

CHYLOTHORAX, A RARE COMPLICATION AFTER ANTERIOR LUMBAR INTERBODY FUSION. CASE REPORT.

QUILOTÓRAX, UNA RARA COMPLICACIÓN LUEGO DE UNA ARTRODESIS LUMBAR INTERSOMÁTICA POR VÍA ANTERIOR. REPORTE DE UN CASO.

QUILOTÓRAX, UMA COMPLICAÇÃO RARA APÓS UMA ARTRODESE LOMBAR INTERSOMÁTICA POR VIA ANTERIOR. RELATO DE CASO.

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Dentro de las complicaciones esperables de una cirugía por vía anterior de la columna lumbar, como son las lesiones vasculares, viscerales, neuropraxias (lesiones nerviosas transitorias), retraso o ausencia de fusión e infecciones, la lesión de los vasos linfáticos y la fuga del quilo es de las menos frecuentes. Este evento se limita, la mayor parte de las veces, a fugas imperceptibles hacia el retroperitoneo luego de lesiones inadvertidas de los vasos perie spinales, y escasos reportes demuestran una conexión hacia la cavidad pleural con quilotorax concomitante. Nosotros presentamos aquí, un caso de quilotorax aislado, sin quilorretroperitoneo, posterior a una fusión lumbar por vía anterior, analizamos todos sus probables orígenes basándonos en la literatura y realizamos un detallado de resumen de su tratamiento y evolución.

Conceptos clave:

Chylous leakage into the retroperitoneum and associated chylothorax has been poorly described after anterior spine procedures.

Isolated chylothorax can present as a very rare perioperative complication after an anterior retroperitoneal lumbar approach.

Direct trauma to the retroperitoneal lymphatic vessels is the main cause but there are several causes described.

The diagnosis is suggestive when a milky secretion is obtained by puncture, and it is confirmed by the analytical examination of the fluid that reveals high triglycerides levels.

Most cases are solved non-operatively with oral rest and nutritional supplementation.

Resumen:

Introducción:

La fuga de quilo en el retroperitoneo es una complicación rara después de los procedimientos anteriores retroperitoneales de columna. El quilotorax es la presencia de líquido linfático en la cavidad pleural y es aún menos frecuente durante estas cirugías.

El objetivo de este trabajo es reportar el primer caso de quilotorax izquierdo aislado después de una fusión intersomática lumbar oblicua izquierda retroperitoneal en posición supina en una paciente adulta.

Caso: Paciente femenina de 30 años de edad en quien se realizó una fusión intersomática anterior L4-L5. Cuatro días después de la intervención, fue diagnosticada con un quilotorax izquierdo aislado, el cual fue drenado y tratado de forma conservadora con buenos resultados.

Conclusión: El quilotorax es una complicación extremadamente rara después de procedimientos por vía anterior de la columna lumbar. Generalmente es secundario a un quilorretroperitoneo. Presentamos un caso único de quilotorax aislado después de un abordaje lumbar retroperitoneal anterior tratado con éxito de manera conservadora.

Palabras Clave: quilotorax; columna vertebral; región lumbosacra; artrodesis

Abstract:

Introduction: Chylous leakage into the retroperitoneum is a rare complication after spinal surgery using an anterior retroperitoneal approach. Chylothorax is the presence of lymphatic fluid in the pleural cavity and it is even less frequent during these surgeries.

The aim of this work is to report the first case of isolated left chylothorax after a retroperitoneal Left Oblique Lumbar Interbody Fusion in supine position in an adult female patient.

Case: A female 30-years-old patient underwent L4-L5 anterior interbody fusion. Four days after the intervention she was diagnosed with isolated left chylothorax that was drained and treated conservatively with good outcomes.

Conclusion: Chylothorax is an extremely rare complication after anterior lumbar spine procedures, and it is usually secondary to a chyloretroperitoneum. We present a unique case of isolated chylothorax after anterior retroperitoneal lumbar approach successfully treated in a conservative manner.

