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Spontaneous cervical chyle fistula: A case report

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ABSTRACT

Background: Cervical chylous fistulae are rare complications usually occurring from iatrogenic injury to the thoracic duct. There have been no reported cases of spontaneous chyle leaks in surgical naïve necks.
Methods: Case report.
Results: A 50 year-old female presented with progressive left neck swelling without fever, dyspnea, or dysphagia. Imaging demonstrated extensive infiltrative changes of the left neck with retropharyngeal fluid extending into strap musculature and the mediastinum. Flexible laryngoscopy revealed posterior pharyngeal wall edema. Differential diagnosis included abscess versus necrotizing fasciitis. Broad-spectrum antibiotics were initiated and she was taken to the OR for neck exploration. Intra-operatively, milky fluid was present around the carotid sheath and in the retropharyngeal space. Fluid analysis demonstrated chylomicrons and triglycerides > 2400 mg/dL. Repeat imaging of the neck, chest, and abdomen did not reveal malignancy or obstructive masses. A lymphangiogram showed dilated lymphatic vessels near the cervical thoracic duct. On post-operative day four, the patient was taken back to the OR for thoracic duct ligation and biopsy of nearby tissue. Pathology demonstrated benign lymph nodes with dilated sinusoids. A low-fat diet was started and she was discharged home on hospital day nine. She has followed up regularly with no signs of recurrence.
Conclusion: A cervical chylous fistula usually results from iatrogenic injury to the thoracic duct. To our knowledge, this is the first reported case of a spontaneous cervical chyle leak.

1. Introduction

Cervical chylous fistulae usually occur from iatrogenic injury to the thoracic duct and are rare complications, occurring at a rate of 1–2.5% [1]. We describe a novel case of an idiopathic, spontaneous cervical chyle leak and evaluation of this entity in a surgically naïve neck.

2. Case report

A 50-year-old female presented to an outside emergency department with rapidly progressive neck swelling without fever, dysphagia, or dyspnea. She denied recent infection or surgery. Her past medical history included obstructive sleep apnea and hypothyroidism. She was in the process of weight training but denied any trauma. Imaging of the neck and chest showed infiltrative changes of the left neck with retropharyngeal fluid extending into the strap musculature and the mediastinum (Fig. 1). She was transferred emergently with concern for necrotizing fasciitis versus retropharyngeal abscess. Physical exam was unremarkable other than soft, non-tender, non-erythematous left neck swelling. Flexible laryngoscopy demonstrated left posterior pharyngeal edema.

The patient was started on intravenous antibiotics and underwent direct laryngoscopy, esophagoscopy, and transcervical incision and drainage of a possible retropharyngeal abscess. During dissection adjacent to the carotid sheath, a large amount of milky fluid was encountered. The fluid was sent for microbial analysis, triglycerides, and chylomicrons. A drain was placed. The patient was transferred to the intensive care unit post-operatively and extubated the next day.

Fluid analysis demonstrated chylomicrons as well as triglycerides > 2400 mg/dL, confirming a chyle leak. Conservative management was initiated with a low-fat diet. Full-body imaging was performed to investigate for a malignancy or lymphoma as the source of the chyle but was negative. A lymphangiogram was performed, demonstrating small cystic spaces near the terminal thoracic duct (Fig. 2). Thus, the patient was taken to the operating room for left neck exploration, and large lymphatic channels and the thoracic duct were ligated. Biopsies were performed of the adjacent tissue and were negative for malignancy. The drain was removed on postoperative day three and she was discharged four days later. She has been seen in follow-up over one-and-a-half years with no evidence of recurrence.

