**CASE REPORT**

# Chylous fistula: management of a rare complication following right anterior cervical spine approach

Federica Novegno[1] · Pierluigi Granaroli[1] · Luigi Ciccoritti[2] · Pierpaolo Lunardi[1] · Mario Francesco Fraioli[1]

**Abstract**

**Purpose** Chylorrhea resulting from injury of the lymphatic system during neck dissection is a well-known complication. It is an uncommon occurrence in spinal surgery, and only one case after right anterior cervical spine surgery has been described so far. Despite its rarity, chylous leakage deserves a particular attention since it may become a serious and occasionally fatal complication if not detected early and managed appropriately.

**Methods** We report the case of a 42-year-old man who underwent a standard anterior cervical discectomy and fusion according to Cloward approach for a C6–C7 disk herniation. The patient developed a delayed prevertebral chyle collection on postoperative day 5, presenting with mild breathing and swallowing difficulties.

**Results** He was managed with conservative care, including bed rest, low-fat diet and drainage pouch positioning, which led to the complete resolution of the fluid collection.

**Conclusions** Knowledge of the normal anatomy of the lymphatic system and of its variations is essential when planning an anterior spinal procedure, and represents the first measure to be adopted in order to avoid such complication. The prompt identification of a postoperative chylous fistula and the applicability of an individually based management's protocol may help in the majority of the cases to reduce the potential morbidity, without significant long-term effects.

**Keywords** Cervical spine · Chyle leak · Lymphatic ducts · Discectomy · Surgical complication

## Introduction

The anterior cervical discectomy and fusion (ACDF) is one of the most common surgical procedures adopted in the treatment of cervical myeloradiculopathy [1, 2]. The most frequent complications include dysphagia, hoarseness, infections, postoperative hematoma, new neurological deficits and laryngeal injuries [2–5]. In this case report, we describe a patient with a postoperative chylous fistula following a standard right C6–C7 ACDF [6].

## Case report

A 42-year-old man was admitted with a 6-month history of left cervicobrachialgia associated with paresthesia and hypoesthesia, resembling a severe C7 radiculopathy, that had failed to improve after adequate medical therapies. The cervical magnetic resonance (MR) scans showed a prevalently left C6–C7 disk herniation and uncovertebral joint osteophyte formation.

The patient underwent a right anterior C6–C7 discectomy and fusion with titanium cage, according to a standard Cloward approach.

The immediate postoperative course was uneventful and characterized by a significant improvement in the radicular symptoms; the patient was discharged on the third postoperative day. On the fifth postoperative day, the patient came back with mild breathing and swallowing difficulties, associated with a non-tender mobile mass superior to the skin incision (Fig. 1a).

Cervical computed tomography (CT) scan and ultrasound examination confirmed the presence of a hypodense fluid

✉ Federica Novegno
federicanovegno@hotmail.it

1 Department of Neurosurgery, Tor Vergata University Medical School, University of Rome "Tor Vergata", V.le Oxford 81, 00133 Rome, Italy

