Case Report

Thoracic duct cyst in supraclavicular region

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SUMMARY

A 28-year-old female attended an outpatient clinic in October, 1989, because of a tumor in the left supraclavicular fossa, detected in a health examination. Following exploratory puncture of the tumor which yielded milky-white fluid, suggesting a cyst in the thoracic duct, she was admitted to our department. The cyst was unilocular measuring about 6 cm in diameter, and the fluid content was chyle-rich in lipids. Lymphography demonstrated a lymphatic structure adjacent to the lesion and scattered lymph vessels on the cyst surface. On November 16 the cyst was resected. A restiform structure was observed between the cyst and the thoracic duct, but the presence or absence of communication was unclear. The histological diagnosis was thoracic duct cyst. Thoracic duct cyst occurring in the cervical region is very rare. Our case may provide useful information as to its pathogenesis and the mode of retention of cyst fluid.

INTRODUCTION

Thoracic duct cyst in the cervical region is extremely rare, and there have been only three reported cases. ¹⁻³ We now report this extremely rare case of cervical thoracic duct cyst in the left supraclavicular fossa, including diagnostical details and its pathogenesis.

CASE REPORT The patient was a 28-year-old female with a cervical mass in the left supraclavicular fossa. She had no past history of trauma in the neck or chest. Her family history was unremarkable. On October 5, 1989, she attended our outpatient department because of a tumor in the left supraclavicular fossa. Imaging techniques suggested a cystic mass. Cytodiagnosis by aspiration puncture yielded milky-white fluid. On November 6, she was admitted to the ward for definitive diagnosis and treatment. The tumor measuring 5.4 x 4.6 cm was oval, well-defined and tense with a smooth surface and rather poor mobility. There was no goitre or cervical lymph node enlargement.

Preoperative examination: Routine blood tests, biochemical data and urine examination showed no abnormalities. The appearance of the fluid aspirated from the cyst was milky-white. Analysis of the fluid revealed triglyceride of 911 mg/dl, chylomicron of 3,155 mg/dl and lactic dehydrogenase (LDH) of 1,026 U (Table).

Ultrasonography revealed a unilocular cyst with increased posterior sounds and a structure with high-luminance sounds in the fluid. Computerized tomography (CT) scanning showed a cystic lesion with a maximum axis of 6 cm at a site posterolateral to the left sternocleidomastoid muscle and anterolateral to the anterior scalenus muscle; infiltration to the left common carotid artery, left subclavian vein, or surrounding organs was absent. Lymphography of the cervical and chest regions 24 hours after infusion showed a lymphatic structure adjacent to the lesion, scattered lymph vessels on the cyst surface, and lymphatic systems in the bilateral supraclavicular fossa and the anterior mediastinum, suggesting lymph regurgitation or collateral vessel formation (Fig. 1). Radiography of the cyst demonstrated a unilocular cyst but no communication with the thoracic duct or other organs (Fig. 2).

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Table

Analysis of the fluid revealed triglyceride 911 mg/dl, chylomicron 3,155 mg/dl and lactic dehydrogenase (LDH) 1,026 U.

Appearance	Fluid of the cyst on admission milkly-white	Normal values for seruum 		
Uric Acid (mg/dl)	6.8	2.5	_	5.4
Blood Urea Nitrogen (mg/dl)	12.9	6.0	_	20.0
Creatinine (mg/dl)	0.7	0.6	_	1.3
Total Cholesterol (mg/dl)	75	150	_	219
HDL* Cholesterol (mg/dl)	4	39	_	93
Free Cholesterol (mg/dl)	15	30	_	60
Lactate Dehydrogenase (U)	1026	230	_	460
Alkaline Phosphatase (U/1)	108	80	_	260
Amylase (U/dl)	211	60	_	200
Free Fatty Acid (mEq/l)	1.13	0.14	_	0.85
Triglyceride (mg/dl)	911	36	_	130
Phosphatide (mg/dl)	140	160	_	260
Phosphatide Fraction				
Lecithin (%)	82.23	66.5		83.1
Sphingomyelin (%)	14.42	0.5	_	24.5
Lysolecithin (%)	3.55	3.1	_	7.9
Lipoprotein Fraction				
LDL (mg/dl)	115	58	_	160
VLDL (mg/dl)	120	0	_	64
Chylomicron (%)	3155	0		
Lipid (mg/dl)	1067	390	_	720
Cytodiagnosis	class II			

^{*} High-density lipoprotein

[†] Low-density lipoprotein

[‡] Very low-density lipoprotein

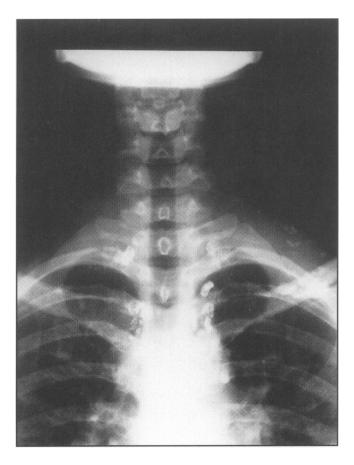


Fig 1. Lymphography of the cervical and chest regions 24 hours after infusion.

Lymphography showed a lymphatic structure toward the lesion, scattered lumph vessels on the cyst surface, and the lymphatic systems in the supraclavicular fossae and the anterior mediastinum, suggesting lymph regurgitation or collateral vessel formation.

