Prone position thoracoscopic management of neck chyle leak following major head and neck surgery.

A case series

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Chyle leak is a major compication following head and neck surgery, with reported incidence of 0.5% up to 8.3% in published literature. Cervical chyle leak may be challenging to manage with significant morbidity, resulting from extensive fluid and nutritional losses.
This manuscript presents four cases of cervical chyle leak after head and neck surgery. Cervical thoracic duct injury had been identified intra-operatively. Conservative treatment failed to reduce chylous output post-operatively.
All patients were offered thoracocscopic thoracic duct ligation in prone position; thoracic duct was dissected above the right diaphragm and ligated. Immediate resolution of their symptoms followed, with no recurrence at the follow-up period.
Intra-operative repair of cervical thoracic duct remains controversial, while when identified early reduces the following comorbidities. Conservative management addresses reduction of chylous output, while amplifying hydration and alimentation. Thoracoscopic thoracic duct ligation offers a safe and feasible treatment for cervival chyle leak following head and neck surgery with all the advances of minimally-invasive surgery.

KEY WORDS: Chyle leak, Head and Neck Surgery, Thoracoscopic, Thorasic Duct, Ligation, Minimally-Invasive

Introduction

Operations of the inferior neck may lead to injury of delicate lymphatic structures, such as the cervical Thoracic Duct. The first incidences of chyle leak following neck surgery were reported by Cheever in 1875 and Allen and Briggs in 1901 [1].
Cervical portion of the thoracic duct is terminated at the confluence of the left subclavian and internal jugular veins, closely adjacent to anatomical landmarks of head and neck surgery, thus being susceptible to iatrogenic injury and resulting to chyle leak during neck dissection and thyroid surgery. Left sided neck surgery, radical or modified neck dissection, as well as thyroid cancer and cervico-mediastinal goitre surgery, are procedures that carry an increased risk of thoracic duct injury [3] (Fig. 1).
The incidence of cervical chyle leak after thyroid surgery and neck dissection varies greatly, reported between 0.5% for thyroid surgery and up to 8.3% for neck dissection, depending mainly on the extent of [3].
Cervical chyle leak may be challenging to manage with significant morbidity, resulting from extensive fluid and nutritional losses, as well as delayed wound healing at the surgical-site [4].
We present four cases of cervical chyle leak treated with Thoracoscopic Thoracic Duct ligation, following head and neck surgery, after failure of conservative treatment.

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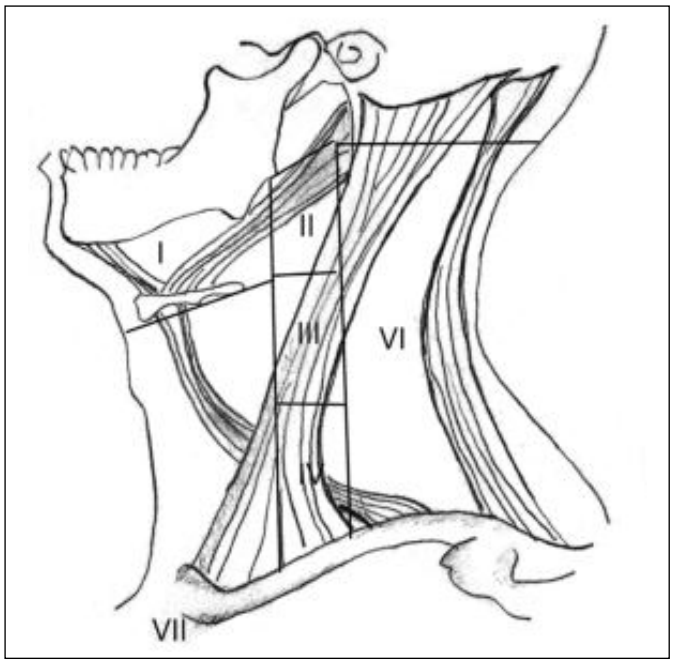

Fig. 1: Cervical lymph-node levels.

