

# The association of unusual site of an idiopathic deep vein thrombosis and chylothorax in an adult

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## Abstract

Deep vein thrombosis (DVT) usually presents with pain, swelling, and tenderness, but very uncommonly does it occur in the left cervical region. The association of a DVT with chylothorax as a complication in adults is very uncommon. We present a 53 year-old female that complained of a progressive painful left lower neck swelling and redness for 2 days. These symptoms were preceded by transient painful and erythematous swelling in her left axillary region one week prior to admission. Physical exam was remarkable for tender erythematous swelling around left lower cervical and supraclavicular regions, and peripheral venous dilatations of left upper chest wall and proximal left upper extremity. Work-up with x-ray and CT scan of the chest revealed mild-to-moderate sized right-sided pleural effusion without mediastinal abnormalities. Thoracentesis revealed findings consistent with chylothorax. An inconclusive mammogram lead to additional imaging with ultrasound that yielded an unusual site of thrombosis in the left subclavian and external jugular veins. Especially in adults, this site of DVT is very uncommon, and more so, was the associated complication of chylothorax, with an unremarkable history and an unyielding work-up, etiology of such an unusual site of DVT was not found.

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## BRIEF DESCRIPTION

Adults presenting with cervical erythematous swelling and tenderness can indicate a variety of significant diseases such as malignancy, infection, or trauma. Deep vein thrombosis and chylothorax alone are two independently rare incidences in adults, and there association can usually always indicate an underlying process present. Although here, we report a case of an extremely rare incidence of an idiopathic DVT of the left subclavian and external jugular veins producing chylothorax in an adult patient with no underlying risk factors.

Key Points: Cervical or upper extremity DVT's have been rarely reported in adults. Of those reported, very small portion of DVT's in this region had even produced chylothorax. Causes have been attributed by a majority cases being due to traumatic events and not idiopathic. Diligent searches for underlying etiology may be prompt due to the

association of malignancies presenting in such a matter in adults. With our report, we find that with ruling out such significant diseases, findings in these patients can also hold an idiopathic nature as well warranting no further testing and just continue treatment with observance.

## QUESTIONS

All of the following can produce a DVT in the cervical region except:

1. Malignancy
2. Central venous catheter
3. Smoking
4. Estrogen therapy

Where does the thoracic duct drain?

1. Superior Vena Cava
2. Subclavian Angle
3. Inferior Vena Cava
4. Brachial vein

(T/F) Chylothorax is a common abnormality in children but very rare in adults.

Answers: 1. C; 2. B; 3. T

## **CASE REPORT**

53-year-old Latin American female with a history of hypertension and osteoarthritis presented to the hospital with 2 days of progressive painful left lower neck swelling and redness. These symptoms were preceded by transient painful and erythematous swelling in her left axillary region one week prior to admission. She denied any swallowing difficulty, or fever.

Patient's current medications included lisinopril, hydrocodone and acetaminophen. She lived in El Paso, Texas, with no recent travel. She denied alcohol and illegal drug use but had a 10 pack-year smoking history. Trauma, stings, or bites were denied.

Temperature was 35 degrees Celsius, pulse was 86 beats per minute, blood pressure was 173/101 mm Hg, and respiratory rate was 18 breaths per minute. Physical exam was remarkable for tender erythematous swelling around left lower cervical and supraclavicular regions, and peripheral venous dilations of left upper chest wall and proximal left upper extremity (Figure 1). No lymphadenopathy was appreciated in axillary, cervical, or inguinal areas. Lung auscultation revealed decreased breath sounds at the right base.

A complete blood count and complete metabolic panel were within normal limits. Hepatitis panel was non-reactive. A chest x-ray indicated moderate right pleural effusion. Cefazolin was administered for a possible cellulitis, and a further testing was initiated to evaluate the pleural effusion. A CT scan of the chest revealed mild-to-moderate sized right-sided pleural effusion without mediastinal abnormalities. Thoracentesis revealed turbid yellow exudates with 64 percent lymphocytes, 719 triglycerides mg/dL, LDH 107IU/L, Glucose 92 mg/dl, Protein 4.7 g/dl, WBC 3500, negative cytology. Results were most consistent with chylothorax.