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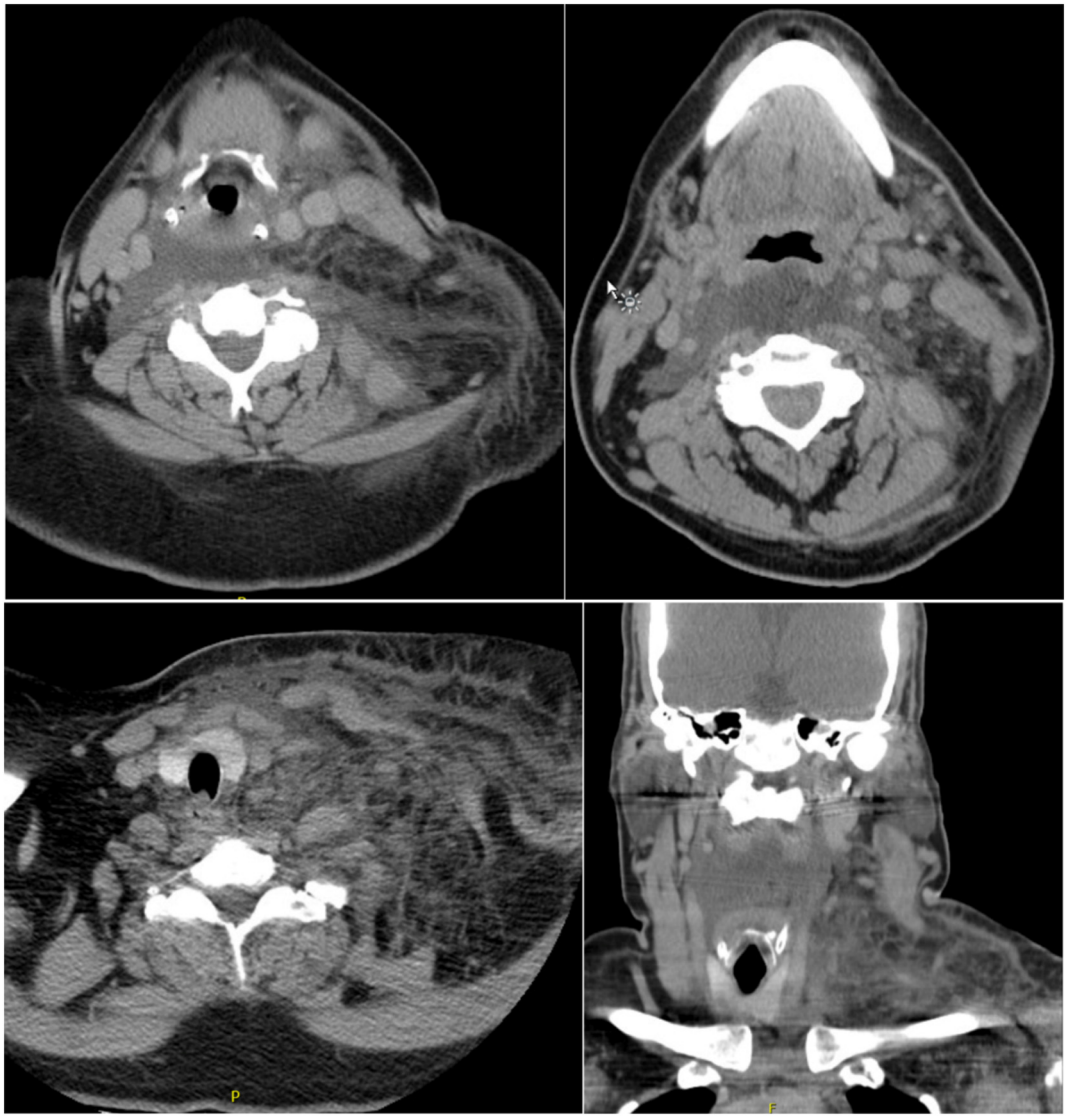

Fig. 1. Representative axial and coronal sections of a pre-operative CT neck with contrast demonstrating infiltrative changes and retropharyngeal fluid.

