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Chylopericardium following thoracoscopic resection of a mediastinal cyst: A case report

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ABSTRACT

INTRODUCTION: Thoracic duct cysts are very rare, and diagnosis is often difficult. We report a rare case of chylopericardium following thoracic duct cyst resection. There are no established guidelines on the management of such cases. We reviewed the literature on postoperative complications after thoracic duct cyst resection, and conducted the first thorough review of the etiology and management of chylopericardium in surgical cases.

PRESENTATION OF CASE: A 54-year-old male presented with cardiac tamponade due to chylopericardium. He had undergone resection of a thoracic duct cyst 2 years previously, which was complicated by postoperative chylothorax. Chyle accumulation resolved with conservative treatment.

DISCUSSION: Chylothorax is a frequent complication following thoracic duct cyst resection, especially in cases where no intraoperative diagnosis is reached. Diagnosis may be difficult due to anomalous location of the cyst, as in our case. Chylopericardium is rarely reported, and may have occurred in our case because of prior pleurodesis. Chyle accumulation can reportedly be managed with diet restrictions in over half of reported cases, especially in cases of lung or mediastinal tumor resection.

CONCLUSION: The most important points highlighted by this rare case of chylopericardium secondary to thoracic duct cyst resection are: 1) pedicles should be ligated in cyst resections, regardless of location; 2) careful assessment in the initial surgery may help identify the point of leakage; 3) low-fat diet is the first choice in the initial management of postoperative chylopericardium, but surgical repair may be considered in cases with no response after > 2 weeks of conservative treatment.

1. Introduction

Thoracic duct cysts are cysts that arise in the thoracic duct through congenital or degenerative processes and are occasionally associated with chylothorax [1]. Herein, we report a rare case of chylopericardium following surgical resection of a thoracic duct cyst performed in a community hospital. We also reviewed the relevant literature, focusing on the etiology of thoracic duct cysts and the management of postoperative chylopericardium. There are no established guidelines for the management of such cases [2], and the present report provides relevant information for surgeons encountering postoperative chylopericardium. This manuscript has been reported in line with the SCARE criteria [3].

2. Case presentation

A 54-year-old Asian male presented at the emergency department with nausea and hypotension. He had a past history of cervical spondylosis, and was receiving medication for hypertension. There was no family history of genetic disorders. He had a smoking history of 26 pack-years.

A mediastinal mass had been detected 2 years previously on computed tomography (CT). Endobronchial ultrasound-guided transbronchial needle aspiration was performed, revealing that the mass contained serous fluid with no evidence of malignant cells. Follow-up CT revealed enlargement of the mass (Fig. 1A). The mediastinal tumor was resected via video-assisted thoracoscopic surgery (Fig. 1B). Pathological diagnosis revealed the mass to be a mediastinal celomic cyst (Fig. 1C). Postoperatively, the patient developed chylothorax; this improved with conservative treatment, comprising a low-fat diet and chemical pleurodesis. Additional immunohistochemical evaluation following the patient's recovery revealed that the epithelium was D2-40-positive

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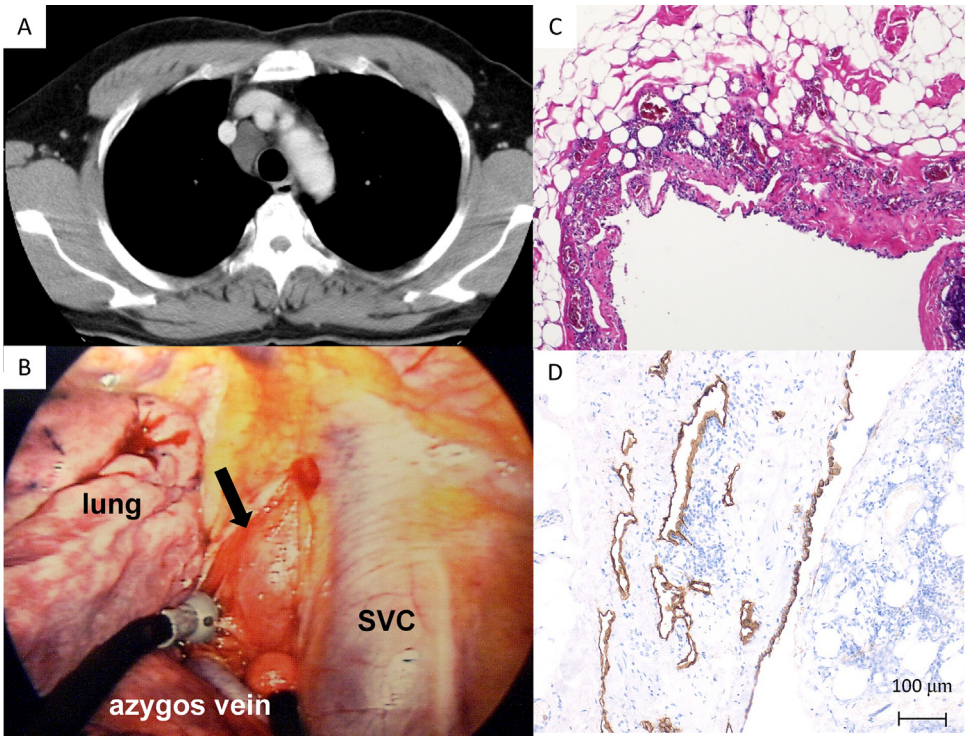

