Bilateral chylothorax without chylous fistula following a left modified radical neck dissection: a case report and review of literature

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Abstract

Bilateral chylothorax following injury to or ligation of the cervical portion of the thoracic duct is a rare but potentially dangerous complication. We report a case of bilateral chylothorax without any evidence of chylous fistula in a 24 year old woman following a left modified radical neck dissection for a primary neck cancer. The patient was effectively treated by conservative management.

Keywords: Bilateral chylothorax, modified neck dissection, neck cancer

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Introduction

Bilateral chylothorax following injury to or ligation of the cervical portion of the thoracic duct is a rare but potentially dangerous complication.[1] Occasional cases of chylothorax following injury to the cervical portion of the thoracic duct in war wounds[2], stabbing[3], and fractures of the first rib[4] has been reported. In all these, there is associated pleura perforation. On the other hand, operative

injury to the thoracic duct not associated with perforation of the pleura and yet complicated with bilateral chylothorax is a very rare clinical condition.

In 1907 Stuart reviewed 42 cases of operative injury to the thoracic duct in the neck and found three patients with chylothorax, all of whom died. Review of literature revealed only 16 cases. To the best of the author's knowledge; this is the first reported case of bilateral chylothorax following neck dissection in Nigeria. We report a case of bilateral chylothorax without any evidence of chylous fistula in a 24 year old woman following a left modified radical neck dissection for a primary neck cancer.

Case Report

A 24-year old lady who presented to our department with four years history of left sided upper neck swelling and two years history of left sided lower neck swelling. The swellings were insidious in onset, initially painless but became painful about ten months prior to representation. There was associated amenorrhea of two years duration and progressively worsening neck pain. There was no otological, rhinological, throat or constitutional symptoms.

Following comprehensive assessment, Computerized Tomography Scan, pan endoscopy and biopsies, no definitive diagnosis was made and the differential diagnoses were cervical metastasis of unknown primary, glomus tumour and brachial cyst anomalies.

She was eventually planned for enbloc excision of the mass and left modified radical neck dissection (sparing spinal accessory nerve). Intra-operative findings of two hard yellowish masses, the upper one at the carotid bifurcation, adhering closely to the walls of the common carotid, the lower one at the level of the lower third of internal jugular vein were noted, both masses were fixed to underlying structures as well as overlying muscles. The masses were excised completely. During surgery, lymph leakage was evident. The thoracic duct and other accessory lymphatic channels that were inadvertently injured were ligated.

First postoperative day was uneventful, wound drained about 200ml of serosanginous fluid and she was commenced on oral intake. However, two days postoperatively, she developed chest pain, difficulty in breathing and decrease air entry bi-basally. Chest X-ray done showed bilateral pleural effusion and she had bilateral closed thoracotomy tube drainage (CTTD) which drained a total of 2.2 litres of chlyous effluent from both pleural spaces. Biochemical analysis of the effusion confirms chyle. The chylothorax was managed conservatively with non-fatty, high protein and high calorie diet. Although the drainage initially increased in quantity per day and climaxed on the third post-operative day at 3.3L of chylous material.

The chest pains as well as respiratory distress started subsiding on the fourth day post-op. Oxygen saturation and other vital signs were stable but she was noticed to have made 2120 ml of urine within six hours despite input of 680ml. Random blood sugar, electrolyte, urea and creatinine were all normal but urine specific gravity was 1.007. An assessment of post-surgical diabetes insipidus was made. She continue making high volume of diluted urine until the seven post-operative day when water deprivation test was commenced after which the urinary output gradually reduced in volume with concomitant increased osmolality. The CTTD was removed on the 8 post-operative day after a check chest x-ray was normal and was discharged home on the 10 day. Patient has been on follow up for two and a half years without any complaint. Histology revealed malignant glomus tumour.