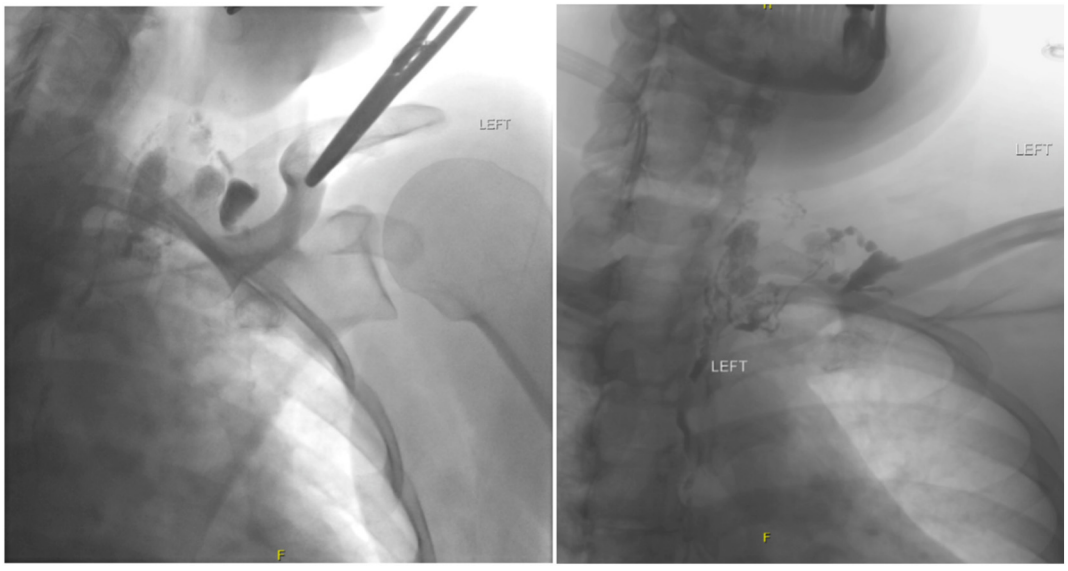

Fig. 2. Representative anterior-posterior and lateral views of a lymphangiogram demonstrating cystic changes in the vicinity of the thoracic duct.

3. Discussion

Spontaneous chyle leaks, including chylothorax and chylous ascites, have been reported in the literature, but are rare [2]. Generally, spontaneous chyle leaks are divided into traumatic versus atraumatic etiologies [3]. Traumatic chyle leaks include iatrogenic cases. In atraumatic cases, obstruction due to malignancy must be excluded. Other causes include tuberculosis, cirrhosis, sarcoidosis or lymphatic malformations [3]. Work-up for this patient did not reveal masses or cirrhosis – however, cystic spaces were seen near the left thoracic duct, concerning for dilation of lymphatic channels.

Although there are no reports of spontaneous cervical chyle leaks, there are multiple reports of dilated cervical thoracic ducts, lymphoceles, or cysts found in patients presenting with a left supraclavicular mass [4]. These were often diagnosed by needle aspiration and imaging. These authors state that high suspicion for a thoracic duct cyst is important with a left-sided supraclavicular mass, as surgical intervention may lead to chyle leak [4]. These cysts are thought to occur secondary to congenital, degenerative, or post-traumatic weakness in the thoracic duct wall [4,5]. A review of 33 reports of thoracic duct cysts indicated that 16 of these cases were primary cysts, while the remaining were secondary to trauma, obstruction, or inflammation. Management usually involves excision of the cyst or ligation of the duct [4]. Given the lack of evidence for malignancy or obstructive masses in this patient, straining during her weight training may have contributed to the rupture of a pre-existing thoracic duct dilation or small cyst, causing the rapid presentation of her symptoms.

To our knowledge, this is the first reported case of a spontaneous cervical chyle leak presenting with rapidly enlarging neck swelling. Although there have been multiple reports of dilations or cysts of the cervical thoracic duct [4], this case represents possible rupture of dilated cervical lymphatic channels. Thus, although rare, if evaluation of a spontaneous chyle leak is negative for malignancy or obstructive masses, this etiology should be considered and managed appropriately.

4. Conclusions

Although chylous leaks have been described in post-surgical necks, this case appears to be the first documented occurrence in a surgically naïve neck with no determined inciting event. Physicians who are presented with a patient with rapidly progressive neck swelling concerning for necrotizing fasciitis should keep chylous effusion among their list of differentials, even in a surgically naïve patient with no inciting event.

Conference Information

This is an original case report which was presented as a poster at the AAO-HNSF 2017 Annual Meeting in September 2017.

Disclosure

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