



Pitfalls in the diagnosis of complicated pulmonary hydatid disease

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The aim of this study was to present the pitfalls in the diagnosis of complicated pulmonary hydatid disease and to discuss the unusual radiological presentations of this endemic disorder in Turkey.

We retrospectively evaluated 34 patients (12 females) aged between 8 and 64 years, who were operated on at our centre between 1991 and 1998 and diagnosed with complicated pulmonary hydatid cyst histopathologically. Computerized tomography (CT) scans of these patients were reviewed double-blind by two radiologists. The patients were then divided into two groups: group 1: initial radiological impression is pulmonary hydatid cyst and group 2: initial radiological impression is not pulmonary hydatid disease. These two groups were evaluated in terms of symptoms, radiographical presentation and laboratory tests.

The presence of cystic appearance, water-lily sign, ring enhancement concomitant with intact cysts unanimously led the radiologists to the diagnosis of complicated pulmonary hydatid cyst, whereas solid appearance and presence of bronchial obliteration made the diagnosis unlikely. In such circumstances patient history, laboratory findings and bronchoscopic evaluation helped the diagnosis.

In conclusion, in endemic regions like Turkey, atypical radiological presentation of complicated pulmonary hydatid disease should be considered in the differential diagnosis of solid pulmonary lesions.

Key words: hydatid disease; lungs; radiology.

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Introduction

Hydatid disease is a parasitosis caused by *Echinococcus granulosus* and is prevalent in sheep and cattle raising areas in the Mediterranean region, South America, Australia and New Zealand (1). It is also endemic in Turkey (2). In humans, the liver is involved in 75% of cases, the lung in 15% and other anatomical locations in 10% (3). Although the presenting chest roentgenogram is almost always diagnostic, hydatid lung disease may present itself in a variety of radiological forms, especially if complicated. When suspected radiologically and/or clinically, in addition to the routine laboratory and serological tests, abdominal ultrasonography should be routinely performed to exclude accompanying cysts. Computed tomography (CT) best demonstrates cyst infection and cyst wall calcification. With atypical presentations, presence of solid lesion and bron-

chial obliteration on CT makes the diagnosis difficult for radiologists. In such circumstances patient history, laboratory findings and bronchoscopic evaluation may help the diagnosis. The aim of this study is to present the pitfalls in the diagnosis of complicated pulmonary hydatid disease and to discuss the unusual radiological presentations of this endemic disorder in Turkey.

Patients and methods

We retrospectively evaluated 34 patients (12 females, aged between 8 and 64 years) with pulmonary hydatid disease who were operated at our centre between 1991 and 1998. Clinical signs, laboratory results (including serology), invasive procedures if performed (bronchoscopy) and type of the operation were all reviewed from the charts. Appropriate CT slices were read double-blind by two experienced radiologists. The results were then compared for the presence of solid, semi-solid, cystic, cavitary lesions, ring enhancement, waterlily-sign, bronchial obliteration, air bubble sign, consolidation, daughter cyst, intact cyst and pulmonary parenchymal involvement. The location and number of cysts were also noted.

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The patients were then divided into two groups: group 1: initial radiological impression is pulmonary hydatid cyst and group 2: initial radiological impression is not pulmonary hydatid disease. These two groups were compared in terms of clinical presentation, radiographical presentation and laboratory tests. Echinococcal indirect haemagglutination test was accepted positive above the titration of 1/320.

Statistical analysis was performed by Fischer's exact test. Statistical significance was accepted for *P*-values above 0.05.

Results

Eighteen patients formed study group 1, including 11 men and seven women, with a mean age of 36.5 years (range 8–64 years). Group 2 consisted of 16 patients: 11 men and five women, with a mean age of 37 years (ranged 8–58 years).

Cough, haemoptysis, chest pain and sputum production were the most common symptoms in both groups but haemoptysis was statistically more common in Group 2. Expectoration of membrane and dyspnoea were other insignificant symptoms. Of 13 patients who underwent fiberoptic bronchoscopy, three patients (one in group 1 and two in group 2) were confirmed as hydatid cyst (yield: 23%). Multiple cysts were more common in group 1 (right lung: one of nine, left lung three of seven) compared to group 2 (none of the 6). The right lung was slightly more frequently involved in group 1 patients (55%) and the left lung for group 2 patients (56%).

Radiographical findings from the two groups are shown in Table 1. Cavitation, cystic lesion and air bubble sign were the most common radiological features in group 1, whereas bronchial obstruction, solid and semi-solid lesion, and consolidation were most common in group 2. Cystic appearance, ring enhancement and water-lily signs were statistically more common in group 1 but solid appearance and bronchial obstruction were more common in group 2. Three patients (33%) in group 1 and two patients (12.5%) in group 2 had concomitant liver cysts. Perforated and infected cysts constituted the majority of group 2 patients

(group 1: 53% vs. group 2: 72%). Cystectomy was the most common operation in two groups (77% vs. 63%). Wedge resection (6% vs. 18%), or lobectomy (6% vs. 12%) were performed more in group 2 than group 1.

