Case Report

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Idiopathic chylothorax in geriatrics: the oldest case report so far

Geriatrik Popülasyonda İdiopatik Şilotoraks: Mevcut Literatürdeki En Yaşlı Şilotoraks Olgusu

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Abstract: Chylothorax is a rare condition characterized by accumulation of chylous fluid in the pleural space resulting in impaired ductus thoracic integrity. It can be an outcome of a traumatic process, although there are a few non-traumatic and/or idiopathic cases in current literature. In this article, we present the oldest case report so far, who is an 87-year-old woman complaining of acute respiratory distress symptoms with pleural effusion having no trauma history. The patient was analyzed for the disease etiology strenuously. Thoracentesis was performed together with imaging modalities and detailed systemic laboratory tests. Non-surgical treatment was successful as the outcome.

Keywords: Chylothorax; Geriatrics; Pleural fluid.

Özet: Şilotoraks, plevral boşlukta şilöz sıvı birikimi ile karakterize, duktus torasikusun bütünlüğünde bozulmayı ortaya çıkabilen nadir bir durumdur. Travmatik bir sürecin sonucu olarak şilototaks ortaya çıkabilse de, mevcut literatürde travmatik olmayan ve/veya idiopatik vakalar da bildirilmektedir. Bu çalışmada, travma öyküsü olmayan, plevral efüzyonlu, akut respiratuar distres semptomları bulunan 87 yaşındaki bir kadın hasta, mevcut literatürdeki en yaşlı şilotoraks olgusu olarak sunulmaktadır. Hastalığın primer etiyolojisinin bulunması için yoğun bir araştırılma yapılmıştır. Görüntüleme yöntemleri, ayrıntılı sistemik laboratuvar testler ile birlikte torasentez uygulanmıştır. Hastamızda, cerrahi girişim olmadan uygulanan tedavi, plevral efüzyonun gerilemesi ile başarılı bir sonuç vermiştir.

Anahtar Kelimeler: Şilotoraks; Geriatri; Plevral sıvı.

Introduction

Chylothorax is the presence of the chyle in the pleural cavity due to thoracic duct disruption [1–3]. Main causes of chylothorax are categorized as traumatic, non-traumatic or idiopathic [3]. Idiopathic cases at geriatric population can be seen rarely in clinical practice. Typically, the pleural fluid is rich in chylomicrons and triglycerides [2, 3]. Diagnosis of chylothorax is based on biochemical analysis of the pleural fluid; the most important parameter is triglycerides level greater than 110 mg/dL. When diagnosed, first of all, conservative treatment should be started and careful follow up is necessary. In cases which do not respond to conservative treatment, surgical integrity of the ductus thoracicus can be chosen as an alternative treatment modality. Herein, we present an 87-year-old woman with chylothorax that we did not find any particular etiological cause of the pleural effusion.
Patients and methods

An 87-year-old woman, complaining of acute respiratory distress symptoms, admitted to our emergency department. In her examination, widespread compression wounds were observed in her back and coccygeal regions. She was not responding to verbal warnings at the time of admittance. She was only making a grimace to painful warnings. The most prominent finding in her medical history was having had a cerebrovascular disease 2 years ago. Afterwards she had been confined to bed. She had been feeding from percutaneous endoscopic gastrostomy (PEG) for 2 years. She had also tracheostomy 2 years ago. Her chest X-ray showed a prominent left pleural fluid at the time of admittance (Figure 1) and the fluid sample was taken by thoracentesis. The pleural fluid was milky-pink in appearance, which was skeptical about having high triglyceride level. Gram stain and bacterial culture results revealed negative. Acid fast bacillus analysis was considered as an important diagnostic tool due to relatively high tuberculosis incidence in the region and debilitated status of the patient. Microbiological assessment for tuberculosis and viral serological markers (HBsAg, anti-HIV and anti-HCV) were negative, as well. Biochemical analysis of the fluid was performed at the same time and it revealed 633 mg/dL triglyceride value. Based on these findings chylothorax was diagnosed. From that point, diagnostic evaluation was deepened by other imaging modalities to enlighten the cause. Thorax tomography of the patient showed only pleural effusion on the left side and there were no other findings such as lymphadenopathy, tumor, space occupying lesion or any other diseases (Figure 2). Laboratory findings of the plasma and chyle of the patient were also compared to establish the final diagnosis (Table 1) [2, 4]. Similar electrolyte results between plasma and the fluid, high triglycerides values and also other parameters strengthened the diagnosis. Lymphangiography was not

Figure 1: Chest X-ray of the patient when she admitted to our hospital: there is a large left sided pleural effusion.

Figure 2: Axial plan thorax CT mediastinal window showing significant amount of pleural effusion (arrow).

