A rare case of metastatic colorectal adenocarcinoma to the thyroid gland after 7 years: An upper airway emergency in a thin neck

Rahmah Nasir, MBBS[1], Danny Kit Chung Wong, MSurg(ORLHNS)[2], Hardip Singh Gendeh, Dr(ORLHNS)[1], Mawaddah Azman, MSurg(ORLHNS)[1], Marina Mat Baki, MSurg(ORLHNS), PhD[1]

[1]Department of Otorhinolaryngology – Head and Neck Surgery, Hospital Canselor Tuanku Muhriz, National University of Malaysia, Kuala Lumpur, Malaysia, [2]Department of Otorhinolaryngology – Head and Neck Surgery, UKM Specialist Children's Hospital, Kuala Lumpur, Malaysia

SUMMARY

Upper airway obstruction is an Ear, Nose, and Throat (ENT) emergency, a life-threatening condition that requires immediate intervention. An 87-year-old female post right hemicolectomy for colon adenocarcinoma 7 years ago presented with stridor. It was a rare case of metastatic colorectal adenocarcinoma to the thyroid causing bilateral vocal fold immobility. Emergency tracheostomy and debulking of thyroid were performed with much difficulty despite the patient's thin neck, due to the distorted laryngeal anatomy caused by the thyroid malignancy. In this case report, we highlight the challenges in acute airway management and role of surgical debulking to management in future cases.

INTRODUCTION

Upper airway obstruction (UAO) is a potentially fatal Ear, Nose & Throat (ENT) emergency that requires immediate intervention. UAO is defined as an anatomic narrowing or blockage of any portion of the airway above the thoracic inlet resulting in decrease ability for ventilation.[1,2] Bilateral vocal fold immobility (BVFI) is a cause of UAO as the vocal cords assume a paramedian static position following injury to the recurrent laryngeal nerve (RLN). This may occur following thyroidectomy or neck malignancy.[3] We describe a rare case of BVFI secondary to inapparent metastatic colorectal adenocarcinoma to the thyroid. The challenges in acute airway management and role of surgical debulking are highlighted for better management in future cases.

CASE REPORT

An 87-year-old female with a background of colon carcinoma with a hemicolectomy 7 years prior presented with progressive noisy breathing and breathlessness for 2 weeks. Her voice was getting increasingly breathy and easily fatigable over the past year. This was associated with a non-productive cough and blood-stained saliva.

She also complained of a 6-month history of progressive dysphagia, choking on solid food. She could only tolerate a soft or liquid diet (e.g.,porridge or soup). She was, however, still independent of all her activities in daily life.

She saw a physician once 6 months ago and was found to have multiple bilateral parenchymal opacities measuring 2–3mm in size on chest X-ray. Metastatic lung disease was suspected however, at that time, she elected not to have further treatment.

She has hypertension and dyslipidaemia. She previously had a right hemicolectomy for a T3N0M0 moderately differentiated adenocarcinoma of the proximal transverse colon. Although there was a perineural invasion, there was no lymphovascular invasion. The tumour was completely excised with clear margins. She refused adjuvant chemotherapy or radiotherapy.

On observation, she had biphasic stridor. Her dysphonia grade was three on the GRBAS scale (grade, roughness, breathiness, asthenia, strain) scale with a predominant breathy component. Her maximum phonation time was only 3 seconds. She was unable to count 1–10 in a single breath and she had poor cough effort.

Physical examination of her neck revealed a thin, slender neck with no neck masses palpable or cervical lymphadenopathy (Figure 1A). Trachea was central and the laryngeal framework was intact. There was, however, loss of laryngeal crepitus.

Flexible nasopharyngolaryngoscopy showed bilateral vocal folds in paramedian position with a slit-like airway. There was a pooling of saliva in the piriform sinuses; however, no lesions were seen in the supraglottic and glottic region. Repeat chest radiograph confirmed metastatic lung cancer with cannonball lesions. Lateral neck radiograph showed narrowing of the subglottic trachea at the level of C7 and below (Figure 1B).

The initial impression was metastatic adenocarcinoma to the lungs or a second primary malignancy such as an upper oesophageal carcinoma causing RLN palsy.

