Türk Pediatri Arşivi
Turkish Archives of Pediatrics

Case Report

Pulmonary hydatid cyst disease mimicking necrotizing pneumonia in a child with leukocytoclastic vasculitis

Sakine Işık[1], Şule Çağlayan Sözmen[1], Eren Güzeloğlu[2], Tülay Öztürk[3], Suna Asilsoy[1]

[1]Department of Pediatrics, Division of Pediatric Allergy-Immunology, Dokuz Eylül University School of Medicine, İzmir, Turkey
[2]Department of Pediatrics, Dokuz Eylül University School of Medicine, İzmir, Turkey
[3]Division of Pediatric Radiology, Dokuz Eylül University School of Medicine, İzmir, Turkey

Cite this article as: Işık S, Çağlayan Sözmen Ş, Güzeloğlu E, Öztürk T, Asilsoy S. Pulmonary hydatid cyst disease mimicking necrotizing pneumonia in a child with leukocytoclastic vasculitis: a case report. Turk Pediatri Ars 2018; 53: 117-9.

Abstract

The diagnosis and management of pulmonary hydatid cyst disease represents an important clinical problem in countries of the world that are endemic to echinococcal infection. Atypical clinical and radiologic findings including multiple cavitary lesions in the lung and pleural effusion may lead to misdiagnosis or delay in diagnosis in these patients. We report a patient who was followed up in our hospital with rashes and, clinical and radiologic findings of necrotizing pneumonia in whom there was no response to broad spectrum antibiotherapy. Lung computed tomography showed multiple thick-walled cystic lesions and pleural effusion and the diagnosis of pulmonary hydatid cyst disease was confirmed by surgical and serologic examinations. Antibiotic treatment was changed to albendazole 10 mg/kg/day. There was no liver involvement in terms of cyst hydatid disease with ultrasonographic examination. Skin biopsy showed leukocytoclastic vasculitis. Complete clinical and radiologic improvement was achieved in three months and albendazole treatment lasted six months.

Keywords: Child, hydatid cyst, leukocytoclastic vasculitis, lung, necrotizing pneumonia

Introduction

In hydatid cyst disease, the most commonly involved organ is the lung in children (1, 2). Many patients with intact lung cysts are asymptomatic. Intact lung cysts are found incidentally with their typical radiologic appearance on lung radiography. Infections and perforation may lead to misdiagnosis and delays in treatment by altering the radiographic appearance of hydatid cysts (3). Herein, a pediatric patient with a rash in whom the diagnosis of hydatid cyst was not considered initially because of extraordinary radiologic and clinical findings including multiple cystic lesions with thick walls and pleural effusion is presented.

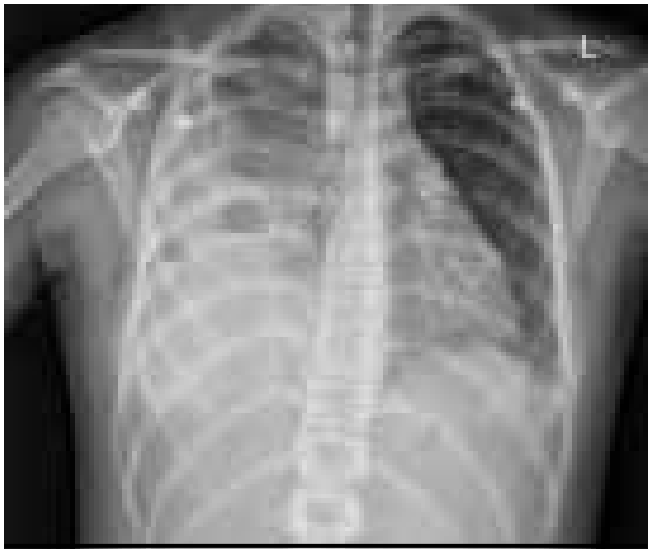
Case

A 10-year-old girl who had no previous history of illness presented to our hospital with symptoms of cough, high temperature, rash, and gradually increasing respiratory distress, which had been continuing for four days. A physical examination revealed fever, tachypnea, reduced respiratory sounds in the right hemithorax compared with the left hemithorax, hypoxia, and numerous palpable purpurae. Complete blood count revealed a white blood cell count of 41,300/mm^3 and a peripheral smear was as follows: granulocytes 92%, eosinophils 1%, and lympho-monocytic cells 7%. The erythrocyte sedimentation rate was found as 115 mm/h and C-reactive protein was found 373 mg/dL. Lung radiography revealed numerous cavitary lesions and pleural effusion in the right hemithorax (Picture 1). The pleural fluid was found to be compatible with exudate. Intravenous ceftriaxone and vancomycin treatment was initiated with a prediagnosis of rapidly progressing necrotizing pneumonia. A purified protein derivative (PPD) test, which was performed in terms of tuberculosis, was

