

## BILATERAL GROIN ADENOLYMPHOCELES: AN UNUSUAL PRESENTATION OF CHYLOUS REFLUX

A.E. Guzmán, A. Silva

Department of Phlebology and Lymphology, Italian Hospital, Mendoza, Argentina

### ABSTRACT

*We report an unusual presentation of a young man with bilateral groin lymph nodal adenolymphoceles and right leg lymphedema as a manifestation of intestinal lymphangiectasia. Chylous reflux was supported by conventional and isotopic lymphography as well as by a total lipid test showing delayed triglyceride absorption 24 hours after ingestion of 60 gm of butter. After excision of groin masses in conjunction with dietary control (short-medium chain triglycerides), manual massage, pneumatic compression, and long-term wearing of a low stretch elastic garments he remains well.*

Chylous reflux is an unusual clinical presentation. Sometimes previous surgery, trauma, or infection with filarial nematodes gives rise to its occurrence. Dilatation of retroperitoneal/mesenteric lymphatics is characteristic, sometimes referred to as lymphatic varices or lymphangiectasia (1). If dilatation involves lymphatics of the nodes, it is termed adenolymphocele and an infrequent presentation but occasionally seen unilaterally in the groin (2). In the patient presented here, adenolymphoceles developed in both groins and was accompanied by chylous reflux (probably congenital) associated with primary lymphedema of the right leg. Satisfactory treatment consisted of resection, hygienic-dietary regimen, elastic compression, and manual manipulation.

Chylous reflux (also termed intestinal lymphangiectasia) is an uncommon condition characterized by accumulation of chyle



*Fig. 1. 24-year old man showing bilateral groin lymphoceles with moderate right leg lymphedema.*

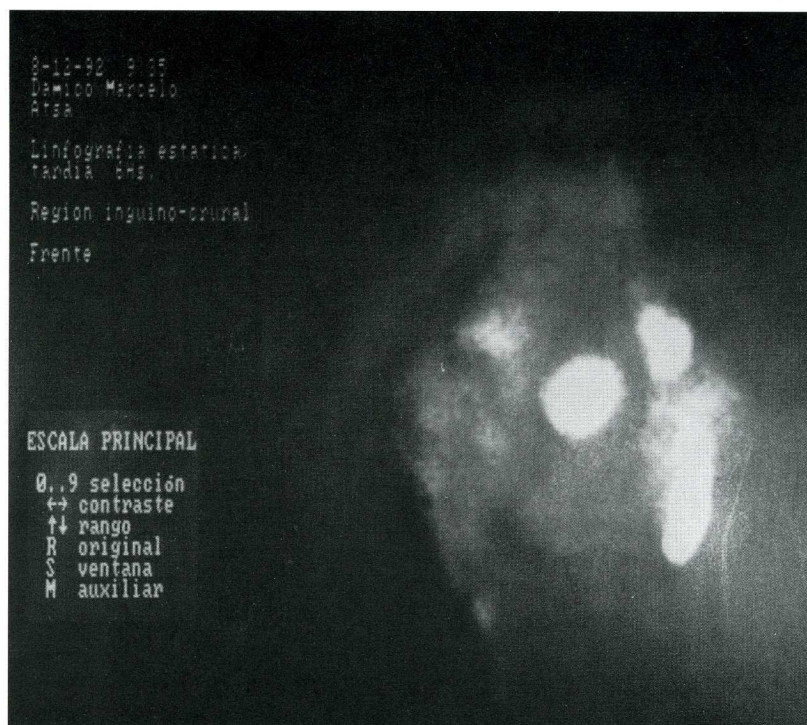


Fig. 2. Radioisotope lymphogram. Note the high uptake of radiocolloid in the groin masses in patient shown in Fig. 1.

