

Chylous ascites in gynecologic malignancies: cases report and literature review

Glauco Baiocchi · Carlos Chaves Faloppa · Raphael Leonardo Cunha Araujo · Elza Mieko Fukazawa · Lillian Yuri Kumagai · Ademir Narciso Oliveira Menezes · Levon Badiglian-Filho

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Abstract

Purpose Chylous ascites is an infrequent postoperative complication after retroperitoneal surgical procedure. Despite its infrequent occurrence, postoperative chylous ascites are associated with significant morbidity. Reports of chylous ascites or fistula after retroperitoneal lymph node dissection for gynecologic malignancies without radiation therapy are rare. A search in the English literature showed only 31 cases of chylous fistula for gynecologic malignancies. Treatment may be conservative with low-fat oral diet with medium-chain triglycerides associated or not to octreotide and total parenteral nutrition. In case of conservative measures failure, it can be managed by surgical intervention or peritoneo-venous shunt.

Methods We report two cases of chylous fistula following systematic pelvic and retroperitoneal lymph node dissection for gynecological cancer without radiotherapy and review the literature.

Results Both were successfully managed with the maintenance of the postoperative drain, total parenteral nutrition, octreotide and dietary intervention.

Conclusions Chylous ascites should be included in differential diagnosis of abdominal distention after surgical retroperitoneal approach or radiotherapy. Most of the patients

may have their chylous ascites successfully treated with conservative management. However, the best policy is to prevent chylous complications by employing meticulous dissection techniques and careful control of the major lymphatics by suture ligation during the primary surgical intervention.

Keywords Chylous ascites · Lymph node dissection · Gynecologic neoplasm

Introduction

Chylous ascites is defined as a pathological accumulation of chylous fluid in the peritoneal cavity after a chylous leakage [1]. Postoperative chylous ascites is an unusual complication of retroperitoneal surgery after unrecognized disruption of major retroperitoneal lymphatics that usually develops as a result of surgical trauma to the lymphatic system [2, 3]. Other conditions may be associated with extensive occlusion of retroperitoneal lymphatics such as malignancies, tuberculosis, cirrhosis and secondary to nephritic syndrome [4]. Chylous ascites is an infrequent postoperative complication after retroperitoneal surgical procedure. In a retrospective series, Press et al. [5] reported a frequency of 1 in 20,464 hospital admissions. For testis cancer, Baniel et al. [6] showed 2% chylous ascites in 603 patients who underwent post-chemotherapy retroperitoneal lymph node dissection (RPLND) and Evans et al. [7], an incidence of 7% in 329 patients. The iatrogenic causes of lymphatic disruption resulting in chylous ascites also include abdominal or pelvic radiation [8].

Despite its infrequent occurrence, postoperative chylous ascites are associated with significant morbidity. Reports of chylous ascites or fistula after RPLND for gynecologic

G. Baiocchi · C. C. Faloppa · R. L. C. Araujo · E. M. Fukazawa · L. Y. Kumagai · A. N. O. Menezes · L. Badiglian-Filho
Department of Gynecologic Oncology,
Hospital do Câncer AC Camargo, São Paulo, Brazil

G. Baiocchi (✉)
Departamento de Ginecologia,
Hospital do Câncer AC Camargo,
Rua Antonio Prudente, 211, 01509-010 São Paulo, Brazil
e-mail: glbaiocchi@yahoo.com.br

malignancies without radiation therapy are rare. Most reports are related with radiation therapy.

We report two cases of chylous fistula following systematic pelvic and retroperitoneal lymph node dissection for gynecological cancer without radiation therapy. Both were successfully managed with total parenteral nutrition (TPN), octreotide and dietary intervention.

Case report

Case 1

A 50-year-old woman was referred to our institution after previous unilateral salpingo-oophorectomy, peritoneal washings and peritoneal biopsies as a suboptimal cytoreduction for a stage IIIC (FIGO) serous-papillary ovarian cancer. Thoracic, abdominal and pelvic computed tomography showed no clear evidence of residual disease. She was submitted to another surgical procedure that consisted in an exploratory laparotomy, total abdominal hysterectomy, contra-lateral salpingo-oophorectomy, infra-colic omentectomy, peritonectomy of right and left diaphragms, pelvic peritonectomy, appendectomy, resection of ileal and colonic implants, and systematic pelvic and retroperitoneal lymph node dissection up to the renal veins. It was considered as an optimal cytoreduction with no visible residual disease.

