INTRODUCTION OF LYMPHANGIOGRAPHY AND PERCUTANEOUS EMBOLIZATION OF THE THORACIC DUCT IN A STEPWISE APPROACH TO THE MANAGEMENT OF CHYLOUS FISTULAS

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Abstract: Background. Chylous fistula occurring after head and neck or thoracic surgery is an uncommon but well-described complication, with a reported incidence of 1% to 2.5%. Conservative management can be successful and consists of dietary measures combined with suction drainage. This article reports on percutaneous embolization of the thoracic duct through catheterization of the retroperitoneal lymph vessels.

Methods. Two patients, in whom conservative management for cervical chylous fistula failed, underwent lymphangiography with opacification of the thoracic duct, followed by radio-guided catheterization and embolization.

Results. Embolization was successful in both patients. In 1 patient the procedure had to be repeated once to stop the chylous drainage.

Conclusions. Radioguided percutaneous catheterization and embolization of the retroperitoneal lymph vessels offers an excellent treatment option for patients with persistent chylous fistulas after failure of conservative management. We revised our stepwise management protocol (de Gier, Head Neck 1996; 18:347–351) and now consider this procedure as the secondary intervention step. ©2007 Wiley Periodicals, Inc. Head Neck 29: 1017–1023, 2007

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Chylous fistula occurring after head and neck surgery is an uncommon but well-described complication. The reported incidence of this complication is 1% to 2.5% in patients undergoing neck dissection.1–5 Since the majority of thoracic ducts drain on the left side of the neck, the risk of chyle leakage is higher on that side. However, up to 25% of the leakages may occur on the right side.1

Parts of this study have been presented at the spring meeting of the Netherlands ENT-Society, Nieuwegein, The Netherlands, in 2004; at the Congress of the European Association for Endoscopic Surgery, Venice, Italy, in 2005; and at the Society for Surgical Oncology meeting, San Diego, USA, in 2006.

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After originating in the chylous cisterns, ascending through the right thorax, and crossing over to the left thorax, the thoracic duct arches superiorly, anteriorly, and laterally as it enters the lower left side of the neck to form a loop before terminating into the venous system. Although this loop is usually 3 to 5 cm above the clavicle, a maximum of 8 cm has also been reported.5,7 Both the site of termination and the number of terminations are variable. In cadaver studies, duct endings were usually found within 1 to 2 cm from the junction of the left internal jugular and left subclavian vein.7,8 However, a considerable number of variations were seen ranging from single to multiple channels ending in the left internal jugular vein.8,9 These many anatomic variations, in combination with the transparent appearance of the ducts’ endings, contribute to increased surgical risk of inadvertent damage to the thoracic duct system.

Chyle (synonym for thoracic duct fluid) consists mainly of lymph and breakdown products of fat digestion. Free fatty acids with a carbon chain length of 12 or less are considered medium chain or short chain; these are water soluble and enter the blood stream directly through the portal system.6 Free fatty acids with a carbon length of more than 12 are considered long chain and are lipid soluble. After uptake from the small bowel, they are converted into triglycerides and transported to the bloodstream in chylomicrons via the thoracic duct. The flow rate of chyle in the thoracic duct can reach 2 to 4 L/24 hours depending on diet, peristalsis, coughing, respiration, and torso movements.1,11

The first signs of a chylous leak are an increase in wound drainage after resuming oral intake, while expecting a decrease.1 The drainage gradually turns from a reddish serous fluid into a milky creamy substance according to the amount of fat in the digested food.2 Persistent chylous fistulas can be potentially life-threatening and may induce fluid, protein, and electrolyte disturbances, lymphopenia, malnutrition, chylothorax, and problems of wound healing, eg, skin flap necrosis.4,10,12

Conservative management by medium chain triglyceride diet or total parenteral nutrition with continuous vacuum drainage resolves the problem in roughly half of the patients. If conservative management fails, surgical reintervention protocols have been advocated, but effective management of chylous fistulas remains difficult.1,3,5 In our previous paper on this subject, we reported a continuation of vacuum wound drainage, leaving surgical intervention only for cases refractory to this conservative approach.13

In this study, we report on a radiological intervention by percutaneous catheterization of the retroperitoneal vessels and embolization of the thoracic duct.14–17

**PATIENTS AND METHODS**

The NKI-AVL is a major reference center in the Netherlands for patients with head and neck malignancies. Yearly 80 to 100 neck dissections are performed, of which one third are en bloc radical neck dissections in patients with malignancies of the oral cavity, oropharynx, larynx, hypopharynx, and thyroid.18 Until 2004, the protocol published by De Gier et al13 was applied for the management of persisting chyle leakage after head and neck surgery, consisting of continued closed vacuum drainage combined with dietary measures. Only when conservative management failed, a muscle flap transposition in the wound bed was performed. By using this regimen, lymph leakage could be stopped in all patients.