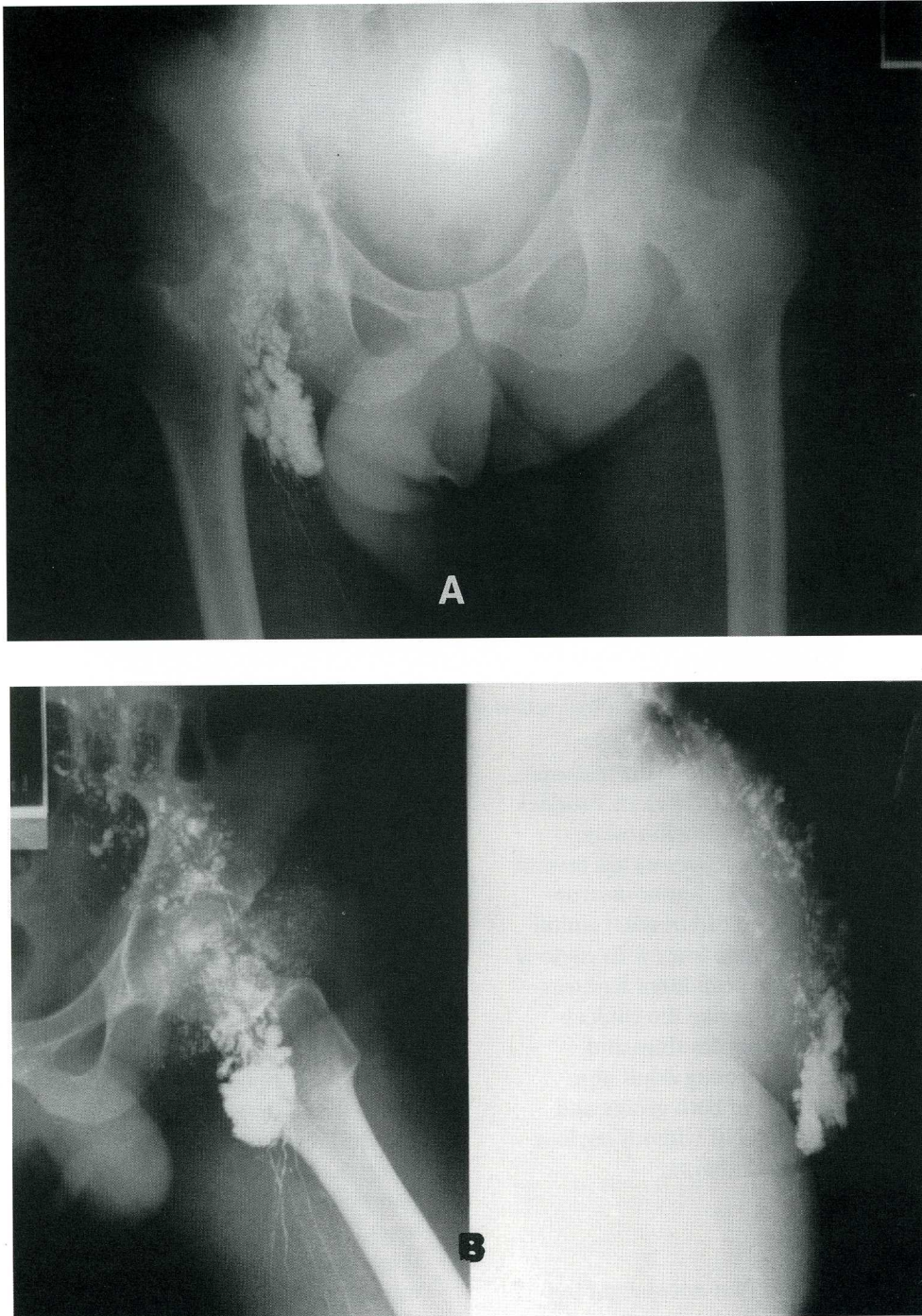
(intestinal lacteal lymph) within the abdomen (ascites) or external leakage (chylous fistulae) often in conjunction with protein-losing enteropathy (3,4). Etiologic factors are several including congenital origin, tumor obstruction (e.g., lymphoma), and filariasis. Genital and leg lymphedema, sometimes with chylous vesicles on the skin, are prominent features.