There was no palpable enlarged lymph node during the lymph node dissection and all 110 lymph nodes were pathologically free of disease. Fifty-three retroperitoneal lymph nodes were resected, of which 35 were above the inferior mesenteric artery.

Oral diet started at the fourth postoperative day and during the ninth day the patient presented symptoms of nausea and vomiting. Then we noticed that the serous fluid from the drain tube previously left in the pelvis, turned onto a milk whitish fluid. It was also associated with an increasing drainage volume from 370 ml in the past 12 h to 1,470 ml during the following 24 h.

Laboratory examination revealed fluid with pH 7.6, triglyceride of 482 mg/dL, albumin of 2.0 g/dL, glucose of 149 mg/dL, amylase of 26 UI/L. Serous triglyceride was 182 mg/dL and albumin 1.7 g/dL.

She was submitted to a computed tomography that did not demonstrate any ascites and then on the tenth postoperative day she started receiving octreotide 100 mcg subcutaneously every 8 h, TPN and water oral intake. After 2 days, the drain fluid became serous and low-fat oral diet with medium-chain triglycerides was introduced. Octreotide was maintained during 5 days and TPN was withdrawn after 14 days. She was discharged on 30th postoperative day and the drain was removed

after a 72 ml drainage volume in 24 h. On 35th postoperative day she stopped the low-fat oral diet and remained asymptomatic.

The adjuvant chemotherapy started on the 53th postoperative day and 6 cycles of carboplatin and paclitaxel were administered. She developed peritoneal recurrence refractory to second-line chemotherapy 6 months after the primary surgery and died because of disease progression after 10 months of follow-up.

Case 2

A 63-year-old woman with a body mass index of 40.4 kg/m² was referred to our institution after postmenopausal bleeding and endometrial thickness of 16 mm. The hysteroscopic evaluation and biopsy diagnosed a grade 3 endometrioid adenocarcinoma of the endometrium. Magnetic resonance of abdomen and pelvis showed possible myometrial invasion of less than 50% and no evidence of extra-uterine disease. The CA125 was 14 UI/dL. She was submitted to a total abdominal hysterectomy, bilateral salpingo-oophorectomy, infra-colic omentectomy, peritoneal washing and systematic pelvic and retroperitoneal lymph node dissection up to the renal veins. A drain tube was left in the pelvis.

The final pathological finding was a stage IBG3 endometrioid adenocarcinoma (FIGO). All 42 lymph nodes were pathologically free of disease. Twenty retroperitoneal lymph nodes were dissected.

Oral diet started at the third postoperative day and during the fourth day the serous fluid drainage turned onto milk whitish. It was also associated with an increasing drainage volume from 655 ml during the past 24 h to 2,470 ml in the following 24 h.

Laboratory examination revealed fluid with triglyceride of 350 mg/dL, total cholesterol of 29 mg/dL, proteins of 1.3 g/dL, glucose of 126 mg/dL. Serous proteins were 4.2 g/dL and albumin 1.5 g/dL.

She started receiving conservative management with the association of low-fat oral diet with medium-chain triglycerides, octreotide 100 mcg subcutaneously every 8 h and TPN. After only 72 h, the drain fluid became serous. Octreotide was used during 5 days and withdrawn after 24 h of serous fluid discharge. TPN was maintained during 10 days. She was discharged on 14th postoperative day and the drain was removed after a 50 ml drainage volume in 24 h. On the 21th postoperative day she stopped the low-fat oral diet.

Adjuvant external beam radiotherapy and high-dose brachytherapy were performed. She remains asymptomatic and without recurrent disease after 10 months of follow-up.

