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Dual Ectopic Thyroid: A Case Report and Review of Literature

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Authors' contributions

This work was carried out in collaboration among all authors. Author NSS designed the study, wrote the protocol and wrote the first draft of the manuscript. Authors YLT and SM managed the literature searches. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

The ectopic thyroid gland is a rare clinical entity originates from the failure of the thyroid gland to descend from the foramen caecum to its normal eutopic pre-laryngeal site. It is further uncommon for two distinct ectopic thyroid tissues to be present simultaneously. We present a case of a 35-year-old lady, who has dual ectopic thyroid with symptoms of dysphagia. As the clinical presentations were non-specific following a series of negative investigations, she was treated as functional dyspepsia. The eventual diagnosis was reached only after performing an ultrasound of the neck, contrast-enhanced CT scan, and thyroid scintigraphy. Its management is still controversial; generally surgery is aim to alleviate bothering symptoms of obstruction. Asymptomatic patients are managed with thyroxine supplements, aiming for clinical improvement of symptoms and gland size reduction. This case report will address concerns of managing dual ectopic thyroid.

Keywords: Lingual thyroid; ectopic thyroid; thyroid scan.

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1. INTRODUCTION

Ectopic thyroid is a rare clinical entity caused by failure of in descendence of thyroid gland to its normal position during embryogenesis. Clinical evidence varies between 1: 3000 and 1:100000 [1]. It is more common in women than men ranging from 4:1 to 7:1 ratio [2]. 33% of these patients have hypothyroidism. The presentation may vary from an accidental finding to life threatening airway obstruction. A Technetium thyroid scan is the most important diagnostic technique to help in the identification of ectopic thyroid. Most cases are treated medically unless obstructive complications present thus warrants surgery.

2. CASE REPORT

A 35 years old Malay lady was referred to a surgical clinic with complaint of difficulty in swallowing for 6 months to solid food. She had no other constitutional symptoms and systemic inquiries were unremarkable. She was diagnosed with subclinical hypothyroidism previously and was started on levothyroxine 50 mcg daily.

On physical examination, it was noticed she had raised base of tongue, with no obvious mass seen. Neck examination revealed neither palpable thyroid gland nor any other palpable mass. A rigid 70 degree scope showed base of tongue occupying the valleculae with no obvious mass.

Her thyroid function tests were suggestive of subclinical hypothyroid with value of T_4= 10.6 (normal=7.9-14.4 pmol/L) and Thyroid Stimulating Hormone (TSH) = 6.4 (normal = 0.34-5.60 mU/L). Other laboratory tests were within normal limits. CT-scan of the neck showed an empty thyroid bed with two well-defined hyperdense lesions demonstrating similar density and enhancement of the thyroid gland, seen at the base of tongue. Technetium (Tc-99 m) thyroid scan revealed isotope uptake was seen in the midline, superior to the hyoid cartilage, with reduced intensity suggestive of hypofunction. No scan evidence of functioning thyroid tissue in the thyroid bed. The final diagnosis of dual ectopic thyroid was made based on thyroid scan findings.

We have started the patient on Levothyroxine 75 mcg OD to control her hypothyroidism. The patient was planned for re-evaluation after 3 months to see any clinical improvement before surgical intervention.

Serial follow up and a repeat CT Neck a year later showed that both the ectopic glands has slightly reduced in size; from 1.7x1.5 to 1.6x1.3 cm and the slightly posteriorly located gland was 1.4x1.8 from the previous size of 1.9x1.8 cm.