In this report we describe the clinical manifestations, laboratory findings and treatment outcome in a young adult man with adenolymphoceles of both groins and peripheral lymphedema but without chylous ascites, chyluria, or chylothorax.

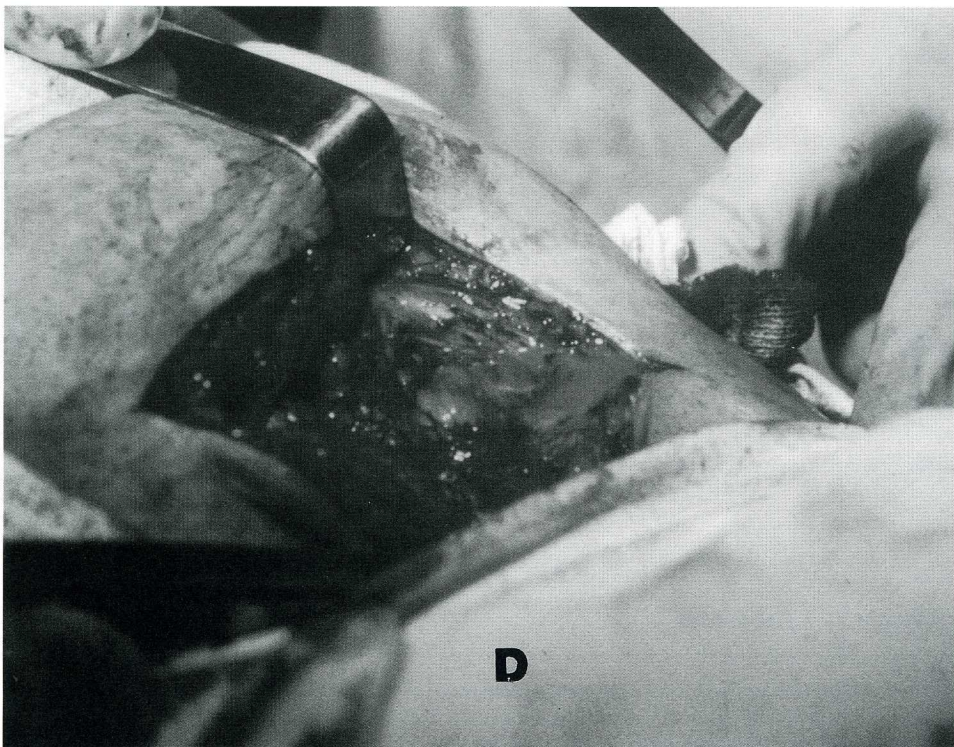
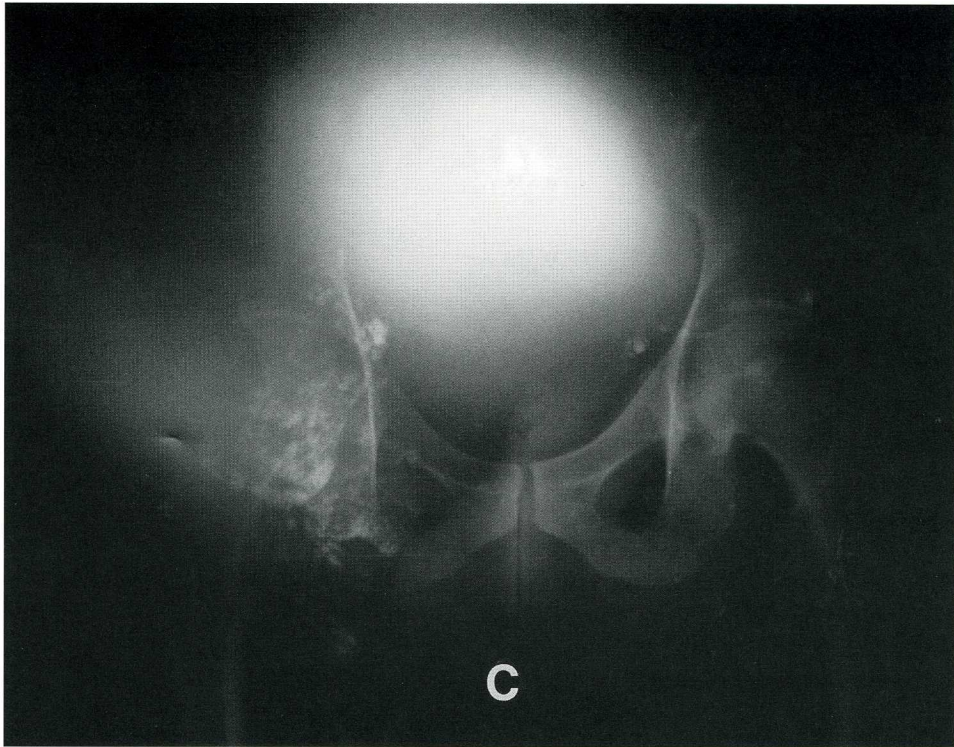
#### CASE REPORT