Discussion

Retroperitoneal surgery is the most common iatrogenic etiology of chylous ascites and is related to surgical injury to the retroperitoneal lymphatic glands or cisterna chyli [9]. It is a rare complication that results in a continuous accumulation of chylous fluid in retroperitoneal and abdominal cavities. Abdominal aortic surgery is the procedure that is most commonly related to chylous complications. Although lymphatic leaks account for only 1% of all complications after aortic surgery, this procedure is implicated in over 80% of cases of postsurgical chylous ascites [9]. Regarding cancer treatment, most are reported after post-chemotherapy retroperitoneal lymph node dissection for testicular cancer [1, 2, 5–7].

A search in the English literature showed only 31 cases of chylous fistula for gynecologic malignancies [10–13]. Chylous ascites has been reported following retroperitoneal lymph node dissection in the absence of radiation therapy in only three cases [11–13]. Here, we present another two cases. Regarding chylous ascites after pelvic lymph node dissection without the addition of either retroperitoneal lymph node dissection or radiation therapy, only two cases have been reported [14].

Chylous ascites was treated only conservatively on 16 (64%) of the 25 cases that the treatment details were reported. Peritoneo-venous shunting treatment associated with gynecologic malignancies has been successfully reported for only two patients [10].

Chyle consists on lymphatic fluid and droplets of fat (chylomicrons) in a stable emulsion [10]. Bowel rest and low-fat elementary diets decrease the lymph flow, while fatty meal ingestion may increase the chyle flow from a fasting baseline of less than 1 mL/min to more than 200 mL/min [3, 6].

The ascending vertical lumbar lymphatic trunks are the major lymphatic channels in the retroperitoneum. They originate from the coalescence of the common iliac lymph vessels derived from the lymphatic glands of the lower extremities and pelvic organs [1]. Ascending vertical lumbar, descending thoracic, intestinal and liver lymphatics join the cisterna chyli, which lies at the level of L1–L2 [10]. In about half of the cases, the cisterna chyli is less defined [3]. At T12–L1, the cisterna chyli ascends as the thoracic duct [1]. Normal lymph flow in the thoracic duct is less than 1.5 L/day [8]. The thoracic duct then ascends through the mediastinum to join the venous system at the junction of the left subclavian and internal jugular veins in 80% of cases and in 20% cases at the right subclavian, internal jugular, or brachiocephalic vein [10]. From the cisterna chyli lymph flows; 50–90% is derived from the intestine and liver and contains absorbed dietary fat in chylomicrons form [15]. Long-chain triglycerides absorbed in the bowel

enter the venous system through the lymphatic system, and they significantly affect the lymph flow [1]. On the other hand, medium-chain triglycerides bypass the lymphatics [1]. They are absorbed directly into the portal venous system and do not increase the amount of lymph flow [2]. Therefore, medium-chain triglycerides diet is expected to contribute to chylous fistula resolution [13].

Chylous ascites may result from congenital malformations of the lymphatic channels, acquired causes of lymphatic occlusion, disruption of lymphatic channels, or hemodynamic causes. Acquired causes of lymphatic occlusion include lymphoma or tumor metastases, radiation, and inflammatory and infectious diseases such as tuberculosis, filariasis, peritonitis, pancreatitis, and sarcoidosis. Disruption of the major lymphatic channels can be due to blunt abdominal trauma or surgical intervention. Hemodynamic causes include cirrhosis and portal vein thrombosis, which increase pressure within the cisterna chyli and thoracic duct that result in potential for rupture within the lymphatic vasculature [4].