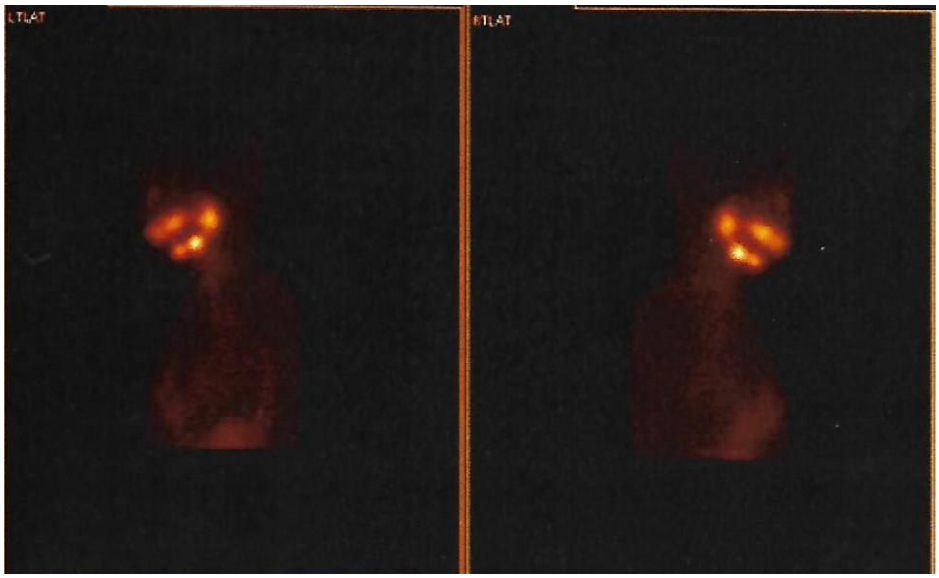

Fig. 1. Scintigraphy – lateral view, focus of increased tracer uptake seen in the midline, posterior to lingual region. Absence of uptake in thyroid bed

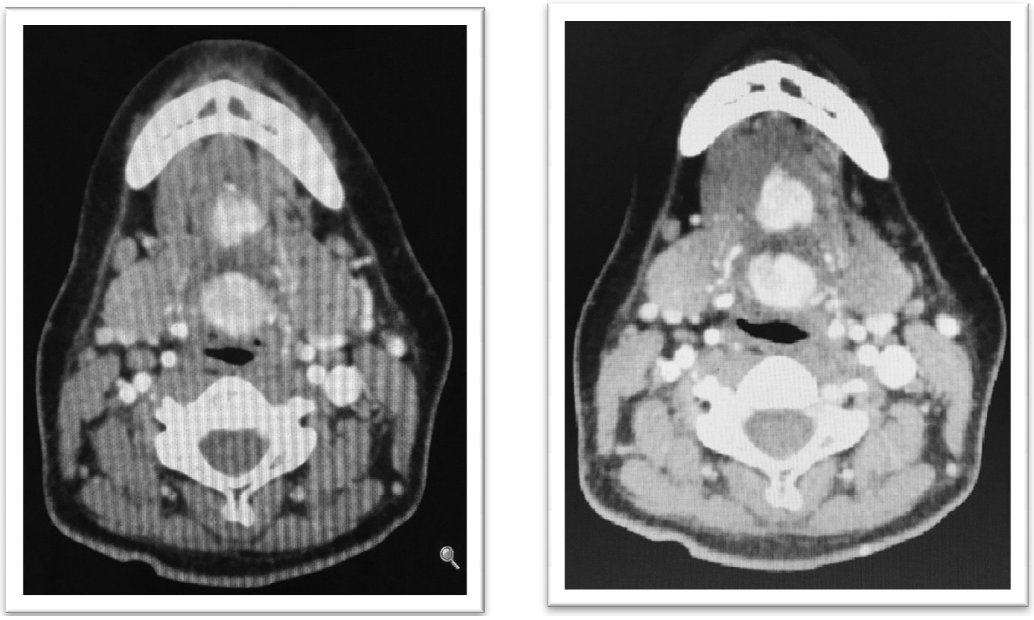

Fig. 2. Comparative CT Neck within a year showed two well defined hyperdense lesions suggesting thyroid gland at suprahyoid level, at the base of tongue showing reduction in sizes in the latter

3. DISCUSSION

Hickman first described the ectopic thyroid in 1869 [1,2]. The clinical incidence varies between 1:3000 to 1:100000 [1]. It is more common in women than men, with a ratio ranging from 4:1 to 7:1 [2]. It is due to the increase in metabolic demand during puberty, pregnancy, or menopause; the plasma TSH level increases in the body leading to the hypertrophy of ectopic tissue [2,3]. Approximately 90% of the cases are ectopic lingual thyroid [2]. The disorder displays two peaks, at the age of 12 and at the age of 50. About 70-80% of patients do not have functional thyroid tissue elsewhere with 33% of them being with hypothyroidism. However, most patients are euthyroid and a few have hyperthyroidism [1].

During embryogenesis, proliferation of endodermal tissue between *tuberculum impar* and *hypobranchial eminence* form the thyroid gland. Normally, the thyroid gland will descend along a path from *foramen caecum* in the tongue to its final position in front and lateral to the second, third, and fourth tracheal ring by 7 weeks of gestation. Arrest of these descent process anywhere along this path result in ectopic thyroid. The term ectopic thyroid refers to functioning thyroid tissue found outside the normal thyroid location. Ectopic tissue has also been found in the mediastinum, heart, esophagus, and diaphragm [4-7]. Lingual thyroid is the result of failure of descent of the thyroid anlage from foramen the caecum of the tongue [8].

