

Chylothorax after cardiac resynchronization therapy-pacemaker implantation: A case report



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Introduction

The benefit of cardiac implantable devices such as cardiac resynchronization therapy pacemaker (CRT-P) has been widely demonstrated in a number of clinical trials, and it remains an important guideline-recommended therapy for the management of patients with symptomatic heart failure with reduced ejection fraction and electromechanical heart muscle dyssynchrony.¹ Despite improved training, advanced techniques, and better experience, device implantation is not without complications. Cannulation of the left subclavian vein, particularly in the context of a stenotic or occluded subclavian vein, can potentially cause injury to the thoracic duct. Disruption of the thoracic duct, at any location along its course, can lead to chylothorax. Thoracic surgery is a major cause of iatrogenic chylothorax, but its incidence rate is relatively low. Other reported etiologies include chest radiation, lung biopsies, subclavian vein occlusion, and lymphangiomyomatosis.² Chylothorax after cardiac implantable electronic device implantation is extremely rare, with a paucity of reported cases. Here, we present a case of recurrent chylothorax secondary to thoracic duct injury during an upgrade from a dual-chamber pacemaker to CRT-P.s

Case report

A 77-year-old man with an extensive past medical history including Hodgkin lymphoma treated with mantle field radiation and splenectomy 40 years prior, atrioventricular block treated with dual-chamber pacemaker implantation 15 years prior, hypertension, hyperlipidemia, and paroxysmal atrial fibrillation developed pacing-induced cardiomyopathy with an ejection fraction of 25% and severe dyspnea at mild exertion (NYHA class III symptoms). Therefore, the device was upgraded to CRT-P.

KEYWORDS Chylothorax; CRT; Complication; Thoracic duct embolization; Thoracic duct injury
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KEY TEACHING POINTS

- Chylothorax is the accumulation of chyle within the pleural space. Chylothorax can occur as a complication after surgery, most frequently after thoracic surgery. Chylothorax after pacemaker implantation is extremely rare.
- Inadvertent thoracic duct cannulation and injury can occur when left upper extremity venous access is utilized for cardiac resynchronization therapy (CRT) procedure, particularly if the left central veins, subclavian vein(s), and/or brachiocephalic vein(s) are stenotic or occluded.
- The usage of the stiff wire and sheath advancement prior to confirming wire position likely led to the thoracic duct injury.
- To minimize risk for this complication during CRT procedure, a detailed central venogram and wire position confirmation should be obtained prior to inserting a larger-bore sheath, particularly in patients with an increased risk for central venous stenosis.
- The lymphangiography and thoracic duct embolization may be effective for diagnosing and treating chylothorax.

Access was gained via left subclavian vein with a 21 gauge Micropuncture needle (Cook Medical, Bloomington, IN). Using modified Seldinger technique, a 5F introducer was advanced over the 0.018-inch wire. A left subclavian venogram was performed during the CRT-P upgrade procedure, showing a patent left subclavian vein (Figure 1A). Then a 0.038-inch J wire was advanced through the 5F introducer into the subclavian vein with some difficulty. Next, a 10F sheath was attempted to be advanced over the J wire, but it was very difficult to advance the 10F sheath over the

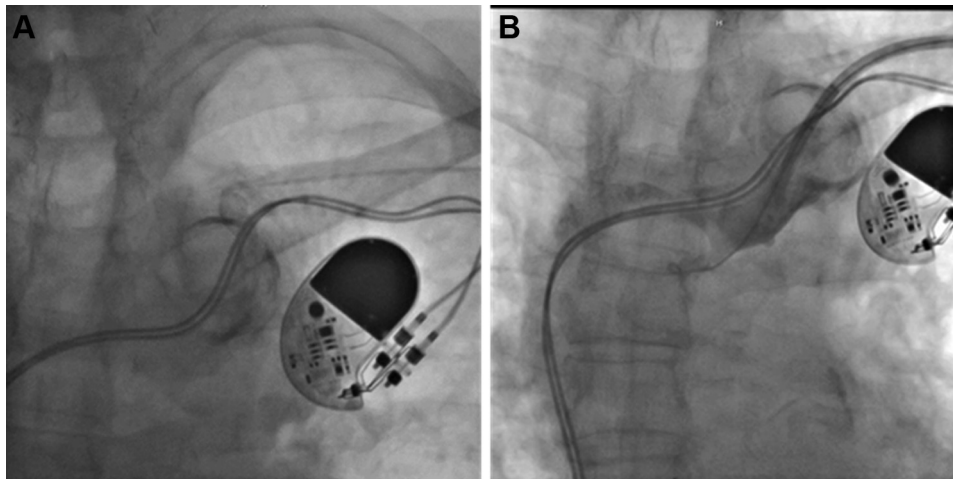


Figure 1 A: Venogram showing the patency of subclavian vein. B: Delayed images of venogram during cardiac resynchronization therapy pacemaker implantation procedure showing left brachiocephalic vein stenosis (arrow) and guidewire coursing extraluminally to left brachiocephalic vein (arrowhead).

