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Case report

Thoracic duct leakage in a patient with type B-Non-Hodgkin lymphoma treated with transvenous retrograde access embolization: a case report

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Abstract. Thoracic duct (TD) is the largest lymphatic vessel in the body and drains the lymph at the junction between the left subclavian and jugular veins. Chylothorax (CTX) represents an accumulation of lymphatic fluid in the pleural space. We present a case of a 65 years-old man with an histologically diagnosed mediastinal type B non-Hodgkin Lymphoma, treated with chemo-immunotherapy. CT scan during follow up showed significant left side pleural effusion, amounting to 2.8 litres after drainage. Conservative treatment with low fat parenteral nutrition was started without reduction of drainage output, then lymphangiography (LP) with Lipiodol was performed demonstrating a leak in the distal TD. CTX increased in the following days, and a further LP was performed. Using transvenous retrograde access we catheterized TD at the left subclavian jugular veins using a microcatheter. The leak was treated with multiple conventional and controlled delivery microcroils and cyanoacrylate, obtaining complete embolization without residual leak. (www.actabiomedica.it)

Key words: lymphangiography, radiology, interventional, embolization, chylothorax, thoracic duct

Introduction

Chylothorax (CTX) is an uncommon cause of pleural effusion related to abnormal accumulation of chyle in the pleuric space. CTX is caused by the extravasation of chyle after obstruction or injury to the thoracic duct or its branches (1). CTX may have different causes, both traumatic and non-traumatic. Traumatic CTX, including iatrogenic, accounts for approximately 25% of cases (2); non-traumatic causes include malignancy (lymphoma and bronchogenic carcinoma are the most common), miscellaneous and congenital disorders. The treatment of low output CTX (<500 mL/day) can be conservative, based on nutritional measures to reduce chyle flow and drainage of CTX with thoracostomy to obtain spontaneous lymphatic closure (3). If average daily chyle exceeds 1500 mL for a five-day period or chyle flow has not diminished in two weeks, conservative treatment is regarded as ineffective and surgical or percutaneous approaches should be considered (3). Percutaneous treatment is based on lymphangiography (LP) with catheterization of cisterna chyli or thoracic duct (TD) followed by percutaneous embolization and direct injection of sclerosing agents. The leakage sites are successfully identified in 64-86% of the patients (4), with percutaneous treatment having a successful rate of 60% (5). Surgical approach, either with classical thoracotomy or thoracoscopic surgery, is aimed at excising the duct at the point of the leak; otherwise, if no chyle leak is noted, surgical treatment consists in ligation above the right hemidiafragm. Overall, surgical therapy is successfull in 67-100% of the cases (6). We presented a case of a man with mediastinal periaortic type B

Non-Hodgkin lymphoma who developed CTX after video-assisted thoracoscopic (VATS) biopsy of periaortic lymphomatous mediastinal mass and first three cycles of immunochemotherapy.

Case report

A 65 years-old man underwent abdominal ultrasound (US) for recurrent epigastralgia in February 2019, with the detection of solid mass surrounding the diaphragmatic aorta. Contrast enhanced CT (CECT) showed an enhancing mass at this site, with maximal axial diameter of 3.7cm and longitudinal extent of 6.7cm. Transthoracic CT-guided biopsy of the mass performed three weeks later was unsuccessful. Thus multiple excisional biopsies of the lesion were obtained by VATS two weeks later, with the diagnosis of type B Non-Hodgkin lymphoma of the marginal zone. Chemo-immunotherapy with bendamustine plus rituximab was started in April 2019. After three cycles of immuno-chemotherapy, the follow up CECT performed in July 2019 identified significant reduction in size of the perioaortic mass, despite an abundant left side pleural effusion was noted, extending from the apex to the lung base with water density (5-15 Hounsfield Unit, HU). The patient manifested mild exertional dyspnea and was admitted to the Pulmonology Unit for effusion treatment. Two days after admission the patient underwent diagnostic thoracoscopy that identified thickened visceral and parietal pleura without nodulations and drained 2.8 litres of milky fluid. At the end of the procedure a 14 French pleural drainage was left onsite through the 7th intercostal space. Laboratory analysis of the pleural effusion showed elevated triglycerides concentration (854 mg/dL, normal values 50-170 mg/dL) consistent with the diagnosis of CTX. A first attempt with conservative treatment consisted in fasting with low fat parenteral nutrition. Despite low fat nutrition a stable daily 500mL milky fluid pleural output was observed during the following two weeks from the gravity drainage. On the 17th day after admission, the patient underwent LP performed via right inguinal lymph node access under ultrasound guidance to unveil the leak site and treat it with Lipiodol (Guerbet, Paris, France). During the procedure a right inguinal node was accessed using a 25-gauge spinal needle (Egemen International, G.Emir Izmir, Turkey), with the needle tip positioned in the transitional zone between the cortex and the hilum of the lymph node. Lipiodol (30 mL) was then injected using an anesthesia injection pump with a rate of 0.2–0.4 mL/minute, leading to opacification of deep iliac femoral lymphatic system and, partially, the cysterna chyli and TD. A leak at the distal TD was detected confirmed by a non enhanced CT (NECT) control after five hours (Figure 1).