Cases

Case 1

A 73-year old male was referred to the Otolaryngology clinic for investigation of a several month history of multiple left sided neck swellings. Examination revealed a left submandibular and multiple left level II swellings. He underwent left tongue base biopsy and modified radical left sided neck dissection (levels I – IV). Intra-operative chyle leak was oversewn with non-absorbable Prolene interrupted sutures and Surgicel and checked with Valsalva manoeuvre. A Jackson Pratt drain was placed alongside the neck incision.
Over the next six days, he had chylous leakage averaging 2300mL; subcutaneous Octreotide (100μg, thrice daily) and enteral medium chain triglyceride (MCT) diet post-operatively were admitted. Given the high output leak and lack of response to Octreotide, thoracoscopic thoracic duct ligation was performed six days postoperatively. The thoracic duct was identified and ligated in the middle third of the chest using a three port approach. Drain output ceased immediately. The patient has made a good recovery with no recurrence of the chyle leak.

Case 2

A 47-year old male presented with unilateral left tonsillar swelling; he was diagnosed with squamous cell carcinoma. He was re-admitted for wide local excision of the left tonsillar bed with a left neck dissection (Levels II – IV). Intra-operative leak from the thoracic duct was identified and controlled using electrocautery, Surgicel appliance and further reinforcement with Prolene interrupted sutures. A Jackson Pratt drain was placed alongside the neck incision. The repair was inspected with Valsalva manoeuvre. A low-fat diet was applied for two days post-operatively (POD). Upon resuming normal diet, there was a rapid increase in daily drain output to an average of n=140mL. MCT diet and subcutaneous Octreotide (100μg, thrice daily) were administrated. Patient presented progressively worsening left-sided neck swelling from the 4th POD and the next eight days.
Patient was treated with Thoracoscopic Thoracic Duct Ligation 13 days later. Neck wound was opened to evacuate the collection but the wound bed was not re-explored for the site of the chyle leak. The operation resulted in immediate cessation of chylous output with normal diet started on the following day. No recurrence of cervical chyle leak ocurred.

Case 3

A 56-year old male ex referred to the Otolaryngology clinic with a right sided laryngeal squamous cell carcinoma. The patient was treated with Total Laryngectomy and Bilateral Neck Dissection (Levels II – IV). Intra-operative leak was identified at the cervical thoracic duct; it was repaired with Prolene interrupted sutures and local appliance of fibrin sealant (Tisseel). The repair was inspected with Valsalva manoeuvre. A Jackson Pratt drain was placed alongside the neck incision.
Despite a post-operative regimen of enteral MCT feed and subcutaneous Octreotide (100μg, thrice daily), the patient had increasing chylous output from his neck drain to a peak of 1100mL (average=500mL). A thoracoscopic thoracic duct ligation was performed on the 8th POD, resulting in immediate cessation of chyle leak. Wound healing was impaired greatly however and the patient continued to have bloody output from the neck drains. Further five days later, the patient required exploration of the neck; ligation of the right internal jugular vein and reconstruction of the anterior pharyngeal wall with a pedicled pectoralis major flap was performed. He was then discharged, with successful healing of the pharynx and no further issues with chyle leak after the thoracoscopic repair.

Case 4

A 72-year old male, with prior medical history of a T2N0M0 squamous cell carcinoma of the right glottis that was treated previously with radiotherapy and debulking, was presented to the Otolaryngology service with rapidly progressive shortness of breath, hoarseness of voice and weight loss. His symptoms were caused by recurrent disease that was treated with exploration and

