CASE REPORT

A Unique Case of Recurrent Thoracic Duct Rupture: A Case Study

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Abstract

Introduction: Thoracic duct rupture is an uncommon presentation and usually occurs through trauma or surgery.

Case presentation: Here, we present an entirely unique case of recurrent spontaneous thoracic duct rupture in a 34-year-old woman who had three separate episodes of thoracic duct rupture. The first two episodes appear to have been caused by a spontaneous internal jugular vein (IJV) thrombosis whereas the final had no demonstrable cause. The initial presentation was with neck swelling, the second with chylothorax.

Management and outcome: This patient was treated with rivaroxaban for the IJV thrombosis while her chylothorax was treated conservatively.

Discussion: This case serves as a learning pearl to consider thoracic duct rupture in patients with neck swellings even without apparent traumatic or iatrogenic cause. It also brings to light the idea of previous insults weakening thoracic duct rupture and predisposing patients to recurrent issues.

Keywords: Case report, Chylothorax, Chyle leak, Thoracic duct.

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Introduction

The thoracic duct is the largest vessel of the lymphatic system. Its drainage at the angle of the left subclavian and internal jugular veins (IJVs) brings it frequently to the attention of head and neck surgeons who are all too aware of the morbidity which results from a leakage of chyle into the neck. This chyle can also accumulate in the pleural space resulting in a chylothorax if produced in sufficient quantity. Rupture of the thoracic duct most often results as a result of trauma from surgery but hyperextension of the spine, blows to the chest, penetrating wounds, and crushing injuries can also result in damage.[1–3] Non-traumatic causes are much less common and include malignancy, and diseases such as tuberculosis and sarcoidosis.[2]

As with any pleural effusion, chylothorax presents with patients complaining of shortness of breath and cough. Treatment of the underlying cause is ideal; however, not always possible. Symptomatic treatment of a chylothorax involves conservative and surgical approaches. The former includes chest drainage and either a strict medium-chain triglyceride diet (these are absorbed through the portal venous system without needing to go through the thoracic diet) or complete parenteral nutrition. Surgical intervention most commonly involves thoracic duct ligation and has a success rate of ~95%.[4]

This is the first reported case of chylothorax resulting from an IJV thrombosis and presenting with recurrent neck swelling.

Case Presentation

Clinical and Radiological Findings

A 34-year-old woman presented to the emergency department on three separate occasions between May 2014 and January 2016. Each time she presented with sudden onset of left supraclavicular neck swelling extending into the posterior triangle. There was no prior trauma, straining, or notable event leading to each of the presentations.

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The first episode was diagnosed and treated as a spontaneous left IJV thrombosis by the hematology unit. An ultrasound scan (USS) demonstrated a small clot within the IJV; however, a CT 24 hours later demonstrated that the clot had recanalized. Numerous investigative tests returned negative including lupus anticoagulant, thrombophilia screen, PNH screen, JACK2 mutation, ANA testing, beta-2 microglobulin, and serum-free light chains. Risk factors for thrombophilia were low: she was 18 months postpartum and was on a non-estrogen contraceptive (Depo Provera). The swelling was self-resolving and the patient was subsequently discharged and treated with a 2-month course of rivaroxaban.

Her second presentation in February 2015 had similar symptomatic onset; due to differing pathology on imaging, she was referred to ear, nose, and throat. An USS demonstrated chronic left IJV thrombus; however, this did not appear on the CT. Instead, the imaging was reported as a cyst in the left lower neck lesion with non-specific appearance. Both CT scan (Fig. 1) and MRI scan (Fig. 2) showed swelling at the insertion of the thoracic duct. Differential diagnoses were suggested including a third branchial cleft cyst and