Key Words: chylothorax; spine; lumbosacral region; arthrodesis.

Resumo

Introdução: O vazamento de quil no retroperitônio é uma complicação rara após os procedimentos anteriores retroperitoneal da coluna. O quilotorax é a presença de líquido linfático na cavidade pleural e é ainda menos frequente durante essas cirurgias.

O objetivo deste trabalho é relatar o primeiro caso de quilotorax esquerdo isolado após uma fusão inter-corpo lombar oblíqua esquerda retroperitoneal em decúbito dorsal em um paciente adulto.

Caso: Um paciente de 30 anos foi submetido à fusão intersomática anterior L4-L5. Quatro dias após a intervenção, ele foi diagnosticado com quilotorax esquerdo isolado, que foi drenado e tratado conservadoramente com bons resultados.

Conclusão: O quilotorax é uma complicação extremamente rara após procedimentos da coluna lombar anterior. Geralmente é secundário a um quilorretroperitônio. Apresentamos um caso único de quilotorax isolado após uma abordagem lombar retroperitoneal anterior tratada com sucesso de forma conservadora.

Palavras-chave: quilotorax; coluna vertebral; região lombossacral; artrodesis.

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Introduction

Chylous leakage into the retroperitoneum is a rare complication after spinal surgery using an anterior retroperitoneal approach. This complication has been attributed to direct injury of the retroperitoneal lymphatic trunk [1]. Only a few cases of this complication have been reported and the real incidence is unknown. Because of the rich lymphatic network that reroutes the disrupted lymphatic flow, chylous leakage usually heals spontaneously and seldom becomes clinically evident [1,2].

Chylothorax is the presence of lymphatic fluid in the pleural cavity. It is less frequent than chyloretroperitoneum after anterior spine fusions. It has been reported in cases where thoracolumbar levels were intervened and the only report in adult patients occurred as a progression of uncontained chyloretroperitoneum, due to an abnormal fistulous connection above and below the diaphragm [1,3].

The clinical importance of this process is that, without an early diagnosis, it can lead to respiratory insufficiency, nutritional and immunological dysfunction and an increase in morbidity and mortality. Considering this, after a critical review of this subject, isolated chylothorax without concomitant chyloretroperitoneum has been reported only in one pediatric patient after combined anterior and posterior thoracolumbar fusion [4].

The aim of this work is to report the first case of isolated left chylothorax after a retroperitoneal left Oblique low Lumbar Interbody Fusion (OLIF) alone in the supine position in an adult female patient.

Case Presentation

A 30-years-old female patient was admitted to the OR for a scheduled anterior L4-L5 interbody fusion due to a 3-years-long history of lumbar back pain refractory to conservative treatment.

The patient was previously healthy with no medical record referred. Preoperative imaging studies revealed an L4-L5 and L5-S1 discopathy, degenerative endplate changes were present at the 4th disc space (Figure 1).

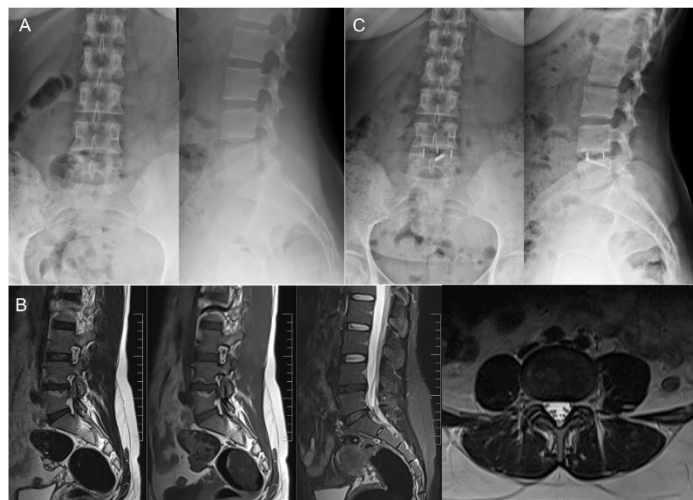


Fig. 1 Case Presentation, 30-years-old female patient. A) anterior-posterior and lateral lumbosacral preoperative X-rays showing mild narrowing of 4th and 5th disc space. B) Sagittal and axial imaging of lumbosacral magnetic resonance showing degenerative changes at the L4-L5 space and mild 5th disc degeneration, c) Anterior-posterior and lateral lumbosacral postoperative X-rays demonstrating a centered L4-L5 interbody cage, filled with autologous iliac crest bone graft.

