CASE REPORT

Bilateral Chylothorax Following Neck Dissection: Case Report & Review of Literature

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Abstract Bilateral Chylothorax following neck dissection is an extremely rare complication. We report a case of bilateral chylothorax detected after neck dissection for carcinoma of lower alveolus. A 61 year Indian female underwent wide excision with segmental mandibulectomy with comprehensive neck dissection for carcinoma of left lower alveolus clinically staged T4N0. Evaluated for dyspnea in post operative period, she was found to have bilateral chylothorax that was managed conservatively. This case report presents potentially life threatening complication following neck dissection that often responds to non surgical management.

Keywords Oral cancer · Neck dissection · Bilateral chylothorax

Abbreviation

SpO2	pulse oximetry saturation
ECG	Electrocardiogram
POD	post operative day
ICD	Intercostal drain
MCT	Medium Chain Triglyceride
TPN	Total Parenteral Nutrition

Introduction

Stuart in 1907 first described three cases of bilateral chylothorax after 40 cases of radical neck dissection, unfortunately none survived [1]. Since then more than 25 cases of bilateral chylothorax have been described. Most of these cases were accompanied with external chyle leaks; however the case that we report here was without external fistula. Chylothorax is more frequently seen in left cervical approaches and its bilateral presentation is extremely rare (1–2 %) [2].

Case Report

A 61 year female with no comorbidity was evaluated for non healing ulcer at left lower alveolus of two months duration; biopsy turned out to be well differentiated squamous cell carcinoma. On examination, she was of good performance status. Local examination of oral cavity showed 3 × 5 cm ulcerated lesion of left lower alveolus occupying last four teeth and encroaching gingivobuccal sulcus. She was staged as c T4aN0 and completed 3 cycles of neoadjuvant chemotherapy with methotrexate and had partial response. She underwent surgery on 27 August, 2014 and intraoperatively she had residual lesion

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of 3.5×1.5 cm of lower alveolus and frozen section reported as all margins clear. Significant nodes in level 1 were present and comprehensive neck dissection was done clearing nodal levels 1 to 4. During level 4 dissection accidental injury of thoracic duct was identified and repaired with prolene 5–0 suture. At the completion of procedure no leak was seen. On post-operative day 2 she developed dyspnea and sweating, breath sounds were decreased bilaterally and SpO₂ was 90 % on room air. Thinking of cardiorespiratory cause, ECG taken was normal. Chest X ray revealed moderate pleural effusion on contralateral side and mild on left side. A diagnostic pleural tap revealed turbid milky white fluid and 750 ml was drained and sent for chylomicron that was elevated. After pleurocentesis her symptoms improved and saturation was 95 %. Serial examination showed refilling effusion bilaterally, this time more on ipsilateral side. Intercostal tube drainage of left side was done. She responded to conservative management with nil by mouth, fat free diet through Ryles tube, high protein diet. ICD was removed on POD 5 and she was discharged on POD 10. Chest x ray was normal at time of discharge.

Discussion

A clear concept of thoracic duct anatomy is crucial for the operating surgeon to avoid complications. Thoracic duct is formed at

the level of L4 lumbar vertebra by cistern chyli and ascends in between aorta and azygous vein over vertebral bodies and crosses at the level of T4–T8 thoracic vertebra towards the left side, then runs behind the aortic arch and left subclavian artery. It ascends behind the carotid sheath to join IJV from behind. Parson et al. [3] in autopsy study found two termination of thoracic duct in 45 % cases; this would predispose to operative injury. A practical point worth remembering is the fact that the duct lies anterior to the fascia overlying the scalenus muscle and therefore in stripping away fat and lymphatic tissue, it is far more susceptible to injury than the phrenic nerve which lies posterior to the fascia [4].

The pathophysiologic mechanism of bilateral chylothorax is not clear but there are two hypotheses:

- Due to direct leak of thoracic duct with subsequent drainage in mediastinum leads to inflammatory reaction that causes effusion to both thoracic cavities.
- Inadvertent ligation of thoracic duct leads to increase in duct pressure. The rising pressure in combination of negative intrapleural pressure is severe enough to cause rupture of thoracic duct. In our case, we presume the second mechanism responsible for bilateral chylothorax.

Following table summarizes the case reports published so far mentioning bilateral chylothorax after neck dissection.