2 Department of Endocrine and Metabolic Surgery, Fondazione Policlinico Universitario A. Gemelli, IRCCS, Rome, Italy

**Fig. 1** **a** Photograph of the patient showing the non-tender mobile mass superior to the skin incision observed on the 5th postoperative day, **b** photograph of a test tube containing the white, milky, creamy fluid drained for laboratory examination
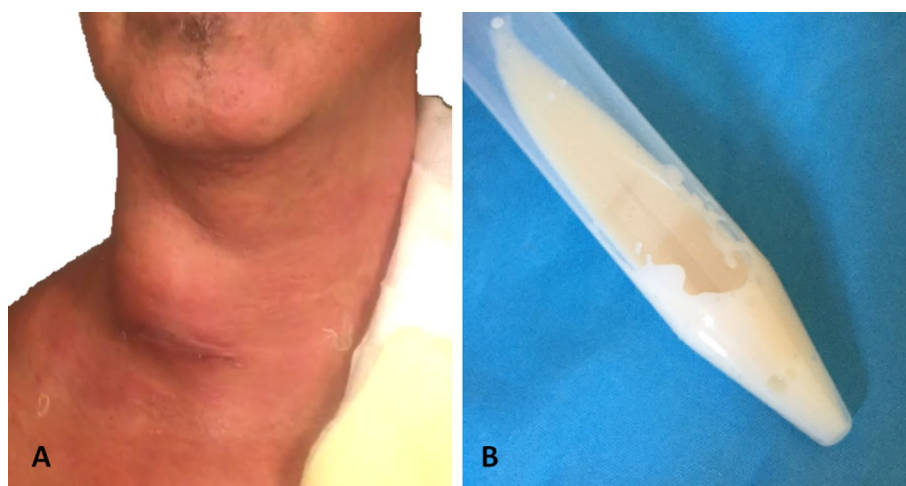
collection in the right supraclavicular region; both trachea and esophagus appeared displaced medially (Fig. 2). We drained about 60 cc of fluid with milky appearance (Fig. 1b). The laboratory analysis of the fluid showed: triglyceride (TG) level 2164 mg/dl, LDL cholesterol 25.00 mg/dl, HDL cholesterol 8.00 mg/dl, glucose 151 mg/dl, albumin 2.1 g/dl, LDH 127 UI/l, total proteins, 6.6 g/dl, lipase 1056 U/l and amylase 58 UI/l. Thus, a diagnosis of chylous fistula was made.

During the following 3 days, the patient underwent daily percutaneous drainage and local compression, with no significant improvement in drain output (about 100 cc/day). Therefore, he was put on a low-fat diet. The wound was partially reopened through the previous incision (about 5 mm), and a drainage pouch was positioned. This allowed a spontaneous constant drainage, avoiding patient discomfort, until an inversion of the pressure gradient was obtained. The drainage volume decreased progressively and, eventually, the wound healed spontaneously after 3 more days. Once

discharged, the patient underwent periodic clinical–radiological evaluations. Seriated ultrasound examinations demonstrated the progressive reabsorption of the fluid collection (Fig. 3). At 2-year follow-up, the patient presents a complete resolution of the radiculopathy and no evidence of chylous fistula recurrence.

## Discussion

The anterior approach to the cervical spine has been widely used with a low rate of major complications (9–20%). The most frequent complications include dysphagia (16.2%), hoarseness (4.8%), epidural hematoma, C5 palsy (2%), CSF leakage (1.9%), infection (0.9%) and implant extrusion (0.8%) [2, 7]. Extremely rare complications include hemothorax, carotid or vertebral artery injury, spinal cord injury, brachial plexopathy, hypoglossal nerve palsy, Horner's



**Fig. 2** Postoperative axial (**a**, **b**) computed tomography (CT) images demonstrating the presence of a right prevertebral fluid collection, with hypodense signal, extending up to the hyoid bone level (**a**) and displacing the cervical trachea and esophagus laterally to the left.

Some small air bubbles are visible in the contest of the collection. **c** Postoperative sagittal reconstruction of the cervical CT showing the proper position of the interbody cage at C6–C7 level