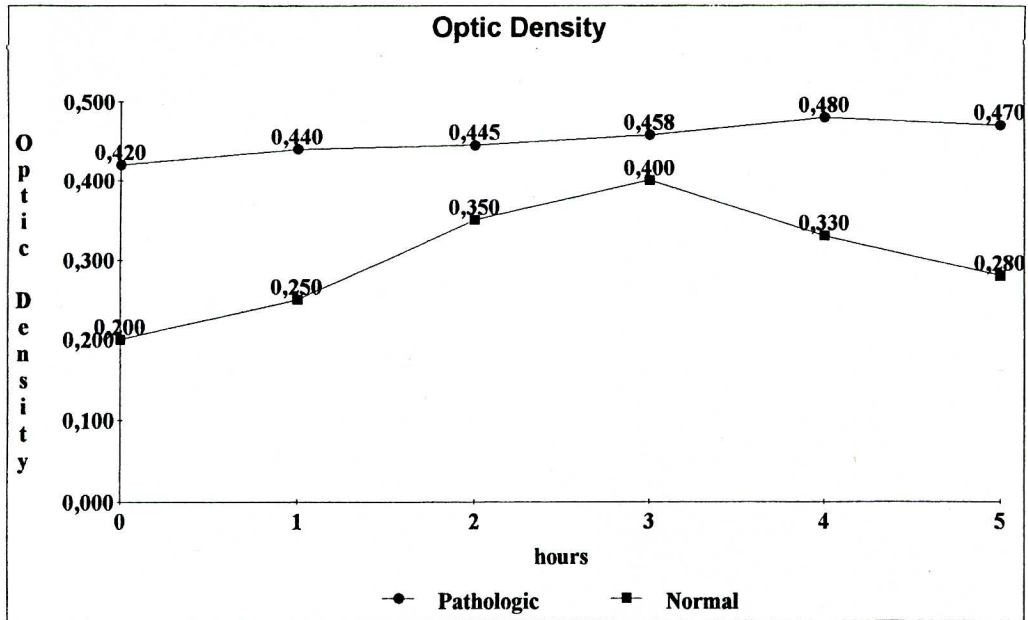
A 24-year old man first noted right leg swelling at 8 years of age. A conventional lymphogram confirmed lymphatic hypoplasia. At 19 years, he noted masses in both groins and when examined 6 months later, the one on the right measured 15x10

