



Case Report / Olgu Sunumu

## Pulmonary metastasectomy with laparotomy in alveolar echinococcosis

### *Alveolar ekinokokta laparotomi ile pulmoner metastazektomi*

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#### ABSTRACT

Pulmonary alveolar echinococcosis is a tumor-like parasitic disease that occurs typically after hepatic involvement. In this article, we present a case of metastatic pulmonary alveolar echinococcosis that underwent laparotomy for hepatic involvement, in which we performed pulmonary metastasectomy with transdiaphragmatic intervention. Thoracic computed tomography revealed a metastatic nodule of approximately 1 cm in the superior segment of the right lung lower lobe. Approximately 7×3 cm diaphragmatic resection was performed due to diaphragmatic invasion. Pulmonary wedge resection for lung metastasis was performed intraabdominally from diaphragmatic defect. We believe that this technique can be applied safely in carefully selected patients with pulmonary involvement.

**Keywords:** Laparotomy, lung, metastasis.

Alveolar echinococcosis (AE) is a chronic and progressive disease that primarily involves the liver and is caused by larval forms (metacestodes) of the *Echinococcus multilocularis* (*E. multilocularis*) tapeworms.<sup>[1]</sup>

In the life cycle of *E. multilocularis*, definite hosts are foxes, wild dogs, wolves, and jackals. The intermediate hosts are rodents, deer, and bison. Pets such as dogs or cats may also be infected and may contaminate humans directly or via the fecal-oral route by ingesting tapeworm eggs.<sup>[1]</sup> Adult parasites live in the intestines of predators, particularly in *Vulpes vulpes*. Tapeworm eggs are released into nature in the feces of these animals. Humans may be infected by

#### ÖZ

Pulmoner alveoler ekinokok, tipik olarak hepatik tutulumdan sonra ortaya çıkan tümör benzeri parazitik bir hastalıktır. Bu yazıda, hepatik tutulum için laparotomi uygulanan, transdiyafragmatik müdahale ile pulmoner metastazektomi yapılan bir metastatik pulmoner alveoler ekinokok olgusu sunuldu. Torasik bilgisayarlı tomografi sağ akciğer alt lob superior segmentte yaklaşık 1 cm'lik bir metastatik nodül görüldü. Diyafragmatik invazyon nedeniyle yaklaşık 7×3 cm diyafragmatik rezeksiyon yapıldı. Akciğer metastazı için intraabdominal olarak diyafragmatik defektten pulmoner kama rezeksiyonu yapıldı. Pulmoner tutulumu olan dikkatli seçilmiş hastalarda bu tekniğin güvenle uygulanabileceğini düşünüyoruz.

**Anahtar sözcükler:** Laparotomi, akciğer, metastaz.

physical contact with pets or wild animals carrying the parasite in their small intestine or by eating food or drinking water that is contaminated. After humans ingest these eggs accidentally, the multi-vesicular metacestodes shows a tumor-like growth, particularly in the liver. It invades and destroys tissues and extends into adjacent structures and may metastasize to the lung, brain, and other organs.<sup>[1]</sup>

Because of the low incidence of AE and the very rare involvement of the lungs, only a few pulmonary AE case reports are found in the literature.<sup>[2]</sup> In this article, we present the case of a patient who underwent pulmonary AE metastasectomy via laparotomy using the transdiaphragmatic approach.

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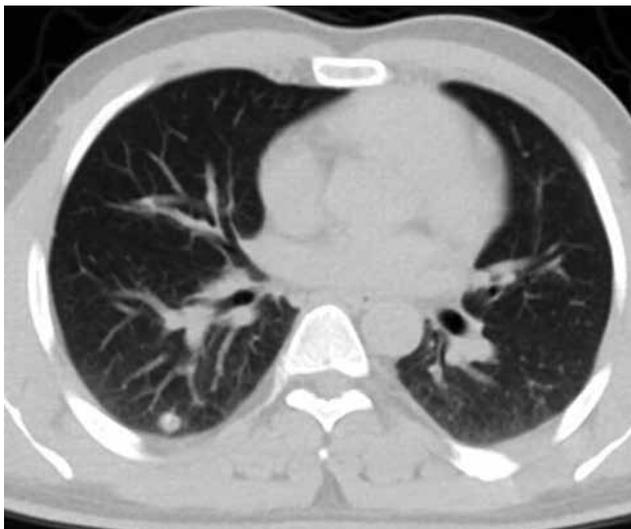
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## CASE REPORT

