

Hydatid cysts of the pulmonary artery

Pulmoner arter kist hidatiği

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ABSTRACT

Hydatid cyst located in the pulmonary artery is rare. The cause is usually rupture of an intracardiac cyst or dissemination from a hepatic focus either spontaneously or secondary to surgery. In this article, we report a 32-year-old patient with a history of hepatic and pulmonary hydatid cyst resection, presenting with multiple hydatid cysts located in the right pulmonary artery, and complaints of dyspnea and hemoptysis.

Keywords: Computed tomography; hydatid cyst; magnetic resonance imaging; pulmonary artery cyst.

Hydatidosis is a parasitic infection caused by ingesting food contaminated with the larvae of *Echinococcus granulosus*. After ingestion, parasite's eggs hatch and the embryos penetrate through the intestinal mucosa and reach the liver via portal venous system. Liver and the lung filter 60% to 70% and 15% to 25% of the embryos, respectively; the remaining embryos may disseminate through systemic vessels and involve any organ system.^[1,2]

Pulmonary artery involvement by hydatid cysts is extremely rare and may be seen due to either rupture of an intracardiac cyst or dissemination from a hepatic focus spontaneously or secondary to surgery. Cysts

ÖZ

Pulmoner arter yerleşimli kist hidatik nadirdir. Nedeni genellikle kalp içi kistin patlaması veya karaciğerdeki odaktan cerrahi işleme bağlı olarak veya spontan yayılımıdır. Bu yazıda, karaciğer ve pulmoner kist hidatik rezeksiyonu öyküsü olan, sağ pulmoner arterde çok sayıda hidatik kist ve dispne ve hemoptizi yakınması ile başvuran 32 yaşında bir erkek hasta sunuldu.

Anahtar sözcükler: Bilgisayarlı tomografi; kist hidatik; manyetik rezonans görüntüleme; pulmoner arter kisti.

within pulmonary arteries are associated with a high mortality rate due to the risk of embolization and anaphylactic shock.^[3]

In this article, we report a case of hydatid cysts located in right pulmonary arteries with a history of hepatic and pulmonary hydatid cysts.

CASE REPORT

A 32-year-old male patient with a history of hepatic and pulmonary hydatid cysts presented with intermittent hemoptysis and dyspnea. Hemoptysis started two weeks prior to presentation, initially as a spoonful blood tinged sputum. At presentation, patient had



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bloody sputum of 150 mL. Medical history revealed a surgical resection of a hepatic hydatid cyst 13 years ago and cystotomy-capitonnage for right sided pulmonary hydatid cysts three years ago. Throughout the years, the patient had received long-term antiparasitic therapy with albendazole; however, indications and the course of therapy were not available in the medical records. For a diagnostic work-up, we performed a contrast enhanced computed tomography of the thorax. We observed a heterogeneous lesion with low attenuation areas and mild peripheral contrast enhancement extending from right main pulmonary artery into segmental arteries. We noted enlargement of right sided pulmonary vessels with hypertrophied bronchial and intercostal arteries (Figure 1a). The lesion was in close proximity to segmental bronchi, while we detected obliteration of right sided lower lobe posterior bronchi by a low attenuation density. We carried out magnetic resonance imaging for differential diagnosis, which revealed cystic lesions extending from right main pulmonary artery into segmental branches that

are heterogeneously hyperintense in T₂ weighted images and hypointense in T₁ weighted image with peripheral contrast enhancement on contrast enhanced T₁ weighted image. We also noted a peripheral hypointense ring surrounding the lesion in T₂ weighted image (Figure 1b, c). Clinical history and imaging findings were suggestive of pulmonary artery hydatid cyst.