During the intervention, the patient was positioned supine with both arms crossing over the chest held with blankets. An OLIF approach in supine position was performed by an access surgeon at the left side of the patient and the intervention was finalized after 120 minutes without any intraoperative complication.

The patient was discharged two days after the procedure with regular painkillers only. Forty-eight hours later the patient was readmitted to the emergency room complaining about left neck tumefaction and pain. No inflammatory signs were present (Figure 2).



Fig. 2 Clinical image of the 30-years-old female patient showing the supraclavicular asymmetry at the left lower part of the neck.

The patient was hemodynamically stable and without fever. The surgical wounds were in good conditions and no pain was referred at the surgical site.

An ultrasound (Figure 3) informed an increase in the adipose component of the left supraclavicular region, with a diffuse heterogeneous appearance in an area of approximately 40x37x13mm.

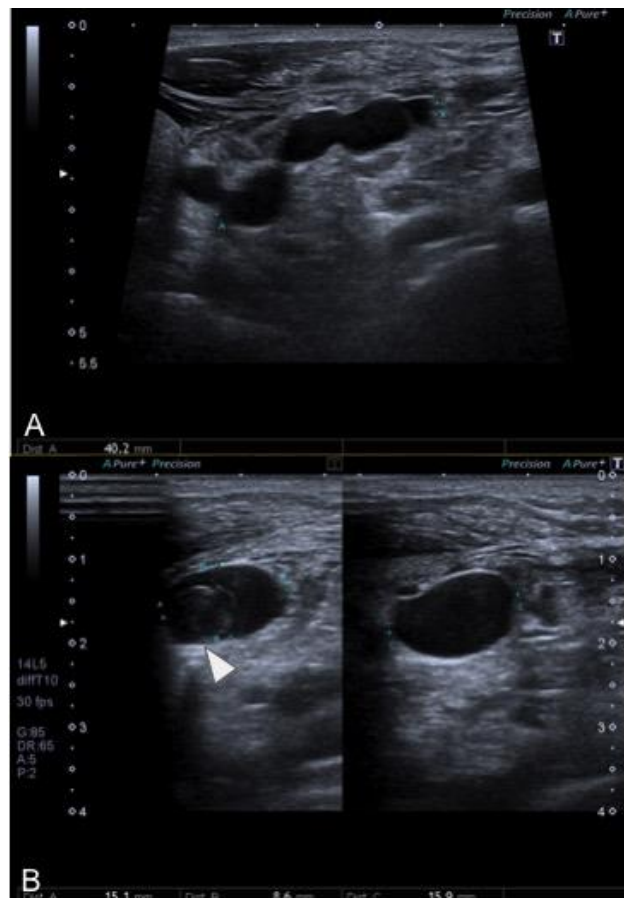


Fig. 3 30-years-old female patient, ultrasound images: A and B) Venous vascular ectasia of irregular varicose-like appearance, 40mm in length, with a 16x15x8mm saccular image that contains a thrombus in its lumen (white arrow head).

Towards the base of the neck, there was a vascular ectasia of irregular varicose-like appearance, 40 mm in length, with a 16x15x8mm saccular image that contains a thrombus in its lumen. Under vascular doppler, there was no evidence of vascularization in this vessel. Permeable left internal jugular vein and left subclavian vein were present. In order to get a better characterization of this lesion, a phlebogram with intravenous contrast was run and a deep vein thrombosis was ruled out. Abdominal CT scan showed no evidence of retroperitoneal effusion or collection. A cervical resonance (Figure 4) and lymphography evidenced a thoracic duct dilatation near the outfall of the yugulosubclavian left confluent and a laminar effusion of the left pleural space.

