapulmonary cystic lesions.

Right lung		left lung	
Upper lobe	31 (11.7%)	upper lobe	37 (13.9%)
Middle lobe	18 (6.8%)	lingula	6 (2.3%)
Lower lobe	89 (33.6%)	lower lobe	84 (31.7%)
Total	138 (52.1%)	total	127 (48.9%)

the endothoracic fascia, one cyst between the parietal pleura and the diaphragm and one diaphragmatic cyst together with a liver dome cyst forming a dumbbell image (figure 4). We believe that the liver dome cyst had opened into the diaphragm and caused two diaphragmatic cysts, and that scoleces had probably been transported by diaphragmatic lymphatics and then parasternal and intercostal lymphatics [6]. There was one secondary cyst in the chest wall with no intrapulmonary cyst in another patient who had undergone surgery for hydatid cyst in the liver 4 years previously. All three patients had both pulmonary and a total of 3 diaphragmatic cysts. One of the diaphragmatic cysts had developed in the thickness of the diaphragm in association with a liver cyst, but the other two did not involve the diaphragm. Six patients had ruptured intrapulmonary cysts and a total of 7 pleural cysts. Secondary cysts had probably developed as a result of intrapulmonary cyst rupture into the pleura. Extrapulmonary lesions were removed by cyst extirpation from surrounding tissue or by pericystectomy. In one patient with chest wall involvement, partial rib resections were carried out in the left 2nd, 3rd and 4th ribs due to rib destruction. In two patients with liver cysts migrating to the thorax through the diaphragm, the diaphragm was incised, cysts on the liver roof were removed and the diaphragm was then repaired. There was no mortality, morbidity or recurrence in these patients.

Three to 6 weeks after surgery for thoracic cysts, laparatomy was performed in the patients with hepatic cyst, except two in whom a thoracic transdiaphragmatic approach was used.

The aetiological agent was identified as *Echinococcus multilocularis* in one case and *E. granulosus* in the other patients. In the patient with *E. multilocularis* three cystic lesions recurred in the right lung six months after the operation. Regression was obtained with albendazole (800 mg daily for three 21-day courses with 10-day rest periods between courses) and no progression was observed for the next 6 years. Nor was any recurrence of *E. granulosus* hydatidosis observed in the follow-up period ranging from 9 to 51 months (mean 34 months).

Figure 1

Computed tomography showing giant hydatid cyst.

Figure 2

A. Multiple intact and ruptured hydatid cysts removed from pleural space in one patient.
B. Multiple intact hydatid cysts in the same patient.

Figure 3

Magnetic resonance imaging of multiple cystic lesions in left cervicothoracic region.

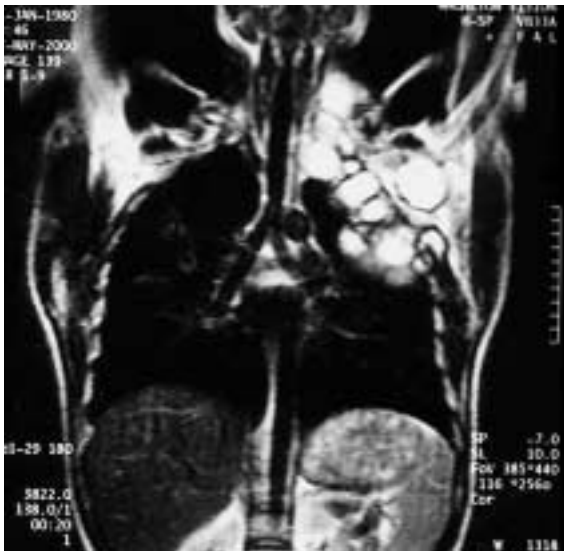

Figure 4

Computed tomography showing multiple cystic lesions between parietal pleura and endothoracic fascia.

Operative and postoperative mortality was nil. The postoperative course in most patients was uneventful. As a postoperative complication wound infection developed in two patients and a prolonged air leak developed in two others. In these two patients VATS was performed on the 5th postoperative day and air leaking was stopped by endostapling. Reoperation was not necessary in other patients. The main postoperative unit stay was 2.3 days and the main hospital stay was 7.2 days. After the operation all the patients' hydatid cyst-related symptoms were improved.

Discussion

Thameur et al. (2000) reported an intrapulmonary hydatid cyst rate of 94.6% and an extrapulmonary rate of 5.62% in 1,619 patients with thoracic hydatidosis [7]. In our 207 patients with thoracic hydatidosis 203 (98%) had intrapulmonary cysts and 13 (6.2%) intrathoracic extrapulmonary cysts. The mean age of our patients was 25.3 years, since 161 were members of the armed forces.

The World Health Organization published an excellent overview of treatment guidelines for echinococcal disease in 1996 [8]. It stated that immediate surgery may be required in patients with impending cyst rupture, compromise of vital organs due to mass effect, haemoptysis, secondarily infected cysts, infection due to obstruction and unmanageable pain [8, 9].

