Received: 29 October 2022 | Revised: 15 March 2023 | Accepted: 4 April 2023

DOI: 10.1002/ccr3.7212

CASE REPORT

Clinical Case Reports Open Access WILEY

Hydatid cyst of the middle mediastinum complicated by fistulated ascending aorta: A case report and review of the literature

Ali Mehri[1] | **Ali Esparham**[2] | **Mohammad Abbasi Tashnizi**[3] | **Reza Rezaei**[4]

[1]Endoscopic and Minimally Invasive Surgery Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

[2]Student Research Committee, Faculty of Medicine, Mashhad University of Medical Sciences, Mashhad, Iran

[3]Department of Cardiac Surgery, Mashhad University of Medical Sciences, Mashhad, Iran

[4]Department of Thoracic Surgery, Endoscopic and Minimally Invasive Surgery Research Center, Mashhad University of Medical Sciences, Mashhad, Iran

Correspondence
Reza Rezaei, Department of Thoracic Surgery, Endoscopic and Minimally Invasive Surgery Research Center, Mashhad University of Medical Sciences, Mashhad, Iran.
Email: rezaeir@mums.ac.ir

Abstract

Clinicians must pay attention to the clinical presentation and consequences of hydatid cysts in rare sites. Cysts close to the aorta require planning and assistance of a cardiac or vascular surgeon before surgical intervention can be considered.

KEYWORDS

aorta, echinococcosis, hydatid cyst, mediastinum

1 | INTRODUCTION

Hydatid cyst or echinococcal disease is a zoonotic disease. A hydatid cyst involving the aortic wall is extremely rare, even in endemic areas. Here, we report a middle-aged woman who presented with atypical chest pain that further examinations showed a mass located in the mediastinum.

Hydatid cyst or echinococcal disease is a zoonotic disease due to Taenia Echinococcus infection.[1] Hydatidosis is a disease in which dogs are the primary hosts. Sheep and cattle are the secondary hosts and sometimes humans are the intermediate hosts. Humans can be infected by parasite eggs or by direct contact with infected hosts.[2] Based on the World Health Organization's[3] estimation, echinococcosis affected more than one million people worldwide in 2015, causing 19,300 deaths.[4] Hydatid cysts are mostly located in the human liver and in the lung, but other organs such as the pancreas, kidney, spleen, and brain were also reported to be involved.[5] However, there are a few reports about cystic hydatidosis in the aortic wall, especially in the ascending and aortic arch, a rare but life-threatening condition requiring special attention. Here, we report a case of a 53-year-old female patient with a mediastinal hydatid cyst that fistulated to the aortic arch.

2 | CASE PRESENTATION

A 53-year-old female patient was referred to our center with a chief complaint of atypical chest pain. She presented to the emergency department with atypical chest pain, which started 5 days ago. The pain was located in the anterior of the left hemi thorax, which lasted for several hours and had a variable radiation pattern. Her vital signs and physical examinations were normal, except for

Ali Esparham should be considered as co first author.

tachycardia. Electrocardiography and echocardiography were performed on the patient several times without any abnormal findings except mild mitral and tricuspid regurgitation. A chest x-ray was performed for the patient that showed a round mass above the heart and adjacent to the aortic arch (Figure 1).

Further investigation with chest CT scan (with and without IV contrast) reported a 51×75 mm mass in the middle mediastinum, to the left side of the aortic arch with peripheral calcification centers, which is suggestive of lymphangioma (Figure 2). An abdominal CT scan with oral and intravenous contrast revealed one cystic mass in the left lobe of the liver, which is highly suggestive of hydatid cysts. A transthoracic needle biopsy revealed the mediastinum mass pathology, and 2 mL fluid was drained from the cyst. Cytology smear investigation reported hypocellular hemorrhagic background with degenerated cells and inflammatory cells that were negative for malignancy. The patient was referred to our center for surgical removal of the mass. Under general anesthesia, a left posterolateral thoracotomy at the fifth intercostal space was undertaken. A mass was observed at the aortopulmonary window in the middle mediastinal space. In the survey of the pleural space, it was observed that the lung parenchyma was attached to the mass, and this adhesion was separated at first. The mass had a white shell color.

