



A Case of Double Ectopic Thyroid in the Mediastinum and Thymus: A Case Presentation and Literature Review

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Abstract

Primary ectopic mediastinal thyroid is a rare presentation. It is only present in a small subset of patients with ectopic thyroid tissue. A patient with two different foci of ectopic thyroid tissue, one in the mediastinum and in the thymus, is an even rarer presentation. We discuss our clinical case as well as review the literature and present a table with cases of double and triple ectopic thyroid tissue.

Case Presentation: In our case report, we present a patient with an ectopic thyroid tissue of the mediastinum and thymus. Our patient originally presented with a multinodular goiter. This was resected unremarkably. The patient presented later with symptoms of continued cough, which prompted a CT scan chest showing a thymic mass and retained thyroid tissue of the anterior neck. This was found to be a mediastinal mass upon surgical exploration. A joint cardiothoracic and general surgical approach was planned for resection of both masses. Pathology of the masses showed thyroid tissue in both the mediastinal and thymic resection sites. The masses were not connected in any way, having us believe that the patient had a multinodular goiter, ectopic thyroid tissue of the mediastinum and ectopic thyroid tissue of the thymus.

Conclusion: Generally speaking, surgical excision of a mediastinal mass is warranted. This proves beneficial in terms of diagnosis and potentially primary treatment. When considering a mediastinal mass, the diagnosis of ectopic thyroid tissue should be in the differential. Patients with thyroid disease are more likely to have ectopic thyroid tissue, and a presentation of two different types of ectopic thyroid tissue is very rare.

Keywords: Ectopic thyroid; Mediastinal thyroid; Thymic thyroid tissue; Mediastinal mass; Thyroid goiter; Multinodular goiter

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Received Date: 15 Nov 2019

Accepted Date: 11 Dec 2019

Published Date: 16 Dec 2019

Citation:

Shortridge K, Brown A, Fang D. A Case of Double Ectopic Thyroid in the Mediastinum and Thymus: A Case Presentation and Literature Review. *Ann Clin Case Rep.* 2019; 4: 1770.

ISSN: 2474-1655

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Introduction

Ectopic thyroid is defined as thyroid tissue in any location other than its position in the anterior neck. The thyroid gland usually sits between the 2nd and 5th tracheal rings [1]. Ectopic thyroid tissue is the result of abnormal gland migration from the foramen caecum to its ectopic location [2]. Dual ectopic thyroid has been described [2]. This is described as having a focus of ectopic thyroid tissue also with a thyroid gland in its normal function and anatomical location [2]. The most common ectopic thyroid location is the lingual region [3]. There are very few cases of dual ectopic thyroid. There are even fewer cases of triple ectopic thyroid cases reported [3]. Embryologically the thyroid originates from the 1st and 2nd pharyngeal pouches. The first case of ectopic thyroid tumor was reported in 1869 by Hickman. The prevalence in the general population is 1 per 100,000 to 300,000. Patients with thyroid disease have a much higher prevalence than the normal population at 1 per 4,000 to 8,000.

Ectopic thyroid tissue is rare, and mediastinal ectopic thyroid tissue is a small subset of this population. In fact, ectopic intrathoracic thyroid tissue is 5.8% of all mediastinal masses [4] and only around 1% of ectopic thyroid tissue [5,6]. Several cases have been reported including intracardiac, intrapericardial, lateral chest wall, pulmonary and the ascending aorta [5,7-9]. In our case, we present a patient with ectopic mediastinal thyroid tissue abutting the trachea. There was no connection to the cervical thyroid gland in our case.

Case Presentation

A 30-year-old female with past medical history of fibromyalgia, chronic anxiety, bipolar depression, migraine, neuropathy, restless leg syndrome, and asthma presented with a multinodular nontoxic goiter. She had a known history of hypothyroidism and goiter for 5 years. She was referred

Table 1: Reported mediastinal ectopic thyroid in the last ten years.

