ORIGINAL INVESTIGATION

Diagnostic Dilemma in Hydatid Cysts: Tumor-Mimicking Hydatid Cysts

Ufuk Çobanoğlu¹, Selvi Aşker², Duygu Mergan¹, Fuat Sayır¹, Salim Bilici³, Mehmet Melek³¹Department of Chest Surgery, Yüzüncü Yıl University Faculty of Medicine, Van, Turkey²Department of Chest Diseases, Yüzüncü Yıl University Faculty of Medicine, Van, Turkey³Department of Pediatric Surgery, Yüzüncü Yıl University Faculty of Medicine, Van, Turkey

Abstract

OBJECTIVES: Hydatid cysts are sometimes confused with different pathologies, and problems arise in their diagnosis and treatment. In this study, cases that are followed up with a diagnosis of lung malignancy and that are detected to have hydatid cysts were retrospectively examined.

MATERIAL AND METHODS: Seven patients with hydatid cysts whose clinical and radiological features were consistent with lung malignancy were retrospectively examined between 2010 and 2014 regarding sex, age, symptoms, diagnostic methods, surgical procedures performed, and postoperative complications. In the diagnosis of the patients, radiological diagnostic methods such as chest radiography, thoracic computed tomography (TCT), and positron emission tomography+computed tomography (PET-CT) as well as invasive diagnostic methods such as bronchoscopy, fine-needle aspiration biopsy, thoracentesis, and video-assisted thoracoscopic surgery were used.

RESULTS: The average diameter of the lesions was determined as 4.14 ± 1.57 cm in TCT. Maximum standardized uptake value (SUV max) was measured as 8.77 ± 3.41 (5.4–15.1) in the PET-CT analysis. Bronchoscopy, fine-needle aspiration biopsy, and thoracentesis yielded no results. Definitive diagnosis was established by performing thoracotomy and video-assisted thoracoscopic surgery.

CONCLUSION: Pulmonary hydatid cysts can appear as malignant diseases such as lung cancer as well as infectious pathologies such as tuberculosis or benign pathologies. Radiologically, it should be kept in mind that pulmonary hydatid cysts can mimic many pulmonary pathologies, particularly malignancies. Necessary examinations towards its differential diagnosis must be performed in the preoperative period.

KEYWORDS: Lung, hydatid cyst, malignancy, diagnosis

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INTRODUCTION

The *Echinococcus granulosus* causes hydatid cyst disease by forming cystic lesions in the organ where it is located. Intact cysts do not create obvious symptoms until they reach a certain size and pressure surrounding structures. Many studies reveal that 30% of hydatid cyst cases located in the lung are asymptomatic [1]. However, when cysts reach large sizes, they cause symptoms by pressuring surrounding vital organs (esophagus, heart, trachea, large veins, etc.) [1,2]. This pressure caused by intact cysts and complications that may arise because of perforated cysts cause hydatid cysts, which are defined as benign diseases, to progress with a high morbidity and mortality [2,3].

Hydatid cysts can be diagnosed by various imaging methods such as direct chest radiography and thoracic computed tomography (TCT) and can be treated with surgeries that are not very complicated, such as cystotomy and capitonnage [4,5].

Certain instances that develop unusually in hydatid cysts cause difficulties in the differential diagnosis with other pathologies and lead to problems in diagnosis and treatment [5]. In particular, the location of hydatid cysts outside the expected localizations leads to different clinical pictures [5]. Furthermore, displaying images other than the radiologically expected homogenous, round or oval, well demarcated lesions, and lesions that are surrounded by normal lung tissues leads to confusion in diagnosis [5,6].

In this study, cases that were initially judged as lung malignancies but detected as hydatid cysts in the definitive diagnosis were retrospectively examined.

This study was presented as a poster in the 7th National Hydatidology Congress (September 4–7, 2014, Ordu, Turkey).