Fig. 1. Computed tomography and surgical findings at initial treatment of the mediastinal cyst 2 years previously. A) Computed tomography revealed a cystic lesion in the upper mediastinum. B) A mediastinal cyst containing serous fluid was located adjacent to the superior vena cava and superior to the azygos vein. C) Histopathological examination confirmed the diagnosis of a celomic cyst. D) Immunohistochemistry revealed that the epithelium was D2-40 positive.

and negative for CD31 and CD34, leading to the diagnosis of a thoracic duct cyst (Fig. 1D).

On physical examination, the patient was pale and had cold extremities. His temperature was 35.6 °C, SpO_2 was 88%, systolic blood pressure was 60 mmHg, and heart rate was 100 bpm. Chest radiography showed an enlarged cardiac silhouette and fluid in the right hemithorax (Fig. 2A). Chest CT revealed fluid collection in the upper mediastinum; there was also fluid in the pericardium and the thoracic cavity (Fig. 2B). Echocardiography led to the diagnosis of cardiac tamponade. A catheter was placed in the pericardium by a cardiologist, and 190 ml of yellowish-white fluid was drained (Fig. 2C). The triglyceride content of the fluid was 560 mg/dL, and therefore chylopericardium was diagnosed.

Hemodynamic support was administered via inotropic agents and fluid replacement. Left thoracentesis was performed on the 8th day after admission, and 400 ml of fluid was drained. Drainage from the pericardium continued at 50–300 ml/day for 2 weeks while the patient was treated with a low-fat diet. The amount of pericardial fluid drainage was reduced from day 16, and the catheter was removed on day 31. The patient was discharged on a low-fat diet on day 34. Echocardiography performed 1 month post-discharge showed no sign of fluid accumulation. The patient has remained asymptomatic with no sign of fluid accumulation in the pericardium on CT for 6 years following treatment.

3. Discussion

The first reported case of surgical resection of a thoracic duct cyst was published in 1950 by Emerson. According to the 39 cases of mediastinal thoracic duct cysts reported in the English literature, many cases were diagnosed during surgery (28 cases, operative diagnosis available in 37) due to the connection of the cyst with the thoracic duct. In these intraoperatively-diagnosed cases, the pedicles were ligated to prevent postoperative chylothorax [4]. Cysts were located at the left diaphragmatic level in three cases, and in the right upper mediastinum in six cases (as in our case) [5–9]; although the cysts may occur along the entire course of the thoracic duct, these locations may be described as atypical when considering the normal route of the thoracic duct. Diagnosis was not reached during surgery in four of these atypical cases (44.4%). Postoperative chylothorax occurred in seven cases [1,5,10–14], and was frequently found in cases in which the cause of the cyst was not diagnosed intraoperatively (Table 1).

Although the thoracic duct usually crosses to the left side at the level of Th 4–6 [15], anatomic variations such as complete or incomplete duplication are reported in over 25% of cases [16]. In some cases, the terminal portion ends in the right internal jugular vein [16]. Cadaveric studies on thoracic duct tributaries have found major lymphatic vessels draining the lung and left ventricle, traveling from the right paratracheal nodes along the arch of the azygos vein into the thoracic duct [17]. In our case, it is possible that anatomic variations of the thoracic duct or its major tributaries were associated with the occurrence of the cyst in the right upper mediastinum.

