CASE REPORT

Intratracheal ectopic thyroid: A rare cause of upper airway obstruction

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Abstract

Intra-tracheal thyroid is a rare ectopic thyroid disease. Presenting symptoms can mimic those of airway obstruction such as bronchial asthma. We report the case of a male patient who presented with airway obstruction due to thyroid ectopia and was misdiagnosed initially with bronchial asthma. After a correct diagnosis of intra-tracheolaryneal thyroid, the case was managed by excision. Surgery is the mainstay of treatment for patients with clinical signs of airway obstruction. We present our experience of his one-month follow-up after resection. Currently, the patient is free of symptoms.

Keywords: Intra-tracheal thyroid, Ectopic thyroid, Airway obstruction.

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Introduction

Ectopic thyroid is a condition in which thyroid tissue can be present outside its normal anatomical location. In the general population, prevalence ranges from one per 100,000 to 300,000 and one in 4,000 to 8,000 individuals who suffer from thyroid illness.[1] Ectopic thyroid tissue can be seen anywhere from the base of the tongue to porta hepatis.[2] In 90% of the cases with ectopic thyroid, the tissue is seen in the base of the tongue, and 70% of the cases do not have a thyroid gland in the typical anatomical site.[3] The most common symptoms include dyspnoea (41.86%), breathlessness (18.60%), and stridor (11.63%), with symptoms often being more severe in pregnant women due to increased physiological demands. Most of these individuals are euthyroid.[4]

The recent visit of a male with upper airway obstruction and ectopic thyroid tissue in the subglottic and epiglottic areas presented challenging diagnostic and surgical technical problems. Early diagnosis of intra-tracheal thyroid is challenging due to its concealed location and initial misdiagnosis as airway pathology, such as asthma. Treatment options available are an endoscopic resection and open surgery with an initial tracheostomy. The open surgery approach includes enucleation of the mass to partial or full-thickness tracheal resection and laryngectomy.[5] Literature review revealed that only limited data is available to guide the management of this condition. Pathogenesis proposed to explain intra-laryngotracheal thyroid is not clear yet. This case report of intra-tracheal thyroid (ITT) will help address the gaps in knowledge of this rare ectopic tissue.

This case report describes the management of a young patient who was being treated for asthma for a long time. Having excluded malignancy, the goal of our surgery was to avoid tracheal resection as much as possible in a very young patient.

Case Report

A 34-year-old male, a non-smoker, had been experiencing shortness of breath, dry cough, and exertional dyspnoea for the past five years. Gradually, the symptoms progressed and he had multiple acute episodes of shortness of breath with a wheeze described as "asthma-like symptoms". He did not have a previous history of asthma or any respiratory or cardiovascular illness. He was being treated for his symptoms with bronchodilators since then, without Pulmonary Function test (PFT). His condition, however, did not improve significantly; instead his symptoms became worse as he started experiencing dyspnoea on rest as well. During the past six months, he visited the hospital multiple times for acute dyspnoea and was treated for asthma. Given his non-responsiveness to treatment, he was referred for further investigations. The case was first seen in Recep Tayyip Erdogan Hospital, Muzaffargarh in September, 2023 where his PFTs were performed by a pulmonologist. The findings were not indicative of asthma, rather suggesting intra-tracheal obstruction. These findings prompted further investigation. The patient underwent tracheobroncho-scopic examination which revealed the presence of subglottic mass causing obliteration of most of the tracheal lumen (Figure 1). A biopsy of the mass and tracheostomy was done to relieve the symptoms. Histopathology and immunostaining studies confirmed benign thyroid tissue.

A thyroid scan showed homogenous tracer uptake in both lobes of the thyroid gland. The patient had no history of thyroid disease, diabetes, or malignancy. Thyroid function

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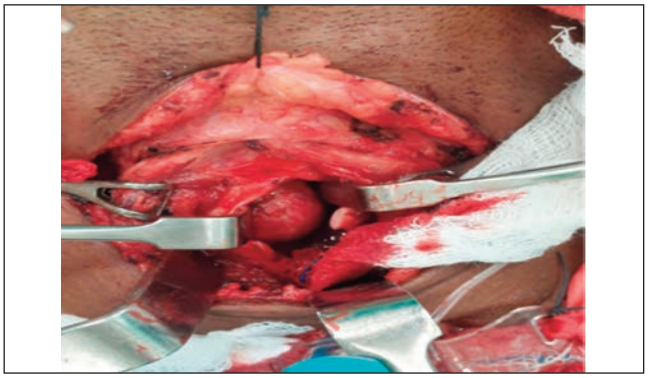

Figure-1: A mass protruding into the tracheal lumen, attached to the right posterolateral wall of trachea.