**Fig. 3** Axial ultrasonography images **a** depicted on the 6th postoperative day, showing the presence of a anechoic cyst partially organized (48 × 47 mm) displacing the right thyroid lobe. Doppler examination shows physiological pulsatility index of common carotid artery. Axial ultrasonography images (**b**–**d**) obtained, respectively, at the 10th, 30th and 60th postoperative days demonstrating the progressive reabsorption of the chylous collection
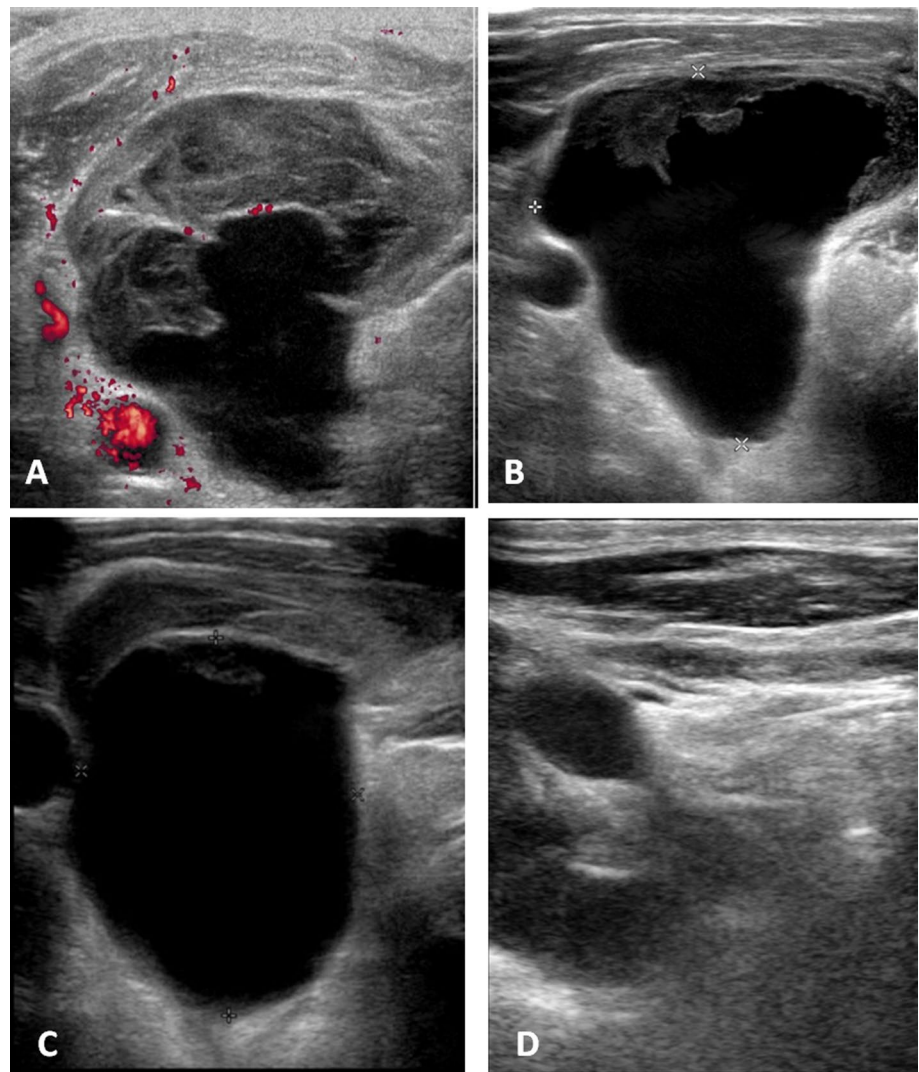
syndrome, vision loss, esophageal perforation and intraoperative death [1, 3, 4, 8–14].

Postoperative chylous fistula represents an even rarer complication in anterior approaches to the cervical spine. Indeed, its prevalence rate has been estimated at 0.02%.

It represents a well-known entity as a serious complication of head and neck surgery that occurs in 0.5–1.4% of thyroidectomies and 2–8% of radical neck dissections [16]. The majority of these cases may be encountered during surgery of the left neck (75–92%), limiting to one-fourth of the cases associated with right neck surgeries [16, 17].

Data from the literature pointed out that this kind of complication has been seldom reported during spinal surgeries, particularly in anterior thoracolumbar approaches, where the anatomical dissection to reach the column involves the biggest lymphatic collectors and the thoracic duct itself [5, 18–21]. Only five cases occurring after anterior cervical surgeries have been described in the literature so far, among which only one was a right side approach (Table 1) [15, 22–24].

