The Vanishing Act: Post-Pericardiocentesis Pericardial-Pleural Fistula in Secondary Hyperparathyroidism

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Abstract

Pericardial-pleural fistula (PPF) may complicate between 0.8% to 6% of echocardiography-guided pericardiocentesis procedures, but reports are sporadic. A 22-year-old gentleman with underlying end stage renal disease complicated by secondary hyperparathyroidism presented to general medical ward with worsening left pleuritic chest pain and shortness of breath for one week. Ultrasonographic investigations revealed global pericardial effusion, with no pleural effusion initially. After echocardiographic-guided pericardiocentesis via the apical approach, the patient developed respiratory distress. Follow-on ultrasound revealed a new pleural effusion with resolved pericardial effusion. The patient recovered with drainage of the pleural effusion. Bacterial and tuberculous cultures were negative, and fluid cytology was benign. Hyperparathyroidism may have contributed to increased risk of PPF due to pericardial calcification. Clinicians should be aware about the potential risk of PPF after pericardiocentesis in patients with hyperparathyroidism.

Keywords: pericardial effusion, pericardial-pleural fistula, secondary hyperparathyroidism

Introduction

Pericardial-pleural fistula (PPF) is a communication between the pericardium and pleural space, resulting in movement of pericardial fluid from the pericardial sac into the lower-pressure pleural space.[1] PPF may complicate between 0.8% to 6% of echocardiography-guided pericardiocentesis procedures.[2,3] In this case report, we report on a 22-year-old gentleman who developed pericardial effusion due to hyperparathyroidism, complicated by pericardial-pleural fistula.

Case report

A 22-year-old gentleman with underlying end stage renal disease complicated by secondary hyperparathyroidism presented to general medical ward with worsening left pleuritic chest pain and shortness of breath for one week. He denied any cough, fever, night sweats, or constitutional symptoms. Chest radiograph on admission revealed globular cardiac shadow with widened cardiothoracic ratio. There were no evolving changes on electrocardiogram and serum troponin was not raised. On examination, he had a blood pressure of 146/71 mmHg, pulse rate of 88 beats per minute, oxygen saturation of 91% under room air and was afebrile. His jugular venous pressure was raised and cardiac auscultation revealed a pericardial rub. Point-of-care echocardiogram demonstrated global pericardial effusion with no right ventricular collapse, while lung ultrasound revealed multiple vertical reverberation artefacts, also known as

B-lines, but no pleural effusion. He was admitted for oxygen supplementation and decongestion therapy. In view of raised inflammatory markers (CRP 76.2 mg/L), antibiotics commenced to empirically treated for pneumonia. However, blood culture, sputum culture, and sputum smear for acid-fast bacilli were all negative.

Formal transthoracic echocardiography on day three of admission revealed mitral annular calcification with extension into the left ventricular outflow track, not causing obstruction, with pericardial effusion measuring two centimeters maximally without evidence of tamponade (Figure 1). On day seven of admission, repeat bedside echocardiography revealed similar pericardial effusion (Supplementary Video 1). In view of persistent tachycardia and due to suspicion of tuberculous pericarditis, echocardiography-guided pericardiocentesis through the apical approach was performed, revealing haemoserous pericardial fluid. Emergent pericardial fluid investigations revealed protein 46 g/L (reference range protein exudate: > 25 g/L) and lactate dehydrogenase of 1237 IU/L (reference range LDH exudate: > 200 IU/L) which was suggestive of an exudative effusion. Post-pericardiocentesis, the patient complained of worsening shortness of breath and required increasing oxygen supplementation. Repeated chest radiograph showed a new left pleural effusion with no pneumothorax (Figure 2 Panel A).