She underwent an emergency tracheostomy under local anaesthesia (LA) due to concerns of difficult intubation due to BVFI, as well as the risk of airway collapse, and prolonged ventilation due to poor lung condition if she was put under

This article was accepted: 10 July 2022
Corresponding Author: Marina Mat Baki
Email: marinamatbaki@ppukm.ukm.edu.my

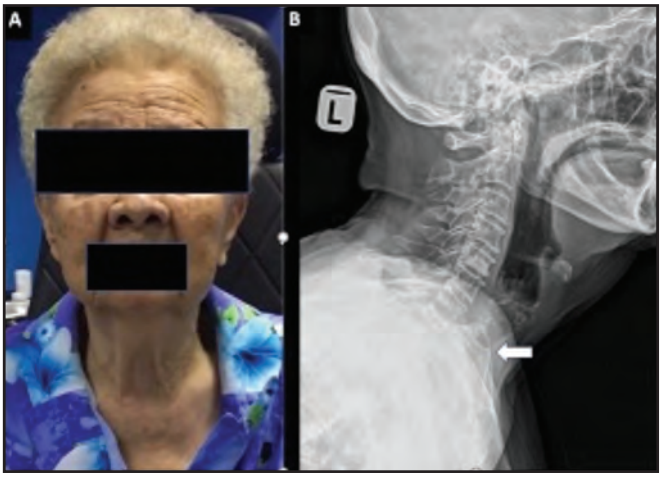

Fig. 1: (A) Patient with a thin neck with no anterior neck swelling palpable (printed with consent). (B) Lateral neck x-ray showing a narrowed subglottic area at level C7 (white arrow)

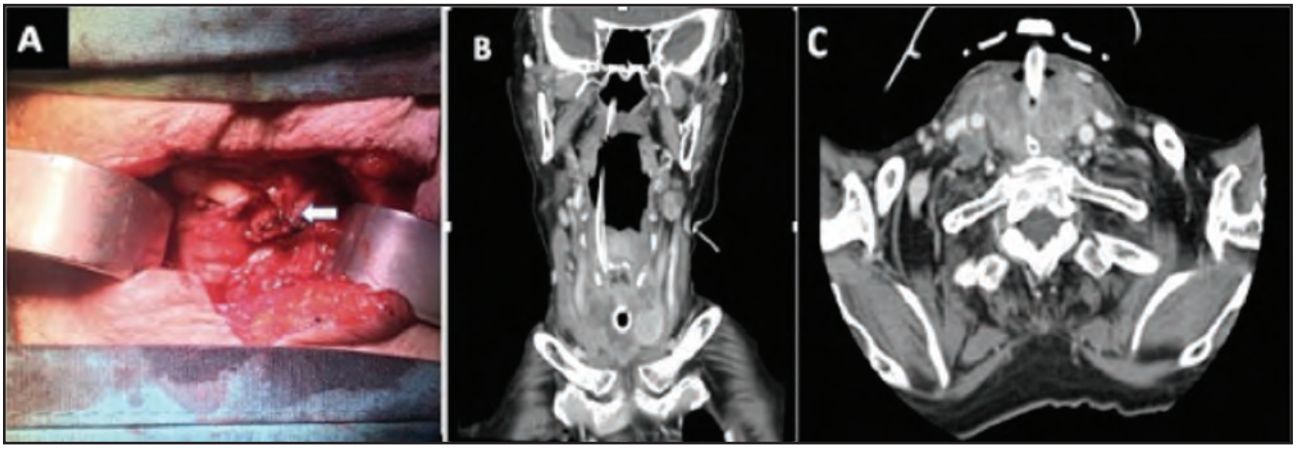

Fig. 2: (A) Intraoperative picture of trachea showing cricoid and tracheal rings one to three after debulking of thyroid tumour (white arrow). (B) Coronal CECT view of right hypodense tumour and left hyperdense thyroid nodule with tracheostomy *insitu*. (C) Axial CECT view of tumour surrounding trachea causing tracheal narrowing at level C7 with tracheostomy *insitu*

general anesthesia (GA) via intubation. Intraoperatively, there was a thick, hard, calcified mass firmly adherent and wrapped around the thyroid cartilage, cricoid, and trachea, conforming to these structures. The mass was identified as a calcified thyroid gland, around 1cm in thickness, and there was no plane of dissection. This distorted the laryngeal anatomy causing difficulties identifying the trachea. After dissecting superiorly to delineate the superior thyroid notch and inferiorly to the level of the sternal notch, a low tracheostomy was not possible. A hypodermic needle and syringe partially filled with normal saline was used intermittently to aspirate air from the lumen ofthe trachea to ensure that it was still midline and to ascertain the depth from the plane of our dissection. Partialdebulking of the mass or an isthmectomy was performed to create a window to expose the cricoid cartilage and upper tracheal rings (Figure 2A) and a size 7.5mm single lumen cuffed tracheostomy tube was inserted. Of note, there was tracheomalacia at rings two to four.The patient was supplemented with oxygen via a face mask throughout the procedure. Once the tracheostomy was completed, the patient was put under GA. Biopsy of the calcified thyroid gland was sent for histopathology examination.A rigid esophagoscopy was initially planned but it was not performed as the patient was not fit for further procedures.

