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Repair of diaphragmatic biliary pleural fistula in a hydatid cyst with pericardial patch: A case report

Bilioplevral fistüle neden olan bir hidatik kistte perikardiyal yama ile diafragma tamiri: Olgu sunumu

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Abstract
Intrathoracic complications are rare events in postoperative recurrence of hepatic hydatid cysts; however, they are serious causes of mortality. In this report, we aimed to present a case of diaphragmatic defect developing as an intrathoracic complication after recurrence, which we repaired using a pericardial patch. Based on the literature, we encountered no use of pericardial patches in such cases.
Keywords: Hydatid cyst, Recurrence, Biliopleural fistula, Diaphragm repair, Autologous graft

Öz
Karaciğer kist hidatiklerinde operasyon sonrası oluşan nükslerde intratorasik komplikasyonlar nadirdir. Oluşan komplikasyonlar ise ciddi mortalite sebebidir. Biz nüks sonrası intratorasik komplikasyon olan diafragma defektini primer onardıktan sonra perikardiyal yama ile güçlendirdiğimiz bir olguyu sunduk. Yaptığımız litaratür taramalarında bu tür vakalarda daha önce perikardiyal yama kullanıldığına rastlamadık.
Anahtar kelimeler: Kist hidatik, Nüks, Bilioplevral fistül, Diafram tamiri, Otolog greft

Introduction

The spread of liver hydatid cysts to the thorax is rare. However, direct compression of the liver hydatid cyst on the diaphragm disrupting its vascular supply, the chemical effect of bile content and negative intrapleural pressure may all cause this complication [1,2]. In this report, we present a case of a diaphragmatic defect, an intrathoracic complication after recurrence of a liver hydatid cyst, that was strengthened with a pericardial patch following primary repair.

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Case presentation

A 48-year-old female patient was admitted to our clinic with complaints of dyspnea, fever, and cough. The patient had been operated due to hydatid cyst of the liver three years ago, and the cysts which could not be removed with USG-guided surgery were denaturized with hypertonic saline serum. An USG-guided drainage catheter was placed in the patient due to liver abscess approximately two months ago, and she was treated with albendazole for 6 months. Physical examination revealed decreased breathing sounds on the right. White blood cell count (WBC) was 5600 cells/mm^3 and direct bilirubin was 0.25 g/dL. Pneumothorax and air-fluid levels were observed in the right hemithorax on chest radiograph (Figure 1). Thoracic computerized tomography (CT) showed atelectasis around the right main, middle, and lower lobe bronchi, along with increases in soft tissue density which were not clearly differentiable from lung parenchyma. Also, a peripherally located lesion with lobulated contours and air-fluid levels containing thin septations was observed in the right hemithorax, presumably within the pleural space, accompanied by pleural thickening. A catheter was seen to extend into the liver parenchyma, and free air was visualized within the intrahepatic biliary tract (Figure 2). Thoracotomy revealed that the parietal and visceral pleura were highly adherent and thick. Cystic membrane was observed between the pleural leaves. The diaphragm defect was 4x4.5 cm in size, and membranes of the hydatid cyst were seen within the cavitary lesions of the liver (Figure 3).

Following removal, cystic membranes were sent for pathology, and pleural decortication was performed. General surgery consultation was requested peroperatively. Upon witnessing no bile leakage, the defect was closed primarily. Approximately 3x3 cm pericardial patch was removed from the area closest to the diaphragm and used as a support patch (Figure 4). Since the defect on the diaphragm was remarkably close to the pericardium, it was easily placed. The pericardium was uneventfully closed end-to-end primarily to prevent cardiac herniation.

Pathological examination revealed a hydatid cyst with acute suppurative inflammation. During the postoperative follow-up period, due to bile leakage from the drain, a sphincterectomy was performed in the gastroenterology unit, and a 10 cm-long plastic 10Fr stent was placed at the hilar level to extend into the choledoch. The patient was discharged uneventfully on the 12th postoperative day without complications.

