CASE REPORT

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SPONTANEOUS CERVICAL LYMPHOCELE

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Abstract: Background. Primary (spontaneous) cervical lymphoceles in adults are extremely rare. More frequently occurring acquired cervical lymphoceles have been described in the setting of a neck trauma or after a neck dissection. We report a case of a spontaneous left cervical lymphocele in a previously asymptomatic female.

Methods and Results. A 44-year-old woman presented with a 2-month history of a left neck mass initially noted by her physician during a routine physical examination. She denied prior head and neck surgery or neck trauma. CT scan of the neck revealed a left cystic mass. Fine-needle aspiration of the cyst yielded chylous material and lymphocytes. The surgical specimen grossly and microscopically was consistent with a lymphocele. The diagnosis was confirmed using D2-40 antibody targeting lymphatic endothelial cells lining the cyst.

Conclusion. Primary cervical lymphocele should be included in the differential diagnosis of a solitary neck mass in an adult.

Keywords: spontaneous; cervical; lymphocele; thoracic duct; cyst

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Spontaneous cervical lymphoceles in adults are rare. Review of the literature yielded only 15 reported cases. Patients ranged from 28 to 68 years of age, and there was no gender preponderance. In addition, Steinberg1,2 reported 2 examples of primary congenital supraclavicular thoracic duct cysts presenting in adults.

Congenital disorders of the lymphatic system are usually caused by duct or lymphatic hypoplasia. A persistent embryonic jugular lymph sac is postulated to result from incompetence or absence of valves safeguarding influx of lymph into the venous circulation. As a result, an increase in intrathoracic pressure during Valsalva maneuvers reverses the pressure gradient resulting in retrograde flow of blood into the thoracic duct. Rarely, a hyperplastic process coupled with valvular incompetence leads to congenital lymphatic pathology.2 Acquired cervical lymphoceles, secondary to blunt cervical trauma3 or thoracic duct injury during neck dissection,4 are more frequent. Interestingly, Barlow and Gracey in 19655 described the first case of a primary noncongenital (ie, spontaneous) cervical thoracic duct dilatation.

CASE REPORT

A 44-year-old woman presented with a 2-month history of a left neck mass initially noted by her physician during a routine physical exam. A CT scan (Figure 1) of the neck revealed a hypodense/
Cystic mass posterior to the lower left sternocleidomastoid muscle and lateral to the left common carotid artery and left internal jugular vein. The lesion measured 4.4 cm in its transverse diameter and 2.5 cm in its anteroposterior diameter.

The patient’s social history was significant for 30-pack-year smoking habit and social alcohol use. The patient denied weight loss, fatigue, dysphagia, prior history of neck trauma, or upper respiratory tract infection preceding discovery of the lesion. On an initial physical examination, a soft, painless, fluctuant mass was noted at the base of the left side of the neck. The remainder of her physical exam and chest X-ray were unremarkable. Fine-needle aspiration (FNA) of the mass yielded 5 cc of creamy, mucoid material. Cytologic examination revealed lymphocytes, macrophages, and the lack of epithelial cells. Because the mass continued to rapidly enlarge, a decision was made to excise it. Under general anesthesia, dissection proceeded in a subfascial plane to the deep musculature of the left neck. Once dissected free from the surrounding soft tissues, the cystic tumor in situ did not reveal any connection to either the thoracic duct or the great vessels medially. After removal of the mass and copious irrigation, a Jackson-Pratt drain was placed, and the wound was closed in 3 layers. The postoperative course was uneventful, and the patient was discharged home the next day after removal of a Jackson-Pratt drain.