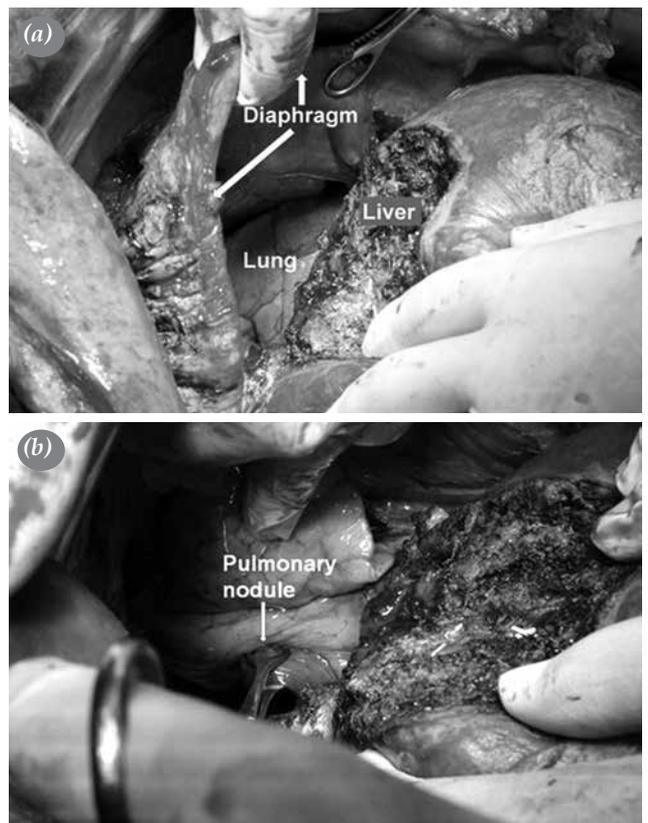
A 32-year-old male patient presented with right upper abdominal pain, fatigue, weakness, and loss of appetite complaints that began approximately two months earlier. Physical examination revealed hepatomegaly. Respiratory system examination was normal. The laboratory values were as follows: white blood cell,  $7.3/\text{mm}^3$ ; hemoglobin, 16.6 g/dL; platelet,  $176,000/\text{mm}^3$ ; bilirubin (total), 0.4 mg/dL; alkaline phosphatase, 512 IU/L; gamma-glutamyl transferase, 159 IU/L; aspartate aminotransferase, 34 IU/L; alanine transaminase, 56 IU/L. Echinococcosis immunoglobulin G enzyme-linked immunosorbent assay test was positive. No pathological findings were found on the posteroanterior chest radiograph. Computed tomography revealed cystic mass lesions; approximately, 3 cm in segment five and 5 cm in segment eight. In addition, the thorax section revealed a nodule of approximately 1 cm in the superior segment of the right lung lower lobe (Figure 1). No other involvement was detected in any part of the body. Due to the prevalence of AE in our region, the hepatic alveolar cyst was suspected radiologically. Preoperatively, the pulmonary nodule was considered to be the metastasis of the hepatic AE.

The patient was operated by laparotomy incision. Non-anatomical liver resection was performed for the primary lesion in the liver. A diaphragmatic resection of about  $7 \times 3$  cm was performed at the level of liver segment eight due to invasion of the diaphragm. The nodule in the right lower lobe was palpated through the opening in the diaphragm (Figure 2). The pulmonary

