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A RARE CASE OF RIGHT-SIDED CHYLOTHORAX INDUCED BY VERTEBRAL OSTEOPHYTOSIS IN A NONAGENARIAN PATIENT

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ABSTRACT

Background: Chylothorax is an abnormal accumulation of lymphatic fluid in the pleural cavity, and it develops because of chyle leaking from the thoracic duct. The clinical presentation can be varied, while symptoms depend on its size and the rate of accumulation in the pleural space. Diagnosis is based on pleural fluid analysis, supported by various imaging studies to help determine the underlying pathology and locate the site of chyle leak.

Case report: We report an extremely rare case of non-iatrogenic right-sided chylothorax in the absence of radiographically demonstrated bony damage which was related to vertebral osteophytosis in a 90-year-old man, who presented with worsening shortness of breath after a blunt trauma. On physical examination, the patient was found to be suffering from substantial right-sided pleural effusion with no radiographic evidence of traumatic injury.

Conclusion: Imaging demonstrated diffused osteophytosis at the level of the lower thoracic vertebrae, which were puncturing the thoracic duct. We adopted conservative treatment, including thoracentesis and total parenteral nutrition, which resulted in the improvement and resolution of symptoms.

KEYWORDS

Chylothorax, vertebral osteophytosis, elderly

LEARNING POINTS

- Vertebral osteophytosis may damage the thoracic duct and cause chylothorax.

INTRODUCTION

Chylothorax is a rare condition that results from thoracic duct damage with chyle leakage from the lymphatic system into the pleural space. It has multiple aetiologies and is usually discovered after it manifests itself as a pleural effusion and can be classified as either traumatic or non-traumatic.

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Chylothoraxes are unilateral in about 80% of all cases (50% right sided, 30% left sided), while bilateral presentation occurs in 16% of cases. However, overall, it accounts for 3% of all causes of pleural effusion[1]. The macroscopic appearance of chylothorax is typically described as milky, though it may sometimes appear serous or blood-stained because of underlying disorders and the nutritional status of the patient. Even though the definitive diagnosis involves the detection with cytological analysis of chylomicrons stained with Sudan III and lipoprotein electrophoresis of the pleural fluid, this remains an expensive and often inaccessible method for centres without available facilities like ours. Therefore, in clinical practice, diagnosis is based on the measurement of pleural fluid triglyceride and cholesterol levels, a widely adopted method. Specifically, a pleural fluid triglyceride level > 110 mg/dl (>1.24 mmol/l), and a cholesterol level < 200 mg/dl (<5.18 mmol/l) are diagnostic for chylothorax. In addition, an important role in clarifying the transudative or exudative nature of chylothorax is also played by the analysis of lactate dehydrogenase (LDH) and protein in pleural fluid[2]. Computed tomography (CT) lymphangiography is considered the gold-standard procedure for detecting the site of leakage or blockage; nonetheless, it is not without risk, especially in elderly patients with comorbidities. CT scan of the chest and abdomen (with or without contrast) remains central and it is useful in identifying and/or excluding non-traumatic causes (such as malignancy, lymphatic disorder) or traumatic injury to the lymphatic system (such as oesophageal, cardiovascular and neck surgery or mediastinal lymph node dissection). The management of chylothorax, especially when it produces significant respiratory symptoms, is primarily based on drainage that can be achieved through therapeutic thoracentesis. Conservative management includes nutritional input and dietary modification, pharmacological therapy and treating the underlying cause, if possible, while surgical intervention may be considered when conservative management fails.

CASE DESCRIPTION

A 90-year-old man presented with progressive dyspnoea, which was worsened by coughing and being in the supine position and associated with right-sided chest pain for 1 week. One week prior, he had fallen to the floor, striking his back and chest against a table; however, he continued with his regular activities. His medical history included heart failure with preserved ejection fraction, chronic kidney disease, atrial fibrillation and insulin-resistant diabetes mellitus. On physical examination, the patient appeared to be in obvious discomfort. His vital signs were as follows: respiratory rate of 22 breaths/minute; heart rate of 85 beats/minute (BPM); blood pressure of 150/90 mmHg; oxygen saturation of 90% on room air. The patient reported no fever, nausea, or vomiting. Sensory function was intact. Chest examination revealed significantly decreased breath sounds on the right-side of the chest, with no chest wall tenderness, rhonchi and rales in the contralateral auscultatory field. Laboratory analysis revealed: hypoalbuminemia (2.1 g/dl) and hypogammaglobulinemia (0.37 g/dl) on serum-protein electrophoresis. Plain chest X-ray demonstrated a large right-sided pleural effusion and consensual atelectasis of lung parenchyma. A chest CT scan without contrast confirmed the above, with no evidence of rib or vertebral fracture *(Fig. 1A)*. A thoracentesis was performed on the right side of the chest with an indwelling a pleural catheter and approximately 1500 ml of milky white effusion was drained. After this procedure the patient experienced rapid improvement of symptoms. Pleural fluid analysis was consistent with chylothorax revealing triglycerides:136 mg/dl, cholesterol: 40 mg/dl, LDH: 295 U/l, and total protein: 5.7 g/l, with the absence of malignant cells on cytological analysis and no growth on microbiological examination. Lymphangiography could not be performed because of the risk it posed to the elderly patient. However, after re-evaluating the CT images, we noticed an important irregularity of the vertebral profile with osteophyte protrusion at T9-T10 levels *(Fig. 1B)*. Vertebral osteophytes are outgrowths that form due to progressive chronic bone degeneration, especially in elderly individuals. Since a non-traumatic aetiology ruled out vertebral and rib fractures, we hypothesized that the stretching of the spine during the previous fall and the pre-existing vertebral osteophytes likely punctured the thoracic duct, causing the extravasation of chyle into the pleural space. Conservative management was adopted, including dietary modification and total parenteral nutrition (TPN) and bowel rest. The drain was kept in place for 10 days and always produced <1000 ml/day, until the chylous drainage consistently decreased and appeared clear and minimal. The patient's condition gradually improved *(Fig. 2)*; he resumed a normal diet, the chest drain was removed, and he was discharged home in a good general condition.

