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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\textbf{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 scarring 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...
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 then. 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, vallsalva or strenuous activity. He had no cardio-respiratory 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 vallsalva 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 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.

| Year and country | Age/ gender | Operation | Indication for thyroidectomy | POD & PPT factors | Symptoms | Cause of tracheal perforation per author | Intervention | Location of injury |
|------------------|-------------|-----------|------------------------------|-------------------|----------|----------------------------------------|-------------|--------------------|
| 2002 [5] Canada  | 30F         | TT        | Hyperthyroidism              | 8 - Persistent cough post-op - atelectasis | Swollen neck, productive cough. Temp 37.8. Needle aspiration of air. But recurrence after coughing. | Prolonged procedure and intubation. Excessive cauteryization, infection, ligation of inferior thyroid artery and residual defect. | NE - Trachea partially closed around Montgomery T-tube with adjacent strap muscles closed in layers around T tube and residual defect. | Right 2nd to 4th rings |
| 2002 [7] Hong Kong | 62M         | TT and selective bilateral neck dissections (Levels 2, 3, 4, and 6) | Papillary carcinoma with Mets in 3 nodes of left neck dissection. | 10 - Vigorous cough after drain removal - COPD | Surgical emphysema over right neck wound - possibly from drain site via coughing. | Multifactorial: Intubation leading to post-op tracheal ischaemia. | FCM + NE and tracheostomy | 2nd and 3rd tracheal rings necrotic |
| 2005 [6] France  | 53F         | TT        | Grave’s disease              | 8                 | Neck swelling, extensive cervical and thoracic emphysema | No definitive cause reported. Attributed to known risk factors: Possible post-op infected haematoma, tracheal intubation, neck surgery | NE, circumferential tracheal excision with anastomosis | Anterior parts of 4 first rings were necrotic. 3rd tracheal ring perforated. Lateral and posterior membranous part of trachea intact. |
| 2009 [1] USA    | 20F         | TT        | Grave’s disease              | 7                 | Sudden pop, rapid swelling of face and neck | Excessive cautery | OT- direct laryngoscopy, bronchoscopy, NE, Primary closure with 3-0 vicryl. Drains placed | 1mmx2 mm perforation at left anterior trachea at 1st tracheal ring |
| 2009 [8] India  | 65M         | TT with bilateral modified neck dissection with maximum excision of lymph node mass as possible | Medullary carcinoma of thyroid T4aN1bM1 (Stage IVc) | 7                 | Surgical emphysema of face, and whole of upper torso, acute respiratory distress and intubated | Cautery and infection | NE, tracheostomy Suction drain in situ and closed. Decannulated 5 days post | 5 mm perforation with necrotic edges in 2nd tracheal ring |
| 2012 [9] Greece | 55F         | TT        | Multinodular goitre          | 40                | Subcut emphysema. No respiratory distress. Anterior neck swelling - gradually worsening over 2 days post admission | No definitive cause reported. Attributed to known risk factors | FCM + NE, debridement and primary closure. No drain | Necrosis in frontal wall of trachea. |
| 2012 [10] Italy | 30 M        | TT        | Grave’s disease              | 10 - Cough        | Rapid swelling in neck and face and an air leak from surgical wound. Cough and dyspnoea present | Cautery leading to ischaemia and necrosis | Bronchoscopy: multiple anterolateral tracheal fissures. DE primary suture repair and Tacho: osil over surface. 2× drains and extubated 24 h post-op Formal bronchoscopy + NE, pre-thyroid muscle flap secured with absorbable suture on defect. Drains placed into neck and anterior mediastinum | CT: below cricoid cartilage. 2× anterior tracheal fissures 3 rings below cricoid cart and 2 anterolateral left and right side immediately below cricoid cartilage |
| 2012 [11] Italy | 45 M        | TT and lymph node dissection | Non-invasive thyroid cancer (no radiation prior) - histo – papillary cancer of thyroid with mets at bilateral recurrent lymph nodes | 4                 | Sudden pop and rapid swelling of face, neck and upper chest | Infected leading to necrosis at a point of relative weakness. Unlikely thermal injury due to use of bipolar forcesps | Unknown | CT: defect in cervical trachea. Virtual bronchoscopy: R anterolateral tracheal wall, 4th ring. 6 mm right anterolateral tracheal wall between 2 rings |
| 2012 [12] Italy | 65 M        | TT        | Thyroid nodules              | 15                | Neck swelling, pneumomediastinum. Subcutaneous emphysema | Conservative management | 1.5 mm anterolateral fibrocartilaginous wall | (continued on next page) |
| 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 supraclavicularly 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 Bronchoscopy. | Location of injury not assessed |
| 2013 [15] Italy| 39F         | Trans-axillary robot-assisted left hemithyroidectomy | Benign 5 cm nodule in left thyroid lobe | 40 - Sneeze | Popping sound and sudden subcutaneous emphysema | Inadvertent injury from harmonic scalpel : ischaemia, necrosis & perforation | 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 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 CT – 2.5 mm defect in right posterior lateral trachea – to the cartilaginous ring. Posterior aspect of trachea |
| 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 20mmx50mm over tracheal defect (Interventional radiologists) | |
| 2016 [18] New Zealand | 29F | Total thyroidectomy in context of moderately enlarged right thyroid lobe and clinically normal left lobe. | Tracheal impingement | 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. Tissue bonding agent also used. Low suction drains inserted | |
| 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 |
| Year and country | Age/ gender | Operation and dissection | Indication for thyroidectomy factors | POD & PPT 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 parapharyngeal 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 | Location of injury not assessed |
| 2018 [21] USA | 25M | Right hemithyroidectomy and isthmusectomy | Papillary thyroid microcarcinoma | 27 - Strenuous exercise and valsalva | Severe swelling | 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. Re-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 tracheal debridged – 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.
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 non-surgically 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 tracheal injury and due to evolution of the tracheal injury further underwent a tracheostomy through the defect [4].

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

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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

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Guarantor

Dr. Vidya Seenarain – Corresponding author – documented in author form.

Declaration of competing interest

Authors have no conflicts of interest to disclose.

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