Based on another hydatid cyst in the liver, a mediastinal hydatid cyst diagnosis was made. According to the new diagnosis, a hypertonic saline long gauze was spread, and the mass was aspirated for confirmation. The aspirated secretion was turbid and had dark chocolate-like color. A 3- to 4-cm incision was made in the mass wall. This incision resulted in the release of primarily daughter cysts. Following the release of the daughter cysts, there were blood clots and a significant amount of fresh blood that were also released.

It was realized at that time that the mass had a fistula to the ascending aorta. After diagnosing a fistulated aorta with a hydatid cyst, the medical team requested the presence of a cardiac surgeon in the operating room. After clamping the aortic arch with a satinsky clamp, bleeding was controlled using the remnant of the pre-cyst. Then, the pre-cyst was sutured with 2/0 Nylon on the site with the formation of a pseudo-aneurysm. Although the above procedures were accompanied by too much of blood loss, with the surgical and anesthesia team efforts and with transfusion of blood and blood products, the patient was finally stabilized and transferred to the ICU. The post-surgery pathology report confirmed the hydatid cyst diagnosis (Figure 3).

Upon consultation with the cardiac surgery team, she was transferred to repair the aortic arch aneurysm. Under general anesthesia and deep hypothermia,

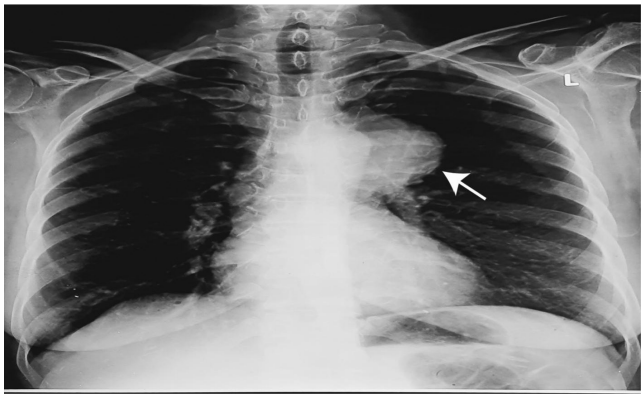

FIGURE 1 Chest X-ray (PA). A round mass in the mediastinum adjacent to the aortic arch.

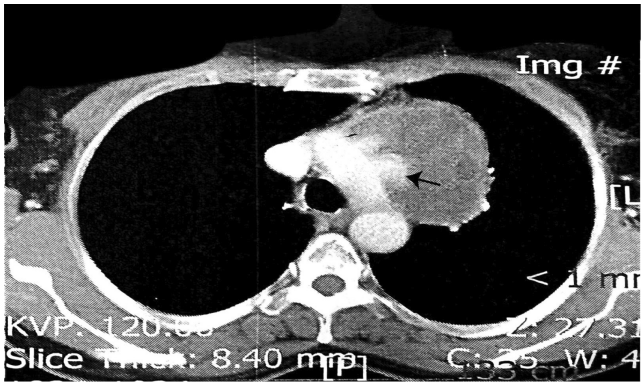

FIGURE 2 Axial section of pre-surgery chest CT scan with IV contrast. A cystic mass in the superior anterior mediastinum along with the evidence of the entry of contrast material into the cyst (arrow) and peripheral calcification centers of the left side of aortic arch.

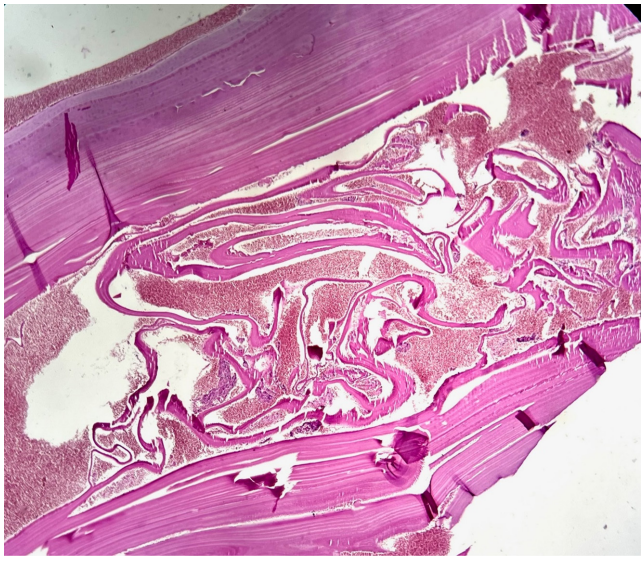

FIGURE 3 Microscopic pathology image of hydatid cyst wall with evidence of hemorrhage (100×).

