Multi-nodular Goiter with Intra-tracheal Thyroid Tissue: Ectopic or Implanted? And Ectopic Nasopharyngeal Thyroid Tissue: A Case Report

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Abstract

We present a case of intra-tracheal thyroid tissue in a 44 year old woman who had been treated for multi-nodular goiter with left hemi-thyroidectomy 26 years earlier, the thyroid tissue was found in the trachea lumen which may appears to have been implanted at the time of this operation or it is a case of ectopic thyroid tissue. The patient presented with progressive right thyroid lobe swelling. Clinical examination and investigations were carried out. Right thyroidectomy was performed first, then after discovering in laryngostroboscopy, CT scan and MRI, the presence of intra-tracheal lesion narrowing the airways, patient had another procedure to remove the residual left thyroid lobe and the intra-tracheal tissue which was thyroid tissue as confirmed by histology examination. Post-operative I131 whole body-scan showed ectopic thyroid tissue in nasopharynx and residual tissue in thyroid bed. Currently the patient is free of complaint, but further investigation: SPECT-CT Scan was requested for better localization.

Keywords: Thyroid gland; Ectopic thyroid; Implanted thyroid tissue; Trachea

Background

The ectopic thyroid is uncommon with the prevalence rate of approximately 1 per 100,000 – 300,000 persons. More than 440 cases have been reported in Europe, America, and Asia to date [1-5]. Anatomically, the most common sites of ectopic thyroid tissue are lingual, sublingual, thyroglossal and laryngotracheal. In rare cases, esophageal, mediastinal, cardiac, aortic, adrenal, pancreatic, duodenal, gallbladder, mesentery of the small intestine, cutaneous and intra-tracheal sites have been found [6-15]. There is also unique cases of ectopic thyroid tissue include pituitary fossa, sphenoid sinus and uterus [16-18]. To our knowledge, this is the first case of ectopic nasopharyngeal thyroid tissue have been reported or published to date. Additionally there have been rare reports of Implantation of thyroid tissue following blunt trauma, fine needle aspiration biopsy and endoscopic thyroid surgery [19-25].

Case Presentation

A 44- year- old woman, nonsmoker, presented with progressive painless right sided neck swelling since more than 2 years, on examination she has right thyroid lobe enlargement, freely movable, not fixed to the skin, nodular, diffuse and about 4*4 cm in size, with other complaints of increasing dyspnea and swallowing difficulty since few months, she had been treated for asthma by her general practitioner. 26 years earlier, the patient had been treated for multi-nodular goiter with left hemi-thyroidectomy with subsequent thyroxin replacement therapy. There is family history of thyroid goiter in her sisters and she is allergic to penicillin. During the current presentation, clinical examination showed painless nodular right thyroid lobe swelling adherent and displacing the trachea, together with tremor, tachycardia and some stridor in deep inspiration, thyroid function tests were normal. Indirect laryngoscopy showed restricted mobility of left vocal cord only. The patient underwent completion thyroidectomy by right hemi-thyroidectomy, during the surgery, there was infiltration of thyroid tissue in between the tracheal rings, and small air leak was detected from trachea after shaving off the thyroid tissue which repaired by using prolene 4-0 suture. Histologically: nodular hyperplasia (multi-nodular goitre), no evidence of malignancy.

On post-operative first day, patient developed biphasic stridor without any sign of neck swelling or hematoma, laryngostroboscopy showed restricted mobility of left vocal cord and submucosal rounded smooth intraluminal lesion on the right side causing marked narrowing in subglottic area, and this low mobility of left vocal cord can be attributed to the previous surgery on left side (Figure 1). Computed Tomography Scanning of the neck showed submucosal intraluminal mass extending from subglottic area to about the level of third tracheal ring, causing narrowing more than 50% of the airways (Figures 2-5). Magnetic Resonance Imaging (MRI) of the neck showed enhancing nodule mainly in the left side of the tracheal lumen in close relation to the left thyroid lobe, suggestive of either an ectopic thyroid nodule or abnormally implanted thyroid tissue post prior surgeries (Figure 6).