The patient continued low fat parenteral nutrition without drainage output for the following ten days. For three days the patient received normal enteral nutrition but at the 14th day after procedure dyspnoea occurred with desaturation and appearance of 700ml milky fluid drainage output. At the same day, NECT displayed increasing left concamerated pleural effusion. Therefore, the day after, the patient underwent TD leak embolization. Through right common femoral vein access using 5 French (Fr) sheath, the tip of a 5 Fr Multipurpose A1 catheter (Cordis, Miami, Florida) over a 0.035" guidewire (Guide Wire M; Terumo) was advanced to the confluence of the left internal jugular vein with the subclavian vein. Then the TD was catheterized by a 2.7 Fr microcatheter (Progreat; Terumo). TD LP confirmed a leak in the distal thoracic tract. Additionally, we demonstrated a TD duplication in its middle-distal tract as anatomical variant. Multiple conventional microcroils, Complex Helical 18, 4x4.0mm (Boston Scientific, Marlborough, Massachusetts) and detachable microcoils, Azur 18, 3x50mm and 4x100mm (Terumo) were released with the embolization of the middle-proximal TD. Embolization of the proximal TD was completed with n-butyl-2-cyanoacrylate (Histoacryl, B. Braun, Melsungen, Germany; figure 2).

At the end of the procedure no residual flow in the TD was observed, without post-procedural complications. Nine days after the embolization, no drainage output was detectable, despite NECT confirmed the presence of multiloculated left pleural effusion that was successfully treated by surgical toilette two days later. One week after surgical intervention thoracic drainage was removed and the patient was discharged 53 days after admission without symptoms. Follow

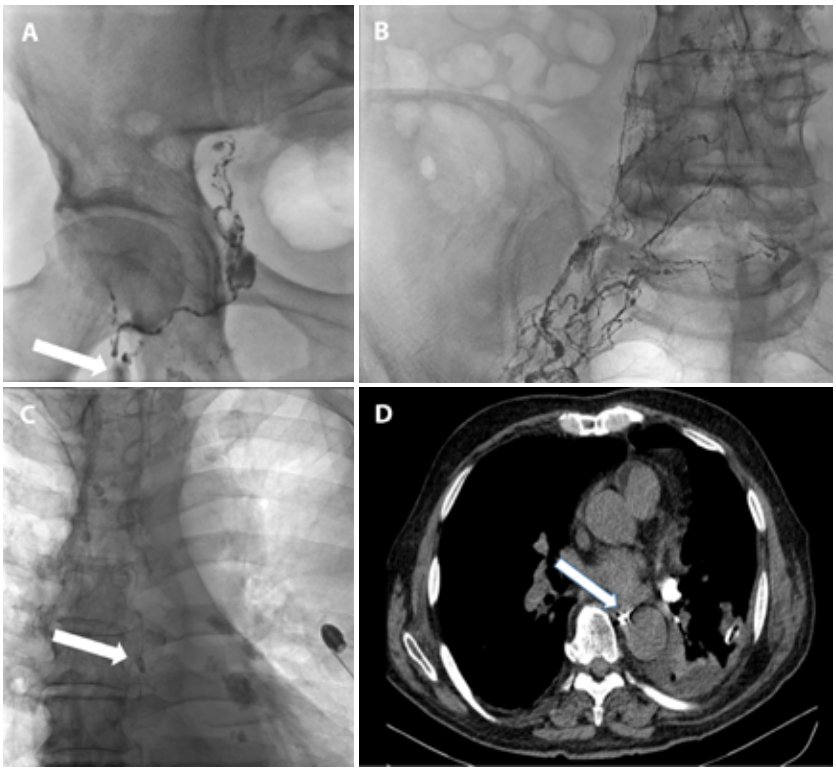

Figure 1. Lymphangiography with Lipiodol, performed via right inguinal lymph node access (arrow in A) with opacification of deep iliac femoral lymphatic system (B) and thoracic duct leak (arrow in C), confirmed by non-enhanced computed tomography). NECT performed the same day (arrow in D).

up with CECT, performed after one, four and twenty months showed no residual pleural effusion.