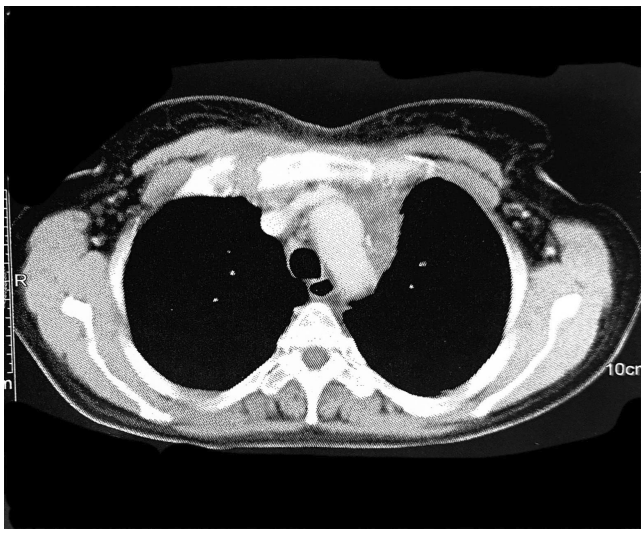

FIGURE 4 Axial section of post-surgery chest CT scan with IV contrast. The repaired aorta, and the mediastinum without residue of hydatid cyst.

cardiopulmonary bypass (CPB) was established. Then the pseudo-aneurysm was removed and reconstruction with Dacron tube graft was done. After the surgery, the patient was transferred to CCU, and after 1 week, she was discharged in stable condition. The patient's condition was uneventful after 6 months, and all physical examinations and investigations were normal except for hydatid cysts in her liver (Figure 4).

3 | DISCUSSION

It has been reported that extrahepatic and extrapulmonary hydatid cysts are rare and account for 15% of cases.[5] Some larvae can pass the hepatic and pulmonary filters and reach systemic circulation. Although cardiac involvement has been reported in 0.5%–2% of patients, there is no available prevalence report for aortic involvement.[6] Nevertheless, hydatid cysts involving the aortic wall are

TABLE 1 Aortic involvement case summary.

Author	Location	Clinical presentation	Treatment
Hendaoui, 1991[15]	Lower mediastinal portion of the thoracic aorta, and splenic and peritoneal cysts	Splenomegaly and several masses in the left flank	Patch
Posacioglu, 1997[17]	Intramural descending aorta	Right-sided chest pain	Dacron graft
Zakhariev, 1997[18]	Descending thoracic aorta	Continuous pain in the left subcostal arch	Dacron graft
Men, 1999[19]	Abdominal and iliac arteries	Claudication of the lower limbs	Aortoiliac graft
Kaynak, 2002[20]	Abdominal aorta	Intermittent claudication and absence of femoral and distal pulses in both lower extremities	Dacron graft
Pulathan, 2004[21]	Abdominal aorta and common iliac arteries	Intermittent abdominal pain	Polytetrafluoroethylene (PTFE) vascular graft
Volpe, 2006[7]	Descending thoracic aorta	Left thoracic and abdominal pain with motor deficit, reduced reflexes, and ataxic movements in the inferior limbs bilaterally	Endograft covered by a Dacron graft
Apaydin, 2007[11]	Aortic arch	Incidental finding by imaging after craniotomy for removing the hydatid cyst	Dacron T-graft
Kamyar, 2008[9]	Descending thoracic aorta	Incidental finding (asymptomatic)	Polytetrafluoroethylene (PTFE)
Dar, 2009[22]	Lower descending thoracic aorta	Gangrenous toes	Definitive surgery for pseudoaneurysm
Buchholz, 2009[23]	Aortobronchial fistula caused by pulmonary hydatidosis	Recurrent significant volume hemoptysis associated and left-sided thoracic pain	Bovine pericardial strips
Gerber, 2012[24]	Ascending aorta and heart	Atypical chest pain and shortness of breath	Dacron graft
Tosyaa, 2015[10]	Ascending aorta	Recurrent arterial embolic events	Dacron graft
Bozok, 2015[25]	Aortic arch	Peripheral artery embolism	–
Chaari, 2018[8]	Descending thoracic aorta	Peripheral arterial embolism	Dacron graft