Table 1 - *Patients demographics and clinical features*

Patients	Age (Years)	Gender (Male/ Female)	Previous Operation	Intra-Operative Repair	Averagechyle Outpout (Ml)	Conservative Management	Days to final treatment	Surgical Treatment	Recurrence
Patient #1	73	Male	Radical left side neck dissection	Oversewn Non-Absorb. Suture, Surgicell, Valsalva Manoeuver	2300	Octreotide 100 MG S:1X3, MCT diet	6	Thoracoscopic Thorasic Duct Ligation	No
Patient #2	47	Male	Local excision of left tonsilar bed + left side neck dissection	Electrocautery, oversewn non-absorb. Suture, surgicell	140	Octreotide 100 Mg S:1x3, MCT diet	13	Thoracoscopic Thorasic Duct Ligation	No
Patient #3	56	Male	Total Laryngectomy + Bilateral Neck Dissection	Oversewn Non-Absorb. Suture, Fibrin Sealant (Tisseel)	500	Octreotide 100 Mg S:1x3, MCT diet	8	Thoracoscopic Thorasic Duct Ligation	No
Patient #4	72	Male	Total Laryngectomy +Resection OF THE Posterior Pharungeal Wall + Bilateral Neck Dissection	Oversewn Non-Absorb. Suture	100	Octreotide 200 Mg S:1x3, MCT diet	11	Thoracoscopic Thorasic Duct Ligation	No

Non-absorb. Suture= prolene, mct diet= enteral medium chain triglyceride diet

tumor debulking. Several days later, the patient was readmitted with stridor, requiring an emergency tracheostomy. His lesion was found to have progressed to a right transglottic tumour with involvement of the posterior pharyngeal wall (T4aN0M0).

Total Laryngectomy with resection of the posterior pharyngeal wall and Bilateral Neck Dissection (Levels II – IV) was performed. The patient also required reconstruction of the subsequent pharyngeal defect with a tubed free radial forearm flap. Chyle leak was identified in the left neck intra-operatively and controlled with non-absorbable Prolene interrupted sutures and inspected with Valsalva manoeuvre. A Jackson Pratt drain was placed alongside the neck incision. The following day, the patient needed evacuation of a cervical haematoma, due to arterio-venous compromise of the free flap. Subcutaneous Octreotide (200µg, thrice daily) and low-fat MCT diet were both administrated; the patient continued to have chylous output averaging over 100mL for the next eleven days. Despite the low output fistula, Thoracoscopic Thoracic Duct ligation was performed, to prevent both further compromise of the neo-pharyngeal reconstruction and to avoid wound healing complications. Normal diet was resumed the next diet with no recurrent chyle leak.

All patients' demographics and clinical features are presented in Table I.

Discussion

Surgical Anatomy: Thoracic Duct

During embryological development, the thoracic duct forms from partial fusion of the separate right and left lymphatic ducts with numerous anastomoses persisting after birth [5]. The thoracic duct is the major conduit for lymph to recirculate into the systemic circulation from the entire left side of the body and sub-diaphragmatic right body. The remainder from the right head and neck, right upper limb and thorax are draining to the right lymphatic duct [6]. At the level of the second thoracic vertebra, dilatations of the lymphatic system known as cisterna chyli, lead to the thoracic duct, which ascends cephalad through the aortic hiatus into the posterior mediastinum. In the lower third of the chest, the duct is most consistent in its anatomy lying to the right of the desceding thoracic aorta, positioned between the vertebral column and oesophagus [4,7,8]. As the thoracic duct courses further cephalad, it lies to the right of the vertebral column before passing under and to the left of the oesophagus at the level of the fifth thoracic vertebra coursing towards its termination at the confluence of the left subclavian and internal jugular veins [2] (Fig. 2). In the neck, the thoracic duct receives further jugular, subclavian and bronchomediastinal lymphatic trunks. The supe-

