Case Report / *Olgu Sunumu*

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Diagnostic Challenges During Pandemic: A Case Report

Pandemide Gerçek Tanı Koyma Zorlukları: Bir Olgu Sunumu

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Abstract

Hydatid cyst is an infection caused by the parasite *Echinococcus granulosus*, which is seen endemic in animal husbandry areas. Due to the heavy burden on physicians during the pandemics, diseases other than COVID-19 took a back seat during the differential diagnosis of patients. Hydatid cyst is located in the bone at a rate of 1-4%. The symptoms and imaging features of the cyst located in the bone are not specific, thus they are less likely to be considered in the differential diagnosis of extremity lesions. The absence of a specific clinical picture and the lack of high sensitivity of the indirect hemagglutination test bring radiological evaluations to the fore in diagnosis. In this case report, we shared diagnostic and treatment process of a patient-who was first admitted to the hospital with edema, pain and fever in the leg, was diagnosed with multisystem inflammatory syndrome in children (MIS-C) later diagnosed hydatid cyst with lung and bone involvement.

Keywords: COVID-19, *Echinococcosis*, hydatid cyst, diagnostic challenges, pandemic

Öz

Kist hidatik *Echinococcus granulosus* isimli parazitin neden olduğu, hayvan yetiştirilen bölgelerde endemik olarak görülen bir enfeksiyondur. Pandemi sürecinde hekimlerin üzerindeki ağır yük nedeniyle, hastalara ayırıcı tanı yapılması aşamasında COVID-19 harici hastalıklar biraz arka plana itilmiştir. Hidatik kist %1-4 oranında kemikte yerleşir. Kemikte yerleşen kiste ait semptomlar ve görüntüleme özellikleri özgün değildir, bu nedenle ekstremite lezyonlarında ayırıcı tanı yapılırken akla gelmesi daha az olasıdır. Özgül klinik tablonun olmaması ve indirekt hemaglütinasyon testinin sensitivitesinin yüksek olmaması tanıda radyolojik değerlendirmeleri ön plana çıkartmaktadır. Bu olgu sunumunda, bacakta ödem, ağrı ve ateş ile hastaneye başvuran, çocuklarda multisistem enflamatuvar sendrom (MIS-C) tanısı aldıktan sonra akciğer ve kemik tutulumlu kist hidatik tanısı alan bir hastamızın tanı ve tedavi sürecini paylaştık.

Anahtar Kelimeler: COVID-19, ekinokokkozis, hidatik kist, pandemi, tanı zorlukları

Introduction

SARS-CoV-2, which emerged in Wuhan, China at the end of 2019, spread rapidly around the world and was declared a pandemic by the World Health Organization in March 2020. During this pandemic, which has been going on for about more than two years, diseases other than Coronavirus disease-2019 (COVID-19) have remained in the background. Many pediatric

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patients presenting to the emergency department with fever are referred to a pediatric infectious disease specialist to be evaluated for COVID-19 or multisystemic inflammatory syndrome (MIS-C) in children, sometimes without even being tested. In this case report, we shared the diagnostic process of a case who was transferred to the intensive care unit of our hospital after MIS-C was considered and treatment was initiated.

Case Report

A 13-year-old female was admitted to another hospital with complaints of severe pain in the left leg and hip and fever reaching up to 39ºC for three days. She was directed to the pediatric intensive care unit of our hospital with the pre-diagnosis of MIS-C, upon the edema in the left gluteal and iliac muscles in the pelvic magnetic resonance imaging (MRI) and findings consistent with COVID-19 pneumonia in the lung CT scan.