Despite its rarity, chylous leakage deserves a particular attention since, without an early detection and appropriate management, it may become a serious and occasionally fatal complication. Delayed wound healing, wound breakdown with fistula formation and infection may result as a consequence of the disruption of the normal biochemical environment [16, 25]. The chyle fluid, indeed, presents erosive properties on the surrounding tissues, leading to possible injuries of the neighboring vessels. Furthermore, the pressure of a chyle collection may decrease tissue perfusion, resulting in flap necrosis [25]. Systemic problems are related to dehydration, nutritional deficiencies (depletion of electrolytes, hypoproteinemia, low blood lipid levels and deficiencies of the fat-soluble vitamins), hypovolemia, respiratory dysfunction and immunosuppression. Chyle leak is burdened by a mortality rate of 50% without supportive therapy [5].

**Table 1** Summary of the cases with chylous leakage after anterior cervical spine procedures reported in the literature

| Authors | Sex/age | Clinical history | Surgical procedure | Chyle leak complication | Treatment | FU |
|---|---|---|---|---|---|---|
| Hart et al. [22] | M/37 | C7 radiculopathy | Left C6–C7 ACDF | Intraoperative occurrence | Intraoperative closure of thoracic duct with sutures | No recurrence at 1 year |
| Warren et al. [24] | F/58 | Advanced cervical degenerative disease | Right multilevel C3–C7 ACDF (plates and screws) | 1 week PO: chyloptysis, dysphagia, respiratory distress. Huge prevertebral fluid collection | First attempt: urgent percutaneous aspiration under US and positioning of drainage. Second attempt: sclerotherapy. Third attempt: percutaneous embolization | No recurrence at 6 months |
| Derakhshan et al. [15] | M/NR | Radicular pain | Left C5–C6 ACDF | Intraoperative occurrence | Intraoperative closure of thoracic duct with clips | No recurrence at 80 days |
| | F/NR | Radiculopathy | Left C5–C6 ACDF | After 2 months fluid collection superior to the incision | Needle aspiration | No recurrence at 3.5 years |
| Mueller et al. [23] | F/59 | Bilateral cervicobrachialgia, already C4–C5 ACDF | Left C5–C6 ACDF and C7 corpectomy | On 1st PO day chyle into the drain | Low-fat diet Octreotide | No recurrence after several years |
| Present case | M/42 | Left C7 radiculopathy | Right C6–C7 ACDF | On 5th PO fluid collection superior to the incision with dysphagia and respiratory distress | Low-fat diet Drainage pouch positioned | No recurrence at 2 years |

*ACDF* anterior cervical discectomy and fusion, *NR* not reported, *PO* postoperative, *yr* year, *US* ultrasound

## Relevant anatomy and variations: surgical nuances

The lymphatic system is accessory to the circulatory system: It transports the products of fat digestion, drains excess fluids from the body back to the blood and filters them through lymph nodes [21, 26]. The thoracic duct drains three quarters of the lymph into the venous blood stream, including that originating from the lower body and the left head and arm. It originates from the cisterna chyli, usually located on the anterior surface of the first and second lumbar vertebral body [22]. Ascending to the thorax, at T7 it deviates obliquely behind the esophagus and crosses the midline to the left at the level of T5–T6 vertebrae. It then passes behind the aorta and to the left of the esophagus, emptying into the confluence of the left internal jugular and subclavian veins, about 2–3 cm above the clavicle [26, 27]. The remaining third of the lymph, originating from the right thorax, arm, neck and head, flows into the adult right lymphatic duct. This duct is 1–2 cm long, closely related to the anterior scalene muscle, and usually empties into a corresponding location on the right, at the level of the junction between the right subclavian and internal jugular veins [27, 28].