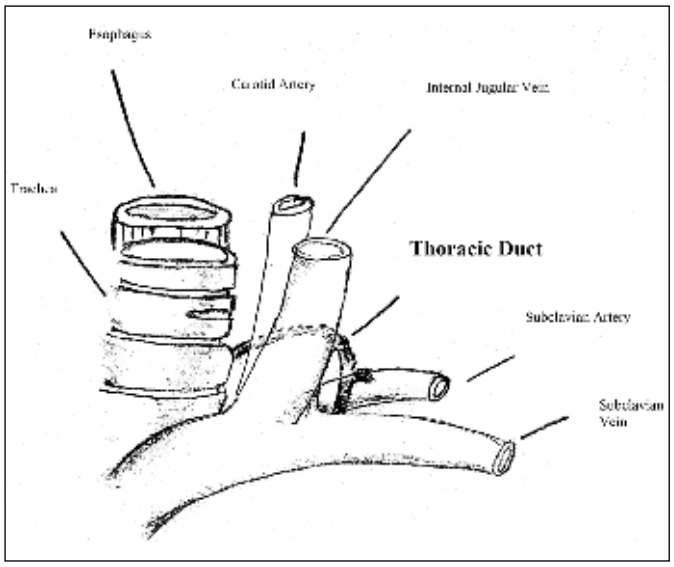

Fig. 2: Cervical thoracic duct and anatomical landmarks.

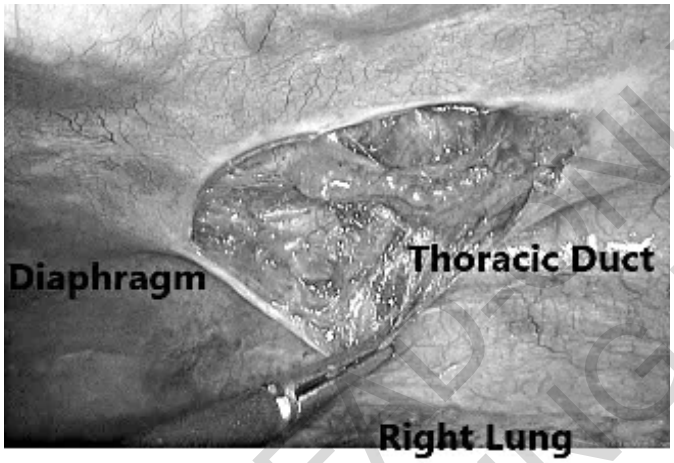

Fig. 3: Dissection of thoracic duct, above the level of the right diaphragm.

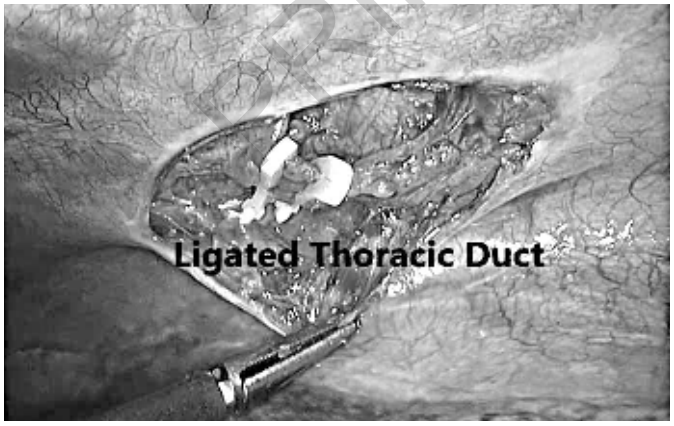

Fig. 4: Thoracic duct ligation.

rior portion of the duct including its cervical termination shows a reported variation of up to 45% in the general population [8]. Common variants include duct duplication with different termination points. Rarely, the thoracic duct may drain into the right side of the neck [5,9] .The thoracic duct is maximal in diameter at its cervical termination measuring up to 10mm but increased intra-thoracic and intra-abdominal pressure will distend the duct further.