Her physical examination of the patient was normal except for pain, edema, and restriction of movement in the left hip. Her fever was 39ºC. Her laboratory investigations were as follows: leukocytes 9370/mm^3, neutrophils 8320/mm^3, lymphocytes 440/mm^3, platelets 177,000/mm^3, ESR 81 mm/hour (normal= 2-20), CRP 252 mg/L (normal< 5), procalcitonin> 100 µg/L (normal< 0.5) , CK 29.800 U/L (normal< 192), ferritin 1322 µg/L (normal< 68), LDH= 890 U/L (normal< 300), d-dimer 12.540 ug/L (normal< 500) and troponin 0.033 mcg/L (normal< 0.014). COVID-19 PCR and COVID IgM-IgG tests were studied twice, with negative results. The pelvic MRI performed at the first center was evaluated again by our radiology department; edema in the left gluteal and iliac muscles and tendons, additionally fluid intensities between the fasciae were reported. No cysts mentioned. Ceftriaxone, vancomycin, and clarithromycin treatment which had been already started before were continued. Since her fever and laboratory markers are not responding to treatment, we consulted the patient with pediatric hematology and nephrology departments. We reached a consensus about starting treatment against macrophage activation syndrome. Methylprednisolone (5 mg/kg), intravenous immunoglobulin (IVIG-1 g/kg), and enoxaparin (1 x 60 mg subcutaneously) were started. Echocardiography was normal. *Staphylococcus aureus* proliferation was reported in hemoculture. The patient was transferred to the general pediatric ward on the eighth day of hospitalization. Antibiotic therapies were stopped after 10 days. No pathological findings were found in the investigations for rheumatological diseases.

On the 12th day of his hospitalization, a thorax CT was performed, and it revealed multiple fluid density cysts with millimetric cavitations in both of lungs. The largest cysts was 22 x 20 mm, and there was no mediastinal or hilar lymphadenopathy (Figure 1). The number of lesions was same as the previous CT, but they were smaller and had less consolidated features. It was also noted that their air-fluid levels and cavitary features become more evident. It was stated that septic embolism or vasculitis should be considered in differential diagnosis. Contrast-enhanced thorax CT angiography performed which did not reveal any intravascular pathology. In the follow-up pelvic MRI, there was an infiltrative lesion with hyperintense lobulated contours in the left iliac bone and an area of signal change showing heterogeneous contrast enhancement in the adjacent muscle group in the periosteum (Figure 2). Bone scintigraphy was evaluated as normal. Tuberculosis and immunological examinations were normal. Bronchoscopy was performed and yielded normal results. The tests on bronchoalveolar lavage samples were normal.

On the 24th day of hospitalization, there was a minimal increase in the size of the cysts in the thorax CT performed upon