Address for Correspondence: Ufuk Çobanoğlu, Yüzüncü Yıl Üniversitesi Tıp Fakültesi, Göğüs Cerrahisi Anabilim Dalı, Van, Türkiye Phone: +90 432 215 04 75 E-mail: drucobanoglu@hotmail.com

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MATERIAL AND METHODS

In this study, seven patients with hydatid cysts whose clinical and radiological features were consistent with lung malignancy were retrospectively examined with respect to sex, age, symptoms, diagnostic methods, surgical procedures performed, and postoperative complications. In the diagnosis of the patients, radiological diagnostic methods such as chest radiography, TCT, and positron emission tomography+computed tomography (PET-CT) as well as invasive diagnostic methods such as bronchoscopy, fine-needle aspiration biopsy, thoracentesis, and video-assisted thoracoscopic surgery were used.

The patients diagnosed with hydatid cysts were started on a postoperative 14-day, 3-course albendazole treatment with a dose of 10 mg/kg, and they were followed up in the first 3 months with chest radiography and biochemical blood tests, including hemogram and liver enzymes. Detailed patient consent forms were obtained, and the results of the cases were discussed in light of the literature.

RESULTS

Seventy-one cases underwent surgery in our clinic between 2010 and 2014 because of pulmonary hydatid cysts. Among these cases, 7 (9.85%) hydatid cyst cases consistent with lung malignancy were detected. In addition, 85.71% of these cases were male, and the average age was 53.85 ± 12.95 years. The most commonly observed symptom was chest pain (57.14%) (Table 1).

Upon detecting a mass image in the direct chest radiographies of patients, they were evaluated first by TCT and then by PET-CT. The lesions were most frequently observed in the lower right lobe, and bilateral multiple lesions were observed in one (14.28%) case (Table 1) (Figure 1, 2, 3, 4). The average diameter of lesions was measured as 4.14 ± 1.57 cm. The average SUV max. (avg $\pm$ SD) value of cases was measured as 8.77 ± 3.41 (5.4–15.1) as a result of PET-CT. In two (28.75%) cases, accompanying pleural effusion was detected in the same side as the mass. In cases with effusion, cytological examination was performed on the pleural fluid obtained by thoracentesis, whereas in the other two (28.57%) patients that displayed a pleural-based mass image, transthoracic aspiration biopsy was preferred. All cases were evaluated by bronchoscopy (Table 1). However, findings that would give a definitive diagnosis could not be obtained by invasive diagnostic interventions. Mass image was not detected in the accompanying organ in any of the cases.

In cases in which open surgery was preferred because of the localization and the diameter of the lesion, the lesion was determined as a hydatid cyst and cystotomy+capitonnage was performed. On the other hand, in patients undergoing minimal invasive intervention, wedge resection was performed in the pathological area, and the frozen result was reported as a hydatid cyst (Table 1).

DISCUSSION

Hydatid cysts, which are an infestation caused by echinococci, frequently arise in areas where stockbreeding is performed [7]. Its contamination route to humans is via foods that are contaminated with the droppings of animals such as dog, wolf, jackal, dingo, or direct contact [8].

Table 1. Demographic, clinical, and radiological features of the cases and treatment methods

Parameters	Features	Number (%)
Sex	Female	1 (14.28)
	Male	6 (85.71)
Age (years)	20–30	2 (28.57)
	31–40	1 (14.28)
	41–50	2 (28.57)
	≥ 51	2 (28.57)
Symptom	Chest pain	4 (57.14)
	Dyspnea	1 (14.28)
	Cough	3 (42.85)
	Weight loss	1 (14.28)
	Asymptomatic	2 (28.57)
Number of lesions	Multiple (bilateral)	1 (14.28)
	Single-solitary	6 (85.71)
Lesion localization	Upper right lobe	3 (42.85)
	Middle right lobe	1 (14.28)
	Lower right lobe	4 (57.14)
	Upper left lobe	2 (28.57)
	Lower left lobe	1 (14.28)
Radiological definition of the lesion	Regularly demarcated multiple nodular lesion	1 (14.28)
	Irregularly demarcated, centrally located, infiltrative lesion	4 (57.14)
	Irregularly demarcated, peripherally located nodular lesion	2 (28.57)
Surgical procedure	VATS (Wedge resection)	5 (71.42)
	Thoracotomy (Cystotomy+capitonnage)	2 (28.57)