Chylopericardium is a rare entity in which chylous fluid accumulates in the pericardial cavity [18]. A previous literature review reported that idiopathic chylopericardium was the most common type of chylopericardium in adult patients (56%), followed by postoperative causes in 9% [18]. We reviewed the English and Japanese literature on postoperative cases of chylopericardium in adult patients (references available in Table 2). This review identified 46 patients with postoperative chylopericardium (including our patient); mean age was 49.3 years (range 23–77 years), and 25 patients (54.3%) were male. Thirty-one (67.4%) patients had undergone cardiac surgery, such as cardiac artery bypass grafting and arterial or mitral valve replacement, five patients developed chylopericardium after lobectomy and lymph node dissection for lung cancer (10.9%) [19], and two patients (including the present patient) had undergone mediastinal tumor resection (Table 2) [20].

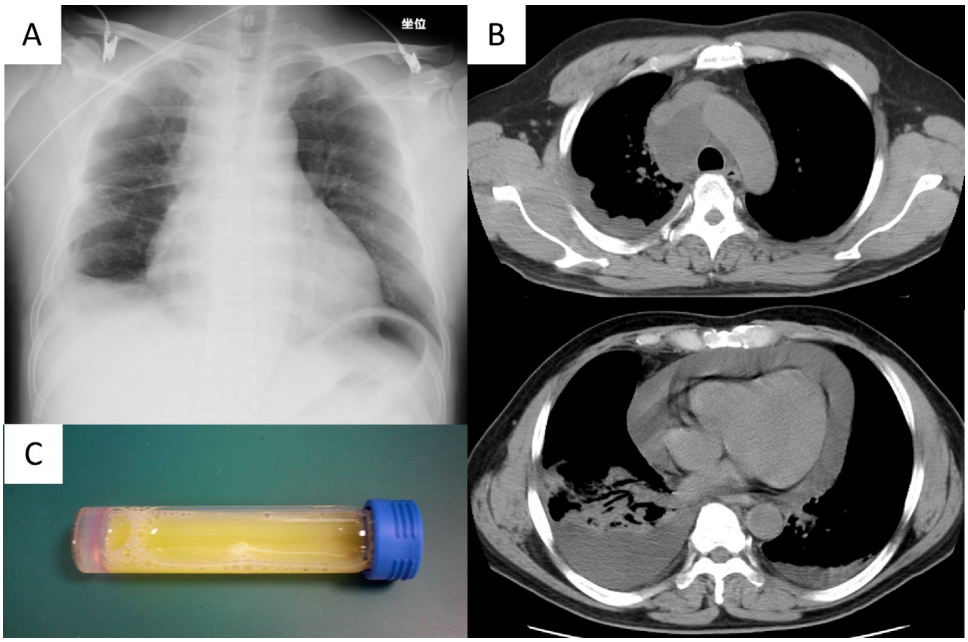

Fig. 2. Diagnostic findings at admission. A) Chest radiography revealed an enlarged cardiac silhouette with signs of fluid in the right hemithorax. B) Chest computed tomography revealed fluid collection in the upper mediastinum with pleural and pericardial effusion. (C) Yellowish-white fluid was obtained at pericardiocentesis.

Table 1
Cases of postoperative chylothorax in thoracic duct cysts.

year	author	age/sex	location	surgical diagnosis
2015	Park et al. [10]	42 F	left diaphragmatic level	no
2011	Taniguchi et al. [11]	29 M	left paratracheal	yes
2009	Mortman [1]	68 F	right retro-cardiac	yes
2009	De Santis et al. [5]	30 F	right paratracheal	no
1993	Okabe et al. [12]	45 M	left adjacent to aortic arch	no
1992	Mori et al. [13]	86 M	right posterior to carina	no
1976	Cervantes-Perez and Fuentes-Maldonado [14]	42 M	retro-cardiac, right of esophagus	no

F: female, M: male.

Table 2
Treatment for post-surgical chylopericardium.

