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PRIMARY LONG-STANDING CHYLOUS REFLUX INTO SKIN: Combined Operative and Non-operative Treatment

A. Zelikovski, M. Haddad, E. Stelman, A. Koren, J. Loewinger

Department of Vascular Surgery, Beilinson Medical Center, Petah Tiqva, Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

ABSTRACT

Chylous discharge into cavities or skin is rare and usually arises after operations in the vicinity of the cisterna chyli (secondary chylous reflux). When chylous discharge is primary, however, chylous reflux has usually been present since birth probably as a consequence of obstruction to lymph flow at or above the cisterna chyli. We report a patient who had had chylous reflux into lacerated skin in a lymphedematous thigh for more than 30 years. The condition was successfully managed using both operative and non-operative methods.

CASE REPORT

A 56 year-old man had long-standing left leg lymphedema and prominent milky discharge from several areas of a chronic skin laceration in the posterior aspect of the thigh. The condition had first appeared 30 years previously. Twenty years earlier, he had undergone excision of the skin of the involved thigh and skin grafting without success. The profuse discharge had persisted, requiring him to wrap towels around the thigh and to change them 8-10 times daily. He also noted an augmentation in the quantity of the milky discharge after meals.

On examination, there was severe lymphedema of the left leg with a skin laceration on the upper posterior aspect of the thigh. From this site, milky fluid discharged at a rate of 100-150 cc per hour. The rest of the physical examination was unremarkable.

Isotopic lymphography demonstrated stagnation of the tracer in the left foot with no passage of the radiopharmaceutical above the knee. Computer tomography of the abdomen revealed dilation of the left iliac vein with fatlike infiltration around the iliopsoas muscle and the femoral neck *(Fig. 1)*. No pathological lymph glands were detected in the pelvis. After diagnosis of chylous reflux into the skin of the left thigh was established, the patient was operated upon using a left retroperitoneal approach. Huge lymphatic collectors were seen parallel to and surrounding the iliac blood vessels, exiting from behind the aortic bifurcation *(Fig. 2)*. Meticulous ligation of all visible megalymphatics was done up to the bifurcation of the aorta on the left side. Chylous leakage from the lacerated skin ceased completely until the third postoperative day when it reappeared, though of a lesser magnitude. Because it persisted more than 7 days, reoperation was performed. The skin of the thigh where the chyle was leaking was excised and small lymphatics that emerged from the lacerated area were oversewn.

The chylous discharge again ceased for two days but then redeveloped, this time with swelling of the scrotum. We assumed that there remained several insufficient lymphatics which drained into the genitalia. After total parenteral hyperalimentation and total fasting, the scrotal swelling rapidly

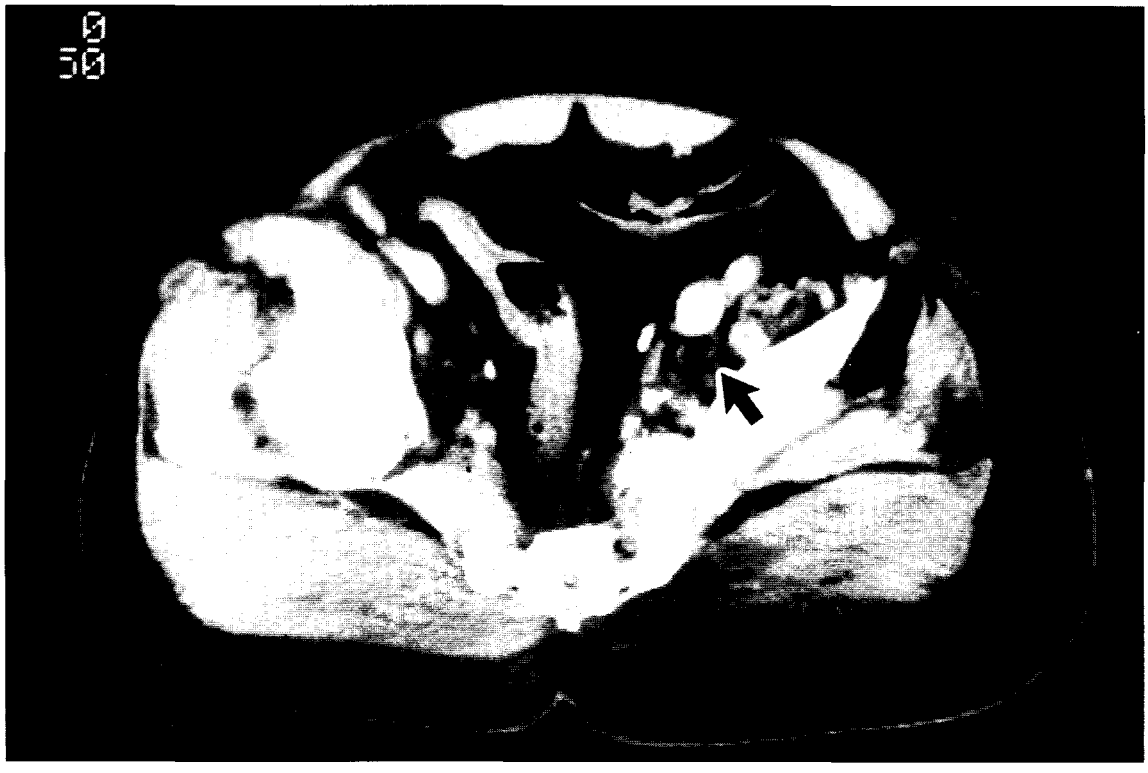

Fig. 1. Fat-like infiltration (arrow) around the iliopsoas muscle.

diminished and the chylous discharge gradually decreased; it completely ceased after 20 days of parenteral hyperalimentation and administration of octreotide (a somatostatin analog) at 0.1 mg subcutaneously twice a day. The patient was then given a low fat diet and discharged 33 days after the first operation with continued treatment with octreotide (0.1 mg x 2 per day subcutaneouly for 30 days).