Figure 1. Irregularly demarcated mass image on the upper right lobe, surrounded by ground glass appearance

Figure 2. Irregularly demarcated mass image on the upper right lobe

Figure 3. Bilateral mass images

Figure 4. Irregularly demarcated mass image on the middle lobe

The initial period of cyst formation is generally asymptomatic, and they do not display any symptoms until they reach a diameter of approximately 5 cm [9]. Symptoms arise with the pressure on neighboring organs or the development of complications [9]. Complaints such as chest pain, coughing, and hemoptysis arise in patients depending on the localization of the hydatid cyst on the lung and the diameter of the lesion, while systemic symptoms such as fever, fatigue, and weight loss are not seen unless capsules are ruptured and cyst contents are opened to the bronchus or intrapleural cav-

ity [5,7]. In our study, the most commonly observed symptom was chest pain, and 28.57% of the cases were asymptomatic.

Serological methods, which are reported with 90% positive results in liver cysts and with 50% positive results in pulmonary hydatid cysts, are used in the diagnosis of the disease. Currently, the most preferred methods are indirect hemagglutination (IHA), indirect fluorescent antibody (IFA), and enzyme-linked immunosorbent assay (ELISA) [5,7,10]. In our cases, serological diagnosis methods are not preferred because hydatid cysts were not considered in the preoperative differential diagnosis.

Because specific clinical symptoms and laboratory findings are absent in hydatid cysts, the use of radiological evaluations in diagnosis is highlighted [5,7]. If hydatid cysts, which are normally observed as well demarcated, round or oval, homogenous lesions, rupture, special radiological findings can be obtained. There is an air-fluid level, sign of meniscus, air crescent sign, water-lily appearance, and appearance of mass or membrane in the cavity [11]. However, what disrupts the nature of the lesion and what makes the radiological appearance a complicated puzzle are consolidation development in the lesion vicinity, occurrence of inflammatory reaction, and cyst becoming a heterogeneous structure [11]. In our cases, the appearances of the nodules were not typical for hydatid cysts. Their borders were irregular, their walls were thickened, and their density was heterogeneous and high.

The homogenous opacity appearance of hydatid cysts in the radiological examinations can lead to erroneous evaluations in the differential diagnosis by giving an image of pulmonary nodules. Many lesions with malignant characteristics, mostly sarcomas, can display a similar image. They can be confused with metastatic lung disease, particularly in the presence of multiple cysts, as seen in our case [12].

Although cystic formations can be identified by chest radiography to a great extent, these images are not pathognomonic. They can mimic many lesions varying in a scale from pulmonary hematoma, fungus ball and benign tumors that are settled in the tuberculosis cavity, to carcinomas [13,14]. In that case, the density of the fluid that the hydatid cyst contains, the internal structure of perforated or complicated hydatid cysts, the collapsed membrane inside the perforated cyst fluid, and the daughter vesicles inside the main cyst that cannot be seen with conventional radiographic methods can be shown by TCT [15,16].

The cyst density is around 3–18 HU in TCT. However, if the cyst is complicated, the density can climb above 20 HU [17]. In ruptured hydatid cysts, infiltration and infection occurs in the lung tissue surrounding the lesion, which raises the density, and in these cases, hydatid cysts can be confused with solid neoplasms, which give the appearance of a soft tissue [18]. In our cases, because of the infiltrations around the cysts, a soft tissue appearance and an increase in its density are detected in TCT, and a diagnosis of hydatid cyst is disregarded.

If TCT is insufficient in distinguishing malignant and benign lesions detected with conventional radiological images, PET-CT, bronchoscopy, and fine-needle aspiration biopsy can be used in differential diagnosis.

PET-CT is an important diagnostic method in differential diagnosis, and it is preferred in differentiating multiple pulmonary nodules with various characteristics, including inflammatory diseases such as echinococcosis [19].

