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Management of cervical thoracic duct cyst with cyst-venous anastomosis

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ABSTRACT

INTRODUCTION: Cervical thoracic duct cyst (CTDC) is a rare cause of lateral neck mass. Surgical excision with ligation of the cervical thoracic duct is the current standard for definitive management with symptomatic patients. We report the first case of an alternative method of management performing a cyst venous anastomosis for decompression.

PRESENTATION OF CASE: A 77 year old female presented with a six month history of left arm pain, swelling and a left-sided cystic neck mass. She was treated with cyst-venous anastomosis between the cyst wall and the left internal jugular vein. At two year follow-up, she has had resolution of pain and no recurrence of the mass.

DISCUSSION: Many potential etiologies have been proposed for CTDC, though surgical management of this rare problem has consistently required cyst excision and thoracic duct ligation. Few innovative modes of therapy have been developed to address this problem in a less invasive manor. Maintaining a more natural thoracic duct anatomy decreases the likely of complications associated with duct ligation.

CONCLUSION: Cyst-venous anastomosis for the management of CTDC provides an effective, novel form of treatment which maintains the integrity of the thoracic duct and avoids potential complications associated with duct ligation.

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1. Introduction

Thoracic duct cysts represent a rare clinical problem with a relatively unclear etiology. The first reported case of thoracic duct cyst was in 1964 by Steinberg et al.¹ Since that time, only 32 cases have been reported in the literature with the largest case series consisting of only 5 patients.² The most common approach to managing these lesions is surgical excision with ligation of the thoracic duct. Few alternatives to surgical excision have been reported in the literature. We present the case of a 75 year-old female with a cervical thoracic duct cyst who was successfully treated by cyst-venous anastomosis. To our knowledge, this is the first reported case of successful cyst-venous anastomosis for the treatment of spontaneous cervical thoracic duct cyst (CTDC).

2. Presentation of case

A 75-year old female presented with a four month history of left neck mass and left arm swelling. She reported no history of trauma

or head and neck surgery. On examination she had edema of the left arm extending from the shoulder to the hand. Neurologic examination revealed no focal motor or sensory deficits. Palpation of the left neck revealed a firm, mobile mass adjacent to the left lobe of the thyroid. Jugular venous distention was noted on the left side distal to the mass, but there was no evidence of venous thrombosis. CT and MRI of the neck and upper extremity were reviewed revealing a cystic mass measuring 3.1 cm × 2.8 cm × 1.8 cm which was not in continuity with the thyroid (Fig. 1). The mass appeared to be medial to the carotid artery and jugular vein in the region of the cervical thoracic duct. Ultrasound confirmed the cystic nature of the lesion without solid intramural components and an FNA was performed showing benign cytology.

Due to concern that the patient's symptoms were caused by vascular or lymphatic compression by the mass she was scheduled for surgical excision. A transverse cervical incision was made over the left sternocleidomastoid muscle (SCM). Subplatysmal flaps were raised and the SCM was retracted laterally to provide exposure. The carotid artery was displaced laterally by the mass which was situated between the artery and internal jugular vein (Fig. 2). The mass appeared to emanate from the thoracic duct and several small lymphoid channels were identified emptying into the mass. It was not adherent to adjacent tissues and easily dissected free from the artery and vein. Given the benign appearance of the

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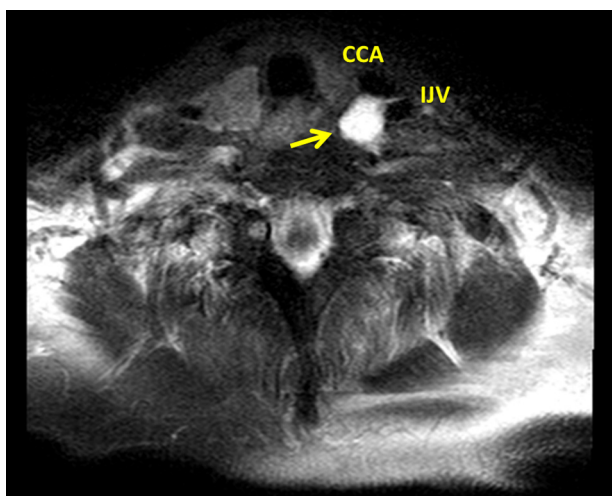