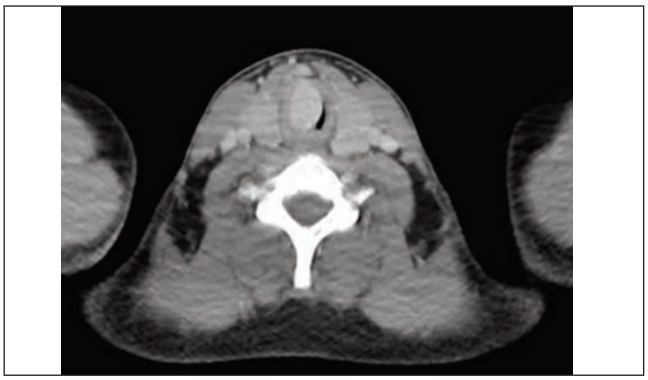

Figure-2: Contrast enhanced axial images of computed tomogram: a mass attached to the right posterolateral tracheal wall, occluding 80-90% of the tracheal lumen.

tests were suggestive of subclinical hypothyroidism. Physical examination showed a young male with a double lumen tracheostomy tube in place, having no dyspnoea, cyanosis, clubbing, or lymphadenopathy. There was no palpable lump in his neck, no lymphadenopathy, and the chest was clear. His deep tendon reflexes were normal and no eye prominence or lid lag was noted.

Contrast-enhanced computed tomography of the neck and chest revealed a well-defined, homogenous, soft tissue mass of 2.2x2.7x1.8 cm (craniocaudal, anteroposterior, transverse) in the subglottic area, opposite to the C6 vertebrae originating from the right lateral wall of the trachea (Figure 2). There was a 2 mm patency between the mass and the opposite tracheal wall. The mass was more enhancing than normally placed thyroid gland on post-contrast images. There was no cervical lymphadenopathy.

Flexible fibre-optic bronchoscopy confirmed the growth and extent of luminal obstruction. Examination showed bilateral normal vocal cord movement, a mass just below the vocal cords originating from the right lateral wall with a broad base and obliterating 80-90% of the lumen of the trachea. The overlying mucosa was normal and the scope was not negotiable; hence, evaluation of the lower trachea and bronchi was not possible. Another growth was noted just below the epiglottis originating from the left lateral wall of the larynx, it was not causing lumen obstruction.

For confirmation of diagnosis and treatment, excision of the mass was planned. On admission, TSH was >100 mIU/L (normal range: 0.27-4.2) and free T4 was 3.88pmol/L (normal range: 12-22). Due to the urgency of relieving airway obstruction, surgery was prioritised, and Levothyroxine was not initiated pre-operatively to avoid delaying the procedure. Excision was done through a transverse incision at the level of the cricoid cartilage. The trachea was opened by cutting the first three tracheal rings. A 3 x 3 cm of mass was excised, and there were no attachments to the normally positioned thyroid tissue (Figure 1). The flaps were re-sutured. The post-operative period remained uneventful and the patient was discharged from the hospital on the fourth post-operative day and recovered well.

Histopathology showed nodular thyroid tissue, with variable-sized thyroid follicles lined by bland cuboidal epithelium, and filled with colloid. There was no evidence of malignancy. The patient was weaned from a tracheostomy tube and remained symptom-free. Thyroxine supplements were started after consultation with an endocrinologist, as the patient's thyroid function tests indicated primary hypothyroidism post-surgery. Periodic follow-up in the endocrine and surgical clinic and direct laryngoscopic examination were advised.

Discussion

The thyroid gland originates from the floor of the pharynx as an ectoderm invagination from the first pharyngeal pouch; this diverticulum subsequently grows into thyroid tissue. Concurrently, the pharyngeal floor gives rise to the thyroglossal duct, which connects to the base of the tongue and atrophies in the fifth week. The developing thyroid gland descends from the base of the tongue down transversally to its pre-tracheal position by the seventh week of gestation as a result of the forward growth of the pharynx. Ectopic thyroid tissue can be present at different locations and one of the rarest locations is intra-laryngotracheal. Ectopic thyroid tissue can be on the descending route of the thyroglossal duct including supra and infrahyoid or the thyroid tissue present away from the normal route of descent of the thyroid gland or thyroglossal duct, such as those associated with mediastinal and subdiaphragmatic organs.4 Intra-tracheal thyroid does not appear to belong to any of these two subgroups and the pathogenesis is still controversial.