There is a wide variation in lymphatic system anatomy due to deviations in normal embryologic development; if present, developmental anomalies of the lymphatic system can provide surgical challenges [28]. Several anatomical variants have been described so far: a complete left-sided thoracic duct, a complete right-sided thoracic duct, a proximal and distal partial duplication of the thoracic duct, a plexiform variation of the thoracic duct or absence of the cisterna chyli [27]. The greatest variation in thoracic duct anatomy is seen in proximity to its termination, with multiple branching in about 40% of cases, thus explaining why the most common site of injury to the ducts during neck dissection is lateral to the inferior portion of the carotid sheath on either side [28]. In 5% of cases, the main thoracic duct may terminate on the right side of the neck. This is the reason that although most injuries of the thoracic duct are in the left cervical area, a smaller percentage of injuries may occur in the right side of the neck [17].

The right lymphatic duct may present likewise anatomical variations (1–5%). Its most frequent origin is from the right jugular, bronchomediastinal and subclavian lymphatic trunks, although these vessels may terminate individually so that the main duct results absent [28].

## Diagnosis

The diagnosis of chylous fistula can be made intraoperatively or postoperatively [16, 29]. In general, some authors recommend a careful inspection of the surgical field after a head and neck procedure, especially in those cases involving the dissection low in the neck; the occurrence of creamy or milky fluid is highly suspicious for a lymphatic system trauma. Intraoperative diagnosis may be challenging because patients are usually in a fasting state before surgery, slowing down significant lymph production [15]. Thus, maneuvers that increase intrathoracic and/or intraabdominal pressure may improve the visibility of the chyle leakage. However, the presence of variant terminations may compromise the efficacy of the ligation resulting in persistent chylous fistula [16].

The majority of cases (about 86%) are diagnosed during the first three postoperative days, owing to the resumption of foods that contain fat, which induce an increment in chyle production and flow [16, 17]. The patient may exhibit a sudden increase in drain output, with a typical milky appearance, and volumes between 300 and 500 ml/day [25]. Nevertheless, low-volume leakages may be underestimated because it was mixed with blood and mistaken as purulent secretions. On inspection, the skin may present a bulge at the level of the supraclavicular fossa, associated with induration, edema, erythema [16, 25].

The drain fluid can be analyzed: The fat content of chyle ranges from 0.4 to 4%, and the main lipid component is triglyceride (greater than 100–200 mg/dl) [16, 17]. Other biochemical elements include percentage of chylomicrons greater than 4% and total protein level greater than 3 g/dl [17]. Leukocytes normally range from 1000 to 20,000/mm$^3$ in thoracic duct lymph [25].

Lymphoscintigraphy with technetium 99 has been proposed in unclear cases with low-volume leakages; data from the literature do not report a consensus on its actual efficacy, and the majority of authors do not utilize this technique routinely [17, 29].

## Treatment

The chylous leakage should be repaired immediately once identified intraoperatively. The thoracic duct may be ligated using either non-absorbable 3-0 or 4-0 sutures or surgical clips [16, 29]. Locoregional myofascial flap may be added to perform a coverage of the area, using the clavicular head of the sternocleidomastoid or the pectoralis major [16, 30]. In addition, the region can be sealed with fibrin, polyglactin or collagen [29].

Since 30–80% of patients treated with conservative therapies healed definitively, the general attitude in these cases is to maintain a non-surgical approach, especially when

dealing with low-volume output fistulae [16, 17]. Thus, the first step is to evaluate the amount of drain output per day: It can be categorized as low output (less than 500 ml/day) or high output (more than 500 ml/day) [16].

## Non-surgical management

As a general disposition, the patient should take bed rest, with elevation of the head (30°–40°), in order to reduce chyle flow; stool softeners can be employed to lessen intraabdominal pressure [16].

Dietary modifications play a crucial role in non-surgical approach, aiming at both decreasing chyle production and flow and replenishing fluid and electrolytes. Diets can include nonfat, low-fat or medium-chain fatty acids (for low output) [16, 30]. Octreotide therapy may represent another step of conservative management: It acts directly on vascular somatostatin receptors to minimize the chyle production [16, 30]. Several authors are reporting encouraging results on its efficacy, but no consensus guidelines are available yet: Its dosage ranges in different reports from 100 to 200 mcg, two or three times per day, with a duration variable between 3 and 24 days [16, 31].