### Table 1: Comparative analysis of serum and pleural fluid [2, 4].

| Parameter       | Serum results of the patient | Reference range of serum | Pleural fluid results of the patient | General composition of chyle | Units   |
|-----------------|------------------------------|--------------------------|-------------------------------------|-----------------------------|---------|
| Triglycerides   | 208                          | 50–150                   | 633                                 | >110 and higher than plasma | mg/dL   |
| Total protein   | 4.7                          | 6.4–8.2                  | 4.1                                 | 2.21–6                      | g/dL    |
| Albumin         | 1.9                          | 3.4–5                    | 2.42                                | 1.2–3.6                     | g/dL    |
| Glucose         | 206                          | 74–106                   | 226                                 | 48–200                      | mg/dL   |
| Sodium          | 133                          | 134–146                  | 132                                 | Similar to plasma           | mmol/L  |
| Potassium       | 5.7                          | 3.5–5.1                  | 5.6                                 | Similar to plasma           | mmol/L  |
| pH              |                               |                          | 7.5                                 | 7.4–7.8                     |         |
Idiopathic chylothorax

The diagnosis of the chylothorax is based on biochemical analysis of the chyle. Pleural fluid triglyceride level which is higher than 110 mg/dL confirms the diagnosis of chylothorax with 99 percent [1–3]. In our case the pleural fluid sample color was pink-white and the sample did not have a bad scent. The biochemical analysis of the fluid showed 633 mg/dL of triglyceride. Based on these findings this patient was diagnosed definitely as “chylothorax”.

Main etiology of chylothorax are divided into traumatic, non-traumatic and idiopathic pathologies. Traumatic chylothorax is the most common reason and it accounts for more than 50% of all cases [3]. Non-traumatic chylothorax can be due to congenital, infectious or neoplastic diseases such as lymphoma [5]. Table 2 shows a detailed etiological classification of chylothorax [1–3]. As aerobic, anaerobic and mycobacterial cultures were negative, infectious chylothorax was ruled out. We excluded neoplastic chylothorax because thorax tomography of the patient showed remarkable pleural effusion on the left side but no other findings such as tumor, lymphadenopathy or other pathological lesions. There were also no remarkable suspicious findings in her repetitive complete blood count and routine biochemistry results. We also excluded traumatic chylothorax because our patient was confined to bed for 2 years and her medical history showed no surgery, blunt trauma to chest wall, penetrating injury to vertebra or radiation exposure. Based on

Discussion

Chylothorax is the presence of the chyle in the pleural cavity due to thoracic duct disruption [1, 3]. Chylothorax cases are usually unilateral which accounts for 83% of all cases [1]. The thoracic duct is the main collector of the lymphatic system which is connected to the lymphatics of various organs such as intestinal trunk that collects lymph from the stomach, intestine, pancreas and spleen. Chyle is consisted of chylomicrons, long chain triglycerides, cholesterol esters, phospholipids, and it is also rich in T cell lymphocytes, immunoglobulins, fat-soluble vitamins, water and electrolytes [1, 2]. Because of these symptoms chylothorax is a very important condition and untreated cases are possibly lethal. So, our clinical team approached the patient aggressively and initiated the treatment immediately.

Table 2: Etiological classification of chylothorax [1–3].

| Traumatic chylothorax | Postoperative chylothorax: |
|-----------------------|-----------------------------|
| Esophagostomy, thoracic surgery, head and neck surgery etc. |
| Blunt trauma to chest wall or vertebra |
| Penetrating injuries (knife wound, gunshot wound etc.) |
| Forceful emesis or cough |
| Radiation |

| Non-traumatic chylothorax |
|---------------------------|
| Congenital chylothorax: |
| Lymphangiectasis, lymphangiomatosis, tuberous sclerosis, congenital heart disease, chromosomal abnormalities such as trisomy 21 or 18 etc. |
| Neoplastic chylothorax: |
| Lymphoma, chronic lymphoid leukemia, lung cancer, esophageal cancer, metastatic carcinoma etc. |
| Infectious chylothorax: |
| Tuberculosis, sarcoidosis, filariasis, histoplasmosis etc. |
| Other conditions and rare diseases: |
| Retrosternal goitre, amyloidosis, haemangiomatosis, lymphangioleiomyomatosis, transdiaphragmatic movement of chylos ascites etc. |

| Idiopathic chylothorax |
these findings, we diagnosed our patient with idiopathic chylothorax. It is already known that idiopathic causes are quite rare and they account for only 10% of all chylothorax cases [3].

Treatment of the chylothorax starts with conservation therapy which consist of nil by mouth or a diet consisted of no long chain triglycerides but medium chain triglycerides which may be absorbed directly to the portal circulation. Total parenteral nutrition (TPN) may also be required [6]. Surgical treatment may be indicated if the chyle leak exceeds 1.5 L per day in an adult, >100 mL/kg per day for a child [7], more than 1 L for 5 days [8] or persistent chyle flow more than 2 weeks after conservative therapy [9]. Aboudo et al. reported two idiopathic chylothorax cases which were managed by different treatment modalities (surgical and non-surgical) in 2015 [10]. In our case, the patient has been feeding from PEG for 2 years. First, we ended feeding from PEG and started TPN that was rich in low fat and consisted of medium chain triglycerides. The amount of the pleural fluid reduced in 2 weeks and chylothorax treatment was successful just like the first case of Aboudo et al.

Conclusion

We present the oldest case of idiopathic chylotorax so far by this report. There is also no information yet which is similar to this case on the aspects of: presence of PEG, cerebrovascular disease, 2 years history of confining bed and finally acute presentation of chylothorax. This knowledge should make clinicians more vigilant and suspicious during approaching pleural effusions. It also enlightens differential diagnosis in geriatric effusion patients in a low prevalent incidence.

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Conflict of interest statement: There are no conflict of interest among the authors.

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