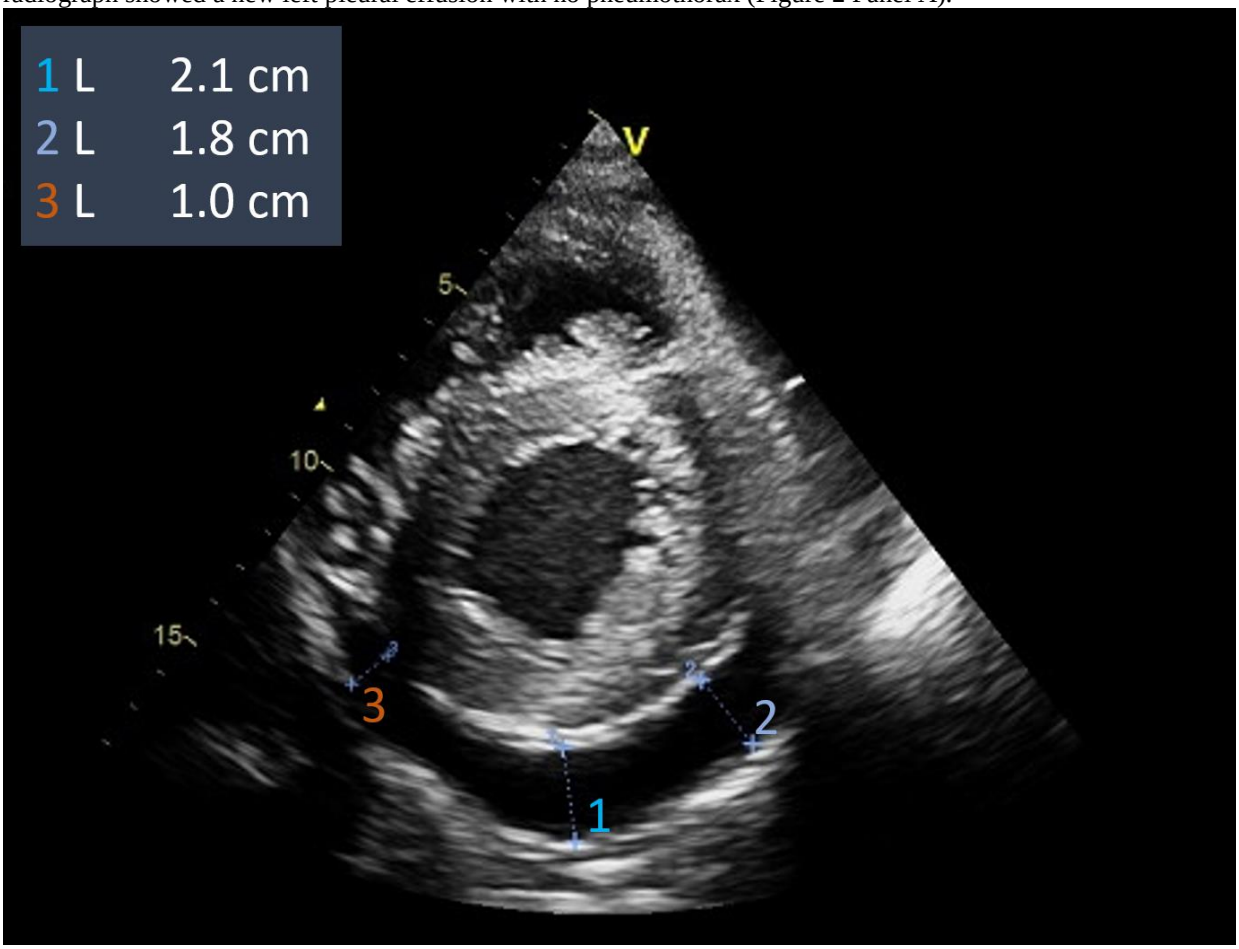

Figure 1: Echocardiogram (parasternal short axis view) demonstrates a global pericardial effusion deepest posteriorly.

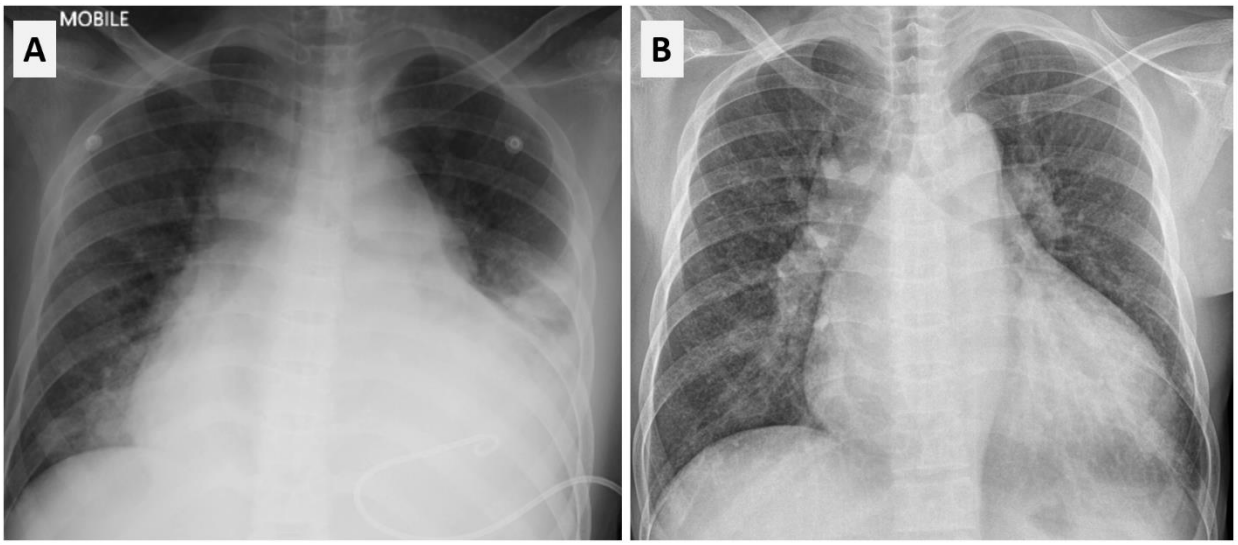

Figure 2: Panel A shows the post-pericardiocentesis chest radiograph which demonstrated a new left pleural effusion. Panel B shows follow-up chest radiograph showing no recurrence of pleural effusion.