0.018-inch wire, so it was exchanged to a long, stiff 0.032-inch Amplatz wire. Contrast injection through the sheath resulted in contrast staining consistent with extravascular positioning of the wire and sheath (Figure 1B). The sheath and the wire were retracted and repositioned in the right atrium under fluoroscopic guidance to complete the procedure.

During the periprocedural period, the patient remained asymptomatic and was discharged 2 days after the procedure. Two months after the procedure, the patient presented to the emergency department with shortness of breath and left-sided chest pain on deep inhalation. Chest radiography revealed a large left-sided pleural effusion (Figure 2). In the ensuing 3 months post procedure, 4 thoracenteses were performed. The first 2 thoracenteses yielded 800 mL and 600 mL of serosanguinous, cloudy fluid. The subsequent 2 thoracenteses removed 850 mL and 950 mL of “milky, cloudy” fluid. The pleural fluid analysis showed a lymphocytic predomi-

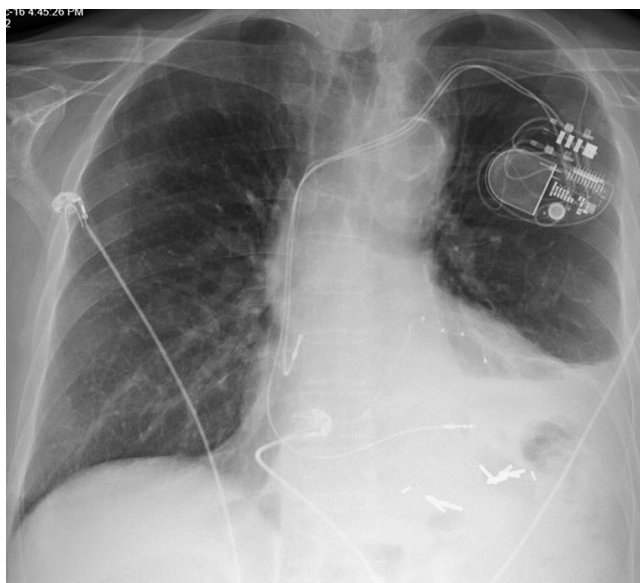


Figure 2 Chest radiograph showing new left pleural effusion 2 months post cardiac resynchronization therapy pacemaker implantation.

nant, exudative effusion with a total triglyceride level of 354 mg/dL, consistent with a chylothous effusion. Microbiology was negative and cytology showed no malignant cells. Positron emission tomography/computed tomography scanning was negative for hypermetabolic lymphadenopathy or distant metastasis. Owing to the rapidly reaccumulating left pleural effusion, a tunneled left chest tube was placed 4 months post procedure. Unfortunately, the patient reported severe left-sided pain and the tube was removed. Conservative treatment measures including a low-fat diet and octreotide were initiated; however, the patient continued to have significant pleural fluid accumulation, causing respiratory symptoms and pain. The patient remained symptomatic, requiring more frequent thoracenteses; therefore, lymphangiography and thoracic duct embolization was pursued. A bilateral intranodal lymphangiogram was performed (Figure 3A). The cisterna chyli was accessed with a 22 gauge needle under fluoroscopic guidance and a thoracic lymphangiogram was performed through a microcatheter positioned in the lower thoracic duct (Figure 3B). Lymphangiography showed a markedly tortuous thoracic duct that bifurcated into 2 main ducts: 1 terminating at the right internal jugular-subclavian vein confluence and the other terminating into the left internal jugular-subclavian vein confluence. Definitive contrast extravasation could not be identified, probably owing to the fact that the microcatheter could not be advanced beyond the inferior aspect of the thoracic duct secondary to its tortuosity. The thoracic duct was successfully embolized with microcoils and gelatin sponge (Figure 3C). The patient’s shortness of breath as well as the recurring left pleural effusion resolved within 1 week post procedure. The patient was discharged to an extended care facility and he remained without left pleural effusion until follow-up, 3 months post procedure (Figure 3D).