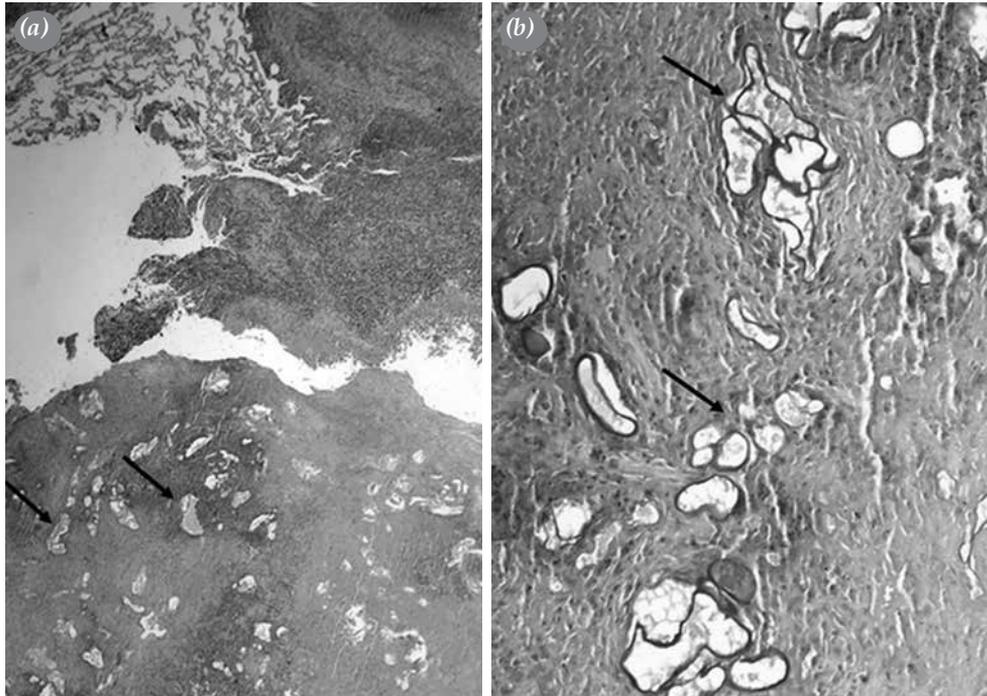
nodule was removed by wedge resection using thick tissue 60 mm linear stapler. A 24 F chest drain was inserted into the right thorax cavity through the eighth intercostal space. The diaphragm was closed primarily with non-absorbable sutures without using a graft. Surgery was performed by a general surgeon and thoracic surgeons. Hepatectomy was performed by the general surgeon, while pulmonary metastasectomy, diaphragmatic resection, and reconstruction were performed by the thoracic surgeons. Diaphragmatic resection was performed due to the invasion of the diaphragm by hepatic AE. Since the diaphragmatic resection was performed, no extra thoracoscopic intervention was performed and transabdominal pulmonary metastasectomy was performed from the defect in the diaphragm. Histopathological examination revealed pulmonary nodule as alveolar echinococcal metastasis (Figure 3). In order to prevent recurrence, treatment was given as 400 mg albendazole twice daily. Albendazole was planned to be continued for a period of two years with a total of three months yearly. No complications were encountered in the postoperative follow-up period of the patient. A written informed consent was obtained from the patient.



**Figure 1.** Axial computed tomography showing pulmonary metastasis in superior segment of right lung lower lobe.



**Figure 2.** (a) Right hemithorax and (b) pulmonary nodule is seen from resected diaphragmatic defect.



**Figure 3.** (a) Multiple cysts (arrows) in lung parenchyma in a necrotic background (H-E  $\times 4$ ). (b) Periodic acid-Schiff positive laminated membranes (arrows) (periodic acid-Schiff,  $\times 20$ ).

## DISCUSSION

Metastasectomy is one of the most frequently performed surgical resections by thoracic surgeons.<sup>[3]</sup> Lung is the second most common location of metastases.<sup>[3]</sup> Today, oncological criteria for pulmonary metastasectomy are: (i) primary cancer must be under control or controllable, (ii) there must not be any extra-thoracic metastases, (iii) metastasis must be resectable with adequate pulmonary reserve and (iv) there must not be any alternative medical treatment options with low morbidity.<sup>[3]</sup>

Primary extra-hepatic localization of *E. multilocularis* is rare.<sup>[1]</sup> Bresson-Hadni *et al.*<sup>[4]</sup> has reported pulmonary metastases in 20% of patients in their 117 AE series. Lung involvement always occurs after hepatic involvement. Pulmonary AE is mainly caused by hematogenous spread from hepatic AE lesions.<sup>[4]</sup> *E. multilocularis* presents a cancer-like appearance in lung with infiltrative growth and metastatic spread.

Laparotomy is not a routine procedure for approaching pulmonary lesions. We applied this approach to a case which had an absolute indication for laparotomy for its hepatic, spleen and lung hydatid cysts.<sup>[5]</sup> In the previous study, it was stated that the

following criteria should be present for approaching lung hydatid cysts by laparotomy: patients must not have thoracotomy history and pleural adhesion, hydatid cyst in lung must be type 1 and must not be a giant cyst, there must be an absolute laparotomy indication, and lung hydatid cyst must be close to diaphragm and easily accessible.<sup>[5]</sup> In our present study, the diagnosis was different, but there was an absolute indication for laparotomy and it was possible to reach and resect the pulmonary lesion transabdominally.

In conclusion, there was an absolute laparotomy indication in the case that we have presented. There was also no need for a separate incision to reach the pulmonary nodule because of the need for diaphragmatic resection. This is a simple, effective and safe method that prevents complications due to another surgical procedure or thoracotomy in patients with right or left pulmonary lesions. This pulmonary intervention must be performed only by a specialist in thoracic surgery. We believe that this method should be considered in the treatment of non-complex pulmonary lesions in cases requiring laparotomy.

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