Discussion

In the present case of CTX, conservative therapy, based on low fat intake diet, was unsuccessful. Successful rate of conservative therapy is reported to be heterogeneous s in the literature (7)(8). As observed in our patient dietary restriction failed in around 70% of the patients with traumatic CTX, in particular in case of high output drainage CTX (7). Surgical approach is reported to be highly successful without significant complications, as demonstrated by Paul et al. (9), although represents an invasive procedure. In our patient, which already underwent VATS for biopsy, we decided for a percutaneous treatment. Our first attempt was Lipiodol LP through inguinal lymph node access. LP in the past was intended only for diagnostic purpose; nevertheless, it has been showed that single or multiple LP alone can be therapeutic, as Lipiodol-induced selective blockage of the pathological lymph duct induces sterile inflammatory reactions leading to duct scarring and leak closure (10). Although this procedure has high successful rate, reported to be around 70% (11), in our case was ineffective. Therefore we decided to perform a direct TD embolization, knowing from the literature that this procedure has a successful rate of 90% (12). The most challenging step of this procedure is TD catheterization, that could be achieved by lymphatic access (cisterna chyli, its major tributary, or an even lower segment of the TD) or by retrograde transvenous access (5)(13). The latter is less invasive because required venous access instead of direct TD puncture, but there are just few cases reported

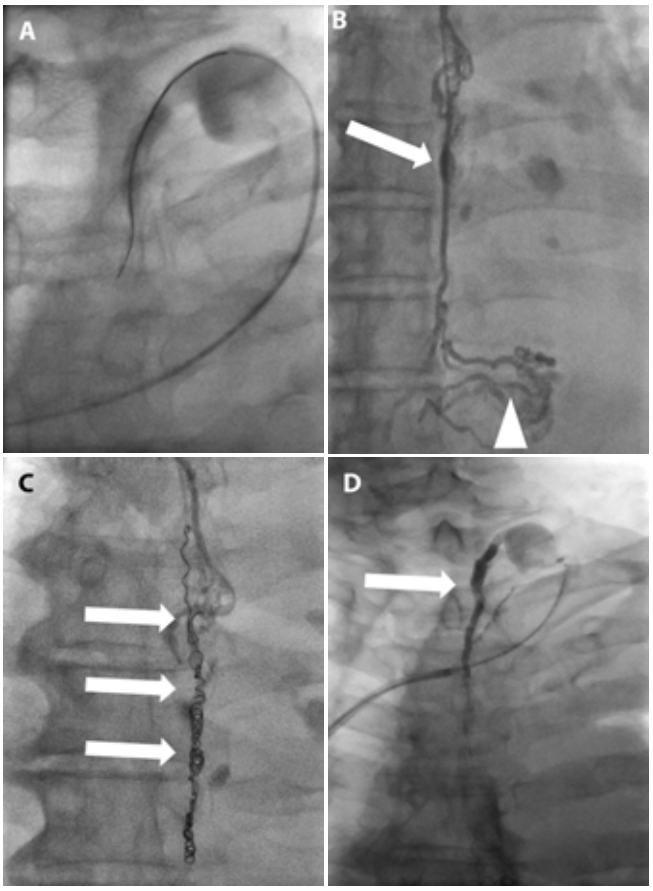

Figure 2. Catheterization of the thoracic duct using microcatheter (A). Thoracic duct lymphangiography confirmed a leak in the distal thoracic tract (arrow in B), duplicated in its middle-distal tract as anatomical variant (arrowhead in B). Embolization of the thoracic duct with multiple microcroils (arrows in C), completed with n-butyl-2-cyanoacrylate (arrow in D).

in the literature (13)(14)(15). The main technical issue is retrograde catheterization of TD, with 61% of successful rate reported by Kariya (16), that is mainly due to anatomical variant at the venous-TD junction, such as plexiform variation or completely left-sided thoracic duct. Moreover, TD is not usually visualized by subclavian venography due to chyle flow in to the venous system and cathetherization required a patient search in the region with microcatheter difficulty traversing the terminal valves. In our case successful catheterization of TD, through venous route, allow us to demonstrate the leak. Embolization was obtained with a combination of liquid embolic agent and coils. Follow up NECT demonstrated no residual pleural effusion.

Conclusion

Our step-by-step approach with conservative management, Lipiodol LP and finally thoracic duct embolization with transvenous retrograde access, lead to the resolution of iatrogenic CTX . Interventional radiology should be considered before the more invasive surgical approach for the treatment of CTX.

Conflict of Interest: Each author declares that he or she has no commercial associations (e.g. consultancies, stock ownership, equity interest, patent/licensing arrangement etc.) that might pose a conflict of interest in connection with the submitted article

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