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Case report

Diagnosis and surgical repair of delayed tracheal perforation post thyroidectomy in context of previous cranio-spinal radiotherapy – A case report

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ABSTRACT

Introduction and importance: This is the first case of delayed tracheal perforation post total thyroidectomy in the context of previous radiotherapy to the neck. Such a presentation can be easily misdiagnosed and managed as a seroma at significant risk to the patient, as the latter had no precipitating factors and cardiorespiratory compromise. There are nineteen previously described cases of delayed tracheal injury post thyroidectomy of variable severity and variable intervention.

Case presentation: A 51-year-old man presented with non-tender anterior neck surgical emphysema initially diagnosed on bedside ultrasound and plain X-ray, 22 days following total thyroidectomy and central neck dissection. His background was significant for childhood acute lymphoblastic leukaemia requiring chemotherapy and cranio-spinal radiotherapy. He underwent total thyroidectomy, for multiple bilateral thyroid nodules found on cranio-spinal MRI surveillance concerning for follicular neoplasm. There were significant amount of adhesions tethering the thyroid secondary to prior radiotherapy but no tracheal injury intra-operatively.

Clinical discussion: At presentation, no source of air leak was identified on Computer Tomography. He failed conservative management. During surgical exploration, a 2 mm tracheal perforation at the right cricothyroid joint was closed with the right sternothyroid muscle due to the proximity of the perforation with the recurrent right laryngeal nerve. Tisseel was applied over the repair. He recovered without further complications.

Conclusion: Sudden onset neck swelling post thyroidectomy in the context of significant scaring from radiotherapy, should raise the suspicion of surgical emphysema in the neck patients and confirmed with plain x-ray. Such patients should have multidisciplinary tertiary care.

1. Introduction

Thyroidectomy is a very commonly undertaken general surgical operation with a 3–5% complication rate [1]. Tracheal perforation after thyroidectomy is rare at a rate of 0.06% [2] and is usually identified and repaired intraoperatively [2]. Delayed tracheal perforation is even rarer with only nineteen case reports published globally with varied management. We describe the first case of delayed tracheal perforation post total thyroidectomy and lymph node dissection in patient with a history of previous radiotherapy to the neck and successful closure of the defect

using the right sternothyroid muscle flap at a tertiary hospital. The literature review emphasises the importance of timely diagnosis and treatment in a tertiary centre, given the risk of airway compromise. This case is reported in line with the SCARE 2020 criteria [3].

2. Presentation of case

A 51-year-old independent, non-smoking man, presented to the emergency department, 22 days post total thyroidectomy with sudden onset neck swelling without any cardiorespiratory instability or signs of

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Fig. 1. Surgical emphysema secondary to delayed tracheal perforation post total thyroidectomy. A: Lateral X-ray of the neck, B: Axial view of upper neck on computer tomography, C: Axial view of lower neck/superior mediastinum. Grey arrows indicating surgical emphysema on imaging. D: Anterior neck lump underlying scar from Total thyroidectomy and central neck dissection, E: Neck exploration, black arrow indicating 2mm perforation at right cricothyroid joint.

infection. He had a background of childhood acute lymphoblastic leukaemia treated with systemic chemotherapy and cranio-spinal radiotherapy between the age of 3 and 10. This was complicated by radiation induced oesophageal stenosis requiring surgical intervention as a child. He had been in remission since. He had also previously undergone resection of radiation induced right frontal lobe meningioma and was under MRI surveillance through which thyroid nodules were incidentally found. Thyroid ultrasound confirmed bilateral thyroid lobes and isthmus nodules. While he was clinically and biochemically euthyroid, the FNA biopsy of the left interpolar thyroid lesion, was of indeterminate cytology. This raised suspicion of follicular neoplasm in the context of previous neck radiation.

