Indian Pacing and Electrophysiology Journal 22 (2022) 161–164

Contents lists available at ScienceDirect

Indian Pacing and Electrophysiology Journal

journal homepage: www.elsevier.com/locate/IPEJ

An atypical pacemaker pocket hematoma containing chyliform fluid

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ARTICLE INFO

Article history:
Received 4 January 2022
Received in revised form
13 February 2022
Accepted 23 March 2022
Available online 1 April 2022

Keywords:
Pocket hematoma
Pseudochylous
Implantable cardioverter defibrillator

ABSTRACT

Subcutaneous hematoma is a complication of cardiac device implantation. In most cases, it is drained or spontaneously reabsorbed. While cases of chylothorax are rare, and cases of pseudochylothorax even rarer, previous cases of accumulation of chyliform material in the subcutaneous pockets of cardiac devices are anecdotal. We present a case of a 60-year-old man with antiphospholipids antibody syndrome and rheumatoid arthritis, who underwent dual-chamber ICD implantation in December 2020; the procedure was complicated by a pocket hematoma, which required surgical drainage. After 7 months, the man returned owing to heart failure, with evidence of the reappearance of a large swelling in the ICD pocket; this was tolerated for months by the patient and was no longer controlled. We drained 100ml of gold-colored, odorless liquid, and found no evidence of blood material in the pocket. The liquid was not pus, as culture testing proved negative for bacterial growth. Chemical-physical examination revealed elevated cholesterol concentration (704 mg/dl) and low levels of triglycerides (80 mg/dl; plasma cholesterol values were 91mg/dl, and triglycerides 48 mg/dl). Microscopic examination revealed isolated leukocytes and rare erythrocytes immersed in mucoid material; cytological analysis showed a carpet of macrophages filled with cholesterol. This evidence supports the diagnosis of pseudochyle fluid, formed by the degradation of a hematoma left intact in a closed cavity for more than 6 months. This is an extremely rare case of chyliform fluid documented in an ICD pocket.

1. Introduction

It has been estimated that about 1.2 million pacemakers and 400,000 implantable cardioverter defibrillators (ICD) are implanted annually worldwide [1]. Pocket hematoma is a common complication, being reported in 2–9% of patients, and has become more frequent with the increasing use of antiplatelet and anticoagulant therapy [2]. In most cases, a pocket hematoma can be drained or spontaneously resorbs; if untreated, there is a risk of infection.

The literature indicates that damage to thoracic lymphatic vessels can cause chylothorax, i.e. the accumulation of chyle in the pleural space [3]; this is most commonly seen after traumatic disruption of the thoracic duct, and is typically diagnosed on the basis of the milky appearance of fluid due to high fat content. Chylothorax must be distinguished from pseudochylothorax; this latter is a rare form of pleural effusion, also called chyliform or cholesterol pleural effusion, which is characterized by its high cholesterol content and milky pleural fluid, as in long-standing exudative pleural effusion of various causes.

There are rare reports of cases of chyle effusion inside the subcutaneous pocket after the implantation of cardiac devices [4]. We present a particular case of drainage of what was supposed to be a "simple" ICD pocket hematoma, but which instead was a collection of gold-colored liquid similar to chyliform fluid, and which posed a very challenging diagnosis.

2. Case report

A 60-year-old man with a history of antiphospholipid antibody syndrome and rheumatoid arthritis, dilated ischemic cardiomyopathy, and a previous acute anterior myocardial infarction in 2010, underwent aorto-coronary bypass surgery (left internal mammary artery - anterior descending artery). He had no clinical signs or family history of dyslipidemia. In 2015, he suffered an episode of atrial fibrillation. Subsequently, owing to the presence of

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Peer review under responsibility of Indian Heart Rhythm Society.

https://doi.org/10.1016/j.ipej.2022.03.002

thrombophilia and antiphospholipid antibody syndrome, oral anticoagulant therapy with warfarin was initiated. In December 2020, the patient underwent primary prophylactic dual-chamber ICD implantation (left ventricular ejection fraction 33%) at another center. The procedure was complicated by the formation of a large pocket hematoma two days later, which required drainage and evacuation. In the following weeks, the patient noticed the reappearance of a large swelling in the ICD pocket; however, as it was painless and not troublesome, he did not seek medical attention.