Three months later, the patient was referred for pulmonary endarterectomy (PEA). Prior to surgery, the patient had no hemoptysis. To remove the cysts in the right pulmonary artery and its segmental branches, we planned PEA surgery, which was performed under general anesthesia through a median sternotomy and using extracorporeal circulation with periods of circulatory arrest under deep hypothermia. Following the arteriotomy on the right side, we noted total occlusion of right pulmonary artery with the cyst at the level of pulmonary trunk (Figure 1d). During endarterectomy, we detected a fistula between intralobar artery and bronchus

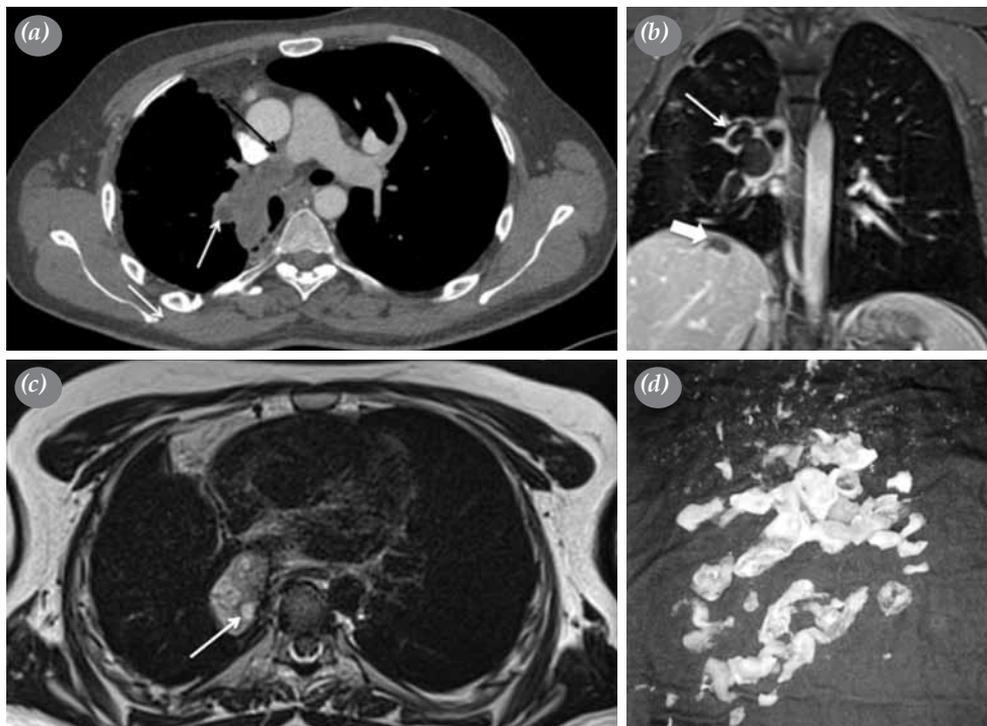


Figure 1. (a) Contrast enhanced computed tomography axial image shows right pulmonary artery occluded (black-arrow) by a hypodense lesion extending into right lower and middle lobe segmental arteries with enlargement of arteries. The lesion has fluid attenuation level with mild peripheral contrast enhancement (white-arrow). (b) Coronal contrast enhanced T₁ weighted image reveals a hypointense lesion with peripheral hyperintensity (thin-arrow) along with hypointense lesions in the liver (thick-arrow). (c) Axial T₂ weighted image shows hyperintense cysts representing daughter vesicles (arrow). (d) Surgical resection of cysts in right pulmonary arteries reveals numerous intact and ruptured hydatid cysts.

which resulted in massive endotracheal bleeding through the fistula. All attempts to close the fistula through the pulmonary artery were unsuccessful. We did not open the right pleura because of the dense pleural adhesions of the right thoracic cavity due to previous thoracotomy. To overcome massive hemoptysis, we decided on physiological lung exclusion since, otherwise, lung resection would be difficult, increase the operating time, and cause more bleeding. We stapled the right main pulmonary artery and the right main bronchus leaving the pulmonary veins intact.