Pathophysiology

Chyle is a milky, opalescent fluid that consists of a mix of electrolyes in a similar balance to plasma, immunoglobulins, lymphocytes and chylomicrons. Chylomicrons contain esterified monoglycerides and fatty acids combined with cholesterol and proteins from the breakdown of long-chain fatty acids in the proximal small bowel by bile salt action. Absorption from the intestine to the lymphatic system is through intestinal lacteals. By contrast, short and medium chain fatty acids are absorbed through the intestinal mucosa directly into the hepatic portal venous circulation and thereby bypass the thoracic duct [4]. The volume of chyle produced is highly variable ranging from 10-100mL/kg body weight daily and is dependent on numerous factors including physical activity, diet and medications use [8].

Diagnosis and Investigation of Cervical Chyle Leak

Cervical chyle leak following head and neck surgery may be identified intra-operatively or post-operatively. Chyle leaks can significantly impact wound healing and cause hypovolemia, malnutrition, electrolyte disturbances, and immunosuppression. Cervical chyle leaks are space-filling and exert pressure on surrounding tissues, creating symptoms that can range from minimal discomfort to life-threatening situations. Due to the potential significant morbidity, chyle leaks identified intra-operatively should be treated immediately.

Intra-operatively, cervical Thoracic Duct damage may cause leakage of chyle (milky coloured fluid). However, due to pre-operative fasting, the consistency of the chyle may be more transparent and less fatty than normally. In case of uncertainty, manual application of pressure to the abdomen [10], Valsalva manoeuvre or Tredelemburg positioning may be performed to increase intra-thoracic pressure [11] and thus further compress the thoracic duct and increase chyle leakage.

If chyle leak is suspected several days after the initial operation, drain fluid or aspirated fluid should be sent for microbiological analysis, as purulent neck collections may have a similar appearance. The fluid can also be examined for presence of chylomicrons and triglycerides.

This can be taken as a standalone reading or compared as a ratio with serum levels. The presence of chylomicrons, triglyceride levels above 100 mg/dL or a fluidserum triglyceride ratio >1, are diagnostic of chylous fistula [12-15]. Furthermore, chylous outputshould be monitored daily.
Following that, various imaging modalities are available in the diagnostic course. Cervical ultrasonography (US), Computed Tomography (CT) scan or Magnetic-Resonance Imaging (MRI) scans 16 can be utilised to identify and possible draining of lymphatic leakage. Furthermore, Bipedal lymphangiography can both be used in diagnosis and therapy to close the leakage [17]. Lymphangiography with thoracic duct embolization is also a well-established technique of refractory thoracic lymphatic leaks for diagnosis and treatment [18]. However, several complications of lymphangiography have been described (tissue necrosis, fat embolism and hypersensitivity) [17], limiting the possible use of this modality.

Treatment options

Intra-Operative Management

Cervical Thoracic duct is prone to injury during thyroid surgery and neck dissection surgery. Purposeful identification and dissection of the cervical portion of the thoracic duct must be avoided, as the thin-walled structure is prone to inadvertent injury [19]. As described earlier, the anatomy of the thoracic duct particularly in its cervical portion can vary significantly. Some authors advocate over-sewing the tissues in the affected area with a non absorbable suture. The topical use of fibrin glue and cyanoacrylate over visible leaks has also been described as a further method to help seal the duct [20-22].
There are several reports of the use of topical sclerosants after neck dissection to increase scarring in the wound bed and to help prevent chylous fistula [4]. Agents such as tetracyclines, cyanoacrylate and doxycycline have been used with varying degrees of success [20,23]. We counsel against the use of these sclerosants as re-exploration of the neck will be more difficult. In addition, there is a report of significant neurotoxicity from the use of sclerotherapy with doxycycline, resulting in unilateral phrenic nerve temporary palsy [24-25].
Where the sternocleidomastoid muscle has not been resected for oncological purposes, one of the heads of the muscle can be sutured onto the wound bed. The clavicular head is usually utilised but the sternal head may be used if it has already been intentionally divided as part of the procedure for total laryngectomy [4,26].
Other loco-regional flaps options include use of the Scalenus Anterior muscle which carries a high risk of damage to the underlying brachial plexus roots [21]. A delayed Pectoralis Major flap has also been described to cover the wound bed [4]. These options aid wound healing and cover the site of the leakage but as they do not definitively address the problem, they carry an inherent risk of failure in addition to the specific morbidity from these procedures. Intra-operative thoracic duct injury, despite successful surgical repair, carries a 27% chance of chyle leak post-operatively [26]. Percentage varies from 25-75% [27,28].