The current treatment for hydatid cyst of the lung is complete excision of the disease process with maximum preservation of lung tissue [1]. The surgical procedure for treatment of hydatid disease may be conservative or radical. The conservative procedures are cystotomy, enucleation of intact cysts, removal of the cyst after needle aspiration and pericystectomy (capsule resection). Radical procedures are pulmonary resection such as wedge resection (lung parenchyma resection of less than one segment), segmentectomy, lobectomy, and pneumonectomy. Peripherally located cysts of any size and small- to medium-sized centrally located cysts should and can be excised without sacrificing the lung parenchyma. In these cysts the procedures should as far as possible be conservative. For cyst removal we adopted conservative techniques in the majority of surgical procedures (93.9%). Our most commonly applied cyst removal techniques were cystotomy (54%) and enucleation (28.3%).

Segmental resection is indicated principally in the treatment of large simple cysts occupying almost the whole of the involved segment. Lobectomy should be performed when the size and number of cysts and the degree of infection exclude lesser procedures. The principal indications for lobectomy are large cysts involving more than 50% of the lobe, cysts with severe pulmonary suppuration unresponsive to preoperative treatment, multiple unilobar cysts and sequelae of hydatid disease such as pulmonary fibrosis, bronchiectasis, or severe haemorrhage. Pneumonectomy is rarely indicated for treatment of hydatid disease of the lung and should be used only when the whole lung is involved in the disease process and no salvageable pulmonary parenchyma remains [1]. Thameur et al. reported a lung resection rate of 14.1% [7]. When the lung parenchyma has been destroyed as a result of bronchiectatic changes and chronic inflammation in the lung tissue surrounding very large or multiple cysts, we employed lung resec-

tion techniques excepting pneumonectomy. We thus carried out lobectomy in only one patient, segmentectomy in 4 patients and 11 wedge resections of cysts and destroyed surrounding tissue in 8 patients. We thus performed lung resection in 13 patients (6.2%).

After the hydatid cyst is removed the bronchial openings should be closed by suturing and the pericystic cavity obliterated to prevent secondary infection [1, 5]. The residual cavity may be obliterated by capitonnage using multiple purse-string sutures from the deepest level to surface level. We used the capitonnage technique after removal of 165 hydatid cysts (62.3%).

Although surgery remains the treatment of choice for hydatid disease, the usefulness of drug therapy has been reported in many studies. Mebendazole and, more recently, albendazole have been used as primary drug therapy and as an adjunct to surgery to diminish recurrence and potential spread of the organism [9]. Horton (1997) reported that albendazole therapy in *E. granulosus* infection can result in apparent cure in up to 30% of cases, with a further 40–50% showing objective evidence of response when followed in the short term [10]. According to recent World Health Organization guidelines chemotherapy is the preferred treatment when surgery is not available, when complete removal of the cyst is impossible, when cyst contents threaten to disseminate due to cyst rupture, or when cysts are too numerous [8–11]. Also, Mawhorter et al. (1997) reported the use of percutaneous aspiration together with albendazole therapy for the treatment of recurrent pulmonary echinococcal infection in a patient [9]. We used albendazole treatment in one patient with recurrent hydatid cysts and in 63 patients who had multiple intrathoracic cysts or additional hepatic cysts.

The surgical procedures for hydatid cyst carry a morbidity of 0 to 13% and mortality of 0 to 5% [9]. Qian (1988) reported operative mortality of 0.6% in 807 patients [12]. Dogan et al. (1989) reported postoperative complications in 37 patients (3.5%) and 30-day mortality was 1.7% in 1,055 patients treated surgically for pulmonary hydatid disease [13]. Recently Thameur et al. (2000) reported operative mortality of 0.78% [10]. In our patients mortality was nil. We believe this situation may be due to our surgical experience and the young patient population.

In conclusion, procedures that conserve lung tissue are appropriate for most patients with pulmonary hydatid cysts. Radical procedures should be avoided, and surgery should preserve lung parenchyma to the maximum.

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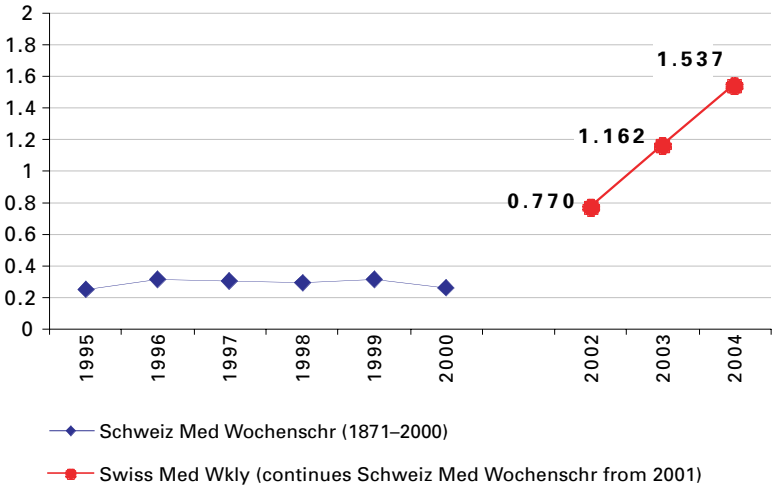

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