From 2001 to 2005, 13 consecutive patients, 6 men and 7 women, with failed conservative management for chylous leakage underwent lymphangiography with visualization of the thoracic duct(s) and subsequent percutaneous transabdominal thoracic duct catheterization and embolization. Eleven patients had a chylothorax after thoracic surgery, and 2 had a cervical chylous fistula after neck dissection. Nine patients underwent their surgery in our institute, whereas the other 4 had been referred from other hospitals. The following primary surgeries had been performed: pleuropneumonectomy/lobectomy (n = 6), esophagectomy (n = 3), thymectomy (n = 2), and thyroideectomy combined with selective neck dissection (n = 2). Six patients had previous unsuccessful thoracic surgical reintervention and 1 patient an unsuccessful surgical reintervention in the neck. The first attempt of embolization following lymphangiography was successful in 6 patients. In 4 other patients, chyle leakage stopped although the thoracic duct could not be coiled. Attempts in 2 patients remained unsuccessful, and they were discharged to the referring hospital, and in 1 patient chyle leakage stopped spontaneously.

In this article, we report on the embolization procedures in patients with head and neck cancer who underwent a neck dissection and developed postoperative chyle leakage.
The technique was first described by Cope\textsuperscript{14} in 1998 and consists essentially of visualization of the lymphatics by (bi)pedal lymphangiography, transabdominal puncture of the cisterna chyli, and catheterization of the thoracic duct, followed by embolization.

We slightly modified the procedure used by Cope. Patients were fasting before the procedure and a pretreatment blood coagulation check was performed. No antibiotics were administered prophylactically. There was no need for barium administration, as in our experience the colon could always easily be identified. Lymphangiography was performed using uni- or bipedal injection of 9 to 12 mL of Lipiodol Ultrafluid (Guerbet Nederland BV, Gorinchem, The Netherlands).

The lower edge of the liver was identified by ultrasound and marked on the skin. The abdominal wall, peritoneum, and deeper soft tissues were lavishly infiltrated with lidocaine 1\% under conscious sedation with fentanyl and midazolam. As soon as the larger retroperitoneal lymph vessels and/or the chylous cisterns were visualized, puncture was performed via an entry point several centimeters above the umbilicus, depending on the estimated needle tract. Puncture was carried out using a coaxial technique with a 22-G 20-cm and a 18-G 9-cm needle during anteroposterior and lateral radio guidance. Once a major lymph vessel or the cistern was punctured, a 0.018-in. Cope wire (COOK, Limerick, Ireland) was introduced. Over this wire, the inner 4F portion of a Neff set (COOK) was introduced to dilate the entry point. Thereafter, a 3F microcatheter (COOK) was introduced and advanced in the thoracic duct. After injection of aqueous contrast to better identify the leak, embolization was carried out using microcoils (Tornado Embolization Microcoils, COOK), followed by tissue adhesive (Histoacryl, B.Braun/Aesculap, Tuttingen, Germany). The catheter was then immediately removed. Embolization was carried out through a microcatheter with 3 microcoils and 1 mL of a mixture Histoacryl/Lipiodol 1:1 on a midthoracic level.

RESULTS

The histories of 2 patients with cervical chylous fistulae are described in detail. In both patients the embolization procedure was successful, although in 1 patient the procedure had to be repeated to stop the chylous drainage.