Year	Author	Country	Title	Diagnosis
2013	Eun Roh, et al. [6]	Korea	A case of mediastinal ectopic thyroid presenting with a paratracheal mass	Paratracheal Ectopic Thyroid Mass. Benign Thyroid Tissue.
2017	Jed Hummel, et al.	USA	Ectopic thyroid tissue in the mediastinum characterized by histology and functional imaging with I-123 SPECT	Mediastinal Ectopic Thyroid Tissue. Benign Thyroid Tissue.
2014	Woo Surng, et al.	Korea	A 7.3 cm × 5.3 cm × 3.5 cm heterotopic thyroid in the posterior mediastinum in a patient with situs inversus totalis	Posterior Mediastinal Ectopic Thyroid. Benign Thyroid Tissue.
2017	Jing Wen Hu, et al.	China	Ectopic thyroid cancer diagnosed by endobronchial ultrasound guided transbronchial needle aspiration	Papillary Carcinoma
2018	Alessio Metere, et al.	Italy	Diagnosis and management of a mediastinal ectopic thyroid laying on the right bronchus: case report and review of literature.	Right Bronchus Ectopic Thyroid Tissue. Benign Thyroid Tissue.
2018	Mohamed Regal, et al.	Saudia Arabia	Mediastinal ectopic thyroid mass with normal thyroid function and location: case report	Mediastinal Ectopic Thyroid Mass. Benign Thyroid Tissue.
2018	Yazdan, Raji, et al.	India	Ectopic thyroid: the great mimicker	Mediastinal Ectopic Thyroid Tissue. Benign Thyroid Tissue.



Figure 1: CT scan showing left paratracheal mass behind the manubrium causing right sided deviation of the trachea.



Figure 3: Paratracheal and thymic mass removed in continuity.



Figure 2: Median sternotomy showing left sided paratracheal mass and thymic mass.

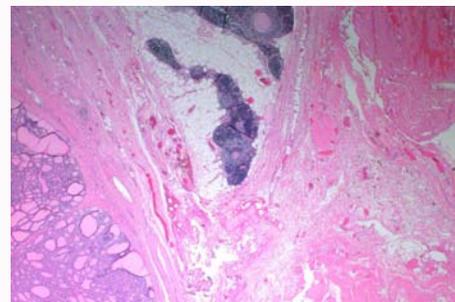


Figure 4: Thymic mass: 20x –left to right: Thyroid tissue, Thymic tissue, Skeletal muscle.

to our general surgery clinic by her endocrinologist when the patient was complaining of cough and obstructive symptoms. She presented in 2017 with an enlarging thyroid mass. She has no history of neck radiation. No family history of thyroid cancer.

On physical examination there was a palpable, enlarged thyroid with the left side being larger than the right. US guided biopsy was ordered and showed indeterminate architecture with more nodules noted on the left lobe than on the right. The patient was consented for surgery and a total thyroidectomy was performed. In surgery the left and right lobe were identified as well as the isthmus. The entire weight was 19 grams. Pathology report showed left lobe with multinodular goiter. The left lobe measured 6 cm × 4.5 cm × 2.4 cm. The right lobe showed multinodular hyperplasia and measured 4 cm × 2.2 cm × 1.2

cm. The specimens were negative for any malignancy.

Patient performed well postoperatively and was discharged home. However, one month after her initial surgery she continued to have symptoms of neck tightness and non-productive cough. A CT chest was ordered to assess for any abnormalities. The CT chest without contrast showed a 3.5 cm mass of the left cervical trachea causing displacement of the trachea to the right. Also, of note on the CT chest was a thymic mass. It was decided to proceed to a CT guided biopsy of the thymic, as this was more easily accessible. CT guided biopsy of the thymic mass revealed residual benign thyroid parenchyma with Hurthle cell metaplasia, degenerative changes, and dystrophic calcifications. No frank papillary carcinoma was noted. The CT scan was read as residual thyroid tissue in the neck, but IR biopsy noted the mass to actually be behind the manubrium and

Table 2: Examples of Ectopic Thyroid Tissue Locations [5].

Year	Author	Country	Title	Diagnosis
2016	Cao L, et al.	China	Clinical Characteristic and molecular pathology of skull ectopic thyroid cancer	Skull Ectopic Thyroid Cancer
2013	Ko HH, et al.	Korea	Ectopic intrapulmonary thyroid: a case report	Intrapulmonary Ectopic Thyroid
2013	Wang SC, et al.	Taiwan	Ectopic Thyroid Tissue in the Adrenal Gland Mimicking a Pheochromocytoma	Adrenal Ectopic Thyroid
2017	Tamaki S, et al.	Japan	Laparoscopic resection of retroperitoneal ectopic thyroid tissue	Retroperitoneal Ectopic Thyroid
2003	Gamblin TC, et al.	USA	Ectopic Thyroid	Thoracic Ectopic Thyroid
2004	De Felice M, et al.	Italy	Thyroid development and its disorders: genetics and molecular mechanisms	Sublingual Ectopic Thyroid
2007	Ozpolat B, et al. [7]	Turkey	Ectopic thyroid gland on the ascending aorta with a partial pericardial defect: report of a case	Ascending Aorta Multinodular Goiter

Table 3: Triple Ectopic Thyroid.

