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Case Report

Pleural Effusion following Yoga: A Report of Delayed Spontaneous Chylothorax and a Brief Review of Unusual Cases in the Literature

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Abstract: Chylothorax is a rare condition where the extravasated chyle accumulates into the pleural space. It is most commonly associated with malignancies, infective or inflammatory disorders and iatrogenic causes. Extremely rarely, it could occur spontaneously. We present the case of a healthy 40-year-old woman who presented with acute right shoulder and neck pain associated with shortness of breath and loss of consciousness. This was preceded by a yoga class two weeks prior. Chest imaging showed right pleural effusion, and tapping revealed a milky fluid which was confirmed to be chylothorax. Conservative management failed and the patient was successfully treated with video-assisted thoracoscopic drainage, thoracic duct ligation and mechanical pleurodesis. Chylothorax association with yoga is not reported in the literature.

Keywords: chylothorax; pleural effusion; VATS; unusual chylothorax

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1. Introduction

Chylothorax is the accumulation of the extravasated chyle into the pleural space which may result from malignant disorders, inflammatory, and infective diseases and thoracic duct injury [1]. Injury of the duct can be caused by indirect trauma as a result of transmission of external forces but, more often, it results from iatrogenic during surgeries, and this contributes to almost 50% of the cases. Esophageal, aortic mediastinal, neck, and thoracic operations such as lobectomies or pneumonectomy are most associated with chyle leak [2]. Moreover, it may occur spontaneously, but this is a far rarer scenario and the most common causes need to be ruled out first (inflammatory, infective, neoplastic processes) [3].

A mimicking case of chylothorax is the pseudo-chylothorax, also described as a cholesterol effusion and chyliform effusion, which is a cholesterol-rich fluid often accompanied by chronic inflammatory disorders [4]. The clinical definition of a pseudo-chylothorax is the association of a milky pleural effusion, pleural cholesterol level greater than 200 mg/dL, pleural triglyceride level generally below 110 mg/dL, a pleural cholesterol/triglyceride ratio of greater than one, and often, the microscopic findings of cholesterol crystals [5].

On the contrary, chyle is an alkaline fluid that mainly consists of triglycerides, proteins, and lymphocytes, with a plasma-like electrolyte content [6]. The pleural fluid appears milky and is characterized by elevated triglycerides > 110 mg/dL or the presence of chylomicrons. Chylothorax is associated with significant morbidity, is potentially lethal, and can result in respiratory, immunologic, respiratory, and nutritional impairment predominantly due to poor fat absorption [7].

Clinically, patients can be asymptomatic or present with moderate respiratory symptoms only since the chyle does not irritate the pleural cavity. Once chyle is infected, they may present with sepsis [8]. In order to prevent severe complications, effective treatments are therefore urgently required. Herein, we describe and discuss the clinical presentation, diagnosis, and treatment of chylothorax, presumably caused by a yoga class in a juvenile lady.

2. Case Presentation

A 40-year-old woman attended the emergency department complaining of acute onset shortness of breath, right sided shoulder pain, and neck swelling. She had no previous medical history nor comorbidities and only reported attendance to her first yoga class 2 weeks prior. Clinical examination revealed lateral neck swelling. Her blood pressure and heart rate were within normal range. However, she did not require additional oxygen support to maintain saturations above 92% and her respiration rate was at 14 breaths per minute. Routine laboratory tests were all within normal limits. A chest X-ray showed minimal right pleural effusion which was conservatively managed, and she was discharged home with analgesia as she was clinically well. Only a few hours later, she was readmitted with an increasing shortness of breath, worsening pain, dysphagia, and an episode of collapse with a loss of consciousness. Within the following 48 h, she had three chest computed tomography (CT) scans including an oral contrast that showed a very interesting sequence of events. Initially, diffuse infiltration and non-specific soft tissue thickening of the right supraclavicular area and the whole mediastinum was found, particularly around the esophagus with radiological suspicion of esophageal perforation (Figure 1).

