Pericardial cyst in a one-year-old boy with ventricular septal defect and patent ductus arteriosus

Nirmal Panthee1*, Sidhartha Pradhan1, Raamesh Koirala1, Anil Dev Pant2, Bishow Pokhrel1, Subhash Chandra Shah3 and Rabindra Bhakta Timala1

Abstract

Background: Pericardial cysts are rare, with the most common etiology being congenital. Ventricular septal defect is the most common congenital heart disease in children. However, the combination of pericardial cyst, ventricular septal defect, and patent ductus arteriosus is extremely rare.

Case presentation: A one-year-old boy with ventricular septal defect and patent ductus arteriosus was planned for surgical correction. Intraoperatively, we made an additional diagnosis of a large pericardial cyst; and the cyst was excised along with ventricular septal defect closure and patent ductus arteriosus ligation.

Conclusions: Pericardial cysts can sometimes be missed with transthoracic echocardiography. Excision of the cyst can safely be done during concomitant cardiac surgery.

Keywords: Pericardial cyst, Congenital heart disease, Ventricular septal defect, Patent ductus arteriosus

Background

Pericardial cysts are rare, with incidence of 1:100,000 [1, 2]. Ventricular septal defects (VSD) have an incidence of 1.56–53.2 per 1000 live births; the detection being dramatically increased due to the use of advanced imaging techniques [3]. Patent ductus arteriosus (PDA) has an incidence of 1 per 2000 term infants [4]. Thus, the combination of pericardial cyst, VSD and PDA is very rare. We have previously reported an adult case of pericardial cyst with atrial septal defect [2]. Here, we report a case of one-year-old boy with pericardial cyst, VSD, and PDA who underwent VSD closure, PDA ligation, and excision of the pericardial cyst.

Case presentation

A one-year-old boy weighing 10 kg was brought to us with repeated lower respiratory tract infections since birth. On examination, he was a playful boy with stable vital signs. Systemic oxygen saturation (SpO2) was 96% in room air. He did not have cyanosis, clubbing, edema, pallor, icterus, or jugular venous distension. Respiratory system examination was unremarkable. Cardiovascular examination revealed normal first and loud second heart sounds with systolic murmur in left lower sternal border.

Baseline hematologic and biochemical profiles were within normal limits. Electrocardiogram showed sinus rhythm with heart rate of 113 beats/min. Chest X-ray showed cardiothoracic ratio (CTR) of 0.55 with plethoric lung fields. Transthoracic echocardiography revealed dilated left ventricle (3.6/2.1 cm) with ejection fraction (EF) of 60%, anterior malaligned perimembranous VSD measuring 1 cm with left to right shunt, 3 mm PDA with left to right shunt, and severe pulmonary artery hypertension (tricuspid regurgitation pressure gradient...
(TRPG): 50 mmHg). With this diagnosis, VSD closure and PDA ligation was planned.

Under general anesthesia, following median sternotomy and pericardiotomy, to our surprise, we noticed a freely mobile cystic lesion (3 × 4 cm) in the inferior surface of heart, which was attached by a stalk to the posterior pericardium near the atrioventricular groove on the left side. (Fig. 1, Additional file 1: video 1 and 2). Cardiopulmonary bypass (CPB) was established with ascending aortic and bicalve cannulation. After initiation of cardiopulmonary bypass, PDA was ligated with number 2 silk suture. After aortic cross clamp, cardiac arrest was achieved by antegrade cardioplegia. Right atrium was opened and VSD was closed with expanded polytetrafluoroethylene (ePTFE) patch using 6–0 prolene suture (Fig. 2). Right atrium was closed and aortic cross clamp was removed. Before weaning from the cardiopulmonary bypass, the apex of the heart was lifted up and the cyst was excised using electrocautery. The cyst contained clear fluid and the resected cyst is shown in Fig. 3. Total CPB time was 117 min and aortic cross clamp time was 80 min. After excision of the cyst, CPB was weaned off. The patient was transferred to pediatric intensive care unit with minimal inotropic support. He was extubated on the same day; transferred to general ward on 2nd postoperative day; and discharged home on 5th postoperative day. The histopathology of the resected specimen showed fibro-collagenous wall with mild inflammation and mesothelial lining consistent with pericardial cyst (Fig. 4).

Discussion
Pericardial cysts are rare, with most common etiology being congenital [5]. Most pericardial cysts are asymptomatic and diagnosed incidentally, although hemorrhage into the cyst leading to chest pain, and cardiac tamponade have been reported [1]. Most of the available literature is in the form of case or case series report in asymptomatic adult patients. Reports of pericardial cysts in children have been scarce [6]. As of 2003, fewer than 20 cases of pericardial cysts were reported in the worldwide literature in children less than 18 years of age [7]. To our best knowledge, the youngest report
was that of a 2-year-old boy [8]. Most of the pericardial cysts reported in pediatric age group (2-year-old boy, 3-year-old girl, 10-year-old girl) are from Japan [1, 8, 9]. The oldest case of pericardial cyst reported in the literature is of a 102-year-old woman from Cyprus [10]. We believe ours is one of the rarest reports of a one-year-old child with associated VSD and PDA.