Postoperatively, she was extubated, put on tracheal mask oxygen support for few days and nursed in general ward. She did not require intensive care unit admission. She was treated with amoxicillin / clavulanic acid 1.2g three times a day for aspiration pneumonia which was caused by the emergency tracheostomy under LA. She also had a left lung pigtail drain inserted for malignant pleural effusion. Cytology of the pleural fluid showed atypical cells. CEA and CA19-9 were elevated.An upper flexible oesophagoscopy and a percutaneous endoscopic gastromy (PEG) tube insertion was scheduled postoperatively but cancelled due to general frailty.

Postoperative contrast-enhanced computed tomography (CECT) scans of her brain, neck, thorax, abdomen, and pelvis revealed metastases to the thyroid gland, cervical lymph

nodes, brain, lung, pleural space, and liver. In the neck, there was a large hypodense right thyroid lobe and isthmus with central necrosis (Figure 2B). There was evidence of extracapsular spread and the lesion surrounded the trachea, causing narrowing of the trachea at the level of C7 (Figure 2C). Posterior to the trachea, there was no clear plane between the tumour and oesophagus at the level of T2; however, there was no overt infiltration of tumour into the lumen of the oesophagus. At that level, the oesophagus was compressed by the tumour. There was the presence of necrotic lymph nodes largest at right level II (1.6cm). There was, however, no evidence of local recurrence in the bowel.

Histopathological examination confirmed metastatic adenocarcinoma to the thyroid gland from colorectal origin. The thyroid gland was completely infiltrated by malignant cells which were positive for gastrointestinal immunohistochemical markers cytokeratin 20 (CK20) and negative for cytokeratin 7 (CK7) and transcription termination factor 1 (TTF-1).

She spent 3 weeks in hospital and was discharged home for hospice care.

DISCUSSION

Thyroid metastases are rare withan incidence of 0.36–2% of all thyroid malignancies.[4-5] The most common site of origin for thyroid metastases is from renal cell carcinoma followed by lung, breast, and then only from the gastrointestinal tract carcinomas.The incidence of thyroid metastases from colorectal carcinoma (CRC) is therefore low and has been reported to be 0.1% in a retrospective audit of 5,862 cases of thyroid metastases detected over a 10-year-period.[6] The principal ways for metastases in colorectal cancer are direct invasion, hematogenous spread, lymphatic spread, and implantation metastasis.However, despite its rarity, one should consider the possibility of an undiagnosed thyroid malignancy in a stridorous patient when faced with the difficulty in identifying normal tracheal rings during tracheostomy.

Thyroid gland and RLN are commonly mentioned together due to their anatomical proximity. The right RLN branches from the right vagus nerve or cranial nerve X (CNX) in front of the first part of the subclavian artery and then hooks below and behind the artery. The right RLN subsequently enters the trachea-oesophageal fascia (TOF) at a level inferior to C7 to T1.[7]

In contrast, the left RLN branches from the left CNX on the left side of the arch of aorta before it hooks around ligamentum arteriosum on the lower surface of the aortic arch in the thorax. The left RLN enters at level inferior to T2.[7] Both RLNs ascend within the trachea-oesophageal groove with the right RLN located more anteriorly and laterally compared to the left RLN, on the medial surfaces of the thyroid lobe.[7] Each RLN passes deep to the inferior constrictor muscle before it enters the larynx. The RLN innervates all the laryngeal muscles except the cricothyroid muscle and the sensation below the vocal cord level.

In the patient above, the CRC had metastasized to the right thyroid gland and the isthmus with extracapsular spread. Based on the CECT of the neck, the tumour had significantly encased and caused narrowing of the trachea at the level C7 with no erosion of the thyroid cartilage seen. Posteriorly, there was no clear fat plane with the trachea at T2 level and tumour compressed the oesophagus. This correlates to the involvement of the trachea-oesophageal groove at the level C7 to T2 which explains the BVFI causing UAO, stridor, hoarseness, and aspiration. Other causes of worsening respiratory distress could include enlargement of thyroid tumour due to haemorrhage or progression of cancer causing rapid increase of external pressure on the trachea or intratracheal invasion of thyroid tumour. In contrast, it was initially thought that the BVFI was due to lung metastases involving both sides of the RLN.

Malignancy of the thyroid gland often causes tethering of the soft tissues due to fibrosis thereby making it difficult to obtain a good laryngoscopic view of the larynx during orotracheal intubation.[8] Although there was a role for awake fibreoptic intubation, this was not possible for this patient due to her severe metastatic lung disease which would put her at a high risk for a complete airway obstruction or a prolonged ventilation period subsequently. Therefore, in our case, we performed a tracheostomy under LA.