He underwent total thyroidectomy with central neck dissection (CND) where significant adhesions tethering the thyroid, secondary to prior radiotherapy were noted. The superior and inferior pole vessels were divided with ligaclips and diathermy. The recurrent laryngeal nerves (RLN) were identified and protected bilaterally. The left superior and inferior parathyroids were sacrificed and left sided central lymph nodes were sent for histopathology. There were no visible signs of tracheal injury intraoperatively. Valsalva manoeuvre confirmed satisfactory haemostasis, surgicel was applied, vacuum drain placed, and surgical site closed in layers. He developed left vocal cord palsy Day 1 post-operatively and was discharged three days post-operatively without any other complications.

He noticed neck swelling, 22 days post-operatively without any preceding coughing, valsalva or strenuous activity. He had no cardiorespiratory distress, no dysphagia, no systemic symptoms of infection, with a notable hoarse voice, unchanged since postoperative Day 1. He had a 7 cm wide soft, fluctuant, non-tender, anterior neck lump underlying the thyroidectomy scar. There was no palpable crepitus or signs of wound infection. An initial opportunistic bedside ultrasound followed by neck/chest X-ray and subsequent computer tomography (CT) of the neck, chest and abdomen confirmed the presence of extensive subcutaneous emphysema within the thyroidectomy bed extending superiorly in the retropharyngeal space and pre-pharyngeal space as well as the left posterior cervical space with gas tracking towards the left axilla and inferiorly into the posterior mediastinum (Fig. 1). However, the source of this surgical emphysema was not identified.

He failed conservative management under the Otorhinolaryngologists, despite an uncomplicated bedside needle aspiration on the second day of admission. During the neck exploration the next day, there were no signs of infection or necrotic tissue and both RLNs were intact. Due to difficulty visualising the defect, the neck was filled with saline and valsalva identified a 2 mm tracheal perforation at the right cricothyroid joint immediately anterior to the intact right recurrent laryngeal nerve insertion point (Fig. 1). Due to the proximity of the defect to the RLN, the right sternothyroid muscle was mobilised superiorly and rotated to cover the right tracheal defect and secured with vicryl sutures. Great care was taken to avoid handling of the RLN, especially in the context of the contralateral nerve palsy. Tisseel was applied over the defect between the trachea and the strap muscle patch. A 10 French Blakes drain was inserted superficial to the muscle patch and wound closed in layers. The drain was placed on high suction for 48 h. He was discharged on the second postoperative day. Both operations were performed by experienced consultant surgeons. He was compliant with instructions to avoid straining; the histopathology was benign; follow-ups were unremarkable and he was satisfied with the treatment provided.

3. Discussion

Thyroidectomies are very commonly performed globally with relatively low surgical risks, performed for malignant and non-malignant indications [4]. There is a reported 0% mortality rate and a less than 3% complication rate. Most tracheal injuries occur near the ligament of Berry where blood vessels are suture ligated or diathermy is used for haemostasis [2]. Delayed tracheal injuries are even rarer. Risk factors include female gender, thyrotoxic goitre [5] especially if there has been long term compression of the trachea by a large goitre. Prolonged tracheal intubation with an elevated cuff pressure can also cause ischaemia and tracheal injury [2]. Extensive fibrosis around the thyroid gland due to multinodular goitre can lead to potentially excessive use of diathermy. Tracheal necrosis may ensue over time from the thermal injury leading to perforation, subcutaneous emphysema with or without respiratory compromise [2]. Post-operative haematoma may become a nidus for bacterial infection leading to tracheal wall necrosis and perforation [2]. The patients' history may include a precipitating event