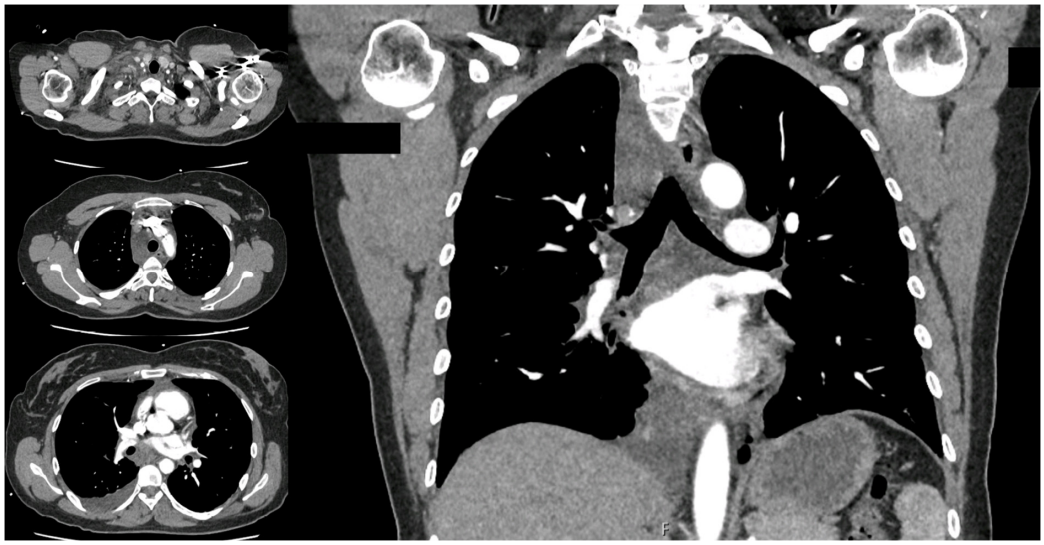

Figure 1. First CT scan axial and coronal views showing predominantly neck and diffuse mediastinal fat and soft tissue swelling with minimal pleural effusion.

The barium swallow scan, however, did not show any extravasation of contrast ruling out esophageal perforation. The third scan showed a significant increase in size of the right pleural effusion (Figure 2).

A diagnostic ultrasound-guided thoracocentesis revealed milky pleural effusion (Figure 3), which tested positive for chylomicrons, and the unexpected diagnosis of spontaneous chylothorax was made.

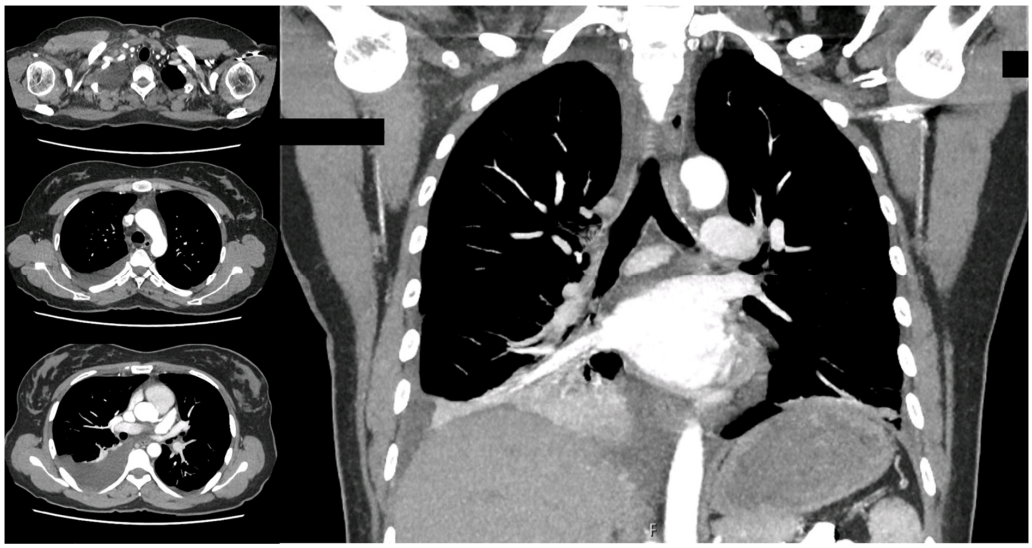

Figure 2. Further CT scan axial and coronal views showing the significant reduction in neck and mediastinal soft tissue swelling and the increase in the pleural effusion.

Figure 3. Aspirated sample of Chyle from thoracocentesis.

