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Case Report

A Case of Incidentally Discovered Congenital Complete Pericardial Defect during Lobectomy for Lung Cancer: A Case Report and Literature Review

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An 82-year-old male patient underwent a left upper lobectomy with anterolateral thoracotomy for lung cancer. Although a complete left-pericardial defect was observed during surgery, the pericardial repair was not performed because the left lower lobe remained and the heart was considered stable. Postoperative pathological examination revealed primary synchronous double-lung squamous-cell carcinoma (pathological stage pT2a(2) N0M0 stage IB). He was discharged without complications on postoperative day 8. Leftward displacement of the heart and left diaphragmatic elevation, suspected of phrenic-nerve paralysis, were found in the chest X-ray after discharge. However, the patient's overall condition remained unaffected at the 5-month postoperative follow-up. To assess the need for pericardial repair, we compared cases of complete pericardial defects observed during lobectomy or pneumonectomy reported in the literature. Only one of 12 cases occurred postoperative death despite pericardial repair, and that case combined pectus excavatum and pericardial defects. Our assessment indicated that pericardial repair might not be necessary, excluding complex cases.

Keywords: congenital complete pericardial defect, lobectomy, pericardial repair

Introduction

Congenital pericardial defects are rare anomalies often first observed intraoperatively.[1)] Most affected individuals are asymptomatic, and finding these defects during preoperative tests can be difficult. Complete pericardial defects are even more rarely diagnosed intraoperatively if only lobectomy or pneumonectomy is performed. Repair of pericardial defects to prevent cardiac herniation after major lung resection is controversial. Here, we report a case of an 82-year-old man who underwent left upper lobectomy for synchronous double-lung squamous-cell carcinoma. A complete pericardial defect was incidentally discovered during the surgery. Although we did not perform pericardial repair, we compiled and compared previous cases of pericardial defects repaired by lobectomy and pneumonectomy to assess whether or not pericardial repair surgery should be performed.

Case Report

An 82-year-old man was referred to us because two solid masses were identified in the left upper lung on

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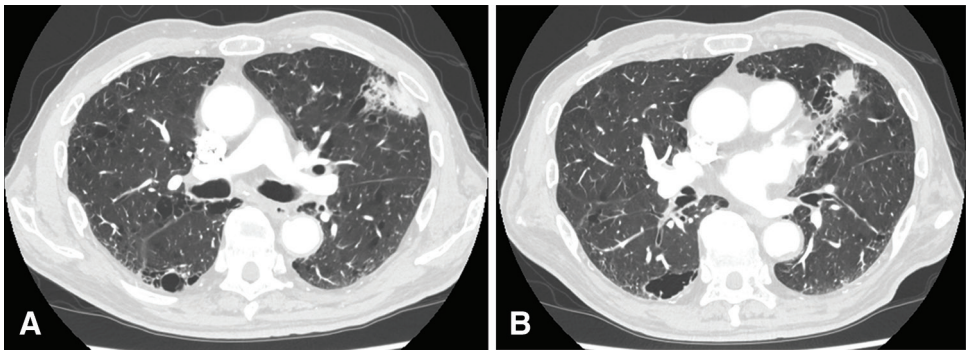

Fig. 1 Preoperative contrast-enhanced CT images. (**A**) A solid tumor shadow measuring 37 mm is observed in the left upper lobe S3. (**B**) A solid nodular shadow measuring 29 mm is observed in the left upper lobe S4. CT: computed tomography

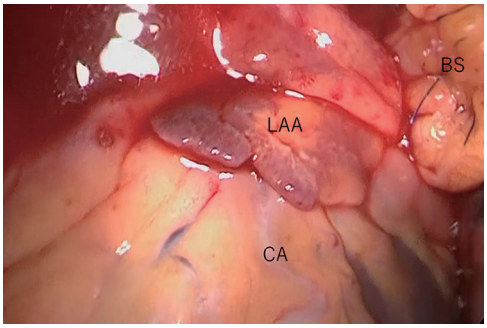

Fig. 2 An image taken during the surgery. The left side of the heart and LAA were exposed. CAs were also visible. The BS was covered with pericardial fat tissue. LAA: left atrial appendage; CA: coronary arteries; BS: bronchial stump

chest computed tomography (CT) performed during a medical checkup as a follow-up for chronic obstructive pulmonary disease. Contrast chest CT showed a 37-mm solid mass in the left lung S3 segment and a 29-mm solid nodule in the left lung S4 segment (**Fig. 1**). Because interstitial pneumonia and pulmonary emphysema were indicated, there was no volume loss in the left lower lobe. We performed a transbronchial biopsy twice, and the tissue diagnosis obtained from S4 was squamous-cell carcinoma. Positron emission tomography with 2-deoxy-2-(^{18}F)fluorodeoxyglucose (FDG-PET) exhibited accumulation of FDG in the subaortic lymph nodes (#5) with a standardized uptake value maximum of 6.3. The accumulation suggested the possibility of lymph node metastasis; however, distant metastasis was not observed, including in the brain and on head magnetic resonance imaging. The preoperative diagnosis was cT2aN2M0, Stage IIIA with single station N2.