Table 1

All published cases of delayed tracheal perforations post thyroidectomy to date. POD & PPT Cause of tracheal Year and Operation Indication for thyroidectomy Intervention Location of injury Age/ Symptoms gender factors perforation per author country 2002 [5] 30F TT Hyperthyroidism 8 - Persistent Swollen neck, productive cough. Prolonged procedure NE - Trachea partially Right 2nd to 4th rings Canada cough post-op -Temp 37.8. Needle aspiration of and intubation. closed around Montgomery atelectasis air. But recurrence after coughing. Excessive cauterization. T-tube with adjacent strap infection, ligation of muscles closed in layers inferior thyroid artery around T tube and residual defect. TT and selective 2002 [7] Hong 62M Papillary carcinoma with 10 - Vigorous Surgical emphysema over right Multifactorial: FCM + NE and 2nd and 3rd tracheal rings Kong bilateral neck Mets in 3 nodes of left neck cough after neck wound - possibly from drain Intubation leading to tracheostomy necrotic dissections (Levels 2, 3, dissection. drain removal site via coughing. post-op tracheal COPD 4, and 6) ischaemia. No definitive cause 2005 [6] 53F TT Grave's disease 8 Neck swelling, extensive cervical NE, circumferential Anterior parts of 4 first rings tracheal excision with France and thoracic emphysema reported. Attributed to were necrotic. 3rd tracheal known risk factors: ring perforated. Lateral and anastomosis Possible post-op posterior membranous part of infected haematoma. trachea intact. tracheal intubation, neck surgery 2009 [1] USA 20F TT Grave's disease 7 Sudden pop, rapid swelling of face Excessive cautery OT- direct laryngoscopy, 1mmx2 mm perforation at left and neck bronchoscopy, NE, Primary anterior trachea at 1st closure with 3-0 vicryl. tracheal ring Drains placed 2009 [8] India 65M TT with bilateral Medullary carcinoma of 7 Surgical emphysema of face, and Cautery and infection NE, tracheostomy Suction 5 mm perforation with modified neck dissection thyroid T4aN1bM1 (Stage whole of upper torso, acute drain in situ and closed. necrotic edges in 2nd tracheal with maximum excision IVc) respiratory distress and intubated Decannulated 5 days post ring of lymph node mass as possible 2012 [9] 55F TT Multinodular goitre 40 Subcut emphysema. No No definitive cause FCM + NE, debridement Necrosis in frontal wall of Greece respiratory distress. Anterior neck reported. Attributed to and primary closure. No trachea. swelling - gradually worsening known risk factors drain over 2 days post admission 2012 [10] 30 M TT Grave's disease 10 - Cough CT: below cricoid cartilage. Rapid swelling in neck and face Cautery leading to Bronchoscopy: multiple Italy and an air leak from surgical ischaemia and necrosis anterolateral tracheal $2 \times$ anterior tracheal fissures 3 wound. Cough and dyspnoea fissures. NE primary suture rings below cricoid cart and 2 present repair and Tachosil over anterolateral left and right surface. $2 \times$ drains and side immediately below extubated 24 h post-op cricoid cartilage Formal bronchoscopy + NE, 2012 [11] 45 M TT and lymph node Non-invasive thyroid cancer Sudden pop and rapid swelling of Infection leading to CT: defect in cervical trachea. 4 necrosis at a point of pre-thyroid muscle flap Virtual bronchoscopy: R Italy dissection (no radiation prior) - histo face, neck and upper chest papillary cancer of thyroid relative weakness. secured with absorbable anterolateral tracheal wall, with mets at bilateral Unlikely thermal injury suture on defect. Drains 4th ring. due to use of bipolar recurrent lymph nodes placed into neck and 6 mm right anterolateral forceps anterior mediastinum tracheal wall between 2 rings 2012 [12] 65 M TT Thyroid nodules 15 Neck swelling, Unknown Conservative management 1.5 mm anterolateral Italy pneumomediastinum. fibrocartilaginous wall

Subcutaneous emphysema

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V. Seenarain et al.