Positivity of laboratory findings (*Echinococcus* haemagglutination test, blood eosinophilia) did not differ significantly between groups 1 and 2 (80% and 70%, respectively).

Discussion

Hydatid disease is endemic in some parts of the world, including Turkey (1,2). It is still an important public health problem, especially in rural areas of Turkey (2). The liver and lungs are most frequently involved organs (3) and the incidence of simultaneous liver and lung involvement has been reported to be between 5.8 and 13.3% according to various reports (4–7). Radiological techniques almost always help the diagnosis, especially if the cyst is intact and uncomplicated, and help to localize the extent of the disease if the cyst is perforated, complicated and difficult to diagnose.

More than 90% of our patients presented with a variety of symptoms, including cough, chest pain, haemoptysis, malaise, fever and even expectoration of cyst material. Of 16 patients in group 2, expectoration of cyst products (19%) confirmed the diagnosis in three patients. Blood eosinophilia may not always be present. Serological tests may help to confirm the diagnosis but a negative serology does not necessarily exclude the diagnosis of cystid hydatid disease. In previous reports, a low positive diagnostic yield has been reported for serological tests (7–10). Haemoptysis was statistically more common in group 2, probably due to the increased number of perforated and infected cysts in this group. In patients presenting with haemoptysis malignancy should be ruled out. There are studies reporting the diagnostic role of bronchoscopy in hydatid disease (11). In our series three patients were diagnosed with hydatid cyst by fiberoptic bronchoscopy.

TABLE 1. Roentgenographical findings

Finding	Group 1	%	Group 2	%	Total	%	<i>P</i> -value
Solid	0	0	5	31	5	15	0.01
Semi-solid	4	22	7	44	11	32	>0.05
Cystic	11	61	2	13	13	38	<0.01*
Cavitation	12	67	7	44	19	56	>0.05
Air bubble	10	56	7	44	17	50	>0.05
Bronchial obstruction	3	17	9	56	12	35	<0.05*
Ring enhancement	9	50	2	13	11	32	<0.05*
Water-lily sign	9	50	2	13	11	32	<0.05*
Consolidation	5	28	7	44	12	35	>0.05
Pulmonary involment	4	22	4	25	8	24	>0.05
Daughter cyst	3	17	0	0	3	9	>0.05
Intact cyst	5	28	0	0	5	15	<0.05*

Surgery is indicated in every symptomatic and/or enlarging or infected cysts. Simultaneous lung and liver cysts should be operated on using a single stage approach (6,12). Bilateral lung hydatid cysts may be managed in consecutive operations. The most frequent complication of pulmonary hydatid cyst is perforation because of degeneration in the wall of the cyst (13). The most serious usual complication of rupture is bacterial infection (4,13). In this series, perforated and infected cysts were more commonly observed in group 2 compared to group 1 (72 vs. 53%), making radiological confirmation difficult. Both rupture and infection can change the appearance of the cyst on routine roentgenograms. In such circumstances CT and fiberoptic bronchoscopy help to reach the final diagnosis. Lewall and McCorkell (14) classified these ruptures into three types: contained, communicating, direct according to the type of rupture and radiological appearance. Diagnostic difficulties related with complicated cysts have been previously reported (15–20). In our study, blinded radiologists had no difficulty in diagnosing intact simple cysts concomitant with complicated ones. Cavitation, cystic lesion and air bubble sign were the most common radiological features in group 1, whereas bronchial obstruction, semi-solid lesion and consolidation were most common in group 2. Simple cysts, cystic appearance, ring enhancement and water-lily signs were statistically more common in group 1, but solid appearance and bronchial obstruction were more common in group 2. Kervancıoğlu *et al.* suggested that ring enhancement and air bubbles are strongly indicative of pulmonary hydatid disease (19). Kokturk *et al.* suggested that the air bubble sign is valuable in the diagnosis of complicated hydatid cyst (20). In our study the air bubble sign was found in 56 % and 44% of the patients in groups 1 and 2 respectively.

In conclusion, clinical picture and typical radiographical appearance of hydatid disease in pulmonary and extra-pulmonary organs are diagnostic for hydatid disease in most occasions, but atypical radiographical presentations of complicated pulmonary hydatid disease (e.g. solid opacification, bronchial obliteration) should always be considered in the differential diagnosis of solid lesions for a prompt diagnosis, especially in endemic regions like Turkey.

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