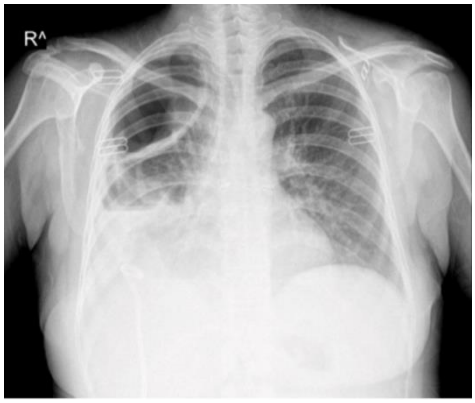

Figure 1: Lung X-ray

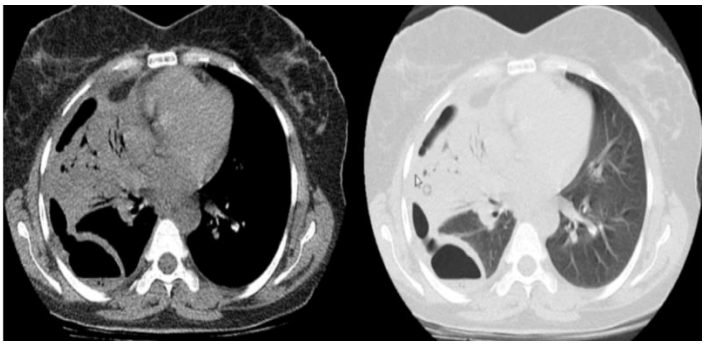

Figure 2: Tomographic section of the mediastinum and lungs

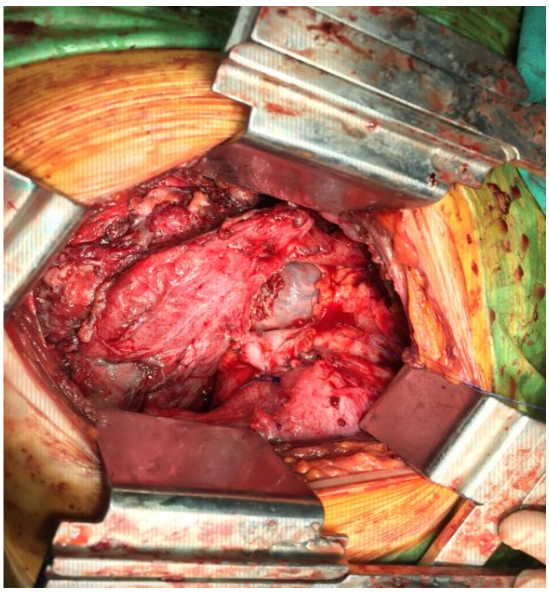

Figure 3: Intraoperative image of diaphragmatic defect and the pericardial patch

Discussion

Intrathoracic complications are rare events in postoperative recurrence of hepatic hydatid cysts; however, they are serious causes of mortality [1].The incidence of spread of liver hydatid cysts to the thoracic area is between 0.6 to 16%, and may be caused by intrapleural negative pressure, disruption in the circulation of the diaphragm due to cyst pressure, and the chemical effect of bile content. Symptoms such as cough, chest pain, shortness of breath, fever may be observed after rupture of liver hydatid cyst into the thoracic cavity [2]. Dyspnea, fever, and cough were observed in our case. Anaphylactic shock may occur if the liver hydatid cyst ruptures into the pleural cavity [3]. In our case, it did not cause anaphylaxis, rather, just a simple itch.

Surgery is the definitive treatment with four main goals: Treatment of the hydatid disease, assurance of free drainage of bile through the common bile duct, cessation of hepatodiaphragmatic communication and ligation of the tracheobronchial fistula [4].

Surgical and percutaneous drainage methods can be utilized in the treatment of the disease. Ultrasonography (USG)-guided percutaneous methods can be used to treat the cysts that cannot be reached during surgery. Recurrence can occur in both surgical and percutaneous methods [5,6]. Percutaneous intervention in hydatid cysts of the liver lowers treatment costs and shortens hospital stay compared to surgical procedures. However, patients treated percutaneously should be well selected. In cases that are not chosen well, serious complications can develop, ranging from abscess and subsequent

bronchobiliary fistula to pneumonia [7]. In our case, the cysts that could not be reached during the operation were treated with percutaneous methods under the guidance of USG. During follow-ups, recurrence and related intrathoracic complications developed. Diaphragmatic defects can be closed primarily [5]. After primary closure of the diaphragmatic defect, we strengthened it with a pericardial patch. During literature review, we saw that strengthening a diaphragmatic defect with a pericardial patch was performed for the first time.

However, the reason we used autologous grafts was that we aimed at strengthening the relatively large defect and the risk of infection is less in autologous grafts, compared with non-autologous grafts. For the patients with postoperative bile leakage, sphincterotomy and biliary catheter placement can be performed endoscopically. This intervention helps reduce and gradually stop the biliary leakage [8]. In our case, sphincterotomy and catheter placement were performed by the gastroenterology department.

Conclusion

Despite being benign disorders, hydatid cysts can cause serious complications. Therefore, we recommend a multidisciplinary approach. Since the risk of infection due to the graft will be lowered, we recommend that the diaphragm be strengthened with an autologous graft, such as a pericardial patch, in relatively large defects of the diaphragm. In addition, even when the diaphragm is sutured primarily, and the patch is placed on, intrathoracic biliary leakage may occur; therefore, endoscopic sphincterotomy should be considered when necessary.

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