Discussion

External chylous fistula is a known complication of radical neck dissection (in 1% to 2%) of cases but chylothorax is extremely rare.[1, 5]

The pathophysiology of bilateral chylothorax that develops after neck dissection was not fully understood, but two hypotheses have been suggested. In the first hypothesis, the chyle escaping from the cervical region flows directly into the mediastinum, leaks into the thoracic cavity and is retained there.[6] This hypothesis might be true if there is an external chylous leakage in the neck.

The other hypothesis states that ligation of the thoracic duct causes the intraluminal pressure in the thoracic duct to increase, and with the presence of negative intra-thoracic pressure during inspiration; extravasation of the chyle into the mediastinum develops. The increase hydrostatic pressure, direct pleural maceration by the chyle and backflow through dilated intrapulmonary lymphatic vessels contributes to the retention of chyle in both thoracic cavities.[7] This hypothesis is most likely be true in our patient as there was chyle leakage during the surgery which necessitated the ligation of the thoracic duct as well as the other accessory lymphatic ducts.

Some authors have reported cases of unilateral chylothorax following radical neck dissection.[8-10] Although, our patient had bilateral chylothorax, it appeared that the left side was involved on the first post-operative day. Bilateral involvement was evident from the second post-operative day. Similar course was noted by other authors.[11, 12] As the chyle accumulated under pressure, it was finally extravasated through both pleura producing a bilateral chylothorax. In the cases reported by Saraceno and Ferrior[13] and Jabbar and AL-Abdulkareem[5], chest films revealed bilateral effusion on the third and fourth day postoperatively; which may suggests that left sided chylothorax occurred prior to bilateral involvement. It should be noted that most patients undergoing neck dissection have an extensive collateral lymphatic network that provides alternative pathways through which the chyle can pass.[14] It appears that these pathways are sometimes initially inadequate with the daily volume of 1500ml-2500ml that normally drain from the thoracic duct directly into the left jugular venous system.[10]

Following radical neck dissection, patient who develops difficulty in breathing, chest pains, decrease air entry and radiological evidence of effusion should be considered to have chylothorax. Since the protein content of chyle is high and electrolyte similar to that of serum[15], prompt recognition is needed to avoid malnutrition, immunodeficiency and fibrothorax. Initial approach to management of bilateral chylothorax involves chest tube drainage of the plural space.[14, 16, 17] Continuous suction drainage helps to relieve the pressure of chyle on the lungs, re-expands the

partially collapsed lungs, obliterate the pleural space and permits accurate measurement of chyle production. Lung re-expansion is sometimes hindered by formation of fibrinous membrane around the lung which may necessitate surgical intervention.[18] Chest tube drainage of less than 500mls during the first 24 hours after complete cessation of oral intake and total parenteral nutrition may predict successful conservative treatment.[19] Because up to 3L of chyle may drain daily, large amounts of fluid, electrolytes, fat, protein and lymphocytes may be lost. A careful measurement of chyle output, patient's weight, serum albumin, total protein, absolute lymphocyte counts and electrolyte levels are essential. A non-fat, high protein and high calorie diet will produce reduction in chyle flow. Administration of medium chain triglycerides (MCTs) is highly essential.[20]

The duration of conservative management is not firmly established but must be tailored to each patient, but generally it is believe that operative intervention is considered when chyle flow has not diminished within 14 days[18], nutritional and immunodeficiency complications are imminent. Ligation of the thoracic duct via a thoracic or abdominal approach can be considered. Others have described a posterior extra pleural approach to ligation of the thoracic duct.[21] Most recently video assisted thoracic surgery has provided an effective and less invasive approach to chylothorax.[22] Successful ligation of the thoracic duct by thoracoscopy using fibrin glue or endoscopic clips at the site of leak has been attempted[23, 24], and has the advantage of less post-operative pain and shorter hospital stay. An alternative to surgical ligation of the thoracic duct may be fluoroscopic percutaneous embolization. The prerequisite for this procedure is pedal lymhography to access the lymphatic duct via trans-abdominal puncture [25].

Conclusion

In conclusion, high index of suspicion, early diagnoses and prompt treatment of chylothorax is needed in a patient that had radical neck dissection to prevent the morbidities associated with it.

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