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Case Report

CERVICAL THORACIC DUCT CYST FOLLOWING TOTAL THYROIDECTOMY AND MRND II WITH REVIEW OF LITERATURE

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ABSTRACT
Thoracic duct cysts are rare. These are more commonly reported in mediastinum and abdomen. Cervical thoracic duct cysts are rarer; they are incidental or occur spontaneously after blunt traumas. Very few cases of cervical thoracic duct cysts are reported and documented. Extensive search yields 48 cases to date. Most of the cases presented were progressively enlarging slow growing otherwise asymptomatic spontaneous swelling in left supraclavicular fossa. Some of them were only growing for few months. In 11 cases some form of trauma has been associated. Penetrating trauma or surgery may injure the thoracic duct and can eventually result in chylous fistula. We are reporting a case of 30 years female two months after surgery presenting with development of true cervical thoracic duct cyst following total thyroidectomy and MRND II left side.

Keywords: *Cervical Thoracic Duct Cyst, Cysts of Neck, Lymphocele*

INTRODUCTION
Thoracic duct drains the chylous lymph from major parts of the body into the venous system at the junction of left internal jugular vein to subclavian vein (Hamilton *et al.,* 2011). It is vulnerable to trauma during clearance of neck or ligation of internal jugular vein at its lower end. Injury to thoracic duct if not taken care properly results in post operative chylous fistula, a dreaded complication following neck dissection. Chylous fistula often requires re-exploration of neck. Injury to small tributaries to lymphatic ducts in neck may result in post operative lymphatic extravasations, forming psuedocysts in neck (Myers *et al.,* 2000). Thoracic or lymphatic duct cysts in the neck are rare and spontaneous in nature. Extensive internet search over Pubmed and Google yielded about 48 reported cases. 11 cases reported some form of trauma associated with swellings rest were spontaneous in origin. We are presenting a case of post operative true lymphatic cyst which occurred two month after total thyroidectomy and modified neck dissection.

CASES
A 30 years female patient presented with swelling in left lower cervical region. Patient was a follow up case of papillary carcinoma thyroid having extra thyroidal spread and neck disease in left level IV, V and in central compartment level VI (T_4N_{1b}). Her earlier FNAC of thyroid swelling was suggestive of papillary carcinoma of thyroid. Patient underwent total thyroidectomy and left MRND II with central compartment clearance. Histopathology report showed papillary carcinoma of thyroid with nodal metastasis, tumor was extending to surrounding soft tissue. Pathological staging was $pT_4N_{1b}M_0$. Post operative period was uneventful. Post operative 100 mCi I^{131} was administered for ablation. Post treatment scan was negative. In her third month of follow up, patient presented with swelling left side of neck growing for 15 days. Examination showed non tender swelling was gradually increasing in size. The finding was quite unusual as presence of recurrent/ residual metastatic node of this much size after total thyroidectomy and MRND II with radioiodine ablation was almost impossible. On USG scan, it showed 40 x 26 mm cystic space occupying lesion in left lower cervical region. FNAC revealed milky white fluid aspiration. Biochemical analysis of fluid showed high levels of triglycerides (757mg/dl). Likely diagnosis

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of chylous cyst was made. Cyst was excised under GA. On exploration 5cm x 4 cm x 4 cm cyst in close proximity of lower end of internal jugular vein was revealed (Figure 1).

Cyst was carefully dissected and both end securely ligated with non-absorbable sutures. Please refer to figure 2 which shows excised cyst and picture of aspirated milky white fluid. It was insured that there was no chyle leak during surgery. Wound was closed over a suction drain. Post operative period was uneventful. Histopathology revealed a cystic lesion lined by wall made up of fibrocollgenous and myxoid tissue lined by endothelium (Figure 3). Cyst wall was 3 mm thick. Cyst wall showed presence of mature lymphoid cells. The fibrocollagenous tissues surrounding the cystic lesion showed presence of foreign body granuloma against ligature material which was used in previous surgery (Figure 4). Currently patient is on follow up of 15 months duration on levothyroxine 100 µgm once a day. TSH level 0.11 µIU/ml and TGA level is 1.2 ng/ml.