cm. Both masses were soft, easily compressed, and painless (Fig. 1). Phlebography was unremarkable. Isotope lymphography ( $^{99m}\text{Tc}$  sulfa colloid) revealed reduced transport in the right leg, normal uptake in the left leg with dramatic uptake in the enlarged regional nodes conforming to the inguinal masses (Fig. 2). Conventional lymphography of the left foot (in the right lymphedematous foot, lymphatics were not depicted) showed a paucity of lymphatics but starting in the groin, the contrast medium (ultrafluid lipiodol) formed ectatic puddles extending along the inguinal canal (Fig. 3a,3b). Twenty-four hours later, puddles within the groin mass (adenolymphoceles) with reflux into the thigh and into the opposite retroperitoneum were seen (Fig. 3c). Standard electrophoresis showed normal albumin and globulin levels. A "hyperlipidemia test" revealed markedly delayed long-chain lipid absorption and delayed clearance from the bloodstream as defined by optical density and total lipids in

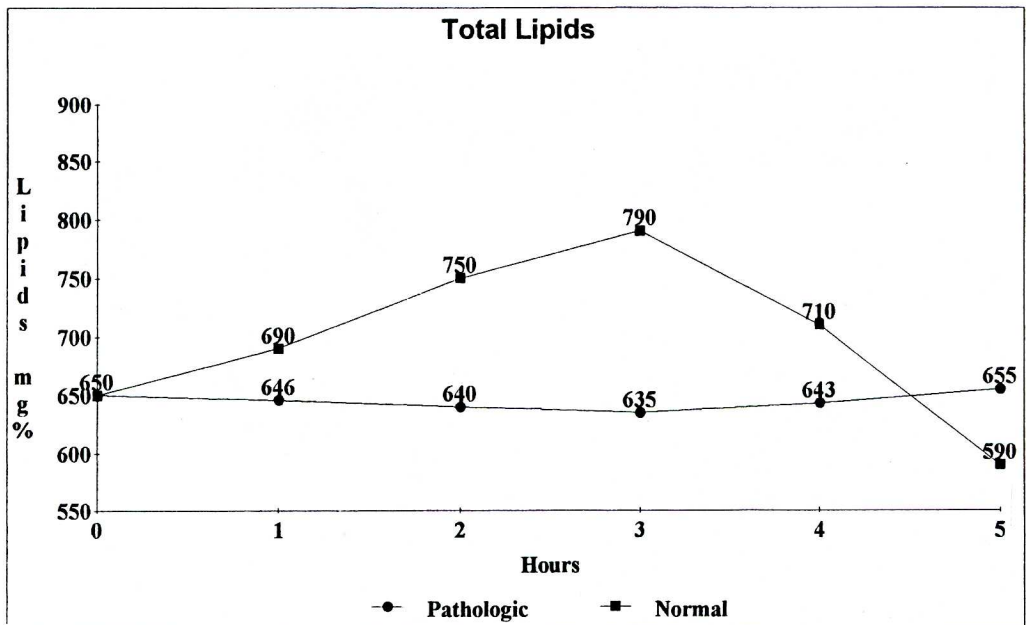


*Fig. 3. Conventional lymphogram (L foot, inverted photographs) showing prominent groin adenolymphocele (A,B) with puddling in the lumbar chain with cross-over into the retroperitoneum (C) (next page). At operation, cystic appearance with blue coloration (patent blue previous instilled into skin of the left thigh) consistent with lymphadenocoele (D) (next page).*