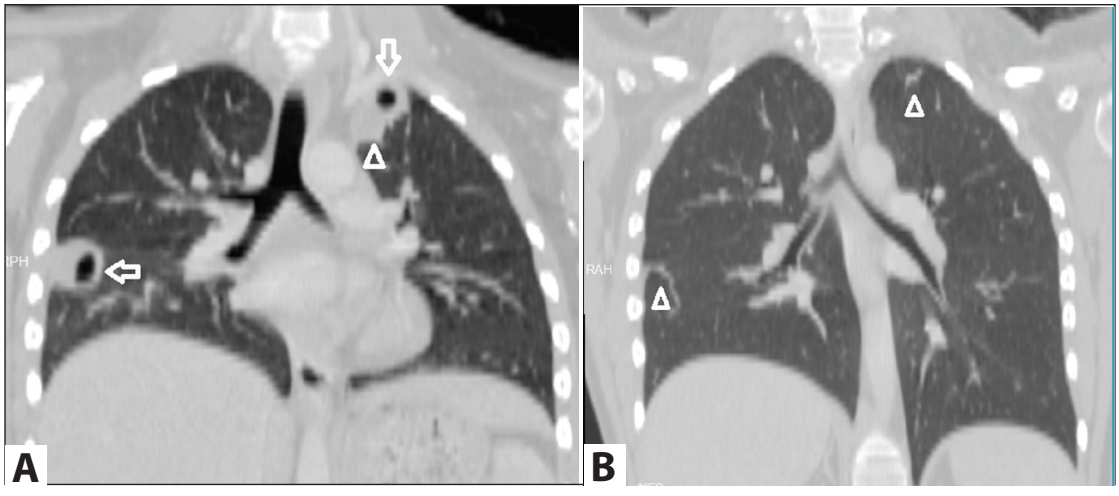

Figure 1. A. Cysts with air-fluid leveling on contrast-enhanced thoracic CT coronal images (closed arrow) and lesions in pure cystic nature (arrowhead) consistent with diffuse pulmonary intraparenchymal hydatid cysts in both lungs. **B.** After two months of albendazole treatment, no residual cavity image is observed in the cystic lesions, and sequelae of linear atelectasis are observed (arrow).

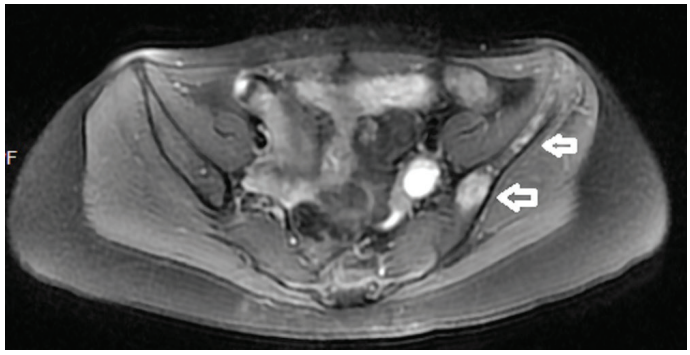

Figure 2. Pelvic MRI: lesions showing hyperintense signal with multifocal geographic contours in the left iliac bone and accompanied by edema in adjacent muscle groups, non-enhancing lesions were evaluated in favor of intraosseous cyst.

the complaint of pain on the right side while breathing. CT scan and pelvic MRI images were re-interpreted by the pediatric radiologist at the another university hospital. Their report of the thorax CT was as follows: "There are fluid-filled thin-walled cysts scattered in all lobes of both lungs, there is an air-liquid level in some cyst lumens that may be due to bronchial opening. A few millimeter-sized sequelae air cysts are seen in the lung. The follow-up pelvic MRI was also re-interpreted as the following: Microabscess formations are observed in the medullotrabecular and adjacent periosteal and gluteal muscle groups in the left iliac wing supraacetabular area. When these images were evaluated together, hydatid cyst disease with diffuse pulmonary involvement and iliac bone involvement was considered. Upon this comment, an indirect hemagglutination test was performed and albendazole (15 mg/kg/day, twice a day orally) treatment was started. The indirect hemagglutination test was negative. Cranial MRI, performed for central nervous system involvement and the abdominal MRI and CT for gastrointestinal system involvement yielded normal results.

We consulted the patient with the pediatric rheumatology department. The clinical course of the patient was found to be consistent with macrophage activation syndrome due to hydatid cyst rupture, and edema in the pelvic muscles was consistent with secondary pyomyositis. Continuing low-dose steroid therapy was recommended. No mutation was observed in the immune dysregulation panel.

The thorax CT performed in the first month of the treatment showed a significant reduction in the pulmonary lesions. Excision was not recommended by the pediatric surgery and orthopedics departments since the lesions are responsive to albendazole. The patient was discharged with oral albendazole and oral methylprednisolone (0.5 mg/kg). Methylprednisolone treatment was discontinued in the fourth month. The imaging performed in the second month, while the patient was still on albendazole, showed that the lung lesions were almost completely responsive to the treatment, the fluid signal intensity decreased in the lesion in the left iliac bone and there was retraction in the lesion contours. The bone lesion was also responsive to medical treatment.

Verbal and written consent was obtained from the parents for this case report.