Year	Author	Country	Title	Diagnosis
2011	Santosh, Konde, et al. [3]	India	Triple Ectopic Thyroid	Lingual (foramen cecum), Sublingual, Infrahyoid
2011	Sjit Nilegaonkar, et al. [17]	India	Triple ectopic thyroid: A rare entity	Base of tongue, Hyoid, Suprahyoid
2013	Kuramoto R, et al. [18]	Japan	Triple Ectopic Thyroid: A case report and review of literature	Three ectopic thyroid glands located near the hyoid bone with normally positioned thyroid

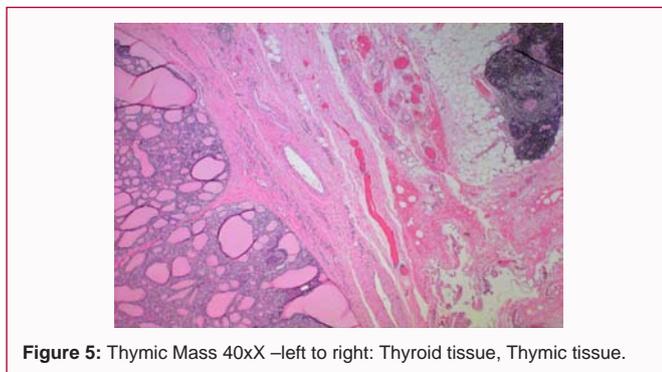


Figure 5: Thymic Mass 40xX –left to right: Thyroid tissue, Thymic tissue.

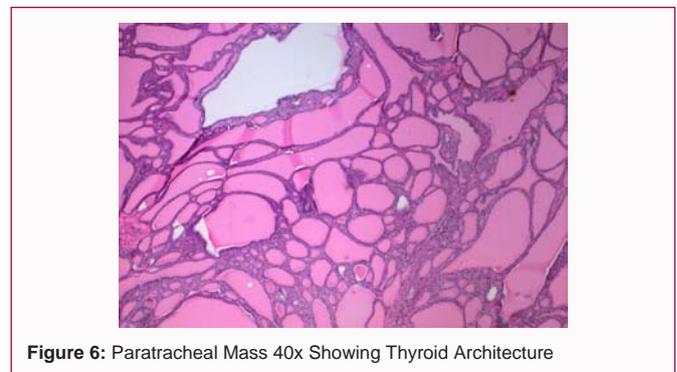


Figure 6: Paratracheal Mass 40x Showing Thyroid Architecture

entering the mediastinum. A bronchoscopy was completed, and there was no invasion into the trachea.

It was decided that removal of the aforementioned masses was necessary in the treatment and diagnosis for this patient. These masses pose concern as they could have malignant potential or foci of malignancy not seen on biopsy. A combined general surgery and cardiothoracic surgery approach through a median sternotomy was decided upon. The left paratracheal mediastinal mass was removed by the general surgery team and the thymic mass was removed by cardiothoracic surgery team. Of note, these two masses did not communicate with each other. They were however removed in one en bloc segment connected by lymph tissue and fibrous tissue. The thymic mass was 9.2 cm × 5.0 cm × 2.2 cm. The histology showed benign thyroid tissue with nodular hyperplasia and benign thymic tissue. The paratracheal mass was 5.0 cm × 3.0 cm × 2.8 cm in size. The histology showed benign thyroid tissue with nodular hyperplasia and benign lymph nodes.

As was the case with the first operation, the patient had an uneventful postoperative course. The patient on follow up had well healing incisional sites and improved symptoms. The patient no longer had a cough or obstructive symptoms. The patient will continue postoperative follow up with her primary care physician for any recurring symptoms.