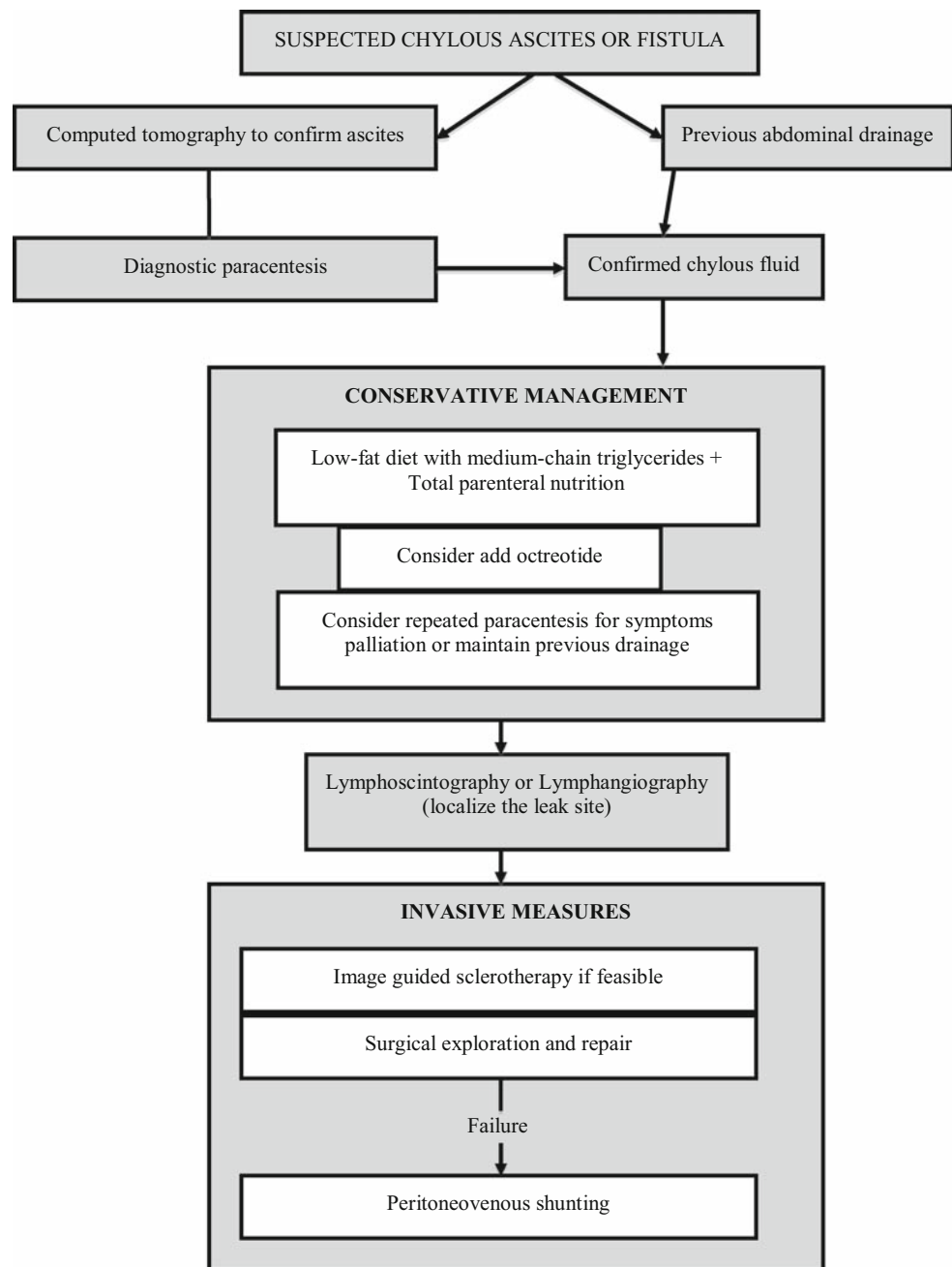
Kaas et al. [16] reported an incidence of 7.4% chylous ascites after complex oncological abdominal procedures. However, the incidence of chylous ascites for gynecologic malignancies remains unclear and the ideal treatment is not well established.

Chylous ascites should be suspected as a differential diagnosis in patients with gynecologic malignancies that develop ascites after RPLND or radiation therapy. Postoperative chylous ascites may become evident from several days to months following surgery. It is also crucial to confirm fluid nature of the ascites and its underlying cause [10]. Patients with chylous ascites usually present abdominal fullness and distention. The typical fluid characteristics of the chylous ascites is clear to milky color, sterile, odorless and the fluid analysis reveals alkaline pH, high triglyceride content (2- to 8-fold above plasma), high protein content (>3 mg/dL) and high leukocyte levels [8, 17].

Computed tomography is useful to identify the ascites and coexistent recurrent disease but is unlikely to identify the site of chyle leakage [8, 10]. Bipedal lymphangiography is the imaging modality traditionally used and has the potential advantage of localize the site of lymphatic leakage, and may facilitate surgical repair in some cases. Unfortunately, cannulation of pedal lymphatics is painful and technically difficult [17, 18]. Lymphoscintigraphy is a less invasive alternative and can confirm an active lymphatic leak [18]. Some authors suggest that a positive bipedal lymphangiography or lymphoscintigraphy might be associated with less likelihood of resolution with conservative measures [1, 18].

