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Secondary Pleural Hydatidosis as an Unexpected Cause of Fever of Unknown Origin

Orijini Bilinmeyen Ateşin Beklenmedik bir Nedeni Olarak; Sekonder Plevral Hidatidoz

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Abstract

Fever of unknown origin (FUO) and hydatid cysts are both complex processes that require a lengthy diagnosis. Patients hospitalized with equivocal pyrexia and pleural effusion in endemic areas should keep this in mind. Hydatid cysts are a cause of FUO. Therefore, we present a rare case. Our patient was presented with an intermittent fever and cough. His left lung's middle and lower zones had reduced respiration noises. The hemogram revealed leukocytosis with neutrophil predominance and normochromic normocytic anemia. The levels of ALT, AST, CRP, and procalcitonin were all elevated. There was increased opacity in the appearance of an abscess on a chest X-ray, as well as pleural effusion. On CT, a 10x9.3 cm pleural-based cystic structure containing an air-fluid level was observed. Thoracentesis fluid was exudative, neutrophil-dominated, and empyema. The indirect hemagglutination test was positive at a 1/2560 titer. Pleural fluid cytology revealed a cyst-hydatid membrane, and the patient was diagnosed with secondary pleural hydatosis.

Keywords: *Fever, Hydatid cyst, Pleural effusion.*

Öz

Orijini bilinmeyen ateş ve kist hidatik klinisyenler için zorlayıcı bir süreçtir ve genelde uzun bir tanı süresi gerektirir. Kist hidatik prevalansı evrensel olarak azalsa da, endemik yerlerde belirsiz ateş ve plevral efüzyonla başvuran hastalarda akılda tutulmalıdır. Kist hidatik, orijini bilinmeyen ateşin bir nedeni olabilir. Bu yüzden çok nadir bir olguyu sunuyoruz. Hastamız bir aydır olan aralıklı ateş ve balgamsız öksürük şikâyetiyle başvurdu. Sol akciğer orta ve alt zonda solunum sesleri azalmıştı. Hemogramında; nötrofil baskınlığı olan lökositozu, normokromik normositik anemisi mevcuttu. ALT, AST, CRP ve prokalsitonin normalin üzerindeydi. Akciğer grafisinde plevral efüzyonun eşlik ettiği apse görünümünde opasite artışı mevcuttu. BT görüntülerinde 10x9.3 cm boyutlarında hava-sıvı seviyesi içeren plevra tabanlı kistik yapı görüldü. Torasentez mayii eksüdatif vasıftaydı. Nötrofil ağırlıklı, ampiyemli sıvıydı. İndirekt hemaglütinasyon testi 1/2560 titrede pozitifti. Plevral sıvı sitolojisinde kist hidatide ait zar görüldü, hastaya sekonder plevral hidatoz tanısı konuldu.

Anahtar Kelimeler: *Ateş, Kist hidatik, Plevral efüzyon.*

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Fever of unknown origin (FUO) is defined as a temperature of more than 38.3 °C on multiple occasions lasting more than three weeks with no diagnosis despite one week of hospital investigations or three outpatient visits (1). The true incidence and prevalence of FUO are unclear. Approximately 3% of hospital admissions are due to FUO. According to a study conducted at a Japanese university hospital, FUO occurred in 153 of 5,245 (2.9%) hospitalized patients. Another study from a community hospital in the United States found that FUO was the cause of one out of every 73 infectious disease consultations (2).

Cystic hydatid disease, which can be seen in many parts of the world, is a zoonotic infection, and its incidence ranges from 1/20000 to 1/50000 in Turkey. Although four different species can cause disease in humans, *Echinococcus granulosus* is responsible for 95% of reported hydatid cases. Humans themselves, regarded as incidental intermediate hosts, have no role in their biological life cycle. They are usually infected after inadvertent ingestion of Echinococcus eggs found in canine feces. First, larval cysts that form in the liver via the portal vein can spread throughout the body via systemic circulation.