4

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|---|----------------|--|--|--|--|--|--|--|
| Year and country | Age/ gender | Operation | Indication for thyroidectomy | POD & PPT factors | Symptoms | Cause of tracheal perforation per author | Intervention | Location of injury |
| 2012 [13] USA | 17F | TT | Grave's disease | 9 - Forceful sneeze while plugging nose day 7 post- operatively with right sided neck pain | Day 9: neck swelling. Stable without respiratory distress. Bilateral subcutaneous emphysema extending supraclavcularly and laterally | Cautery | FCM + NE: primary repair with interrupted sutures and Sternothyroid muscle flap for reinforcement. Suction drain. | 2.5 cm linear tear along anterior surface of trachea. From 2nd ring down to 4th ring. No necrosis. |
| 2012 [14] USA | 55F | TT | Papillary thyroid carcinoma | 30 - Cough | Worsening neck swelling, dysphagia and dyspnea | No definitive cause | Conservative management. Bedside aspiration of 300 cc of air. But spontaneous recurrence post. Admitted and drainage seroma catheter inserted and recovered uneventfully | Location of injury not assessed |
| 2013 [15] Italy | 39F | Trans-axillary robot- assisted left hemi- thyroidectomy | Benign 5 cm nodule in left thyroid lobe | 40 - Sneeze | Popping sound and sudden subcutaneous emphysema | Inadvertent injury from harmonic scalpel -> ischaemia, necrosis & perforation | Bronchoscopy. Endoscopically curetted hole and fibrin glue applied. Follow up bronchoscopy – healed. | 2 mm rounded defect in anterior wall of trachea between 1st and 2nd rings |
| 2014 [16] Italy | 17F | Total thyroidectomy with functional right lymphadenectomy. | Papillary thyroid carcinoma with lateral cervical adenopathies and lung Mets | 7 | Dyspnea, dysphonia, cough and subcutaneous emphysema extending to supraclavicular area and neck bilaterally | Malignancy, cautery and endotracheal cuff pressure leading to ischaemia and necrosis | Bronchoscopy and Neck exploration with muscle flap – clavicular part of left SCM and left and right pre- thyroid muscles used over tracheal defect. | CT and Bronchoscopy: suspected tear on right lateral wall 2 cm under glottis caudally extended for 3 cm. Intraop– 3 cm wide irregular tracheal defect on right antero-lateral wall from 2nd ring down to 5th ring with ++ necrotic tissue on tear's edge |
| 2016 [17] China | 39F | Right lobe thyroidectomy and right recurrent laryngeal nerve exploration. | Solid nodules in right thyroid lobe | 3 - Severe cough day 1 post- operatively | Day 3 – haemoptysis, dyspnea, neck swelling, pain and fever, worsening cough, choking and deglutition. | Infection | Bronchoscopy with covered tracheal stent 20mmx60mm over tracheal defect (Interventional radiologists) | Tracheography – rupture on right wall 25 mm below throat. CT 13 days post op showed abscess 50x60x40mm in right antero-superior mediastinum with gas/liquid interface- aspirated |
| 2016 [18] New Zealand | 29F | Total thyroidectomy | Tracheal impingement in context of moderately enlarged right thyroid lobe and clinically normal left lobe. | 14 - Cough | Day 14 - bitter tasting fluid in throat, anterior neck swelling with coughing and audible noises with inspiration. Palpable surgical emphysema around wound. | Metal ligating clips associated with infected seroma (D11 post-op) leading to tracheal perforation | FCM + NE: Found moderate mucus, inflamed tissue, free air. Sternohyoid muscle transposition myovascular flap to cover defect. Tisseel tissue bonding agent also used. Low suction drains inserted | CT – 2.5 mm defect in right posterior lateral trachea – posterior to the cartilaginous ring. Posterior aspect of trachea |
| 2017 [19] Country unknown - published in Saudi Journal of Anaesthesia | 20M | Total thyroidectomy with central compartment lymph node dissection | Papillary thyroid carcinoma | 2 - Cough/ sneeze | Surgical emphysema, swelling with respiratory distress | No definitive cause. Attributed to known risk factors | Neck exploration with debridement of necrotic edges and tracheostomy fashioned | 1 cm hole with necrotic edges found at anterolateral surface of trachea at level of 1st and 2nd tracheal ring |