A pressure dressing is generally not recommended because the supraclavicular region has no solid basis to support it and there is a potential risk of damaging the skin flap perfusion [16]. Suction drainage is generally employed to evacuate the chyle collection while monitoring drain output. Negative wound pressure medications have also been used in some cases. However, the application of a permanent negative pressure to the entire wound bed may prevent the closure of the chyle fistula, with significant collateral risks, namely major bleeding and infections [16, 32].

## Invasive procedures

Surgical re-exploration is generally recommended once conservative approach fails. In particular, surgery should be performed in high output volumes (> 1000 ml/day) lasting for more than 5 days, when low output leakage continues after more than 10 days, or when there are serious complications [29, 30]. The main goal is to identify the leakage site and isolate the lymphatic duct to perform a definitive ligation; however, given the poor state of tissues, this may result extremely difficult even for expert surgeons. Hence, some authors recommend to apply automatically fibrin sealant, followed by a layer of muscle and fascia [29].

Further options of treatment include minimally invasive techniques, often employed as second-line procedures, namely thoracic duct embolization and therapeutic lymphangiography [30]. Despite its promising results, these techniques are both time-consuming and technically challenging, requiring often several attempts [24, 30]. In patients who underwent failed

surgery, ligation of the thoracic duct by means of a thoraco-scopic approach can be an effective salvage procedure, avoiding the significant morbidity of major thoracic access [16, 30].

## Chylous fistula after anterior cervical spine surgery: lesson learned

According to the literature review, there are only five cases describing lymphatic duct injuries during cervical spine surgery reported so far (Table 1) [15, 22–24]. All but one was left-approached surgical procedures; the only right-approached case was a right multilevel C3–C7 ACDF [24], which required an extensive neck exposure. Indeed, despite being more easily comprehensible, the left laterality cannot alone explain the occurrence of such a complication in cervical spine surgery. The possible anomalous locations or anatomical variations of the terminal branches of the thoracic duct may have contributed to the inadvertent injury, most likely in our case, which was a standard single-level C6–C7 ACDF, requiring minimal neck dissection [22, 24]. In particular, we hypothesized that a damage of the right cervical duct may have resulted in either excessive traction or improper coagulation of redundant fat in proximity of the esophagus during the soft tissues dissection. Fortunately, in our case the leakage resolved in a relatively short time with conservative treatments, so that we excluded a major laceration of the main thoracic duct (even suspecting its anomalous right side termination) not requiring further invasive diagnostic procedures.

In order to avoid the lymphatic system's injury, a sound understanding of its anatomy and an intraoperative awareness of the potential interpatient variations are, therefore, advisable (Fig. 4) [15]. With concern to anterior cervical approaches, this knowledge is important for spinal surgeons, since preoperative radiographic methods (namely sonography, CT, MR lymphography and MRI) are considered neither determinant nor useful in clinical practice [18].

In two cases, the chyle leakage occurred intraoperatively and required an immediate surgical repair [15, 22]. In these two cases, the fistula presented with a mild amount [15, 23] and, as in our case, a conservative approach resulted effective. The last case required several procedures, including positioning of a drainage, catheter-based sclerotherapy treatments and percutaneous thoracic duct embolization [24]. In conclusion, all the patients had a good prognosis, with no recurrences at the last follow-up.