Conservative management

Chyle flow is modulated by a number of factors such as physical activity, diet, changes in respiration and intra-thoracic and intra-abdominal pressure. Conservative measures, therefore, aim to address these factors to minimise chylous output. In the immediate post-operative period, minimising strenuous activity is important. This includes the addition of stool softeners to help prevent straining that would increase intra-thoracic and intra-abdominal pressure [5].
Dietetic input is essential for the safe management of these patients not only to address the large volume and fat losses but to replace trace elements, vitamins and electrolyes [5]. The elimination of long chain fatty acids from the diet is essential as their breakdown products are absorbed into the lymphatics as chylomicrons [2].
Enteral feeding regimens typically use water-soluble short and medium chain fatty acids with replacement of fat-soluble vitamins, minerals and trace elements. These work on the principle that they are not absorbed by enteral lymphatics but directly into the hepatic portal venous circulation thereby decreasing chyle flow [5]. Alternatively, total parenteral nutrition has been shown to be a useful adjunct in management. However, this must be balanced with the potential side effects and complications that it carries such as systemic infectionandvillous atrophy [29,30].
Another major key in cervical chyle leakage, is the decrease of chylous output. Lipid absorption and breakdown can be reduced by the use of Orlistat, a pancreatic lipase inhibitor but this does not seem to be in widespread use to manage chylous leakage [5]. Somatostatin, a neuro-endocrine hormone, with well established effects on the digestive and lymphatic systems; Ulíbarri JI et al [31] were the first to describe the use of somatostatin for the treatment of chyle leakage from thoracic duct injury during a supraglottic laryngectomy in 1990s.Somatostatin decreases chyle production; it reduces the secretion of gastric, pancreatic, and intestinal fluids [32]. Somatostatin has a major drawback in trerapeutic algorithm as it has short half-life that requires continuous intravenous infusion. This problem has been sorterd out with the has been obviated by the introduction of its synthetic analogue, Octreotide, which is administrated subcutaneously. Octreotide has gained considerable popularity in the management of CL, first in thoracic surgery and more recently with head and neck surgery [13].

Subcutaneous Octreotide is now more commonly used in the management of chyle leak but there is great variance in the dosage amongst reported cases ranging from 100micrograms twice daily to 200micrograms subcutaneous thrice daily [33]. Again, there is no evidence base for duration of treatment which can vary from several days, for low output chyle leaks, to several weeks for chronic higher output leaks [2,5]. Patients undergoing conservative management received on average two additional days of Octreotide after cessation of their leaks [5]. If there is no change in chylous output in five days, there is consensus to treat the chyle leak with more invasive measures such as surgical exploration [4]..In our case series, Octreotide administration was terminated on the day that definitive ligation of the thoracic duct was performed.

Surgical Management

The gold standard therapy of chyle leakage still remains controversial. Considering our results, the efficacy of surgical intervention is clear. It is invasive, but can reduce not only chyle output volume but also length of hospital stay. Comparing to surgical treatment, conservative management is non-invasive, but has an unpredictable hospital course and requires long-term avoidance of oral intake [34].

Some case reports describe attempts at cervical re-exploration to identify and ligate the chyle leak [35]. This usually involves preparation with a fatty meal one to four hours pre-operatively to increase chyle production. Limited success has been reported with re-exploration of the neck. This is partly due to alteration of surgical anatomy, post-operative inflammatory changes and presence of chyle in the post-operative wound. In addition to this finding and ligating the thoracic duct in the neck is challenging. In our experience, we only explore the neck for washout of the chyle collection from the neck, placement of surgical drains or for other complications.