The liver is the most commonly affected organ, accounting for 54% of cases, followed by the lungs (35%) (3). Intrathoracic extrapulmonary hydatid cysts are extremely uncommon (7.4%), with pleural hydatid disease being the most prevalent site of involvement. Patients may also have infections of the mediastinum, pericardium, and the chest wall.

There are two types of pleural hydatid disease: primary and secondary. Pleural involvement in hydatid illness occurs largely by direct larval infestation of the pleura via the hematogenous or lymphatic route or secondarily through the contents of the cyst across the pleura because of a pulmonary or hepatic neighboring cyst rupture.

Infections are the most common cause of FUO, accounting for 37% of all cases. To the best of our knowledge, hydatid illness has not been reported as a cause of FUO, although a few cases have. Therefore, we present to you this rare case.

CASE

A 17-year-old male presented to the emergency department complaining of intermittent fever, a nonproductive cough, and a 1-month-long chest pain. When the patient's medical history was thoroughly investigated, it was discovered that he was a high school student, that he had lived in Ağri (one of Turkey's rural cities) until a year ago, that he had no other diseases, that he fed dogs, and that his family was involved in farming. No specific feature was taken in his spare time activities. The patient's vital signs were stable during the physical examination, with a pulse rate of 110 beats per minute, an oxygen saturation of 97%, a blood pressure of 110/65 mmHg, and a temperature of 37.1°C. Thoracic vibrations were reduced, as were breathing noises in the middle and lower zones of the left lung. Other organs were examined and no pathological symptoms were discovered.

His full blood count revealed leukocytosis with neutrophil predominance (WBC: 13.45 x 10(-3)/L) and normochromic normocytic anemia (HG: 11 g/fL, MCV: 80.8 fL). The results were as follows: ALT 319 U/L, AST 339 U/L, total bilirubin 0.57 mg/dl, CRP 192 mg/L, procalcitonin 1.47 ng/ml, sedimentation 33 mm/hour, and total IgE 218 IU/L. On his chest X-ray, there was increased opacity, which indicates abscess formation with pleural effusion. On the left side of the lung, diaphragm contours were erased. Cavity walls are not precisely measured and have an air-fluid level within (Figure 1). An exudative pleural effusion was discovered during a diagnostic thoracentesis. It was a neutrophil-predominant fluid with ampyematous effusion with a pH less than 7.1, LDH of 700, glucose of 40, total protein of 7, and albumin of 4.2. A thoracic tube was used, as well as closed underwater drainage. Sultamisilin IV and clindamycin were administered as medical treatments. A pleural-based cystic structure measuring 10x9.3 cm in size was discovered on a CT scan (computerized tomography) (Figure 2). In repeated blood cultures, no infective focus or signal was found. The indirect hemagglutination test yielded a positive result with a titer of 1/2560. An abdominal ultrasound was performed to rule out liver cyst hytatid disease due to his elevated liver function test on admission. In pleural fluid culture, no microorganisms grew. Acid-fast bacilli samples were likewise negative. Pleural fluid cytology indicated a cyst hydatid membrane, and the patient was diagnosed with secondary pleural hydatosis as a result of a pulmonary cyst rupture. Thoracic surgeons performed therapeutic video-assisted thoracoscopy (VATS). Postoperatively, the antihelmintic drug albendazole was administered (Figure 3).

DISCUSSION

Pleural hydatosis is the most prevalent manifestation of intrathoracic extrapulmonary hydatid disease, accounting for 53–72% of cases (4).

Although cough, fever, and chest pain are the most common symptoms of lung cyst hydatid disease, studies show that 20–30% of patients may be asymptomatic. Since they are asymptomatic, uncomplicated hydatid cysts are usually discovered by chance. Cysts grow more quickly and show symptoms earlier in organs with weak connective tissues, such as the lungs and brain. At the time of admission, 80% of the lung hydatid cysts were symptomatic. Patients can be diagnosed with various symptoms,

such as hemoptysis, respiratory distress, or anaphylaxis, based on the location, rupture, or advancement of cysts (5).