Fig. 1. Pre-operative MRI of cystic neck mass. The image is a T2 weighted MRI showing the left-sided cystic neck mass (arrow) located posterior to the common carotid artery (CCA) and internal jugular vein (IJV).

mass, the decision was made to create a lymphovascular shunt. It was felt that this approach would preserve lymphatic function and maintain a more natural anatomic configuration than excision and duct ligation. Proximal and distal control of the internal jugular (IJ) vein was obtained and a Satinsky clamp was placed on the cystic mass. The mass and the IJ were opened and a side-to-side anastomosis was made with 7-0 Prolene suture. Immediate cyst decompression was achieved and there was no evidence of leakage from the anastomosis. A 7 mm drain was left in place and the skin was closed in a layered fashion. The drain was removed at 1 week and at 1 month follow-up the patient noted improvement of both swelling and pain in her left arm. On follow-up at two years the patient has had no mass recurrence, but left arm swelling has persisted to a lesser degree.

3. Discussion

The pathogenesis of CTDC is not well defined, but many potential etiologies have been proposed. Lymphatic cysts of the neck are known to develop as a complication of previous neck surgery or other form of trauma to the cervical portion of the TD.³ Others have postulated that congenital wall weakness or degenerative processes such as inflammation or atherosclerotic changes in the cyst wall may contribute to cyst development.^{2,4,5} Distal obstruction of the duct has been reported as a cause for cyst development,⁶ but other authors are more skeptical of this possibility.^{5,7} TD ligation is often performed without subsequent development of cysts proximally, theoretically supporting the view that other factors must be present for the development of CTDC to occur.

CT and MRI are both useful modalities for initially diagnosing cystic neck masses. Typical features include lack of nodularity, septations or contrast enhancement as well as a well-circumscribed round or oval shape. Radiodensity is close to CSF with a mean of 10.2HU reported in the literature.⁸ The most critical test for diagnosis is ultrasound guided fine needle aspiration of the identified cystic mass. Fluid with lymphocyte predominance and very high levels of triglycerides and chylomicrons (fat content 0.4–4%) is very suspicious for lymphatic cyst.⁵ Differentiation between lymphocele and CTDC is difficult based on imaging.⁹ Endothelial and epithelial markers have been described⁵ which confirm the presence of an epithelial lining. This histologic finding differentiates CTDC from lymphocele which lacks epithelium. Lymphangiography is the “gold standard” for imaging and diagnosis of malformations of the TD,^{2,9} but with the ease and availability of high resolution imaging and needle aspiration this is not often pursued.

Both medical and surgical approaches have been proposed for the management of CTDC, though the most widely employed method of treatment is surgical ligation. In the review by Brauchle et al. only 2 of 15 patients reported in the literature were treated with observation. A follow up report on one of these patients 25 years later showed spontaneous regression of the cyst with conservative management,¹⁰ suggesting that observation is a reasonable option. The psychological angst associated with a mass of unknown