Surgically, this was a difficult case due to poor anatomical landmarks. Therefore, a total or subtotal thyroidectomy would have helped delineate the anatomy better. However, given the patient's comorbidities and the emergency airway obstruction requiring quick airway secure, we performed an isthmectomy and partially debulked the thyroid mass instead to enable us to access the trachea. This was possibly the most appropriate decision at the time to secure the airway, given the urgency of situation.

In a caseseries by Testini et al,[9] they have advocated for an emergency thyroidectomy with or without an emergency tracheostomy for airway obstruction caused by a thyroid mass.[9] This is to relieve the compression on the trachea. In their series, tracheostomy was reserved for patients with tracheomalacia or when there was tracheal infiltration of tumour.[9] The rationale was that a tumour of the thyroid gland would completely obscure the landmarks of the thyroid cartilage and trachea and this would carry a high risk of bleeding when performing a tracheostomy alone. Therefore, if a subtotal, near-total or total thyroidectomy was unable to be performed at the same setting, a debulking of thyroid tumour would be appropriate to gain access for a tracheostomy and relieve the airway obstruction.[9]

Another method of a tracheostomy was to combine the use of a rigid bronchoscopy and percutaneous tracheostomy.[10] This technique involved tracheal intubation usinga rigid bronchoscopein a dark light off operation theatre. A 30-degree telescope wasintroduced and facedanteriorly. The xenon light intensity was set to the maximum level. Anopen surgical collar incision was made following a standard skin preparation. A partial debulking of tumour might be required if the pre-tracheal mass was thick until the illumination was visualised. A 23 Gaugehypodermic needle

was inserted into the trachea with an illumination guide. Once the endoscopic control has confirmed needle position in the midline of trachea, percutaneous tracheostomy would proceed. The advantages of this technique are the airway is under control throughout the procedure, it allows reopening of airway in the case of partially obstructing endoluminal tracheal tumour, it provides suctioning of the blood and secretion adequately, and the positioning of tracheostomy tube can be performed under direct visualisation.[10] Therefore, this technique should be considered as a management of upper airway in an obstructing tumour.

CONCLUSION

This was a rare presentation of BVFI due to metastatic CRC to the thyroid. Although rare, surgeons should be aware of its possibility in spite of a patient's thin neck. This is to avoid the undesirable consequences of discovering an advanced thyroid malignancy intraoperatively. When there is difficulty identifying the tracheal rings during a tracheostomy intraoperatively, one should always consider the diagnosis of a thyroid malignancy. This will ensure that the operating team is well-prepared to handle a potentially hazardous airway when a patient presents with obstructive symptoms.

REFERENCES

1. Brady MF, Burns B. Airway obstruction. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2021 [cited 2021 Nov 22]. Available from: http://www.ncbi.nlm.nih.gov/books/NBK470562/
2. Sasidaran K, Bansal A, Singhi S. Acute upper airway obstruction. Indian J Pediatr. 2011; 78(10): 1256-61.
3. Sairin ME, Mat Baki M, Manap RA, Puteh SEW, Azman M, Mohamed AS. Body plethysmography for measuring airway resistance in patients with bilateral vocal fold immobility. Auris Nasus Larynx. 2020; 47(5): 842-8.
4. Ghossein CA, Khimraj A, Dogan S, Xu B. Metastasis to the thyroid gland: a single-institution 16-year experience. Histopathology. 2021; 78(4): 508-19.
5. Nixon IJ, Coca-Pelaz A, Kaleva AI, Triantafyllou A, Angelos P, Owen RP, et al. Metastasis to the thyroid gland: a critical review. Ann Surg Oncol. 2017; 24(6): 1533-9.
6. Lièvre A, Leboulleux S, Boige V, Travagli JP, Dromain C, Elias D, et al. Thyroid metastases from colorectal cancer: the Institut Gustave Roussy experience. Eur J Cancer. 2006; 42(12): 1756-9.
7. Haller JM, Iwanik M, Shen FH. Clinically Relevant Anatomy of Recurrent Laryngeal Nerve. Spine. 2012; 37(2): 97-100.
8. Bajwa SJS, Sehgal V. Anesthesia and thyroid surgery: the never ending challenges. Indian J Endocrinol Metab. 2013; 17(2): 228-34.
9. Testini M, Logoluso F, Lissidini G, Gurrado A, Campobasso G, Cortese R, et al. Emergency total thyroidectomy due to non traumatic disease. Experience of a surgical unit and literature review. World J Emerg Surg. 2012; 7: 9.
10. Puma F, Ceccarelli S, Potenza R, Italiani A, Melis A, Cagini L, et al. Rescue Tracheostomy for Patients With Unresectable Large Growing Neck Masses. Ann Thorac Surg. 2021; 112(5): e383-6.