Upon review of all the cases readily published and the present case (from Table 1 [2-23]), we found that the mean age of these patients was 29 years with a range from 4 years to 62 years. It is more common in women than in men with a M/F ratio of 1:31.The presentation varies from asymptomatic [9] to anterior neck swelling with or without altered thyroid status. Most of the cases reported as hypothyroidism while only a case reported with Graves disease [10].

In children, they may suffer from failure to thrive [11,12] and mental retardation (neuro-psychomotor development) caused by hypothyroidism pathology or severe respiratory distress due to obstruction [8].

Most of the patients usually diagnosed as a result of symptomatic manifestation related to the obstruction of oropharynx such as dysphagia, dysphonia, dyspnea, feeling fullness at the throat, stridor and obstructive sleep apnea [9]. Due to the vascularized structure some may present as bleeding [13] but rarely described.

Table 1. Presentation and management of ectopic thyroid cases

Case	Author	Age	Gender	Presenting symptom	Thyroid hormone level		Management		Site of ectopic thyroid
					TSH	T4	Surgical intervention	Conservative	
1	Iqbal et al. [2]	16	female	dysphagia	N	N	transoral approach		lingual
2	A.Toso et al. [3]	62	female	FB sensation	↑	N	endoscopic micro-laryngo surgery		lingual
		42	female	Fb sensation	N	N	external approach with tracheostomy		lingual
3	Tuncay et al. [4]	20	female	ant neck swelling	N	N	excision of infrahyoid thyroid	conservative of lingual thyroid	Infrahyoid lingual
4	Valeria et al. [6]	30	female	neck pain (incidental)	↑	N	N/A		lingual
5	Sood et al. [7]	5	Female	Midline neck swelling	↑	N		Levothyroxine	Subhyoid Lingual
6	Bassem et al. [8]	5	female	dysphagia	N	N	transoral approach excision		lingual
7	Eder et al. [19]	8	female	dysphagia FB sensation	↑	N		Levothyroxine	lingual
8	Col SS Anand et al. [12]	4	female	reduced appetite constipation	↑	↓		Levothyroxine	lingual
9	Ramzisham et al [22].	57	female	dysphagia dysphonia stridor	↑	N	midline mandibulectomy and tongue splitting		lingual
10	Surej Kumar et al. [14]	40	female	FB sensation	↑	↓	split mandibulectomy		lingual
11	Kumar et al. [18]	32	female	mass at back of tongue dysphagia	↑	↓		levotyroxine	lingual
12	S. Rabie et al. [21]	19	female	dyspnea dysphagia	N/A		transoral coblation		lingual
		10	female	dysphagia cough	N/A		transoral coblation		lingual

Case	Author	Age	Gender	Presenting symptom	Thyroid hormone level		Management		Site of ectopic thyroid
					TSH	T4	Surgical intervention	Conservative	
		20	female	speech problem swallowing problem	N/A		transoral coblation		lingual
		23	female	dysphagia	N/A		transoral coblation		lingual
		8	female	massive bleeding	N/A		transoral coblation		lingual
13	Saad et al. [23].	5	female	midline neck swelling	↑	N		levothyroxine	subligual subhyoid
14	Tan et al. [10].	25	female	swelling at throat	↓	↑		PTU	lingual subhyoid
15	Yang et al. [24]	26	female	rt side anterior neck swelling	N	N		levothyroxine	rt at thyroid bed left at lingual (hemiagenesis)
16	Lin et al. [16]	43		lump at throat FB sensation	N/A		N/A		lingual
17	Pokhraj et al. [15]	12		dysphagia, dysphonia	N	N	transoral approach		lingual
18	Sudke et al. [13]	24		hematemesis	N	N		Observation	lingual
19	Ralli M et al. [17]	16		dysphagia	↑	N		levothyroxine	lingual
20	Patil et al. [9]	12	Female	Stomatolalia	Hypothyroid			levothyroxine	Lingual
		25	Female	Dysphagia/ dyspnaoe	Hypothyroid			levothyroxine	Lingual
		30	Male	Asymptomatic	Hypothyroid			levothyroxine	Lingual
		16	Female	Dysphagia	Euthyroid		Suprahyoid approach		Lingual
		19	Female	Blood stain sputum	Hypothyroid			levothyroxine	Lingual
		22	Female	Dysphagia	Hypothyroid			levothyroxine	Lingual
21	Present	35	Female	dysphagia	N	N		levothyroxine	Lingual sublingual

As previously mentioned, the most common site of ectopic thyroid is in the lingual area, where almost all the literature reported ectopic lingual thyroid. On physical evaluation, lingual thyroid usually presents itself as midline nodular mass, painless, soft, smooth or irregular surface, with highly vascularized or red-colored structure at the base of tongue. Palpation of the neck is extremely essential in order to check the presence or absence of the thyroid gland in a normal position. The rigid and/or flexible endoscopy allows photo-documentation of the size and position of the lesion, observation of the larynx and assessment of adequate airway.

