Lymphology 24 (1991) 130-134

THORACIC DUCT CYST OF THE NECK: A Case Report

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ABSTRACT

A thoracic duct cyst was excised from the left side of the neck of a 63-year-old woman. It was 6.5cm in length and 3.5cm in width, and was located behind the common carotid artery and jugular vein. The proximal portion of the thoracic duct with a narrowed lumen was in direct contact with the lateral surface of the cyst and the venous angle. The lower end of the cyst extended into the mediastinum to immediately above the aortic arch; the inferior portion of the thoracic duct descended from the bottom of the cyst into the mediastinum behind the aorta. The cyst was unilocular with a fluid content high in triglyceride (3350mg/dl). Its wall was muscular and permeated with lymphatic vessels.

A cyst of the thoracic duct is an uncommon phenomenon with most occurring in the mediastinum (1-15). We describe an adult woman with a large cyst in the left neck, which based on its anatomical location, histopathological appearance, and chylous fluid was consistent with an origin from the thoracic duct.

CASE HISTORY

A 63-year-old Japanese woman presented with a tumor in the left anterior neck. She had no history of trauma or hepatic dysfunction nor was there evidence of malignancy. Neck echography and computer tomography showed a hypoechoic cystic mass with smooth surface behind the left common carotid artery and jugular vein (*Fig. 1A*). The

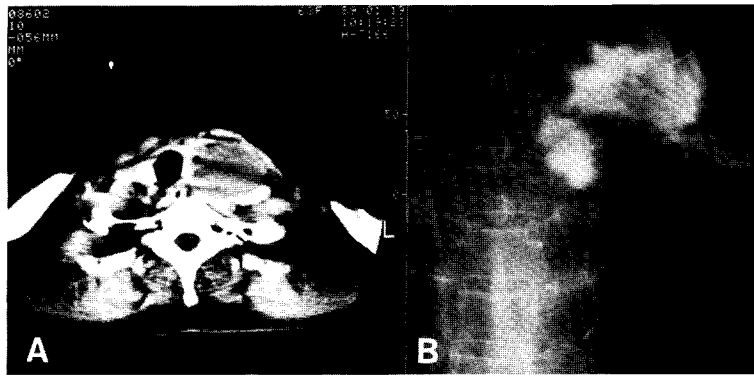

*Fig. 1. **A**, Computer tomography of a large cystic mass (arrowheads) at the left side of the neck; **B**, Large cystic mass filled with contrast medium after percutaneous injectionnote the narrow, irregular thoracic duct inferior and contiguous with the cyst mass.*

cyst was 4cm in diameter and its lower end extended to just above the aortic arch. Fine needle aspiration yielded 50ml of brownish milky fluid, which microscopically contained a small number of lymphocytes and erythrocytes, along with a substance soluble in ether. On biochemical analysis, the fluid was found to contain 3.35g/dl triglyceride, 7.7g/dl total protein, 4.1g/dl albumin, and on agarose gel electrophoresis, high concentrations of chylomicrons and free fatty acid (*Fig. 2*). The serum of the patient contained 99mg/dl triglyceride, 149mg/dl cholesterol and 3.4g/dl albumin.

Histologically, the cyst wall consisted of an inner layer of fibrous connective tissue of varying thickness, a central layer of smooth muscle, and an outer fibrous connective tissue layer (*Fig. 3A*). A few reticulin fibers were observed in and around the muscle layer (*Fig. 3B*). Part of the cyst lacked a central layer of muscle and was rich in granulation tissue and blood vessels. The lumen contained proteinaceous fluid with some red blood cells and foamy macrophages; endothelial cells were not observed on the inner cyst surface. The cyst wall was permeated by lymph ves-

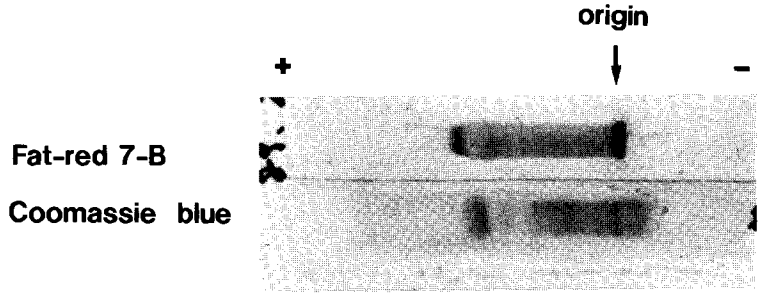

Fig. 2. Agarose gel electrophoresis of fluid obtained from the cyst. Chylomicra are stained positive by Fat red 7B at the starting point (origin) whereas free fatty acid appears as a band moving faster than albumin as shown by Coomassie blue stain.

Although dorsal pedal lymphography failed to show an influx of the contrast media into the cyst via the thoracic duct, percutaneous injection of contrast medium directly into the cyst revealed that the cyst and the thoracic duct were in close contact (*Fig. 1B*). When the cyst was exposed at operation, the thoracic duct was delineated along the lateral surface of the cyst, and joined the central venous system at the "venous angle." In spite of focal adhesions, the cyst was easily removed. The lower portion of the thoracic duct extended from the inferior margin of the cyst into the mediastinum. The excised cyst was 6.5cm in length and 3.5cm in width. On cut section, it was unilocular and filled with chylous fluid.

sels (*Fig. 3C*); the inner surface was covered by endothelial cells which failed to stain for Factor VIII associated antigen. Around the cyst was a focal proliferation of lymph vessels with smooth muscular walls (*Fig. 3D*). The portion of the thoracic duct located between the cyst and the venous angle contained a thickened intima with loose fibrous connective tissue and marked luminal narrowing (*Fig. 4A,B*).