Mammogram showed inconclusive results so additional imaging with breast ultrasound yielded an unusual site of thrombosis in the left subclavian and external jugular veins. Confirmation of acute deep venous thrombosis was confirmed with Doppler studies showing extent of thrombosis through left external jugular, subclavian,

brachial, and basilic veins. Cefazolin was discontinued and low molecular weight heparin was initiated.

CT chest, abdomen and pelvic showed moderate right side pleural effusion and moderate abdominal and pelvic ascites. No other abnormality was identified.

Further studies showed normal levels of protein C and S, antithrombin antibodies, rheumatoid factor, anti-nuclear antibody, cardiolipin IgG and IgM, and antiphospholipid antibodies.

## **Figure 1**

Figure 1: Left Anterior Cervical and Shoulder view (Image depicts skin hyperpigmentation, erythematous edema, along with prominent superficial venous congestion)



## **DISCUSSION**

Deep vein thrombosis (DVT) usually presents with pain, swelling, and tenderness, but very uncommonly does it occur in the left cervical region. The incidence of DVT can occur secondary to recent surgery, hospital admission, trauma, immobility, antiphospholipid antibody, familial thrombophilias, and serious illnesses including malignancy, chronic heart failure, stroke, chronic lung disease, acute infections, and estrogen therapy<sup>1-2</sup>. However, none of these were appreciated. In an extensive review of literature by Sajid et al.(3) it was found that incidence of an axillary-subclavian DVT occurred in 1-4% of the total cases reported with majority (80%) derived secondarily and only 20% being primary. Consequently, the rarity of the association of DVT with chylothorax as a complication in an adult has led us to report this incidence.

Occurrence of chylothorax is very uncommon(4) and is usually elicited by the compression, rupture, or leakage of the thoracic duct anywhere along its course into the pleural

space. In a review by Skandalakis et. al.(5), a description of its course in the thoracic and cervical regions begins as it ascends at the level of T7 in which it transverses behind the esophagus, then at the base of the neck, passes posterior to the common carotid artery, internal jugular vein, and vagus nerve, continuing its journey to a level of C7 before it descends across the subclavian artery and ends at the venous angle between the left subclavian vein and left internal jugular vein(5).

In chylothorax, a variable appearance of pleural fluid may occur but it is frequently nonmilky, exudative, and contains triglycerides, characterized by presence of chylomicrons(3) Etiologies of chylothorax has been described in many reviews(6,7,8) with vast majority induced by malignancy (ex. Lymphomas, bronchogenic carcinomas (8), lymphangioliomyomatosis(20), trauma (including surgery(7,9,10), catheter placement(7,11), blunt chest trauma(7,8), etc.), infections (such as tuberculosis(12), histoplasmosis(13), or filariasis(14)), and the minority of causes resulting from mediastinal radiation exposure, congenital syndromes (ex. Noonan syndrome, Down syndrome, congenital lymphangiectasia, tracheoesophageal fistula, and thoracic duct hypoplasia(15-16)) or even idiopathic. Although chest and neck surgery alone contributes to the majority of these cases (8), they are still considered uncommon complication such of that radical neck surgery producing a reported rate of 1-3%(10).

With the common association of mediastinal malignancies, chylothorax may be the presenting symptom of lymphomas(17-18). Therefore, nontraumatic chylothorax is an indication for a diligent search for malignancy. CT chest, abdominal and pelvis did not identify any masses. Thrombosis of the superior vena cava or the subclavian vein has become one of the more common causes of chylothorax(15-16,21) but in the pediatric population not the adult. Berman et al.(21) reviewed case histories of 37 infants and children with thrombosis of their superior vena cava in a newborn and pediatric intensive care unit and reported that nine (24%) had a chylothorax.

While the association of thrombosis in external jugular and subclavian veins with chylothorax is identified and isolated in pediatric care unit, it is extremely rare in adults.

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