Figure 1: Laryngostroboscopy showed right sided intraluminal submucosal lesion narrowing the airways.

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days later, starting with surgical tracheostomy and then we were unable to remove the intra-tracheal lesion sub-mucosally using debrider and bronchoscope, so we did an external approach (Laryngofissure) by median cricoidotomy extending through first three tracheal rings, exposing the endo-subglottis area and endo upper trachea, sub-mucosal removal of lesion, re-approximating and suturing mucosal edges, suturing of cricoid and trachea bu absorbable sutures, following by completion thyroidectomy in left side and finishing by closure of tracheostomy, frozen section of this intra-tracheal lesion confirmed thyroid tissue.

Following surgery, the patient was kept intubated in ICU for 6 days to keep endotracheal tube in place as a stent, and then Micro-laryngotracheoscopy under general anesthesia was done and revealed patent airways so patient was extubated with uneventful postoperative course without any stridor or respiration difficulty, patient was discharged few days later. Endoscopic control after two weeks showed fixed left vocal cord, mobile right vocal cords and clear subglottic area (Figure 7).

I131 whole body Scan was performed after 1 month and showed

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Drain was removed next day, patient still complains of stridor, so patient was planned for excision of the tracheal lesion with or without surgical tracheostomy and tracheal stenting. Surgery was done two
Intra-tracheal mass and biopsy is required for a histologic diagnosis. Laryngoscopy is recommended in order to visualize directly the extension and location of the ectopic thyroid gland and fiberoptic tool for ectopic thyroid but CT scan and MRI may help in defining the presence of thyroid remnants in the bed of the previous surgery so endocrinologist advised the radioiodine ablation for the ectopic nasopharyngeal thyroid tissue in our present case. A variety of surgical techniques have been applied in the management of ectopic intra-tracheal thyroid tissue. Patients have two options: removal of the lesion via an open cricoid procedure, as we did in this case, or removal via an endoscopic laser-assisted approach.

In this case report, the intra-tracheal thyroid tissue was most probably dual ectopic foci with the other foci in the nasopharynx, but there is another weak probability that it was directly implanted into the tracheal lumen during the patient’s initial treatment for multi-nodular goitre in right lobe of the thyroid gland, while details of that surgical procedure were not available, implantation might occurred during surgery and the resultant remnant remained quiescent for 26 years before enlarging and ecching symptomatic. There are rare reported cases of thyroid tissue implantation following blunt trauma, fine needle aspiration biopsy and endoscopic thyroid surgery, and there are only five documented cases of intra-tracheal thyroid tissue associated with previous thyroidectomy [33-36]. These patients presented 4 to 21 years after their original thyroid surgery, with dyspnea or stridor, the tissue was resected or iopsied in all cases, and found to contain benign thyroid tissue.

In this case a pre-op indirect laryngoscopy to assess vocal cord movement and airway patency was not enough to put a pre-op diagnosis and therefore a better surgical strategy and plan, also there was no any preoperative radiologic investigations such as ultrasound or CT Scan that would be helpful in planning surgery and would know the presence of thyroid remnants in the bed of the previous surgery so it may prevent severe subsequent morbidity.

Conclusions

Even ectopic intra-tracheal thyroid tissue is rare, it should be considered as a possible cause of airway obstruction, particularly in those who have goiters. The current case highlights the possibility of Intra-operative thyroid tissue implantation in the trachea and shows the need for careful tissue handling during thyroid surgery, so ectopic thyroid tissue should be considered in the differential diagnosis even in the presence of a normal thyroid gland.

Standard thyroid workup before any surgical intervention should include imaging such as thyroid ultrasound or CT Scan and an assessment of the vocal cords by fiberoptic nasolaryngoscopy or laryngostroboscopy to ensure the status following previous surgery and prior to any further surgical treatment.

Consent

Written informed consent was obtained from the patient for publication of this Case Report and any accompanying images.
Authors’ Contributions

MSJ: conception and design, writer, revision and approval of the final version. JK, HT: conception and design, revision and approval of the final version. GY: conceived of the study, and participated in its design and coordination and helped to draft the manuscript. AA: revision and approval of the final version.

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