The probability of malignancy is low for nodules that do not retain FDG in PET-CT. FDG retention increases in malignant pulmonary nodules in parallel with the increase in glucose metabolism, and the SUV max value above 2.5–3.0 is accepted as sensitive and specific regarding malignancy [20]. However, even apart from tumor cells, given that FDG retention will increase in all infections, inflammations and infestations that include active macrophages, particularly in granulomatous diseases, yield false-positive results (10%–25%) [20]. Tuberculosis, sarcoidosis, coccidiomycosis, aspergillosis, and parasitic diseases are the most frequent pathologies among the ones that yield the most false-positive results in PET-CT [20,21]. This shows that FDG-PET is not very effective regarding positive predictive values and that histological diagnosis is needed in every nodule that shows FDG retention [22].

There are many studies in the literature that support the foregoing explanations. For instance, Kurt et al. [23] later reported that the high FDG retention detected lesions as hydatid cysts, which they initially evaluated as metastasis in female patients in whom they performed PET-CT for breast cancer staging. In another study, Rangarajan et al. [24] detected, in the postoperative histopathological examination, that the mass lesion that showed hypermetabolic involvement in the lung in the PET-CT of 40-year-old male patients is a ruptured, infected, and consolidated hydatid cyst. Demir et al. [25] reported similar findings in their presentations. In our study, similar to the literature, the SUV max value is detected as high in hydatid lesions, and our cases are taken into operation with a provisional diagnosis of malignancy.

Another diagnostic intervention that can be performed for the diagnosis of pulmonary lesions is bronchoscopy. Although bronchoscopy is performed in all our patients, hydatid cyst diagnosis was not reached. Kılınc et al. [26] saw the cystic membrane in the bronchoscopic biopsy performed on a pulmonary mass that they interpreted as a malignant lesion and established that the lesion is a hydatid cyst. In two separate studies, Tor et al. [27] and Saygı et al. [28] performed cytological and histopathological examinations on the lesions that they reached via fiber optic bronchoscopy and reached the hydatid cyst diagnosis.

In fact, observing protoscoleces or their hooks in the microscopic examination of the hydatid cyst fluid aspirate is a definitive diagnostic method [29]. However, because of its possibility of causing anaphylaxis and dissemination, it is accepted as contraindicated. However, there are studies in which these processes are performed because of considering different provisional diagnoses and in which it was reported that the hydatid cyst diagnosis was established [29]. Differing

from this literature, in our study, transthoracic aspiration biopsy that was performed on two of our cases that displayed pleural-based mass image was reported as non-diagnostic. This can be explained by the fact that cysts, particularly ones that are pulmonary located, can be non-fertile in humans.

Mistaking pulmonary hydatid cysts with pulmonary malignancies is a common diagnostic error. However, mistaking pulmonary malignancies with hydatid cysts is very unusual. Lung tumors can sometimes display cystic formations, and in some of these malignant masses, anti-echinococcal antibody levels can, albeit rarely, get elevated, similar to hydatid cysts [30]. In a study that supports this literature finding, Singh et al. [31] reported a large-cell type lung cancer case that displayed a positive serological test for *Echinococcus granulosus* and that radiologically mimicked the pulmonary hydatid cyst. Forming a hydatid cyst image in a malignant lesion in the lung is caused by necrosis and exfoliation of the nucleus part that is at the center of the tumor, by proximal tumors causing air trapping in distal with check-valve effect and malignant cell formation settling in the air pocket created here, or by malignant cell infiltration into the wall of a pre-existing benign bullous lesion [32].

In conclusion, radiologically, it should be kept in mind that pulmonary hydatid cysts can mimic many pulmonary pathologies, particularly malignancies, and this should be considered during differential diagnosis, particularly in endemic regions.

Ethics Committee Approval: We have not received ethical approval because we had detailed consent from all patients including small number of cases of this article (The article contains the 7 cases).

Informed Consent: Written informed consent was obtained from patients who participated in this study.

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