extremely rare, even in endemic areas. Hydatid cysts that involve the aorta were reported with different clinical presentations based on the hydatid cyst location, extension, and complications. Previous case reports noted chest pain, recurrent arterial embolic events, and even asymptomatic presentation.[7–9] Preoperative diagnosis of extrahepatic and pulmonary hydatid cysts is challenging, even in endemic areas, unless there is a specific consideration for a hydatid cyst. Previous studies recommended that digital subtraction angiography, CT scan, MRI, and ultrasound as effective approaches for detecting the exact relation of cysts and aorta walls in thoracic and aortic hydatid cysts.[10] However, in our case, a CT scan showed a mass located in aortopulmonary window, and the involvement of the aorta was not considered preoperatively. In rare cases, as in ours, the diagnosis of hydatid cysts in uncommon locations may only be confirmed during surgery by visualizing daughter cysts or through pathological examination. Due to the rarity of thoracic hydatid cyst, which involves aorta, there is no specific recommendation for the treatment approach, whether to use a Dacron patch or a graft interposition after excision. Recent studies have shown that using a patch for hydatid cysts involving the aorta can result in various outcomes, including good postoperative condition, false aneurysms, and even fatal bleeding.[11–13] There are several suggested mechanisms for aortic involvement by hydatid cysts. It can be caused by embryos entering the vasa vasorum through an intima defect.[14] Also, it has been suggested that scolices can erode the aortic wall from adjacent organs.[15] In addition, a direct invasion of the hydatid cyst to the aortic wall causing a pseudo aneurysm was reported, which is probably the main mechanism of aortic involvement in our case.[16] As a summary in Table 1, a review of reported aortic hydatid cysts has been provided with accurate location, clinical presentation, and treatment.

4 | CONCLUSION

In conclusion, the hydatid cyst should be considered an important differential diagnosis for each cystic mass, especially in endemic areas. In addition, in cysts near to the aorta, surgery should be carried out under the guidance of a cardiac or vascular surgeon. Also, physicians should pay attention to clinical presentation and complications of hydatid cysts in uncommon locations.

AUTHOR CONTRIBUTIONS

Ali Mehri: Project administration; writing – original draft; writing – review and editing. **Ali Esparham:** Project administration; writing – original draft; writing – review and editing. **Mohammad Abbasi Tashnizi:** Resources. **Reza Rezaei:** Resources; supervision; writing – review and editing.

ACKNOWLEDGMENTS

We thank the patient for giving consent for this case report.

FUNDING INFORMATION

None.

CONFLICT OF INTEREST STATEMENT

All authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

CONSENT

The patient gave written informed permission for publication of this report in line with the journal's patient consent policy.

ORCID

Ali Mehri https://orcid.org/0000-0002-8989-974X

REFERENCES

1. Aribas OK, Kanat F, Turk E, Kalayci MU. Comparison between pulmonary and hepatopulmonary hydatidosis. *Eur J Cardiothorac Surg*. 2002;21(3):489-496.
2. Douba Z, Sinno JA, Jawish H, et al. Unusual localizations of hydatid cysts: a rare case report from Syria. *J Surg Case Rep*. 2020;10:rjaa438.
3. Alexandrou A, Davis PA, Law S, Murphy S, Whooley BP, Wong J. Squamous cell carcinoma and adenocarcinoma of the lower third of the esophagus and gastric cardia: similarities and differences. *Dis Esophagus*. 2002;15(4):290-295.
4. World Health Organization (WHO). Charateristics and details of echinococcosis. http://www.who.int/mediacentre/factsheets/fs377/en/
5. Wani RA, Wani I, Malik AA, Parray FQ, Wani AA, Dar AM. Hydatid disease at unusual sites. *Int J Case Rep Images*. 2012;3(6):1-6.
6. Kir A, Baran E. Simultaneous operation for hydatid cyst of right lung and liver. *Thorac Cardiovasc Surg*. 1995;43(1):62-64.
7. Volpe P, Dalainas I, Ruggieri M, Nano G, Paroni G. Endovascular treatment of the descending thoracic aorta in a patient with a hydatid pseudoaneurysm. *J Vasc Surg*. 2006;44(5):1097-1100.
8. Chaari Z, Damak A, Kardoun N, et al. Primary hydatid cyst of the thoracic aorta revealed by peripheral arterial embolism. *Ann Thorac Surg*. 2018;106(5):e257-e259.
9. Kamyar MM, Ravari H. Thoracic aortic involvement in cystic echinococcosis: a case report. *Electron Physician*. 2017;9(9):5257-5260.
10. Tosya A, Uymaz B, Celebi S, Aybek T. A rare presentation of cystic echinococcosis: aortic involvement. *Interact Cardiovasc Thorac Surg*. 2015;21(4):548-549.