Discussion

Although CRT has become a well-established treatment for heart failure with reduced ejection fraction and conduction

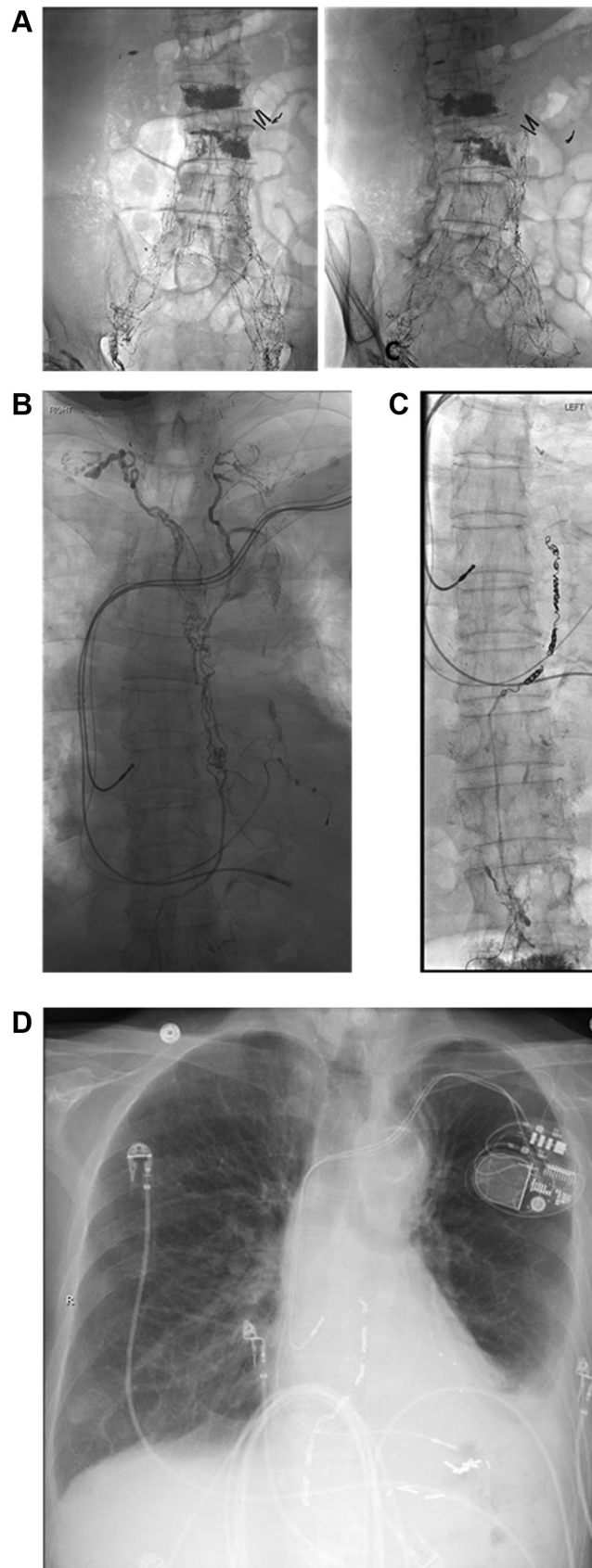


Figure 3 **A:** Frontal and right anterior oblique images of intranodal lymphangiogram showing cranial progression of ethiodized oil towards the cisterna chyli. **B:** Frontal lymphangiogram of the thorax showing a markedly tortuous thoracic duct with bifurcation of the thoracic duct into the left and right medial subclavian veins. **C:** Frontal spot image of the caudal aspect of the thoracic duct and cisterna chyli post microcoil and gel-foam embolization. **D:** Chest radiograph shows no recurrence of pleural effusion 3 months after thoracic duct embolization.

abnormalities, device implantation is not without complications such as bleeding, infection, pneumothorax, and cardiac tamponade.^{3,4} Chylothorax after pacemaker implantation is extremely rare, and there have been few reports in the literature caused by vein thrombosis.⁵⁻⁷ Thoracic duct injury that resulted in a chylous effusion of the pacemaker pocket has been reported only in a limited number of case reports.^{8,9} To the best of our knowledge, this is the first case of recurrent chylothorax secondary to thoracic duct injury during an upgrade from a dual-chamber pacemaker to CRT-P.