Address for Correspondence: Sakine Işık E-mail: drsakinekar83@hotmail.com
Received: 13.12.2015 **Accepted:** 29.08.2016

DOI: 10.5152/TurkPediatriArs.2018.3670

found to be negative. Bacterial growth was not found in a culture of the pleural fluid, which was performed in terms of aerobic/anaerobic bacteria, and polymerase chain reaction (PCR) for M. tuberculosis was negative. Computed tomography (CT) of the lung revealed cystic lesions with thick walls in both hemithoraces and consolidation and pleural effusion in the right hemithorax (Picture 2). Thoracotomy was performed because the clinical and radiologic response to treatment was inadequate. The cystic lesions, which were perforated, were repaired with cystectomy and the empyema was drained. An enzyme-linked immunosorbent assay (ELISA) performed in terms of echinococcus infection was found to be positive (1/640). Antibiotic treatment was switched to albendazole at a dose of 10 mg/kg/day. Involvement of the liver with hydatid cysts was not found on abdominal ultrasonography. A skin biopsy from skin lesions, which persisted for longer than 24 hours, was found to be compatible with leukocytoclastic vasculitis. Complete clinical and radiologic recovery of the patient occurred in three months and albendazole treatment was continued for six months. Written informed consent was obtained from the patient's family.



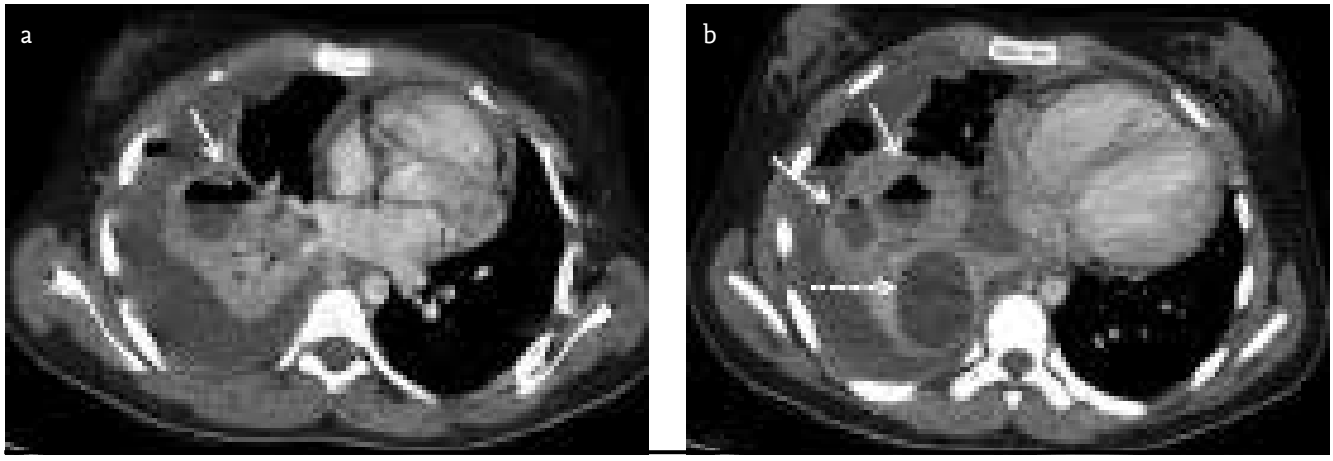
Picture 1. Cavitary lesions with air-fluid level are observed in the right hemithorax on lung radigraphy. In addition, pleural effusion and an area of consolidation is seen in the right lower-middle zone

Discussion

Pulmonary hydatid cyst disease is still an important clinical problem in countries where echinococcus infections are endemic (1). Approximately 10-20% of subjects with hydatid cyst disease are diagnosed in childhood (3). The lung is the most commonly involved organ in children (1-3). Lung cysts generally occur by way of hematologic spread of metacestodes as a result of transphranic spread of larvae through hepatic sinusoids or perforation of parasite eggs in the stomach (1). Lung cysts may rarely occur as a result of spread of the parasites into the lymphatic circulation (2). Patients with intact cystic lesions are generally asymptomatic. Spontaneous perforation of the cyst in the lung or in relation with trauma or secondary infection may lead to sudden onset of cough, fever, hemoptysis, and hypersensitivity reactions (4).

Although serologic tests are used in the diagnosis, a positive result may not be found in all subjects with hydatid cyst disease (5). Serologic examination of our patient was found to be positive in terms of echinococcus infection.

