

Contents lists available at [ScienceDirect](http://www.sciencedirect.com)

## International Journal of Surgery Case Reports

journal homepage: [www.casereports.com](http://www.casereports.com)

# Refractory hepatic lymphorrhea after total pancreatectomy. Case report and literature review of this uncommon complication



Michele Bartoli\*, Gian Luca Baiocchi, Nazario Portolani, Stefano Maria Giulini

Department of Medical and Surgical Sciences, Surgical Clinic, University of Brescia, Brescia Civil Hospital, P.le Spedali Civili, 1, 25123 Brescia, Italy

## ARTICLE INFO

### Article history:

Received 20 August 2015

Received in revised form

14 September 2015

Accepted 18 September 2015

Available online 26 September 2015

### Keywords:

Refractory hepatic lymphorrhea

Chylous ascites

Lymphadenectomy

Hepatoduodenal ligament

Lymphangiography

## ABSTRACT

**INTRODUCTION:** After extended abdominal lymphoadenectomy, lymphatic vessel injury may cause lymphorrhea that usually disappears spontaneously. However, intractable ascites sometimes develops. Although there are many reports describing persistent chylous ascites from intestinal lymphorrhea, little is known about hepatic lymphorrhea, not containing chyle. It is caused by injury of the lymphatic vessels during hepatoduodenal ligament lymphadenectomy.

We present a case of massive ascites due to hepatic lymphorrhea after total pancreatectomy and extended lymphoadenectomy for Ampullar adenocarcinoma. We successfully treated it with prolonged medical therapy after surgical relaparotomy.

**PRESENTATION OF CASE:** A 65-year old man underwent total pancreatectomy with extended nodal dissection. Massive clear-colored ascites (2000–9000 mL per day) developed since the second postoperative day and persisted despite conservative therapy. At re-laparotomy no lymphatic leakage was found. Similarly lymphangiography was showed no contrast spreading. We treated this hepatic lymphorrhea with intermittent opening of the abdominal drainage until spontaneous resolution.

**DISCUSSION:** The standard treatment of hepatic lymphorrhea is an aggressive medical treatment. After such approach the most effective therapy seems to be surgical exploration. Other option are peritoneovenous shunt or intraperitoneal administration of OK-432.

**CONCLUSION:** In our experience the intermittent abdominal drainage until spontaneous resolution is an useful approach to hepatic lymphorrhea.

© 2015 The Authors. Published by Elsevier Ltd. on behalf of Surgical Associates Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

## 1. Introduction

During abdominal surgery, especially after extended lymphoadenectomy for gastric or pancreatic cancer, lymphatic vessel injury causes lymphorrhea. The postoperative lymphorrhea usually disappears spontaneously in a few days. However, intractable ascites may develop especially in patients with liver cirrhosis, heart failure or renal failure. Although there are many reports describing the diagnosis, causes and treatment of persistent chylous ascites from intestinal lymphorrhea, little is known about hepatic lymphorrhea following abdominal surgery. It is caused by injury of the lymphatic vessels especially during the hepatoduodenal ligament lymphadenectomy. Usually conventional medical treatments such as cessation of oral intake, total parenteral nutrition, diuretic therapy, albumin support, intravenous reinfusion of ascites, or therapeutic paracentesis are unsuccessful. We present a case of intractable massive ascites due to hepatic lymphorrhea after total pancreatectomy and extended lymphoadenectomy for

Ampullar adenocarcinoma, successfully treated with conservative approach after ineffective surgical relaparotomy.

## 2. Presentation of case

A 65-year old man was hospitalized due to obstructive jaundice. An adenocarcinoma of Vater papilla with nodal involvement was diagnosed by abdominal TC scan and ERCP. Preoperative staging was T2N+. ASA grade was 3. Intra-operative finding of soft pancreas and little Wirsung duct (<3 mm) prompted us to perform total pancreatectomy. Suspected nodal involvement till interaortocaval nodes was found, so extended lymph node dissection was carried out (over D2). Postoperative staging was T3N1 with involvement of 2 of 27 nodes. Massive ascites, ranging from 2000 to 9000 mL per day, developed since the second postoperative day. The ascites was unresponsive to albumin and electrolyte support, administration of maximal diuretics dose, diet, total parenteral nutrition (TPN), continuous drainage, and octreotide infusion. Cytological examination revealed no malignant cells, and culture was initially negative. The ascites was clear-colored and was diagnosed biochemically as non chylous lymphorrhea with total protein concentration of 3 g/dL, albumin concentration 1.5 g/dL and serum-ascites gradient

\* Corresponding author.

E-mail address: [michelebartoli83@gmail.com](mailto:michelebartoli83@gmail.com) (M. Bartoli).



Fig. 1. Lymphangiography showing no contrast spreading.