Chylothorax refers to the accumulation of chyle in the pleural space and is an infrequent but potentially life-threatening complication with profound respiratory, nutritional, and immunological consequences. Disruption or damage to the thoracic duct or its lymphatic tributaries within the thorax is the most common cause of chylothorax. The resultant pleural effusion typically has a high triglyceride concentration, and often a turbid or milky-white appearance. Most commonly, chylous effusions are iatrogenic in etiology, particularly after thoracic surgery involving the posterior mediastinum and/or nearby structures. Chylothorax is reported to occur at a rate of 0.42% for all general thoracic surgery and up to 3.9% after esophagectomy.⁴ In the present case, the venogram showed the guidewire and sheath migrated outside of the subclavian vein, probably into the thoracic duct. Based on the imaging and chronicity of the chylous effusion, we suspect that the thoracic duct injury was caused by wire manipulation into the thoracic duct-venous junction, resulting in a left chylous effusion. Cannulation of the thoracic duct during subclavian vein access is exceedingly rare; however, we believe this occurred because of several factors, including the presence of existing pacemaker leads causing brachiocephalic vein stenosis and synechiae, as well as engorgement of the thoracic duct-venous confluence owing to history of lymphoma and mantle cell radiation. The usage of the stiff wire and sheath advancement prior to confirming wire position likely led to the thoracic duct injury. Placing the guidewire in the inferior vena cava under fluoroscopic guidance prior to sheath advancement would have ensured intravascular positioning. Moreover, performing a detailed venogram with the sheath placed in the peripheral aspect of the subclavian vein may have prevented this complication, particularly in patients with an increased risk of upper-extremity venous stenosis or obstruction.

Chylous effusion is a rare complication of thoracic surgery and subclavian vein procedures. The treatment strategy of chylothorax remains controversial. In general, conservative treatment such as chest tube drainage, and either bowel rest with total parenteral nutrition or a high-protein, reduced-fat diet with medium-chain triglyceride supplementation is customarily tried prior to more invasive procedures for low-output chylothorax (<1000 mL/day).² Somatostatin and octreotide reduce intestinal chyle production and have proved beneficial in the treatment of chylothorax.⁴ If conservative treatment measures are unsuccessful, surgical ligation of the thoracic duct or percutaneous thoracic duct

embolization can be considered based upon the severity of underlying disease.² Prolonged leakage of chyle, rich in nutrients, T cells, and electrolytes, can significantly worsen a patient's clinical status.¹⁰ In this case, the patient had severe left ventricular dysfunction and poor performance status; therefore, after conservative management failed, it was decided to pursue definitive treatment with the lesser invasive percutaneous thoracic duct embolization. Percutaneous thoracic duct embolization is considered a relatively safe procedure with low complication profile and no reported deaths related to the procedure.¹¹ Percutaneous thoracic duct embolization has a reported success rate of 70%.^{12,13} Moreover, lymphangiography is useful for identifying the site of the thoracic duct injury as well as for treatment. Early lymphangiography is therefore recommended for patients who have failed conservative measures.¹⁴

In this case report, lymphangiography was performed and there was technical success in percutaneous cannulation of the cisterna chyli; however, a definitive leak from the thoracic duct could not be identified. We believe that the site of the thoracic duct injury could not be identified because the patient's variant thoracic lymphatic anatomy and thoracic duct tortuosity prevented extension of the microcatheter cranially towards the thoracic duct junction with the left subclavian vein. Furthermore, the patient's thoracic duct bifurcated at the level of the manubrium, resulting in further dilution of hand-injected contrast. The suspected site of injury to the thoracic duct is near the left subclavian vein and left internal jugular vein confluence, based on the contrast extravasation seen on venogram from CRT-P implantation procedure. The wire perforation near the thoracic duct entry site at the level of the central subclavian vein is much more difficult to identify, particularly if the lymphangiogram is performed from a caudal position. Despite not being able to identify the site of chyle leak on the lymphangiogram, the thoracic duct was successfully coil embolized. The patient's chylothorax resolved post procedure and he was discharged to an extended care facility.

Conclusion

Inadvertent thoracic duct cannulation and injury can occur when left upper extremity venous access is utilized for CRT procedure, particularly if the left central veins, subclavian veins, and/or brachiocephalic vein(s) are stenotic or occluded. Thoracic duct injury causing recurrent chylous effusion is a rare but potentially serious complication after cardiac implantable electronic device implantation. To minimize risk for this complication during CRT procedure, a detailed central venogram and wire position confirmation should be obtained prior to inserting a larger-bore sheath, particularly in patients with an increased risk for central venous stenosis.

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