## Conclusions

Complications involving the lymphatic system are rarely encountered in anterior cervical spine surgery; however, a prompt identification and use of an individually based
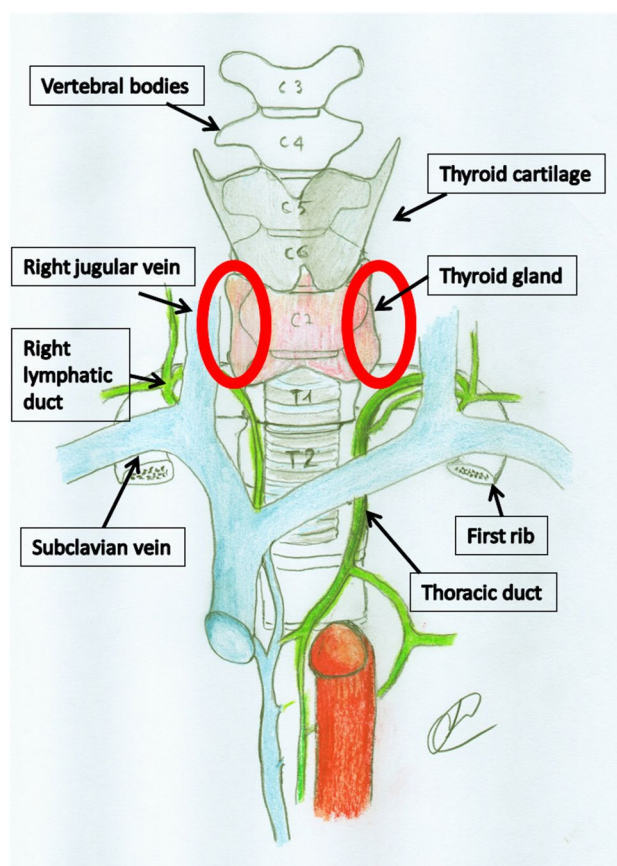


**Fig. 4** Schematic drawing showing the relevant anatomy of the neck. The ellipses indicate the likely region of most lymphatic system injuries on both sides, given the anatomical variability of the ducts pathways in the cervical area described in the text

protocol of management may help reduce potential morbidity without significant long-term effects, achieving the complete resolution of symptoms [23].

## Compliance with ethical standards

## References

1. Harhangi BS, Menovsky T, Wurzer HA (2005) Hemothorax as a complication after anterior cervical discectomy: case report. Neurosurgery 56(4):E871
2. Wang T, Tian XM, Liu SK, Wang H, Zhang YZ, Ding WY (2017) Prevalence of complications after surgery in treatment for cervical compressive myelopathy: a meta-analysis for last decade. Medicine (Baltimore) 96(12):e6421

3. Ames CP, Clark AJ, Kanter AS et al (2017) Hypoglossal nerve palsy after cervical spine surgery. Glob Spine J 7(1 Suppl):37S–39S

4. Daniels AH, Hart RA, Hilibrand AS et al (2017) Iatrogenic spinal cord injury resulting from cervical spine surgery. Glob Spine J 7(1 Suppl):84S–90S

5. Denaro L, Longo UG, Papalia R, Denaro V (2010) Complications related to antero-lateral approaches. In: Denaro L, D'Avella D, Denaro V (eds) Pitfalls in cervical spine surgery. Springer, Berlin, pp 91–108

6. Cloward RB (1958) The anterior approach for removal of ruptured cervical disks. J Neurosurg 15(6):602–617

7. Smith GA, Pace J, Corriveau M et al (2017) Incidence and outcomes of acute implant extrusion following anterior cervical spine surgery. Glob Spine J 7(1 Suppl):40S–45S

8. Gabel BC, Lam A, Chapman JR et al (2017) Perioperative vision loss in cervical spinal surgery. Glob Spine J 7(1 Suppl):91S–95S

9. Härtl R, Alimi M, Abdelatif Boukebir M et al (2017) Carotid artery injury in anterior cervical spine surgery: multicenter cohort study and literature review. Glob Spine J 7(1 Suppl):71S–75S

10. Hilibrand AS, Nassr A, Arnold PM et al (2017) Epidemiology and outcomes of vertebral artery injury in 16 582 cervical spine surgery patients: an AOSpine North America Multicenter Study. Glob Spine J 7(1 Suppl):21S–27S

11. Hsu WK, Kannan A, Mai HT et al (2012) Chyloretroperitoneum following anterior spinal surgery. J Neurosurg Spine 17(5):415–421

12. Than KD, Mummaneni PV, Smith ZA et al (2017) Brachial plexopathy after cervical spine surgery. Glob Spine J 7(1 Suppl):17S–20S