Discussion

Hydatid cyst is an infection caused by the parasite *Echinococcus granulosus*, which is endemic in regions involved in animal husbandry. Dogs and other carnivorous animals are definitive hosts, while sheep, cattle, horses, and goats are intermediate hosts. Humans are incidentally infected and are the final host. Although *Echinococcus* most commonly affects the liver in adults, lung involvement is more common in children than in adults (1). Other organs such as the spleen, heart, and brain are rarely affected, while bone and soft tissue involvement is 1-5% of all cases (2).

The symptoms and imaging findings of hydatid cysts in the bone are not specific, thus they are less likely to be considered in the differential diagnosis of extremity lesions, as in our patient. *Echinococcal* cysts located in the bone may remain dormant for years. Symptoms are usually caused by a pathological fracture, a neurological deficit, or an infection (3). In addition, *Echinococcus* is somewhat overlooked by physicians who do not live in rural areas. For these reasons, the diagnosis of *Ehinococcus* is considered late and the final diagnosis is usually made after surgery.

Our patient was admitted with complaints of leg pain, swelling, and high fever during the pandemic and was referred to our hospital with a preliminary diagnosis of MIS-C, although she did not have COVID-19. Multiple lesions were mistakenly interpreted as COVID-19-related in the pulmonary CT performed at an first admission, which interpreted in favor of MIS-C. A very low incidence of cavitation has been reported in COVID-19-associated pulmonary lesions (4). The MRI performed at the same time, which evaluated the pelvic bones and muscles, showed edema, but there were no cystic findings so it was interpreted as myositis.

Radiological studies are prioritized in the diagnosis because there is no distinctive clinical presentation and the indirect hemagglutination test is not sensitive enough (60%). It can be challenging to diagnose *Echinococcosis* radiologically. Although CT is the most efficient radiological method in hydatid cyst disease involving the lung, the lesions can sometimes be confused with other diseases. In the literature, there are many cases where tumor or tuberculosis is considered and the diagnosis is later changed after histological examination (6-8). In our case, a large number of ground-glass findings were found in the first

pulmonary CT, and these lesions were considered as viral pneumonia or COVID-19 pneumonia. The presence of ground glass findings and or consolidation around hydatid cysts can occur due to secondary pulmonary infection around the cyst, alveolar hemorrhage due to cyst growth, or secondary findings to cyst rupture (5,9). However, the main pattern and secondary findings should be carefully distinguished from each other. The fact that the patient was admitted to the hospital during the peak of the pandemic, led us, clinicians, more towards COVID-19 and its complications. In a study published in February 2021, it was shown that the number of patients diagnosed with tuberculosis during the pandemic decreased by 43% and there were significant delays in diagnosis (10). Indeed, a review of the literature shows case reports of misdiagnoses or delayed diagnoses during the COVID-19 pandemic (11,12). During the pandemic, due to patients' reluctance to visit the hospital for minor complaints and their tendency to only go when things got worse, as well as clinicians' primary focus on the diagnosis or exclusion of COVID-19, there were delays and misdiagnoses in treatment.

Hydatid cysts are located in the bone at a rate of 1-4%. The spine, pelvis, and long bones are the most typical sites. Due to their slow growth rate, they are usually diagnosed at a later age and are rarely encountered in childhood. Although a systematic treatment algorithm is not yet available for bone involvement, treatment options consist of surgical excision followed by the use of albendazole (13). In some studies, albendazole was used in combination with praziquantel or nitazoxanide (14,15). There is no definitive protocol for the duration of albendazole use. Since our patient showed radiological response to albendazole treatment, surgical excision was not recommended by the pediatric surgery and orthopedics departments. The literature shows some cases where surgical treatment was not possible or involved high risk, and were followed up with medical treatment (13,14).

Conclusion

The concern of physicians not being able to recognize and treat COVID-19 patients in the pandemic has caused clinicians to focus more on COVID-19 when evaluating patients. Although our patient did not have any contact with COVID-19 and the repeated COVID-19 PCR and antibody tests resulted negative, the patient was diagnosed with MIS-C and her treatment was started. Even though COVID-19 is the a top priority during the pandemic, it's important to remember that other illnesses should also be taken into account when making a differential diagnosis.