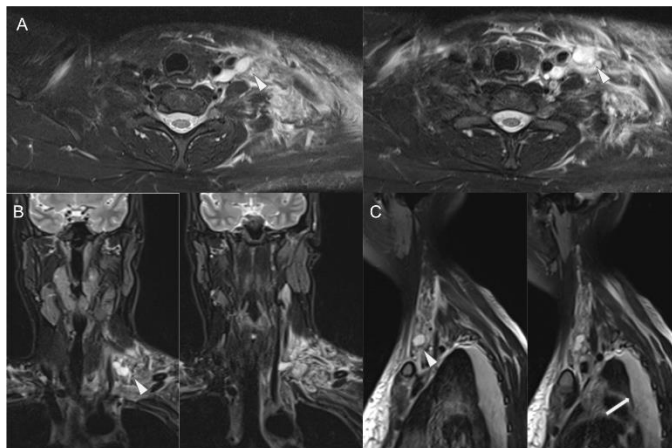


Fig.4 30-years-old female patient, cervical and upper thoracic magnetic resonance. A) T2 axial images showed a cystic dilatation of the proximal portion of the thoracic duct (white arrow head) nearby the outfall of yugulosubclavian confluent, peripheral lymphedema was evident, B and C) coronal and sagittal images revealed a cystic formation and its relationship with the vascular structures. The white arrow shows the left pleural effusion.

An ultrasound-guided pleurocentesis obtained a whitish and milky liquid. Analytics showed proteins concentration of 4.3 gr/dl, triglycerides 1643 mg/dl and 410 leukocytes/ml (60% mononuclear, 40% polymorphs). This was consistent with chylothorax.

The diagnosis of thoracic duct cyst, lymphedema and chylothorax was made. The patient was immediately restricted oral feeding and started on octreotide and parenteral feeding pump with a formula with <3% long-chain TG and supplementation of medium-chain TG, proteins and fat-soluble vitamins. Octreotide (somatostatin analog) was administered intravenously at a dose of 150 mcg every 8 hours. The usual dose is between 100 to 600 mcg per day divided into 2 to 4 doses. Treatment was maintained until oral feeding was reinstated. The proposed mechanism of action is based on the inhibitory effect of digestive secretions (gastric, pancreatic and biliary), reducing the absorption of triglycerides and fats at the intestinal level with the consequent decrease in the amount chyle volume that is drained through the thoracic duct.

As no respiratory complications were developed, no additional chyle drainage was required, either by pleurocentesis or by placing a chest tube.

The patient's clinical and nutritional status was controlled daily. By the 5th day after admission, there was a significant improvement in pain and tumefaction at the neck level. At the 14th day, a chest CT scan confirmed the resolution of chylothorax, and the patient started oral feeding with a protein-rich and low-fat diet.

The patient was discharged on the fifteenth day and controlled clinically every two months for recurrence. One year after the procedure, the patient evolved favourably resolving her low back pain without any further complication related to the thoracic duct cyst or the chylothorax conservatively treated.

Discussion

We present a case of isolated unilateral chylothorax after anterior lumbar arthrodesis, without evidence of previous chylous leakage into the retroperitoneum. To our knowledge, this is the first report of this complication with this particular presentation in an adult female patient.

The major function of the lymphatic ductal system is to transport 60-70% of ingested fat, it is the main pathway for the return of extravascular protein to the circulation and lymphocytes constitute the main cellular element of the thoracic duct lymph (between 400 and 600 cells/ml). The rate of chyle transport is dramatically affected by the amount of fat in the diet, particularly long-chain triglycerides [3]. In addition to producing mechanical compression of adjacent structures, chylous leakage can lead to fat, protein, fat-soluble vitamin and lymphocytic depletion [1]. Chylothorax is an infrequent cause of pleural effusion, unilateral in 80% of cases. It is a potentially lethal disorder that may cause respiratory, nutritional, and immunologic complications. Currently, the most frequent cause is traumatic (50%) (vertebral fractures [5], abrupt hyperextension of the spine, penetrating trauma, increased intra-abdominal pressure in closed trauma, iatrogenic injury [6,7]).