After 7 months, in June 2021, he came to our observation following the appearance of initial signs of heart failure. Clinical examination revealed a large swelling in the left pectoral area (Fig. 1), extending to the nipple, soft to the touch, not painful, not hot or red, with overlying skin of a normal color. The patient had no fever or signs of infection or inflammation (WBC 5000/μL, Hb 13.8 g/dl, ESR 4mm, creatinine 1.5 mg/dl, CRP 0.5 mg/dl, Procalcitonin 0.43 ng/ml, Quantiferon TB gold negative).

An ultrasound examination revealed a large fluid component in the pocket. We decided to drain the hematoma, given its size. After incising the skin and subcutis, and reaching the defibrillator pocket, we made a small incision, from which 100ml of a gold-colored, odorless liquid leaked out (Fig. 2). We decided to open the pocket and completely revise it in the doubt that the drained fluid was purulent, even if it had atypical characteristics. There was no blood material in the pocket. The capsule surrounding the effusion appeared fibrotic, non-erythematous, intact and smooth. After drainage of all the liquid, the pocket normalized. The case of the ICD

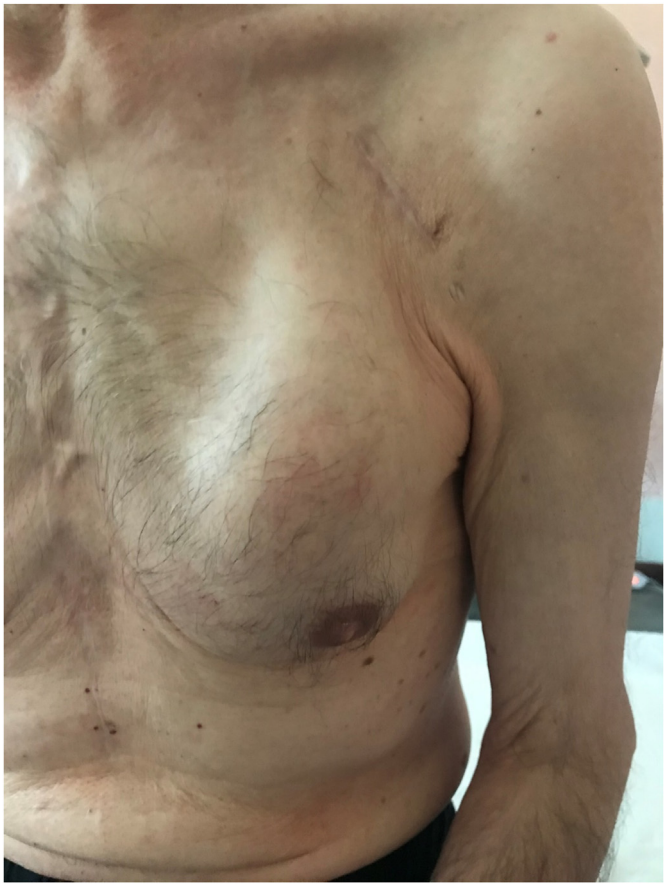

Fig. 1. Large hematoma in the left pectoral area, extending to the nipple, soft to the touch, not hot or red, with overlying skin of a normal color.

Fig. 2. Syringes containing the gold-colored, chyliform fluid, without a blood component.

was prudently then wrapped in a rifampicin-releasing antibacterial bag and reinserted into the subcutaneous pocket, and a triple-layer suture was carried out to close the surgical wound; no complications occurred.

Biochemical, cultural and cytological analyses were carried out on the drained liquid. The chemical-physical examination revealed a cloudy, gold-colored liquid, with a density of 1020, pH 8.5, high protein content (>400 g/l), and absence of glucose. Microscopic examination revealed isolated leukocytes and rare erythrocytes immersed in material of a mucous nature; this was not pus, as culture testing was negative for GRAM +, GRAM-, mycobacteria and anaerobic bacterial growth. The concentrations of liquid cholesterol and triglycerides were 704 mg/dl and 80 mg/dl, respectively (the patient's total plasma cholesterol value was 91 mg/dl, and triglycerides 48 mg/dl).

