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Review Article

Spontaneous Cyst of the Cervical Segment of the Thoracic Duct: Literature Review

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Abstract

Objective: Cervical cysts are common pathologies in Otorhinolaryngology head and neck surgery. Cysts of the cervical segment of the thoracic duct are poorly understood, but require very specific treatment, as there is a risk of complications. In this work, we outline the particularities of this pathology and the debate surrounding its management.

Materials and Methods: Personal experience of one case and a literature review.

Results: Thirty-four cases of cervical cysts of the thoracic duct have been reported. They are characterized by dilation of the terminal portion of the thoracic duct, just above the left subclavian jugular confluence or in one of the two vessels of which it is composed. They are typically asymptomatic and discovered by chance. Diagnosis of these cysts is based on MRI, perioperative observation (cysts that communicate with the thoracic duct) and anatomopathologicial examination (vascular wall of the cyst). Their treatment remains controversial. Surgical excision becomes an option if the cysts are symptomatic, unsightly or if they are at risk of rupture. Identification and ligation of the thoracic duct are then essential in order to prevent lymphorrhoea.

Conclusion: Cervical cysts of the thoracic duct are benign but must be recognized by the ENT surgeon in order to avoid iatrogenic complications.

INTRODUCTION

Cervical thoracic duct cysts are rather rare. Before 2015, 34 cases had been reported in the literature since the first description in 1964 [1-3]. We report the case of an asymptomatic 61-year-old female patient in whom a Doppler ultrasonography of the supra-aortic trunks fortuitously revealed a cyst in the left supraclavicular cavity. The diagnosis of cervical thoracic duct cyst was suspected intraoperatively and confirmed by histological examination. We describe the features, investigations, diagnosis and treatments of these cysts.

A 61-year-old woman received a Doppler of the supra-aortic trunks as part of follow-up for a cardiac valvulopathy revealing a mass in the left supraclavicular region. It was anechoic, comprised of a thin 50-mm wall on the major axis, and without Doppler vascularization.

The patient was asymptomatic, had a smoking history of 40 pack-years and was not a chronic alcohol drinker. She reported no history of trauma or surgery. Questioning and clinical examination revealed asthma, night-time sweating and a large swelling in the left supraclavicular cavity that was firm, homogeneous, mobile and painless. There was no vascular murmur. Palpation of the other lymph nodes, the rest of the physical examination and standard laboratory tests were normal.

The cervicothoracic CT scan (Figures 1,2) evidenced two adjoined oval shapes without fat at the interface and of different densities. The bulkier measured 34 mm on its long axis and was hypodense (14 HU), while the second had soft tissue density (40 HU) and measured 25 mm. Thoracic slices were unremarkable.

The radiologist concluded that they were adenomegalies.

The patient was referred to the hematology department for management of these cervical lymphadenopathies. A PET scan did not reveal any hyper-metabolic site.

Surgical excision was performed. It revealed a cystic mass beneath the clavicular head of the sternoclidomastoid muscle and behind the clavicle. It was encapsulated, well-delimited, adherent to the adjacent tissues and supplied by the thoracic duct. Careful dissection and ligation of the thoracic duct were performed. Dissection was continued downwards to the subclavian vein, thus allowing complete excision of the cyst. One
drain was set up, the postoperative course was uneventful and the patient returned home two days later.

Macroscopic pathologic examination of the excised tissue revealed a thin-walled bilocular cyst measuring 50 x 20 x 15 mm that contained serous light yellow matter. The wall was fibromuscular and smooth, corresponding to a vascular wall with interstitial fibrosis (Figure 3). The intima was lined with fibrinous exudate containing small lymphocytes. The endothelium appeared attenuated. Immunohistochemical examination confirmed the presence of a media, containing actin + smooth muscle cells. Immunostaining with anti D2-40 antibody did not reveal the native endothelial lining, which was completely bare owing to intra-cystic alterations. The diagnosis of a cervical thoracic duct cyst was made after clinicopathological correlation.

**DISCUSSION AND CONCLUSION**

The thoracic duct is the largest of the lymph vessels in the body [4,5] (Figure 4) and is 45 cm long and 5 mm in diameter in adults. It also collects most of the lymph in the body as it drains the fluid from the sub diaphragmatic region and the left half of the supradiaphragmatic region. It starts between T11 and L2 from the confluence of the left and right lumbar lymphatic trunks and the intestinal trunk. It forms a bulge at this level called the “cisterna chyli” which consists of three segments. The abdominal segment is pre-vertebral and retro-aortic. The thoracic duct passes through the diaphragm by the aortic hiatus. Its thoracic segment describes an upward path in the posterior mediastinum. In its cervical segment, the thoracic duct passes behind the common carotid artery, the internal jugular vein and the vagus nerve, and in front of the vertebral artery and the sympathetic trunk. It rises to C7 forming a concave curve downwards, forwards and to the left, known as the “arch of the thoracic duct”, then crosses the subclavian artery before joining the posterior surface of the left subclavian jugular confluence or one of the two vessels of which it is composed [6-8].