Due to its rarity, the management of chylous ascites remains controversial and variety of successful strategies have been reported. Aalami et al. [4] published a large

Fig. 1 Algorithm for the management of chylous ascites following retroperitoneal surgery



review of 156 cases. They reported 67% resolution rate with conservative measures and 33% required surgical procedure. Conservative management of chylous ascites is based on low-fat diet with medium-chain triglycerides. It can take weeks or even months to succeed. The objective is to decrease the intestinal lymphatic flow and triglycerides transport, and consequently prevent lymph and triglyceride accumulation [8]. This diet may also be considered for maintenance therapy for several months after resolution of chylous ascites [1, 6]. TPN associated with no oral intake may neither not diminish chyle production nor effectively reduce lymph flow, but might be important to restore nutri-

tional and metabolic impairments caused by chylous ascites, underlying disease and low oral intake [1, 10, 13]. However, it may have a success rate of 60–80% [6, 9] and should be continued for a minimum of 3 weeks if clinical improvement is desired [4, 13]. Conservative measures may include repeated paracentesis that can provide transitory palliation of abdominal symptoms. Unfortunately, it has been implicated in prolongation of leaks, worsening of nutritional status and infectious complications [6].

Somatostatin and its analogs (octreotide) have been described with or without adjunctive TPN. The exact role remains controversial, but it seems to decrease the intestinal

absorption of fats, decreases the triglyceride concentration in the thoracic duct, and attenuates lymph flow in the major lymphatic channels. It also decreases splanchnic blood flow and decreases gastric, pancreatic, and intestinal secretions. As the intestinal absorption decreases, a further decrease in lymph production is expected. Various success rates have been described and it has been recommended in cases not responsive to the initial measures of dietary modification and TPN. Responses are typically achieved within 24–72 h [10, 19].

In cases of failure of conservative measures, lymphoscintigraphy may localize the leak site and guide either image guided sclerotherapy or surgical intervention [16]. Aalami et al. [4] reported that 41% of 51 patients were successfully treated with direct surgical ligation of the leaking site. Surgical exploration success depends on finding the leak site and this can also be facilitated by ingestion of preoperative heavy fatty meal or by intraoperative lipophilic dye administration [3, 6, 17]. The optimal timing of surgical intervention remains controversial. Most authors suggest 4–8 weeks of conservative measures before surgery [9, 17, 19]. Evans et al. [7] reported a resolution rate with conservative measures of 65% for 23 patients, within a median time of 13 weeks.

The use of peritoneo-venous shunt remains a satisfactory therapeutic or palliative option for the management of severe refractory chylous ascites, mainly in patients with poor performance status who cannot undergo surgical exploration [6, 10, 12, 19]. It can be associated with several complications, especially occlusion of the shunt due to the high viscosity of the chyle, sepsis, disseminated intravascular coagulation, hypokalemia, ascites leak, pulmonary edema, and air embolism [7, 10, 19, 20].

Based on our experience and literature review, we propose an algorithm shown in Fig. 1. Our two cases were successfully treated with conservative measures with association of dietary intervention, TPN and octreotide started on the same day. With these measures, we had chylous fistula resolution in less than 7 days. Both were related to systematic retroperitoneal lymph node dissection without radiotherapy. We usually leave a drain after retroperitoneal lymph node dissection and it was very useful to diagnose the postoperative chylous fistula and monitor its resolution. Although some reports suggest that continuous paracentesis may perpetuate chylous ascites [6, 17], the drain maintenance in our report did not confirm this data.

Chylous ascites is a rare, but an important complication following retroperitoneal surgery. It should be included in differential diagnosis of abdominal distention after surgical retroperitoneal approach or radiotherapy. The best policy is to prevent chylous complications by employing meticulous dissection techniques and careful control of the major lymphatics by suture ligation during the primary surgical intervention [2, 3, 13, 19].

Conflict of interest statement None.

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