The patient was referred to the thoracic surgery department for treatment. Initially, conservative management was established with intravenous octreotide (300 µg/ daily, up to 600 µg/daily after three days of ineffective treatment) and a medium chain triglyceride (MCT) diet. Since the chylous drain output remained high (over 1000 mL for five consecutive days), surgical exploration was then scheduled and a 2-port right video-assisted thoracoscopic surgery (VATS) was performed. The pleural cavity was debrided and the chylothorax fully evacuated. No damage to thoracic duct nor obvious leak were found. We decided to perform the mass control of the thoracic duct together with the azygos vein by ligating both vessels and performing total pleurectomy to achieve pleurodesis. Satisfactory lung expansion was observed prior to skin closure. The patient remained on the MCT diet and octreotide for 5 days as the chyle output drastically reduced from post-operative day (POD) 2 and turned serous on POD 4. On POD 5, a normal diet was restarted and a further 48 h observation did not reveal an increase in drain output nor chyle leak. The drain was therefore removed, and the patient discharged home.

The patient did not exhibit malnutrition or weight loss and remained in a good general health condition throughout the treatment. At 3 months follow up, she was asymptomatic and chest X-ray showed complete resolution of the effusion and full lung expansion.

3. Discussion

Chylothorax is a rare cause of pleural effusion with several etiologies grouped broadly into traumatic and nontraumatic (spontaneous) causes. Most cases of spontaneous chylothorax are associated with minor trauma such as coughing, vomiting, stretching, and hiccups after a high-fat meal [2]. Infiltrating neoplasms located in the mediastinum or neck, or expansive solid lesions of another nature, can cause thoracic duct lesions and result in chyle loss, some of which are anecdotal, as reported by rare cases in the literature [9]. Trauma-related chylothorax contributes to almost 50% of all cases. Surgeries that involve posterior or superior mediastinum such as a foregut or aortic surgery or mediastinal lymph nodes sampling and lymph node dissection are also common causes. Idiopathic and medical causes account for 6% and 44%, respectively [2]. About 50–60% of chylothorax occur on the right side, 33.3% on the left side and about 16.6% will present with bilateral pleural effusions [8,10].

Chylothorax can be diagnosed either clinically or biochemically. Clinically, milky pleural aspirate suggests the presence of chyle; however, it has to be confirmed biochemically to exclude pseudochylothorax [10]. Triglyceride levels with a value of greater than 110 mg/dL is highly suggestive (99%) of chylothorax, whereas a value of <50 mg/dL is unlikely to be chyle [10,11]. Fluid cholesterol-to-triglyceride ratio of <1 is considered diagnostic as well [12]. Usually, careful history taking is the key to reaching the offending cause in patients with chylothorax. However, a CT chest scan is a very useful tool to assess for underlying malignancy as a cause of chylothorax.

Further diagnostic approaches, such as MR-lymphangiography, should be critically considered regarding feasibility, clinical benefit, and patient safety. Lymphatic system imaging studies remain challenging, though recently, non-enhanced MR-lymphography has been attempted in nontraumatic chylothorax [13]. It has also been reported that combining intermittent digital X-rays and live near-infrared imaging with microsurgical techniques, the "dual imaging lymphangiography" might be an option for refractory chylothorax [10]. However, such imaging techniques should be reserved for unresponsive or complicated cases [14,15].

Patients with chylothorax may be asymptomatic or present with non-specific symptoms such as chest discomfort, shortness of breath, and fatigue [12]. Moreover, if there was a chronic chyle loss, this could lead to nutrients, electrolytes, and volume loss in addition to the depletion of lymphocytes [10]. Ultimately, this will lead to malnutrition and immunosuppression.

The cisterna chyli is a lymphatic sac located at the lumbar level to the right of the abdominal aorta and behind the right diaphragmatic crus, at the level of L1–L2. The lymphatic flow from the lower half of the body converges to this saccular structure [16,17]. The cisterna chyli receives multiple lymphatic afferents and gives rise to the thoracic duct to the right of the aorta at the level of T12–L2. This primary lymphatic duct crosses the aortic diaphragmatic hiatus to enter the thorax, passing by the aorta, the esophagus, and the azygos vein. In 50% of people, the thoracic duct crosses from right to left at the level of T5–T6, passing the left brachiocephalic vein on the transverse plane and rotates inferiorly to the end in the subclavian vein at the confluence of the left jugular vein [16,17]. Therefore, a thoracic duct injury below the T-5 level results in a right chylous effusion, while a thoracic duct injury above T-5 results in a left chylous effusion. Because of the usual transition from the right to the left side of the thorax at the level of T3–6, a vessel injury at this point is expected to result in a chyle effusion in both pleural cavities.