Cytological analysis showed amorphous paucicellular material, composed of similproteinaceous debris and a carpet of numerous foamy macrophages filled with cholesterol crystals (Fig. 3).

Our conclusion was that the fluid accumulated in the pocket was pseudochyle, and had resulted from the evolution of an untouched hematoma in a fibrous, thickened subcutaneous pocket for more than 6 months.

3. Discussion

This is a rare case in which the presence of chyliform fluid has been documented in an ICD pocket. The most difficult point in the management of this clinical case was understanding the origin of

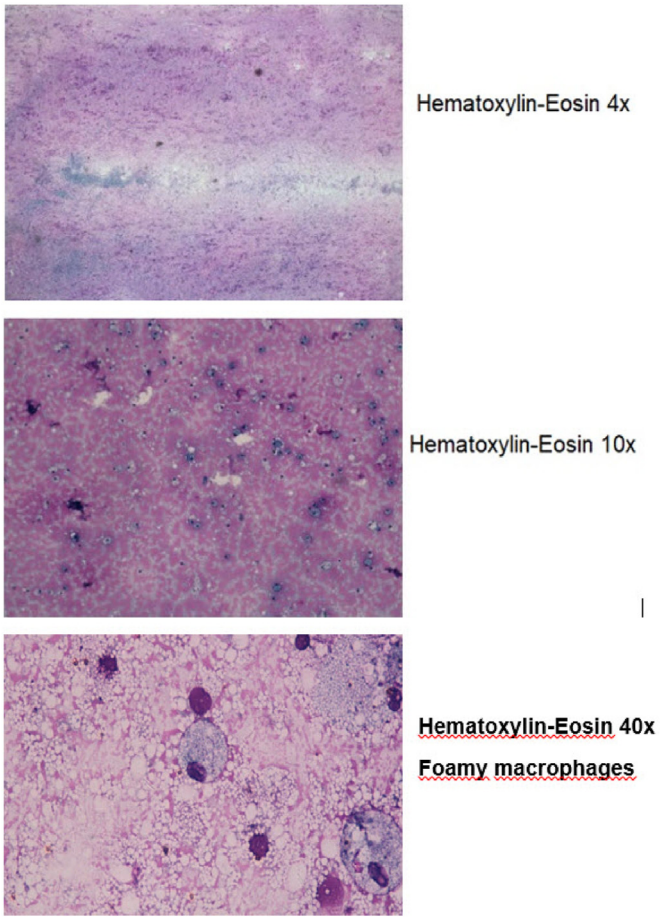

Fig. 3. Cytological analysis showed amorphous paucicellular material, composed of similproteinaceous debris and a carpet of numerous foamy macrophages filled with cholesterol crystals.

the fluid. We believe that the material was not exactly "chyle", since the patient had already undergone drainage of a pocket hematoma a few days after ICD implantation. On that occasion, the evacuated fluid was exclusively blood, and it was not necessary to reposition the ICD leads. If a large lymphatic vessel had been punctured in the attempt to cannulate the left subclavian vein, thereby creating a communication with the subcutaneous pocket, chyiform material would already have been found during the first drainage.

Furthermore, the presence of chyliform fluid in an ICD pocket is very unlikely, not least for anatomical reasons. Indeed, the thoracic duct, enters the thorax through the aortic hiatus of the diaphragm and ascends in the posterior mediastinum. Located between the descending thoracic aorta on the left and the azygos vein on the right, it ascends behind the aortic arch and the thoracic part of the left subclavian artery to the thoracic inlet. Finally, the duct terminates by opening into the junction of the left subclavian and jugular veins. It is approximately 3 mm–5 mm in diameter and its entire course is intrathoracic; thus, a breach in the integrity of the thoracic duct anywhere along its course, even near the left subclavian vein, will cause the effusion of chyle into the pleural cavity, leading to chylothorax [3]. Only if a fistula were created between the duct and the subcutaneous pocket could the chyle reach the ICD pocket. Bokhari et al. have reported chyle collection in pacemaker pocket in a patient with a background of radiotherapy, where the local anatomy may have been altered, with the possibility of forming a fistula [4]. Other cases of chyloma were reported in patients with anomalous thoracic duct drainage into the left subclavian vein with injury during device implantation [5]. On the other hand, cases of chylothorax and chylopericardium due to obstruction of the brachiocephalic vein by the pacemaker leads have been reported, with superimposed thrombosis, and subsequent cascade of events that resulted in chyle leakage [6].