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Table 1 (continued)

| Year and country | Age/ gender | Operation | Indication for thyroidectomy | POD & PPT factors | Symptoms | Cause of tracheal perforation per author | Intervention | Location of injury |
|--------------------|----------------|---|---|--|--|--|---|---|
| 2018 [20] India | 48M | Total thyroidectomy and ipsilateral neck dissection | Papillary carcinoma of thyroid T4a N1b M0 - extension into para- pharyngeal space, invading strap muscles and internal jugular vein. Ipsilateral recurrent laryngeal nerve grossly involved and sacrificed. Trachea and hypopharynx not involved. Tumour scraped off cricothyroid membrane superiorly | 5 during index admission | Emphysema appeared intermittently and reduced on deflation of suction drain | Cautery | Conservative management with pressure dressing | Location of injury not assessed |
| 2018 [21] USA | 25M | Right hemithyroidectomy and isthmusectomy | 4 cm Papillary carcinoma – classical variant and 8 mm Papillary thyroid microcarcinoma | 27 - Strenuous exercise and valsalva | Seen in clinic Day 21 for routine follow up - compliant with activity restrictions. Advised can restart exercise gradually. Day 27 – neck lump immediately after set of pullups. Improved on its own but recurrence with Valsalva – seen in ED and discharged. Day 29 seen in clinic – needle aspiration – pressure dressing. Admitted for observation | Cautery and devascularisation near Ligament of Berry - weakness exacerbated by pullups | Conservative management. Bronchoscopy. Bedside neck exploration – Evacuation of air pocket successful. Penrose drain inserted under strap muscles at level of tracheal injury. Skin closed and pressure dressing applied. Discharged next day | CT: right lateral trachea at level of 1st and 2nd tracheal ring. Bronchoscopy – 1 mm mucosal irregularity along right lateral tracheal wall inferior to cricoid + scant overlying granulation tissue. |
| 2020 [4] USA | 24M | Total thyroidectomy with bilateral neck dissections | Metastatic papillary thyroid cancer - Left recurrent laryngeal nerve sacrificed due to tumour involvement. <i>Re</i> -intubated due to stridor and respiratory distress on table. Extubated day 1 post. Vocal fold injection augmentation on Day 5 - noted granulation tissue on anterior tracheal wall extending inferiorly approx. 5 mm. suspected due to recent intubation. Biopsy taken – fibropurulent exudate and few reactive squamous cells | 10 but presented to follow up clinic day 12 - Cough and valsalva | Neck swelling with cough or valsalva. CT – ventral cervical and mid tracheal defects with extensive mediastinal and subcutaneous emphysema | Endotracheal tube cuff pressure | Neck exploration with partial sternotomy – superior defect closed with SCM flap and inferior defect with pectoralis flap. Due to evolution of tracheal injury inferior repair had to be revised approximately 2 weeks later – additional necrotic trachea debrided – closed with thick acellular dermal matrix allograft over silicone stent and reinforced with omental flap. Stent removed after 1 week. Tracheostomy done through previously repaired proximal defect | 1 cm defect of anterior wall at rings 2 and 3. 2nd defect at anterior inter-cartilaginous membranes from rings 9–12. |

TT: total thyroidectomy, Mets: metastasis, NE: neck exploration, FCM: failed conservative management, POD: post-operative day of presenting symptoms, PPT: precipitating.

International Journal of Surgery Case Reports 91 (2022) 106761

such as a cough or sneeze. Common presenting signs include face and/or neck swelling, haemoptysis, and retrosternal pain. Computed tomography of the neck is the preferred imaging modality to identify the cause and extent of subcutaneous emphysema.