Discussion

Mediastinal ectopic thyroid is a rare entity and account for only

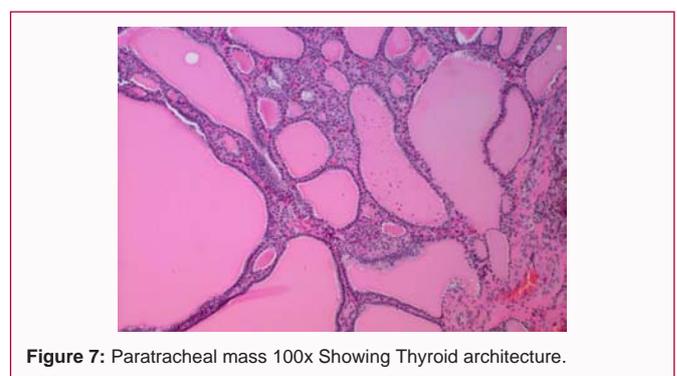


Figure 7: Paratracheal mass 100x Showing Thyroid architecture.

1% of mediastinal masses [10]. Very few cases are reported yearly. When there is no connection between the cervical thyroid, it is termed PMG or primary mediastinal goiter [11]. The blood supply for primary mediastinal goiters are derived from intrathoracic vessels and not from cervical thyroid vessels [11]. A large percentage of PMGs originate in the anterior mediastinum [11].

Approximately 90% of all ectopic thyroid is found on the wall of thyroglossal duct cysts. This means that the majority of ectopic thyroid tissue will be located in the neck. Occasionally there will be ectopic thyroid tissue present in the mediastinum. The thyroid may or may not be in continuity with the original thyroid tissue [12]. Often times they present alongside a normal sized thyroid in the neck [12]. Ectopic thyroid tissue has been described in the presence of benign

and malignant thyroid cancer [12].

In the absence of thyroid disease, a PMG is generally an incidental finding [11]. Today, many CT scans are ordered on a regular basis in the emergency room or other settings. The increased availability of high-resolution imaging is responsible for the increased incidence of incidental mediastinal masses found on imaging. If there is enlargement of the glandular tissue there could be a symptomatic presentation. If a PMG was to press against the trachea, like in our patient, the patient could present with symptoms such as cough, shortness of breath, dyspnea, stridor or others. In our case, the patient presented with a cough [11]. Patients could present with many other possible symptoms depending on the exact location and nearby anatomy.

A reasonable differential diagnosis for this condition would be lymphoma, lymphadenopathy, teratoma, thymoma, neurogenic tumors, primary esophageal tumors, primary tracheal tumors, vascular malformation & aortic aneurysm dilatation [13]. This is a short list of some common mediastinal mass diagnoses; however there are many other possible etiologies for a mediastinal mass.

In a primary mediastinal goiter, there is concern for potential cancer development. There are minimal guidelines for the management of PMG [11,14]. If a mediastinal mass is encountered incidentally, a specific tissue diagnosis should be attained [14,15]. Depending on the location of the mass, a percutaneous tissue biopsy can be performed. This will give definitive histological diagnosis and help differentiate between the many possible causes. Management will depend on the final diagnosis, whether the lesion is benign or malignant, whether there is atypia, dysplasia or signs of a high-grade lesion. If a benign, low risk lesion in an asymptomatic patient is found, there is an argument that could be made for regular imaging and watchful waiting. However, several authors who have treated patients with PMG state that surgical resection is the gold standard [14,15]. Most authors state that primary mediastinal goiter is an indication for surgical resection [4,6,10,14,15]. Most surgical resection will be performed through either a sternotomy or thoracotomy depending on the location and other factors. Some authors have advocated for a cervico-mediastinal approach for successful removal of large thyroid masses extending from the cervical region to the mediastinum [4]. In our case, the cervical thyroid tissue was removed initially, and then a secondary sternotomy was performed to remove the other two ectopic tissues. If three foci of ectopic thyroid were known initially, a cervico-mediastinal approach would have been an excellent option as it provides excellent visualization and ability to remove multiple foci [4].

A literature review of PubMed for the last ten years revealed 7 reported cases of ectopic thyroid tissue in the mediastinum. Although rare, the most common site outside of the neck for ectopic thyroid tissue is in the mediastinum. Typically, these are reported to be treated with resection due to the broad differential of cancers that can present as mediastinal masses [16]. However, most of the reported cases of ectopic thyroid tissue in the mediastinum are benign without hyperplasia or concerning features. Thus, if a tissue diagnosis of thyroid tissue can be obtained prior to excision, serial monitoring by radiographic imaging may be appropriate in patients in whom thoracic surgery is a high risk (Table 1).