An important adjunct to surgical treatment of chylous leakage is the incorporation of thoracoscopic thoracic duct ligation. This treatment has been translated from the thoracic surgery literature regarding refractory chylothorax; thoracoscopic thoracic duct ligation is nowadays performed routinely as one of the first stages during Minimally-Invasive esophagectomy for cancer.

Historically, the classic technique involved a right-sided thoracotomy with identification and ligation of the thoracic duct just above the supradiaphragmatic esophageal hiatus [35]. Selle et al instructed this surgical modality for chyle drainage after failure of conservative management for 14 days, chylous outpout exceeding 500 cm^3 per day over a 5-days period, or systemic metabolic complications. Thoracic duct ligation has been reported to be successful up to 90% of the cases and could be performed without harmful effects. The thoracoscopic method decreases the morbidity rates of the classical open thoracotomy, and minimize the prolonged hospital stay [36]. Thoracoscopic thoracic duct ligation was performed in all of our presented cases following iatrogenic lesion during neck surgery [37]; patients were placed in prone position, using a double-lumen endo-tracheal tube to facilitate single-lung ventilation; access to the chest was achieved from the right-hand side. This approach is utilized in many of thoracic procedures, due to no need of lung retractor use with decreased rates of pulmonary complications and better visualization of the posterior mediastinum. Right-sided video-assisted thoracoscopic surgery (V.A.T.S.) through three ports was perfrormed, with dissection of the thorasic duct between azygos vein and thoracic aorta, above the level of the right diaphragm (Fig. 3). Two Hem-o-lok clips were applied in the dissected thoracic duct (Fig. 4). After careful haemostasis, 28 Fr chest tube was placed in the right costophrenic angle.

There are cases supporting a uniportal thoracoscopic approach [38,39] compared to three-ports thoracoscopy due to reduction of surgical trauma stress. However, three port thoracoscopic approached can offer excellent both instruments and tissues handling.

Conclusions

Chyle leak, particularly when high output, carries significant morbidity due to nutritional, fluid, electrolyte and lymphatic losses and can prolong the hospital stay. As demonstrated in our series and other reports, intra-operative identification and repair of iatrogenic thoracic duct injury still carries a significant risk of post-operative chyle leak. Octreotide alongside a low-fat diet is commenced to reduce chyle production. It seems that thoracoscopic approach is feasible, safe, and effective. In all of our patients, chyle leak decreased immediately after surgery.

Riassunto

La perdita del Chilo è una delle principali complicanze dopo un intervento chirurgico al capo e al collo, con un'incidenza che va dallo 0,5% fino all'8,3% come riportato nella letteratura. Questa perdita a livello cervicale del chilo può essere difficile da gestire, con una significativa morbilità dovuta ad ampie perdite di liquidi e di componenti nutrizionali.

In questo articolo vengono presentati quattro casi di perdita di chilo a livello cervicale dopo un intervento chirurgico al capo o al collo. La lesione del dotto toracico cervicale era stata identificata già durante l'intervento, ma il trattamento conservativo non è riuscito a ridurre la produzione di chilo dopo l'intervento.

A tutti i pazienti è stata eseguita la legatura del dotto

toracico sopra il diaframma a destra, previa sezione, e procedendo in posizione prona. Alla legatura ha fatto immediato seguito la risoluzione delle perdite, senza recidiva al follow-up.
La riparazione intraoperatoria del dotto toracico cervicale rimane controversa, mentre una volta che il danno sia stato identificato tempestivamente vengono a ridursi le morbilità conseguenti. La alternativa conservativa è rivolta alla riduzione della produzione di chilo, amplificando 'idratazione e integrazione alimentare.
La legatura toracoscopica del dotto toracico rappresenta un trattamento sicuro e fattibile in caso di fistola chilosa cervicale a seguito di un intervento chirurgico al capo o al collo grazie a tutti i progressi della chirurgia mininvasiva.

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