The uncommon nature of intra-tracheal ectopic thyroid makes the diagnosis tough. Careful examination and the patients presenting with airway obstruction but not responding to bronchodilators can be a clue to this rare disease. An ITT could be suspected in the case of an external goiter and has been observed in three-quarters of the cases.[5] Among all the endotracheal tumours, intra-tracheal ectopic thyroid tissue accounts for 6% to 7%. The differential diagnoses include papilloma, enchondroma, osteoma, chondrosarcoma, squamous cell carcinoma, adenoid cystic carcinoma, lymphoma, and thyroid cancer with intra-tracheal invasion, and these should be differentiated from benign intra-tracheal thyroid. Biopsy is critical in the diagnosis. Bleeding, multiple nodules, and ulceration should raise the suspicion of cancer. Special attention should be given to lymph node enlargement in case of suspicious findings.[6]

The thyroid status in ectopic thyroid cases is usually euthyroid or hypothyroid. Imaging provides anatomical clues.

The first case of intra-tracheal thyroid was reported by Ziemssen in 1875, and the first successful removal of the thyroid was performed by Heise in 1988.[7] The prevalence of malignant change in intra-tracheal thyroid may be as high as 11% in previously cited reports, however, recent systemic review has shown it to be 4.6% owing to the better pathological technique.[4] The most common malignancy is papillary thyroid carcinoma.[6]

Theories proposed to explain the intra-laryngeotracheal thyroid are not completely appealing. One of them "malformation theory" describes it as foetal malformation of the thyroid gland, arising during early embryonic life, where the developing thyroid gland gets encroached upon and divided by later developing tracheal cartilage rings. Another theory known as "ingrowth theory" describes it as an ingrowth of thyroid tissue into the tracheal lumen through tracheal cartilage by a properly formed mature thyroid.ii Imaging and intra-operative findings have shown intra-tracheal thyroid connected to the extra-tracheal normally positioned thyroid gland, in relation to the position of Berry's ligament connected with it through a fibrous stalk in 59.5% of cases. Such findings suggest that Berry's ligament can be involved in the development of intra-tracheolaryngeal ectopic thyroid.[4]

In 96.7% of the cases, intra-tracheal thyroid is seen in the upper trachea between the first and third rings, while in 2.3% it is seen above the carina. The prevalence of ectopic endotracheal thyroid disease is 10:3 more in females than in males. It can present from day one to 85 years of age; however, third to fifth decade is the most prevalent presentation age.[4]

Diagnosis of intra-tracheal thyroid is based on radiological and endoscopic features. Radionuclide studies are highly sensitive and specific in detecting functional ITT.[6]

An unusual aspect of this case was the presence of thyroid tissue in the upper trachea as well as the larynx at the level of epiglottis. Moreover, there was no evidence of nodularity in the normally placed thyroid tissue. The treatment options available to date for ectopic thyroid tissue are open or endoscopic excision, radioiodine ablation, and thyroid suppression therapy.[6] The choice of treatment depends on the size and location of the lesion, symptoms, age of the patient, thyroid function status, and histological findings.

Post-mortem anatomic studies suggest that intra-laryngeotracheal thyroid is not very rare, it becomes evident in a person's life when it is associated with airway obstruction or endocrine dysfunction. Intra-laryngeo-tracheal thyroid should be considered in patients with gradually progressive upper airway obstructive symptoms with no obvious external compression. The current patient had hypothyroidism as shown by low T4 and high TSH levels, this physiological alteration can lead to thyroid stimulation and growth.[2] However, in the present case, only ectopic thyroid tissue showed increased growth and the normally placed thyroid did not have any enlargement.

Before definitive surgery is undertaken an elective tracheostomy is a prerequisite. The cartilaginous skeleton of the trachea should be preserved in order to provide support. Hemithyroidectomy is not an absolute indication of benign ITT. Endoscopic surgery, laser-assisted approach, or radioiodine ablation have only limited success and are offered to patients with poor health conditions, advanced disease, and those who deny open procedure.[4]

Conclusion

Intra-tracheal thyroid cases are rare, and the presenting symptoms could mimic airway disease, such as asthma, and can lead to misdiagnosis until it causes severe airway obstruction. The mechanism of ITT pathogenesis is still unclear and needs further research. It poses a unique challenge to clinicians in terms of diagnosis and management. Treatment is excision through an endoscopic or open approach, the open approach being the conventional definitive treatment, which includes excision of ectopic thyroid tissue along with the stalk if present and reconstruction of tracheal flaps, it has low post-operative complications and better results.

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Disclaimer: Written consent from the patient was taken to publish this case report, including any accompanying images. The authors and Recep Tayyip Erdogan Hospital accept no liability for the accuracy, completeness, or reliability of the information in this case report. The medical decision should be based on sound clinical judgment and the latest evidence-based guidelines.

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Author Contribution:
SEU: Concept, design, data collection, analysis and writing.
MIH: Drafting, revision, final approval and agreement to be accountable for all aspects of the work.
FM: Data collection and design.