On postoperative day (POD) three, hypoxia and hypercarbia developed with a left lower lobe infiltration. First, we performed percutaneous extracorporeal membrane oxygenation (ECMO) using femorofemoral vein circuit. However, we instituted a peripheral veno-arterial ECMO because of hypoxia and hemodynamic instability. The patient was successfully weaned from ECMO on POD seven. Unfortunately, the patient died on POD 12, due to septic shock.

DISCUSSION

Hydatid cysts locations in the pulmonary arteries are exceptional and may be secondary to dissemination from intracardiac cysts or a hepatic focus spontaneously or secondary to surgery.^[3] During hepatic cystectomy, clamping of inferior vena cava and avoidance of hepatic traction may decrease the risk of pulmonary dissemination.^[4]

Our patient was symptom free for three years after cystotomy-capitonnage for right sided pulmonary hydatid cysts which can be explained by the slow growth rate of cysts located in an artery which may last long enough time for collaterals to develop. Collaterals may maintain adequate pulmonary perfusion and the patient may remain asymptomatic. However, symptoms may develop in the long-term due to further cyst growth, compression of a vital structure by cyst or rupture of the cyst with resultant anaphylactic shock.^[1] In our patient, imaging prior to pulmonary cystotomy did not show pulmonary arterial involvement; however, we noticed a fistula between segmental pulmonary arteries and bronchi during surgery. Based on these imaging and surgical findings, our impression for this patient was that pulmonary artery hydatid cyst was related to cystotomy for pulmonary hydatid cysts. However, as the patient had hydatid cysts in the liver, spontaneous dissemination through liver cannot be excluded.

On CT, intra-arterial cysts appear as rounded intravascular masses with levels of fluid attenuation,

with contrast enhancement at periphery. The differential diagnosis for such intraluminal defects includes pulmonary thromboembolism and primary arterial tumors, such as sarcomas. However, an arterial tumor would show more diffuse contrast enhancement and a faster rate of growth. Thromboembolism may present with similar symptoms of cough, hemoptysis, and acute onset of chest pain. Absence of predisposing factors for thromboembolism may be helpful in differential diagnosis. Furthermore, magnetic resonance imaging may assist in diagnosis by showing the cystic nature of lesions by revealing homogeneously hypointense lesions in T₁ weighted image and hyperintense lesions in T₂ weighted image. A typical finding is a low signal ring on T₂ weighted image indicating cyst wall.^[5]

Preoperative bronchoscopy is an important tool for patients presenting with hemoptysis. It helps to localize the origin of the hemoptysis and also rules-out any presence of intrabronchial lesion prior to surgery. Our patient did not complain of hemoptysis before the surgery although his previous symptom was hemoptysis three months ago. Following the intubation of the patient, intratracheal aspiration was clean which led us not to perform preoperative bronchoscopy.

In our patient, we planned PEA to remove the cysts in the pulmonary artery. This operation is more challenging than regular PEA performed for chronic thromboembolic pulmonary hypertension. Since the cystic inflammation is more firmly attached to the intima layer of the artery, finding a true dissection plane was very difficult. On the other hand, massive hemoptysis led us to perform physiological exclusion of the right lung. This surgical option is applicable in cases of: (i) really tight pleural adhesions which would increase the operating time and cause more bleeding; (ii) desaturated patients; and (iii) hemodynamically unstable patients.^[6] Preoperative anti-helminthic medication may be employed to decrease the risk of spillage during surgery. Our patient had also received long-term antiparasitic therapy before the surgery. Surgery may be complicated by dissemination of cystic contents, embolism, anaphylactic shock, pseudoaneurysm formation, and uncontrollable bleeding, as demonstrated in this case.

Computed tomography and magnetic resonance imaging are helpful in diagnosis and required for patient follow-up to investigate for recurrences and formation of pseudoaneurysm. In cases with diffuse and severe pulmonary arterial involvement, reported mortality in the literature is high.^[7]

In conclusion, hydatid cyst located in pulmonary artery is rare and awareness of imaging and clinical features of this entity provides a better differential diagnosis.

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