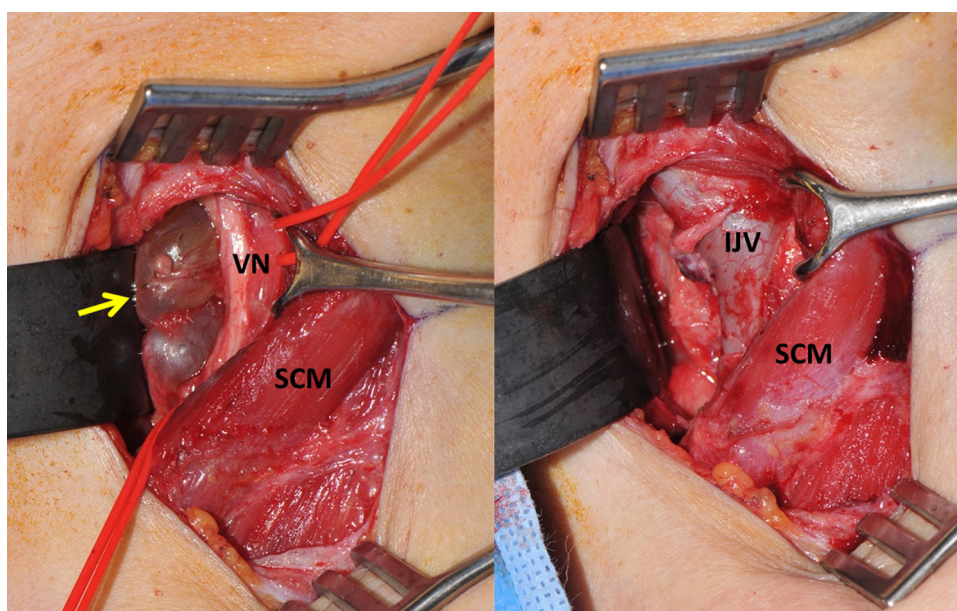


Fig. 2. Pre and post-operative images of thoracic duct cyst. [Left] The thoracic duct cyst (arrow) is exposed with medial retraction of the common carotid artery and lateral retraction of the internal jugular vein (encircled by red vessel loops). The vagus nerve (VN) is seen lateral to the cystic mass. [Right] Image taken after cyst-venous anastomosis showing decompression of the cyst with suture line on medial aspect of internal jugular vein (IJV). Common carotid artery is retracted medially.

origin may be a contributing factor to the generalized practice of surgical resection, but other benefits of excision include definitive histologic exclusion of malignancy and potential relief of symptoms. Chyle fistula is a serious potential risk factor associated with cyst excision; however this has not been reported in the literature at this point. Other potential risk factors include injury to the nearby nerves or major vessels (e.g. vagus n., phrenic n., carotid a., internal jugular v.). Sclerotherapy is one alternative form of treatment that has been proposed for both cervical lymphoceles and CTDC.¹¹ This treatment modality reserves the option of surgical excision if sclerosing agents are ineffective. Cyst aspiration and diet modification have also been discussed. However, both of these reports resulted in eventual surgical excision for definitive management.^{3,12}

This case represents a unique scenario in that no previous patients have presented with unilateral arm swelling ipsilateral to the CTDC. Given the initial improvement in the patient's symptoms after cyst drainage we hypothesize that the etiology of her symptoms included an element of lymphatic obstruction caused by the cystic mass. The exact location of obstruction is unclear as one would expect that the extensive collateral network of lymphatics in the arm to allow adequate drainage. She also had moderate persistent edema at 2 years post-surgery suggesting the presence of other factors contributing to her left arm edema. It is important to note that the patient had no evidence of venous thrombus pre-operatively and did not have hypercoagulable risk factors. Doppler ultrasonography would be a potentially valuable adjunct in this case, but this has not been arranged thus far. Reverse lymphatic mapping of the arm may also reveal anatomic variations responsible for this patient's obstruction. However, this was not pursued due to her initial improvement post-operatively.

Prior to this case, there have been no reports of CTDC management via cyst-vein anastomosis despite the common application of similar surgical techniques to other clinical problems. Several studies have reported success with the treatment of lymphatic disorders via lymphovascular shunting^{13–15} but most of the present studies focus on repairing secondary injury to lymphatic structures. This case illustrates that lymphovascular anastomosis is a potential alternative to surgical excision of CTDC, which allows preservation of a more natural lymphovascular anatomy.

4. Conclusion

CTDC are very rare entities which may present with a wide variety of symptoms. We report a novel application for lymphovascular anastomosis for decompression and definitive treatment of this unusual pathology.

Conflict of interest

The authors have no conflicts of interest to report.

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Ethical approval

Verbal consent was obtained directly from the patient who was not physically present for written consent.

Author's contributions

John Dortch, MD – Manuscript preparation, patient follow-up.

Dustin Eck, MD – Manuscript editing and preparation.

Albert Hakaim, MD – Performance of operation.

John Casler, MD – Performance of operation, patient follow-up and care.

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