The diagnosis of chylous leakage is suggestive when a milky and opalescent secretion is obtained by puncture, and it is confirmed by analytical examination of the fluid that reveals exudative characteristics with cholesterol (CH) levels below 200 mg/dL and triglycerides (TG) levels above 110 mg/dL. TG levels lower than 50 mg/dL exclude the diagnosis, but at intermediate values, the presence of chylomicrons or cholesterol crystals should be investigated. The CH/TG ratio below 1 is also diagnostic [6]. The lymphocyte fraction should be over 80%.

Concerning spine surgeries, Bae et al. [8] reported an unusual case of bilateral chylothorax following a right-sided anterior cervical approach for a two-level cervical fusion as a consequence of direct trauma to one of the main branches of the lymphatic circulation. It was solved by bilateral chest tubes and total parenteral nutrition for two weeks.

During anterior lumbar surgeries, the cisterna chyli and its rich lymphatic tributaries surrounding the vertebral column are theoretically prone to injury. However, complications of postoperative chylous ascites remain rare [9,10]. The real incidence of chylothorax is unknown and it has only been published as case reports and small case series.

In low lumbar surgeries with the diaphragm intact, a chylothorax is, almost always, confined exclusively to the retroperitoneal cavity. When cranial levels are intervened, dissection of the diaphragm is required, which loses the ability to isolate both cavities. In these situations, it is difficult to assess if the chylothorax is due to a lesion of the cistern with retroperitoneal drainage and subsequent stiffening to the thoracic cavity, or to a primary lesion of the duct in its ascending portion.

Even though retroperitoneal chylous leakage has been reported after anterior thoracolumbar or lumbar spinal surgeries [2,3,11,12], only one case described a concomitant unilateral chylothorax as a consequence of a diaphragmatic and parietal pleural defect [1].

Mora de Sambricio et al. [4] reported a case of a 9-years-old boy who sustained an isolated chylothorax six weeks after a double approach thoracolumbar surgery due to congenital scoliosis. The fluid was drained through a chest tube and the drainage was maintained until no residual leakage was demonstrated. They do not discuss the etiopathology of the event but they consider it a consequence of direct trauma during the anterior approach.

To the best of our knowledge, isolated chylothorax after low lumbar OLIF arthrodesis has not been described previously. The etiopathology of this case has not been defined accurately, even though we resume our findings and theories.

The possibility of an unidentified thoracic duct injury and its leakage communication through the diaphragmatic hiatus exists. However, we could not observe chylorrhea intraoperatively and absence of free fluid in the abdominal CT scan made us think that the real source of the leakage came from the anatomical structures above the diaphragm.

One of our most probable theories states that the patient had an asymptomatic thoracic duct dilatation or cyst previously to the procedure and that some kind of trauma or pressure increase during the operation could end up in lymphedema and chylous leakage into the pleural cavity. This is an extremely infrequent abnormality but the cervical thoracic duct cysts have been described in the neck, in the mediastinum and even in the abdomen [13]. The usual presentation of the cyst is like a soft mass, painless and asymptomatic, in the left supraclavicular region, in adults, without sex preference. This aetiology is sustained by the imaging findings in our patient of a cyst-like formation at the left side of the neck near the yugulosubclavian confluent (Figure 4). We could not assess if this was a pre-existent pathology or if it occurred after the spine surgery.