Since excision 18 months ago, she has remained well without ascites, hydrothorax, or peripheral edema.

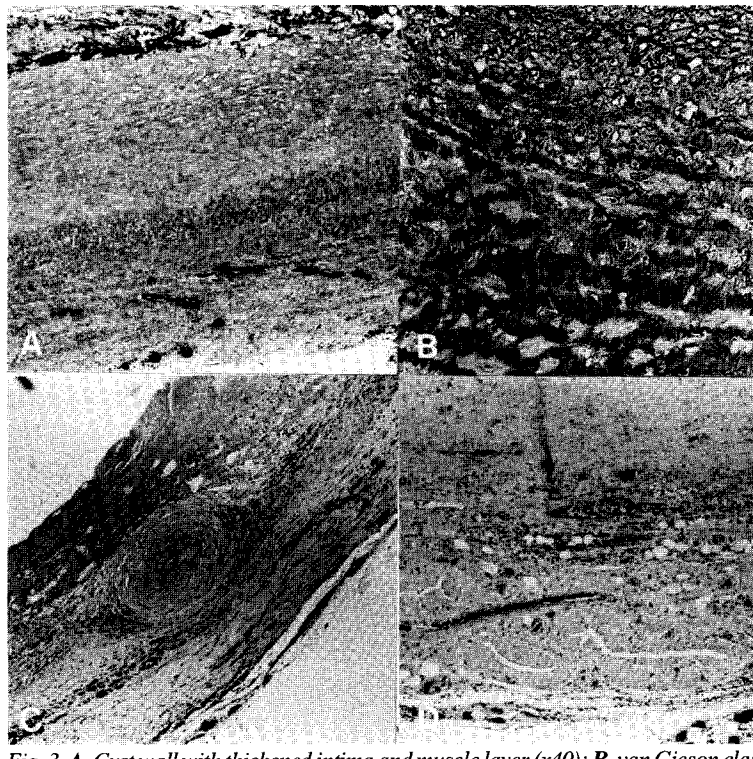

Fig. 3. ***A****, Cyst wall with thickened intima and muscle layer (x40);* ***B****, van Gieson elastic stain showing sparse distribution of reticulin fiber around smooth muscle cells (x100);* ***C****, Lymphatic channels permeating the cyst wall (x40);* ***D****, proliferation of lymphatics around the thoracic duct cyst (x40).*

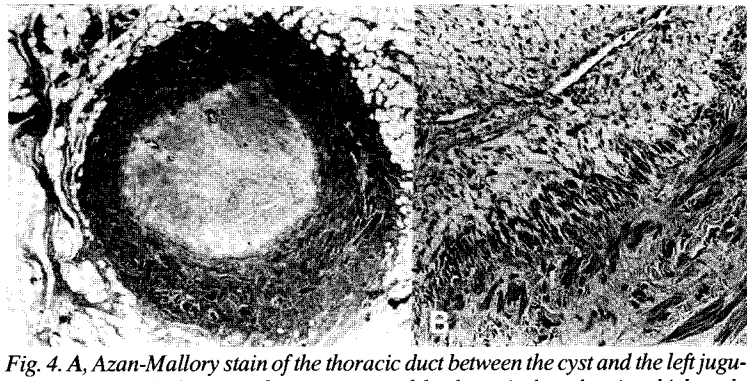

Fig. 4. ***A****, Azan-Mallory stain of the thoracic duct between the cyst and the left jugular angle (x20);* ***B****, hematoxylin-eosin stain of the thoracic duct showing thickened intima with loose fibrous connective tissue (x100).*

COMMENT

Thoracic duct cyst is a rare disorder which usually occurs in the mediastinum. To our knowledge, only 15 mediastinal thoracic duct cysts have been reported between 1950 and 1990 (age range 17 to 76 years; mean age 41 years; male/female7/8) (1-15). The earliest

record of thoracic duct cyst formation is that described by Carbone in 1892 (16) and Priesel in 1914 (17). The cervical segment of the thoracic duct is much shorter than its mediastinal component which may account for its infrequency in the neck. Thus far, only two patients heretofore have been described with a cervical thoracic duct cyst (6,15). Our patient's cyst was located in the left neck, contained chylous fluid, and possessed a smooth muscle layer, features consistent with an origin from the thoracic duct.

Persistent jugular lymph sac in the left neck arises as a cystic mass and can be opacified by angiography (18-20). It is unlikely that our patient's cyst was derived from a persistent jugular lymph sac because of the narrowness of the thoracic duct lumen between the cyst and the jugular angle. Differentiation between a parathyroid cyst and a simple lymph cyst from a thoracic duct cyst can be readily made by comparison of the cyst fluids; the first two have clear fluid without chylomicra. Moreover, the wall of the parathyroid cyst contains parathyroid glandular tissue (21). In contrast to these findings, our patient's cyst contained chyle, was permeated with lymphatics, and considerable smooth muscle. The cyst location between the thoracic duct and the jugular venous angle with a narrowed lumen and thickened intima suggests that it developed in response to stenosis or thoracic duct obstruction from "thrombosis" or "sclerosis" within the intima. It has been previously suggested that sclerotic changes in the thoracic duct is a pathogenetic factor in cyst occurrence (3,6,7,9). Nonetheless, the proliferation of lymph vessels around the cyst in our patient raises the possibility of a congenital origin related to narrowing of the proximal portion of the thoracic duct.

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