Of interest, although ectopic thyroid of the mediastinum is rare, other cases have been presented this last century in even more rare

locations. Below is a table extracted from a literature review by Metere et al. [5] discussing other ectopic thyroid locations both in and out of the mediastinum (Table 2).

Dual ectopic thyroid gland presentation is rare. There are however several cases of triple ectopic thyroid tissue that have been presented. Below is a list of three cases that have been presented in the past decade (Table 3).

References

- Nasiru Akamnu I, Idowu Olusegun F. Ectopic thyroid: etiology, pathology and management. *Hormones*. 2011;10(4):261-9.
- Roh E, Hong E, Ahn H, Park S, Yoon H, Park K, et al. A case of mediastinal ectopic thyroid presenting with a paratracheal mass. *Korean J Intern Med*. 2013;28(3):361-4.
- Konde SR, Singh H Retd, Pawar A, Sasane A. Triple ectopic thyroid. *Med J Armed Forces India*. 2012;68(2):173-5.
- Di Crescenzo V, Vitale M, Valvano L, Napolitano F, Vatrella A, Zeppa P, et al. Surgical management of cervico-mediastinal goiters: Our experience and review of the literature. *Int J Surg*. 2016;28:S47-S53.
- Metere A, De Giacomo T, Vergine M, Biffoni M, Giacomelli L. Diagnosis and management of a mediastinal ectopic thyroid laying on the right bronchus: case report and review of literature. *BMC Surg*. 2018;18:19.
- Roh E, Hong E, Ahn H, Park S, Yoon H, Park K, et al. A case of mediastinal ectopic thyroid presenting with a paratracheal mass. *Korean J Intern Med*. 2013;28(3):361-4.
- Özpolat B, Do an O, Gökaslan G, Ereku S, Yücel E. Ectopic Thyroid Gland on the Ascending Aorta with a Partial Pericardial Defect: Report of a Case. *Surg Today*. 2007;37(6):486-8.
- Peng E, Oxenham, H, Foley M, Goodwin A. Right ventricular outflow tract tumour: an unsuspected intracardiac ectopic thyroid mass. *Interact Cardiovasc Thorac Surg*. 2013;17(5):903-5.
- Besik J, Szarszoi O, Bartonova A, Netuka I, Maly J, Urban M, et al. Intracardiac Ectopic Thyroid (Struma Cordis). *J Card Surg*. 2014;29(2):155-8.
- Regal M, Kamel MM, Alyami H, Al-Osail EM. Mediastinal ectopic thyroid mass with normal thyroid function and location: Case report. *Int J Surg Case Rep*. 2018;52:5-7.
- Vargas-Abrego B, Alvarez-Maldonado P, Alcazar-Lopez R, Lorente-Ludlow F, Redondo C. Primary Mediastinal Goiter Resected *via* Cervical Approach. *Int J Respir Pulm Med*. 2015;2(1):1-3.
- Adelchi C, Mara P, Melissa L, De Stefano A, Cesare M. Ectopic thyroid tissue in the head and neck: a case series. *BMC Res Notes*. 2014;7:790.
- Koduri P. The Diagnostic Approach to Mediastinal Masses. *Ann Thorac Surg*. 2004;78(5):1888.
- Oueriachi FE, Hammoumi MM, Arsalane A, Slaoui O, Diouri H, Kabiri E. Primary mediastinal goiters. *Springer Plus*. 2014;3:503.
- Cichoń S, Anielski R, Konturek A, Baczyński M, Cichoń W, Orlicki P. Surgical management of mediastinal goiter: risk factors for sternotomy. *Langenbecks Arch Surg*. 2008;393(5):751-7.
- Li W, van Boven W, Annema J, Eberl S, Klomp H, de Mol B. Management of large mediastinal masses: surgical and anesthesiological considerations. *J Thorac Dis*. 2016;8(3):E175-E184.
- Nilegaonkar S, Naik C, Sonar S, Hirawe D. Triple ectopic thyroid: A rare entity. *Indian J Nucl Med*. 2011;26(4):194-5.
- Kuramoto R, Oikawa K, Fujita K, Oridate N, Fukuda S. Triple Ectopic Thyroid: A Case Report and Review of Literature. *Thyroid Disorders Ther*. 2013;2(2):126.