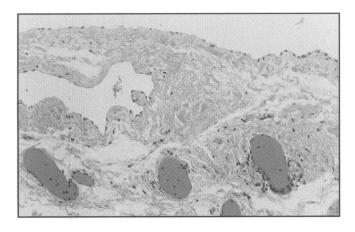


Fig 3. Histopatholpgical findings (Magnification x 100). The cyst was unilocular with an unclear adventitia, lined by a layer of endothelial cells. Smooth muscle bundles were discontinuously observed around the wall of the cyst. There were no findings suggestive of malignancy.



Fig 2. Radiography of the cyst.

Radiography of the cyst demonstrated a unilocular cyst but no communication with the thoracic duct or other organs.

Operative findings: The patient underwent surgery with a preoperative diagnosis of thoracic duct cyst on November 16, 1989. After a collar incision, the platysma and superficial cervical fascia were cut. A cyst containing white fluid, covered with fine lymphatic vessels was observed in the area dorsal-lateral to the sternocleidomastoid muscle. There was no infiltration to the surrounding vasculature or neural system. The cyst was readily dissected. The thoracic duct was observed dorsal to and immediately below the cyst. A restiform structure was present between the thoracic duct and the cyst, but the presence or absence of communication was unclear.

Resected specimen: The tumor was elastic and soft measuring 4.6 x 4.0 x 4.5 cm. There were networks of arterioles, venules and fine lymph vessels on the cyst surface. Granulation-like pieces of tissue were suspended in the cyst fluid. Histopathological findings: The cyst was a unilocular one. It had an unclear adventitia and was lined by a layer of endothelial cells. Smooth

muscle bundles were discontinuously observed around the wall of the cyst. There were no findings suggestive of malignancy. The granulation-like tissues were lymph clots (Fig. 3). Based on these findings, a diagnosis of thoracic duct cyst was made.

DISCUSSION

Well-known cysts in the cervical region include median cervical cyst, lateral cervical cyst and cystic lymphangioma. However, thoracic duct cyst is very rare. In particular, there are only about 20 reported cases of thoracic duct cyst occurring cephalic to the mediastinum. Of them, only three were cysts in the supraclavicular fossa, including one reported in 1965 by Barlow et al. as cystic dilatation of the thoracic duct. The pathogenesis of cyst formation in this area is unknown.

The differentiation of thoracic duct cyst from cystic lymphangioma is most important. In general, cystic lymphangioma is congenital and frequently observed in infants, developing by two years of age in 90% of the cases.⁴ The cyst is multilocular and lined by smooth epithelium and contains serous or yellow transparent fluid.⁵

In the present case, the cyst developed at the age of 28 years and was unilocular with a content fluid of lipid-rich chyle. In addition, lymphography showed fine lymph vessels on the cyst surface. These findings strongly suggested thoracic duct cyst. However, such a pathologic state has been noted when cystic lymphangioma became continuous with the thoracic duct due to secondary changes such as inflammation and trauma.^{6, 8} In our case, secondary changes of cystic lymphangioma were excluded because of the absence of history of inflammation or trauma in the neck, adhesion of the cyst to the surrounding organs at the time of operation, or inflammatory cell infiltration.

The cyst in this patient may have developed by obstruction of the thoracic duct for some reason at a site where it empties into the left subclavian vein, and a cyst arose from a fragile part of the duct. Concerning the mode of retention of cyst fluid, lymphography showed scattered lymph vessels on the cyst surface, and radiography of the cyst showed a unilocular cyst but no communication with the thoracic duct or other organs. Therefore, retention of chyle in the cyst via parietal lymph tissue cannot be excluded.

The general management of asymptomatic thoracic duct cyst is observation of its progress. 8,9 There are no reports of malignant changes in asymptomatic cases. Surgery is indicated in patients with clinical symptoms such as pain or compression of the surrounding tissue. 2, 3, 6-8 Repeated cyst puncture and infusion of a sclerotic agent into the cyst may be undertaken. However, the risk of infection increases in the former, and there are no precedent cases in the latter. Further evaluation of this method is needed.

REFERENCES

- Kolbenstvedt A, Aanesen J. Cystic dilatation of the thoracic duct presenting as a supraclavicular mass. Br J Radiol 1986; 59: 1228-9.
- 2. Wax M K, Treloar M E. Thoracic duct cyst: an unusual supraclavicular mass. *Head Neck* 1992; **14**: 502-5.
- 3. Barlow D, Gracey L. Cystic dilatation of the thoracic duct. *Br J Clin Pract* 1965; **19**: 101-2.
- 4. Feutz E P, Yune H Y, Mandelbaum I, Brashear R E. Intrathoracic cyst hygroma. a report of 3 cases. *Radiology* 1973; **61**: 61-6.
- 5. Willis R A, ed. Pathology of tumours. 3rd ed. London: Butterworth, Co, 1960: 714-6.
- 6. Gowar F J S. Mediastinal thoracic duct cyst. *Thorax* 1978; **33**: 800-2.
- 7. Emerson G L. Supradiaphragmatic thoracic-duct cyst. an unusual mediastinal tumor. *N Engl J Med* 1950; **242**: 575-8.
- 8. Hori S, Harada K, Morimoto S, Uchida H, Okumura K. Lymphangiographic demonstration of thoracic duct cyst. *Chest* 1980; **78**: 652-4.
- 9. Morettin LB, Allen TE. Thoracic duct cyst: diagnosis with needle aspiration. *Radiology* 1986; **161**: 437-8.