The most important diagnostic tool is radioactive iodine thyroid scan and thyroid scan with Technetium-99 m [3,6,7]. The radioactive iodine thyroid scan is the most sensitive and specific test for detection of existence, size, distribution and activity of ectopic thyroid tissue and to see the presence or absence of thyroid tissue at usual location [1]. While thyroid scan with Technetium-99 is less specific in view of false-positive results in case of physiological uptake by salivary gland and nasal mucosa, pathological uptake as seen in sialo-adenitis[14], but with better cost-benefit. It also can provide the information regarding presence of other thyroid tissue at other sites. Approximately 75% of patients, the ectopic is the only functioning thyroid tissue in the body [3]. Therefore, follow up for patient is crucial to avoid the complication of post-operative hypothyroidism. Other modalities such as Ultrasound, CT scan and MRI may help in defining the extension and location of the ectopic thyroid gland before surgery [3,6,15]. The head and neck ultrasonography is a non-invasive image tool for evaluation of the presence of ectopic and orthotopic thyroid tissue, especially when scintigraphy is unavailable or contraindicated [16].

Thyroid function tests are also essential in the initial investigation, due to the high possibilities with hypothyroidism. The guided FNAC (fine needle aspiration cytology) can also confirm to differentiate benign from malignant [17].

Management of ectopic lingual thyroid is still controversial [3,18]. Treatment modalities employed in lingual thyroid depends on factors such as the general condition of the patient, size, and degree of discomfort. Euthyroid patients and asymptomatic patients are followed up regularly to be aware of complications without any treatment. Supplemental thyroxine should be given in symptomatic hypothyroid patients [9,12, 14,19].

Exogenous hormones could cause a suppressive action in the gland, reducing its size or preventing hypertrophy or goitrous enlargement. The goal of this treatment is to suppresses the production of TSH to avoid increase volume of ectopic gland [1].

When medical therapy fails and patients are symptomatic and have complications, surgery is the treatment of choice. The malignant transformation also has been described [3,20] and for this reason, some authors consider complete surgical removal of the gland as appropriate treatment.

In literatures, several surgical approaches have been described; divided into a transoral approach or external approach. Transoral approach is the most widely employed technique for smaller lesions as they avoid injury to deep neck structures such as lingual nerve, fistula formation, and deep cervical infection; however it provides limited accessibility. Cold instruments with monopolar coagulation, laser CO2 and coblation assisted excision [21] has been used in this approach.

External approaches such as lateral pharyngotomy, transhyoid techniques [9] and midline mandibular split osteotomy with tracheostomy [14,22] is recommended for larger mass. These approaches have better control of bleeding and accessibilities.

Ablative radioiodine therapy is an alternative approach recommended in older patients or patients who are deemed unfit for surgery [3,9]. However, this treatment is less suitable for children and young adults as it may damage the gonads or other organs, especially the required dose is generally high [3].

4. CONCLUSION

Lingual ectopic thyroid is a rare developmental anomaly, hence the controversial treatment option for these patients. Conservative with substitutive hormone treatment should be considered in patients with mild symptoms, while severe cases with airway obstruction and dysphagia should opted for surgical removal. Evaluation of thyroid function is recommended to assess hypothyroidism. In the present case, we opted for conservative management with control

of thyroid hormones status; concurrent with her level of symptoms severity, which warranted off surgery. However, re-evaluation of clinical improvement during follow up will guide the decision of surgical intervention. A Technetium thyroid scan is the most important diagnostic technique to help in the identification of ectopic thyroid before any treatment. When complications such as dysphagia or dyspnoea occur, surgery is necessary. The treatment approach should be tailored according to the patient's age, the status of the thyroid gland, and the presence of complicating factors such as obstructive symptoms.

CONSENT

As per international standard or university standard, patient's written consent has been collected and preserved by the author(s).

ETHICAL APPROVAL

It is not applicable.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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