Nineteen case reports describing delayed tracheal injury post thyroidectomy were found to date (Table 1). The average age of patients was 38. 52% were female. 84% of cases had total thyroidectomies. 47% of thyroidectomies were for malignant indications and none had previous radiotherapy of the neck. Patients developed symptoms of delayed tracheal perforation at post-operative day 2 and as late as post-operative day 40. 58% of patients had a documented trigger for perforation such as coughing, sneezing, valsalva or strenuous exercise. Cautery contributed to 47% of delayed tracheal perforations. No definite cause was attributed to 21% of cases with some describing a combination of surgical risk factors contributing to tracheal perforation [6]. Infection contributed to 26% of tracheal perforations. 47% of cases were managed surgically from the outset, 32% were managed non-surgically, and 21% failed conservative management, requiring neck exploration. Two nonsurgically managed patients underwent bronchoscopic interventions (Table 1) and two did not require any form of bedside intervention. The commonly affected areas of the trachea were right anterolateral and anterior surface with the most proximal tracheal ring levels. At neck exploration, 23% were managed with debridement and formation of a tracheostomy as their definitive treatment. 31% underwent primary suture closure with or without overlying muscle flap. 23% underwent transposition of a myovascular flap to cover the tracheal defect without primary suture closure. One case underwent partial sternotomy due to the extent of the tracheal injury and due to evolution of the tracheal injury further underwent a tracheostomy through the defect [4].

Most patients were managed surgically involving a multidisciplinary input, however, there is scope for conservative management. Difficulty in identifying the correct planes for dissection especially in a setting of extensive fibrosis between the thyroid and trachea in patients with multinodular goiter was also a factor contributing to tracheal perforation [2]. Thyroid malignancy not thought to be a major contributing factor [2]. Amongst the published case reports (Table 1) there were slightly more non-malignant indications for thyroidectomy associated with tracheal injury and none had undergone any radiotherapy preoperatively. However total thyroidectomy was clearly a major risk factor contributing to tracheal perforation.

In this patient, extensive adhesions secondary to previous radiotherapy was a major risk factor for delayed tracheal perforation. Due to the suspicion of potential malignancy, central neck dissection in this setting was warranted. The injury in this case occurred close to the ligament of berry. While the defect size was 2 mm wide, its close relation to the RLN, lead to the choice of a sternothyroid patch repair of the defect rather than primary suturing. The preservation of the right RLN was paramount given the pre-existing contralateral nerve palsy. Tisseel was used as an adjunct to seal the defect between the trachea and the strap muscle. A 10 French Blakes drain overlying the muscle patch on high suction for 48 h was placed for additional negative pressure within the thyroidectomy bed to minimise recurrence. These techniques successfully treated the perforation without complication.

4. Conclusion

Rapid onset of neck swelling post thyroidectomy, and presence of subcutaneous emphysema are indicators of delayed tracheal perforation. Suspicion for delayed tracheal perforation should be raised if the index surgery was difficult due to adhesions or issues with haemostasis as this may indicate increased use of cautery and other dissecting instruments. An x ray of the neck can be used to rule out surgical emphysema within the thyroidectomy bed. CT is the imaging of choice to determine the source of perforation but may not always be successful. Such patients with delayed tracheal perforation post thyroidectomy should be managed in a tertiary hospital in a multidisciplinary setting.

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Ethical approval

This article is exempt from ethical approval per the Head of Department of the Acute surgical Unit at Fiona Stanley Hospital, Perth, Western Australia. The exemption of ethical approval for this article will be indicated in the article.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution statement

Corresponding author: Dr. Vidya Seenarain – Patient interview, primary author and editor.

Other authors

- Mr. Anand Trivedi proof-reader and editor of the manuscript
- Dr. Stephanie Flukes proof-reader and editor of the manuscript
- Mr. William Edward Tjhin proof-reader and editor of the manuscript

Ethics

This article is exempt from ethics approval.

Guarantor

 $\label{eq:Dr.Vidya} Dr.\ Vidya\ Seen arain-Corresponding\ author-documented\ in\ author\ form.$

Declaration of competing interest

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V. Seenarain et al.

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