Although there are a few reported cases of chylothorax after light exercises or a workout (Table 1), chylous effusion resulting from only the hyperextension of the neck secondary to the yoga exercise has not been reported in the literature [18–23]. In our case, the patient presented with respiratory symptoms 2 weeks following a yoga class, hence this was also a delayed presentation. Upon detailed history taking, there was no other obvious cause for the chyle leakage. We are raising the potential correlation between the yoga hyperextension of the neck and a spontaneous rupture of the thoracic duct. The duct more often drains

into the confluence of the left internal jugular and subclavian veins which is located at the cervical base in close relationship to the stretched cervical muscles over the left side. Interestingly, however, our case of chylothorax was on the right side. In this setting, one must be aware than anatomical variations of the thoracic duct pathway have indeed been described and that a right lymphatic duct that drains the right upper trunk, arm, and neck does exist as well [24].

Table 1. Unusual cases of chylothorax following nontraumatic physical activity.

Authors, Year	Subjects (*n*)	Supposed Cause	Location of Chyle Leakage	Intervention
Meade RH, 1972 [25]	5	Hyperextension of the spine	Unilateral chylothorax	N/A
Reilly et al., 1975 [26]	1	Hyperextension of the spine during stretching exercise and yawning	Bilateral chylothorax	Conservative
Gullane et al., 1984 [27]	1	Hyperextension of the spine while swimming	Bilateral chylothorax	Conservative
Tankanow et al., 1986 [28]	1	Hyperextension of the spine while climbing into a bathtub	Bilateral chylothorax	Conservative
Bocquel et al., 1997 [23]	1	Overstretch of subclavius and anterior scalenus muscle during stretching exercise	Unilateral chylothorax	Conservative
Torrejais et al., 2006 [18]	1	Hyperextension of the neck during light physical activity at a fitness center	Bilateral neck swelling and bilateral chylothorax	Conservative
Fehr et al., 2007 [29]	1	Hyperextension of the neck and overstretch of thoracic outlet during routine vacuum cleaning	Left-sided neck swelling and bilateral chylothorax	Conservative
Garcìa-Aparicio et al., 2009 [21]	1	Hyperextension of the neck during light physical activity	Bilateral chylothorax	Conservative
Bottet et al., 2019 [22]	1	Overstretch of subclavius and anterior scalenus muscle during stretching exercise	Bilateral chylothorax	Conservative
Kolbas et al., 2020 [19]	1	Overstretch of subclavius and anterior scalenus muscle while lifting 40 kg at a fitness center	Left-sided chylothorax	Conservative
Akbar et al., 2021 [20]	1	Hyperextension of the spine during stretching exercise	Left-sided neck swelling and bilateral chylothorax	Conservative

According to a brief literature review, the leading causes of thoracic duct rupture related to light physical activity are associated with forced postures of the backbone or overstretched junctions. The first report was presented by Meade [25], describing five cases of spontaneous unilateral chylothorax, believed to be correlated to the hyperextension of the spine with the rupture of an inherently weakened thoracic duct. In 1975, Reilly and Tsou [26] reported a case of bilateral chylothorax apparently associated with stretching exercises and yawning, assuming that a thoracic duct disruption was created by the hyperextension of the spine. Similarly, Gullane and Marsh [27] reported a case of bilateral spontaneous chylothorax presenting as an anterior neck mass after swimming and assumed the mechanism to be the same as that reported by Reilly and Tsou. In another reported case report, due to the absence of any demonstrable pathology, trauma, or structural abnormality, Tankanow et al. considered the sudden hyperextension of the spine while climbing into the bathtub as the cause that resulted in injury to a weakened portion of the thoracic duct, with the leakage of chyle in both pleural spaces [28]. An unusual presentation of chylothorax

was also reported by Fehr et al., who described a left-side neck mass and bilateral chylous pleural effusion occurred in a patient during routine vacuum cleaning, potentially related to an unnatural movement of the scapular girdle and consequent extension of the cervical spine while using the cleaning tool [29]. In all the reported cases, a conservative treatment (cessation of fatty food consumption and medium chain triglyceride diet supplement) was the choice that resulted in complete patient healing.