Histologically, the wall of the thoracic duct is composed of an endothelial inner lining (intima), a thin layer of smooth muscle (media) and an external connective tissue (adventitia) which is adherent to the adjacent tissues (Figure 3), making dissection difficult. Injury to the thoracic duct during neck dissection may occur (2%) [9,10], mainly due to the high variability of its anatomical terminations [9,11].

Thoracic duct cysts are uncommon and are mainly located in the mediastinum. Cervical locations are the most rare [3,12] with only 34 cases in the literature [3].

On a histological level, thoracic duct cysts have a thin vascular wall. The internal surface of the cyst is lined by a type of lymphatic endothelium that can be thinned or abraded by means of intra-cystic hemodynamic forces. With immunohistochemistry, the endothelium is positive for the vascular markers CD31, CD34 and factor VIII, as well as for D2-40 and LYVE-1, the specific markers of the lymphatic endothelium [13,14]. Thoracic duct cysts contain lymph. Their puncture reveals a milky [15] or a light yellow fluid, depending on their protein and triglyceride content [13]. Cytological analysis reveals the contents to be mainly lymphocytes (90-95%) of which the vast majority are of the T type [2,16] and there are no polynuclear cells or macrophages [15,17].

Cervical thoracic duct cysts are often asymptomatic [10] and become apparent by gradual swelling of the left supraclavicular region. Pain is sometimes a preliminary symptom. Compressive symptoms such as dyspnea, dysphagia, dysphonia, and coughing have also been reported [18].

The diagnosis of a cervical thoracic duct cyst is suspected on clinical and radiological criteria, after ruling out the more common differential diagnoses of cervical cyst such as branchial cyst, cystic lymph node metastasis of squamous cell carcinoma, lymphangioma, hygroma, thymic cyst, parathyroid cyst and thyroid nodule [14,15,18].

Ultrasoundography reveals an anechoic mass and confirms the cystic nature of the lesion. CT scan demonstrates its characteristics (size, location, intrathoracic extension, boundaries) and relationship with the adjacent structures, and is essential in the preoperative work-up for guiding the surgical procedure [18]. Multiplanar reconstructions demonstrate the passage between the cyst and the thoracic duct, thus confirming its origin. We report here (Figure 1,2), the most demonstrative example ever published in the literature, with evidence of this communication in several planes. Fine-needle aspiration to obtain lymph may be coupled with a retrograde injection of contrast product to demonstrate the passage [11]. Although some authors argue that lymphangiography is the gold standard for revealing thoracic duct abnormalities [12,18,19], the current performance of MRI provides sufficient detail to make the correct diagnosis [14]. MRI reveals a hypo intense or isointense structure on T1-weighted images and a hyper intense structure on T2 images. Gadolinium may be used to differentiate the lymph channels from the small veins [13] and is the most efficient examination for determining the borders of the lesion [20]. Lymphoscintigraphy is also of assistance as it reveals an accumulation of contrast product at the level of the cyst.

The final diagnosis is clinical and pathological, as confirmed by the correlation between intraoperative findings (cystic lesion...
developing at the expense of, and in communication with, the thoracic duct) and the pathologic examination (confirming the vascular organization of the cyst wall) [13,21].

The pathogenesis of this lesion remains unknown although several mechanisms have been proposed: a) congenital weakness in the vessel wall, b) an acquired degenerative process due to inflammation, infection, trauma or atherosclerosis, or c) upstream dilation of an obstacle to lymphatic drainage that is located in the angle formed by the internal jugular vein and the subclavian vein. Surgery and external trauma [2,22] are classically considered as factors promoting the onset of a thoracic duct cyst.

The management of such cysts remains controversial. Watch-and-wait monitoring can be offered to asymptomatic patients. Several conservative treatments have been described, including a low-fat diet, fine needle aspiration of the cyst or the injection of sclerosing agents such as tetracycline, povidone iodine, sodium tetradecyl sulfate and OK-432 [12,18,23]. However, their efficacy is limited. Surgery should be considered only when the cyst is large (esthetic issues, risk of rupture) [16,18] or symptomatic (pain, compression) [12]. In such cases, the cyst is excised and the thoracic duct is ligated to avoid the occurrence of lymphorrhoea. Ligation has no functional effect on the lymphatic system owing to the existence of many collateral channels that terminate in the intercostal, lumbar and azygos veins [10,13] (Figure 4). No recurrence of the lesion after surgery has been reported to date.

Recently, some authors reported the first case of an alternative method of management performing a cyst venous anastomosis for decompression [24].

REFERENCES