No	Year	Authors(ref)	Disease	Neck dissection	Treatment	Prognosis
1	1951	Frazell et al. [5]	Cutaneous	Left	Conservative	Recovery
2	1976	Coates et al. [6]	Larynx	Left	Conservative	Recovery
3	1981	Saraceno et al. [7]	Thyroid	Left	Conservative	Recovery
4	1985	Har et al. [4]	Thyroid	Left	Conservative	Recovery
5	1985	Ng et al. [8]	Floor of mouth	Left	Conservative	Recovery
6	1988	Pace Balzet et al. [9]	Adenolymphoma	Left	Conservative	Recovery
7	1989	Oi et al. [10]	Maxilla	Left	Conservative	Recovery
8	1992	Biurrun et al. [11]	Unclear	Unclear	Conservative	Recovery
9	1995	Jabbar et al. [12]	Thyroid	Bilateral	Conservative	Recovery
10	2000	Gregor et al. [13]	Nasopharynx	Right	Conservative	Recovery
11	2001	Al Sebeih et al. [14]	Larynx	Bilateral	Conservative	Recovery
12	2001	Jortay et al. [15]	Unclear	Left	Conservative	Recovery
13	2003	Kamasaki et al. [16]	Tongue	Bilateral	Conservative	Recovery
14	2004	Busquets et al. [17]	MUO	Left	Conservative + Surgery	Recovery
15	2006	Srikumar et al. [18]	Nasopharynx	Left	Conservative + Surgery	Recovery
16	2007	Tsukahara et al. [19]	Thyroid	Left	Conservative	Recovery
			Tongue	Left	Conservative	
			Larynx	Bilateral	Conservative	
17	2007	Bae et al. [20]	Thyroid	Left	Conservative	Recovery
			Thyroid	Left	Conservative	
18	2009	Han et al. [21]	Thyroid	Bilateral + mediastinal	Conservative	Recovery
19	2009	Khurana et al. [22]	Thyroid	Bilateral + mediastinal	Conservative	Recovery
20	2010	Al Khudaris et al. [23]	Larynx	Bilateral	Conservative	Recovery
21	2010	Tallon-Aguilar et al. [24]	Thyroid	Left	Conservative + Surgery	Recovery

22	2010	Soodin et al. [25]	Unclear	Unclear	Conservative	Recovery
23	2011	Rodier et al. [26]	Thyroid	Left	Conservative	Recovery
24	2011	Tian et al. [27]	Thyroid	Bilateral	Conservative	Recovery
			Thyroid	Bilateral	Conservative	
25	2013	Amusa et al. [28]	MUO	Left	Conservative	Recovery
26	2014	Runge et al. [29]	Thyroid	Bilateral	Conservative	Recovery
27	2014	Yang et al. [30]	Breast	Left	Conservative + Surgery	Recovery

The morbidity following chylothorax arises from two affects- firstly cardiorespiratory and secondly metabolic. Fluid accumulation in pleural cavities results in compression of lung with reduction of vital capacity and shifting of mediastinum with kinking of great vessels. The metabolic effects result from loss of lymphatic system fluids, including electrolytes (Na and Ca most common), proteins, fats, fat soluble vitamins and circulating lymphocytes [4].

Management of chylothorax is mainly conservative and focuses on decreasing the amount of chyle production and preventing concomitant nutritional, immunological and cardiopulmonary complications. Drainage of effusion by inserting chest tubes to relieve respiratory and circulatory distress is the priority. Conservative measurements include postural recommendation (Bed rest, head elevation, compression bandaging of surgical area), discontinuation of drainage aspiration, fat free, high protein and calorie diet. Some surgeons prefer MCT oil and even TPN without oral intake [31]. Role of octreotide is controversial [32].

Indication for surgical management includes chyle leak of more than 1 L persisting for more than 5 days, chylothorax persisting for more than 4 weeks or wherein severe metabolic complications sets in.

However, there is a group of surgeons who prefer early operative intervention to avoid long term nutritional morbidity and prolonged hospital stay. There are basically two surgical approaches. In the cervical approach, the leakage site can be closed by direct ligation or a PMMC flap transfer after local application of fibrin sealant [13, 33]. In the videothoracoscopic approach, thoracic duct is identified in sub diaphragmatic area where it can be ligated en block with periaortic tissue and azygous vein, either with sutures or clips [34]. Percutaneous lymphangiography guided cannulation with embolization of the leakage is another minimally invasive option [35].

From oncologic point of view, delay in adjuvant therapy is potential side affect of chylothorax and its associated morbidity.

Conclusion

Taken together, our result is similar to those available in literature for management of bilateral chylothorax following neck dissection. High index of suspicion, good physical examination, chest imaging, pleurocentesis might be sufficient for diagnosis and further management.

Competing Interests The authors declare that they have no competing interests.

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