13. Traynelis VC, Malone HR, Smith ZA et al (2017) Rare complications of cervical spine surgery: Horner's syndrome. Glob Spine J 7(1 Suppl):103S–108S

14. Wang JC, Buser Z, Fish DE et al (2017) Intraoperative death during cervical spinal surgery: a retrospective multicenter study. Glob Spine J 7(1 Suppl):127S–131S

15. Derakhshan A, Lubelski D, Steinmetz MP et al (2017) Thoracic duct injury following cervical spine surgery: a multicenter retrospective review. Glob Spine J 7(1 Suppl):115S–119S

16. Delaney SW, Shi H, Shokrani A, Sinha UK (2017) Management of chyle leak after head and neck surgery: review of current treatment strategies. Int J Otolaryngol. https://doi.org/10.1155/2017/8362874

17. Priego Jiménez P, Collado Guirao MV et al (2008) Chyle fistula in right cervical area after thyroid surgery. Clin Transl Oncol 10(9):593–596

18. Akcali O, Kiray A, Ergur I, Tetik S, Alici E (2006) Thoracic duct variations may complicate the anterior spine procedures. Eur Spine J 15(9):1347–1351

19. Bhat AL, Lowery GL (1997) Chylous injury following anterior spinal surgery: case reports. Eur Spine J 6(4):270–272

20. Su IC, Chen CM (2007) Spontaneous healing of retroperitoneal chylous leakage following anterior lumbar spinal surgery: a case report and literature review. Eur Spine J 16(Suppl 3):332–337

21. Upadhyaya CD, Park P, La Marca F (2007) Chyloretroperitoneum following anterior spinal deformity correction. Case report. J Neurosurg Spine 7(5):562–565

22. Hart AK, Greinwald JH Jr, Shaffrey CI, Postma GN (1998) Thoracic duct injury during anterior cervical discectomy: a rare complication. Case report. J Neurosurg 88(1):151–154

23. Mueller K, Syed HR, Rhee JW, Nair MN (2017) Delayed chyle leak following anterior cervical spinal surgery: a case report and management algorithm. Cureus 9(5):e1231. https://doi.org/10.7759/cureus.1231

24. Warren PS, Hogan MJ, Shiels WE (2013) Percutaneous transcervical thoracic duct embolization for treatment of a cervical lymphocele following anterior spinal fusion: a case report. J Vasc Interv Radiol 24(12):1901–1905

25. Erisen L, Coskun H, Basut O (2002) Objective and early diagnosis of chylous fistula in the postoperative period. Otolaryngol Head Neck Surg 126(2):172–175

26. Phang K, Bowman M, Phillips A, Windsor J (2014) Review of thoracic duct anatomical variations and clinical implications. Clin Anat 27(4):637–644

27. Johnson OW, Chick JF, Chauhan NR et al (2016) The thoracic duct: clinical importance, anatomic variation, imaging, and embolization. Eur Radiol 26(8):2482–2493

28. Smith ME, Riffat F, Jani P (2013) The surgical anatomy and clinical relevance of the neglected right lymphatic duct: review. J Laryngol Otol 127(2):128–133

29. Santaolalla F, Anta JA, Zabala A et al (2010) Management of chylous fistula as a complication of neck dissection: a 10-year retrospective review. Eur J Cancer Care (Engl) 19(4):510–515

30. Campisi CC, Boccardo F, Piazza C, Campisi C (2013) Evolution of chylous fistula management after neck dissection. Curr Opin Otolaryngol Head Neck Surg 21(2):150–156

31. Prabhu S, Thomas S (2015) Octreotide for conservative management of intractable high output post operative chylous fistula: a case report. J Maxillofac Oral Surg 14(Suppl 1):21–24

32. Kadota H, Kakiuchi Y, Yoshida T (2012) Management of chylous fistula after neck dissection using negative-pressure wound therapy: a preliminary report. Laryngoscope 122(5):997–999