Expectoration of cyst fluid and membranes is a key diagnostic finding. A microscopic examination of the cyst fluid and the finding of the parasite scolexs is pathognomonic. Routine laboratory tests can help guide diagnosis, but there is no disease-specific test. The total blood count can reveal eosinophilia. In our case, however, we found no eosinophilia or elevated IgE levels. Serological diagnostic tests include ELISA, indirect hemagglutination, indirect fluorescent antibody, and latex agglutination. Because of its high sensitivity, ELISA is frequently used for postoperative monitoring. A titer of 1/160 or greater is considered positive in the IHA test. In our situation, the IHA test was positive at a titer of 1/2560.

The radiography of lung hydatid cysts can vary depending on the cyst wall's integrity. Intact cysts are spherical, homogeneous, and have a well-defined density. Secondary pleural hydatosis should be considered in the differential diagnosis of well-defined nodular lesions in endemic areas. Ruptured cysts may signal an elevated air-fluid level, the meniscus sign, or the water lily flower sign, all of which are symptoms of hydatid illness, pneumothorax, pleural effusion, or empyema. As a result, radiologic characteristics differ after cyst rupture into the pleural cavity, and clinicians should be aware of this when managing patients.

A hydropneumothorax occurs when a pneumothorax and pleural effusion coexist in the pleural space, which is commonly shown as a gas or fluid level on an upright screen. On the supine screen, a sharp pleural line is bordered by increasing opacity lateral to it within the pleural space, which may occasionally suggest the diagnosis.