Six months later the patient was free of chylous discharge and the wounds were healed. He continues to use an elastic support for the lymphedematous leg.

DISCUSSION

Chylous reflux is a rare condition. It occurs when lymphatic back pressure is high causing microvessel bursting into adjacent cavities (e.g., peritoneum, urinary bladder, vagina, thorax) or rupture into the skin. There is often a history of operation, irradiation or tumor to explain secondary chylous discharge (1-3) but understanding of the primary disorder remains perplexing (4). The aim of treatment is to stop the chylous reflux by diminishing retrograde flow of lymph emanating from intestinal lacteals and by centralizing leakage from disrupted discharging lymphatics.

Our patient was successfully managed using the following combined operative and non-operative modalities: 1) ligation of the retroperitoneal megalymphatics through which chyle refluxed and burst into the thigh skin, and ligation of dermal discharging lymphatics; 2) parenteral hyperalimentation and administration of octreotide to minimize chyle production (5).

The pathophysiology of this patient's condition remains unclear because he never

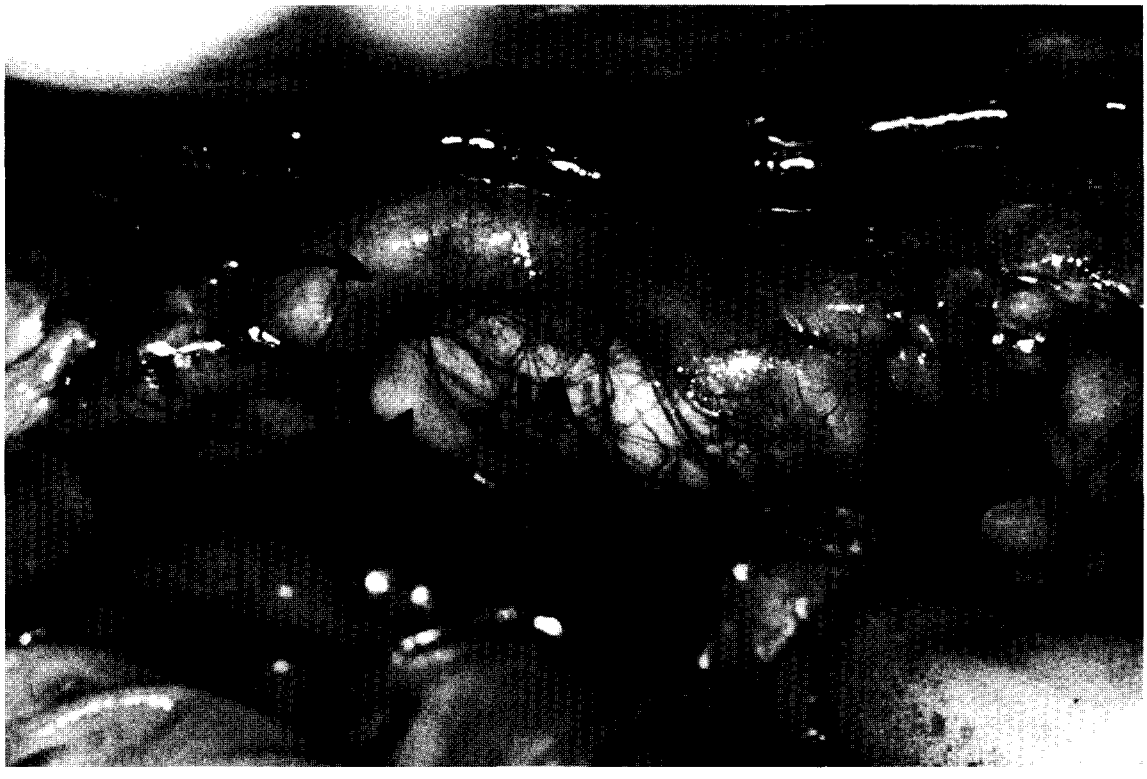

Fig. 2. Huge megalymphatics (arrows) containing chyle surrounding the left iliac artery (IA).

underwent abdominal surgery or sustained visceral trauma. We speculate that congenital lymphatic insufficiency was responsible for the chylous reflux syndrome.

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Prof. A. Zelikovski, M.D.
Head, Department of Vascular Surgery
Beilinson Medical Center
Petah Tiqva, 49100
Israel

Editor's Note:

An alternative approach has recently been demonstrated in management of similar patients using percutaneous sclerotherapy of ectatic retroperitoneal lymphatic collectors under computed tomographic guidance [Molitch, HI, EC Unger, CL Witte, E vanSonnenberg, Radiology 194 (1995), 343]. With monitoring by lymphangioscintigraphy and computer tomographic imaging, percutaneous sclerotherapy of incompetent lymphatic collectors can be carried out and serially repeated as necessary and laparotomy and conventional operative and reoperative ligation and oversewing of these lymphatics circumvented altogether.