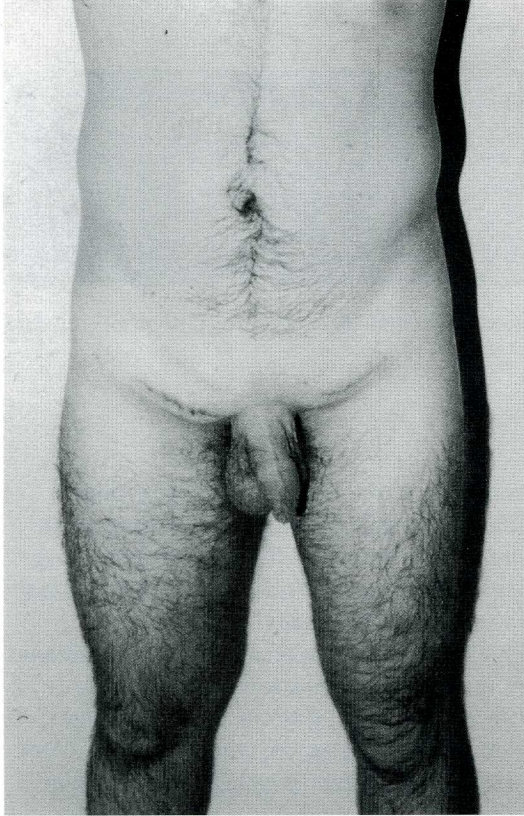


Graph 1 - Optic Density



Graph 2 - Total Lipids

Fig. 4. Total blood lipid absorption curve and optical density of serum in patient (pathologic) compared with normal after ingestion of 60 gm/butter (long-chain triglycerides) 24 hours earlier. Note the slow absorption of total lipids in patient (below) and overall higher optical density (above).



*Fig. 5. 3 years after excision of groin masses and non-operative treatment using diet (short and medium chain triglycerides), manual massage, pneumatic compression, and wearing of elastic garments.*

serial blood samples after ingestion of 60 gms of butter (*Fig. 4*). Fecal fat loss was also high (8.78 g/24 hrs) (normal <6g/24 hrs).

Initially the adenolymphocele of the right groin was excised (L. Pereyra, M.D.) revealing several spongy cavities consistent with lymphangiectasia. The excised specimen revealed fibroadipose tissue with dilated lymphatic vessels infiltrated with mononuclear cells. One year later, the adenolymphocele of the left groin was excised with similar histologic findings.

The patient had a good recovery (*Fig. 5*). Lymphedema of the right leg has continued but with manual manipulation, pneumatic compression, elastic stockings, programmed exercises, and benzopyrones, the edema has remained stable.

## COMMENT

Our patient was unusual in that adenolymphocele of the groins were bilateral in conjunction with chylous reflux, right leg lymphedema and without filariasis. In light of the leg lymphedema documented since childhood, it seems reasonable that the primary disorder was congenital intestinal lymphangiectasia although chylous ascites, chyluria, and/or chylothorax was absent. Support for this diagnosis was by the "hyperlipedema test" (3). In normal individuals, the basal lipid concentration in the blood is 400-900 mg/dl and after long chain fatty intake it increases ~22% at 3 hours and then decreases to a basal level or slightly lower. With abnormal chyloferous vessels (*Fig. 3d*), lipid absorption is diminished with a flattening of the absorption curve and loss of a post-prandial spike at 3 hours. Basal lipid levels are usually normal. The optical density of the serum, on the other hand, is usually higher in those with intestinal lymphangiectasia initially and remains higher after several hours (*Fig. 4*). The findings in our patient were consistent with abnormal chyloferous lymphatics.

Although this condition remains unusual, a combination of surgical excision, compression therapy, dietary control (short-medium chain triglycerides) was successful in managing this young adult male's congenital disorder.

## REFERENCES

1. Olszewski, W: Lymphatic stasis. In: Physiopathology and Treatment. CRC Press, 1991.
2. Pereyra, L: Surgery of lymphedema and of penis-scrotal elephantiasis. Doctoral Thesis. Recife, Brazil, 1985.
3. Jimenez Cossio, JA: Diagnosis and Treatment of Lymphedema. Uriach, 1987.
4. Martorell, F: Angiology. Vascular Diseases. Salvat Editors. 1972.

**Angel E. Guzmán, M.D.**  
**Emilio Civit 356**  
**(5500) - Mendoza, ARGENTINA**