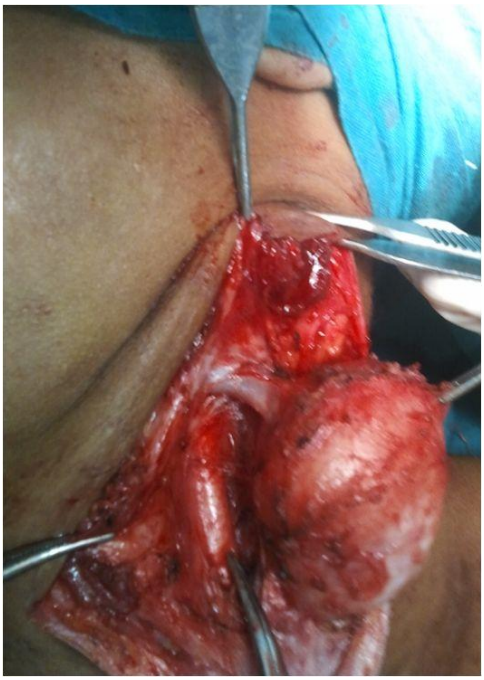

Figure 1: Thoracic duct cyst in close association of internal jugular vein

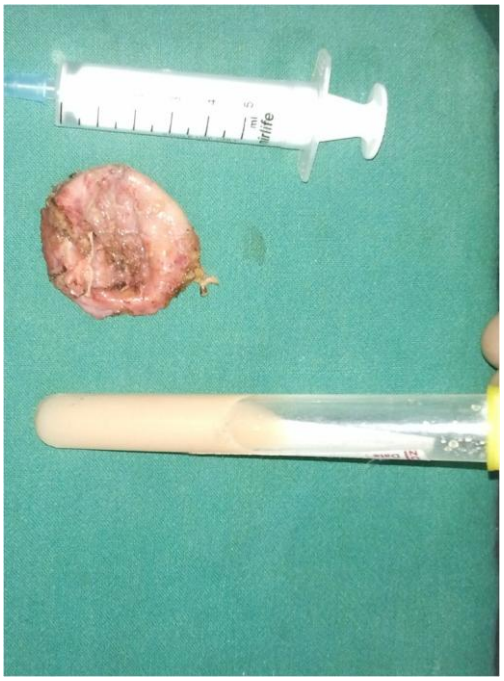

Figure 2: Excised cyst with aspirated milky white fluid

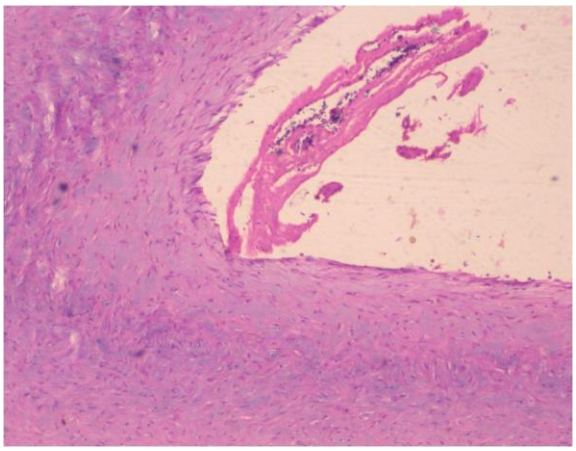

Figure 3: Endothelial lined cyst having lymphoid content in lumen

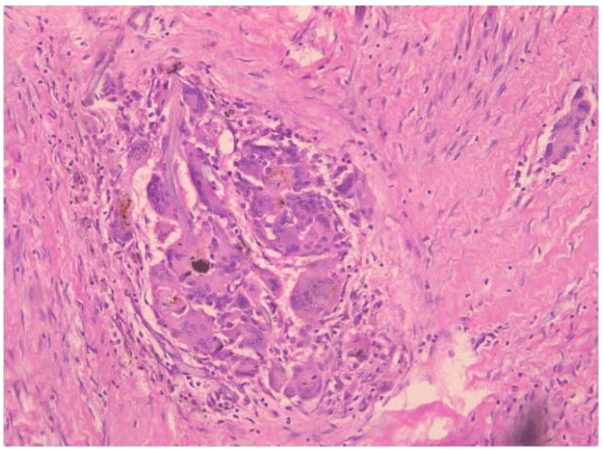

Figure 4: Foreign body granuloma around ligature material in cyst wall

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DISCUSSION

Lymphatic system at root of neck drains the lymph and chyle from whole of the body into venous circulation. Subclavian, jugular and bronchomediastinal lymphatic trunks on either side of neck drains into venous system at or near the junction of the internal jugular vein and subclavian vein. Large thoracic duct drains the chylous lymph from gastrointestinal tract into left lymphovenous portal. These lymphatic trunks may drain directly or join to form lymphatic ducts. At the junction dilated lymphatic ducts form ampullae where the lymph enter into the venous system (Hamilton *et al.,* 2011; Pan *et al.,* 2010)

Thoracic duct cysts are rare in neck. Most of the cases presented with asymptomatic swelling of few month duration. Dull pain occasional dyspnea, hoarseness and difficulty in breathing have been reported (Okazaki *et al.,* 1996; Gupta *et al.,* 2005; Qureishi *et al.,* 2012). Study reviewed 48 cases of cervical thoracic cysts; average age of the patients was 48 years (between 17 and 79 years). The age distribution curve shows bimodal presentation at 28 yrs and 55 yrs. Duration of cystic swelling was of few months, however Brauchle *et al.,* (2003), Hiraumi *et al.,* (2003) and Wang *et al.,* (2009) reported duration of 2, 5 and 10 years respectively.

Numerous theories have been suggested to explain the etiology of lymphocoele of the terminal thoracic duct. Both congenital weakness in the thoracic duct wall and acquired degenerative process from inflammation have been proposed as causative mechanisms. Trauma can be considered as the underlying etiology. In majority of cases swelling arise spontaneously, however in 11 patients history of preceding trauma was present. These cases were associated with blunt trauma (Livermore *et al.,* 1993), hyperextension of neck (Mosahebi *et al.,* 1998), fall (Lecanu *et al.,* 2001), weight lifting (Walker *et al.,* 2010), neck massage (Ceylan *et al.,* 2007) and physiotherapy (Nouwen *et al.*,2004). The swelling noted after 2 days to few months of suspected trauma.