We believe that, in our patient, the fluid was pseudochyle or chyliform. Pseudochyle is an effusion with a gross appearance similar to that of chyle: milky white; this is less common than the classical chyle and has been reported in long-standing exudative pleural effusions of various causes. Pseudochyle fluid contains a high concentration of cholesterol, giving it its characteristic milky or white appearance. In long-standing exudative pleural effusion, the cell is ruptured, releasing cholesterol from the cell membrane into the fluid, which gets trapped in the pleural cavity. However, unlike classical chyle, this fluid does not contain chylomicrons or long-chain fatty acids. The cholesterol concentration in pseudochyle is typically more than 200 mg; the triglyceride level is less than 110 mg/dL, and the cholesterol to triglycerides ratio is always more than 1. High cholesterol content leads to golden rather that whitish appearance. Cholesterol crystals may be visible in dried slides from pleural fluid when viewed under polarized light, and appear as rectangular plates with notched edges. The presence of cholesterol crystals is virtually diagnostic of pseudochyle. [3–7].

Both pseudochyle and chyle classically have a turbid or milky appearance, due to their high lipid content. Apart from that, their etiologies, pathogenesis and clinical implications differ, making it important to distinguish between them. Chyle is primarily made up of chylomicrons, an aggregate of long-chain triglycerides, cholesterol esters and phospholipids. It is also rich in lymphocytes, primarily T lymphocytes, as the main cellular component, with concentrations that range from 400 to 6800 cells. The absence of a milky appearance does not exclude chyle. If the fluid is kept undisturbed for some time on centrifugation, the supernatant of the fluid is clear. In empyema, the supernatant is not clear. Chyle is rich in lymphocytes, which account for 80% of all the cells. A pleural fluid triglyceride concentration greater than 110 mg/dL confirms the diagnosis of chylothorax. However, 15% of cases of chylothorax are known to have triglyceride concentrations less than 110 mg, depending on the time of the last meal and the fat content of the diet. Detection of chylomicrons in the pleural fluid by means of lipoprotein electrophoresis confirms chylothorax. Typically, the total cholesterol level in chyle is less than 200 mg/dl. The other fluid composition is similar to plasma. Chyle is usually alkaline, with pH ranging from 7.4 to 7.8 [3]. The biochemical and cytological differences between chile and pseudochile are summarized in Table 1.

The most frequent causes of pseudochylothorax include tuberculosis, rheumatoid arthritis, paragonimiasis, echinococcosis, neoplasia, or trauma [8]. The pathogenesis of pseudochylous has never been investigated in depth. However, the presence of an effusion for at least 5 years in the setting of a fibrous, thickened pleura due to chronic pleuritis is characteristic [8]. It is believed

Table 1
Biochemical and cytological differences between Chylous and Pseudochylous.

	Chylous	Pseudochylous
Biochemical analysis		
Triglycerides	>110 mg/dl	<110 mg/dl
Cholesterol	<200 mg/dl	>200 mg/dl
Cholesterol to triglycerides ratio	<1	>1
Chylomicrons	present	absent
pH	<7,8	>7,8
Cytological analysis		
Cholesterol crystals	absent	present
Cellular predominance	Lymphocytes (>80%)	variable

that, as a result of the pleural thickening that blocks the drainage of fluids to the pleural wall lymphatic system, the cholesterol and lecithin-globulin complexes released after red cell and neutrophil lysis into the pleural fluid become trapped in the pleural cavity. Indeed, some authors consider pseudochylothorax to be a form of "lung entrapment" in the context of chronic inflammation [9]. In our patient, the same pathophysiological mechanism may have occurred in a closed subcutaneous pocket, without fluid drainage, untouched for several months. In addition, the patient was affected by rheumatoid arthritis, a pathology present in 50% of the published cases of pseudochylothorax [8].

Publications on pseudochylothorax are rare and involve isolated case-reports and small case-series [10]. There are no publications on similar phenomena in the extrapleural area, nor in subcutaneous cardiac device pockets.

Declaration of competing interest

None.

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