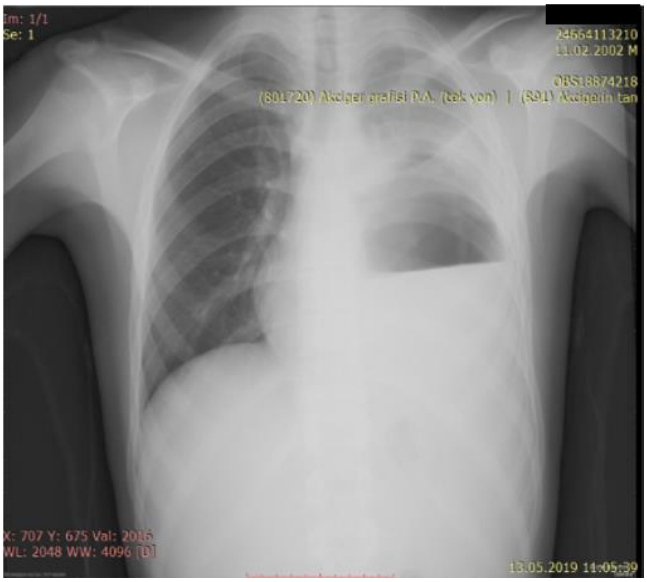

Figure 1: *Chest X-ray on admission*

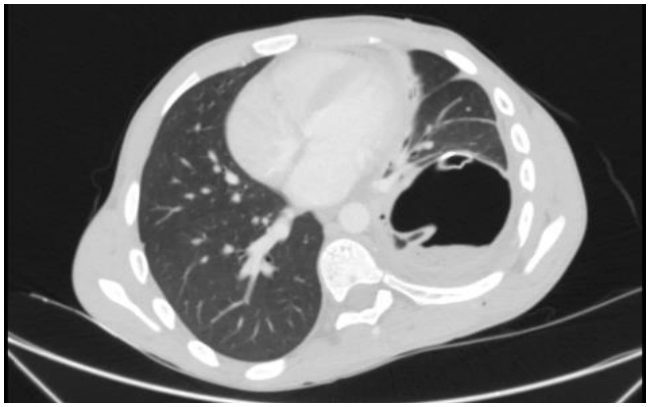

Figure 2: *CT-scan of the patient*

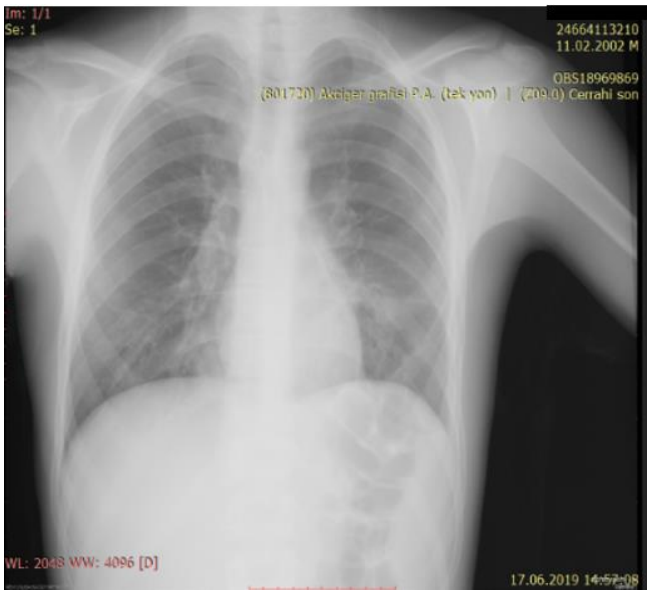

Figure 3: *Chest X-ray 1 month later*

When hydatid cysts in the lung extend into the bronchi or pleural space, they are classified as complex cysts. In complex cases, inflammation interferes with wound healing and can lead to consequences such as persistent air leakage, empyema, and pneumonia. In our case, we started treatment for empyema first, and the fact that hydatid cyst disease was a possible cause made it easier to figure out what was wrong.

Surgery is the initial treatment option, whether the lesion is symptomatic, asymptomatic, or complicated but intact. The procedure's purpose is to enucleate the lesion completely, avoid cyst perforation to prevent recurrence, and resect as much lung tissue surrounding the cyst as possible to maintain an acceptable breathing capacity. Some individuals may benefit from video-assisted thoracoscopic surgery instead of a thoracotomy. Decortication, segmentectomy, or lobectomy may be required, especially in complex cysts. As a result, open surgical intervention in pleural hydatid disease is strongly advised. Intraoperative cyst therapy can be radical, such as segmentectomy, lobectomy, and pneumonectomy, or conservative, such as cystectomy and cyst enucleation. Although extensive

resection of the germinative membrane and pericysts is the gold standard treatment method, in many experienced institutions, a simple cystectomy procedure produces the same satisfying results as a radical approach. The patient was operated on using VATS because our hospital's thoracic surgeons are experienced in the enucleation of hydatid cysts. During the patient's 12-month follow-up, no late problems or recurrences were observed.

CONCLUSION

For doctors, FUO is regarded as a persuasive process that necessitates a lengthy diagnostic period. The same is true for hydatid cyst disease if it is not included in the differential diagnosis while the condition is being investigated. In developed countries, hydatid cyst disease may be uncommon. Although its global prevalence has declined, it remains a public health risk in some regions, particularly those with ineffective veterinary control and insufficient sanitary conditions. In such endemic areas, patients admitted with unclear severity pyrexia and radiologically confirmed pleural effusion or pyopneumothorax should be evaluated for pleural hydatid disease in addition to normal hydatid cyst characteristics.

CONFLICTS OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

Concept - K.K., B.K., E.A., H.A., F.T.A., S.T.Ö.; Planning and Design - K.K., B.K., E.A., H.A., F.T.A., S.T.Ö.; Supervision - K.K., B.K., E.A., H.A., F.T.A., S.T.Ö.; Funding - H.A., F.T.A.; Materials - F.T.A., E.A.; Data Collection and/or Processing - K.K., B.K., H.A., S.T.Ö.; Analysis and/or Interpretation - B.K., S.T.Ö, K.K.; Literature Review - K.K., E.A..; Writing - K.K., B.K., E.A., S.T.Ö.; Critical Review - K.K., S.T.Ö.

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