Lateral thoracotomy with video-assisted thoracic surgery was performed. No pleural effusion or dissemination was observed. Left upper lobectomy and lymph node dissection (ND2a-2) were then performed. When we reexamined the thoracic cavity after lymph node dissection, we noticed that the entire left side of the heart was exposed (**Fig. 2**). Although we were concerned about cardiac hernia related to decreased cardiac bearing capacity due to decreased lung volume, we decided not to repair the defect because the remaining left lower lobe after the left upper lobectomy would adequately support the heart. The chest drainage tube was removed on postoperative day 2, and the patient was discharged on postoperative day 8. Subsequent pathological examination revealed primary synchronous double-lung squamous-cell carcinoma pT2a(2)N0M0, Stage IB. Leftward displacement of the heart and left diaphragmatic elevation, suspected of phrenic-nerve paralysis, were observed on the chest X-ray after discharge (**Fig. 3**) and at the 5-month postoperative follow-up. However, the patient's overall condition remained good at the 5-month postoperative follow-up.

Discussion

Pericardial defects, which are either acquired or congenital, are uncommon. The congenital variant is estimated to occur in <1/10000 individuals.[2)] This variant predominantly affects males, with a male-to-female ratio of 3:1, and familial cases are infrequent. Congenital pericardial defects are categorized based on the location and size of the pericardium absence. Complete left-sided defects are the most prevalent, accounting for approximately 70% of all cases of pericardial defects.[2)] Perna et al. proposed that left-side congenital pericardial defects arise from premature degeneration of the left duct of Cuvier, leading to incomplete formation of the left pleuropericardial fold. By contrast, the right duct of Cuvier typically matures into the superior vena

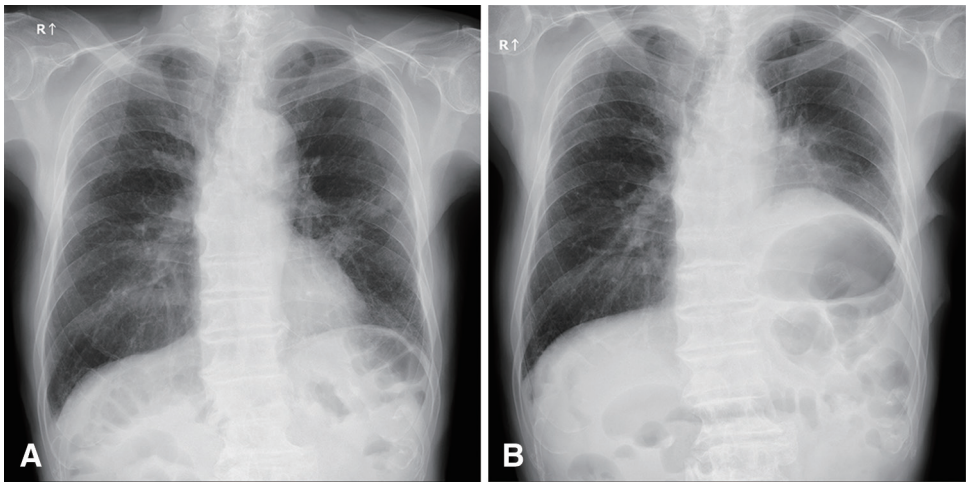

Fig. 3 The chest X-rays were taken before and 22 days after the surgery. (**A**) There were no indications of pericardial defects before the surgery. (**B**) After the surgery, leftward displacement of the heart and left diaphragmatic elevation, suspected of phrenic-nerve paralysis, were observed.

cava, contributing to the closure of the right pleuropericardial membrane. Consequently, defects on the right side are considerably less common.[3] We were unable to identify the complete left-sided pericardial defect preoperatively; however, it was noted upon postoperative review. No defect was observed on the right side (**Supplementary Fig. 1**).

In certain instances, patients may exhibit symptoms such as chest pain, palpitations, dyspnea, and absence of seizures. Typically, however, the majority of patients with congenital pericardial defects remain asymptomatic. The presence of a defect is often discovered incidentally during surgical procedures or autopsy.[4] Although one-third of pericardial defects are linked to congenital anomalies involving the heart, lungs, bronchi, chest wall, or diaphragm, our patient did not exhibit any of these associated conditions. Ogawa et al.[5] documented a case of left-phrenic-nerve paralysis attributable to compression of the left auricle caused by a congenital partial left-pericardium defect. By contrast, Kaneko et al.[6] reported a case with a complete pericardial defect and dual passage of the phrenic nerve. They demonstrated that congenital pericardial defects may also influence phrenic-nerve development or function. In our case, we observed that the left phrenic nerve passed through the ventral side of the defect during the operation. Therefore, we concluded that there were no significant alterations noted in the phrenic nerve. However, we found postoperative diaphragmatic elevation suspected of phrenic nerve paralysis on X-ray radiography, but it remains unclear whether or not the postoperative phrenic-nerve paralysis, in this case, was caused by the surgical procedure or by compression due to cardiac deviation.