1 g/dL. The main constituent of the ascites was hepatic lymph. It derived from surgical injury of the lymphatics in the hepatoduodenal ligament. In the 19th post operative day (POD) we decided to perform surgical re-exploration. However no obvious lymphatic leakage point was found even after administration of fatty meal through naso-gastric tube and Lipofundin (B Braun; Melsungen; Germany) injection in lymphatic vessels. Meticulous exploration of hepatoduodenal ligament was performed; even in absence of clear leakage, surgical ligation of inter aortocaval tissues and Atossiclerol (Chemische Fabrik Kreussler & Co.; Wiesbaden; Germany) application in hepatoduodenal ligament were carried out. Also Tachosil (Takeda; Linz; Austria) was applied to treat lymphatic leakages.

After surgical treatment there was no change in the amount of the drainage (about 4000–9000 mL/day). The patient's daily activities worsened, due to disturbance of mobility and fever compromising respiratory function till acute respiratory failure.

In the 34th POD, due to the increasing of lymphorrhea, lymphangiography was performed in an attempt to identify (and treat with lymphatic embolization) an unrecognized leakage. However no contrast spreading was found (Fig. 1).

The patient developed sepsis sustained by *Pseudomonas Aeruginosa* at first, and then by multi-resistant *Klebsiella Pneumoniae* and *Candida Albicans*. He also suffered from septic chorioretinitis that required surgical treatment (vitrectomy) in addition to targeted antibiotic therapy.

The postoperative course was characterized by progressive weight loss, hypoalbuminemia, electrolyte imbalance and diffuse edema. We decided to close the abdominal drainage for a few days. This option caused asthenia, appetite loss, nausea, vomiting, fullness sensation, abdominal distention, peripheral edema and required parenteral support and careful enteral nutrition with nose gastric tube.

The intermittent opening of the drainage, caused the progressive decrease of the lymphorrhea. After 132 days we could remove the tube. The patient was discharged in the 181th POD. At 12 months follow-up, the patient is alive, disease free and has fair general conditions without lymphorrhea recurrence.

### 3. Discussion

Intra abdominal lymph pathways are mainly classified into hepatic and intestinal. Both these pathways drain into the cisterna

chyli and subsequently into the circulatory system through the thoracic duct. The hepatic lymphatic system has two major pathways (ascending and descending) of drainage. Via the ascending pathways, lymph from the surface of the upper part of the liver flows along the diaphragm into the cisterna chyli while lymph from the liver bed and in the liver flows along the hepatic veins. The descending pathway runs through the hepatoduodenal ligament with portal vein, hepatic artery and bile duct. Intestinal lymph drains 50–75% of intra abdominal lymph and contains mainly lipid droplets of long chain fatty acids; thus its color is milky. On the other hand, hepatic lymph drains 25–50% of intra abdominal lymph. This lymph contains proteins of the same density as plasma without lipid droplets and is clear-colored [1,2].

Intra abdominal lymphorrhea, without chylous ascites, is a rare complication after abdominal surgery. It results from injury of the lymphatic vessels particularly in the hepatoduodenal ligament. Although small lymph leaks are commonly left open during lymphadenectomy, they rarely cause lymphorrhea because of the abundance of collateral lymphatic channels and the regeneration ex-novo of lymphatics. Therefore they usually remain clinically asymptomatic or unrecognized. Lymphorrhea becomes intractable in cases of injury to major lymphatic vessels and when reparative mechanisms become insufficient. Little is known about the management of postoperative hepatic lymphorrhea. To our knowledge, there is only one report of hepatic lymphorrhea following abdominal surgery in the Western medical literature [3], and less than twenty cases in Eastern medical literature [4]. After diagnosis of postoperative hepatic lymphorrhea, all patients were treated conventionally by diet, TPN, diuretics and therapeutic paracentesis.

We performed an aggressive medical treatment since the presentation of lymphorrhea by TPN, massive diuretics therapy, continuous drainage, albumin and electrolyte support and octreotide infusion. The mechanism by which octreotide affects lymphorrhea is unknown. It has also been speculated that somatostatin inhibits lymph fluid excretion through specific receptors found in the normal lymphatic vessels.

After conventional medical treatment, various regimens are used to treat the intractable hepatic lymphorrhea (Table 1). The most effective therapy appears to be surgical ligation of lymph fistula with sclerotherapy and/or fibrin glue sprinkle even if the preoperative imaging study does not show an obvious leakage point [4].

We carried out surgical ligation earlier than other authors, even in absence of obvious leakage point, because of amount of lymphorrhea and resistance to all conservative therapies. No author reported ascites more than 4500 mL per day. At the time of reoperation the patients bothered lymphorrhea ranging from 4000 to 8000 mL per day, drained by the abdominal drainage tube. It caused severe and potentially life-threatening hypokalemia, albumin depletion and dehydration despite medical therapy.

Inoue [5] reported the use of peritoneovenous shunt (PVS) for intractable ascites due to hepatic lymphorrhea. The placement of a PVS is mainly used for intractable ascites due to decompensated liver cirrhosis and for refractory chylous ascites. It is a simple and cost-effective procedure even if it is linked to high morbidities such as obstruction, vascular thrombosis, infections, sepsis and disseminated intravascular coagulopathy. Hence it has become a less common technique. Moreover our patient showed signs of sepsis (leukocytosis, fever, high PCR and procalcitonin value) associated with *P. Aeruginosa* and *Klebsiella pneumoniae* colonization of the ascites. This condition highly contraindicated PVS placement.