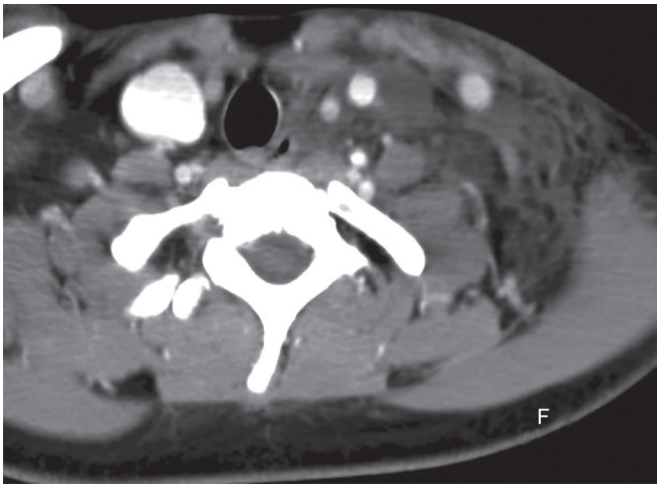

Fig. 1: Both CT scan showed swelling at the insertion of the thoracic duct

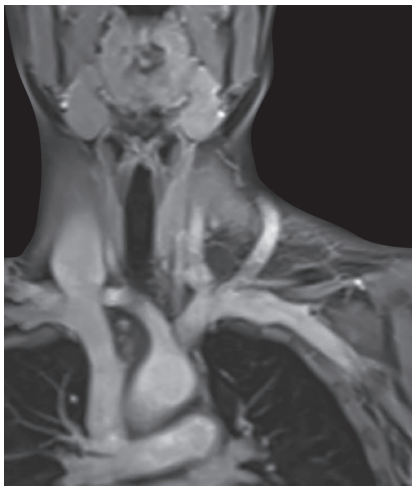

Fig. 2: MRI scan showed swelling at the insertion of the thoracic duct

cystic necrotic lymph node. Fat stranding and edema of the left posterior triangle with subsequent lymphocele and lymphedema were in keeping with a thoracic duct disruption. Bilateral small pleural effusions were revealed and subsequent USS guided pleural tap elicited fluid with a triglyceride level of 17.1 mmol/L diagnostic of chylothorax. Again, the swelling resolved with only conservative management.

In January 2016, she presented to the emergency department with symptoms of sudden onset of swelling to her left neck; however, this time, she reported associated mild dysphagia. An USS neck showed a collection at the site of insertion of the thoracic duct but no evidence of a thrombus and no apparent cause. The collection resolved at day 6; USS scan demonstrated a normal thoracic duct.

Management and Outcome

As discussed above, in this case report, conservative management was the mainstay with the neck swellings resolving within a week. The chylothorax was drained to assist in the second presentation but otherwise again treated conservatively. Only the IJV thrombosis was actively managed with a 2-month course of rivaroxaban. At time of writing, this patient was yet to re-present for ongoing issues and/or recurrent rupture.

A Medline (1946-present) and Embase (1947-present) search was conducted for case reports using keywords "rupture, spontaneous" or "internal jugular vein" and "thoracic duct". Additional references of selected papers were also reviewed. Only one case has been reported of spontaneous rupture of the thoracic duct as published by Connerley in 1955.[1] Six cases have been reported as result of IJV cannulation resulting in chylothorax.[5–9] In a case review by Doerr et al., three patients had chylothorax secondary to subclavian thrombosis.[10] This is the first reported case of thoracic duct rupture secondary to IJV thrombosis.

Discussion

Thoracic duct rupture is rare and most commonly caused by trauma. However, there was no evidence of prior trauma in this case study. Although no formal diagnosis was made at the time, the first rupture of the thoracic duct in this patient may have been precipitated by an internal jugular thrombus. It is hypothesized that a spontaneous IJV thrombus over the insertion of the thoracic duct may create a high enough pressure environment for the duct to rupture. Internal jugular vein thrombosis itself is rare and usually due to IV drug abuse, prolonged venous catheterization or deep head–neck infections or trauma; none of which was demonstrated in this patient. The presence of a chylothorax with a diagnostic pleural tap triglyceride level was the greatest contributing factor to the diagnosis of thoracic duct rupture. However, interestingly, a thrombus was only present in the first two presentations; in the final presentation, imaging demonstrated no such thrombus suggesting that even if this had been the precipitating factor in the previous presentations, this was not the cause of the final thoracic duct rupture. Whether the initial insults weakened the structure to allow a seemingly benign insult result in the final rupture is unknown.

Conclusion

This case is unique as this patient has two presentations, one that has never been presented before—thoracic duct rupture precipitated by an IJV thrombus, and a second rarely seen: subsequent spontaneous rupture with no obvious cause. At this stage, it is difficult to surmise whether this last rupture is due to weakness from the previous ruptures; if it were to rupture again with no obvious precipitants, then this would strengthen this theory but only time will tell.

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