Case 1. A 55-year-old woman underwent a total thyreoidectomy and selective neck dissection on the left (levels II–V), with preservation of the internal jugular vein, sternocleidomastoid muscle, and accessory nerve for removal of her papillary thyroid carcinoma. The thoracic duct was identified during the operation and ligated with multiple clips. Two closed high vacuum suction drains were left behind. On the first postoperative day, 520 mL of fluid with a milky appearance was observed in the drain reservoir. The patient was put on a diet of medium chain triglycerides (MCTs). During the following days, serum electrolytes were carefully monitored and adequately supplemented. Serum glucose levels were found to be markedly raised, and treatment with intravenous insulin was started. Because of a gradual increase of drain production up to 2600 mL/24 h on postoperative day 4, the MCT-diet was replaced by total parenteral nutrition (TPN, Fresenius Kabi Nederland, ’s-Hertogenbosch, The Netherlands) on the seventh postoperative day. Wound production continued to be high, with volumes up to 1840 mL/24 hours. On postoperative day 16 the patient was returned to the operating room, where the wound was re-explored under general anesthesia. A substantial chyle-leakage was observed at the lower neck, with the patient in Trendelenburg’s position; therefore the thoracic duct was ligated with 10 clips and sealed off with Tissuecol (Baxter, Utrecht, The Netherlands). A pectoralis major muscle flap was prepared and, after a second application of Tissuecol, fixed to the sternocleidomastoid muscle to fill up the defect. After inserting 2 suction drains, the wound was closed in layers.

Because of an increased drain production of up to 3300 mL/24 hours on the 19th postoperative day, an unsuccessful attempt was made to stop the leakage by injection of 300 mg doxycycline for sclerosis purposes.

Thereafter drain production increased to a maximum of 4010 mL/24 hours on the 20th postoperative day. Because standard institutional complication treatment (de Gier et al) proved to be failing, the thoracic surgeon was consulted for intrathoracic clipping of the thoracic duct.

Considering the patient’s deterioration, with diabetes and catabolic changes, lymphography followed by percutaneous catheterization and embolization of the thoracic duct was proposed. This procedure was carried out on the 21st postoperative day, 5 days after the surgical intervention. After right-sided lymphangiography and...
infusion of 9 mL Lipiodol, the 4F Neff catheter was introduced with delivery of only 2 microcoils. More microcoils could not be placed, since the catheter slipped out off the thoracic duct. Serum glucose levels normalized rapidly and drainage production decreased gradually to a minimum of 15 mL/24 hours within 5 days. However, after these 5 days, leakage increased to a maximum of 1150 mL/24 hours.

On the 31st postoperative day, a second percutaneous embolization was performed, using bipedal lymphangiography with a total volume of 12 mL Lipiodol. Embolization was carried out using 3 microcoils combined with 1 mL of a mixture of Histoacryl/Lipiodol (1:1) at midthoracic level. The drainage production rapidly halted. The patient was discharged on the 42nd postoperative day, with continuation of the MCT-diet for 2 more weeks. Twenty-five months after treatment, the patient is still alive without signs of recurrent disease. The course of chylous leakage and intervention are shown in Figure 1.

Case 2. A 63-year-old woman underwent a total thyreoidectomy with bilateral selective neck dissection (levels II–V, sparing of the right internal jugular vein, both sternocleidomastoid muscles, and accessory nerves), for removal of a Hurtle-cell carcinoma.

After sacrificing the left internal jugular vein, a leakage of the thoracic duct was noticed; this was treated by oversewing the leakage site with atraumatic Prolene 5 × 0 (Ethicon/Johnson & Johnson Medical BV, Amersfoort, The Netherlands). Before closure of the skin, no chyle leakage could be detected after a local check with the patient in Trendelenburg’s position. Two closed high vacuum suction drains were left behind.

Already on the first postoperative day, a chyle production of 1640 mL/24 hours occurred and an MCT diet was started immediately. During the following days, serum electrolytes were carefully monitored and low calcium was adequately supplemented. On the second postoperative day, the MCT diet was replaced by TPN. Nevertheless, drainage production remained high, with volumes up to 2400 mL/24 hours.

On the 14th postoperative day, unilateral lymphangiography with 12 mL Lipiodol was performed. Because no cisterna chyli was present, the distal thoracic duct was directly punctured. This resulted in some leakage of Lipiodol to the right pleural space without any significant sequelae. A successful embolization with 3 microcoils and Histoacryl was carried out. The patient was discharged in good condition on the 29th postoperative day, after antibiotic treatment for an intercurrent sepsis caused by wound infection. Sixteen months after treatment, the patient is still alive with stable disease.