Primary Surgery	Patients	no surgery		surgical treatment	
		low fat diet	TPN	pericardial window, shunt	ligation, repair
Cardiac surgery	31	7 *Pollard* 1981 (2) *Schiessler* 1984, *Bar-El* 1989 *Nwaejike* 2009	9 *Fudge* 1982, *Schactman* 1994 *Sharpe* 1999, *Sasaki* 2002 *Pitol* 2004 *Sachithanandan* 2008 *Koutsogiannidis* 2013 *Savas* 2013, *Karaca* 2014	3 *Kansu* 1977 *Bakay* 1984 *Soon* 2007	12 *Thomas* 1971, *Pollard* 1981 *Rose* 1981, *Lee* 1987 *Kawahira* 2003, *Mood* 2006 *Selimoglu* 2008, *Chaloob* 2008 *Mundra* 2011, *Navaratnarajah* 2011 *Gyoten* 2014, *Likaj* 2014
Cardiac transplantation	4	–	1 *Jackson* 2010	1 *Mailander* 1992	2 *Gelsomino* 2002 *Wierzbicki* 2015
Lobectomy	5	2 *Kimura* 1978, *Mizushima* 1996	1 *Hasumi* 1995	1 *Murakawa* 2012	1 *Jeon* 2013
Mediastinal tumor resection	2	2 *Morota* 1974, **present case**	–	–	–
Esophagectomy	2	–	1 *Nakamura* 1996	–	1 *Stewart* 2008
Lung transplantation	1	1 *Wait* 2013	–	–	–
Pulmonary Endarterectomy	1	–	–	–	1 *Niznansky* 2015
total	46	12 (26.1%)	12 (26.1%)	5 (10.9%)	17 (37.0%)

TPN; total parental nutrition, shunt; pericardial-peritoneal shunting.

Over half of the reported patients with postoperative chylopericardium were successfully treated conservatively, and surgical repair was required in only one patient who underwent lung or mediastinal tumor surgery (Table 2). Five patients also received

administration of somatostatin. Of the 24 patients treated by dietary restriction, six underwent lymphangiography or magnetic resonance thoracic ductography. These procedures were useful in excluding the possibility of injury to major vessels, subsequently indicating that immediate surgery was unnecessary; they were also useful in ascertaining that the leakage was unidentifiable following nonsurgical treatment in some cases. Duration of drain insertion was reported in 18 of the conservatively-treated patients, with a mean of 15.3 days (range 7–46 days). In the 17 patients treated with surgical ligation, mean time to operation was 22.8 ± 16.5 days (range 5–63 days). Nine of these patients (52.9%) received either preoperative fat administration or lymphangiography to determine the point of leakage. Lymph vessels in the mediastinum, mainly found in the thymus, were the leak point in seven (41.2%) cases, while the cause was damage to the thoracic duct in two (11.8%) and undetermined in eight (47.1%). Many of the patients underwent mass ligation of the thymus and mediastinal fat, in addition to suturing of the damaged vessel. A total of eight patients (47.1%) received thoracic duct ligation.

The cause of chylopericardium has been associated with the opening of the pericardium at surgery, thrombosis of the subclavian vein leading to obstruction of thoracic duct flow [21], and valve dysfunction in lymphatic vessels with reflux of chyle [19]. Other possible causes include extensive dissection of posterior mediastinal structures [21], and injury of the lymphatic plexus in the thymus [2]. In our case, the original chylothorax occurred due to resection of a thoracic duct cyst. The leakage and reaccumulation of chyle at the point of injury after 2 years may have happened due to a change in diet. Previous chemical pleurodesis and healing of the mediastinal pleura may have prevented the chyle from initially leaking into the thoracic cavity, and caused reflux into the pericardium.

4. Conclusion

We report a rare case of chylopericardium secondary to thoracic duct cyst resection. Chylous effusions are relatively common following surgical resection of thoracic duct cysts, especially when an operative diagnosis is not reached. However, chylopericardium is rare, and the initial management (pleurodesis in this case) may have been related to its onset. The main points highlighted by the present case are: 1) pedicles should be ligated in cyst resections, regardless of cyst location; 2) careful assessment in the initial surgery and radiographic studies such as thoracic ductography may help identify the point of leakage; 3) a low-fat diet is the first choice in the initial management of postoperative chylopericardium, especially in cases of lung or mediastinal tumor resection. Surgical repair may be considered when patients do not respond to more than 2 weeks of conservative treatment.

This is the first thorough review of cases of postoperative chylopericardium. Although further analysis of more cases is needed, this report may provide information on the optimal management of this rare complication.

Conflicts of interest

None.

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Ethical approval

Publication was approved by the ethics committee of Kimitsu Chuo hospital (#328).

Consent

Written informed consent for the publication of this manuscript was obtained from the patient. Publication was approved by the ethics committee of our institution (#328).

Authors contribution

Toshiko Kamata; data collection, data analysis, writing of paper.
Mitsutoshi Shiba; data interpretation, manuscript review.
Taiki Fujiwara; data collection.
Kaoru Nagato; data collection.
Shigetoshi Yoshida; data analysis and interpretation.
Tohru Inoue; pathological analysis.
Tomohiko Iida; manuscript review, study design.

Registration of research studies

UIN researchregistry2410.

Guarantor

Toshiko Kamata and Mitsutoshi Shiba.

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