Most of the cysts are diagnosed by transthoracic echocardiography; however, in our case, we missed to detect during preoperative echocardiography. Echocardiography is operator-dependent; and most of the times, we look for intracardiac lesions; and extracardiac lesions could be overlooked. Also, chest X-ray would show cardiomegaly or abnormal shaped cardiac silhouette; [9] however, in our case, the primary diagnosis of VSD and PDA would itself result into cardiomegaly, and due to location of the cyst (immediately behind the heart), abnormal silhouette was not expected. Computed tomography (CT) scan would definitely have detected the cyst. However, we do not routinely perform CT scan for patients with isolated VSD unless the VSD is part of tetralogy of Fallot.

Immediately after opening the chest, we were not sure about the content of the cyst. Keeping in mind the rare possibility of hydatid cyst, we deferred to excise the cyst until after the intracardiac procedure was completed and cardiac chambers were closed to avoid spillage of the cyst contents inside the heart itself. We do not believe that the management would have been significantly different from ours in any other cardiac center. We also do not believe that we have done a marvellous job by resecting a pericardial cyst in a child with VSD and PDA. However, we do believe that reporting our case would definitely add to the available literature given that this is a rare entity.

Conclusions

We encountered pericardial cyst as an incidental finding during cardiac surgery and we have successfully resected a pericardial cyst as a concomitant operation during VSD closure and PDA ligation in a very young child.

Abbreviations

CPB: Cardiopulmonary bypass; CT: Computed tomography; CTR: Cardiothoracic ratio; EF: Ejection fraction; ePTFE: Expanded polytetrafluoroethylene; H&E: Hematoxylin and Eosin; PDA: Patent ductus arteriosus; TRPG: Tricuspid regurgitation pressure gradient; VSD: Ventricular septal defect.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s43044-022-00278-6.

Acknowledgements

The authors would like to thank Dr Ashish G Amatya for taking intraoperative videos.

Author contributions

NP: concept, original manuscript draft, data collection, patient management; SP: manuscript revision, supervision, patient management; RK: manuscript revision, supervision, patient management; AP: manuscript revision, data analysis; BP: manuscript revision, data collection, patient management; SS: manuscript revision, data collection, patient management; RT: manuscript revision, supervision, patient management. The final manuscript was approved by all authors prior to submission.

Funding

None.

Availability of data and materials

Upon reasonable request, the corresponding author will share the data and materials used in the manuscript.

Declarations

Ethics approval and consent to participate:

Not applicable.

Consent for publication

Informed consent for publication of the report was obtained from the parents of the child.

Competing interests

The authors declare that they have no competing interests.

Author details

1 Department of Cardiac Surgery, Shahid Gangalal National Heart Center, Bansbari, Kathmandu, Nepal. 2 Department of Pathology, Tribhuvan University Teaching Hospital, Maharagunj, Kathmandu, Nepal. 3 Department of Pediatric
Cardiology, Shahid Gangalal National Heart Center, Bansbari, Kathmandu, Nepal.

Received: 27 January 2022   Accepted: 28 April 2022
Published online: 16 May 2022

References

1. Tanoue Y, Fujita S, Kanaya Y, Tominaga R (2007) Acute cardiac tamponade due to a bleeding pericardial cyst in a 3-year-old child. Ann Thorac Surg 84:282–284. https://doi.org/10.1016/j.athoracsur.2007.01.058
2. Acharya A, Koirala R, Rajbhandari N, Panthee N (2018) Pericardial cyst causing cardiac compression and shunt reversal at atrial level. Nepalese Heart J 15(2):37–38. https://doi.org/10.3126/njh.v15i2.21475
3. Minette MS, Sahn DJ (2006) Ventricular septal defects. Circulation 114:2190–2197. https://doi.org/10.1161/CIRCULATIONAHA.106.618124
4. Schneider DJ, Moore JW (2006) Patent ductus arteriosus. Circulation 114:1873–1882. https://doi.org/10.1161/CIRCULATIONAHA.105.592063
5. Kar SK, Ganguly T, Dasgupta S, Mitra M, Bhattacharya R (2015) Pericardial cyst: a review of historical perspective and current concept of diagnosis and management. Interventional Cardiol J 1:1
6. Kumar S, Jain P, Sen R, Rattan KN, Agrawal R, Garg S (2011) Giant pericardial cyst in a 5-year-old child: a rare anomaly. Ann Pediatr Cardiol 4(1):68–70. https://doi.org/10.4103/0974-2069.79629
7. Noyes BE, Weber T, Vogler C (2003) Pericardial cysts in children: Surgical or conservative approach? J Pediatr Surg 38:1263–1265. https://doi.org/10.1016/S0022-3468(03)00285-9
8. Masuoka A, Sakurai H, Shiraishi M, Yoshiba S, Katogi T, Suzuki T (2015) Giant epicardial cyst presenting as acute cardiac tamponade in a 2-year-old boy. Asian Cardiovasc Thorac Ann 23(7):866–868. https://doi.org/10.1177/0218492314566331
9. Eto A, Arima T, Nagashima A (2000) Pericardial cyst in a child treated with video-assisted thoracoscopic surgery. Eur J Pediatr 159:889–891. https://doi.org/10.1007/PL00008362
10. Collins KL, Zakhanious F, Mandal AKJ, Missouri CG (2018) Pericardial cyst: never too late to diagnose. J Clin Med 7:399. https://doi.org/10.3390/jcm7110399

Publisher’s Note
Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.