Another possible cause, is the occlusion of the thoracic duct outfall into the yugulosubclavian confluent due to a thrombus or venous embolism. This rare event could have led to a lymphatic vessel disruption due to increased luminal pressure and consequent lymphatic oedema and chylothorax. Few cases of chylous effusion secondary to thrombosis of neck veins were reported [14]. Primary subclavian venous thrombosis

(Paget-Schroetter syndrome) is the name that has been given to this entity [15]. This syndrome is also called effort thrombosis, stress thrombosis or venous thoracic outlet syndrome because it frequently develops after a strenuous effort of the superior limbs [16]. The subclavian vein runs through a small corridor that can be narrowed by an excessive abduction of the superior limbs, excessive cervical extension, and a backward expansion or dropping of the shoulders. This could be favoured by the two hours-long cross arms position that the patient held during the operation and the initial ultrasound performed in our patient with a thombo-like intravascular image visualization (Figure 3) lead us to think towards this pathology, however this structure could not be identified during the phlebotomography.

Secondary deep venous thrombosis of the superior limbs is the most frequent cause of this obstruction but no catheterization or direct injuries to the veins were done during the procedure. What is more, venoclysis where made on the right arm of the patient, so the possibility of occlusion by a venous embolism was ruled out.

Whatever was the real aetiology, management of chylothorax is initially conservative. The main goals are the reduction of chyle formation and the prevention of concomitant nutritional and immunological problems [8]. Due to the low incidence of chylothorax, a consensus on treatment becomes difficult since the data comes from retrospective case series [17]. Chest tube utilization depends on the magnitude of pleural effusion and clinical repercussions.

Dietary treatment decreases chyle formation by avoiding lymphatic circulation with parenteral nutrition, or by enteric formulas with <3% long-chain TG and medium-chain TG supplementation, which are absorbed directly into the portal circulation without stimulating the lymphatic flow and preserving the nutritional contribution [7]. Supplementation of fat-soluble vitamins and proteins should be considered [3].

There is no consensus on the duration of dietary treatment, which should be maintained for at least 2 weeks or until the resolution of the chylothorax. Subsequently, a normal diet can be resumed.

We must assume a failure of conservative treatment and raise the need for surgical resolution in the following scenarios: 1) excessive drainage for more than 5 days, which involves > 500 ml / day in adults and > 10 ml / kg / day in children; 2) any production for more than 14 days in adults; and 3) appearance of signs of metabolic complications [2].

If medical management is insufficient, the patient should undergo imaging to identify the origin of the leakage, such as lymphography, CT lymphangiography, or magnetic resonance lymphography. The surgical options are pleurodesis, pleurectomy, ligation of the thoracic duct or its repair, lymphovenous anastomosis and pleuro-peritoneal shunt in cases of non-pulmonary reexpansion despite fluid evacuation [7].

In this case report we describe a not-previously known presentation of a very infrequent complication after anterior lumbar interbody fusion, a case of isolated unilateral chylothorax without any evidence of concomitant chyloretroperitoneum. We acknowledge that the principal limitation of this paper is that the real aetiology could not be identified. Nevertheless, after a thoughtful literature review, we present and analyze every possible cause of isolated chylothorax that could be directly related to our procedure (congenital thoracic duct cyst, yugulosubclavian confluent obstruction, left subclavian vein embolism or thrombosis).

Conclusion

Chylothorax is an extremely rare complication after anterior lumbar spine procedures. It has always been described secondary to a chyloretroperitoneum. We present a unique case of isolated unilateral chylothorax after anterior retroperitoneal lumbar approach without evidence of retroperitoneal leakage. The management of this type of chylothorax should be the same as in traumatic ones with dietary electrolytic and nutritional support, with or without chest drainage until more evidence become available.

Limitaciones de responsabilidad

La responsabilidad del trabajo es sólo de los autores

Conflictos de interés

Ninguno

Fuentes de apoyo

No hubo fuentes de financiación ni apoyo.

Originalidad del trabajo

Este artículo es original y no ha sido enviado para su publicación a otro medio de difusión científica en forma completa ni parcialmente.

Sesión de derechos

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Participación de los autores

Todos los autores han participado en la concepción del diseño, recolección de la información y elaboración del manuscrito, haciéndose públicamente responsables de su contenido y aprobando su versión final.

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