Study shows that even trivial form of trauma can cause lymphatic duct injury eventually resulting in cyst formation. Roh *et al.,* (2008) reported four cases of postoperative cervical lymphatic swelling developed 3 weeks to 12 months after surgery. Nouwen *et al.,* (2004) reported a case of left supraclavicular lymphatic swelling six days after total thyroidectomy and neck dissection.

Positive history of smoking was seen in six patients. Out of those six patients, one patient had history of physiotherapy few months back. Smoking can attribute to atherosclerotic weakness of lymphatic wall leading to formation of cyst. Aging and smoking are associated risk factor besides some degree of trauma which may even remain unnoticed. However as these lesion are rarely seen, we assumed that unnoticed trauma to these weak lymphatic may be the triggering event which progresses to form a lymphatic cyst. Bimodal presentation of age can be due to congenital or acquired weakness of the wall of lymphatics. Therefore we conclude that development of cyst, either spontaneous or acquired, occurs in weak lymphatics with degenerative changes (Brauchle *et al., 2003*; Ducic *et al.,* 1999; Dool *et al.,* 2007; Hekiert *et al.,* 2007; Preuss *et al.,* 2006; Wax *et al.,* 1992).

Ligation of thoracic duct is being done for ages and it does not result in cystic dilatation of thoracic duct. However in the presented case, foreign body granuloma was seen around suture material of previous surgery in the cyst wall. It could be due to trauma to the wall of surrounding lymphatic channel or junction area causing weakness of its wall which progressed to cyst formation.

Lymphographic evaluation is a traditional diagnostic method for thoracic duct anomalies. CT and MRI are less affordable and require contrast medium. If enhanced CT or MRI shows unilocular, nonseptated, fluid density or intensity, and nonenhancing supraclavicular cyst in the posterior cervical space, lymphocoele is important to be considered as one of the differentials.

Lymphoceles are rare unilocular cystic neck masses that may mimic other congenital, infectious, and malignant neck cysts (Hamilton *et al.,* 2011). Majority of these are true cysts, cystic metastatic lesions, or pseudocysts. A true cyst is a fluid-filled, epithelial-lined structure, whereas a pseudocyst lacks a true epithelial lining (Myers *et al.,* 2000). Franceschi *et al.,* (2012) used echo-colour-Doppler (ECD) for recurrent spontaneous cervical swelling of supraclavicular fossa in four patients. Franceschi *et al.,* (2012) found thoracic duct dilation with hyperechogenic content during the swelling period which was

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undetectable during asymptomatic periods. They related chyle stasis to be due to malformation of the valve, located at the jugular–subclavian confluence resulting in recurrent cervical swellings.

Conservative management of small otherwise asymptomatic lesions can be done by USG guided aspiration and injection of sclerosent followed by fat frees diet and pressure dressing. Absolute alcohol (Dool *et al.,* 2007; Walker *et al.,* 2010), OK-432 (Preuss *et al.,* 2006; Roh *et al.,* 2008), povidone iodine (Seelig *et al.,* 1998) and talc (Okazaki *et al.,* 1996) have been used as sclerosent with variable success rate. Zätterström *et al.,* (2009) reported spontaneous complete regression of a cystic dilatation of the thoracic duct in a patient with 25 years of follow up. The advantage of conservative management is that it avoids the potential risk of chylous leak, risk of injury to nerves and great vessels with almost no side effect and no cosmetic compromises. Only slight fever for few days has been mentioned after the use of OK-432 (Roh *et al.,* 2008). Excision was done in majority of cases and in almost all cases followed by complete cure with no case of post operative chylous fistula or recurrence. Therefore surgery is the definite treatment for lymphatic cysts.

Aspiration of milky white fluid rich in triglycerides and chylomicron and presence of lymphocytes is diagnostic of chylous cyst of thoracic duct origin (Mattila *et al.,* 1991). Reviewed cases report high levels of triglycerides in aspirate ranging from 670 mg/dl to 3350 mg/dl (Sakamoto *et al.,* 1991; Nouwen *et al.,* 2004; Ceylan *et al.,* 2007).

Cyst wall lined by endothelium is diagnostic of true lymphatic cyst. Endothelial lining can be confirmed by immunohistological examination showing CD 31 positive cells. Immunohistochemical staining with D2-40 monoclonal antibody readily targets lymphatic endothelial cells lining the cyst (Kahn *et al.,* 2002). The presence of lymphocytes and triglycerides differentiate thoracic duct cyst from cystic hygroma which is also lined by endothelium (Hekiert *et al.,* 2007).

Conclusion

Cervical thoracic duct cysts are very rare. Small asymptomatic lesions can be kept under observation or conservatively managed by aspiration and sclerosent injection followed by fat free diet. Symptomatic and progressively increasing swelling should be surgically excised. The thoracic duct should be carefully ligated to avoid post operative chylous fistula.

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