11. Apaydin AZ, Oguz E, Zoghi M. Hydatid cyst involving the aortic arch. *Eur J Cardiothorac Surg*. 2007;31(3):558-560.
12. Viver E, Bianchi L, Callejas J, Martorell A. Hydatid cyst fistula into the aorta. *J Cardiovasc Surg (Torino)*. 1989;30(4):624-626.
13. Hadjiat N, Graba A, Mansouri H. Rupture of retroperitoneal hydatid cyst into the abdominal aorta. *Ann Vasc Surg*. 1987;1(4):483-485.
14. Alper H, Yünten N, Scsener R. Intramural hydatid cysts of pulmonary arteries: CT and MR findings. *Eur Radiol*. 1995;5(6):666-668.
15. Hendaoui L, Siala M, Fourati A, Thameur M, Hamza R. Case report: hydatid cyst of the aorta. *Clin Radiol*. 1991;43(6):423-425.
16. Papacharalambous G, Panoussis P, Andrikopoulos V, Pappas P, Liapi G. Chronic arterial occlusion produced by hydatid cyst development in the lumen of the femoral artery. *Eur J Vasc Endovasc Surg*. 1998;15(6):544-546.
17. Posacioglu H, Bakalim T, Cikirikcioglu M, Yuce G, Telli A. Intramural hydatid cyst of descending aorta complicated by false aneurysm. *Scand Cardiovasc J*. 1999;33(4):242-244.
18. Zakhariev T, Stankev M, Baev B, Iliev R, Tschirkov A. A rare case of hydatid cyst perceived as false aneurysm of thoracic aorta. *J Thorac Cardiovasc Surg*. 1997;113(4):792-793.
19. Sl M, Cn Y, Edgüer TR, Hekimoglu B. Intraaortic growth of hydatid cysts causing occlusion of the aorta and of both iliac arteries: case report. *Radiology*. 1999;213(1):192-194.
20. Kaynak M, Koksal C, Arslan C. Hydatid disease of the abdominal aorta mimicking Leriche syndrome. *EJVES Extra*. 2002;3(1):4-5.
21. Pulathan Z, Cay A, Güven Y, Sarihan H. Hydatid cyst of the abdominal aorta and common iliac arteries complicated by a false aneurysm: a case report. *J Pediatr Surg*. 2004;39(4):637-639.
22. Dar AM, Kawoosa NU, Ahangar AG, et al. Hydatid embolism from a thoracic aortic pseudoaneurysm presenting as gangrenous toes. *Gen Thorac Cardiovasc Surg*. 2010;58(7):344-347.
23. Buchholz S, Sowden D, Stapleton T, Pohlner P, Wright C. Massive haemoptysis due to aortobronchial fistula caused by pulmonary hydatidosis. *Med J Aust*. 2009;190(4):222.
24. Gerber BL, Pasquet A, El Khoury G, et al. Echinococcosis of the heart and ascending aorta. *Circulation*. 2012;125(1):185-187.
25. Bozok Ş, Karakişi SO, Yılmaz Y, et al. Peripheral arterial embolism due to involvement of the aortic arch with hydatid cyst. 2015 Peripheral arterial embolism due to involvement of the aortic arch with hydatid cyst.

How to cite this article: Mehri A, Esparham A, Tashnizi MA, Rezaei R. Hydatid cyst of the middle mediastinum complicated by fistulated ascending aorta: A case report and review of the literature. *Clin Case Rep*. 2023;11:e7212. doi:10.1002/ccr3.7212