Few authors [4,6] reported effective treatment of hepatic lymphorrhea using local intraperitoneal administration of OK-432 which is a heat and penicillin-treated preparation of streptococcus pyogenes A3. It is largely used in the treatment of lymphatic malformations and for malignant ascites. OK-432 promotes

**Table 1**  
Characteristics, therapies and clinical outcome of the patients with hepatic lymphorrhhea.

Case	Author	Age/sex	Operation	Treatment	Time to complete resolution
1	Miyagawa, 1983	65/M	TG	Surgical ligation	13
2	Nakashima, 1985	58/M	DG	Surgical ligation + sclerotherapy	30
3	Nakano, 1987	49/M	TG	Surgical ligation	14
4	Kawata, 1989	52/M	DG	Surgical ligation + fibrin glue + sclerotherapy	37
5	Umehara, 1989	59/M	TG	Surgical ligation	28
6	Kaneko, 1991	44/M	DG	Surgical ligation + PVS	30
7	Imai, 1992	34/M	TG	Reoperation + sclerotherapy	7
8	Shimizu, 1992	62/M	DG	Surgical ligation	30
9	Ota, 1993	70/M	DG	Surgical ligation + fibrin glue	50
10	Mitsuno, 1993	42/M	DG	PVS	ND
11	Kawahira, 1994	58/M	DG	Surgical ligation + fibrin glue + OK-432 sclerotherapy	10
12	Matsumoto, 1995	44/M	DG	Re-re-surgical ligation + fibrin glue	14
13	Tanaka, 1998	49/M	DG	Surgical ligation + fibrin glue + OK-432 sclerotherapy	12
14	Tanaka, 2004	66/M	TG	Surgical ligation + fibrin glue + OK-432 sclerotherapy	67
15	Inoue, 2011	73/M	HR	PVS	12

TG: total gastrectomy; DG: distal gastrectomy; PVS: peritoneovenous shunt; HR: hepatic resection ND: not described.

reabsorption of ascitic fluid increasing capillary permeability and inducing local inflammatory action which causes occlusion of injured lymphatics. We have no experience of this kind of sclerotherapy and the ethics Committee didn't authorize the use of this kind of drugs.

Our strategy was to open and close intermittently the abdominal drainage. At first this option caused discomfort to the patient but allowed a gradual reduction of spontaneous lymphorrhhea until its resolution.

**4. Conclusion**

In our experience, this approach increases intra abdominal pressure and promotes trans-peritoneal reabsorption. This solution may be useful when the other options mentioned by literature prove to be ineffective or contraindicated.

**Conflict of interest**

None.

**Funding**

None.

**Ethical approval**

Not needed.

**Consent**

Written inform consent was obtained from the patient for publications of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

**Author contribution**

Michele Bartoli was the principal investigator and wrote the paper. Stefano Maria Giulini, Nazario Portolani and Gian Luca Baiocchi treated the patient. All authors critically reviewed the paper and approved the final version.

**Guarantor**

Michele Bartoli.

**Acknowledgements**

None.

**References**

- [1] H. Ota, T. Miyazawa, I. Hiizu, et al., A case report of intractable ascites due to hepatic lymphorrhhea from hepatoduodenal ligament after radical gastrectomy for gastric cancer, *Jpn. J. Gastroenterol. Surg.* 26 (1993) 1115–1119.
- [2] Y. Kawahira, K. Nakao, M. Nakahara, et al., A case of intractable hepatic lymphorrhhea after gastrectomy for gastric cancer, *Jpn. J. Gastroenterol. Surg.* 27 (1994) 117–120.
- [3] D. Guez, B.S. Gregory, J. Nadolski, et al., Transhepatic lymphatic embolization of intractable hepatic lymphorrhhea, *J. Vasc. Interv. Radiol.* 25 (1) (2014) 149–150.
- [4] K. Tanaka, Y. Ohmori, Y. Mohri, et al., Successful treatment of refractory hepatic lymphorrhhea after gastrectomy for early gastric cancer, using surgical ligation and subsequent OK-432 (Picibanil) sclerotherapy, *Gastric Cancer* 7 (2004) 117–121.
- [5] Y. Inoue, M. Hayashi, F. Hirokawa, et al., Peritoneovenous shunt for intractable ascites due to hepatic lymphorrhhea after hepatectomy, *World J. Gastrointest. Surg.* 3 (1) (2011) 16–20.
- [6] Y. Inaba, Y. Arai, K. Matsueda, et al., Intractable massive ascites following radical gastrectomy, treatment with local intraperitoneal administration of OK-432 using a unified CT and fluoroscopy system, *Australas. Radiol.* 47 (2003) 465–467.

**Open Access**

This article is published Open Access at [sciedirect.com](http://sciedirect.com). It is distributed under the [IJSCR Supplemental terms and conditions](#), which permits unrestricted non commercial use, distribution, and reproduction in any medium, provided the original authors and source are credited.