However, the management of chylothorax depends on the etiology. Other than conservative treatment, there are different approaches, such as non-surgical operative intervention or surgery [8].

Dietary fat restriction and the use of octreotide to reduce the production of chyle constitute conservative management. In clinically stable patients, it seems reasonable to begin therapy with a noninvasive, fat-free oral diet, and MCT supplement. Saturated fatty acids of eight-to-twelve carbon chain lengths are absorbed through the portal venous system, overriding the lymphatic drainage [28]. In our algorithm, we used octreotide early, as diet alterations alone did not lower the fluid patient's output.

Compared to somatostatin, octreotide has a longer half-life in circulation and can be administered subcutaneously [30]. The concurrent administration of octreotide or somatostatin with chest tube drainage and enteric rest in postoperative adult patients with chyle leaks has been shown to decrease the need for surgical intervention [31]. Although the octreotide mechanism of action is not entirely clear [32], it is known that its vasoconstrictor effect reduces intestinal blood flow due to the vasoconstriction of the splanchnic circulation, thus reducing gastric, pancreatic, and biliary secretions. Furthermore, fat absorption from the intestine is also reduced [33,34]. It has also been reported that the expression of somatostatin receptors SSTR2 and SSTR5 in the human thoracic duct and their stimulation may decrease lymphatic flow and lymph production [35,36]. In most reports, the benefit of octreotide treatment was seen within 2–3 days [37]. As recently published in a case series, orally administered propranolol, commonly used to treat childhood hemangiomas, appears to be an alternative treatment for chylothorax, especially in children [38]. The advantages of propranolol therapy include ease of administration, broad experience, and easy accessibility.

It is a matter of debate among authors as to when to shift the treatment from a nonoperative approach to operative means [39]. Once introduced by Cope et al., thoracic duct embolization (TDE) was accepted as the treatment of choice or at least as a viable alternative to surgical ligation in treating intractable postoperative chylothorax [40–43]. Thoracic duct cannulation (TDC) is the primary determining factor for the technical success of TDE due to its technical difficulty [40]. Different studies reported the conventional antegrade TDC's technical success rate to be about 70% [41,44,45]. Hence, significant numbers of patients for whom TDC had failed were forced to settle for the unproven therapeutic effect of Lipiodol during lymphangiography or thoracic duct disruption. However, neither procedure is as clinically efficient as TDE is [46,47]. Recently, novel retrograde transvenous or percutaneous cervical TDC techniques, with the aid of a bail-out retrograde approach, significantly improved overall technical success [48]. The main limitation of this nonsurgical operative approach lies in the availability of the necessary equipment and devices. For this reason, although recommended in selected cases, thoracic duct embolization techniques are to be reserved for high-volume and experienced centers [49].

Surgical interventions for managing chylothorax include thoracic duct ligation or clipping by thoracotomy or VATS, mass ligation, pleurodesis, pleurectomy, or pleuroperitoneal shunt. Thoracic duct ligation can be performed at the site of leakage or in the supradiaphragmatic region; in the latter case, success rates of up to 90% were reported [50]. In our case, the conservative treatment failed, and because leakage from the thoracic duct could not be identified intraoperatively, the mass ligation of the duct and azygos vein was performed by minimally invasive technique, followed by total pleurectomy. The aim of pleurectomy is to reduce the risk of recurrence and need for repetitive exploration. Alternatively, some authors report high success rates with talc pleurodesis for cases in which the thoracic duct

could not be identified [51]. Others suggest pleurodesis with iodopovidone in cases where leakage was identified and ligated [9]. With the adoption of surgical measures, the mortality rate of chylothorax decreased from 50% with conservative management to 10% [52].

4. Conclusions

Chylothorax, although rare, should be treated with a high index of suspicion. It should be considered as a differential diagnosis in any patient presenting with spontaneous pleural effusion in the context of very light physical exercise. Our case was quite unique in that it occurred after a prolonged time (2 weeks) following modest exercise after a neck extension only, during introductory beginner yoga techniques. Other cases of spontaneous chylothorax after exercise have been described, but more likely to be attributable to sudden, stressful movements than those predicted by yoga relaxation techniques. As defined by experiences reported in the literature, surgical management aided by conservative therapy was also successful in our case.

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