Computed tomography is successful in terms of detecting hydatid cyst disease in the lung and can give infor-



Picture 2. a, b. Axial lung CT sections reveal cystic lesions with air-fluid level (arrow) and well-demarcated cystic lesions surrounded by a consolidation area are seen in the lower lobe of the right lung (dashed line). In addition, pleural effusion containing air bubbles due to thoracentesis is observed in the right hemithorax. Radiologic findings indicate suppurative infectious pathologies

mation related to the nature and location of the cyst (3, 5). The diagnostic CT characteristics related with perforated hydatid cyst in the lung include detached or collapsed endocyst membrane, collapsed daughter cyst membrane or intact daughter cysts (5). It may be difficult to diagnose infected perforated cysts in the lung on CT (3, 5). Computed tomography imaging revealed numerous cystic lesions with thick walls, pleural effusion, and consolidation in the right lung in our patient.

Misdiagnosis is frequently observed in hydatid cyst disease of the lung (6). In one study, it was reported that surgical intervention was performed because of misdiagnoses including pleural abscess or empyema in about 8.3% of patients who were diagnosed as having hydatid cyst disease of the lung (7). Broad-spectrum antibiotic treatment was initiated in our patient because clinical and radiologic findings related to necrotizing pneumonia were observed at the time of hospital presentation and surgical intervention was performed when adequate treatment response could not be obtained.

Cystectomy, segmentectomy, wedge resection, lobectomy or pneumonectomy are performed in the surgical treatment of hydatid cyst disease of the lung (6). In our patient, the perforated cyst was treated with cystectomy.

Approximately 15-40% of patients with hydatid cyst disease of the lung have cysts in the liver simultaneously (8). No cystic lesions in the liver were found on abdominal ultrasonography in our patient.

Leukocytoclastic vasculitis is an inflammatory disease of small vessels, also known as hypersensitivity vasculitis or hypersensitivity angiitis. This disease mostly occurs in relation with drugs that stimulate the immune system or secondary to infections including hepatitis, chronic bacteremia (e.g., endocarditis, infected shunt) and HIV (9). Skin findings are classically characterized by palpable purpura, and urticarial plaques, and vesicles; bulla and pustules are observed more rarely. In our patient, leukocytoclastic vasculitis was related with infection and there are no cases of leukocytoclastic vasculitis secondary to echinococcus infection in the literature.

In conclusion, our patient presented with a clinical picture of leukocytoclastic vasculitis and necrotizing pneumonia, and a diagnosis of hydatid cyst disease of the lung was made. In countries where hydatid cysts are endemic, perforated hydatid cysts in the lung should be kept in mind in patients in whom necrotizing pneumonia unresponsive to broad-spectrum antibiotic treatment is considered, and echinococcus infection should be kept in mind in the etiology of leukocytoclastic vasculitis.

Informed Consent: Written informed consent was obtained from patient's parents.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - S.K., S.Ç.Ş.; Design - S.K., S.A.; Supervision - S.K., S.A.; Data Collection and/or Processing - S.K., Ş.Ç.S., E.G., T.Ö.; Analysis and/or Interpretation - S.K., T.Ö., S.A.; Literature Review - S.K., E.G., S.A.; Writing - S.K.; Critical Review - S.A.

Acknowledgement: We would like to thanks all author who took part of diagnosis and treatment phase of patient.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References

1. Anadol D, Gocmen A, Kiper N, Ozçelik U. Hydatid disease in childhood: a retrospective analysis of 376 cases. Pediatr Pulmonol 1998; 26: 190-6. [CrossRef]
2. Rebhandl W, Turnbull J, Felberbauer FX, et al. Pulmonary echinococcosis hydatidosis in children: results of surgical treatment. Pediatr Pulmonol 1999; 27: 336-40. [CrossRef]
3. Turgut AT, Altin L, Topcu S, et al. Unusual imaging characteristics of complicated hydatid disease. Eur J Radiol 2007; 63: 84-93. [CrossRef]
4. Solak H, Yeniterzi M, Anil N, Göktogan T, Ceran S. The hydatid cyst of the lung in children and results of surgical treatment. Thorac Cardiovasc Surg 1990; 38: 45-7. [CrossRef]
5. Biava MF, Dao A, Fortier B. Laboratory diagnosis of cystic hydatid disease. World J Surg 2001; 25: 10-4. [CrossRef]
6. Kayhan S, Akgünes A. Histopathologically diagnosed pulmonary complicated hydatid cyst cases. Turkiye Parazitol Derg 2011; 35: 189-93. [CrossRef]
7. Cevik M, Boleken ME, Kurkcuoglu IC, Eser I, Dorterler ME. Pulmonary hydatid disease is difficult recognized in children. Pediatr Surg Int 2014; 30: 737-41. [CrossRef]
8. Doğan R, Yüksel M, Cetin G, et al. Surgical treatment of hydatid cysts of the lung: report on 1055 patients. Thorax 1989; 44: 192-9. [CrossRef]
9. Martinez-Taboada VM, Blanco R, GarciaFuentes M, Rodriguez-Valverde V. Clinical features and outcome of 95 patients with hypersensitivity vasculitis. Am J Med 1997; 102: 186-91. [CrossRef]