If a partial pericardial defect is detected during an operation, repair should be considered according to the defect's location and size and the lung volume to be resected.[1,7] Borrie argued that partial defects should be closed because of the dangers of herniation of the heart with strangulation.[8] Treatment options include patch closure of the defect, enlarging the defect to prevent herniated-tissue incarceration, pericardiectomy, or pericardioplasty. In cases of herniation of the left atrial appendage, appendage excision and closure of the pericardial defect with a patch can be performed.

We incidentally detected a complete left-sided pericardial defect during left-upper lobectomy for lung cancer and decided to leave it untreated because the remaining left lower lobe after the left-upper lobectomy would adequately support the heart. We have compiled previous reports on the intraoperative discovery and management of congenital complete pericardial defects during lobectomy or pneumonectomy (**Table 1**). All cases were left-sided. Including the current case, there were seven cases of upper-lobe resection, four cases of lower-lobe resection, and one case of pneumonectomy.[9–19] Repair procedures were performed in two cases of upper-lobe resection and one case of lower-lobe resection.[10,13,19] In the case of lower-lobe resection in which repair was performed, the authors reported that repair was performed after considering the effect of decreased lung volume on cardiac support.[19] In the cases in which repair was

Table 1 Summary of case reports on lobectomy and pneumonectomy with congenital complete pericardial defects

Case	Published year	Age	Sex	Surgery	Side	Disease	Treatment for pericardial defect	Postoperative complication and course	Deviation of the heart	References
1	1985	14	F	LUL	L	Bronchogenic cyst	+	Sudden death	Sudden death	Honda et al.[10]
2	1989	61	M	LLL	L	LC	–	No complication	N/D	Tsukada et al.[11]
3	2000	70	M	LUL	L	LC	–	Satisfactory at 7 years postoperatively	+	Hanaoka et al.[12]
4	2001	74	M	LUL	L	LC	+	N/D	N/D	Yamaguchi et al.[13]
5	2011	61	M	LPn	L	LC	–	Satisfactory at 7 months postoperatively	+	Shimada et al.[9]
6	2019	49	M	LUL	L	LC	–	Satisfactory at 1 month postoperatively	–	Sakaguchi et al.[14]
7	2021	75	M	LUL	L	LC	–	No complication	N/D	Nakamura et al.[15]
8	2021	78	M	LLL	L	LC	–	No complication	N/D	Shiikawa et al.[16]
9	2022	63	M	LUL	L	LC	–	Satisfactory at 6 months postoperatively	–	Fiorelli et al.[17]
10	2023	75	M	LLL	L	LC	–	Satisfactory at 20 months postoperatively	–	Miura et al.[18]
11	2023	N/D	N/D	LLL	L	Metastasis	+	Satisfactory at 7 years postoperatively	N/D	Takeda et al.[19]
12	2024	82	M	LUL	L	LC	–	Left diaphragmatic elevation, satisfactory at 5 months postoperatively	+	Our case

M: male; F: female; LUL: left upper lobectomy; LLL: left lower lobectomy; LPn: left pneumonectomy; L: left; LC: lung cancer; N/D: not described

not performed, none of the patients experienced postoperative complications regardless of the surgical procedure.[9,11,12,14–18] Shimada et al. suggested that repair is unnecessary in cases of asymptomatic left-sided complete pericardial defects during ipsilateral pneumonectomy.[9] However, there have been cases of sudden death in left-sided cardiac herniation caused by unrepaired complete pericardial defect after left-upper-lobe resection for an inflammatory bronchogenic cyst with pectus excavatum.[10] Pericardial repair was performed after the patient suddenly deteriorated, but the patient could not be saved. In cases with complex complications, such as pectus excavatum, pericardial repair should be considered.

In conclusion, we encountered an incidental finding of a complete pericardial defect during a left upper lobectomy for lung cancer. In this patient, pericardial repair was not performed because of the presence of the remaining lower lobe. Except for the complex cases mentioned above, it has been generally considered that pericardial repair is not necessary in cases of complete congenital pericardial defects, regardless of the extent or location of lobe resection.

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from the patient for the publication of this report and its accompanying images.

Funding

None.

Data availability statement

Not applicable.

Author contributions

TS conceived and designed the study and drafted the manuscript and figures. TK and HW performed surgery. TK, SN, TM, and TFC-Y participated in the study conception and design and reviewed the manuscript.

Disclosure statement

There is nothing to declare.

Supplementary Material

Supplementary Fig. 1 The postoperative review of the CT image revealed the absence of pericardial continuity between the yellow arrowheads. CT: computed tomography

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