Bedside scan showed a new left pleural effusion up to upper midzone, with absence of pericardial effusion. Thoracocentesis and pigtail drain insertion was performed. Pleural fluid gross appearance and biochemical picture correlated well with pericardial fluid; haemoserous in colour, and exudative with pleural fluid LDH 1123 IU/L, protein 45 g/L, adenosine deaminase (ADA) 21.67 U/L (cut off value pleural ADA 29.6U/L). Both cytology and culture were negative for malignant cells and no organisms were isolated. Post-thoracocentesis, patient was weaned to room air and discharged home. Pericardial and pleural fluid tuberculosis cultures were both negative. A repeat chest radiograph six weeks post-thoracocentesis revealed revealed no recurrence of pleural effusion (Figure 2 Panel B). With bacterial, malignant, tuberculous effusion ruled out; and considering the echocardiogram findings of mitral annular calcification, a final diagnosis of secondary hyperparathyroidism-related pericardial effusion was made with multidisciplinary input from the cardiology team.

Table 1: Serial biochemical investigations revealed increasing iPTH levels over time.

Biochemical markers	**Reference range**	**April 2021**	**Sept 2021**	**April 2022**	**July 2022**
Sodium (mmol/L)	135 - 145	134	133	135	132
Potassium (mmol/L)	3.5 -5.1	3.5	3.8	3.6	4.1
Urea (mmol/L)	2.5 - 6.7	17.3	16.5	17.6	18.3
Creatinine (umol/L)	54.0 - 98.0	698.9	722.4	758.2	742.5
iPTH (pg/ml)	15.0 - 68.3	435	782	659	1473
Calcium (mmol/L)	2.10 - 2.55	2.12	2.16	2.12	2.10
Phosphate (mmol/L)	0.74 - 1.52	1.95	1.78	1.96	2.54

Discussion

This is the first reported incidence of pericardial-pleural fistula (PPF) in the setting of secondary hyperthyroidism. Post-pericardiocentesis PPF has been previously reported in only four case reports and one case series.[1,2,3,4] Of these four reports, two of the pleural effusion occurred on the left side,[1,4] while the other two sources did not report on the laterality of the pleural effusions.[2,3] One of the cited risk factors that may have led to PPF in our case was the apical approach; this is due to the adjacent pleural space, which is absent in the subxiphoid approach.[3] In one case, it was proposed at the use of a rigid dilator for pericardial drain placement may have pulled vertically through an area of pleuro-pericardial overlap, resulting in PPF.[4]

Aside from being a complication of pericardiocentesis, pericardial-pleural fistula (PPF) has been reported in cardiac procedures, namely epicardial catheter ablation for ventricular tachycardia,[5] implantable cardioverter defibrillator insertion,[6] or due to a stab injury which penetrated the right ventricle.[7]

Identifying PPF is crucial to prevent repeated attempts to aspirate an empty pericardial sac, which may result in disastrous injury to the myocardium or coronary vessels.[1] Traditionally, there are no technical means to avoid PPF, hence high level of vigilance and early detection of PPF complication is needed.[3]

Pericardial effusion associated with secondary hyperparathyroidism was first reported in 1985 in Japan [8], and may exist concurrently with a calcified pericardium.[9] The calcification is attributed to the process of extraskeletal calcification due to secondary hyperparathyroidism.[9] Two pathophysiological pathways have been proposed; first, alterations in calcium metabolism result in ectopic calcification, or second, dystrophic calcification leading to tissue damage.[10] Yet another etiological explanation to consider is pericardial calcification due to haemodialysis itself.[9] In patients found to have calcified pericardium with normal serum phosphate, calcium, and parathyroid hormone levels; it can be inferred that the calcification of pericardium is associated with hemodialysis.[9]

We posit that our patient's calcified pericardium is more susceptible to fistula formation due to it being less malleable. Insertion of the pericardiocentesis needle may have caused a residual "flap" that may have been unable to oppose back due to its rigid, calcified nature. Furthermore, as our approach was apical, this may have caused the needle to traverse through an area of both pleura and pericardium.

Conclusion

Clinicians should be vigilant in performing pericardiocentesis on patients with underlying hyperparathyroidism. Further studies are needed to understand the predictive factors of PPF and how to avoid this complication from occurring.

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Conflict of interest

The authors declare that they have no conflict of interest.

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Authors' contribution

TSE – Data curation, investigation, writing – review and editing; LEN – conceptualization, data curation, visualization, writing – original draft; CWL – writing – review and editing, resources, MNW – Data curation, investigation, NA –supervision ;PN –supervision

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