DISCUSSION

Chylothorax, whether traumatic or non-traumatic, is a rare condition and an atypical presentation of pleural effusion and is not normally suspected in an old patient with several comorbidities, who presents to the hospital complaining of dyspnoea and coughing. It is characterized by the accumulation of chyle in the pleural space, and results from the rupture of the thoracic duct. Amongst traumatic cases, the leading cause is iatrogenic, because of damage caused to the thoracic duct during chest surgery, especially oesophageal surgery. However, traumatic chylothorax in the absence of obviously significant traumatic injury is less commonly reported as protection of the thoracic duct is guaranteed posteriorly by the thoracic and lumbar spine, and anteriorly by the mediastinum[3,4]. In our case we suggest that pre-existing vertebral osteophytosis was the likely culprit responsible for the thoracic duct being punctured, following the blunt trauma that occurred to our patient 1 week prior to admission to the hospital. In trying to explain this pathogenetic mechanism, we conducted a thorough analysis of the literature. In 2009, Apostolakis et

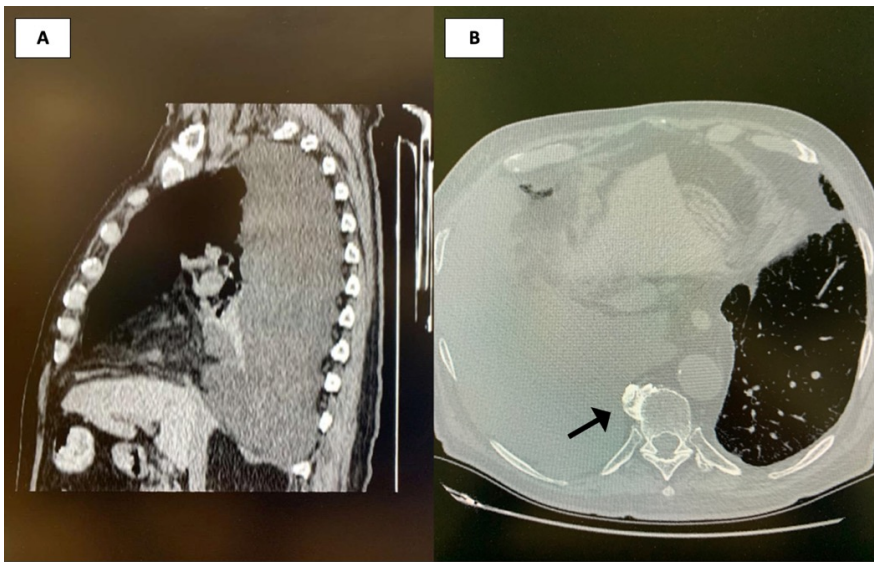


Figure 1. A computed tomography scan of the chest showing (A) massive pleural effusion and (B) vertebral osteophytes (arrow).



Figure 2. Patient phenotype and chyle.

al.[5] described a case of a 75-year-old female with bilateral chylothorax following blunt trauma which appeared to be due to thoracic duct perforation by existing osteophytes. In this case conservative management was adopted, which resulted in a good response from the patient[5]. In 2019, Horiuchi et al.[6] described an interesting case of bilateral chylothorax associated with osteophytosis of the lumbar vertebrae in a 71-year-old male. After a thoracic trauma 3 months earlier, the usefulness of lymphangiography in detecting the thoracic duct lesion was confirmed and the patient was given conservative treatment[6]. In our case, considering the patient's advanced age (90 years) and his comorbidities, re-evaluation of the radiographic images highlighted the large osteophytes of the lower thoracic vertebrae puncturing the thoracic duct. Furthermore, due to his advanced age and comorbidities and to prevent some of the complications that can easily occur in a frail patient such as malnutrition, immunosuppression, and dehydration, we preferred to use conservative management including thoracic drainage and TPN, preferring not administer octreotide or somatostatin.
Overall, considering the patient's sex and the age of onset, the pleural effusion, the abundant vertebral osteophytes, and the use of CT scan alone for their identification, in our case is, to the best of our knowledge, the first case of traumatic non iatrogenic unilateral (right-sided) chylothorax in the absence of obvious traumatic injury to be reported in the English language medical literature.

CONCLUSION

Non-iatrogenic traumatic unilateral chylothorax is rare, and its occurrence in the absence of posterior rib or thoracic vertebral fractures, as in our nonagenarian patient, is even more rare. Vertebral osteophytes may damage the thoracic duct by blunt trauma and cause chylothorax. Thus, although it is a rare occurrence, we suggest that chylothorax associated with osteophytosis at the lower thoracic vertebrae should be considered in the differential diagnosis of patients presenting with shortness of breath and dyspnoea after experiencing blunt trauma, even in the absence of traumatic fracture or injury on radiographic imaging.

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