**RESULTS**

Gross examination of the specimen demonstrated a yellow-tan, thin-walled cystic structure measuring 5.3 × 3.2 × 2.6 cm (see Figure 2). The cyst was opened to reveal approximately 20 mL of chylous fluid. A translucent cyst wall was less than 0.1-cm thick and exhibited patchy, trabecular areas. Histological sections of the cyst stained with hematoxylin–eosin demonstrated cystic spaces lined by benign endothelial cells separated by a scant intervening connective tissue stroma (see Figure 3). Immunohistochemical staining with D2-40 antibody readily targeted lymphatic endothelial cells lining the cyst. Clinical, gross, and histological findings supported the diagnosis of a cervical lymphocele.

**DISCUSSION**

A lymphocele is an abnormal circumscribed collection of lymphatic fluid lined by endothelium. This benign tumor is typically composed of massively dilated cystic spaces lined by endothelial cells that are separated by a scant intervening connective tissue stroma. Lymphoid follicles or infiltrates may also be observed in the intervening septa. Since these tumors lack true epithelial capsule and do not have discrete margins, surgical removal can be difficult.

Lymphatic circulation arises from venous vasculature between the sixth and tenth week of gestation. Six lymphatic primordia (lymph sacs) bud off from venous endothelium in a human embryo. By the end of the ninth week, all but the pair of jugular sacs lose connection with their parent veins. A mature thoracic duct conveys the majority of the lymph and chyle into the blood. It originates from the cisterna chyli, enters the thorax via a diaphragmatic hiatus, and runs cephalad.

**FIGURE 1.** CT scan of the left cervical lymphocele; (A) internal jugular vein, (B) common carotid artery, (C) sternocleidomastoid muscle, (D) lymphocele.
Obstruction at the lymphatic-venous angle may also predispose to cyst formation. Simple ligation of thoracic duct at any level is not thought to cause either aneurysmal dilation or cysts. In the review of 15 cases of cervical thoracic duct cysts by Brauchle et al,11 the etiology was unclear in 3 cases while congenital pathology accounted for 4 cases. Trauma was the cause in 2 instances. Obstruction, degeneration, or both were the etiology in 2, 1, and 3 patients, respectively. In the present case, there was no evidence of an inflammatory process. This finding, combined with the lack of communication between the cyst and thoracic duct at the time of surgery, suggests obstructive etiology of the lymphatic flow with or without congenital wall weakness.

As in this case, the vast majority of primary cervical thoracic duct cysts are asymptomatic at the time of presentation. Only 2 of the reviewed 17 patients had complaints other than a left supraclavicular mass. When present, symptoms relate to compression of adjacent structures and range from a vague pressure sensation to apnea.12 Preoperative evaluation should include a thorough history and physical exam. Physical examination often reveals a fluctuant left supraclavicular mass. Valsalva maneuvers will accentuate a persistent jugular lymph sac. FNA and cytologic and/or biochemical fluid analysis13 may provide additional diagnostic information, because lymph appears milky, contains lymphocytes and macrophages, and has a high fat concentration. Imaging modalities play a role in determining anatomical boundaries of the mass and help distinguish multilocular from unilocular cysts. The usual location of a cervical thoracic duct cyst is near the junction of the left internal jugular and subclavian veins, superior to clavicle, deep to sternocleidomastoid muscle, and lateral to the carotid sheath. Plain film of the chest should be obtained to rule out mediastinal involvement. Sonography is a useful noninvasive tool to confirm the cystic nature of the mass. The “gold standard” test for determining lymph flow is lymphogram.2,13,14 However, in the era of CT and MRI, it is rarely indicated. Since the 1990s, CT scan has been the most common imaging modality used to delineate surrounding anatomy and characterize the tumor. Since the differential diagnosis of a solitary neck mass is extensive and includes both benign and malignant neoplasms, pathologic evaluation is always warranted. The treatment of choice for this benign neoplasm remains surgical resection. Only 2 cases of a conservative follow-up are re-
ported. In those instances, patients declined surgical management.

Spontaneous cervical lymphocele is an extremely rare yet important entity to consider in the differential diagnosis of a solitary neck mass in an adult patient.

REFERENCES