Figure 2 shows details of the course of chylous leakage before and after intervention.
**DISCUSSION**

During surgical procedures in the lower neck, the thoracic duct is prone to iatrogenic injury because of the wide variation of the thoracic duct anatomy and its fragile transparent wall. The head and neck surgeon should be aware of the difficulties in treating this postoperative complication. In general, it is not recommended to search routinely for the thoracic duct during neck surgery; but should the duct be identified during the dissection and still be intact, then it is advisable to leave it untouched unless it frustrates the oncological resection. If by occasion a creamy, watery fluid fills up the lower part of the dissection, an active search for the lower part of the duct is required. If the leak can be identified, it is advisable to ligate the duct (endings) with Prolene 0. Application of Histoacryl combined with Surgicel (Ethicon/Johnson & Johnson Medical BV) or Vicryl mesh (Ethicon/Johnson & Johnson Medical BV) might be helpful. The surgeon can make a final check for chylous leakage at the end of a neck dissection before closure of the skin, by placing the patient in Trendelenburg's position and thereby creating an increased intrathoracic pressure.

For the last decade, we treated chylous leakage conservatively with continued vacuum drainage, MCT diet, and if necessary, followed by TPN. In the years after our publication, this regimen proved to be effective. In a series of 394 neck dissections (345 patients) performed in our institute between 1996 and 2001, 16 patients (4%) developed a chylous fistula postoperatively. In 7 of these cases, the thoracic duct was ligated during surgery. By using our stepwise approach, it was observed that the leakage stopped spontaneously in 2 patients, and by use of an MCT diet in 11 patients. A switch to TPN proved to be successful in another 2 patients with unsuccessful MCT diet. Although some authors question the use of TPN because of the lack of experimental evidence, the costs, and the potential danger of infections by parenteral feeding, we have used this technique successfully. Only 1 patient required a muscle flap transposition to stop the leak.

On the basis of the described data, we discourage attempts to locate the leak by reexplorative surgery because the local wound conditions impair proper identification of major blood vessels and nerves. Similar concerns have been expressed by others. Closed-vacuum drainage is essential for wound healing in the neck and promotes fixation of skin flaps to the under layer. Local application of pressure dressings is not effective in case of chyle leakage, as they are difficult to keep in place and may interfere with the precarious blood supply to skin flaps. Many adjunctive measures have been proposed in the literature, such as injection of tetracycline solution in the wound bed, but none of these have developed into routine practice. There are some case reports suggesting that there might be a beneficial effect from subcutaneous or venous administration of somatostatin (analougues) on the cessation of lymph leakage. Therefore, we feel that it could well be justified to add this drug in combination with parenteral feeding.

In cases of persistent chylous fistula, ligation of the thoracic duct by thoracic surgery (or by thoracoscopic techniques or video-assisted thoracic surgery) has been described as an alternative for local explorative surgery. However, in the international literature only a few cases have been reported in relation to chronic chyle fistulas after neck surgery. Although the thoracoscopic method seems attractive as an alternative for open thoracic surgery and intrathoracic ligation of the duct, the advantages of video-assisted thoracic surgery procedures over open thoracic surgery are modest and the many intrathoracic anatomic variations and duplications may interfere with straightforward localizing and clipping of the duct(s).

Percutaneous catheterization and embolization of the retroperitoneal lymph vessels, a technique first described by Cope in 1998, is a minimally invasive alternative for management of this complication. Depending on the diameter, the thoracic duct can be catheterized for embolization with microcoils and Histoacryl. If the diameter is too small for catheterization, it is assumed that scar tissue formation caused by (multiple) perforations of the thoracic duct may finally lead to cessation of lymph leakage. Cope et al published a series of 60 procedures for chylothorax, with a success rate of 70% without any morbidity related to this intervention procedure. If catheterization was impossible or a chylous fistula persisted after multiple embolizations, the thoracic duct was ligated by surgery or pleurodesis.

Although we did not register major morbidity associated with this treatment, the procedure can be time-consuming and painful. In our experience, analgesia by local anesthesia and sedation (fentanyl and midazolam) appeared to be insuffi-
cient, and in future cases epidural anesthesia will be considered. Analgetic medication should be continued for some days. With the advances of Intervention radiology, percutaneous embolization of the retroperitoneal lymph vessels can be offered much earlier in the course of management of chyle fistulas.

We revised our stepwise management protocol (Figure 3). Now, we consider lymphangiography and percutaneous embolization as the secondary intervention step for patients with high-output fistulas and longstanding wound drainage after a primary conservative management period of roughly 10 days with MCT or TPN. Based on our own limited experience and literature data, in the majority of patients chyleous leakage can be stopped by this minimally invasive procedure.14–17 If not successful, visualization of the thoracic duct(s) gives important additional anatomic information, which might be helpful in case of subsequent surgical intervention.

REFERENCES

18. Balm AJ, Lohuis PJ, Copper MP. Surgical technique—unwrapping the neck node levels around a sternocleido-