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References

1. *Kanat F, Turk E, Aribas OK. Comparison of pulmonary hydatid cysts in children and adults. ANZ J Surg 2004;74(10):885-9. [CrossRef]*
2. *García-Díez AI, Ros Mendoza LH, Villacampa VM, Cózar M, Fuertes MI. MRI evaluation of soft tissue hydatid disease. Eur Radiol 2000;10(3):462-6. [CrossRef]*
3. *Zlitni M, Ezzaouia K, Lebib H, Karray M, Kooli M, Mestiri M. Hydatid cyst of bone: Diagnosis and treatment. World J Surg 2001;25(1):75-82. [CrossRef]*
4. *Selvaraj V, Dapaah-Afriyie K. Lung cavitation due to COVID-19 pneumonia. BMJ Case Rep 2020;13(7):e237245. [CrossRef]*
5. *Çobanoğlu U, Aşker S, Mergan D, Sayır F, Bilici S, Melek M. Diagnostic dilemma in hydatid cysts: Tumor-mimicking hydatid cysts. Turk Thorac J 2015;16(4):180-4. [CrossRef]*
6. *Mitrovic M, Tadic B, Kovac J, Grubor N, Milosavljevic V, Jankovic A, et al. Pancreatic hydatid cyst misdiagnosed as mucinous cystadenoma: CT and MRI findings. Medicina (Kaunas) 2020;56(3):124. [CrossRef]*
7. *Gadelkareem RA, Elqady AA, Abd-Elshafy SK, Imam H, Abolella HA. Isolated renal hydatid cyst misdiagnosed and operated as a cystic renal tumor. Med Princ Pract 2018;27(3):297-300. [CrossRef]*
8. *Zhang Q, Huang TM, Li BZ, Li ZL, Liao KX. [Misdiagnosis of pulmonary hydatid cyst rupture: Report of 38 cases] Zhonghua Jie He He Hu Xi Za Zhi 2003;26(8):474-6.*
9. *Garg MK, Sharma M, Gulati A, Gorsi U, Aggarwal AN, Agarwal R, et al. Imaging in pulmonary hydatid cysts. World J Radiol 2016;8(6):581-7. [CrossRef]*
10. *Di Gennaro F, Gualano G, Timelli L, Vittozzi P, Di Bari V, Libertone R, et al. Increase in tuberculosis diagnostic delay during first wave of the covid-19 pandemic: Data from an Italian Infectious Disease Referral Hospital. Antibiotics (Basel) 2021;10(3):272. [CrossRef]*
11. *Novak CB, Scheeler VM, Aucott JN. Lyme disease in the era of COVID-19: A delayed diagnosis and risk for complications. Case Rep Infect Dis 2021;2021:6699536. [CrossRef]*
12. *Yousefzai R, Bhimaraj A. Misdiagnosis in the COVID-19 era: When zebras are everywhere, don't forget the horses. JACC Case Rep 2020;2(10):1614-9. [CrossRef]*
13. *Cattaneo L, Manciulli T, Cretu CM, Giordani MT, Angheben A, et al. Cystic Echinococcosis of the bone: A European Multicenter Study. Am J Trop Med Hyg 2019;100(3):617-21. [CrossRef]*
14. *Monge-Maillo B, Olmedo Samperio M, Pérez-Molina JA, Norman F, Mejía CR, Tojeiro SC, et al. Osseous cystic Echinococcosis: A case series study at a referral unit in Spain. PLoS Negl Trop Dis 2019;13(2):e0007006. [CrossRef]*
15. *Perez-Molina JA, Diaz-Menendez M, Gallego JI, Norman F, Monge-Maillo B, Ayala AP, et al. Evaluation of nitazoxanide for the treatment of disseminated cystic Echinococcosis: Report of five cases and literature review. Am J Trop Med Hyg 84:351-6. [CrossRef]*